PARENTS WITH MENTAL AND/OR SUBSTANCE USE DISORDERS AND THEIR CHILDREN, VOLUME II

EDITED BY: Joanne Nicholson, Jean Lillian Paul, Joanne Louise Riebschleger
and Anja Wittkowski

PUBLISHED IN: Frontiers in Psychiatry and Frontiers in Public Health

frontiers Research Topics

ISSN 1664-8714
ISBN 978-2-83250-344-7
DOI 10.3389/978-2-83250-344-7

About Frontiers

Frontiers is more than just an open-access publisher of scholarly articles: it is a pioneering approach to the world of academia, radically improving the way scholarly research is managed. The grand vision of Frontiers is a world where all people have an equal opportunity to seek, share and generate knowledge. Frontiers provides immediate and permanent online open access to all its publications, but this alone is not enough to realize our grand goals.

Frontiers Journal Series

The Frontiers Journal Series is a multi-tier and interdisciplinary set of open-access, online journals, promising a paradigm shift from the current review, selection and dissemination processes in academic publishing. All Frontiers journals are driven by researchers for researchers; therefore, they constitute a service to the scholarly community. At the same time, the Frontiers Journal Series operates on a revolutionary invention, the tiered publishing system, initially addressing specific communities of scholars, and gradually climbing up to broader public understanding, thus serving the interests of the lay society, too.

Dedication to Quality

Each Frontiers article is a landmark of the highest quality, thanks to genuinely collaborative interactions between authors and review editors, who include some of the world's best academicians. Research must be certified by peers before entering a stream of knowledge that may eventually reach the public - and shape society; therefore, Frontiers only applies the most rigorous and unbiased reviews.
Frontiers revolutionizes research publishing by freely delivering the most outstanding research, evaluated with no bias from both the academic and social point of view. By applying the most advanced information technologies, Frontiers is catapulting scholarly publishing into a new generation.

What are Frontiers Research Topics?

Frontiers Research Topics are very popular trademarks of the Frontiers Journals Series: they are collections of at least ten articles, all centered on a particular subject. With their unique mix of varied contributions from Original Research to Review Articles, Frontiers Research Topics unify the most influential researchers, the latest key findings and historical advances in a hot research area! Find out more on how to host your own Frontiers Research Topic or contribute to one as an author by contacting the Frontiers Editorial Office: frontiersin.org/about/contact

PARENTS WITH MENTAL AND/OR SUBSTANCE USE DISORDERS AND THEIR CHILDREN, VOLUME II

Topic Editors:
Joanne Nicholson, Brandeis University, United States
Jean Lillian Paul, Medizinische Universität Innsbruck, Austria
Joanne Louise Riebschleger, Michigan State University, United States
Anja Wittkowski, The University of Manchester, United Kingdom

Citation: Nicholson, J., Paul, J. L., Riebschleger, J. L., Wittkowski, A., eds. (2022). Parents With Mental and/or Substance Use Disorders and Their Children, Volume II. Lausanne: Frontiers Media SA. doi: 10.3389/978-2-83250-344-7

Table of Contents

frontiers | Frontiers in Psychiatry

TYPE Editorial
PUBLISHED 13 September 2022
DOI 10.3389/fpsyt.2022.1020660

OPEN ACCESS

EDITED AND REVIEWED BY
Rahul Shidhaye,
Pravara Institute of Medical
Sciences, India

*CORRESPONDENCE
Joanne Nicholson
jnicholson@brandeis.edu

SPECIALTY SECTION
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

RECEIVED 16 August 2022
ACCEPTED 29 August 2022
PUBLISHED 13 September 2022

CITATION
Nicholson J, Paul JL, Riebschleger J
and Wittkowski A (2022) Editorial:
Parents with mental and/or substance
use disorders and their children,
volume II.
Front. Psychiatry 13:1020660.
doi: 10.3389/fpsyt.2022.1020660

Editorial: Parents with mental and/or substance use disorders and their children, volume II

Joanne Nicholson[1]*, Jean Lillian Paul[2], Joanne Riebschleger[3] and Anja Wittkowski[4]

[1]The Heller School for Social Policy and Management, Institute for Behavioral Health, Brandeis University, Waltham, MA, United States, [2]Division of Psychiatry I, Department of Psychiatry, Psychotherapy and Psychosomatics, Medical University Innsbruck, Innsbruck, Austria, [3]School of Social Work, Michigan State University, East Lansing, MI, United States, [4]Division of Psychology and Mental Health, Faculty of Biology, Medicine and Health and the Manchester Academic Health Science Centre, School of Health Sciences, The University of Manchester, Manchester, United Kingdom

KEYWORDS

parents, mental health, substance use disorders, children, families, family-focused practice

Editorial on the Research Topic
Parents with mental and/or substance use disorders and their children, volume II

Introduction

The first Frontiers eBook compilation of Research Topic articles on Parents with Mental and/or Substance Use Disorders and their Children, published in 2020, included 27 papers, with over 100 contributors from 15 countries. Investigators employed diverse designs and methods to explore the experiences of parents and their families, and to develop and test interventions. While the prior Research Topic was a significant contribution to the field, families living with parental mental and/or substance use disorders remain vulnerable. The present, second volume of papers on this Research Topic fills gaps identified in the first volume, and moves the field forward by highlighting significant relationships and experiences of key stakeholders; the description and application of conceptual models and frameworks; recent innovation in intervention development, adaptation, testing, and sustainability; shifts in policy and practice paradigms toward more integrated models; and further developments in the research process, measures, and methods, particularly given the impact of the COVID-19 pandemic on parents, families, and practice.

The 34 articles in volume II of this Research Topic represent the work of 151 authors from 13 countries, with reviewers from many more, contributing to cutting-edge knowledge and identifying next steps in research, policy, and practice. Rich material is provided as supplements to several of the papers, which readers are encouraged to explore. The articles reflect progress in the field, in the development and application of theory, and intervention specification, sustainability, and impact. Contributions have shifted from describing prevalence to exploring solutions to supporting families, parents, children, and professionals at both policy and practice levels. Several groups of investigators who contributed protocol papers to volume I have summarized their findings in volume II. The research measures and methods papers in volume II provide evidence of greater stakeholder involvement in research, as co-designers and collaborators. There is increasing focus on improving outcomes for adults with mental and/or substance use disorders who are parents, as well as for the children of parents with mental illness (COPMI) across the lifespan, from considering parenthood, to pregnancy and the perinatal period, to adulthood. Colleagues are reporting on the longer-term impact of policy and practice paradigm shifts promoting the identification of the needs of whole families and collaborative efforts to address them. Innovative solutions to the challenges facing whole families may require the support of the larger context and communities in which families reside—"the village." Families may benefit from the support of both professional and natural resources in their "village," accessed formally through service delivery channels and informally through family, neighborhood, and community networks.

Significant relationships and impact

Important, comprehensive review papers contribute to the state of our knowledge. Radley et al. in the UK provide a scoping review of interventions relevant to parents with psychosis, focusing on five groups of diverse intervention components, from talking about to improving parenting skills and experiences, to support for the whole family. The authors underscore the need for RCTs, and the need to identify components effective in changing outcomes for both parents and children. In their systematic review, Reid et al., also from the UK, provide evidence for the relationships between experiences of abuse and maternal suicide ideation, attempted suicide and death, focusing on outcomes for mothers and the perinatal period. They recommend that women with experiences of domestic violence or childhood abuse be identified and provided emotional and practice support during this crucial period. Children's experiences of stigma-by-association are the focus of a systematic mixed studies review by Dobener, Fahrer, et al., investigators in Germany. The authors provide a comprehensive framework of identified aspects of stigma related to parental mental illness and group these into four dimensions (i.e., experienced, anticipated, and internalized stigma and structural discrimination); the importance of anti-stigma interventions and campaigns is emphasized. The potential for expressed emotion to contribute to the transgenerational transmission of mental disorders is examined by Fahrer et al. also in Germany. Their systematic review highlights the dearth of studies on expressed emotion in families in which a parent has a mental illness.

Experiences of key stakeholders

Fathers are the focus of a single paper in this volume, in which Doi et al. in Japan examine the relationship between workplace and community social capital, and fathers' postpartum depression and anxiety. Community social capital (i.e., social support and resources) was found to be inversely related to symptoms of depression and anxiety, suggesting the potential benefit of promoting paternal social support in the perinatal period. Sabella et al. in the USA employ life story qualitative research methods to explore the experiences of young adult parents with serious mental health conditions. Young adult parents were actively involved as researchers in this community-based participatory research study in which participants described their challenging but motivated parenting journeys. Young adults are also the focus of the study by Villatte et al. in Canada regarding the perceived social support of youth whose parents have a mental illness. Participants described themselves as important sources of support for their parents, and emphasized the need for having other support figures in their lives, a potential target for intervention. Gregg et al. compare expressed emotion and attributions in parents with and without serious mental illness. Parents with schizophrenia exhibited significantly more hostility and criticism toward their children, and less warmth, and made more child-blaming attributions. These findings suggest targets for intervention with parents and families.

Conceptual models, frameworks, and program theory

Reupert et al. place the notion of "the village" in the context of Bronfenbrenner's ecological theory to underscore the importance of promoting the capacity at all levels (e.g., individual, family, services, government) to provide support and guidance to families living with adversity. They call for further research to explore ways in which village concepts and components may play out in diverse settings with diverse families to develop interventions and evaluate impact. In-depth interviews by Bauer et al. with program implementers inform theory development, illustrating the interconnectedness between changes that need to co-occur in practitioners, parents, and children, and fragmented health systems to enable practitioners to focus on parents' strengths. Drawing from a realist approach and complex systems thinking, the authors link contextual factors with action mechanisms to disrupt the status quo and transform practice. Family-focused practices support adults in their parenting role and mental health recovery, and focus on protecting children and promoting their resilience (Allchin et al.). These investigators engaged stakeholders within adult mental health services to inform the development of a model of key elements influencing the sustainability of a particular intervention, Let's Talk about Children, and, ultimately, a sustainability model for family-focused practice, placing the work in a wider context. The authors underscore the potential benefit of recognizing the parenting status of adult clients to benefit parents, children, and families.

Intervention development, adaptation, testing, and sustainability

Articles in this volume regarding interventions focus on the engagement of and outcomes for children, parents and families. Hagström analyzed narrative structured interviews with children and parents regarding their experiences in a grief support camp in Sweden for families affected by a parent's suicide. Parents and children reported the benefits of a psychoeducational approach, open communication, and opportunities to connect with others with similar experiences, which contributed to destigmatization of their experiences. Vetri et al. conducted a formative evaluation, examining children's, parents', and workers' perceptions of bibliotherapy using a book with strategies and activities specifically targeted to the elementary school age group. The authors conclude that bibliotherapy may help children learn concrete strategies for coping with challenges, and help families initiate sensitive discussions when a parent has mental illness. A Norwegian team of investigators investigated the rate and characteristics of children's participation in Child Talks, conducting quantitative and qualitative analysis of electronic patient journal entries by healthcare professionals (Kristensen et al.). While sessions with children were relatively rare, participating children knew more about their parents' illnesses and treatment, suggesting the benefit of studying factors influencing their participation. Petzold et al. report findings from an observational study evaluating adherence to an integrated care program (i.e., "Mommy think of me") for methamphetamine-related mental disorders (e.g., ADHD, depression) in pregnant women and parents. The 15-session intervention draws from motivational interviewing, psychoeducation, and cognitive behavior therapy. Depression and ADHD were significantly related to lower participation in treatment, underscoring the importance of disseminating integrated care concepts to counter the increasing methamphetamine crisis.

Two groups of investigators report on adaptations of Triple P Positive Parenting Program resources. Outcomes of implementing the Triple-P Self-Help Workbook with guidance and support in 10 sessions with parents with psychosis were investigated by a team in the UK (Wolfenden et al.). Improvements in mental health, parenting and child behavior measures were reported and maintained by parents completing all 10 sessions. The authors provide preliminary evidence that symptoms of psychosis may be reduced by improving family functioning. A second team of UK researchers studied the feasibility and acceptability of delivering the Baby Triple-P Positive Parenting Program (BTP) to mothers with severe mental illness in an inpatient Mother and Baby Unit (Wittkowski et al.). They compared characteristics, participation and outcomes for women in two conditions: (1) treatment as usual and (2) BTP in addition to treatment as usual. The authors provide a thorough overview of their study procedures, preliminary findings, and lessons learned to inform wider implementation in existing perinatal mental health services and a future, larger RCT.

Two papers provide insight into the perspectives of families and practitioners implementing the Family Talk intervention in 15 sites in Ireland (Furlong et al.; Mulligan et al.). The two studies reported here are nested within an RCT. The vast majority of families reported substantial benefits from participating in Family Talk (e.g., increased confidence, improved communication), and identified key facilitators (e.g., non-judgmental clinician) and barriers to participation (e.g., stigma). The authors provide a comprehensive discussion of implementation issues, with recommendations for addressing them across phases of participation. Mental health clinicians and managers were interviewed to investigate their experiences implementing Family Talk and perspectives on longer-term sustainability. Participants described key factors to successful implementation, including organizational support, clinician skills, and appreciating the benefits for families. The benefits of a structured, manualized approach are highlighted, along with a call for the development of a multi-level public-health response to address societal and systemic barriers to change.

The adaptation of Let's Talk about Children (LTC) in the Massachusetts USA adult mental health services context—the ParentingWell Practice Profile—is described in detail by Nicholson et al., who delineate program theory and action mechanisms. Supplementary materials provided with this article include the ParentingWell Practice Profile, a Workbook of activities for practitioners and parents, and self-assessment resources for use in training, supervision, and coaching. The development and adaptation of LTC in various contexts, alongside the developing evidence base, is documented by Allchin and Solantaus. Drawing from their review of the literature regarding LTC, the authors identify three forms of LTC, with outcomes related to parents, and family and child wellbeing and evidence of effectiveness in implementation contexts. The contribution of this paper lies in the use of LTC as an example of an evidence-based practice developed in the context in which it was implemented, rather than the academic setting or laboratory, to guide and inspire future innovation, and support sustainability over time.

A Research Topic in psychiatry or public health in 2022 would not be complete without an article on the impact of the COVID-19 pandemic on policy, research and practice. Obradovic and Nicholson provide a perspective on pandemic-related adaptations in family-focused service delivery given the dramatic changes in people's lives, with implications for research measures, methods, and outcomes. The authors couch their perspective in the EASE Framework to highlight consequences for engagement, assessment, support, and education of family members. Treatment targets and timeframes may have shifted, and routine outcomes may have to be re-evaluated. Hopefully, pandemic-induced changes in access to and participation in services and research (e.g., virtual strategies) will help to promote engagement, and address inequities and disparities.

Cross-sector and systems level approaches

Concerns have been raised about the potential over-representation of parents with mental health and substance use disorders in the child welfare system. Effective supports for families living with parental challenges may well require cross-sector efforts as well as within-sector or within-system identification and response. Vis, Lauritzen C, Havnen, et al. in Norway tested their hypotheses regarding child protection and welfare reports in a case file study. Reported concerns about mental illness and substance abuse problems were substantiated in over half of the cases. Services were provided in just over a third of the cases, and were not more or less likely in cases about mental illness and substance abuse than in other types of cases. A second study by these researchers focuses on the involvement of children in child welfare and protective services investigations (Vis, Lauritzen, Christiansen et al.). In situations in which the parent's mental health was a concern, conversations with children were conducted much less frequently than in situations when the child's problem was the focus of the report. Investigations based on concerns regarding parental mental health took more time and effort than other investigations. The authors call for a national knowledge-based system and a focus on children's needs in child welfare. In a third study by this team, the investigators explored the extent to which children were identified in the records of patients with mental illness and substance use disorders (Reedtz et al.). The identification of minor children has increased since the Norwegian Health Personnel Act (2010), with over half identified in 2020. In slightly fewer than one-third of the cases, health personnel provided support to children. The authors conclude that children remain unidentified and underserved, and recommend enhancements in the skills of clinicians.

Everts et al. evaluate the implementation of the mandatory identification of the children of adult patients receiving mental health services in the Netherlands. The Dutch COPMI check is part of the first step in a five-step protocol, in which parental mental health is a warning sign of risk for child abuse. Patient files were examined to extract data for the study, which were complemented by focus group discussions with professionals. For the majority of adult patients, the COPMI check tool was not used. The authors recommend that a shift to a "needs/support" focus could be geared to helping children when there is no immediate threat to their safety.

An integrated family approach in mental health services often requires the collaboration of professionals from adult and child mental health services to support family members and prevent the intergenerational transmission of psychopathology (Stolper et al.). This reflects a paradigm shift from an individual practice model to a family centered model, for which many professionals are unprepared. Group interviews with professionals were conducted to explore their experiences working with families and identify the challenges in implementing a family centered model. Differences in perspectives (i.e., adult service provider vs. child provider) and loyalties contributed to challenges in setting treatment targets and in information exchange. A focus on the whole family, flexible treatment planning, and multidisciplinary consultation were perceived as contributing to success.

Contributions to research measures, methods, and processes

Several papers in this volume highlight comprehensive measurement development processes. Riebschleger et al. describe the development and initial testing of the Youth Mental Health Literacy Scale for ages 11–14. Drawn from theoretical perspectives on mental health literacy, with input from diverse stakeholders, further psychometric analyses suggested refinements in subscales and reductions in items. The result is a scale that can be useful with the general population as well as with youth with a family member with mental illness in assessing needs and testing the effectiveness of mental health literacy programs. Dobener, Stracke, et al. hone in on the challenges conveyed by stigma in developing the Children of Parents with Mental Illness—Stigma Questionnaire (COPMI-SQ) for youth aged 12–19 years. Based on extensive literature review, and discussions with experts and youth, the investigators report pilot data on the measure's psychometric properties. They describe next steps in reliability and validity testing. These measurement developments will contribute to rigorous research on the experiences and needs of youth, and to building the evidence based of effective prevention and intervention approaches.

Community/stakeholder engagement in research, co-design and co-production reflect cutting-edge approaches to the implementation of research *per se*, as well as to the development, adaptation, and testing of interventions. The facilitated, transdisciplinary process supported by the Ludwig Boltzmann Gesellschaft (LBG) is described by Kaisler and Grill. The governance structure for funded projects included diverse stakeholders—researchers, individuals with lived experience, and an open innovation expert—along with a competence group of young adult offspring of parents with mental illness. The authors highlight the challenges to researchers, including the complexity of the process and the integration of various perspectives and skillsets. Goodyear et al. reported on steps in the co-development and implementation of the "It takes a Village" collaborative practice model to promote child-focused support networks in Austria for families in which a parent experiences a mental illness. They highlight the importance of regional, context-specific solutions in designing care models. A similar co-design, co-development process is detailed by Nicholson et al., as they adapted the Let's Talk about Children model—the ParentingWell Practice Profile—as described above. A specific method for engaging mothers with mental health and/or substance use conditions in research is provided by Zisman-Ilani et al. The Virtual Community Engagement Studio (V-CES) approach was developed and piloted in the USA during the pandemic, when accessible virtual strategies for actively engaging research participants and patients became essential. The V-CES toolkit is provided as supplementary material, and offers a step-by-step, accessible, supportive approach to mothers and others from underserved or marginalized populations as research collaborators.

Several research teams highlight the importance of using data to support the development, implementation, and sustainability of preventive and supportive interventions for children, youth, and families living with parental mental illness. The team collaborating on the Danish High-Risk and Resilience Study continued in the third wave of assessment to collect a wide range of data on multiple domains of children's functioning over time (Thorup et al.). Their goal is to develop a comprehensive understanding of the developmental trajectories of children at familial risk for mental illness to identify optimal time points and domains for targeted preventive and early intervention approaches. Finally, Takalo et al. provide an example of the use of data from multiple sources in Finland, including population level, regional, and local data, to inform the implementation of the collaborative Let's Talk about Children Service Model in a pilot region. The inclusion of diverse services sectors, guided by a collective impact framework, provides the context for the sustainability of stand-alone interventions.

Next steps

Articles in this volume represent innovation in approaches and advances in our thinking about how best to work together with parents with mental and/or substance use disorders and their families to ensure positive outcomes for all family members. Challenges remain in specifying interventions and their action mechanisms in greater detail, to facilitate rigorous research with a focus on outcomes for both adults and children. Innovative perspectives on adapting and studying interventions in new contexts and sustaining them over time suggest the importance of further research, not only focused on outcomes *per se*, but on the characteristics of collaborations, contexts, and communities that support the scaling up and out, and sustainability of these efforts. Next steps in the field must focus not just on what to do, but how to do it—how to engage stakeholders effectively—parents, children, practitioners, policymakers, funders, legislators—as partners in this endeavor. The development of initiatives and collaborations within and across countries underscores a growing commitment to promoting positive outcomes for whole families, and offers increasing opportunities for researchers, policymakers, practitioners, and family members to work together to achieve this goal.

We would like to dedicate this volume to Dr. Mary Seeman, MDCM, FRCPC, DSc, Professor Emerita in the Department of Psychiatry at the University of Toronto. Dr. Seeman is a tireless, committed leader in the field, focusing in particular on gender, psychosis, and the experiences of women and mothers with serious mental illnesses. As a co-editor of each of the three editions of Parental Psychiatric Disorder, Dr. Seeman's contributions have spanned decades (1). She has personally supported many of us in our professional development and research careers. Dr. Seeman's many seminal papers bring attention to the needs of women with schizophrenia and treatment considerations, including the importance of addressing reproductive issues and effective parenting. It is her contention that comprehensive treatment of schizophrenia in women means remembering that all women of childbearing age are potential new mothers, and that women with schizophrenia who are parents benefit from ongoing support (2). In 2013, Dr. Seeman wrote that "useful services for parents with schizophrenia need to bridge the adult/child mental health divide and provide family-centered care with full interagency cooperation" (p. 19), citing references from the early 2000's, and presaging conclusions and recommendations in this current volume, nearly a decade later. Dr. Seeman provided thoughtful reviews for many of the papers in the current volume, for which we are grateful. Dr. Seeman's commitment and contributions inspire us to move forward with this important work.

Author contributions

JN initiated the Research Topic. JN, JP, JR, and AW were topic editors and wrote the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

Acknowledgments

The authors acknowledge the contributions of the many international guest editors and reviewers to this Research Topic.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman MV. *Parental Psychiatric Disorder: Distressed Parents and their Families* (3rd ed.). Cambridge: Cambridge University Press (2015).

2. Seeman MV. Clinical interventions for women with schizophrenia: pregnancy. *Acta Psychiatr Scand.* (2013) 127:12–22. doi: 10.1111/j.1600-0447.2012.01897.x

frontiers
in Psychiatry

ORIGINAL RESEARCH
published: 22 October 2021
doi: 10.3389/fpsyt.2021.741225

Towards a Program Theory for Family-Focused Practice in Adult Mental Health Care Settings: An International Interview Study With Program Leaders

Annette Bauer[1]*, *Stephanie Best*[2], *Juliette Malley*[1], *Hanna Christiansen*[3], *Melinda Goodyear*[4], *Ingrid Zechmeister-Koss*[5] *and Jean Paul*[6]

[1] Care Policy and Evaluation Centre, Department of Health Policy, London School of Economics and Political Science, London, United Kingdom, [2] Australian Institute of Health Innovation, Macquarie University, North Ryde, NWS, Australia, [3] Department of Psychology, Clinical Child and Adolescent Psychology, Philipps University, Marburg, Germany, [4] School of Rural Health, Monash University, Melbourne, VIC, Australia, [5] Austrian Institute for Health Technology Assessment GmbH, Vienna, Austria, [6] Department of Psychiatry, Psychotherapy and Psychosomatics, Division of Psychiatry I, Medical University Innsbruck, Innsbruck, Austria

Objectives: In several high-income countries, family-focused practice programs have been introduced in adult mental health care settings to identify and support children whose parents live with mental health problems. Whilst their common goal is to reduce the impact of parental mental illness on children, the mechanisms by which they improve outcomes in different systems and settings are less well known. This kind of knowledge can importantly contribute to ensuring that practice programs achieve pre-defined impacts.

Methods: The aim of this study was to develop knowledge about relationships between contextual factors, mechanisms and impact that could inform a program theory for developing, implementing, and evaluating family-focused practice. Principles of a realist evaluation approach and complex system thinking were used to conceptualize the design of semi-structured in-depth interviews with individuals who led the implementation of programs. Seventeen individuals from eight countries participated in the study.

Results: Interviewees provided rich accounts of the components that programs should include, contextual factors in which they operated, as well as the behavior changes in practitioners that programs needed to achieve. Together with information from the literature, we developed an initial program theory, which illustrates the interconnectedness between changes that need to co-occur in practitioners, parents, and children, many of which related to a more open communication about parental mental health problems. Stigma, risk-focused and fragmented health systems, and a lack of management commitment were the root causes explaining, for example, why conversations about parents' mental illness did not take place, or not in a way that they could help children. Enabling practitioners to focus on parents' strengths was assumed to trigger changes in knowledge, emotions and behaviors in parents that would subsequently benefit children, by reducing feelings of guilt and improving self-esteem.

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Anneli Silvén Hagström,
Stockholm University, Sweden
Scott Yates,
De Montfort University,
United Kingdom

***Correspondence:**
Annette Bauer
a.bauer@lse.ac.uk

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *14 July 2021*
Accepted: *27 September 2021*
Published: *22 October 2021*

Citation:
Bauer A, Best S, Malley J, Christiansen H, Goodyear M, Zechmeister-Koss I and Paul J (2021) Towards a Program Theory for Family-Focused Practice in Adult Mental Health Care Settings: An International Interview Study With Program Leaders.
Front. Psychiatry 12:741225.
doi: 10.3389/fpsyt.2021.741225

Conclusion: To our knowledge, this is the first research, which synthesizes knowledge about how family-focused practice programs works in a way that it can inform the design, implementation, and evaluation of programs. Stakeholder, who fund, design, implement or evaluate programs should start co-developing and using program theories like the one presented in this paper to strengthen the design and delivery of family-focused practice.

Keywords: program theory, family-focused practice, evidence-supported practice, implementation, evaluation, parental mental health, theory of change

INTRODUCTION

Family-focused practice approaches, which recognize the family as a unit in the treatment of a person's mental health problems, have been developed and introduced in mental health services internationally. They have in common that they seek to combat the limitations of services that are focused only on the individual, and do not consider the impact of mental disorders on other family members, in particular children (1, 2). Examples of policy initiatives or national flagship programs introducing such approaches include: the 'Effective Family Program' in Finland (3), 'Think Family' initiative in the United Kingdom (4), 'Children of Parents with a Mental Illness' (COPMI; https://www.copmi.net.au) and 'Families where a Parent has a Mental illness' (5) in Australia. Governments in Scandinavia even made legal amendments to their health and social care acts, that requires practitioners in adult mental health services to identify and look after the needs of children whose parents are using their services (6–8).

The term family-focused practice (FFP) has been used differently in different contexts, and can refer to (mental) health, social care and other sectors. For the purpose of the paper, and in line with Foster et al. (1), we define FFP as the way, by which mental health practitioners or services respond to the family members of the person in treatment for their mental illness. More specifically, we focus on FFP in adult mental health settings and therefore use the term to refer to how adult mental health practitioners and services respond to children.

Even though attempts to transform adult mental health services to incorporate FFP began decades ago (1), most mental health systems still do not operate in this manner (9–11). Reasons for this are manifold, and include fragmented service systems, inadequate funding to address needs beyond the individual's most urgent problems, lack of organizational commitment and leadership reflected, for example, in a lack of policies or guidelines on identifying parenting status, and limited knowledge or skills among practitioners (12, 13).

Whilst the above-mentioned policies and legal changes seek to address barriers, their success depends on efforts to implement changes in local systems and organizations. Such efforts, to change practice at an organizational and local system level, are the subject of our investigation in this paper. We define these efforts as practice change programs, which are complex interventions that require or demand some form of professional behavior change at an individual or collective level (14). From here onwards, we refer to them simply as programs. Most programs have multiple components, which can include the documentation of parenting status, assessment of family relationships and the children's situation and providing or referring to psychological, -educational, -social interventions to support adults in their parenting role or to support children directly (1, 4, 15). Whilst findings from systematic reviews (16, 17) suggest that psychoeducational and psychological interventions can lead to improved mental health for children, evidence is still largely lacking for such multi-component programs that have been implemented under real-world conditions, and which take place in complex ecological systems (18, 19).

In this study, we wanted to understand how the different components of programs have been implemented, and the mechanisms or processes by which they were expected to lead to changes in outcomes for practitioners, parents and children. The goal of our study was to gather knowledge that could inform the development of an initial program theory for FFP. We sought to surface some of the conscious and subconscious processes of how programs have been developed and implemented by interviewing people who had led the implementation of programs in this field. We expected that this kind of explorative knowledge could inform the development of future frameworks that are theory-driven whilst empirically focused, a gap that has been highlighted by various implementation scientists (20–22).

In our understanding of a program theory, we borrowed from two theoretical frameworks developed or commonly used in the public health field - a realist approach and complex systems thinking. Both approaches suggest that interventions cannot be uncoupled from the systems in which they are operating, and interventions need to be developed and evaluated considering contextual factors (23). In public and mental health, a realist approach has been central in shifting the focus of intervention development and evaluation from whether something works to what works, for whom, how, and in which context (24). Whilst a realist approach proposes the development of a theory by linking contextual factors with mechanisms that are expected to lead to desirable outcomes (25–28), a complex systems perspective offers ways to theorize interventions as disruptions to dynamic and complex systems (29, 30). The latter includes the analysis of an intervention's ability to change relationships between key players that make up such systems, displace entrenched practices and transform or redistribute resources (31).

Whilst the realist synthesis guided both the design and analysis of the study, a complex systems thinking perspective, together with insights from behavior change theories, informed mainly the analysis. Following a realist synthesis, we gathered knowledge to understand what works, for whom, in which context and why. In addition, following both, realist and complex systems thinking perspectives, we also wanted to gather knowledge about the role of actors and resources in influencing the interaction between programs and local systems. Combining these aspects, we set out the following research questions: (1) What is the context in which programs take place, and how is it modified? (2) Which program components can be distinguished? (3) What are the expected program outcomes and for whom? (4) What are the assumed mechanisms leading to expected outcomes? Or, in other words, why and how do programs work? (5) What are the resources employed for the delivery of programs?

METHOD

General Approach

Realist approaches suggest several methods for extracting knowledge to inform the development of initial program theories. Reflecting the current state of the evidence base, we initially sought to apply a dual approach, which would have consisted of an initial synthesis of the literature and then interviews with individuals who led the implementation of programs (27, 32). Exploring the international academic and gray literature on FFP, we found detailed descriptions of programs, rich accounts of how they had been implemented, and the challenges (4, 7–11, 33, 34). However, we only identified limited information about expected changes for parents or children and mechanisms or processes leading to those. None of the papers set out or referred to a program theory or explained the rationale for evaluating changes in practitioners' behaviors, and the mechanisms leading to improved child and parent outcomes, a gap that has been highlighted (35). We therefore did not conduct a synthesis of the papers. Instead, we drew from the literature for a description of possible programs components to guide our interviews with program leaders. We also used the information more informally to guide the conduct and interpretation of the findings from qualitative interviews.

Semi-Structured In-Depth Interviews

We conducted semi-structured in-depth interviews to elicit the perspectives of individuals who had been developing, managing, implementing (and evaluating) FFP programs, and explored their first-hand experiences of driving and implementing practice change in this area.

Sampling

A two-stage purposive sampling process, using snowballing principles, was adopted to identify individuals who were leading the implementation of programs. First, we approached a selected group of researchers in the field of FFP. We first contacted a handful of researchers who had been invited as experts to a workshop on the topic of parental mental illness in Austria called Ideas lab, which had been organized by the funder of this research with the aim to conceptualize new research in this area (36). We asked those researchers to recommend other researchers to us, who they thought would know about programs internationally. When contacting those researchers, we also invited them to recommend other researchers. At the end of this snowballing process, we had a group of twenty researchers, all of whom had expertise in FFP as evidenced by their publication record in this area. Next, we asked them to recommend individuals who had been leading the implementation of FFP programs. We did not set out specific criteria as we wanted to allow for diverse programs, including, for example, those that had evolved more organically. Whilst we originally had set out that programs should refer to adult mental health settings, we allowed for the inclusion of programs that spanned across settings or originated from child mental health and social care settings. This decision was made as it became clear from the feedback we received from researchers that the question in which part of the care system the program started or was anchored depended on national or regional funding structures and arrangements. It also became clear that roles of developing, implementing, evaluating, or advocating for programs were overlapping, and that recommended individuals often had more than one role. Often their role was not a formal program administrator role. We therefore did not specify the role or function of individuals should have. Researchers identified altogether forty individuals, who we then invited to participate in the study. Invited participants were from the following countries: Austria, Australia, Germany, Netherlands, Norway, Sweden, United Kingdom, and the US.

Study Participants and Data Collection Procedure

We conducted interviews with the 17 individuals who agreed to participate, who were from seven countries (none of the invited participants from Sweden responded to our emails). The rest of the potential participants ($n = 23$) did not respond to our emails. Most interviewees were employed by organizations that provided publicly funded adult or child mental health services. A few were – either additionally or solely – employed by universities or charities or worked in private practice. In addition to clinical and therapist roles, part of their job descriptions covered service improvement, implementation management, or research. Interviewees had professional backgrounds in psychiatry, psychology, occupational therapy, social work, or teaching.

Interviews were conducted via Skype or telephone and, in one instance, in person, as requested by the interviewee. One interview involved two persons from the same program. Interviews lasted between 1.5 and 2 hours. In one case the interview had to be ended earlier than planned, after about thirty min, because the interviewee needed to attend to an emergency concerning a family at their practice. Fully informed verbal consent was obtained at the beginning of each interview, and in writing, which participants completed before or after the interview. The study of interviews was reviewed and approved by the Research Ethics Committee of the London School of Economics and Political Science.

The interview schedule was informed by ideas from realistic synthesis. It included questions about how the program components identified by us from the literature work in practice, the kind of resources their implementation involved (e.g., training), and how they were linked to improved outcomes. We included questions and prompts about 'how' and 'why' interviewees thought that outcomes were achieved. This was done to generate knowledge about potential processes and mechanism leading to improved outcomes, and to distinguish between short-, medium- and long-term outcomes. We applied the following order of questions First, we asked interviewees how they had become involved in this area as well as their roles and responsibilities in programs. Next, we asked about their views concerning the key components of programs which we identified from the literature: identifying and documenting parenting status; leading conversations with parents about their parenting and their children; initiating conversations with children about their parent's mental health problems; offering or signposting to interventions and support. We asked them whether they thought some components were more important than others or were more challenging to implement than others. We then asked about the types of outcomes and impacts that they expected from the program, and the processes leading to such outcomes. Finally, interviewees were asked about the context in which the programs took place, the drivers and challenges for change, and the resources and support required to achieve change and overcome challenges. The interview guide is presented in the **Supplementary Material**.

Interview questions were sent in advance to interviewers, to overcome potential language barriers as not everyone was fluent in English. All but four interviews were conducted in English. The four were conducted in German, which was the preferred language for these interviewees, and the mother language of the lead researcher (AB) who conducted the interviews.

Recordings, Translations, and Data Analysis

Audio-recordings were produced for all interviews. Full transcripts of each audio recording were generated and uploaded on NVivo11 software. The coding framework was developed and refined in an iterative process, led by AB and in consultation with members of the research team, with main inputs from a specialist qualitative researcher (JP).The lead researcher (AB) coded the data in NVivo11 following principles of the Framework Method (37), a method that is commonly applied in qualitative health research. JP read a sample of the interviews and provided critical inputs to the development of the coding framework, and into the coding of the data. Main categories of the coding framework reflected the key concepts for developing program theories following a realist synthesis (38): components, context, mechanisms, outcomes, activities, actors, and resources. Data was indexed according to this framework. Within each of the indexed categories, we looked for further themes and created additional (sub-) categories inductively to, allowing, for example, a distinction into practitioners, parent, and child perspectives. Sub-categories were iteratively constructed through conversations between two authors (AB and JP), informed by ideas from behavior change and complex systems theories. Data were then summarized in a matrix by categories using a spreadsheet. For each sub-category a short descriptive summary was generated, which was presented alongside example quotes. In several meetings throughout the study, researchers from the team discussed emerging themes and findings, applying their multi-disciplinary backgrounds in health and social care research to the interpretation of the data.

RESULTS

We present the findings structured by key concepts. This includes a description of the contextual factors that influence the delivery and outcomes of the program (research question 1), the components of programs, including what those should encompass (research question 2), the expected outcomes for practitioners, parents and children (research question 3), and processes leading to these outcomes ('mechanisms') (research question 4). Whilst information about resource inputs (research question 5) are provided under the headings of components and contextual factors, we also summarized them briefly in a separate section. At the end of the section, we present an initial program theory that was developed based on these findings.

Contextual Factors

From interviewees' responses, we identified a range of factors that influenced the successful delivery of programs and outcomes for children. Interviewees described how the stigma, discrimination and social isolation children experienced, often prevented or hindered effective engagement of families with services.

> "The degree to which they [families] were avoidant of mental health services because of (...) shame and stigma is massive." (Interview 8)
> "A lot of the children grow up thinking that they are the only child of a parent with a mental illness (...). A lot of these families are isolated or fragmented or stigmatized." (Interview 9)

Whilst none of the interviewees described a role for programs in changing stigma or awareness at a community level, they emphasized the importance of psychoeducation and helping families to find a language in which they could talk about parental mental illness within the family and to others. (This is described in more detail in the section on mechanisms and outcomes of programs).

Interviewees offered detailed accounts on what had hindered and facilitated practice change at a system and organizational level. They reflected how, traditionally, professional workforce development, education, funding, and performance systems were all focused on the medical treatment of a person's crisis rather than on preventing problems through integrated solutions. Such systems had led to or facilitated certain attitudes, beliefs and behaviors of mental health practitioners, which included them being highly protective of their relationships with 'their patients'. Most interviewees described what they thought were exaggerated fears among practitioners that if they started to ask detailed questions about parenting and children, this would bring up safeguarding issues, which would require involvement of child welfare agencies and ultimately lead to children's removal from home. Some interviewees reported how they had addressed such barriers by providing accurate information to practitioners about the role of child and youth welfare agencies and safeguarding procedures. This included information about the role of those agencies in supporting families to prevent child removal, and about the risk that children would be removed permanently, which was very small. Some interviewees explained how they had organized cross-sector training with practitioners from adult mental health and child and youth welfare agencies in order to reduce misconceptions that practitioners had about each other's roles.

> "So, I have to address that very clearly when talking about this to adult psychiatry personnel that this is not about alerting child protection. That this is the last resort that will be necessary for only a few (...)." (Interview 10)
> "(...) there is a lot of misconceptions about child protection services and their work, but I think just reframing it all and saying we want to come into the family as early as possible because there is this possibility of prevention (...)." (Interview 5)

Interviewees described how drivers for successful program delivery had included policies and legislation that were supportive of prevention- and family-focused practice, in particular if those were accompanied with ring-fenced funding for this population. Interviewees explained how their own persuasion or advocacy efforts needed to take place at many different levels in order for change to happen: from policy makers and commissioners of services to senior managers, and frontline practitioners. They described how they had successfully used stories of lived experiences, research data, and legislation on child rights to get the attention of politicians and commissioners. At an organizational level, interviewees referred to the support that managers needed in order to implement changes and the need for organizational capacity to make changes sustainable. This was particularly challenging in organizations that had weak leadership, and in which managers were not skilled to manage organizational change. They described a diverse range of training and workforce development programs that they had implemented. However, according to interviewees training on its own was not sufficient to achieve change in a context, in which frontline practitioners were burnt out and in which there was high staff turnover.

> "(...) training works a bit but it doesn't really work to change culture. We have to have lots of things. We have to have the service, the development. You have to have some interventions to help. You have to have the combination. So, it's a whole combination that is needed so that you get that kind of light bulb moment." (Interview 11)

Program Components

Routine Questions About Parenting Status and Children

Most interviewees explained that, whilst formally and routinely asking parents about their children, and recording this information should be standard practice in adult mental health services, this was commonly not the case. Instead, this was often left to the discretion of the individual practitioner. Recording data on children in the clinical notes (e.g., how many; what age; where they live) was regarded an important starting point for potential further changes in practice. For example, it could lead to sharing information in meetings where case records were reviewed, and to further signposting to support. Some interviewees believed that introducing routine documentation required performance management systems to check compliance.

> "We know that parental mental illness has consequences [for children], but we need to find them [the children] in order to help them. So, the idea is to get all the services to systematically ask "Do you have children?", and to record that, so we can find the children who need help. That has been the main issue, the first step, because we can't provide any family-focused practice if we don't know if the patient has a family." (Interview 1)

Engaging parents and children, the latter often referred to by the interviewees as "invisible" or "hidden" (terms commonly used in the literature), was described as a major challenge. Therefore, asking the right questions, which could include questions about the wider family network, was regarded as important.

Some interviewees emphasized that practitioners also needed to understand why they were asking those questions, and what they would do with the information.

> "In some cases, some of the government policies say you need to ask about children and to find out in which care they are and find out different things. But sometimes people were asking the question, but they didn't have the knowledge and understanding to interpret the information they got back." (Interview 2)

Conversations With Parents About Impact of Mental Health Problems on Children

Interviewees described how discussions between practitioners and parents about the impact of mental illness on their parenting role was a 'natural' starting point, which could then lead to further conversations about how children were doing, and the impact the parent's mental illness had on them.

> "The first conversation, the conversation with the adults is easier for them [adult mental health practitioners], because (...) they already have a relation with the patient." (Interview 3)

Whilst some interviewees thought that parents were just "waiting for therapists to ask" (Interview 5) about their children, as this was an "existential" part of their identity (Interview 4), others thought that practitioners needed substantial time and efforts to encourage parents to see the benefits of talking with their children about their mental disorder. Some described how motivating the parents to have these discussions could be extremely challenging especially when parents had a limited awareness of their mental illness, which they explained was particularly common among parents with personality disorder. At the same time, interviewees believed that not asking about parenting was potentially harmful, because it reinforced the taboo around the subject.

Interviewees emphasized that conversations needed to follow a strengths-based approach focusing on what the parent was doing well and their needs rather than an assessment of their parenting skills.

A few interviewees also thought that it was important to talk to the wider family as they brought in a different perspective that was not covered by talking to parents or children alone. Since parents with mental disorders often distanced themselves from their wider families, talking to them could help children become less isolated.

Conversations With Children About Parental Mental Health

Interviewees described the opportunities for supporting children through adult mental health services. Whilst interviewees agreed that adult mental health services had an important role in facilitating support for children, they had differing views concerning the nature of such involvement. Most interviewees thought that practitioners should encourage parents to have conversations with their children about the impact of their mental health on them. Some interviewees thought that this could or should include talking to children directly.

> "I do think quite strongly that adult mental health workers should be able to do that [talking to children]. (...) Because children do slip through the net (...)" (Interview 2)
>
> "Because they know the parent's diagnosis and how this is affecting the parent they [adult mental health practitioners] are the key personnel to explain this to the child." (Interview 1)

However, other interviewees expressed concerns about practitioners talking to children as this, in their view, required specialist knowledge, skills and dedicated time. Interviewees mentioned some practical barriers in offering help to children in adult mental health settings, such as the need for parental consent, or that some children did not want to talk to professionals involved in their parent's care, as they were worried that something they would say would then be shared with the parent.

> "We don't want the adult practitioner of the parent to talk also with the children because for the children, it is important they feel they can talk to someone, who is not connected to the parent." (Interview 6)

Supporting Children, Including in Collaboration With Other Services

Interviewees talked about a wide range of interventions and activities that had been implemented as part of programs to support families and children, ranging from psychosocial and -educational support, to peer support, help with school, leisure and fun activities.

> "(...) we came up with [activities] to do with the children... and then, while the children were in class next door, we were educating them [the parents] about child development and about children's experiences of mental illness" (Interview 7)

Whilst some interviewees described informal activities or therapeutic approaches that they had developed themselves in response to what they perceived families needed or wanted (e.g., a fun day, or a support group), others referred to more structured interventions that followed manuals and tools. The latter included genograms for the systematic assessment of social relationships and support needs, evidence-based interventions, such as the Beardslee family intervention (39) and family conferences. Some mentioned a collaboration with researchers in the field, which had informed the development of their support offers and therapeutic methods.

Although some thought there needed to be a specific 'offer' for this population of children and parents to which practitioners could refer directly, others thought that most communities had existing support offers for families and children in place and that those should be better utilized for these families.

Interviewees believed it was important that adult mental health services collaborated with services and agencies in contact with the family such as child welfare agencies, schools, and mental health services. They thought that the responsibility for supporting this group of children needed to be a shared responsibility between various services. This required a system, in which providing information about mental illness and signposting parents to support was the responsibility of all agencies involved with families. They explained that this required the commitment of all agencies and could only be achieved through wider system changes.

Program Mechanisms and Outcomes

Interviewees reported on a wide range of behavior changes in practitioners, parents, and children that programs sought to achieve. The following provides description of those, highlighting the connections between outcomes for practitioners, parents, and children as they became apparent to us during the analysis.

Practitioners

Interviewees described how practitioners needed to feel confident in talking to parents and motivating them to engage in conversations about parenting and children, as their confidence projected on to the parent. To do this, they also needed to believe in the importance and benefits of doing so and required appropriate skills in delivering strengths-based practice and knowledge about parenting and child development. Whilst changing practitioners' knowledge of the impact of parental mental health problems on children was seen as an important first step by some, others reported that most practitioners knew this but thought that this, on its own, did not lead to changes in practice. In addition to the organizational support structures that needed to be in place, practitioners also needed to experience the impact of parents' mental disorder on children's lives, including the positive impact as a result of their own changes.

> "Having information and having knowledge does matter, but what is more important is being able to see the connection between general knowledge and their [parents and children] daily life situations." (Interview 10)

Parents

Interviewees explained that parents needed to understand the importance of talking to their children about their mental health problems as some parents did not think that their mental health problems had an impact on children.

> "What is important is that parents realize that they need support and that their children need support." (Interview 4)

Awareness alone was, however, not always enough, according to interviewees. Parents needed to be willing and able to talk and listen to their children.

> "When children ask questions [about parents' mental health problems] it is important, that parents are prepared and that parents are willing to answer questions." (Interview 12)

Interviewees thought that once families were able to talk openly about parental mental illness, many positive outcomes could be achieved (although they did not further specify which ones, or how they would be achieved), and that this was the change they were focusing on.

> "I do think that helping parents and children and other family members to understand what is happening in the family is one of the most important things." (Interview 13)
> "Making this something we can talk about and not making this a big dark secret (...) making them [the parents] able to talk about the problems in their family that's the behavior change we want to achieve." (Interview 1)

Interviewees explained that by focusing on parents' strengths in their therapy, this would enable them to feel more confident in their parenting skills, and reduce their feelings of shame and guilt, which in turn would improve their mental health symptoms. They described that talking about parenting could lead to improvements in their therapy goals, which in turn changed practitioners' motivation to include family discussions in their therapy.

Children

Whilst interviewees were giving comprehensive and coherent accounts of the changes they expected to occur in practitioners and parents, their accounts of changes in children were more diverse. In their reflections on what and how support to children should be provided, the age of children was a main consideration. Interviewees described how discussions with children, initiated by the parent or the practitioner, needed to be conducted using age-appropriate language, and approaches that were focused on the child, their needs, and what mattered to them.

Interviewees described the importance of helping children to understand parents' mental illness, and to enable them to make sense of what was going on at home. Children were feeling relieved once they had more accurate information about their parents' mental illness because they were better able to understand their parents' behaviors and place it outside themselves.

> "For the children, the main outcome will be to reduce feelings of guilt and shame (...)." (Interview 1)

A few interviewees described how this new understanding had also improved relationships between children and parents.

> "It [talking about parent's mental illness] opened-up a level of trust that had not been there before and it reduced a lot of resentment that had built over the years." (Interview 9)

Some interviewees thought that these changes led to resilience in the long-term. Other long-term outcomes mentioned by interviewees included improved school performance, prevention of child removal, and reduced trauma (associated with child removal). Some interviewees were convinced that positive long-term prevention effects occurred for children but did not offer an explanation about the types of outcomes, and how those were achieved.

> "So, if the parents feel like they are confident and they can do this. They talk to their children about what is going on and it [has] a big prevention effect for the children." (Interview 1)

Not everyone was certain whether long-term outcomes, such as breaking the cycle of poor mental health between family members, was ultimately achievable, but that it was more about providing children with the tools to cope with adversities. This included children's increased ability to ask for help by helping them to find a language to talk about their parent's mental disorder without shame.

> "Obviously we want children, who grow up well, who have resilient lives, and who are able to go on and function well and don't end up with their own mental health issues but (...) [even with support] you could end up with one [mental illness] (...) But [with support] it is more like that - if things go wrong - [the children] are resourceful enough to be able to find, get support and help to work through things." (Interview 7)

One interviewee reflected on the challenges of evidencing long-term outcomes.

> "We are not tracking parents over historic periods, so we are left with relatively short snapshots." (Interview 9)

Resources

As mentioned above, a lack of dedicated resources to FFP was seen as a major barrier towards the adoption of FFP. Resource inputs required to implement the program, included different types of training, ongoing supervision, and various opportunities for knowledge exchange between professionals from different agencies, including child and youth welfare, schools, and primary health care. Interviewees considered the commitment from the organization's senior management essential, but explained how a lack of funding for activities that were not core business (together with a lack of change management or general leadership skills) prevented such commitments. Interviewees also talked about commitments required from insurance companies and local, regional, or national governments. Buy-in from these parties were needed to mobilize the necessary resources. Most interviewees thought that, in addition to workforce development, the introduction of new and consistent policies and procedures, which outlined the expectations towards managers and practitioners, as well as (amendments to) reporting and performance systems to monitor those were needed. In addition, interviewees explained that it required a shared vision and care pathways, which needed to be implemented at a system level.

Initial Program Theory

Based on the findings from the interviews, we developed an initial program theory in the form of a logic model, depicted in **Figure 1**. The logic model illustrates the relationships between resource inputs required to deliver the program components, the contextual factors, which enable or constrain the delivery of the program, and the mechanisms assumed to lead to final long-term outcomes for the child. In the model, we assume that contextual factors are potentially amenable to the programs, and that all or some of them might need to be modified to achieve the desired impact. For example, system and organizational factors, such as stigma, risk-focused and fragmented systems, and lack of management commitment, were assumed to be the root causes of the problem, which impact on practitioners', parents' and children's situations and behaviors, explaining, for example, why they would not have conversations about parents' mental illness. Their knowledge, attitudes, and beliefs, such as those manifested in shame and guilt, present factors at the individual level that need to be addressed by programs.

It is hypothesized that a successful program triggers changes in knowledge, emotions and behavior in practitioners, parents, and children, which are closely interconnected. For example, as practitioners start applying their newly gained knowledge and skills in asking about parenting using a strengths-based approach, they find that parents respond positively, which in turn encourages them to continue with their new practice, and to further enhance their knowledge and skills. As parents are enabled to talk with children about their mental health problems, and learn to listen to the child's needs, children start to develop trust towards their parents, and feel better about themselves. It is expected that this encourages the parent further to talk about their mental illness more openly, both in their relationship with practitioners and their children. More immediate changes in children's feelings and behaviors, such as help-seeking, are then assumed to lead to some longer-term outcomes for children, such as resilience and improved relationships.

Arrows in the **Figure 1** illustrate the spiral effects between mechanisms or short-term outcomes for practitioners, parents, and children, as well as possible feedback loops between them and contextual factors. In line with complex system thinking (40), the logic model shows how programs need to activate a virtuous circle where an initial success creates the conditions for further successes. The non-linear way, in which change may be created, was well illustrated by one interviewee:

> "[The question is] whether you need to change systems before you can change practitioners before you can change outcomes of the family, or whether you can use changes in families to create changes in practitioners as well. And I used to think they are quite linear (…) but I am less [convinced] by it now and I think that changes in a client can create change in their [practitioners] practice and that enables them to put more things in place organizationally as well." (Interview 3)

DISCUSSION

Programs seeking to introduce FFP in adult mental health settings need to be informed by appropriate evidence, which includes evidence about what works in different contexts for different populations, and why it works. This paper contributes to the literature by providing a synthesis of the potential components that constitute FFP programs, and how programs might lead to improvements. To our knowledge, this is the first paper in this field, which synthesizes such knowledge in a way that it can inform the design, implementation, and evaluation of programs. Applying realist and complex system perspectives to our interviews with individuals, who were leading the implementation of programs, allowed us to identify potentially important spiral effects and feedback loops between changes in the behavior of practitioners, parents and children. We were able to identify contextual factors that programs might need to target to trigger such spiral effects. Ultimately, program theories need to be developed for individual programs in collaboration with relevant stakeholders. However, we hope that the knowledge generated in this study provides a useful starting point for such exercises.

Our study was exploratory. Several limitations in our data hindered the development of a more comprehensive and ultimately more robust program theory. A first limitation relates to our main data source, which was a relatively small number of interviews, conducted with interviewees based in a small number of selected countries, all of which were high-income countries. We were not able to reach interviewees from some of the countries in which FFP programs have been implemented, such as Canada, Finland, and Sweden. Whilst selecting a small sample of individuals based on their knowledge and expertise is considered appropriate for the purpose of developing a program theory (41), it might mean that important perspectives from individuals not involved in those networks or movement(s) have been missed. For example, future inquiry is needed to understand whether including a larger number of individuals, including study participants who did not respond to our emails, would validate the initial program theory developed in this study. In addition, we relied in our choice of interviewees on recommendations from expert researchers, and we did not apply clearly defined inclusion criteria to guide their recommendations. It might be that a more refined inclusion of individuals would have led to richer information, such as information about child outcomes. For example, it might be useful to select interviewees by their level of competence and experience in the field, or by certain characteristics of programs they implemented such as size. However, despite this limitation, it was possible to identify commonly held views and common experiences, especially concerning practitioners' attitudes and behaviors, and how those

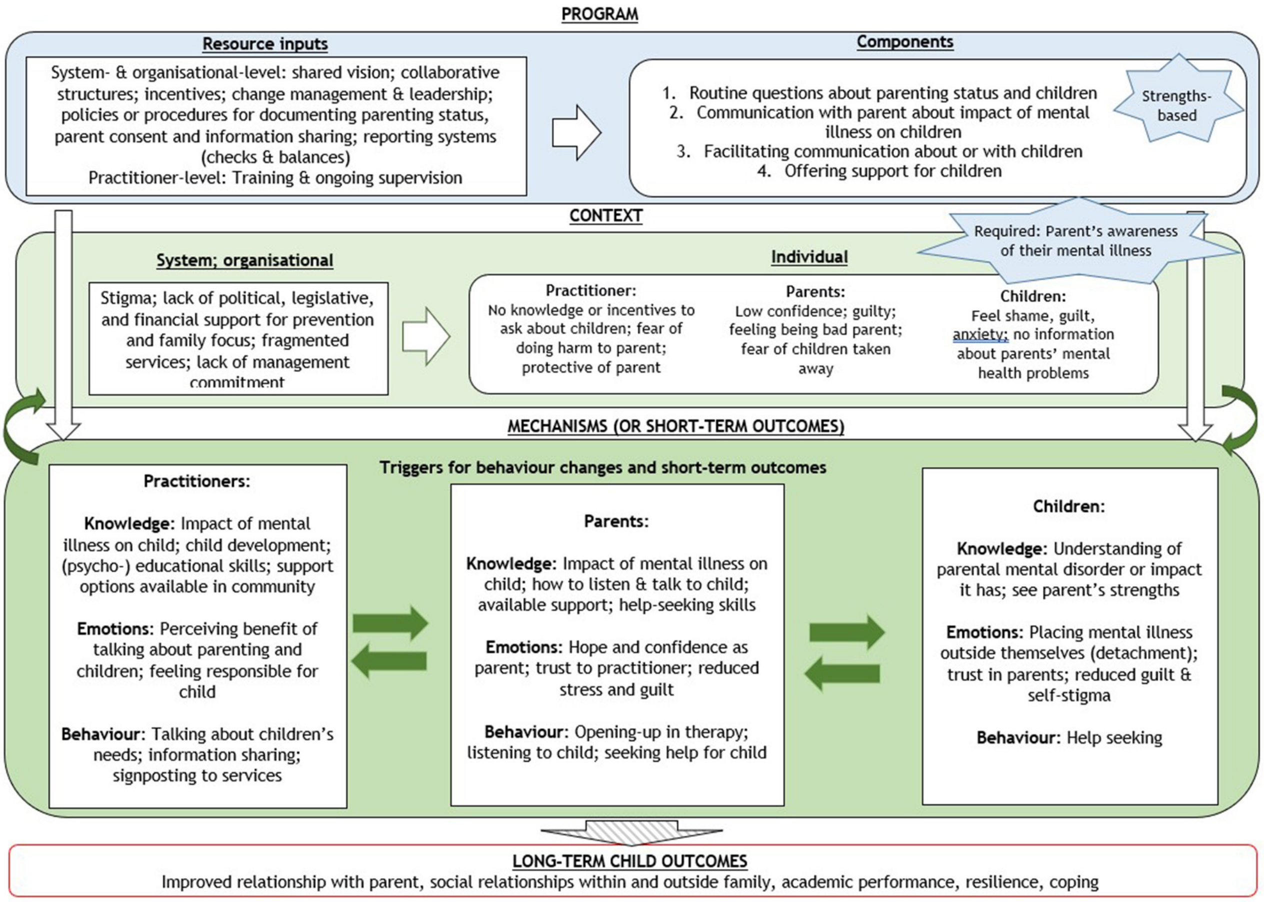


FIGURE 1 | Initial program theory for family-focused practice.

needed to be changed. Whilst it was beyond the remit of this study to include the views of service users' representatives, future research should involve families using services.

Overall, in our data we observed that information was much richer for the short-term outcomes of programs, which is not uncommon in program theories as many interventions only seek to achieve intermediate outcomes (42). However, program theories should be transparent about which outcomes they seek to achieve, how short-term or intermediate outcomes are linked to long-term outcomes (if at all) and highlight evidence gaps. For example, the focus on short-term outcomes might be indicative of an insufficient evidence-base for child-focused practice and of an uncertainty about what kind of outcomes can be expected for children of different age groups (35, 43). It also is possible that, especially in some adult mental health settings, where the focus is naturally on the adult, the final outcomes of FFP are perceived to be about achieving parent's outcomes [potentially alongside children's outcomes). In addition, other outcomes such as those for partners or siblings, might be considered important too. A program theory should make the expectations as to what are viewed as final outcomes clear, and set out the pathways that are supported by evidence and can be realistically assumed to be causal vs. those that are less well established (42). For FFP, future enquiry is needed to assess which types of evidence should be considered when developing the initial program theory further.

The findings from our study also highlight the importance of including the expected relationships between behavior changes in practitioners, parents, and children into program theories, and how those (in combination) influence longer-term child outcomes. For example, the role of trusting, non-judgmental relationships between practitioners and families have been found to lead to improved parents' mental health (35), and good interpersonal relationships between children and their parents have been found to lead to improved child behavior (19). Additional actors might be useful to include, such as individuals managing, funding, or influencing FFP. Integrating theories of behavior change, which describe the dynamic relationships between players at different organizational or system levels, into program theories might be particularly valuable. Methods that support the development of this knowledge, such as actor-based change framework (44), social network analysis (31, 45) and the Capability, Opportunity, Motivation towards Behavior change approach (COM-B) (46) might be particularly useful for developing context-sensitive strategies as part of practice change programs (47).

Another area that program theories should address (but commonly do not) refers to economic evidence. Economic evidence in FFP is largely lacking (35). Whilst we identified cost pressures as a key barrier that prevented change in this area, a finding that is commonly cited in the relevant literature (12), only a few interviewees mentioned the importance of developing an economic case for programs in this area. Program theories, in particular if they include economic evidence, can be an important tool to address accountability demands of funders and tax payers in systems that are under financial pressure (44). They can also be an important tool to help building a collation for change in systems in which many stakeholders from different organizations and sectors are involved, and which require democratic processes to agree on common goals and actions to achieve those.

Different types of program theories may be developed using a range of methodologies, for example supporting the specific purpose of each of the stages of the program development, implementation, and evaluation cycle (40, 47). An initial program theory, such as the one we developed, might play a particular important role during the early development stage, which benefits particularly from theories that consider the interactions of the program with contextual factors (47). Developers might first set out the contextual factors that are most pertinent to the successful delivery of their program, the components they want to focus on as a result, and describe those in detail, together with the resource inputs they require. In the case of FFP this might include discussions about: whether and how mental illness stigma needs to be addressed through the program, whether reporting and performance systems are fit for purpose, how managers might need to be supported to lead change processes. Without such planning, it is possible that programs fail. For example, introducing new staff roles in adult mental health settings to take on additional responsibilities to look after children largely failed in the context of the strongly hierarchical Swedish and Norwegian systems, in which important decisions are traditionally only made by doctors (48).

Actions to prevent child and youth mental health problems are expected to lead to long-lasting improvements in wellbeing, health, and employment (49). Considering that one in four to five children live with parents with mental health problems (50) and that the risk for those children to develop mental health problems is as high as forty per cent (51), use of evidence-based practice in this area is important. Our paper provides a starting point for an increased use of program theories in this important area of practice.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by London School of Economics and Political Science. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

AB was primarily responsible for the study design and conduct and analysis as well as manuscript preparation for this study. JP also informed the study design and supported the conduct and analysis of the study. All authors contributed to the conceptualization of the study, final analysis and preparation of the manuscript, and approved the submitted version.

FUNDING

The research described in this paper was conducted as part of the research project How to raise a village to raise a child, which received funding from the Austrian Federal Ministry of Health, Science and Research through the Open Innovation in Science Centre at the Ludwig Boltzmann Gesellschaft GmbH. The project is hosted by the Medical University of Innsbruck, which provided in kind contributions to the research project. The funders did not influence the collection, analysis, and interpretation of data and played no role in writing the manuscript.

ACKNOWLEDGMENTS

First, we would like to thank Dr Geoff Wong at the Nuffield Department of Primary Care Health Sciences for the training on realist synthesis he provided to the research team and his invaluable advice with regards to the design of the interview guide. Second, we would like to acknowledge the support from individuals, who provided management, supervision, or administrative support to the research project How to raise a village to raise a child as part of which this study was conducted. In particular, we would like to thank Professor Alex Hofer at the Medical University Innsbruck Department of Psychiatry, Psychotherapy and Psychosomatics, Division of Psychiatry, for hosting the research project within his department. Finally, we would also like to thank the individuals who helped with the translation of the interviews: Johanna Krönner, Julia Kapferer, Anna-Elena Pingerra and Elena Seis.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.741225/full#supplementary-material

REFERENCES

1. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child and Youth Services.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
2. Nicholson J, Reupert A, Grant A, Lees R, Maybery D, Mordoch E, et al. The policy context and change for families living with parental mental illness. *Parental Psychiatr Diso*. (2015). 3:354–64. doi: 10.1017/CBO9781107707559.034
3. Solantaus T, Toikka S. The effective family program: preventative services for the children of mentally ill parents in finland. *Int J Mental Health Promot.* (2006) 8:37–44. doi: 10.1080/14623730.2006.9721744
4. Grant A, Lagson S, Devaney J, Davidson G, Duffy J, Perra O, et al. *A study of health and social care professionals' family focused practice with parents who have mental illness, their children and families in Northern Ireland*. Final report. Retrieved from Belfast, Ireland. (2018).
5. Goodyear M, Hill T-L, Allchin B, McCormick F, Hine R, Cuff R, et al. Standards of practice for the adult mental health workforce: meeting the needs of families where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120
6. Afzelius M, Plantin L, Östman M. Families living with parental mental illness and their experiences of family interventions. *J Psychiatr Ment Health Nurs.* (2018) 25:69–77. doi: 10.1111/jpm.12433
7. Lauritzen C, Reedtz C, Van Doesum K, Martinussen M. Factors that may facilitate or hinder a family-focus in the treatment of parents with a mental illness. *J Child Fam Stud.* (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
8. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness: a necessity to provide relevant support. *Front Psychiat.* (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
9. Goodyear M, Maybery D, Reupert A, Allchin R, Fraser C, Fernbacher S, et al. Thinking families: A study of the characteristics of the workforce that delivers family-focussed practice. *Int J Ment Health Nurs.* (2017) 26:238–48. doi: 10.1111/inm.12293
10. Maybery D, Foster K, Goodyear M, Grant A, Tungpunkom P, Skokoy BE, et al. How can we make the psychiatric workforce more family focused? *Parental Psychiatr Diso*. (2015) 3:301–11. doi: 10.1017/CBO9781107707559.029
11. Maybery D, Goodyear M, O'Hanlon B, Cuff R, Reupert A. Profession differences in family focused practice in the adult mental health system. *Fam Process.* (2014) 53:608–17. doi: 10.1111/famp.12082
12. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
13. Shah-Anwar S, Gumley A, Hunter S. Mental health professionals' perspectives of family-focused practice across child and adult mental health settings: a qualitative synthesis. *Child Youth Services.* (2019) 40:383–404. doi: 10.1080/0145935X.2019.1591947
14. Johnson MJ, May CR. Promoting professional behavior change in healthcare: what interventions work, and why? A theory-led overview of systematic reviews. *BMJ Open.* (2015) 5:e008592. doi: 10.1136/bmjopen-2015-008592
15. Liangas G, Falkov A. Use of structured clinical documentation to identify patients' parental concerns and their childrens' wellbeing. *Commun Ment Health J.* (2014) 50:646–55. doi: 10.1007/s10597-013-9684-5
16. Siegenthaler E, Munder T, Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17.e18. doi: 10.1016/j.jaac.2011.10.018
17. Thanhäuser M, Lemmer G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. *Curr Opin Psychiat*. (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342
18. Nicholson J. Guest editorial. *Aust e-J Adv Mental Health.* (2009) 8:222–6. doi: 10.5172/jamh.8.3.222
19. Solantaus T, Paavonen EJ, Toikka S, Punamäki R-L. Preventive interventions in families with parental depression: children's psychosocial symptoms and prosocial behavior. *Eur Child Adolesc Psychiat.* (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
20. Greenhalgh T, Papoutsi C. Studying complexity in health services research: desperately seeking an overdue paradigm shift. *BMC Med.* (2018) 16:95. doi: 10.1186/s12916-018-1089-4
21. Lawless A, Baum F, Delany-Crowe T, MacDougall C, Williams C, McDermott D, et al. Developing a framework for a program theory-based approach to evaluating policy processes and outcomes: health in all policies in South Australia. *Int J Health Policy Manag.* (2018) 7:510–21. doi: 10.15171/ijhpm.2017.121
22. Reed JE, Howe C, Doyle C, Bell D. Simple rules for evidence translation in complex systems: a qualitative study. *BMC Med.* (2018) 16:92. doi: 10.1186/s12916-018-1076-9
23. Craig P, Di Ruggiero E, Frohlich K, Mykhalovskiy E, White M, on behalf of the Canadian Institutes of Health Research (CIHR) National Institute for Health Research (NIHR) Context Guidance Authors Group. *Taking account of context in population health intervention research: guidance for producers, users and funders of research.* Retrieved from Southampton: NIHR Evaluation, Trials and Studies Coordinating Centre. (2018). doi: 10.3310/CIHR-NIHR-01
24. Duncan C, Weich S, Fenton S-J, Twigg L, Moon G, Madan J, et al. A realist approach to the evaluation of complex mental health interventions. *The British Journal of Psychiatry.* (2018) 213:451–3. doi: 10.1192/bjp.2018.96
25. Blamey A, Mackenzie M. Theories of change and realistic evaluation: peas in a pod or apples and oranges? *Evaluation.* (2007) 13:439–55. doi: 10.1177/1356389007082129
26. Byng R, Norman I, Redfern S. Using realistic evaluation to evaluate a practice-level intervention to improve primary healthcare for patients with long-term mental illness. *Evaluation.* (2005) 11:69–93. doi: 10.1177/1356389005053198
27. Jackson SF, Kolla G. A new realistic evaluation analysis method: linked coding of context, mechanism, and outcome relationships. *Am J Evaluat.* (2012) 33:339–49. doi: 10.1177/1098214012440030
28. Pawson R. Nothing as practical as a good theory. *Evaluation.* (2003) 9:471–90. doi: 10.1177/1356389003094007
29. Moore GF, Evans RE. What theory, for whom and in which context? Reflections on the application of theory in the development and evaluation of complex population health interventions. *SSM - Population Health.* (2017) 3:132–5. doi: 10.1016/j.ssmph.2016.12.005
30. Moore GF, Evans RE, Hawkins J, Littlecott H, Melendez-Torres GJ, Bonell C, et al. From complex social interventions to interventions in complex social systems: Future directions and unresolved questions for intervention development and evaluation. *Evaluation.* (2018) 25:23–45. doi: 10.1177/1356389018803219
31. Hawe P, Shiell A, Riley T. Theorising interventions as events in systems. *Am J Community Psychol.* (2009) 43:267–76. doi: 10.1007/s10464-009-9229-9
32. Mukumbang FC, van Belle S, Marchal B, van Wyk B. Towards developing an initial program theory: program designers and managers assumptions on the antiretroviral treatment adherence club program in primary health care facilities in the metropolitan area of western cape province, South Africa. *PLoS ONE.* (2016) 11:e0161790–e0161790. doi: 10.1371/journal.pone.0161790
33. Korhonen T, Vehviläinen-Julkunen K, Pietilä AM. Implementing child-focused family nursing into routine adult psychiatric practice: hindering factors evaluated by nurses. *J Clin Nurs.* (2008) 17:499–508. doi: 10.1111/j.1365-2702.2007.02008.x
34. Tchernegovski P, Reupert A, Maybery D. "Let's Talk about Children": A pilot evaluation of an e-learning resource for mental health clinicians. *Clin Psychol.* (2015) 19:49–58. doi: 10.1111/cp.12050
35. Bee P, Bower P, Byford S, Churchill R, Calam R, Stallard P, et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. *Health Technol Assessment.* (2014) 18:1–250. doi: 10.3310/hta18080
36. Ludwig Boltzmann, Gesellschaft (2021). Ideas Lab. Available online at: https://ois.lbg.ac.at/en/projects/ideas-lab. (accessed September 13, 2021)
37. Ritchie J, Lewis J. *Qualitative Research Practice: A Guide for Social Science Students and Researchers*. London: Sage. (2003).
38. Pawson R, Tilley N. Realistic Evaluation (Vol. 2). London: SAGE Publications. (1997).
39. Beardslee WR, Gladstone TR, Wright EJ, Cooper AB. A family-based approach to the prevention of depressive symptoms in children at

risk: evidence of parental and child change. *Pediatrics.* (2003) 112:e119–131. doi: 10.1542/peds.112.2.e119

40. Rogers PJ. Using program theory to evaluate complicated and complex aspects of interventions. *Evaluation.* (2008) 14:29–48. doi: 10.1177/1356389007084674
41. Manzano A. The craft of interviewing in realist evaluation. *Evaluation.* (2016) 22:342–60. doi: 10.1177/1356389016638615
42. Wight D, Wimbush E, Jepson R, Doi L. Six steps in quality intervention development (6SQuID). *J Epidemiol Commun Health.* (2015) 70:520–5. doi: 10.1136/jech-2015-205952
43. Caldwell DM, Davies SR, Hetrick SE, Palmer JC, Caro P, López-López JA, et al. School-based interventions to prevent anxiety and depression in children and young people: a systematic review and network meta-analysis. *Lancet Psychiat.* (2019) 6:1011–20. doi: 10.1016/S2215-0366(19)30403-1
44. Koleros A, Mulkerne S, Oldenbeuving M, Stein D. The actor-based change framework: a pragmatic approach to developing program theory for interventions in complex systems. *Am J Eval.* (2018) 41:34–53. doi: 10.1177/1098214018786462
45. Shelton RC, Lee M, Brotzman LE, Crookes DM, Jandorf L, Erwin D, et al. Use of social network analysis in the development, dissemination, implementation, and sustainability of health behavior interventions for adults: a systematic review. *Soc Sci Med.* (2019) 220:81–101. doi: 10.1016/j.socscimed.2018.10.013
46. Michie S, van Stralen MM, West R. The behavior change wheel: a new method for characterising and designing behavior change interventions. *Implement Sci.* (2011) 6:42. doi: 10.1186/1748-5908-6-42
47. Mills T, Lawton R, Sheard L. Advancing complexity science in healthcare research: the logic of logic models. *BMC Med Res Methodol.* (2019) 19:55. doi: 10.1186/s12874-019-0701-4
48. Östman M, Afzelius M. Children's representatives in psychiatric services: what is the outcome? *Int J Soc Psychiat.* (2009) 57:144–52. doi: 10.1177/0020764008100605
49. Knapp M, Evans-Lacko S. Health economics. *In Rutter's Child and Adolescent Psychiatry*. (2015). p. 227–238. doi: 10.1002/9781118381953.ch18
50. Abel KM, Hope H, Swift E, Parisi R, Ashcroft DM, Kosidou K, et al. Prevalence of maternal mental illness among children and adolescents in the UK between 2005 and 2017: a national retrospective cohort analysis. *Lancet Public Health.* (2019) 4:e291–300. doi: 10.1016/S2468-2667(19)30059-3
51. Foster K. One-third of children of parents with severe mental illness are at risk of developing severe mental illness. *Evid Based Mental Health.* (2014) 17:73–73. doi: 10.1136/eb-2014-101807

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

BRIEF RESEARCH REPORT
published: 25 October 2021
doi: 10.3389/fpsyt.2021.762041

Integrated Care for Pregnant Women and Parents With Methamphetamine-Related Mental Disorders

Johannes Petzold[1,2*†], *Maik Spreer*[1], *Maria Krüger*[1], *Cathrin Sauer*[1], *Tobias Kirchner*[1], *Susanna Hahn*[1], *Ulrich S. Zimmermann*[1,3] *and Maximilian Pilhatsch*[1,4†]

[1] *Department of Psychiatry and Psychotherapy, Carl Gustav Carus University Hospital, Technische Universität Dresden, Dresden, Germany,* [2] *Department of Psychiatry and Biobehavioral Sciences, University of California, Los Angeles, Los Angeles, CA, United States,* [3] *Department of Addiction Medicine and Psychotherapy, kbo-Isar-Amper-Klinikum München-Ost, Haar, Germany,* [4] *Department of Psychiatry and Psychotherapy, Elblandklinikum Radebeul, Radebeul, Germany*

Background: Methamphetamine use is a rapidly increasing cause of morbidity and mortality. Pregnant women and new parents who consume methamphetamine are at high risk since they seldom seek health services despite having multiple needs. We addressed this care gap by implementing an easily accessible program that pools resources from psychiatric, obstetric, and pediatric departments as well as community and government agencies.

Method: This real-life observational study evaluated an integrated care program in 27 expecting parents and 57 parents of minors. The outcome criteria were treatment retention, psychosocial functioning, and abstinence. We compared participant demographics according to outcome and applied ordinal logistic regression to predict treatment success.

Results: Patients received integrated care for almost 7 months on average. Nearly half achieved stable abstinence and functional recovery. Only one pregnant woman dropped out before a care plan could be implemented, and all women who gave birth during treatment completed it successfully. Three-fourths of patients had psychiatric comorbidities. Patients with depressive disorders were almost 5 times less likely to succeed with treatment. Attention-deficit hyperactivity disorder (ADHD) was diagnosed in nearly 30% of patients who dropped out of a care plan, which was about 4 times more often than in the successful outcome group.

Conclusion: Our program engaged pregnant women and parents in treatment and helped them recover from methamphetamine-related mental disorders. Management of comorbid ADHD and depression should be an integral part of care initiatives to counter the methamphetamine crisis that affects parents and children across the globe.

Keywords: methamphetamine use disorder, drug dependence, addiction, ADHD, depression, pregnancy, multimodal therapy, outcome prediction

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
Michael Klein,
Catholic University of Applied
Sciences of North Rhine –
Westphalia, Germany
Ingrid Zechmeister-Koss,
Austrian Institute for Health
Technology Assessment
GmbH, Austria
Rebecca Kate Allchin,
Eastern Health, Australia

Correspondence:
Johannes Petzold
johannes.petzold@
uniklinikum-dresden.de

†ORCID:
Johannes Petzold
orcid.org/0000-0003-4163-9014
Maximilian Pilhatsch
orcid.org/0000-0003-4323-3309

Specialty section:
This article was submitted to
Addictive Disorders,
a section of the journal
Frontiers in Psychiatry

Received: *20 August 2021*
Accepted: *27 September 2021*
Published: *25 October 2021*

Citation:
Petzold J, Spreer M, Krüger M,
Sauer C, Kirchner T, Hahn S,
Zimmermann US and Pilhatsch M
(2021) Integrated Care for Pregnant
Women and Parents With
Methamphetamine-Related Mental
Disorders.
Front. Psychiatry 12:762041.
doi: 10.3389/fpsyt.2021.762041

INTRODUCTION

Methamphetamine use continues to rise on a global scale (1), and methamphetamine-involved overdoses claim lives in staggering numbers (2). This humanitarian crisis is further fueled by maternal, fetal, and child deaths related to methamphetamine exposure (3–6). Moreover, ample evidence implicates methamphetamine in lasting health and psychosocial problems that severely affect parents and their children (3–5). Women who use methamphetamine are burdened by mental disorders in the perinatal period (5), and parents who use methamphetamine are overstrained by their caregiving roles in often adverse living conditions (7). Infants can experience serious damage, such as microcephaly, due to intrauterine methamphetamine exposure (5, 8). Child development is compromised as a result of these factors, as evidenced by lower IQ scores as well as internalizing (e.g., depression) and externalizing symptoms (e.g., aggression) (5, 7, 8). Despite the urgency for prevention and intervention, the uptake of antenatal, pediatric, and mental health services is low and too late in this population (5, 9, 10). Moreover, pregnant women who use methamphetamine leave substance use treatment against professional advice even more often than pregnant women who use other illicit drugs or alcohol (11).

The stark contrast between care needs and utilization highlights the importance of developing programs that are easily accessible and appealing to those impacted by methamphetamine. Pregnancy and parenthood can create a strong motivation for abstinence (7), but treatment may not be accessed due to fear of stigma and punitive measures (5, 12, 13). Thus, transparency and the commitment to keeping families together are paramount. To reach women of childbearing age, expecting parents, and parents of minors, different avenues within the healthcare system, social services, and the community should be used. Linking these avenues also promises to help families achieve lasting health benefits as continuity of care is instrumental in sustaining abstinence (14, 15).

We are aware of only one study that at least partly addressed this care gap. This study compared methamphetamine-specific psychoeducation with a program also covering relationship and parenting skills tailored to South-African pregnant women (12). Both group interventions reduced methamphetamine use and risky sexual behavior, and 92% of participants completed the 4-session comprising interventions (12). Only 14% had been in substance use treatment before, indicating that access to care can be improved substantially (12).

Acknowledging the unmet needs of families affected by substance use disorders, the concept "Mama denk an mich" (MAMADAM, "Mommy think of me") was developed to deliver coordinated care across disciplines and settings. After having demonstrated the feasibility of MAMADAM (9, 16), we here present its potential to improve the mental health of pregnant women and parents who use methamphetamine. The findings on outcome prediction provide critical information for patient assessment and program optimization. With this report, we hope to encourage the implementation and study of similar initiatives to promote the well-being of parents and children impacted by methamphetamine.

METHOD

This is a real-life observational study of integrated care for expecting and new parents with methamphetamine-related psychiatric disorders, which was approved by the ethics committee at the Carl Gustav Carus Faculty of Medicine at the Technische Universität Dresden, Germany.

Care Model

MAMADAM is a family-centered concept that pools resources from psychiatric, obstetric, and pediatric departments as well as local drug counseling and child welfare services. The concept and its elements draw on the available evidence and best practice for the management of methamphetamine-related disorders (8). Services provided through MAMADAM are easily accessible and flexible, ranging from health information for women of childbearing age to comprehensive support for families across care sectors. Shared decision-making and the participation of patients in multidisciplinary meetings ensure that treatment is matched to their preferences and needs. Other engagement strategies are calling patients who missed appointments and offering provider continuity whenever feasible. The following outlines the parts that are directed to the mental health of expecting and new parents. The coordination and specifics of MAMADAM, including information on obstetric and pediatric services, are described elsewhere (9, 16).

All expecting and new parents who use methamphetamine and present to our psychiatric department are considered for enrollment in MAMADAM. Access is facilitated by referrals from healthcare providers and by psychiatric consultations at the obstetric and pediatric departments. Motivational interviewing is used to develop personalized care plans after assessing mental illness and methamphetamine-related medical sequelae. Patients are seen by psychiatrists and psychotherapists on an outpatient basis from several times a week to once a month as needed. Inpatient and day treatments are provided whenever necessary.

An individual session introduces a methamphetamine-specific relapse prevention program that combines aspects of psychoeducation, motivational interviewing, and cognitive behavior therapy (17, 18). Psychotherapists deliver this program in 15 sessions of 50 min to a maximum of 5 patients. Individual psychotherapy and other group therapies are also available (e.g., exercise classes, social skills training, psychoeducation for major depression).

Social workers and occupational therapists help patients enhance their functioning and well-being in areas such as work, housing, and childcare. They partner with local and government agencies to furnish services ranging from community connections to intensive home support. Home care includes random drug screening, but most patients are called into the clinic once in 6 days on average. Urine is collected

---

Abbreviations: ADHD, attention-deficit hyperactivity disorder; MAMADAM, "Mama denk an mich," "Mommy think of me".

under direct observation followed by temperature measurement to minimize manipulation.

Analytic Strategy

We studied a naturalistic sample of patients with methamphetamine-related disorders who received psychiatric care within MAMADAM since its start in 2016 and left or completed treatment before September 13, 2019. Outcome was classified as early dropout (before implementation of a care plan), partial completion of the program (late dropout), and successful completion. Successful completion was defined as a mutually agreed program discharge, which required continuous abstinence, stable housing, financial security, psychosocial functioning, and a support system. Psychosocial functioning required patients to perform daily activities in ways that were gratifying to them while meeting the demands of their dependents (e.g., safe environment, loving relationship) and the community (e.g., engagement in employment). Support usually involved primary care physicians, private psychiatrists, drug counseling centers, and child welfare services.

We used SPSS 27 (IBM, Armonk, NY, USA) and a significance level of 0.05 (two-tailed) for all analyses. Statistics were performed on complete data from all patients unless stated otherwise. We compared participant demographics and the duration of psychiatric care within MAMADAM according to outcome, using Pearson's chi-square-test, Fisher's exact-test, and Bonferroni-adjusted pairwise comparisons for categorical variables. Histograms, normal quantile-quantile plots, and normality tests (Kolmogorov-Smirnov, Shapiro-Wilk) determined the tests for continuous variables (one-way independent ANOVA, Kruskal-Wallis, Mann-Whitney). To identify predictors of outcome, we built a base model with all participant demographics that had complete data and met the assumptions of ordinal logistic regression. We then progressively removed non-significant variables to produce a parsimonious model. Variables that differed considerably between outcome groups and significant predictors were tested for associations, reporting the phi or Spearman's coefficient. These associations were not corrected for multiple comparisons.

RESULTS

We studied 84 patients with methamphetamine-related mental disorders (1 × F15.0, 8 × F15.1, 74 × F15.2, 1 × F19.2; diagnosed according to the International Classification of Diseases, 10th revision). This sample comprised 27 expecting parents and 57 parents of minors (mean age of child ± SD, min-max: 19.02 ± 30.59, 1–144 months; newborns counted as 1 month). Sixteen patients (19.0%) dropped out before receiving a care plan, 27 (32.1%) completed part of the program, and 41 (48.8%) transitioned successfully to community care. Average program participation was over 6 months (mean ± SD, min-max: 202.49 ± 167.42, 0–793 days) and not statistically different between patients who partially and those who successfully completed treatment ($n = 68$, $U = 655.00$, $z = 1.27$, $p = 0.203$).

Table 1 displays demographics according to outcome, showing no statistical differences in sex, age, years of methamphetamine use, and prior addiction rehab. The proportion of pregnant women was significantly lower in the early than in the late dropout group, and all women who gave birth during treatment completed it successfully. Three-fourths of patients had psychiatric comorbidities. Groups were comparably affected except for attention-deficit hyperactivity disorder (ADHD). Although there was only a trend to an overall significant difference between groups, ADHD was significantly less common in patients successfully than in those partially completing the program.

When accounting for the order of outcomes (worst to best: early dropout, partial completion, successful completion), depression, substance use comorbidity, and prior addiction rehab emerged as significant predictors. Patients with substance use comorbidities were more likely to have a better outcome, whereas the opposite applied to patients with depressive disorders and patients with prior addiction rehab. **Table 2** lists the unique (net) contribution of each significant predictor to variations in outcomes (controlled for the other significant predictors).

None of the outcome predictors or variables with considerable group differences were significantly related to one another (see **Table 3**). Yet, there was one trend-level significant association with 59.3% of expecting parents compared with 36.8% of parents having prior addiction rehab. Of note, expecting parents, patients with prior addiction rehab, and patients with ADHD featured a significantly longer use of methamphetamine. Regular methamphetamine use was also numerically longer for patients who dropped out compared with those who completed MAMADAM successfully.

DISCUSSION

This study evaluated the real-world adherence to integrated care by pregnant women and parents with methamphetamine-related mental disorders. Despite being challenged by their roles and psychiatric comorbidities, nearly half of our patients completed treatment successfully, and only one pregnant woman dropped out before receiving a care plan. These data support the notion that pregnancy and parenthood can be motivators for abstinence (7, 8, 19). We are not aware of any study that evaluated a comparable concept for this population, but methamphetamine use is generally associated with high dropout rates (8, 19). Less than one-fourth of patients with methamphetamine as their primary drug completed 180 days of outpatient drug treatment, a duration previously identified as necessary for treatment success (20). Moreover, pregnant women using methamphetamine left substance use treatment against professional advice even more often than those using other illicit drugs or alcohol (11).

Three-fourths of our patients had at least one other mental disorder, which reflects the high psychiatric morbidity reported in association with methamphetamine use (8, 19). Patients with depressive disorders were almost 5 times more likely to have less treatment success than patients without such a diagnosis. This aligns with an outpatient treatment study for methamphetamine dependence in which higher baseline depression predicted methamphetamine use before discharge

TABLE 1 | Participant demographics.

	Early dropout ($n = 16$)	**Partial completion ($n = 27$)**	**Successful completion ($n = 41$)**	**Group differences**
Sex				$X^2_{(2)} = 0.921$, $p = 0.696^{F}$
Women	13 (81.3)	23 (85.2)	37 (90.2)	
Men	3 (18.8)	4 (14.8)	4 (9.8)	
Age	31.63 ± 5.25 (21–38)	28.89 ± 5.02 (18–38)	28.51 ± 6.19 (18–41)	$F_{(2,81)} = 1.809$, $p = 0.170^{A}$
Expecting parents[#]	1 $(6.3)_{a}$	12 $(44.4)_{b}$	14 $(34.1)_{a,b}$	$X^2_{(2)} = 6.867$, $p = 0.032^{C*}$
Pregnant women	1 $(7.7)_{a}$	11 $(47.8)_{b}$	14 $(37.8)_{a,b}$	$n = 73$ women, $X^2_{(2)} = 5.995$, $p = 0.048^{F*}$
Becoming a parent during treatment	0 $(0.0)_{a,b}$	0 $(0.0)_{b}$	6 $(42.9)_{a}$	$n = 27$ expecting parents[#], $X^2_{(2)} = 7.163$, $p = 0.023^{F*}$
	0 $(0.0)_{a,b}$	0 $(0.0)_{b}$	6 $(42.9)_{a}$	$n = 26$ pregnant women, $X^2_{(2)} = 6.686$, $p = 0.026^{F*}$
Years of regular methamphetamine use	6.43 ± 6.27 (0–23)	7.62 ± 6.34 (0–24)	6.11 ± 5.92 (0–25)	$n = 76$, $H_{(2)} = 1.122$, $p = 0.571^{K}$
Prior addiction rehab	9 (56.3)	13 (48.1)	15 (36.6)	$X^2_{(2)} = 2.077$, $p = 0.354^{C}$
Current psychiatric comorbidity				
Due to substance use (2 × F10.1, 8 × F10.2, 1 × F11.2, 3 × F12.1, 35 × F12.2, 4 × F19.2)	6 (37.5)	14 (51.9)	26 (63.4)	$X^2_{(2)} = 3.256$, $p = 0.196^{C}$
Depressive disorder (1 × F32.0, 3 × F32.1, 1 × F33, 1 × F33.4, 1 × F33.8)	2 (12.5)	4 (14.8)	1 (2.4)	$X^2_{(2)} = 3.713$, $p = 0.151^{F}$
Personality disorder (2 × F60.30, 7 × F60.31, 3 × F60.8)	4 (25.0)	3 (11.1)	5 (12.2)	$X^2_{(2)} = 1.869$, $p = 0.465^{F}$
Attention-deficit hyperactivity disorder (F90.0)	3 $(18.8)_{a,b}$	8 $(29.6)_{b}$	3 $(7.3)_{a}$	$X^2_{(2)} = 5.897$, $p = 0.059^{F}$
Any (the above plus 1 × F40.1, 2 × F43.2, 1 × F55.2, 1 × F63, 1 × F63.0, 2 × F63.8, 3 × F70.0, 1 × F70.8, 1 × F91.1, 1 × F91.3, 1 × Q86.0)	11 (68.8)	20 (74.1)	32 (78.0)	$X^2_{(2)} = 0.549$, $p = 0.757^{F}$
Any except due to substance use	6 (37.5)	14 (51.9)	14 (34.1)	$X^2_{(2)} = 2.191$, $p = 0.334^{C}$

Mental illness was diagnosed according to the International Classification of Diseases, 10th revision. Any psychiatric comorbidity comprised social phobias, adjustment disorders, abuse of laxatives, habit and impulse disorders, mild intellectual disability, conduct disorders, and fetal alcohol syndrome. Statistics are based on complete data from the entire sample (N = 84) unless stated otherwise. Data are number of patients (percentage within outcome category) or group mean ± SD (min-max). Percentages with the same subscript do not significantly differ from each other (Bonferroni adjusted).

A, one-way independent ANOVA; C, Pearson's chi-square test; F, Fisher's exact-test; K, Kruskal-Wallis test.

[#] *Twenty-six pregnant women + one expecting father.*

**Statistically significant.*

and poorer treatment attendance (21). Although not predicted by baseline depression scores, methamphetamine use 3 years post-treatment was associated with major depression at follow-up (21). Together, these findings emphasize the need for routine assessment and treatment of depression. Exercise and psychological interventions can reduce depressive symptoms, but methamphetamine-induced cognitive deficits complicate the latter (8, 19). Medications including antidepressants have largely failed and their potential for drug interaction effects has been highlighted (8, 19). Yet, recent research suggests that glutamatergic modulators with antidepressant and pro-cognitive properties may prove effective in treating methamphetamine use disorder and comorbid depression (22).

ADHD was diagnosed in almost one-third of patients who dropped out of an established care plan. This proportion was about 4 times as high as in the successful outcome group. Moreover, ADHD was associated with more years of regular methamphetamine use, which was also numerically longer for patients who dropped out. Routine assessment for ADHD seems therefore imperative, not least because some patients report using methamphetamine to treat ADHD symptoms (8). The risk of misuse and drug interactions warrants close monitoring of pharmacotherapy, with long-acting formulations recommended if stimulant medication is prescribed (19).

A diagnosis of substance use comorbidity predicted a better outcome, which indicates that the benefits of MAMADAM extend to other drugs. Experiencing a substance use comorbidity might increase readiness for change and treatment. Patients with prior addiction rehab were, in comparison, more likely to have a worse outcome. Similarly, previous drug treatment

TABLE 2 | Ordinal logistic regression model.

	Parameter	*B* (SE)	OR	*p*
Threshold	Early dropout to partial completion	−0.16 (0.77)		
	Partial to successful completion	1.50 (0.79)		
Prior addiction rehab	No (vs. yes)	0.92 (0.45)	2.52	0.039*
Current substance use comorbidity		−1.14 (0.45)	0.32	0.012*
Current depressive disorder		1.58 (0.78)	4.87	0.041*

This model was built from a base model by progressively removing the non-significant variables (first to last: current personality disorder, sex, expecting parents, current attention-deficit hyperactivity disorder, age). N = 84 with complete data on all variables. $X^2_{(3)} = 11.452$, $p = 0.010$, *Nagelkerke pseudo* $R^2 = 14.6\%$.
**Statistically significant.*

TABLE 3 | Associations between correlates of outcome.

	Expecting parents	Prior addiction rehab	Current substance use comorbidity	Current depressive disorder	Current attention-deficit hyperactivity disorder	Years of regular methamphetamine use
Expecting parents		$r_\phi = 0.211$ $p = 0.053^C$	$r_\phi = 0.165$ $p = 0.131^C$	$r_\phi = -0.208$ $p = 0.091^F$	$r_\phi = 0.034$ $p = 1.000^F$	$r_S = 0.258$ $p = 0.024$*
Prior addiction rehab			$r_\phi = 0.180$ $p = 0.099^C$	$r_\phi = -0.007$ $p = 1.000^F$	$r_\phi = 0.054$ $p = 0.623^C$	$r_S = 0.372$ $p = 0.001$*
Current substance use comorbidity				$r_\phi = 0.101$ $p = 0.449^F$	$r_\phi = -0.043$ $p = 0.695^C$	$r_S = 0.157$ $p = 0.175$
Current depressive disorder					$r_\phi = 0.096$ $p = 0.595^F$	$r_S = -0.066$ $p = 0.572$
Current attention-deficit hyperactivity disorder						$r_S = 0.285$ $p = 0.013$*

Categorical variables (expecting parents, prior addiction rehab, comorbidities) are coded as 0 = no and 1 = yes. Pearson's chi-square (C) and Fisher's exact-tests (F) are based on complete data from the entire sample (N = 84). Spearman's correlations (r_S) *are based on n = 76 due to missing data on years of methamphetamine use.*
**Statistically significant.*

predicted treatment attrition in pregnant women using methamphetamine weekly or more (11). Prior treatment could indicate greater disease severity or longer disease duration, as patients with prior addiction rehab featured more years of regular methamphetamine use. Of note, more years of regular methamphetamine use predicted poor outcome in a study of the methamphetamine-specific group psychotherapy that we provide in MAMADAM (18). The data collectively highlight that patients with previous drug treatment require close monitoring and greater support. Recognizing the value of their treatment experiences may be a way to better meet their needs and expectations, which should improve retention in care and health outcomes.

Strengths and Limitations

To our knowledge, this is the first work evaluating the adherence to integrated care by pregnant women and parents with methamphetamine-related mental disorders. We did not include a control condition but the exploratory study of a naturalistic sample in the real world can provide outcome predictors of direct importance for patient care. Of note, we report significant predictors from a parsimonious regression model that did not control for non-significant variables and only considered variables with complete data. ADHD was not among the significant predictors in our regression model yet more prevalent in patients who dropped out, with the late dropout group statistically differing from the successful outcome group. We believe that ADHD and other comorbidities, such as depressive and personality disorders, are underreported for patients dropping out of MAMADAM early since these diagnoses require thorough assessments in drug-free intervals. Lastly, we did not collect follow-up data, but treatment success required stable abstinence and continuing care, which are critical factors for long-term recovery.

CONCLUSION

Integrated care is a promising strategy for pregnant women and parents with methamphetamine-related mental disorders. Pregnancy and parenthood provide opportunities to motivate change and engage a population that hardly accesses treatment despite the high psychiatric morbidity. Comorbid ADHD and depression warrant close monitoring as they jeopardize treatment engagement and success. Given the little information on the management of these conditions in the context of methamphetamine use, research is imperative to provide evidence-based interventions. Moreover, integrated care concepts should be disseminated to counter the increasing methamphetamine crisis that affects parents and children across the globe.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee at the Carl Gustav Carus Faculty of Medicine at the Technische Universität Dresden, Germany. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

UZ, MP, MS, and SH contributed to the development of the care concept. MS, SH, TK, UZ, and MP were members of the care team. MP, UZ, CS, and MK designed the study. MP and JP obtained funding. MK, CS, MS, MP, and TK collected the data. JP analyzed and interpreted the data and wrote the manuscript. All authors contributed to manuscript revision, as well as read and approved the final version.

FUNDING

This work was supported by the German Research Foundation [TRR 265 (23)]. The article processing charge was paid from the Publication Fund of the Technische Universität Dresden. The funders were not involved in the conceptualization of the study, the collection, analysis, interpretation of data, the preparation of the manuscript, or the decision to submit it for publication.

ACKNOWLEDGMENTS

We thank the staff at the psychiatric outpatient department for assistance with data collection and Matthew Belanger for diligent proofreading.

REFERENCES

1. UNODC. *World Drug Report 2020, Booklet 2 Drug Use and Health Consequences*. United Nations publication, Vienna (2020).
2. Kariisa M, Scholl L, Wilson N, Seth P, Hoots B. Drug overdose deaths involving cocaine and psychostimulants with abuse potential - United States, 2003-2017. *Morb Mortal Wkly Rep.* (2019) 68:388–95. doi: 10.15585/mmwr.mm6817a3
3. Admon LK, Bart G, Kozhimannil KB, Richardson CR, Dalton VK, Winkelman TNA. Amphetamine- and opioid-affected births: incidence, outcomes, and costs, United States, 2004-2015. *Am J Public Health.* (2019) 109:148–54. doi: 10.2105/AJPH.2018.304771
4. Dinger J, Hinner P, Reichert J, Rüdiger M. Methamphetamine consumption during pregnancy - effects on child health. *Pharmacopsychiatry.* (2017) 50:107–13. doi: 10.1055/s-0042-122711
5. Perez FA, Blythe S, Wouldes T, McNamara K, Black KI, Oei JL. Prenatal Methamphetamine- impact on the mother and child - a review. *Addiction.* (2021). doi: 10.1111/add.15509. [Epub ahead of print].
6. Tse R, Kesha K, Morrow P, Glenn C, Stables S. Commentary on: Kenneally M, Byard RW. Increasing methamphetamine detection in cases of early childhood fatalities. *J Forensic Sci.* (2020) 65:1384. doi: 10.1111/1556-4029.14459
7. Dyba J, Moesgen D, Klein M, Leyendecker B. Methamphetamine use in German families: parental substance use, parent-child interaction and risks for children involved. *Subst Use Misuse.* (2019) 54:583–91. doi: 10.1080/10826084.2018.1528459
8. Braunwarth W-D, Christ M, Dirks H, Dyba J, Härtel-Petri R, Harfst T, et al. S3 *Practice Guideline Methamphetamine-Related Disorders*. 1st ed. Berlin: Agency for Quality in Medicine (2016).
9. Dinger J, Näther N, Wimberger P, Zimmermann US, Schmitt J, Reichert J, et al. Increasing consumption of crystal meth in saxony and its risks for mother and child - experiences at a level I perinatal center from a pediatric viewpoint. *Z Geburtshilfe Neonatol.* (2017) 221:73–80. doi: 10.1055/s-0043-102953
10. Stevens S, Rogers J, Dansereau L, DellaGrotta S, Lester BM, Wouldes TA, et al. Prospective study of service use in the year after birth by women at high risk for antenatal substance use and mental health disorders. *Int J Ment Health Addict.* (2019) 19:1005–1018. doi: 10.1007/s11469-019-00207-w
11. Lindsay B, Albrecht J, Terplan M. Against professional advice: treatment attrition among pregnant methamphetamine users. *Subst Abuse Rehabil.* (2011) 2:189–95. doi: 10.2147/SAR.S25083
12. Jones HE, Myers B, O'Grady KE, Gebhardt S, Theron GB, Wechsberg WM. Initial feasibility and acceptability of a comprehensive intervention for methamphetamine-using pregnant women in South Africa. *Psychiatry J.* (2014) 2014:929767. doi: 10.1155/2014/929767
13. Lewis D. We were wrong about "crack babies": are we repeating our mistake with "meth babies"? *MedGenMed.* (2005) 7:30.
14. Chi FW, Parthasarathy S, Mertens JR, Weisner CM. Continuing care and long-term substance use outcomes in managed care: early evidence for a primary care-based model. *Psychiatr Serv.* (2011) 62:1194–200. doi: 10.1176/ps.62.10.pss6210_1194
15. Manning V, Garfield JB, Best D, Berends L, Room R, Mugavin J, et al. Substance use outcomes following treatment: findings from the Australian Patient Pathways Study. *Aust N Z J Psychiatry.* (2017) 51:177–89. doi: 10.1177/0004867415625815
16. Groß C, Hahn S, Spreer M, Behrendt S, Dinger J, Reichert J, et al. "Mama denk' an mich" (MAMADAM) - ein multimodales Therapieprogramm für suchtkranke Schwangere, Mütter und Väter im Rahmen der psychiatrischen Institutsambulanz. *SUCHT.* (2018) 64:97–108. doi: 10.1024/0939-5911/a000533
17. Groß C, Schützwohl M, Mayer-Pelinski R, Hasler H, Kirchner T, Scheck A, et al. "CrystalClean" - a German-language manual for qualified detoxification and motivation treatment in cases of "crystal meth" dependency - feasibility and acceptance. *Psychiatr Prax.* (2020) 47:22–8. doi: 10.1055/a-1003-5148
18. Petzold J, Weber B, Bassett TR, Bauer M, Bernhardt N, Gross C, et al. Effectiveness of the first German-language group psychotherapy manual to accompany short-term treatment in methamphetamine dependence. *Front Psychiatry.* (2020) 11:130. doi: 10.3389/fpsyt.2020.00130
19. Grigg J, Manning V, Arunogiri S, Volpe I, Frei M, Phan V, et al. *Methamphetamine Treatment Guidelines: Practice Guidelines for Health Professionals*. 2nd ed. Richmond, VA, Victoria: Turning Point. (2018).
20. Maglione M, Chao B, Anglin MD. Correlates of outpatient drug treatment drop-out among methamphetamine users. *J Psychoactive Drugs.* (2000) 32:221–8. doi: 10.1080/02791072.2000.10400232
21. Glasner-Edwards S, Marinelli-Casey P, Hillhouse M, Ang A, Mooney LJ, Rawson R. Depression among methamphetamine users: association with outcomes from the methamphetamine treatment project at 3-year follow-up.

J Nerv Ment Dis. (2009) 197:225–31. doi: 10.1097/NMD.0b013e31819db6fe

22. Petzold J, Szumlinski KK, London ED. Targeting mGlu(5) for methamphetamine use disorder. *Pharmacol Ther.* (2021) 224:107831. doi: 10.1016/j.pharmthera.2021.107831
23. Heinz A, Kiefer F, Smolka MN, Endrass T, Beste C, Beck A, et al. Addiction research consortium: losing and regaining control over drug intake (ReCoDe)-From trajectories to mechanisms and interventions. *Addict Biol.* (2020) 25:e12866. doi: 10.1111/adb.12866

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 15 November 2021
doi: 10.3389/fpsyt.2021.783189

The Family Talk Programme in Ireland: A Qualitative Analysis of the Experiences of Families With Parental Mental Illness

Christine Mulligan[†], *Mairead Furlong*[*†], *Sharon McGarr, Siobhan O'Connor and Sinead McGilloway*

Centre for Mental Health and Community Research, Maynooth University, Maynooth, Ireland

Background: Parental mental illness is common, costly, can lead to children developing mental disorders and impaired lifetime outcomes, and places a substantial burden on caregiving partners. Family Talk (FT) is a widely implemented, 7-session, whole-family programme, with promising evidence of effectiveness in targeting the intergenerational transmission of mental illness. However, to date, very little qualitative research of family experiences of FT has been undertaken. The objectives of this study were to: (1) investigate the experiences of families attending FT; and (2) explore the key facilitators and barriers to engagement in mainstream mental health settings.

Methods: This study was nested within a randomised controlled trial (RCT) of Family Talk [$N = 86$ families (139 parents, 221 children)] implemented in 15 adult, child and primary care mental health sites in Ireland. Semi-structured interviews were conducted with a purposive sample of 45 participants, including 23 parents with mental illness (PMI), 7 partners and 15 children/young people aged 9 to 18 years. Interview data were transcribed verbatim and analysed using constructivist grounded theory.

Results: Over two thirds of families across sites reported substantial benefits from participation in FT, including reduced stigma, giving children and partners a voice, increased service-user confidence, and improved family communication/relationships. Key facilitators identified by families included: programme delivery by a competent, non-judgmental clinician; the whole-family approach; and family readiness to engage. Barriers to engagement included stigma, family crises/relapse, service constraints, impact of COVID-19, and a need for further child, family and follow-up sessions/supports.

Conclusion: This study is the first qualitative analysis of family experiences of FT to be conducted within the context of an RCT and national programme to introduce family-focused practise for families with PMI. The findings illustrate that FT is beneficial across cultural/policy contexts, different mental disorders and can be implemented

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Tariq Munshi,
University of Toronto, Canada
Joanna Fox,
Anglia Ruskin University,
United Kingdom

***Correspondence:**
Mairead Furlong
Mairead.Furlong@mu.ie

[†] *These authors have contributed equally to this work and share first authorship*

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *25 September 2021*
Accepted: *22 October 2021*
Published: *15 November 2021*

Citation:
Mulligan C, Furlong M, McGarr S, O'Connor S and McGilloway S (2021) The Family Talk Programme in Ireland: A Qualitative Analysis of the Experiences of Families With Parental Mental Illness. Front. Psychiatry 12:783189. doi: 10.3389/fpsyt.2021.783189

across adult and child mental health settings, including children with existing mental health challenges. Key barriers and facilitators to implementation were identified by families, all of which should help to inform the future implementation of FT, and other similar interventions, both in Ireland and elsewhere.

Keywords: children, COPMI, Family Talk, mental health, mental disorder, mental illness, parents, qualitative research

INTRODUCTION

It is estimated that 23% of all families have at least one parent who has, or had, a mental illness; this has been shown to increase the risk of children developing a mental disorder during their lifetime (range 41 to 77%), whilst multiplying five-fold their utilisation of health and social services, and placing a substantial emotional, financial and parenting burden on caregiving partners (1–3). In the Republic of Ireland (RoI), 20% of adults experience a mental health illness—the third highest incidence across 36 countries in Europe—costing the Irish state €11 billion per year (4). Furthermore, it is estimated that 280,000 children in the RoI are dependent on parents who have a mental illness (5).

The transmission of risk from parents to children involves a complex interplay of genetic, prenatal, family and environmental/social influences and is significantly mediated by the impact of parental symptoms on parent-child interactions (e.g., insensitive and erratic attunement)(2). Worryingly, these vulnerable families are often not identified or supported by mental health professionals in the RoI, or in other jurisdictions, due to: a lack of policy/practise guidance; little or no collaboration between Adult Mental Health Services (AMHS) and Child and Adolescent Mental Health Services (CAMHS); an individualised, crisis-oriented approach to assessment/treatment; competency and confidentiality concerns amongst mental health professionals who may feel ill-equipped to undertake family work; and parental stigma/fear of social services and losing custody of their children (6, 7).

Although the prevalence and burden of parental mental illness (PMI) is a cause for public concern, there is increasing evidence that integrated prevention and early intervention family-focused programmes/practise (FFPs) can help decrease the risk of developing mental disorders for children by up to 40% (8) and reduce referrals to child protection services (9). The Family Talk programme, in particular, has been identified in several systematic reviews (8, 10, 11) as a key intervention with promising evidence of effectiveness in improving parent and child understanding of mental illness and child internalising symptoms (12–16), with one study indicating enhanced family functioning and parental mental health recovery 4.5 years later (14).

Family Talk (FT) was developed by William Beardslee and colleagues in the USA in the 1980's and is a manualised, 7-session, strengths-based, psycho-educational, whole-family approach designed to enhance family understanding and communication about parental mental illness, improve family interpersonal relationships, and promote family resilience and utilisation of social supports (12). The intervention involves a clinician meeting with each individual family, i.e. with parents (sessions 1, 2, 4, 6, 7), with each child individually (session 3), and with the whole family (session 4) (see **Figure 1**). Sessions typically last 60–90 min. The current evidence base for FT is limited by the small number of RCTs conducted to date and within only three countries (USA, Finland, Germany), generally small sample sizes, and mixed support for effectiveness in improving child externalising symptoms, parental mental health and family functioning (14, 15, 17, 18).

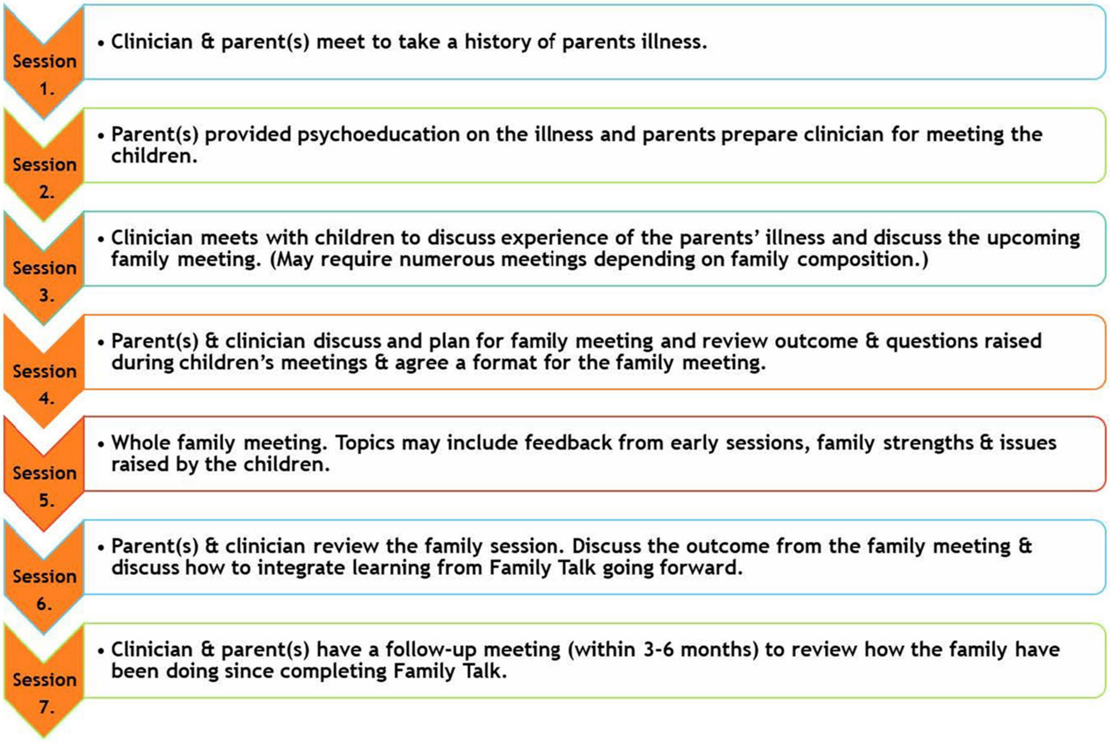
Due to its small but growing evidence base, FT has been implemented in recent years in several countries to support families where a parent has mental illness [e.g., the USA (Chicago), Costa Rica, Colombia, the Netherlands, Greece, Scandinavia (Norway, Sweden, Finland), Iceland, and Australia (Victoria)] (19). Internationally, there has been a growing trend, informed by the United Nations Convention on the Rights of the Child, to introduce legislation mandating support for children when a parent suffers from serious mental illness [e.g., the Swedish Health and Medical Service Act (20, 21)]. This legislation means that psychiatric services are obliged to take patient's children into consideration, including meeting their needs for information and support, and discussing issues of parenthood and the children's well-being (21). However, the continuing stigma around mental illness, especially as a parent, coupled with service provider constraints, often means that these policies are not implemented in practise (22).

Within the Irish context, whilst national practise guidelines have recently re-oriented toward a recovery,[1] strengths-based model of care that recognises the needs of family carers and the value of family-focused mental health practise (23–25), there is no specific policy/practise guidance to support families with PMI in the RoI. Consequently, the national Health Service Executive (HSE) provided funding for the current research programme—called "PRIMERA" (**P**romoting **R**esearch and **I**nnovation in **M**ental h**E**alth se**R**vices for f**A**milies and children), the primary aims of which were to: (1) identify/develop, implement, and

---

Abbreviations: ADHD, attention deficit hyperactivity disorder; AMHS, adult mental health services; ASD, autism spectrum disorder; CAMHS, child and adolescent mental health services; FFP, family-focused practise/programmes; FT, Family Talk; HSE, Health Service Executive; MI, Mental illness; PMI, parents with mental illness; PRIMERA, **P**romoting **R**esearch and **I**nnovation in **M**ental h**E**alth se**R**vices for f**A**milies and children; PTSD, post-traumatic stress disorder; RCT, randomised controlled trial; RoI, Republic of Ireland; SUP, Service-user parent.

[1] The approach argues against just treating or managing symptoms but focusing on building the resilience of people with mental illness and a change in outlook that is related to leading a meaningful, purposeful life, with or without ongoing episodes of illness (23).



- Clinician & parent(s) meet to take a history of parents illness.

- Parent(s) provided psychoeducation on the illness and parents prepare clinician for meeting the children.

- Clinician meets with children to discuss experience of the parents' illness and discuss the upcoming family meeting. (May require numerous meetings depending on family composition.)

- Parent(s) & clinician discuss and plan for family meeting and review outcome & questions raised during children's meetings & agree a format for the family meeting.

- Whole family meeting. Topics may include feedback from early sessions, family strengths & issues raised by the children.

- Parent(s) & clinician review the family session. Discuss the outcome from the family meeting & discuss how to integrate learning from Family Talk going forward.

- Clinician & parent(s) have a follow-up meeting (within 3-6 months) to review how the family have been doing since completing Family Talk.

FIGURE 1 | Family Talk sessions.

evaluate family-focused interventions for families with PMI; and (2) inform a "think family" care delivery agenda within mental health services in Ireland. Following an initial scoping study that demonstrated a lack of structured support for this population in the RoI, it was agreed with stakeholders that clinicians across 15 AMHS, CAMHS and child protection/welfare service sites would deliver Family Talk as part of a randomised controlled trial (RCT), with embedded qualitative and economic analyses (6, 26). FT was chosen for implementation as it: incorporates a structured "whole family" evidence-based approach; can be used with a range of mental disorders; provided freely available and high quality online training/resources[2]; and was replicable and capable of being implemented across sites in Ireland (6).

Despite the growing number of trial evaluations of FT, very few qualitative studies to date have investigated the experiences of families in attending FT. This means that little is known about the barriers and facilitators of change, intervention characteristics or contextual factors that may influence implementation and trial outcomes, particularly when delivered in real-world service settings (27). Indeed, the voices of service users, their families and particularly children, are rarely heard in controlled evaluations of FFPs (10, 28). Previously, it has been found that children may have a different perspective on "what helps" compared to parents and mental health practitioners (29). In addition, partners of service users have reported feeling uninvolved in research, which compounds their experience of feeling unsupported in their care burden by mental health services (30, 31). Thus, eliciting the views of children and other family members regarding FT delivery is important for informing the future development and refinement of this, and other similar, programmes.

Five qualitative studies eliciting family experiences of FT have been conducted, to date, all undertaken in Sweden, three within outpatient psychiatric settings (32–34), one within a substance misuse clinic (35), and another in an open care psychosis unit (36). With regard to the last of these, a companion study of clinician reports of family experiences of FT was also conducted (37). **Table 1** summarises the participant characteristics across these studies). Collectively, the findings from these studies from both parents and children show that: the silence around mental illness in their home had been broken, they had greater understanding of mental illness, and more open family communication and closer relationships, although the level of improvements varied across and within families (32–36). Service-user parents felt more equipped and empowered in their parenting role and children expressed relief from fears, less monitoring of their parents, less carework in the home, and being able to spend more time with friends and other interests (33, 35, 37).

Arguably, these findings are potentially biassed in that they did not interview families who refused to attend or disengaged from the programme. High rates of refusal and attrition have been noted elsewhere, often due to competing needs for daily survival

---

[2] https://emergingminds.com.au/online-course/family-focus

TABLE 1 | Qualitative studies of Family Talk.

References	Cohort interviewed	Recruitment agency	Parental diagnosis	Method & analysis	Country
Pihkala et al. (32)	10 service-user parents (SUPs)	Adult psychiatry	Depression	Qualitative interviews, grounded theory	Sweden
Pihkala et al. (33)	14 children from 9 families, aged 6-17 yrs	General psychiatry	6 depression, 1 psychosis, 1 anxiety and ADHD, 1 with PTSD	Qualitative interviews, content analysis	Sweden
Pihkala et al. (34)	17 SUPs & 8 partners from 18 families	General psychiatry	11 depression, 2 personality disorder, 2 bipolar, 1 anxiety and ADHD, 1 psychosis and PTSD	Qualitative interviews, grounded theory	Sweden
Pihkala et al. (35)	7 SUPs, 7 partners & 10 children, aged 8-15 yrs	Clinic for substance use disorder	All 7 parents diagnosed with substance misuse comorbid with depression, anxiety and/or bipolar disorder.	Qualitative interviews, content analysis	Sweden
Strand and Meyersson (36)	8 SUPs & 7 children, aged 8-15 yrs	Open care psychosis units	4 schizophrenia and 4 schizoaffective disorder	Qualitative interviews, content analysis	Sweden
Strand and Rudolfsson (37)	11 Family Talk clinicians	Open care psychosis units	Parental psychosis	Qualitative interviews, thematic analysis	Sweden

ADHD, attention deficit hyperactivity disorder; PTSD, post-traumatic stress disorder; SUP, Service-user parent.

and fear of judgement (15, 37). A limited range of informants (e.g., mostly PMIs with depression, limited data from partners or those who disengage from FT), small sample sizes, and an overall lack of cultural diversity, underscore the need for qualitative analyses to be undertaken across a wider variety of settings and contexts. For instance, FT is not always delivered in countries with specific policy/practise guidance for this population.

This qualitative study was nested within an RCT of the Family Talk intervention in Ireland for families with parental mental illness and children aged 5–18 years; the aim of the RCT was to assess the nature and extent of any pre-post intervention changes in child and family psychosocial functioning (26) and data analysis is currently underway. The objectives of the current study were to: (1) investigate the experiences of families attending FT; and (2) explore the processes of change, contextual factors or intervention characteristics that may influence trial outcomes in mainstream mental health settings (26).

METHODS

Participants and Settings

The larger RCT included 86 families (139 parents, 221 children) in 15 sites across the RoI, involving adult, child, and primary care mental health services, and Tusla child protection services (26). Families (parents and children aged 5–18 years) were recruited by clinicians in each site from their existing waiting lists, and written informed consent/assent was obtained for their participation in the research (26). FT was delivered in a mental health outpatient clinic and/or in the home by a mental health professional, typically a social care worker, social worker, or psychologist. Families were eligible where a parent had a formally diagnosed mental disorder, with 80% of service-users attending AMHS for various mental disorders and 20% receiving antidepressant medication or primary care psychological support under the governance of a General Practitioner (26). Due to the high risk of intergenerational transmission of mental disorders (2), and a desire among stakeholders to increase family-focused collaboration between traditionally segregated adult (AMHS) and child mental health services (CAMHS) (6), we included families where children attended CAMHS or primary care services for mental health issues, as well as families where children were not involved with mental health services (26).

Participants were block randomised, on a 2:1 ratio, to the FT intervention ($n = 56$) or to a treatment as usual control group ($n = 30$). Assessments were carried out at baseline and at six month follow-up periods. At six-month follow-up, attrition was 37%, the rate of which doubled due to the impact of the COVID-19 lockdown restrictions (22.8 vs. 45%). More details on study parameters can be seen in the study protocol (26). The flow of participants from recruitment through the RCT to the qualitative studies is shown in **Figure 2**.

For the qualitative study, a purposive sampling method was used to approach prospective participants ($n = 34$ families) for interview on the basis of key demographic variables (e.g., age, gender, lone parent, mental disorder, number of children, site location and (mainly socially deprived) socioeconomic status). A series of 37 one-to-one semi-structured interviews and 3 group-based family interviews were undertaken at 6 month follow-up with a total of 45 participants from 23 families, including 23 parents with mental illness (PMI), 7 partners and 15 children/young people aged 9 to 18 years. (While children over five could participate in FT, only those aged over 8 years could participate in the research process as the assessment measures were not suitable for the younger age group). Fourteen families attended all FT sessions while nine dropped out after completing less than three sessions, and were interviewed to provide a "negative case" analysis. In the RCT sample, mean attendance in the intervention group was 4.4 sessions ($SD = 1.2$), with 53% attending all sessions.

The qualitative sample had a largely similar profile to the larger RCT cohort in terms of the demographics indicated above. Twelve of the interviewed families were recruited by AMHS and eleven by CAMHS. Service-user parents (i.e., parent was attending mental health services, usually AMHS, for their mental health challenges) had a mean age of 41.6 years ($SD = 8.2$) and were predominantly female (18/23), Caucasian

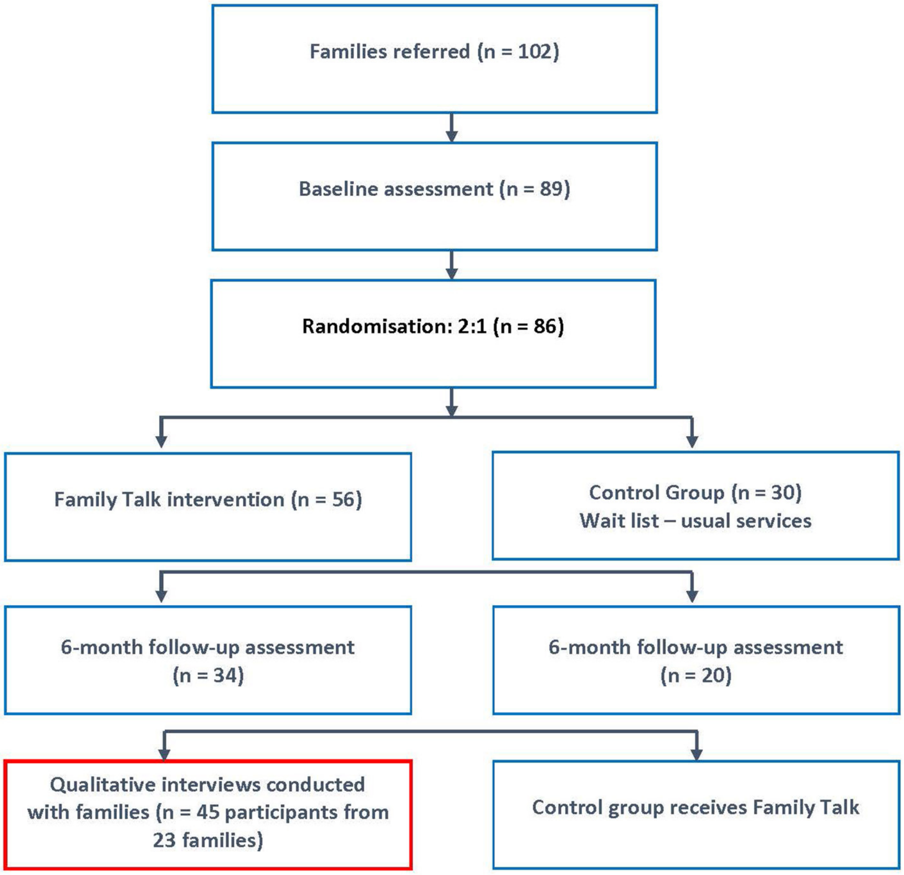


FIGURE 2 | Study flow diagram.

(22/23), and socially disadvantaged (15/23); approximately half (11/23) were lone parents and the largest proportion reported anxiety/depression ($n = 14/23$), followed by bipolar disorder ($n = 4$), Borderline Personality Disorder ($n = 2$) and psychosis ($n = 3$). Six of the seven partners who agreed to be interviewed were married. Ten of the 15 child participants were male with a mean age of 13.2 years ($SD = 2.8$), and approximately half (8/15) reported attending, or were on a wait list for, CAMHS. The children in the larger RCT tended to be more evenly distributed by gender and were also a little younger ($M = 10.3$; $SD = 5.3$), whilst 42% were attending either CAMHS or psychology/family services (**Table 2**). More details on the characteristics of interviewed families can be seen in **Table 2**.

Data Collection and Analysis

Ethical approval (for both the RCT and qualitative study) was obtained from four research ethics committees including the HSE, the research institution where the research was carried out [name withheld for anonymous peer review], and two of the services with whom the research team worked (called Tusla and Saint John of God's Hospitaller Services).

Consent/assent forms were administered to parents and children, outlining details of the PRIMERA study, its potential benefits/risks, and where to seek help if necessary. Parents provided written informed consent for their children to participate and then their children provided written informed assent. Interview schedules were devised for each of the three participant groups in order to guide, and provide a framework for, interviews. These included questions such as "Tell me about your experience of FT, "What did you like about it?," "What would you change about FT?" and "Would you recommend FT to other families?" Families who completed <3 sessions were asked their reasons for not completing FT. Interviews lasted between 15 and 40 min, with 33 (73%) conducted in participant's homes, and 12 (27%) via online platforms during the COVID-19 lockdown restrictions. There was some evidence of possible gatekeeping in three families with the PMI limiting access to family members. In addition, two parents requested to sit in on the children's interview. The interviews were conducted by experienced researchers [CM ($n = 38$), SMcGa ($n = 7$)], with lived experience of PMI, and with qualifications in psychology, mediation and psychotherapy. Given the stigma and impact of PMI, every effort was made to create a warm and non-judgemental atmosphere to ensure that participants felt understood. In addition, rapport had been established prior to the interviews as both researchers had prior contact with families

TABLE 2 | Characteristics of interviewed families.

Family ID	Referring agency	PMI	Marital Status	Mental diagnosis	Age	Partner mental health	No. Children	Children's mental health/services	Interview configuration	FT attendance
1	AMHS	Female	Living together	Schizophrenia	45	Substance use disorder & anxiety	5	One child has anxiety, attends family support service	Dyad with parents	Completed FT
2	AMHS	Female	Single parent	Anxiety	31	History of domestic violence	3	Son and daughter in CAMHS with ASD and anxiety/self harm	1:1 with PMI 1:1 with eldest son	Completed FT
3	AMHS	Male	Married	Bipolar	49	None identified	3	One son in CAMHS with ASD	1:1 with each family member	Completed FT
4	AMHS	Male	Married	Depression/ PTSD	38	None identified	3	Not in services. Eldest child on waitlist due to anxiety symptoms	1:1 with both parents & two children	Completed FT
5	AMHS	Male	Married	Low mood/ Anxiety	47	None identified	3	Not in services	1:1 with PMI	Completed FT
6	CAMHS	Female	Married	BPD	42	None identified	3	One child in CAMHS with self-harm & emotional deregulation	1:1 with each family member	Completed FT
7	AMHS	Female	Divorced	Schizophrenia	52	None identified	3	Not in services	1:1 with PMI	Left after 3 sessions
8	CAMHS	Female	Married	Depression	48	Depression symptoms	3	Eldest in CAMHS with depression and youngest with behavioural difficulties	1:1 with eldest child; group interview with other family members	Completed FT
9	AMHS	Female	Married	Depression	36	None identified	3	Eldest in CAMHS, feeling suicidal	1:1 with PMI	Completed FT
10	CAMHS	Female	Married	Anxiety/ Depression	40	None identified	2	Eldest in CAMHS with depression	1:1 with both parents & eldest child	Completed FT
11	CAMHS	Female	Widowed	Depression	37	N/A	3	Eldest in CAMHS, suicidal	1:1 with PMI	Did not start FT
12	AMHS	Male	Separated	Depression	43	N/A	1	Not in services	Dyad with father & son	Completed FT
13	CAMHS	Female	Single parent	Bipolar/ADHD	39	N/A	2	Eldest in CAMHS for stress/ADHD	1:1 with PMI	Completed FT
14	CAMHS	Male	Married	Depression	50	None identified	5	Three children in CAMHS – all with anxiety	1:1 with PMI	Completed FT
15	CAMHS	Female	Single parent	Depression	44	N/A	5	Two children in CAMHS – depression/anxiety and ADHD/ASD	1:1 with parent & youngest child	Completed FT
16	CAMHS	Female	Married	Depression	51	Depression & history of panic attacks	3	Not in services. One child on CAMHS waitlist, suicidal thoughts	1:1 with both parents	Completed FT
17	AMHS	Female	Lone parent	Psychotic depression	36	N/A	1	Not in services	1:1 with PMI	Did not start FT
18	AMHS	Female	Lone parent	Depression	48	N/A	2	Both children in CAMHS – social anxiety, self-harm	1:1 with PMI	Left after 2 sessions
19	CAMHS	Female	Married	BPD	35	None identified	4	One child in CAMHS with anxiety	1:1 with PMI	Left after 3 sessions
20	AMHS	Female	Married	Depression & anxiety	37	Depression & anxiety	2	Not in services	1:1 with PMI	Left after 1 session
21	CAMHS	Female	Lone parent	Bipolar	41	N/A	2	One child in CAMHS with anxiety	1:1 with PMI	Did not start FT
22	AMHS	Female	Lone parent	Bipolar affective disorder	42	N/A	2	Not in services but says child is depressed	1:1 with PMI	Did not start FT
23	CAMHS	Female	Lone parent	Anxiety	34	N/A	1	Child in CAMHS with social anxiety	1:1 with PMI	Left after 2 sessions

ADHD, attention deficit hyperactivity disorder; AMHS, Adult Mental Health Services; ASD, autism spectrum isorder; BPD, Borderline Personality Disorder; CAMHS, Child and Adolescent Mental Health Services; FT, Family Talk; PMI, parent with mental illness; PTSD, Post traumatic Stress Disorder.

during the baseline and 6 month RCT assessments (blinding was broken after the 6 month assessment to complete the qualitative interviews). Parents received a €25 gift voucher as a token of thanks for participating in the qualitative interview and children received a €10 voucher. Interviews were audio recorded with consent and transcribed verbatim.

The data from the interviews were analysed using constructivist grounded theory and MAXQDA software in order to identify and organise themes (38, 39). Analysis was also informed by the Medical Research Council guidance for complex interventions (27). Data were analysed using line-by-line and focused coding, constant comparison of codes to find similarities and variations within categories and hierarchical linking of categories to generate super-ordinate (or overarching) themes. The epistemological stance of constructivist grounded theory is more explicit than grounded theory in acknowledging the interpretive or constructivist nature of generating themes (38). The research interviewers were sensitised to honouring the lived experience of all participants (and particularly children) given the lack of data from this often invisible cohort but also due, in part, to their history of PMI. All interview transcripts were read by CM and MF, CM coded and analysed all of the data, while three authors (MF, SMcGa, SOC) independently assessed the reliability of coding on 12 of the 45 (27%) interviews. Reporting conforms to COREQ (Consolidated Criteria for Reporting Qualitative Research) guidelines (40).

RESULTS

Two overarching themes were identified from the analysis: (1) Benefits and experiences of FT and (2) Key barriers to participation (**Table 3**). A number of subthemes were also identified within each.

TABLE 3 | Qualitative themes and subthemes of family experiences of Family Talk.

Theme 1: Benefits and experience of FT	
From fear and silence to sharing and empowerment	Experiences of service-user parents - Reduced stigma and worry - Deeper understanding of impact of MI on children - Better family relationships (communication, support) - Parental confidence and enhanced wellbeing
	Hearing the child's voice - Disclose hidden concerns and burdens - Better understanding of PMI - Relief and less worry - Warmer, more open family relationships
	Partners' experiences - Relief at having burden validated - Enhanced team approach to supporting PMI - Closer family relationships
Facilitators of change	Clinician skill
	Whole-family approach Timeliness/readiness
Theme 2: Key barriers to participation	
Initial engagement phase	Parental stigma and beliefs
	Lack of clarity for children on purpose of FT Service constraints
Intervention phase	Emotionally challenging, but in a good way
	Varied within-family experiences
	Covid complications Disengaging from FT
Ending phase	More child, family and follow-up sessions Need for additional supports

FT, Family Talk; MI, Mental illness; PMI, Parental mental illness.

Theme 1. Benefits and Experiences of FT: From Fear and Silence to Sharing and Empowerment

Despite initial reluctance and fear about discussing mental health in a family context, the majority of families who attended FT (14/16) reported substantial benefits from participation, including: reduced worry and stigma, a greater understanding of the impact of PMI on family members; giving children a voice; improved parental confidence and support; improved family communication, problem solving, and warmer relationships. A total of four sub-themes were identified here.

Benefits to PMI

Three quarters of service-user parents (12/16) reported a reduction in shame, stigma and worry about being a "bad parent" following the intervention, which helped to improve their sense of well-being and parental confidence. Labelling was a common source of stigma. One service-user parent, for instance, agreed to participate only on the condition that the term "bipolar" was not used with his children. Another parent recalled the pejorative names used by his wife, such as "crazy" or "mentaller". Such labelling encouraged the PMI not to share their suffering and to try to appear "normal."

> *"I became very good at hiding things, trying to adapt and fit in and mirroring other people that were deemed to be socially acceptable."* (PMI 5)
>
> *"After coming out and saying it to them, and talking to them about it, there is nothing to be ashamed of."* (PMI 13)
>
> *"It was hard. But it was very relieving because there was a lot of stuff that I would have been fearing to talk about or say out loud."* (PMI 12)

FT also helped parents to have a better understanding of the impact of their mental illness on their children. While most parents feared that discussion of their mental illness would burden their children, they were relieved to learn that more open communication enabled them to better understand their child's perspective, and empowered them to address child concerns and unspoken inaccurate beliefs. For example, one son panicked if his mother mentioned the doctor or heard an ambulance siren, fearing she would be re-hospitalised. Another secretly feared his mother was dying from cancer, while children in another family felt that they were somehow to blame for their mother's illness. The FT sessions also allowed parents to explain frightening past behaviour to their children, thereby allaying anxieties. Listening to their children's accounts was an emotional experience for all parents who completed FT.

> *"It helped us as a family to see from their [kids] vantage point how it affected them and try to give them what they wanted to try to move on."* (PMI 4)

> *"She [*daughter aged 11 years*] said she wasn't a very good daughter. When I was getting cross or why things weren't harmonious in the house, she felt that it was her fault."* (PMI 16)

> *"I was able to think of them more as people rather than my children that I'd be trying to protect, keep them safe from everything... Anything that they needed to talk about or worried about, and without fear of repercussion. Being able to say it in a safe place was good, for all of us really. There were tears and everyone at the end of it felt good and felt heard and respected in it."* (PMI 13)

> *"I just felt brilliant after it and I was able to tell them how proud I was of them and how much I love them. I can't just put words on it. But it has changed us for the better."* (PMI 15)

The direct involvement of children also helped parents to re-evaluate their understanding of children's prior behaviour; instead of assuming that silence indicated the child's lack of awareness of the illness or lack of care for the parent, the PMIs realised that a child's silence is more often an attempt to protect the family and/or to avoid burdening them.

> *"Beforehand I was saying, oh they don't want to talk to me... It's that they didn't want to be putting extra worry on me about anything because I had a mental health issue."* (PMI 6)

> *"I did not realise my eldest was being bullied for 2 years in school during my illness. He kept it to himself because at the time, he worried about me killing myself."* (PMI 1)

Service-user parents also indicated that the sessions improved their communication with, and support from, their partners, as well as from their children. Overall, improved family interactions and relationships appeared to assist mental health recovery and personal and parental confidence

> *"It gave me a sense of kind of, well not accomplishment... it was a huge sense of like, I'm doing this, I'm going to help* [the children]... *Having been through it, it gave me strength in a way."* (PMI 2)

Hearing the Child's Voice

All but three the children (12/15) reported that they found FT to be helpful despite their initial reservations about attending and the emotional challenge of participating in individual/family session (Three of the younger children found it difficult to recall FT as they were interviewed 4 months following FT). Children indicated that they felt empowered by the opportunity to: voice "hidden" concerns about PMI, family dynamics, and other issues; and to develop a deeper understanding of their parent's mental illness. Previously undisclosed concerns included: distress with the PMI's behaviour (e.g., anger, social withdrawal, self-harm) and/or with arguments and tension at home, feeling overburdened by caretaking activities, being bullied, educational disruption, child depression, suicidal ideation, feelings of blame, fear, sadness and injustice/anger. Eldest and only children were more likely to carry a largely unacknowledged burden in caring for family members, especially during heightened presentation of symptoms. Caretaking responsibilities included: caregiving, cleaning, shopping, cooking, financial responsibilities, and looking after the emotional well-being of siblings and the other parent. The dominant unspoken message of silence around PMI, combined with an often unavailable partner (due to work pressures, absence from the home, and/or emotional disconnection), left children feeling overwhelmed and unsupported.

> *"Dad was absolutely working his ass off to try and get money for us and taking care of Mam and running in and out of hospital. And doing school with us, trying to get us to do our homework and everything. He needed a lot of help. From a young age, myself and my sister had to take on a role, me more so because Dad was working and trying to provide. Mom was either in bed or in hospital, so I'd be like at school, have to come home, mind my siblings, my sister had to cook."* (Eldest child, 16 years old)

> *"I do get in a terrible mindset when it comes to my Mum's mental health because it's not nice... impacts on all of us as a family."* (Eldest child, 18 years old)

> *"I ask Mum for a lot of days off school because I get fed up with all the bullies...I used to have dark thoughts, not wanting to wake up...I do worry a lot. Because my Mum doesn't really have another person to help her, I'm normally that other person."* (Middle child, 10 years old)

> *"I pretty much have been in a really bad state since I was about nine, really low depression and suicidal. The only reason I didn't tell my parents was because my Mum's sick, my Dad doesn't care. It's not that they don't care, it's just that my Dad was working. I was kind of like, my Mum's in bed sick. I can't be talking to her, she needs to get better first."* (Middle child, 14 years old)

Many children indicated that their parents were unaware how much they had been affected by tense/volatile home situations, and had hidden their concerns to avoid burdening parents. As parents became more cognisant of children's needs, family members were motivated to reduce levels of anger/arguments, and to relate to each other in more warm, caring and fun ways, thereby leading to reduced stress and worry and increased child well-being. For instance, family members made more efforts to connect with each other by having regular meals, spending time with each other, and being generally more cooperative and supportive. Siblings also advocated for each other's well-being in sessions, which helped to improve sibling relationships following FT. While there was still some evidence of parentification among children following FT, several expressed relief that FT had broken the silence, secrecy and stigma around mental illness within their families, and that as well as feeling that parents were now listening to them, they also experienced increased empathy and compassion for their parents whom they perceived as "trying their best" in challenging circumstances.

> *"I found it was helpful for the family, like to talk about this, because usually when we're home, we don't really talk about it properly."* (Youngest child, 12 years old)
>
> *"I felt like the course has helped quite a lot to be honest. Family life has just got a lot easier. We're not arguing as much, we're not shouting. It's just easier to talk to people now...You have the resources to actually talk about it and try and sort it out."* (Eldest child, 16 years old)
>
> *"It was definitely worth doing. Because without the course we mightn't have known anything about it. And for him* [Dad] *to understand that we understand what he has. It's kind of improving him and us."* (Middle child, 14 years old)
>
> *"He* [FT clinician] *was asking how I was getting along with my Dad and my Mam, and I said, "Ok, we fight a lot, then it improved" and then we were called back in, and we were just saying, "Me and my Dad improved," and he* [FT clinician] *said, "That's good."* (Youngest child, 9 years old)
>
> *"I think everyone's being a lot less aggressive, everyone's just trying to be a bit nicer to each other. I think everything's kind of been a lot calmer, especially with my Dad, he's been a lot calmer recently and he's started to kind of take other people's opinions and ideas into account."* (Eldest child, 16 years old)
>
> *"It helped me a lot. It made me feel better."* (Eldest child, 10 years old)

Partners' Experiences

While partners corroborated the benefits noted above, the largest single gain from their perspective, was that FT provided them with a forum in which they could voice their experience of partnering and co-parenting with a PMI, often for the first time, despite their partner being in mental health services sometimes for up to 20 years. All partners spoke of the stresses of caregiving, financial and household responsibilities, feelings of loneliness, frustration and helplessness, and strains on their marital relationships. Partners indicated that their isolation was amplified by exclusion from the PMI's treatment/care plan, and that they lacked the knowledge or skills to help their partner. Maintaining the focus on the service user's ill health also negatively influenced some partners' self-care, with three partners (3/7) managing their own mental health difficulties including anxiety, depression and alcohol misuse.

> *"You feel like you're carrying a whole house on your shoulders. You feel like a right tool. I can't do anything right. I can't say anything right. I can't help... I don't know what to do."* (Partner 6)
>
> *"I needed to ring someone, just for advice or help. But there was nobody."* (Partner 4)
>
> *"Over the last 4 or 5 years, we even talked about splitting up."* (Partner 3)

All partners described relief at having their experiences validated by the FT clinician and acknowledged by the PMI. While heated discussions and angry outbursts were common in the initial sessions, they were seen as worthwhile as it increased understanding and empathy between parents on the burdens that each was carrying. Five partners indicated that their relationship with the PMI had improved following FT. Partners also expressed to the PMI that they wanted to know how best to support them and wanted to be involved in their careplans. The dialogical approach of FT sessions helped to encourage a team approach to supporting the PMI, helping both parents feel more connected.

> *"It's an opportunity for him* [husband] *to hear me voicing the impact that it has on me in a very calm manner, because I'm in front of somebody else. It also takes away some of the guilt or the blame for me on his side... when you are more involved in the treatment."* (Partner 3)
>
> *"These sessions were great because we were both able to see where the other person was coming from."* (Partner 1)
>
> *"I think it [Family Talk] is 100% needed. As I said, there was nobody out there for me or the kids that I knew about... I can't compliment it enough. It's just the best thing that happened."* (Partner 4)

Facilitators of Change

Clinician Skill

The majority of PMIs and partners indicated that it was the skill of FT clinicians that mediated the benefits for families. Parents welcomed the non-judgemental and strengths-based approach adopted by clinicians, and their skill in facilitating multiple perspectives across several developmental ages. In addition, the PMIs (12/16) appreciated the clinician-led, psycho-educational aspect of the programme, which led to a deeper understanding and normalisation of their mental health challenges.

> *"Family Talk was very positive because there was somebody, a trained professional who had seen this before-it was in a way normalised. The kids were worried they were the only family in Ireland who had this problem."* (PMI 5)
>
> *"Everything that I talked about and went through, I had full support from her* [FT clinician]. *I can't even tell you how good she was. I can't say it enough. She was unbelievable."* (PMI 12)
>
> *"Everyone can say how they felt without any fear...Everyone felt very good afterwards and it was like a weight lifted... It's like a friendship with someone* [clinician] *that knows what you're talking about."* (PMI 13)

Whole Family Approach

All family members believed that FT worked because it involved the whole family, and allowed multiple, often hidden, stigmas, concerns and burdens to be revealed and shared, thereby validating each person's lived experience, whilst also empowering them to be more supportive of each other. Participants indicated that the focus on the family unit had helped them to look beyond

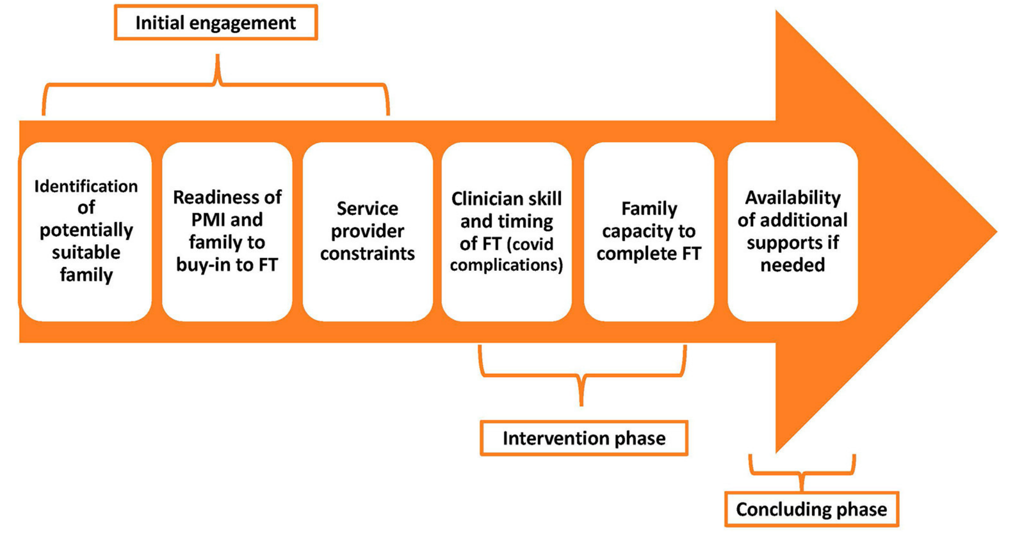


FIGURE 3 | Challenges to participation in Family Talk during the engagement, intervention and ending phases.

their individual burdens and to feel deeper understanding and empathy for each other.

> *"It* [FT] *opened up the family and they talked about what they wanted to say and everything and you knew exactly where you stood, and it was up to you then to change the wrong things to try change them to the right things...It was brilliant because it brought out everything, the good and the bad, which was good."* (Partner 7)
>
> *"I just remember it was good for our family to actually talk properly without any kind of aggression, without any blame... everyone could just say how they saw things and people would put in their input without anyone kind of being upset about it. It was good to have like outside influences making sure everything was just calm."* (Eldest child, 16 years old)

Timeliness/Readiness

Parents also indicated that timing, setting and their readiness for FT were important factors in engagement. If approached too early, they said that they might have denied the impact of their illness on their family/children. They also required a lead-in time to build up the courage and find words for the difficult initial conversations with their partners and children in order to convince them to participate. Furthermore, in order to engage properly with the programme, they indicated that they needed to have recovered from their worst symptoms. Parents also valued the flexibility of holding sessions within their homes or within clinics.

> *"We did it during the summer and it was ideal, we walked to it* [clinic] *through the park and went for coffee afterwards, just the whole experience of going was great for the family."* (PMI 4)
>
> *"This time last year I wasn't feeling well so I was able to focus on it this time."* (PMI 9)

Theme 2: Key Barriers to Participation

Key barriers to participation occurred during one of three phases, each of which was identified here as a subtheme including: (a) initial engagement; (b) attending the intervention; and (c) the concluding phase. An overview of challenges to participation can be seen in **Figure 3**.

Initial Engagement Phase

The most common barrier for families attending FT was parental attitudes/beliefs and stigma around mental illness. Parents were hesitant about participating for the following reasons: they felt stigma and shame in openly discussing their mental health challenges in the family context; they believed they were protecting children from the impact of their illness by not discussing it; they feared what their children would say; and a few were not ready to acknowledge that their illness might affect their children. Families required extensive preparatory input from clinicians to allay their fears to persuade them to engage with FT.

> *"The hardest part was actually admitting to him* [the child] *that I suffer with mental health problems. I hid away because for a long time, I have suffered with the stigma that goes with it."* (PMI 15)
>
> *"There's a very big stigma there. To make it easier to get through to him* [partner], *I think having someone else outside* [clinician]. *Through that, I felt a lot safer to try Family Talk."* (PMI 12)

Several children also highlighted their reservations about attending FT in terms of not understanding its purpose, fearing the focus would be on their wrongdoings (perceived or otherwise), and distrusting the viability of open discussion with typically uncommunicative parents. A couple of parents admitted that they had deliberately not clearly explained the purpose of FT to their children, for fear they would not attend.

> *"I would have preferred a bit more information. I didn't know what to expect and when we went in to speak with the girl who was doing it, I didn't know if the questions were going to be directed at us or about our parents. I was getting agitated because I was confused."* (Eldest child, 15 years old)

> *"I wasn't clear what it was about so I didn't know what to say."* (Middle child, 13 years old)

In addition, service constraints (e.g., staff turnover, uncovered maternity leave) disrupted/delayed FT delivery which meant that some families had an FT clinician with whom they were unfamiliar, meaning that additional time was needed to build rapport and trust before starting the intervention.

> *"I had never met her* [FT clinician] *so I think if maybe we had one or two sessions before just to get her to know a little bit better."* (Middle child, 17 years old)

Four of the interviewed families did not start FT due to family challenges and service constraint issues including: relapse in parental mental health; dealing with family crises (e.g., sexual assault, facing homelessness); deciding not to inform children about FT; being discharged from CAMHS before they could start FT; and suspension of FT delivery due to COVID-19 lockdown restrictions. It was interesting to note that discharge from CAMHS was cited as a reason for non-engagement because this reflects the lack of managerial priority given to family work and a perception by some CAMHS clinicians that working with parents is outside their service remit.

> *"We really wanted to do Family Talk but our daughter told us she was sexually assaulted so it wasn't the right time."* (PMI 20)

> *"We were meant to go to it* [FT] *but then we were discharged* [from CAMHS] *and that was the end of it."* (PMI 18)

Intervention Phase

The findings suggest that FT was challenging for many families despite the non-judgemental support provided by FT clinicians. Several parents/partners reported difficulties in speaking openly in sessions and/or listening to the experiences of family members, although most ultimately felt that it was worthwhile because it improved family communication. Partners and parents were shocked by their children's previously undisclosed revelations, what children had observed, and the internal narratives being used by children to make sense of the family situation (e.g., "Mum has cancer," "I made Dad ill"). There was also some unease that clinicians might "selectively" reveal what children had disclosed in the child session, leaving parents feeling uninformed. Furthermore, a couple of service-user parents believed that their partners had undiagnosed mental illness, and consequently were unhappy that the focus was primarily on their own mental health challenges.

> *"It was an eye opener. It was upsetting at the time, because to hear your child say certain things, it's really upsetting. But upsetting in a good way."* (Partner 4)

> *"We think we're protecting and sheltering them and, in the meantime, we're doing more damage."* (Partner 6)

Similarly, most children (10/15) reported emotional challenges in engaging with FT. For instance, most children had no prior relationship with the FT clinician and some perceived them as asking too many personal questions too quickly, without allowing time for trust and rapport to build. Four children said that they felt shy and/or embarrassed when answering clinician's questions. A couple of children also indicated that they did not want to answer questions as their parent had not discussed their mental illness with them before the FT child session. Another child left the individual session and informed his mother that he had not revealed anything because the school encourages pupils "not to talk to strangers." A few children found the family session emotionally volatile but ultimately helpful, while one teenager was initially upset in learning details of their parent's illness. Despite these challenges, the skill of clinicians in engaging children (e.g., allowing time to adjust, facilitating their voice being heard in the family session) had helped build a safe space for all but two to open-up and to engage with the process.

> *"Sometimes she'd ask personal questions that I wouldn't feel comfortable answering...My mouth goes shut."* (Middle child, 10 years old)

> *"I was just sitting there quietly not really talking or saying anything. I felt completely thrown under the bus. I wasn't surprised. I would have liked more information."* (Eldest, 16 years old)

> *"It was kind of emotional but then it got helpful and pleasurable. We got to say what we wanted to say. Dad has become more open. He shows his emotions now. He used to bottle them up a lot before."* (Only child, 12 years old)

Interestingly, within four families, there were widely varying experiences of the perceived utility of FT. In three families, both children and partners reported considerable benefits but the PMI did not. One PMI said that she "did not want to hear what others [in her family] were saying and blanked out," whilst another was wary of discussing mental health with his children in terms of diagnostic labels and believed the intervention, particularly with the children, was of insufficient duration and should have been delivered years earlier. Both of these parents had severe/enduring mental illness, were currently feeling very unwell, and had a history of being unhappy with mental health service provision. In the fourth family, FT was delivered in the morning when the PMI was heavily medicated, thereby limiting her level of engagement. Furthermore, while the child reported many benefits, both the PMI and her partner found it less helpful. The PMI was disappointed that FT had not focused on her daughter's mental health difficulties or her husband's "control issues," while her partner said he found it difficult to share his concerns as he believed it would aggravate his wife's emotional instability. These varied experiences highlight that each family member presents with a unique history and motivations and can present a range of challenges for clinicians when considering a family's readiness for FT.

> *"Family Talk might not have helped Mum as much but it helped us."* (Eldest, 18 years old)
>
> *"I just don't think we got a whole lot from it. It is very one sided to be honest... when an issue did come up, if there was something with regards to myself or my husband, they just constantly kept bringing it back to "Well, how does that affect* [child]*?"* (PMI 10)
>
> "[The PMI] *was getting so emotional because of her own opinions about things and stuff...I wasn't going to start dumping my own out there because it could have got messy and emotional. I didn't want to escalate any kind of like emotions. It was emotional enough. I was just kind of dealing with what was being brought up by* [partner] *and* [child]*."* (Partner 1)

The COVID-19 pandemic restrictions have been shown to have had a considerable impact on population mental health and on those with pre-existing mental illness, both in Ireland and internationally (41–44). Seven families in this study were interviewed during the COVID-19 emergency, with three reporting sustained benefits from FT and that they were coping well with pandemic stresses, while four families reported increased mental distress and challenging child misbehaviour as a result of the restrictions; two of these families had disengaged from FT due to stigma/relapse issues and two indicated that FT delivery had been suspended due to the restrictions. Therefore, it appeared that the level of prior vulnerability and ability to engage with FT predicted how well families had coped with the stresses of the COVID-19 restrictions. In addition, one parent reported attending online sessions of FT for PMIs, partners, and older teenagers (16+) but these were not considered suitable for younger children or for family sessions and they had to wait until it could be delivered safely again in person and in line with COVID-guidelines.

> *"I don't think we could have dealt with months of isolation if we hadn't done FT. We make time for each other now at this stage. We watch family films or to sit down for dinner, meals."* (PMI 13)
>
> *"It wasn't the same but we were able to talk with him [clinician] on zoom. It was a while before the children could be seen so it wasn't ideal."* (PMI 5)

Disengaging From FT

Families who disengaged from FT after three or fewer sessions ($n = 5$) gave the following reasons. One said that FT was too emotionally upsetting, with another feeling a sense of blame for causing her children's mental health issues. A number of other factors also contributed to disengagement including family crisis, relapse in symptoms, and having too many competing priorities. Additional delays/disruption in FT delivery due to the COVID-19 restrictions also led to some degree of disillusionment and disengagement from mental health services. This was more common in areas where mental health clinicians were partially redeployed to frontline COVID-19 duties and could only provide minimal telephone support to service users (41). Interestingly, those who disengaged from FT were almost twice as likely as "completers" to be lone parents (6/9 vs. 5/14)–suggesting that the stresses of lone parenting may also have been a barrier to engagement.

> *"With covid, we are far less a priority for them. I don't know when or if we're ever going to get it."* (PMI 22)
>
> *"It felt like she was attacking me and it was my fault how the girls are...I don't need that."* (PMI 19)

Concluding Phase

Despite benefiting from FT, most attendee families ($n = 12$) found the programme to be too short and expressed a desire for more child, family and follow-up sessions to build family communication. Families had high expectations of FT, which appeared to be linked to their need for more (often unavailable) psychological and family support from mental health services. Three families reported that they were referred to further mental health supports (e.g., dialectical behaviour therapy), while two others were given a list of alternative supports including national mental health charities. Most, typically, parents reported that FT clinicians provided closure by affirming their availability if future issues arose but the lack of follow-up was problematic for some partners who were not offered additional support and who were unable to pay for private treatment outside of statutory service provision. However, it is important to note that three attendee families were interviewed during the first COVID-19 lockdown which severely limited their access to mental health and community services and to other social/family supports at that time. Nonetheless, there were numerous indications that this population would likely benefit from longer-term (family-focused) mental health support.

> *"It didn't feel like* [it] *was ready to be finished."* (Partner 3)
>
> *"I think more sessions with the family...and more time with the children would have really helped. The three of them went in one by one for 20 min. So it might have been a little bit rushed for them, they might not have had enough time."* (PMI 16)
>
> *"Family Talk is minimal... A taster...I think the hospital might have family therapy...But on a private basis so..."* (PMI 2)

Discussion

This study is the first qualitative analysis of family experiences of FT conducted outside Sweden, the first situated within the context of an RCT of FT, and as part of the first nationwide endeavour to introduce FFP to adult and child mental health services in the RoI. The qualitative findings, in line with those reported in Swedish psychiatric settings (32–37), indicated many benefits for families who attended FT, including: reduced worry and stigma; a greater understanding of mental illness; giving children and partners a voice; improved parental confidence and family communication; and warmer relationships. Notably, the current study placed a greater emphasis on the caregiving, parental and financial stresses experienced by partners, all of whom for the first time had a forum in which to have their burdens validated, and a space in which to develop a more constructive team-based approach to supporting the PMI and the family unit. In addition, unlike previous qualitative studies, this study reported on the experiences of families who refused to attend or disengaged from the programme.

Importantly, the findings reported here, indicate that FT is acceptable and beneficial for families across different cultural/policy contexts, mental health settings, types of mental disorders, and among children with and without existing mental health challenges. Firstly, unlike Scandinavian countries where legislation has been introduced to safeguard children of PMI and where FT is implemented on a national level, the RoI lacks any "think family" policy/practise guidance for this population whilst service and public awareness of the need to support this population is erratic/unsystematic. There are also continuing high levels of mental health stigma in Ireland, which is an important barrier to help seeking (6, 45, 46). Given the challenges have been reported within Scandinavian (and other) countries in terms of translating family-focused legislation/policy into practise (22, 47), it was reassuring to find that FT was perceived as helpful by the vast majority of the families in this study, thereby highlighting the need for, and value of, such FFP supports for families where there is PMI.

Secondly, even though AMHS may appear the most natural fit for FT/FFP and provide a common context for the implementation of FFPs (45, 48, 49), families in this study reported benefits across both AMHS and CAMHS settings. FT is typically delivered as a preventive intervention to families whose children do not attend mental health services (14, 15) but in this study, most of the child participants, including those who attended CAMHS, reported that FT had improved their well-being and family relationships. Therefore, FFPs such as FT may also be helpful for children with existing mental health challenges, as well as promoting collaboration between adult and child mental health services, and increasing the identification of families through a "no wrong door" approach to family access, as promoted in the "Think Family" model in Northern Ireland (50).

Thirdly, there did not appear to be any notable variations across family experiences here in terms of the PMI diagnosis, thereby highlighting the suitability of FT for disorders beyond parental depression, which was the original focus of the programme (13, 14). Whilst a small number of adult service users who attended all FT sessions felt that it had not been helpful, their children/partners, and other service users with similar disorders, reported a range of benefits. Similarly, two PMIs, suffering from depression and Borderline Personality Disorder respectively, indicated that they disengaged from FT after two to three sessions as they felt blamed/upset by the idea that their mental illness may affect their children. Previous qualitative studies have indicated that while FT may work across a range of disorders, those with Borderline Personality Disorder or low-functioning psychosis are more likely to struggle with establishing a therapeutic alliance and/or exhibit a lack of understanding/insight into the impact of their mental illness on their children (34, 37). Low functioning service users may possibly require additional psycho-educational sessions and/or complementary groups for patients and children, in order to share experiences and learn about their mental illness and its impact on their children (37).

The findings reported here also highlight a number of important facilitators and barriers to engagement, which should help inform the future implementation of FT/FFPs and could be tested as mediators/moderators of RCT outcomes. Key facilitators included timeliness, clinician attributes and expertise, and involvement of the whole family. The clinician's role was key in: providing a setting for parent, partner and child voices to be heard and validated; normalising the family's lived experience; reducing fear and stigma through psycho-education; and teaching a strengths-based, problem-solving approach to improve family communication and interactions. Previous qualitative studies of FT likewise, highlight the value of clinician attributes of confidence, competence, warmth and non-judgmentalism in contributing to better family experiences (33, 34), whereas conversely, a perception that clinicians lack competency has been linked to disengagement (Unpublished data, Preventive Intervention Project, 2013, https://fampod.org/file.php/1/collaborations/Columbia_Application_of_PIP.pdf). An interesting study by Marston et al. showed that when parents were given a family-focused DVD, but did not receive the support of a clinician, their understanding of the impact of their illness improved, but they did have the confidence to open up a dialogue about the illness with their children (51). Thus, the presence of a trained clinician with appropriate attributes is a critical element of FT (and other FFPs) in terms of engaging families and potentially producing more positive outcomes.

Families particularly valued the whole family approach. As noted in previous qualitative studies, they felt that this had enabled all perspectives to be surfaced and heard, and improved mutual understanding and family relationships (32–34). Nevertheless, whole-family programmes appear to be less commonly delivered than parent-only models (48, 49), most probably due to the additional buy-in and logistics required in involving children. Our findings suggest that without the involvement of the child(ren), parents may not become aware of their worries and burdens as demonstrated here by the surprise and alarm that parents reported when their children opened up during the FT sessions. In addition, within parent-only interventions, parents may not be sufficiently supported to find the right words to have a conversation with their children, and may also lack a forum to realise just how much their children/partners want to support them, but have been prevented from so doing by the silence and stigma associated with discussing mental illness within the home. Two RCTs which have compared FT with the parent-only *Let's Talk about the Children* found FT to be more effective in reducing child emotional symptoms and improving the parent–child relationship (13, 15).

Similarly, child programmes that exclude parents may reduce the likelihood of mutual understanding as parents lack the opportunity to discuss their stigma concerns, to gain insight into the impact of their mental illness on their children, and/or to build parental confidence. Enhancing parental confidence and competence has been identified as key to shaping the quality of parent-child relationships (52). Significantly, the involvement of partners is less common in the implementation of FFPs (3). The findings from the current study indicated that FT provided a forum, usually for the first time, for partners to express their burdens and stresses and to communicate with the PMI about how they can better support them. Given the level of burden and stress reported, and the protective boundary provided by a healthy second parent (2), it is imperative that services/FFPs help to strengthen the "safety net" that co-parents provide in families with PMI. Recent filicide tragedies in Ireland (e.g., McGinley case) highlight the ultimate cost of not involving family members in the service user's treatment (53).

Significant barriers to participation were also reported in the current study. Most family members, including both completers and drop-outs, indicated that they had experienced challenges in engaging with FT in the initial phase. Parental fears and stigma around mental illness were the most commonly reported barriers to participation. Children also expressed reservations about attending, indicating that they were uninformed about the purpose of FT and wanted prior contact with the clinician prior to commencing FT sessions. Other family barriers to engagement were also noted, including relapse in symptoms and family crises. Previous qualitative studies have similarly highlighted that fear of judgement and/or competing needs for daily survival may impede family engagement (32, 37). These findings suggest that clinicians may benefit from the development of FT/FFP training videos/protocols to promote effective engagement strategies and address potential barriers to participation and retention. For instance, addressing issues of stigma, readiness/timeliness, consent and confidentiality during the recruitment process and including quotes/videos from previous FT attendees may help to improve engagement (54). In addition, a child-friendly recruitment approach that used age-appropriate marketing literature and involved a meet-and-greet session with the facilitating clinician might help to address children's concerns about attending. Lastly, low functioning PMIs may benefit from additional psycho-educational sessions and complementary group supports to promote engagement (37).

Service constraints were also an inhibitive factor in family engagement with the programme. The capacity of FT clinicians to build rapport and familiarity with the family beforehand was undermined by high turnover of personnel and under-resourced mental health teams. In addition, a small number of families were discharged from AMHS/CAMHS before they could start FT, while several other families disengaged due to their unhappiness with long waiting lists, and delays/disruptions due to the COVID-19 lockdown restrictions. These difficulties reflect general underfunding of mental health services in the RoI, alongside a lack of policy/practise priority given to supporting this population in an Irish context (26, 45, 55).

Some challenges were also noted during the intervention phase. Firstly, while most families reported that FT was ultimately worthwhile, it was also seen as emotionally challenging at times. Many reported difficulties in speaking openly in sessions and/or listening to other's experiences and indeed, this was also shown in research by Pihkala et al. (33) in Sweden. The clinician's skill in facilitating multiple perspectives was instrumental in ensuring that family members could listen to each other without becoming overly defensive or upset. Secondly, there was some evidence that children within two families did not receive sufficient time in their individual child session (e.g., 20 min each). Moreover, while children largely reported benefits from FT, there was little mention of fun within sessions. Therefore, children may benefit from the inclusion of some light relief at the beginning or close of sessions (e.g., ice breakers, child-friendly videos, closing "fun" take-home exercise), as used in, for example, the Kidstime intervention (56). Thirdly, the COVID-19 lockdown restrictions had implications for the delivery of FT, including blended adaptation (both in-person and online sessions), as well as family disengagement following repeated delays to delivery. Notably, there was considerable variation in the capacity of sites to deliver FT during the lockdowns with some mental health staff partially redeployed to frontline COVID-19 duties and providing minimal phone support to patients while clinicians in other areas were able to continue home visits and outpatient clinics, following COVID-19 guidelines (41). Reassuringly however, it is likely that the future implementation of FT will be conducted in person in view of the >90% uptake of vaccination in the RoI (57).

Lastly, while most families benefitted from FT, they indicated a desire for additional child, family and follow-up sessions, thereby suggesting that some of their needs had not been met. This was also noted by FT clinicians working with lower functioning psychosis patients in Sweden (37), although it was not reported by families experiencing depression (32–35). Where possible, the concluding phase of FT should signpost families to additional family and mental health supports as required. Given the complexity of service user needs, a flexible spectrum of family-focused services may be necessary, as demonstrated internationally (58), although this level of family resources is not currently available in Ireland (6).

Strengths and Limitations of the Study

This study is the first qualitative analysis of family experiences of FT conducted outside Sweden and the first conducted within the context of an RCT and national programme to introduce FFP for families with PMI, in this case within the RoI. Service-user parents are typical of those who take part in RCTs and qualitative studies of FFPs (28), but the current study involved the recruitment of a large and diverse sample (in qualitative terms) of both child and adult participants, including PMIs, partners, children, and "drop-out" families. In addition, our sample was recruited from a number of mainstream adult and child mental health services and encompassed a variety of mental disorders as well as including both children who were and were not attending CAMHS. The analysis yields further important insights into the barriers and facilitators of implementation, as perceived by families, and will help to support and amplify the clinician experiences of FT which are reported here in a companion paper (as well as the RCT results when they become available).

The study was limited in a number of ways, including firstly, the transferability of the findings across different cultural contexts. However, the description of the study context should help in this regard. It is also possible that the findings may be biassed in that families who agreed to be interviewed had a more positive experience of FT, and in a small number of instances, we believe that gatekeeping from the PMI may have potentially excluded feedback from other family members who were invited to participate in the research. Importantly though, we interviewed nine families who had disengaged from FT after three or fewer sessions as well as families who had completed FT. In addition, the interval between FT and the child interviews (ranging from 3 to 5 months) created recall difficulties for three of the younger children in our sample, although the remainder ($n =$ 12) had much to say about their involvement. This interval was necessary due to blinding in the RCT which had to be retained until after the 6 month assessment had been completed. Lastly, 7 of the 23 families were interviewed during the first COVID-19 lockdown, which severely restricted access to services and led to increased levels of psychological distress in the general population in Ireland (42), both of which may have impacted their experiences and views.

Implications for Policy, Practise and Research

Our findings highlight the value of a whole family approach when a parent has mental health challenges, particularly in revealing the hidden burdens that children carry, reducing fears and stigma, and improving empathy and communication among parents and children. The findings illustrate that FT can be successfully implemented across adult and child mental health settings and with families experiencing different mental disorders, thereby reflecting, at least to some extent, a "no wrong door" approach to identifying and supporting families. Key facilitators to implementation included delivery by a competent, non-judgmental clinician and family readiness to participate. The primary barrier to FT implementation was recruiting and engaging with families in the initial phase due, in large part, to family challenges and service constraints. Engagement may be improved if clinicians address issues of stigma, readiness, consent and confidentiality during the recruitment process and use quotes/videos from previous FT attendees. In addition, children's concerns about attending could be addressed using age-appropriate marketing literature and an initial meet-and-greet session with the facilitating clinician.

Our findings also suggest that FT may not be suitable/sufficient for all families (e.g., low functioning service users) and should, ideally, be implemented as part of a suite of lower and higher intensity FFPs (58). There is an urgent need in the context of the RoI, to introduce "think family" practise guidelines and to provide dedicated funding to develop a multi-level, public-health response to identifying and supporting these families, as has been done in, for example, Scandinavia and Australia (33, 47). Under the United Nations Convention on the Rights of the Child (20), children have a right to a childhood and not be used as unpaid/unsupported carers filling gaps in service provision. Moreover, when child welfare is not considered in the treatment of service-user parents, it increases their risk of developing mental disorders and becoming the next generation of service users, and, in the most tragic (but thankfully rare) cases, can lead to their death by filicide (53). Internationally, systemic barriers to change need to be addressed, including mandatory auditing of the parenting status of adult mental health users, balancing the priority given to patient confidentiality with unmet family needs, increased collaboration between traditionally segregated AMHS and CAMHS services, and equipping clinicians with time and resources to undertake FFP.

Further qualitative and quantitative research on family and clinician experiences of FT implementation is required across different cultural/policy contexts, mental health and family settings, types of mental disorders and level of child mental health difficulties. Further research is also needed on the types of families that are more likely not to engage with FT, and to identify measures and/or supports that might increase engagement. For instance, there may be value in developing and evaluating training videos that teach recruitment strategies to see whether they improve engagement. In addition, qualitative analyses may inform RCT evaluations of FT/FFPs; for instance, RCTs could include as outcome measures, benefits identified in qualitative analyses, such as reduction in stigma, parental confidence/competence, service-user mental health, partner well-being, and family functioning. Moreover, facilitators and barriers to implementation identified in qualitative studies could be tested as moderator/mediator variables in quantitative research.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because where participants provided consent/assent, an anonymised version of their data will be stored in the Irish Qualitative Data Archive. Ethical approval from an approved higher level institution will be required if the data is to be used in future research. Requests to access the datasets should be directed to Mairead.Furlong@mu.ie.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by four ethics committees: the Social Research Ethics Committee in Maynooth University, Ireland (Reference number SRESC- 2018-100), the HSE Research Ethics Committee, Tusla Ethics Review Committee, and the Saint John of God's Research Ethics Committee. Parents/legal guardians provided written informed consent for their children to participate in the study, followed by their children providing written informed assent to participate.

AUTHOR CONTRIBUTIONS

CM, MF, and SMcGi conceived and designed the study. CM conducted interviews and coded transcripts, with 25% of transcripts independently coded by MF, SMcGa, and SO'C. CM prepared the initial draft, with subsequent drafts undertaken by MF, with input from SMcGi, CM, and SMcGa. All authors contributed to the article and approved the submitted version

FUNDING

The PRIMERA research programme is funded by the Health Service Executive and the Maynooth University Higher Education Authority COVID-19 Costed Extension Fund.

ACKNOWLEDGMENTS

We are very grateful to all the families who generously gave their time to participate in this study. We also owe special thanks to all of the dedicated clinicians and managers who agreed to participate in this research and to introduce and deliver Family Talk within their settings, including the Health Service Executive, Saint John of God Hospitaller services, and Tusla child protection agency.

REFERENCES

1. Maybery D, Reupert A, Patrick K. Prevalence of children whose parents have a mental illness. *Psychiatr Bull.* (2009) 33:22–6. doi: 10.1192/pb.bp.107.018861
2. Hosman CMM, van Doesum KMT, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. the scientific basis to a comprehensive approach. *Aust e-J Adv Ment Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
3. Afzelius M, Östman M, Råstam M, Priebe G. Parents in adult psychiatric care and their children: a call for more interagency collaboration with social services and child and adolescent psychiatry. *Nord J Psychiatry.* (2018) 72:31–8. doi: 10.1080/08039488.2017.1377287
4. OECD/EU. *Health at a glance: Europe 2018: State of Health in the EU cycle.* (2018) (accessed September 5, 2021).
5. Central Statistics Office. *Census of Population 2016-Profile 4 Households and Families*. (2016). Available online at: https://www.cso.ie/en/releasesandpublications/ep/p-cp4hf/cp4hf/fmls/ (accessed September 5, 2021).
6. Mulligan C, Furlong M, McGilloway S. Promoting and implementing family-focused interventions for families with parental mental illness: scoping and installation. *Adv Ment Health.* (2020) 18:202–16. doi: 10.1080/18387357.2019.1614466
7. Jones M, Pietilä I, Joronen K, Simpson W, Gray S, Kaunonen M. Parents with mental illness–a qualitative study of identities and experiences with support services. *J Psychiatr Ment Health Nurs.* (2016) 23:471–8. doi: 10.1111/jpm.12321
8. Siegenthaler E, Munder T, Egger M. Effects of preventive interventions in mentally ill parents on the mental health of the mental health of offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:817. doi: 10.1016/j.jaac.2011.10.018
9. Nicholson J, de Girolamo G, Schrank B, editors. *Parents with Mental and/or Substance Use Disorders and Their Children*. Lausanne: *Frontiers Media SA* (2020).
10. Bee P, Bower P, Byford S, Churchill R, Calam R, Stallard P, et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. *Health Technol Assess.* (2014) 18:1–250. doi: 10.3310/hta18080
11. Reupert A, Cuff R, Drost L, Foster K, van Doesum KTM, van Santvoort F. Intervention programs for children whose parents have a mental illness: a review. *MJA Open.* (2012) 1:18–22. doi: 10.5694/mjao11.11145
12. Beardslee WR, Wright E, Rothberg PC, Salt P, Versage E. Response of families to two preventive intervention strategies: long-term differences in behavior and attitude change. *J Am Acad Child Adolesc Psychiatry.* (1996) 35:774–82. doi: 10.1097/00004583-199606000-00017
13. Beardslee WR, Salt P, Versage EM, Gladstone TRG, Wright EJ, Rothberg PC. Sustained change in parents receiving preventive interventions for families with depression. *Am J Psychiatry.* (1997) 154:510–5. doi: 10.1176/ajp.154.4.510
14. Beardslee WR, Wright EJ, Gladstone TR, Forbes P. Long term effects from a randomized trial of two public health preventive interventions for parental depression. *J Fam Psychol.* (2007) 21:703–13. doi: 10.1037/0893-3200.21.4.703
15. Solantaus T, Paavvonen EJ, Toikka S, Punamaki RL. Preventive interventions in families with parental depression. *Eur Child Adolesc Psychiatry.* (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
16. Christiansen H, Anding J, Schrott B, Röhrle B. Children of mentally ill parents—a pilot study of a group intervention program. *Front Psychol.* (2015) 6:1494. doi: 10.3389/fpsyg.2015.01494
17. Punamäki RL, Paavonen J, Toikka S, Solantaus T. Effectiveness of preventive family intervention in improving cognitive attributions among children of depressed parents: a randomized study. *J Fam Psychol.* (2013) 27:683. doi: 10.1037/a0033466
18. Compas BE, Forehand R, Keller G, Champion JE, Rakow A, Reeslund KL, et al. Randomized controlled trial of a family cognitive-behavioral preventive intervention for children of depressed parents. *J Consult Clin Psychol.* (2009) 77:1007. doi: 10.1037/a0016930
19. Beardslee WR, Solantaus TS, Morgan BS, Gladstone TR, Kowalenko NM. Preventive interventions for children of parents with depression: international perspectives. *Med J Aust.* (2013) 199:23–5. doi: 10.5694/mjao11.11289
20. United Nations Treaty Series. *United Nations convention on the rights of the child. Treaty no. 27531.* (1989) 1577. p. 3–178. Available online at: https://treaties.un.org/doc/Treaties/1990/09/19900902%2003-14%20AM/Ch_IV_11p.pdf (accessed September 5, 2021).
21. Swedish Health and Medical Service Act, SFS. (2010:662). *Requirements on Health Care. Stockholm: Ministry of Health*. Available online at: https://www.riksdagen.se/sv/dokument-lagar/dokument/svensk-forfattningssamling/halso--ochsjukvardslag-1982763_sfs-1982-763 (accessed September 5, 2021).
22. Lauritzen C, Reedtz C, Van Doesum KT, Martinussen M. Implementing new routines in adult mental health care to identify and support children of mentally ill parents. *BMC Heal Ser Res.* (2014) 14:58. doi: 10.1186/1472-6963-14-58
23. Health Service Executive. *BFT National clinical programme.* (2019). Available online at: https://www.hse.ie/eng/about/who/cspd/ncps/mental-health/psychosis/resources/hse-early-intervention-in-psychosis-model-of-care-executive-summary-june-2019.pdf (accessed September 5, 2021).
24. Health Service Executive. *National Framework for Recovery in Mental Health.* (2017). Available online at: https://www.hse.ie/eng/services/list/4/mental-health-services/advancingrecoveryireland/national-framework-for-recovery-in-mental-health/ (accessed September 5, 2021).
25. Health Service Executive. *Family, carer, and supporter guide*. (2020). Available online at: https://www.hse.ie/eng/services/list/4/mental-health-services/mentalhealthengagement/family-carer-and-supporter-guide/ (accessed September 23, 2021).
26. Furlong M, McGilloway S, Mulligan C. McGuinness, C, Whelan, N. Family talk versus usual services in improving child and family psychosocial functioning in families with parental mental illness (PRIMERA—Promoting Research and Innovation in Mental hEalth seRvices for fAmilies and children): study protocol for a randomised controlled trial. *Trials.* (2021) 22:243. doi: 10.1186/s13063-021-05199-4
27. Moore GF, Audrey S, Barker M, Bond L, Bonell C, Hardeman W, et al.: Process evaluation of complex interventions. UK *Medical Research Council (MRC) guidance. BMJ.* (2014) 350:h1258. doi: 10.1136/bmj.h1258
28. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
29. Maybery D, Ling L, Szakacs E, Reupert AE. Children of a parent with a mental illness perspectives on need. *Advanc Ment Health.* (2005) 4:78–88. doi: 10.5172/jamh.4.2.78

30. Oltedal S, Garratt A, Johannessen JO. Psychiatric outpatients' experiences with specialized health care delivery. a Norwegian national survey. *J Ment Health.* (2007) 16:271–9. doi: 10.1080/09638230701279832
31. Bateson K, Darwin Z, Galdas P, Rosan C. Engaging fathers: acknowledging the barriers. *J Health Visiting.* (2017) 5:126–32. doi: 10.12968/johv.2017.5.3.126
32. Pihkala H, Johansson EE. Longing and fearing for dialogue with children—depressed parents' way into beardslee's preventive family intervention. *Nord J Psychiatry.* (2008) 62:399–404. doi: 10.1080/08039480801984800
33. Pihkala H, Sandlund M, Cederström A. Children in beardslee's family intervention: relieved by understanding of parental mental illness. *Int J Soc Psychiatry.* (2012) 58:623–8. doi: 10.1177/0020764011419055
34. Pihkala H, Sandlund M, Cederström A. Initiating communication about parental mental illness in families: an issue of confidence and security. *Int J Soc Psychiatry.* (2012) 58:258–65. doi: 10.1177/0020764010392088
35. Pihkala H, Dimova-Bränström N, Sandlund M. Talking about parental substance abuse with children: eight families' experiences of Beardslee's family intervention. *Nord J psychiatry.* (2017) 71:395–401. doi: 10.1080/08039488.2017.1308009
36. Strand J, Boström P, Grip K. Parents' descriptions of how their psychosis affects parenting. *J Child Fam Stud.* (2020) 29:620–31. doi: 10.1007/s10826-019-01605-3
37. Strand J, Rudolfsson L. A qualitative evaluation of professionals' experiences of conducting Beardslee's family intervention in families with parental psychosis. *Int J Ment Health Promotion.* (2017) 19:289–300. doi: 10.1080/14623730.2017.1345690
38. VERBI Software. *MAXQDA 2020*. Software. (2019). Available online at: www.maxqda.com (accessed March 30, 2020).
39. Charmaz K. *Constructing Grounded Theory: A practical Guide Through Qualitative Analysis*. London: Sage Publications (2006).
40. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care.* (2007) 19:349–57. doi: 10.1093/intqhc/mzm042
41. Furlong M, McGilloway S, Mulligan C, Killion MG, McGarr S, Grant A, et al. Covid-19 and Families With Parental Mental Illness: Crisis and Opportunity. *Front Psychiatry.* (2021) 12:567447. doi: 10.3389/fpsyt.2021.567447
42. Kelly BD. Impact of Covid-19 on mental health in Ireland: evidence to date. *Ir Med J.* (2020) 113:214. Available online at: http://imj.ie/wp-content/uploads/2020/12/Impact-of-Covid-19-on-Mental-Health-in-Ireland-Evidence-to-Date.pdf (accessed September 23, 2021).
43. Mental Health Foundation. *The COVID-19 pandemic, Financial Inequality and Mental Health (*2020). Available online at: https://www.mentalhealth.org.uk/our-work/research/coronavirus-mental-health-pandemic/covid-19-inequality-briefing (accessed September 5, 2021).
44. Quittkat HL, Dusing R, Holtmann FJ, Buhlmann U, Svaldi J, Vocks S. Perceived impact of Covid-19 across different mental disorders: a study on disorder-specific symptoms, psychosocial stress and behavior. *Front Psychol.* (2020) 11:586246. doi: 10.3389/fpsyg.2020.586246
45. Grant A, Goodyear M, Maybery D, Reupert A. Differences between Irish and Australian psychiatric nurses' family-focused practice in adult mental health services. *Arch Psychiatr Nurs.* (2016) 30:132–7. doi: 10.1016/j.apnu.2015.07.005
46. St Patrick's Mental Health Services. *Attitudes to Mental Health Annual Survey*. (2020). Available online at: https://www.stpatricks.ie/media/2941/attitudes-to-mental-health-survey-findings-2020-st-patricks-mental-health-services.pdf (accessed September 5, 2021).
47. Falkov A, Goodyear M, Hosman CM, Biebel K, Skogøy BE, Kowalenko N, et al. A systems approach to enhance global efforts to implement family-focused mental health interventions. *Child Youth Serv.* (2016) 37:175–93. doi: 10.1080/0145935X.2016.1104104
48. Maybery D, Goodyear M, Reupert A, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry.* (2017) 17:1–7. doi: 10.1186/s12888-017-1357-4
49. Allchin B, O'Hanlon B, Weimand BM, Boyer F, Cripps G, Gill L, et al. An explanatory model of factors enabling sustainability of Let's Talk in an adult mental health service: a participatory case study. *Int J Ment Health Syst.* (2020) 14:1–6. doi: 10.1186/s13033-020-00380-9
50. Grant A, Lagson S, Devaney J, Davidson G, Duffy J, Perra O, et al. *A Study of Health and Social Care Professionals' Family Focused Practice With Parents Who Have Mental Illness, Their Children and Families in Northern Ireland*. (2018). Available online at: https://pureadmin.qub.ac.uk/ws/portalfiles/portal/154075911/Think_Family_Final_Long_Report_27.2.18_Final_Final.pdf (accessed September 5, 2021).
51. Marston N, Maybery D, Reupert A. Empowering families where a parent has a mental illness: a preliminary evaluation of the 'family focus' DVD. *Adv Ment Health.* (2014) 12:136–46. doi: 10.1080/18374905.2014.11081891
52. Kamis C. The long-term impact of parental mental health on children's distress trajectories in adulthood. *Soc Ment Health.* (2021) 11:54–68. doi: 10.1177/2156869320912520
53. Wayman S. *Mental Illness: Where Should Family Involvement Begin and End When a Parent Needs Treatment*? (2021). Available online at: https://www.irishtimes.com/life-and-style/health-family/parenting/mental-illness-where-should-family-involvement-begin-and-end-when-a-parent-needs-treatment-1.4588412Irish~Times~15~June~2021 (accessed September 5, 2021).
54. Van Doesum KT, Riebschleger J, Carroll J, Grové C, Lauritzen C, Mordoch E, et al. Successful recruitment strategies for prevention programs targeting children of parents with mental health challenges: an international study. *Child Youth Serv*. (2016) 37:156–74. doi: 10.1080/0145935X.2016.1104075
55. Mental Health Reform. *Pre-budget Submission*. (2020). Available online at: https://www.mentalhealthreform.ie/wp-content/uploads/2019/07/Full-MHR-pre-budget-submission-2020_-FINAL.pdf (accessed September 5, 2021).
56. Cooklin A. "Mental health, mental illness, the family and others: How do we think about them together?" In: Cooklin A, Barnes GG, editors. *Building Children's Resilience in the Face of Parental Mental Illness*. London: Routledge (2020). p. 6–42.
57. Towey N. *Covid-19 Vaccine Tracker: How Many People in Ireland Have Been Inoculated in Ireland?* Available online at: https://www.irishtimes.com/news/health/covid-19-vaccine-tracker-how-many-people-have-been-inoculated-in-ireland-1.4481095#how%20many%20vaccinated~Irish~Times (accessed September 8, 2021).
58. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Family options for parents with mental illnesses: a developmental, mixed methods pilot study. *Psychiatr Rehabil J.* (2009) 33:106. doi: 10.2975/33.2.2009.106.114

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 23 November 2021
doi: 10.3389/fpsyt.2021.783161

A Family-Focused Intervention for Parental Mental Illness: A Practitioner Perspective

*Mairead Furlong**[†]*, Christine Mulligan*[†]*, Sharon McGarr, Siobhan O'Connor and Sinead McGilloway*

Centre for Mental Health and Community Research, Maynooth University, Maynooth, Ireland

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Christine Grové,
Monash University, Australia
Frances Louise Dark,
Metro South Addiction and Mental Health Services, Australia

Correspondence:
Mairead Furlong
mairead.furlong@mu.ie

[†]These authors have contributed equally to this work and share first authorship

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *25 September 2021*
Accepted: *01 November 2021*
Published: *23 November 2021*

Citation:
Furlong M, Mulligan C, McGarr S, O'Connor S and McGilloway S (2021) A Family-Focused Intervention for Parental Mental Illness: A Practitioner Perspective.
Front. Psychiatry 12:783161.
doi: 10.3389/fpsyt.2021.783161

Background: Parental mental illness (PMI) is common and can lead to children developing mental disorders. Family Talk (FT) is a well-known and widely implemented intervention designed to reduce the risk of transgenerational psychopathology. However, given the research to practise "gap," very little qualitative research, to date, has investigated practitioner experiences in implementing FT. This study aimed to explore the practitioner-perceived barriers and facilitators to the implementation and sustainability of FT within mainstream mental health settings.

Methods: This qualitative study was nested within a randomised controlled trial (RCT) of Family Talk [$N = 86$ families (139 parents, 221 children)] within 15 adult (AMHS), child (CAMHS), primary care mental health, and child protection sites in Ireland. Semi-structured interviews and focus groups were undertaken with a purposive sample of clinicians ($n = 31$) and managers ($n = 10$), based on their experiences of implementing FT. Interview data were transcribed verbatim, analysed using constructivist grounded theory, and informed by Fixsen's implementation science framework.

Results: Service providers highlighted a number of benefits for approximately two thirds of families across different diagnoses and mental health settings (AMHS/CAMHS/primary care). Sites varied in their capacity to embed FT, with key enablers identified as acquiring managerial and organisational support, building clinician skill, and establishing interagency collaboration. Implementation challenges included: recruitment difficulties, stresses in working with multiply-disadvantaged families, disruption in delivery due to the COVID-19 global pandemic, and sustainability concerns (e.g., perceived fit of FT with organisational remit/capacity, systemic and cultural barriers to change).

Conclusion: This study is only the second qualitative study ever conducted to explore practitioner experiences in implementing FT, and the first conducted within the context of an RCT and national research programme to introduce family-focused practise (FFP) for families living with PMI. The findings illuminate the successes and complexities of implementing FFP in a country without a "think family" infrastructure, whilst highlighting a number of important generalisable lessons for the implementation of FT, and other similar interventions, elsewhere.

Keywords: family talk, implementation, mental disorder, mental illness, parents, qualitative, COPMI, children

INTRODUCTION

Parental mental illness (PMI) is common, with 23% of all families having at least one parent who has, or had, a mental disorder (1), and a 41–77% lifetime risk for children of developing serious mental illness, physical illness, and impaired educational and occupational outcomes (2). Traditionally, both in Ireland and in other jurisdictions, these families have remained "invisible" and unsupported due to the segregation of adult and child mental health services (3, 4). Data on parenting status within mental health services is scarce (5), but early studies estimate that 25–68% of adult mental health service users are parents, and 35–60% of children presenting at child and adolescent mental health services have a parent with mental illness (6, 7).

Given the prevalence and burden of PMI–and in the context of the principles and values enshrined within the United Nations Convention on the Rights of the Child–there has been a growing recognition in many countries of the need to support families in order to protect children from developing mental disorders (8, 9). Reassuringly, a range of interventions has been developed (e.g.,targeting parents, children, whole family or peers), with evidence that they can decrease the risk of developing mental disorders for children by up to 40% and reduce referrals to child protection services (10, 11). Family Talk (FT), in particular, has been identified as a key intervention with promising evidence of effectiveness in improving parent and child understanding of, and communication about, mental illness and child internalising symptoms (9, 10, 12–14). FT is a whole-family, 7-session, manualised, clinician-facilitated, psycho-educational, and strengths-based approach designed to improve family communication and resilience (15), and has been implemented in recent years in several countries as part of national initiatives to support families where a parent has mental illness (e.g., the USA, Costa Rica, Colombia, the Netherlands, Greece, Scandinavia, Iceland, and Australia) (15).

Nevertheless, we know from the translation of other evidence-based psychosocial programmes that positive outcomes achieved in controlled research settings may not always be replicated within mainstream service settings (16, 17). According to Fixsen, the implementation of practise change typically involves a lengthy recursive process of six (non-linear) stages, including "exploration," "installation," "initial implementation," "full implementation," "innovation," and "sustainability," with each stage presenting its own unique set of challenges (18). Within the context of family-focused practise (FFP) for families with PMI—and including our own research–a number of implementation barriers have been noted, including: (1) the socio-political context (e.g., lack of policy/practise guidelines, dedicated funding); (2) organisational culture (e.g., siloed adult and child mental health services, ideological differences, under-resourced mental health teams); (3) clinician skill/attitudes (e.g., professional training typically based on a biomedical, crisis-oriented, individualised model of care); and (4) service user/families' willingness to participate (e.g., stigma, fear of losing custody, lack of awareness of impact of PMI on children) (5, 19–23). Research has found that implementation of FFP is erratic and unsystematic even within countries with established "think family" initiatives and legislation that mandates the identification and support of families with PMI (24–26). For instance, less than half of all clinicians in adult mental health services (AMHS) in Norway identified the parental status of service users despite acknowledging it to be a mandatory task (25), thereby indicating that changes in legislation or attitudes alone, do not necessarily lead to change in practise.

To date, only one published qualitative study of clinicians/managers' experiences in implementing FT has been conducted, despite FT being delivered as part of national initiatives in several countries (15). Eleven clinicians in Sweden were interviewed to explore their experiences of delivering FT to families living with parental psychosis. Several benefits were indicated, including increased family understanding of, and communication about PMI, and the utility of the FT manual in equipping clinicians to ask about patients' parenting capacity and children's well-being. Nevertheless, high rates of refusal and attrition were noted, and clinicians reported that some parents with psychosis lacked insight into the impact of their mental illness on their children. In addition, in a recent paper, the FT programme developer, William Beardslee, reported on his team's experience of delivering FT to parents with depression in the US and while this was not a qualitative paper involving interviews with clinicians, the importance of the clinicians' skill was highlighted, including their capacity to engage parents in the initial phase, build a partnership with families, and develop a shared, strengths-based, family narrative (27).

Whilst only one previous study has examined service-provider experiences of implementing FT, a small number of studies have reported on family experiences, which may help to inform workforce practise (28–32). Work by Pihkala et al. (30) and Strand et al. (32) showed that families (parents and children) have reported a number of benefits across a range of mental disorders, although there was some indication that those with BPD or low-functioning psychosis were more likely to struggle with establishing a therapeutic alliance and/or exhibit a lack of understanding/insight into the impact of their mental illness on their children. Parents indicated that factors enabling engagement included having a trusted and skilled professional to mediate family conversations, and timeliness, structure and flexibility of the intervention, while stigma and fear (e.g., being perceived as an incompetent parent) were often significant barriers to participation. However, it should be noted that all of these studies were conducted in psychiatric settings in Sweden, a country with legislation to support families with PMI and which has implemented FT as part of a national "think family" initiative since 2006 (29). In addition, small sample sizes, a limited range of informants, and an overall lack of cultural diversity, restricts the transferability of the findings and underscore the need for

---

Abbreviations: AMHS, adult mental health services; BPD, Borderline Personality Disorder; CAMHS, child and adolescent mental health services; FFP, family-focused practice/programmes; FT, Family Talk; HSE, Health Service Executive; LT, Let's Talk about the Children; MDT, multi-disciplinary team; PC, Primary Care; PMI, parents with mental illness; PRIMERA, **P**romoting **R**esearch and **I**nnovation in **M**ental h**E**alth se**R**vices for f**A**milies and children; PTSD, Post-traumatic stress disorder; RCT, randomised controlled trial; RoI, Republic of Ireland.

qualitative analyses to be undertaken across a wider variety of settings and contexts.

Ireland lags behind most European countries and also Australia, in its lack of legislation and/or a national "think family" policy/practise guidance to support families with PMI (24, 26, 33–36). Moreover, mental health provision in Ireland is severely underfunded when compared with European counterparts, with services operating at between two-thirds to three quarters of recommended staffing levels (37, 38). In the earlier phase of this research (2017–2018), we conducted a scoping study of FFP across adult ($n = 114$) and child ($n = 69$) mental health services in the Republic of Ireland and found that support for families was either non-existent, in the planning stages or *ad hoc* and small scale (4). In addition, the 2019 census for psychiatric units in Ireland provided statistics on 2,000+ inpatients (e.g., age, marital status, diagnosis, socioeconomic status), but failed to include their parental status (39), thereby highlighting a persistent lack of service awareness. Similarly, a recent qualitative study conducted with psychiatric nurses in Ireland ($n = 14$), identified several barriers to FFP, including lack of practise standards to identify service users as parents, no available structured approach, and an absence of appropriate training (3).

The funding provided by the national Health Service Executive (HSE) for the current "PRIMERA" research (**P**romoting **R**esearch and **I**nnovation in **M**ental h**E**alth se**R**vices for f**A**milies and children) was crucial in supporting the first endeavour to systematically implement FFP for families with PMI in Ireland. The aims of PRIMERA were to: (1) identify/develop, implement, and evaluate family-focused interventions for families with PMI; and (2) inform a "think family" care delivery agenda within mental health services in Ireland. Therefore, following an initial scoping and installation phase, we sought to introduce FFP into mental health provision in Ireland through the implementation and evaluation of FT (utilising a randomised controlled trial, qualitative and economic analyses) (4, 40). This qualitative study is one of two which were nested within a randomised controlled trial (RCT) of FT. The objective of this study was to identify and explore with clinicians and managers the barriers and facilitators to implementing and sustaining FT across adult, child (AMHS/CAMHS) primary care and child protection services in Ireland. A companion paper reports family experiences of FT across sites.

METHODS

This qualitative study of practitioner experiences of implementing FT was conducted in the context of an RCT of FT, and was analysed using constructivist Grounded Theory to identify and organise themes, and informed by Fixsen's implementation science framework and the Medical Research Council (MRC) guidance for complex interventions (18, 41, 42). Details of the RCT protocol and registration can be seen at the following link https://trialsjournal.biomedcentral.com/articles/10.1186/s13063-021-05199-4; (40).

Participants and Settings

A purposive sample of mental health clinicians ($n = 31$), and managers ($n = 10$) were identified and recruited for participation in the study, based on their experiences of delivering FT to 55 families within the RCT.

The larger RCT included 86 families (139 parents, 221 children) in 15 sites across Ireland, involving AMHS, CAMHS, primary care psychology, and child protection/welfare services (40). Families were block randomised, on a 2:1 ratio, to the FT intervention ($n = 56$) or to a treatment as usual control group ($n = 30$), and assessed at baseline and 6-month follow up. At follow up, attrition was 37%, the rate of which doubled due to the impact of the COVID-19 lockdown restrictions (23 vs. 45%). Eligible families were those with a child aged 5–18 years and a parent with a formally diagnosed mental disorder. Eighty per cent of service-users were attending AMHS and 20% were receiving antidepressant medication or primary care psychological support under the care of a General Practitioner (40). Due to the high risk of intergenerational transmission of mental disorders (2), and a desire among stakeholders to increase family-focused collaboration between traditionally segregated adult (AMHS) and child mental health services (CAMHS) (4), we included families where children attended CAMHS or primary care services for mental health issues, as well as families where children were not involved with mental health services. Families were excluded if the parent/family was in a state of crisis/instability (e.g., hospitalised, active psychosis/addiction, contentious separation) (40). The 55 families included in service provider reports, had a similar profile to the larger RCT sample in terms of age, gender, mental disorder, and site/location (**Table 1**).

Participating sites were eligible to participate in the research if they had secured managerial support to implement FT, and had identified a lead person to coordinate clinicians, oversee training, plan family recruitment, organise regular peer supervision and be a point of contact with the research team. Clinicians delivering FT were required to have at least 3 years' experience in working within adult, child mental health and/or protection services; have completed the online training in FT (www.emergingminds.com.au), attend monthly supervision, and recruit families and/or facilitate FT. Families were recruited by clinicians in each site from their existing waiting lists. FT was delivered in an outpatient clinic and/or in the home by an FT clinician (40). Ethical approval (for both the RCT and qualitative study) was obtained from four research ethics committees including the research institution where the research was carried out [name withheld for anonymous peer review], the HSE, Tusla child protection agency and Saint John of God's Hospitaller Services. The flow of participants from recruitment through the RCT to the qualitative studies is shown in **Figure 1**.

Clinicians and managers were selected for interview based on service setting (e.g., AMHS, CAMHS, primary care, Tusla child protection agency), professional discipline (e.g., social work, psychology) and site location. All 15 sites were approached and interviews were secured with participants from five sites that recruited 10+ families each, from 3/5 sites that recruited ≤3 families, and from 1/5 sites that did not recruit any

TABLE 1 | Characteristics of families in RCT ($N = 86$).

	N (%)
PMI gender (female)	73 (85)
PMI mean age (SD)	41.01 (7.09)
Lone parent	42 (49)
Mental illness	
– Anxiety/depression	55 (64)
– Bipolar	15 (18)
– BPD	9 (10)
– Psychosis	5 (6)
– PTSD	2 (2)
Length of episode	
– <6 months	16 (18)
– 6–12 months	15 (17)
– 1–2 years	11 (13)
– >2 years	44 (52)
Child gender (female)	120 (55)
Child mean age	10.27 (5.28)
Child mental health	
– CAMHS	42 (19)
– Other psychology/family service	50 (23)
– No service	127 (58)
Family social disadvantage[a]	65 (76)

BPD, Borderline personality disorder; CAMHS, Child and adolescent mental health service; PMI, Parent with mental illness; PTSD, Post-traumatic stress disorder.

[a] *Social disadvantage compared to Irish norms and calculated based on: income, employment, family size, lone parenthood, education and household ownership. In 2019, 17.8% of the population were defined as being socially disadvantaged (43).*

families. Three sites could not be contacted and three declined interview due to FT clinicians either leaving the service or having competing demands on their time due to the COVID-19 pandemic. Most of the 31 clinicians interviewed were female ($n = 27$), parents ($n = 25$), aged 31–50 years ($n = 26$), with 14 employed in AMHS, 14 in CAMHS and 3 in primary care and the Tusla child protection agency. More than three quarters were employed as social workers, three as social care workers, and the remaining five as clinical nurse specialists and psychologists. On average, they had been employed as practitioners for 15 years (SD = 6.7), with most (24/31) having worked in multiple settings (e.g., AMHS, CAMHS, and child protection services).

Ten managers were also interviewed, half of whom were female, six employed in AMHS, three in CAMHS and one in primary care psychology. Most ($n = 6$) were principal/senior social workers, two senior clinical psychologists, one systemic family therapist, and one general manager.

Data Collection and Analysis

All participants provided written informed consent to participate in a one-to-one, semi-structured interview or focus group. Eight managers and eight clinicians participated in an individual interview while two managers and 23 clinicians were interviewed across five focus groups. The focus groups typically lasted ∼1.5 h (with a break if so required), while one-to-one interviews with clinicians and managers lasted 30–45 min. Most interviews were conducted using online platforms (all managers, 24/31 of clinicians) due to the COVID-19 pandemic restrictions. An interview schedule/topic guide was devised to investigate: (1) stakeholders' experiences of facilitating/implementing FT; (2) key barriers and enablers to implementation; and (3) factors mediating the longer-term sustainability of FT/FFP in their service/in Ireland. The interviews were conducted by CM, who had met with all service providers several times previously during the exploration and installation phases of FT implementation (4). Interviews were audio recorded and transcribed verbatim.

The data were uploaded to MAXQDA software (44) and analysed using constructivist Grounded Theory in order to identify and organise themes (41). Analysis was informed by Fixsen's implementation science framework and the MRC guidance for complex interventions (18, 42). Data were analysed using line-by-line and focused coding, constant comparison of codes to find similarities and variations within categories and hierarchical linking of categories to generate super-ordinate (or overarching) themes. All of the interviews were read by CM and MF, CM coded and analysed all of the data, while three authors (MF, SMcGa, SOC) independently assessed the reliability of coding on 25% of the transcripts, with more than 90% inter-rater agreement. Disagreements were resolved by discussion. Trustworthiness of the analytic process was also enhanced by audiotaped interviews, verbatim transcription, audit trail of code generation, clear description of sampling procedures, participants and settings, theoretical saturation, and seeking disconfirming cases. Reporting adhered to COREQ guidelines (Consolidated Criteria for Reporting Qualitative Research) (45).

RESULTS

Three main themes and a number of subthemes therein, were identified, as outlined below (**Table 2**).

Theme 1: Facilitators to Implementation

Clinicians indicated a number of factors as key to the successful implementation of FT including: organisational/managerial support; the structured approach of the intervention; clinician experience and skills; seeing the benefits of the work for families, clinicians and the wider service; and being part of a high profile and well-funded research programme.

Organisational and Managerial Support

Ten of the 15 sites recruited families for the RCT, with five sites recruiting 90% of all families (See **Table 3**). Sites that were more successful were more likely to be led by an FFP champion with strong networking and team-building skills, who had secured support from a Consultant Psychiatrist. In addition, FFP champions promoted interagency liaison amongst AMHS, CAMHS, Tusla, and primary care services which, in turn, facilitated recruitment, shared delivery, and learning. They also engaged in regular awareness-raising and buy-in efforts with management/colleagues to raise the profile of FT within their organisation through, for instance, promoting FT successes during multi-disciplinary team (MDT) meetings. They also

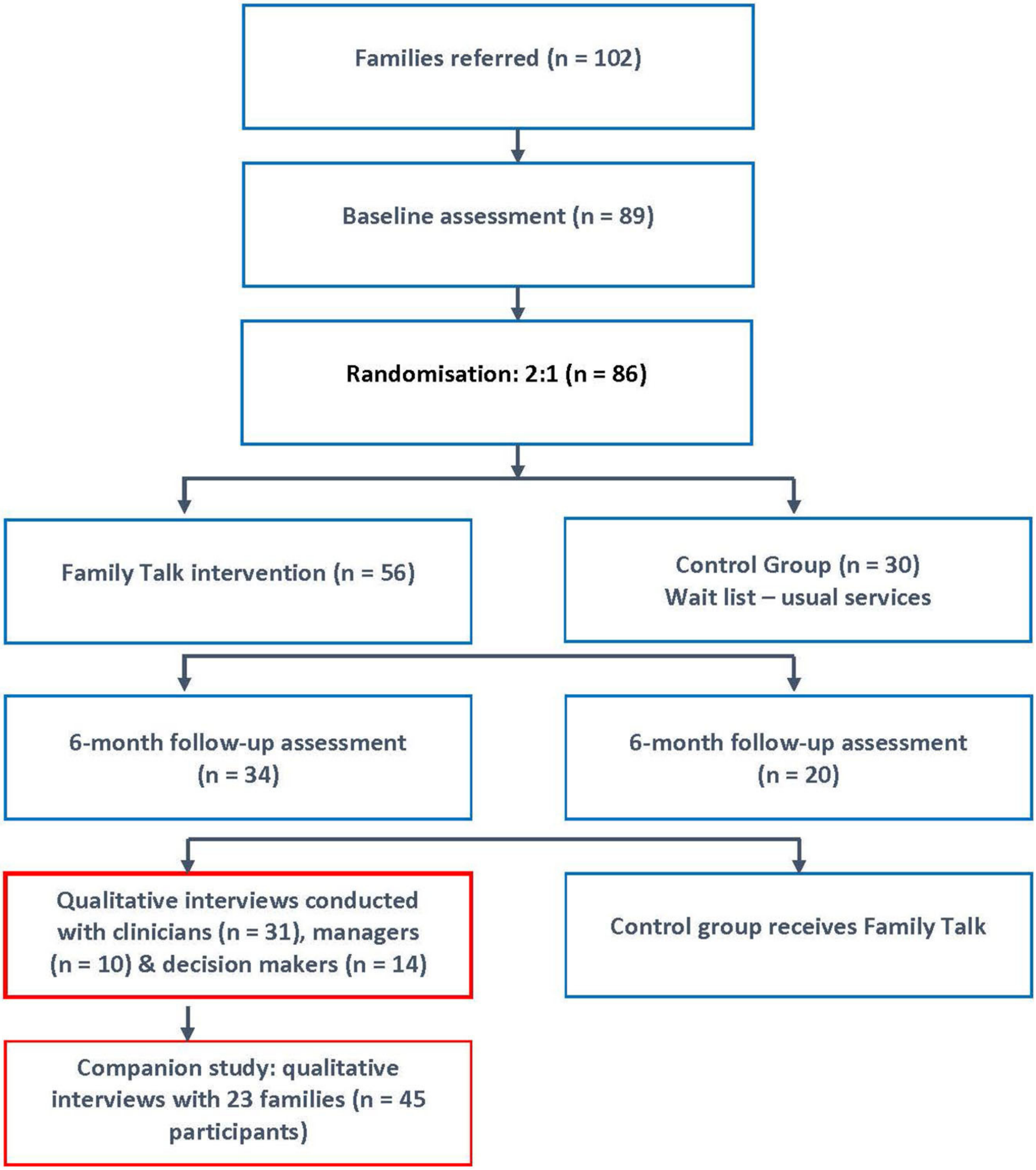


FIGURE 1 | Study flow diagram from RCT to qualitative studies.

established a referral structure for FT and held regular FT peer supervision meetings. Supervision was seen as important in increasing clinician competence and sharing storeys of successful outcomes for families helped to motivate clinicians in their recruitment and delivery efforts. Moreover, clinicians in these areas were given time to complete the training, engage in recruitment and FT facilitation, and attend supervision. It should be noted that sites that recruited more families were more likely to have joined the PRIMERA collaborative research programme in 2018, which gave them more time to train suitable clinicians and identify families, compared to other sites that only joined in mid/late 2019 and only a few months before the onset of the COVID-19 pandemic restrictions.

> *"Bringing them* [AMHS, CAMHS, Tusla and primary care] *all together for supervision every five weeks... discussing cases of dynamics and challenges. They also have the peer supervision and support... The work was seen as important."* (Manager 5, AMHS, Site 1)

> *"My consultant psychiatrists and my team are excellent–and she hears that this work is done. And she's delighted! But she's one of the few psychiatrists who I've seen think systemically."* (Clinician 10, AMHS, Site1)

> *"More recent referrals have come from team members... that probably has a lot to do with a few more of the talks from myself, a team meeting generating referrals... one of the consultants in the team was quite eager."* (Clinician 3, CAMHS, Site 1)

Structured Approach of FT

All clinicians/managers appreciated the structured, yet flexible, approach that FT provided in working with families. They also valued its evidence base and its manualised, no-cost, online training. Most also highlighted the importance of the psycho-education provided, and indicated that the skills they had gained were transferrable, although some noted that additional face-to-face training might be helpful for managing more complex cases.

> *"I thought the training was really good. I thought it was very accessible... I see the children and the parents get a lot from it... The checklist is really helpful... The structure is invaluable. It's really easy to evidence the work that I'm doing."* (Clinician 8, AMHS, Site 2).

> *"The fact that it was free, it was online, it's brief, that we could do it ourselves, it didn't require investment from the services–all those things appealed to us."* (Manager 9, AMHS, Site 5)

> *"I do think given the complexity of cases, you do need to modify, but the structure is there, and the structure is very accessible to most people. And that's one of the big strengths to it."* (Clinician 14, AMHS, Site 1)

Clinician Skills and Experience

Clinicians with prior experience of working in both AMHS and CAMHS were more committed to FFP implementation, having observed at first hand the transgenerational effect on

TABLE 2 | Experiences of implementing Family Talk.

Themes	Subthemes
Facilitators to implementation	Organisational and managerial support Structured approach of FT Clinician skills and experience Seeing the benefits of the programme Role played by research/research team
Barriers to implementation	Engaging and retaining families – Family challenges – Clinician and organisational barriers – COVID-19 and research barriers – Variation across sites Delivery challenges
Sustainability of FT/FFP in Ireland	Site continuity plans FT fit with service remit and as part of FFP suite of supports Longer-term sustainability of FFP

FFP, Family-focused practise; FT, Family talk.

TABLE 3 | Site characteristics.

Site	Date joined study	*N* family recruits	% family withdrawals	No. trained family talkers	Interagency effort	Service(s) involved
1	Mid 2018	39	33	16	Yes	AMHS, CAMHS, PC, Tusla
2	Late 2018	15	19	5	No	AMHS
3	Mid 2018	14	12	18	Yes	CAMHS, AMHS, Tusla
4	Early 2019	13	17	10	Yes	CAMHS, AMHS, PC
5	Mid 2018	10	7	5	No	AMHS
6	Late 2019	2	2	1	No	AMHS
7	Late 2019	2	0	8	Yes	AMHS
8	Mid 2018	1	0	6	Yes	AMHS, recovery college
9	Mid 2019	2	5	1	No	Tusla
10	Late 2018	3	5	3	No	CAMHS
11	Mid 2018	0	–	3	Yes	AMHS, CAMHS
12	Late 2018	0	–	1	No	Tusla
13	Late 2018	0	–	2	No	Tusla
14	Late 2019	0	–	2	No	Tusla
15	Early 2019	0	–	3	No	AMHS

AMHS, adult mental health services; CAMHS, child and adolescent mental health services; PC, primary care.

children when they became service users. In addition, cross agency experience gave clinicians confidence and competency in working with the whole family, and facilitated interagency collaboration and co-delivery of FT, which considerably enhanced family recruitment and the quality of programme provision. Furthermore, most participants were social workers and believed that their professional training equipped them to be more persistent with family work when compared with other disciplines on mental health teams; for instance, they felt more competent in assessing family readiness for FT; establishing a positive relationship with families before and during FT; and in working with multiple family members.

> *"I spent time in both AMHS and CAMHS. You would see people being referred and you would see there was an inter-generational connection. What you often see is a history of parental mental illness and how that's impacted on them growing up."* (Clinician 29, AMHS, Site 1)

> *"What I liked about it was having the mum and dad and the others all in the room together because while this may be new for some clinicians, it's not odd for the family, because that's the way they work as their every day."* (Manager 1, Primary Care, Site 4)

> *"From my point of view, co-working works really well. The adult mental health practitioners being involved is really important because the children are very badly affected, so having this model of working on those cases, I'll be working with that going forward."* (Clinician 11, Tusla, Site 1)

Seeing the Benefits of the Programme

Benefits to Families

An important and frequently reported implementation driver for clinicians/managers, was the benefits they had witnessed in approximately two thirds of the families with whom they were working; these included: reduced worry and stigma, a greater understanding of the impact of PMI on family members, a new family narrative around the parents' illness, and improved family communication. Clinicians indicated that parents/partners were typically surprised/upset by how much their children had been affected by tense/volatile home situations, and had hidden their worries and concerns to avoid burdening parents. For children, having their reality acknowledged, was significant as children were usually told nothing was wrong. As parents became more cognisant of their children's needs, family members were motivated to reduce levels of anger/arguments, and to relate to each other in more warm, caring and fun ways, thereby leading to reduced stress and increased family well-being. Clinicians further indicated that the improved family interactions/relationships assisted the PMI's personal and parental confidence and well-being.

> *"I think parents being able for the first time to hear their kid's opinions, and that they have opinions on it, they do have questions, and they're not in the dark–that does have a positive impact. Parents can become upset. I have had parents who cry in the feedback session. They can't believe they* [children] *knew what was happening, but there is some motivating factor in that for recovery. One parent I was working with for over a year had not shown a massive shift, but whatever it was about hearing feedback from her kids, and questions about her mental health, it seemed to motivate her. It did make a difference to her recovery."* (Clinician 4, AMHS, Site 3)

> *"Their life is totally different. The mum had a lot of guilt and shame around her being a mental health patient. It was the first time she talked to the girls and they talked about the frustrations of mum not being available. She's able to speak to both the girls now. Mum is able to cook everyday when she couldn't before so life has become a lot more predictable, which is exactly what they wanted–so hugely beneficial for them."* (Clinician 10, AMHS, Site 1)

> *"For the kids themselves, just to be given that space to talk and have their own voice heard is huge… Because the kids know without maybe knowing what the words are for it, but they know that there's something going on in the household…Takes a huge weight off their shoulders…In one family, both girls were actually blaming themselves for mum's illness because their aunt had told them it was their fault that mum was having relapses."* (Clinician 1, CAMHS, Site 3)

> *"That was the best thing he [service user] had done he said and because of the communication with his family, he's doing quite well again. He's more aware of the need to communicate."* (Clinician 15, AMHS, Site 1)

Benefits for Clinicians and the Wider Service

Most clinicians also believed that FT was beneficial for themselves and for their service. FT was reported to be enjoyable and rewarding and had helped to allay long-held ethical concerns about not addressing the needs of family members. In addition, several clinicians noted that FT worked well as a stepping stone for early identification of vulnerable families within their service, could be easily added to treatment plans, and was useful in signposting families to additional supports if required.

> *"It's definitely a hugely beneficial piece of work… I could feel it as a practitioner, and they could feel it as a family."* (Clinician 1, CAMHS, Site 3)

> *"If you think about it, this intervention is almost social justice. We're doing what we believe is right in developing children's rights."* (Clinician 10, AMHS, Site 1)

> *"It was overall positive and really valuable work."* (Clinician 3, CAMHS, Site 3)

The Role Played by the Research/Research Team

Clinicians and managers indicated further that a significant motivating factor for their involvement in FT training and delivery—and another key implementation driver—was the fact that the research was funded by the HSE (national health service in Ireland) and involved a multi-site, national programme aimed at addressing a major service gap in Ireland (i.e., developing FFP for families with PMI). Participants also clearly appreciated the wide range of advocacy and support activities undertaken by the research team to scaffold site buy-in, implementation, and family engagement. These included: co-developing a complementary online resource hub to assist clinicians in working with families; co-producing brochures and posters to recruit families; hosting/facilitating access to FFP workshops/masterclasses; co-delivering presentations to site managers and MDTs; providing regular updates by e-zines; and promoting the study through local and national media to raise public and service awareness on the topic (4). Thus, the early installation and implementation of FT was a joint collaboration between the research team and site stakeholders (4).

> *"What attracted it to us was the fact that it was supported by research, it was multi-site, it was a broader 'Think Family' agenda which appealed to us… The sense of being part of something bigger. There was a support structure there and we wouldn't have done this in a systematic way unless we were part of the research study."* (Manager 9, AMHS, Site 5)

> *"More recent referrals have come from team members, and that has a lot to do with a few more of the talks by the research team coming into the service."* (Clinician 3, CAMHS, Site 3)

> *"It was great to be part of the research. I feel it was a very exciting time and you guys are doing such an incredible job…I definitely intend to keep going. I would absolutely love to see it more evolved in Ireland. I'm a big believer in it."* (Clinician 4, AMHS, Site 3)

Theme 2: Barriers to Implementation

Engaging and Retaining Families

Engaging and retaining families was the primary challenge faced by service providers, and was one which was exacerbated by the COVID-19 restrictions. Clinicians indicated that three to four families had to be approached for every one successfully recruited, and in ten sites there were three or less families recruited (**Table 3**). Overall, 16% (16/102) of referrals to the RCT were withdrawn before randomisation due to their unsuitability for FT (e.g., child protection issues, parent relapse, family crises). Of the 56 families allocated to the intervention group, 6 did not start FT and 5 disengaged after attending <3 sessions, with 53% attending all sessions [mean attendance was 4.4 sessions (Sd 1.2)]. Participants identified a range of barriers to engagement and retention covering multiple family, clinician, organisational, pandemic, research, and systemic/cultural levels.

Family Challenges

Clinicians indicated that for many parents–including those who agreed to attend FT–mental health stigma and concerns about involving their children, was a major concern and key barrier to implementation. Many parents disagreed about what should be discussed with their children, while concerns around social worker involvement with their children, also inhibited engagement. A small number of children in CAMHS also were anxious about discussing the issue with their parents. In addition, many of the cases on waitlists were complex (e.g., long-term service users, socially disadvantaged) which may also have affected engagement and retention. Thus, extensive preparatory work by clinicians was needed to allay all of these concerns and fears. Clinicians also reported that some families disengaged before FT commenced/completed due to family crises (e.g., threat of homelessness, job loss), relapse in mental health symptoms, having other priorities or finding it too emotionally painful to hear from their children about the impact of their illness on them.

> *"As much as we're trying to reduce the stigma of mental illness… It's a massive thing still in Ireland. Especially I think for the parents. I definitely think more open communication is essential in families."* (Manager 3, AMHS, Site 2)

> *"I think it's probably about five or six families that said no. Their reason for saying no was, 'don't like social workers'… or fear that I'm going to start doing a parent assessment and that someone will be speaking to their child."* (Clinician 15, AMHS, Site 1)

> *"The family withdrew… Maybe it was the difficulty of having to talk to her mum about how she was feeling about their relationship. They disengaged with CAMHS… And then COVID hit and to be honest, the crisis of the last couple of months… so that has been it."* (Clinician 21, CAMHS, Site 4)

Other Barriers

All sites experienced a number of organisational barriers that affected the engagement and retention of families, although some struggled more than others. Resistance to FFP from colleagues was reported as common due to: heavy workloads, staff shortages/high turnover, ideological differences (e.g., perceiving FFP to be outside their service remit), and feeling ill-equipped to undertake family work due to the individualised, crisis-oriented focus of their professional training. Other barriers included: slow referral processes; difficulties in identifying PMIs; needing to re-secure buy-in with new consultants who rotated on a 6-monthly basis; and colleagues being supportive in theory but not in practise as demonstrated, for example, by their unwillingness to train in FT or to refer families, a tendency to discharge suitable families without notice, and being risk adverse in balancing service-user confidentiality/data protection concerns with family needs.

> *"We've had locum six-month positions who are very good psychiatrists, but then they're gone. And they don't have any weight when they're here for six months and they are very dismissible"* (Manager 8, CAMHS, Site 10)

> *"Some will say that's not our job, it's a luxury, it's time consuming…Most other disciplines are trained just to work with an individual. So whereas we're going into the messy family life and that's a very frightening thing for services and they'll say to you, 'oh GDPR'… It's very much a pushback, people aren't comfortable with it at all."* (Manager 4, CAMHS, Site 3)

> *"Health services are reactionary. They deal with crisis after crisis… Which shows how slow we have been to look at preventive intervention… The other reluctance around this is that if you start looking at the psychological and social aspects of mental health, that may potentially reveal the delusion of psychiatry and the medical model."* (Manager 10, AMHS, Site 8)

In addition, there was evidence in some sites that insufficient effort may have been invested in recruiting families, which led to some not engaging with FT. For instance, it was reported that FT may have been poorly explained to families, or that parents had been informed by "cold calling" rather than through the building of a prior relationship with them. In addition, several clinicians indicated that negative past experiences of mental health/child protection services amongst some families, had led to their disengagement. Furthermore, some families dropped out following lengthy delays to FT delivery as a result of the COVID-19 pandemic restrictions in Ireland, and particularly in sites where mental health clinicians were redeployed to frontline COVID-19 duties (46). While involvement in the research promoted implementation and recruitment in some regards (as discussed earlier), being involved in a time-limited RCT also hindered recruitment to some degree. For instance, some families did not wish to be in the control group or to complete questionnaires. One site conducted FT with several families ($n = 7$) but not as part of the RCT and, despite support from the research team, struggled to communicate to families the value of taking part in the research.

> *"The main challenge was recruitment. It's because they* [colleagues] *didn't explain it properly to the parent."* (Clinician 31, CAMHS, Site 3)

> *"We have been hugely affected by COVID... And after so much work put into it* [FT]. *That's been hugely challenging."* (Manager 6, AMHS, Site 1)

> *"We had a certain amount of time to complete it because of the* [research] *timelines so there's that added pressure to find families and get them seen. Once that is gone, it will be very good to see this as an integral part of AMHS. I really hope that happens."* (Clinician 12, AMHS, Site 1)

Variation Across Sites

Ten sites recruited three or fewer families, only one of which (site 11) withdrew from the research; they did so because clinicians did not see FT as being a fit with the type of systemic family work which they wanted to undertake. The remaining nine sites were all characterised by limited resources (e.g., few FT clinicians with little dedicated time), ideological differences, lack of a champion or practical support from colleagues, and/or lack of organisational readiness to engage families due to joining the study later in its lifetime and especially with the onset of the COVID-19 restrictions. Furthermore, eight of the ten sites had little history of interagency work, which possibly impeded recruitment. Notably, those sites in which child protection services collaborated with AMHS and CAMHS were more successful in engaging families than those who attempted to deliver FT without such interagency support; the latter struggled with clinician buy-in and family recruitment. Child protection practitioners/service providers in Ireland are typically not trained in mental health, and without interagency support, they may have felt less equipped to undertake family-focused mental health practise. In addition, given their limited resources and crisis-oriented focus, they may not have considered families with PMI to meet their criteria/threshold of a child being at risk.

> *"I felt a bit overwhelmed...I was the only one that took on the Family Talk intervention even though I spent a lot of time advocating for it... If I was rolling out Family Talk maybe in six- or seven-months' time, I think I would have had more space and the team would have gotten to know me better and trusted me with some of the families to see what social work can do. Within the team, the role of social work was a very basic view of the role of social work* [e.g., form filling and applying for benefits/services rather than engaging families in interventions]." (Clinician 26, AMHS, Site 7)

> *"Mum has mental health problems, a lot of trauma from her background... The family would really benefit from it [FT]. But Tusla said, no, it doesn't meet our threshold as Dad's a protective parent."* (Clinician 28, Tusla, Site 13)

Delivery Challenges

A small number of clinicians indicated that the family meeting, in particular, was stressful, due to the emotional content being shared, and the requirement to support parents and children spanning a broad age range.

> *"What I found difficult was the family meetings, you were sitting with mum, a 16-year-old, an 11-year-old and a six-year-old in the room. You speak differently... You're still getting the essence across, but you're not being as frank about certain issues, or you're making it more child friendly because a child is there."* (Clinician 4, AMHS, Site 3)

Fidelity to FT protocols was also a challenge, with frequent delays/disruptions due to the COVID-19 restrictions. In a small number of cases, clinicians adapted FT using online platforms, which facilitated individual parent and older teen sessions, but was not considered suitable for younger children or family sessions, and therefore completion of FT was delayed. In addition, for families with more complex needs, one third of clinicians indicated that they provided additional parent, child and family sessions beyond the 7-session model, and referred families to further services (e.g., individual/relationship counselling, family supports). As FT was frequently the first (and perhaps only) time parents and children spoke about living with PMI, parents/partners were often angry/upset during initial sessions, while some service users needed time to adjust to not being the sole focus of care. Child meetings were also extended (if time permitted) when complex issues or concerns were raised.

> *"Due to the pandemic, I was unable to recommence Family Talk. It was impossible to start the individual meetings again and it just didn't flow straight into the family meeting. Otherwise, I feel the Family Talk would have been very successful."* (Clinician 7, AMHS, Site 2)

> *"A couple of families had a lot of issues, and they needed time–one session with the kids wasn't going to be enough... And*

> *they needed follow-on supports that I was able to refer them to."* (Clinician 11, CAMHS, Area 3)

Theme 3: Sustainability of FT/FFP in Ireland

Site Continuity Plans

Despite the disruptive long-run impact of COVID-19 (e.g., increased waitlists), six sites have continued to deliver FT beyond the research programme, while the remaining areas hope to use its principles in practise, subject to resource limitations. The top five recruiting sites (**Table 3**) appear best placed to sustain FT as managers/clinicians have: (1) introduced practise guidelines for engaging families to FT as part of routine service provision (e.g., during initial patient assessments); (2) promoted FT using service-user feedback; (3) encouraged new staff/colleagues to train in FT; (4) continued to deliver FT to families; and (5) held regular FT peer supervision.

> *"We have continued receiving referrals for Family Talk and are continuing to deliver it to families. I am delighted that staff want it to become embedded in practice and our peer supervision group has become an established forum."* (Manager 6, AMHS, Site 1)

> *"We still continue here in CAMHS. I still fly the Family Talk flag as much as I can."* (Manager 4, CAMHS, Site 3)

> *"We are going to continue using it in CAMHS. I think it's a very useful service. But definitely the challenge is the recruitment."* (Clinician 21, CAMHS, Site 4)

> *"I still use it. I use it in everyday work now."* (Manager 10, AMHS, Site 8)

> *"The intervention is really great so it's definitely something that we're going to continue to do with families. It should have been here a long time ago."* (Clinician 29, AMHS, Site 5)

FT "Fit" With Service Remit and as Part of FFP Suite of Supports

A key sustainability issue concerned the perceived "fit" of FT with service remit; while many stakeholders expected AMHS to be the most natural fit for FT–and with CAMHS/Tusla perceived as being more proficient at family work–success in implementing FT appeared to be mediated more by local site resources, organisational culture and the availability of a strong champion. A small number of CAMHS clinicians within one site viewed FT as a mid-level intervention which was not sufficient for complex cases while CAMHS clinicians in other areas, working with equally complex cases, believed FT was appropriate. In addition, four AMHS and CAMHS clinicians believed that while FT principles would inform their future practise, the FT intervention would be better delivered at primary care level, given their lower threshold for access (i.e., mild to moderate mental health presentations). Conversely, a clinician working in primary care psychology indicated considerable recruitment challenges due to a lack of willingness among parents with moderate anxiety/depression to acknowledge the impact of their difficulties on their children. This participant indicated that recruitment should be easier in AMHS where patients generally have a more defined diagnosis. Therefore, while FT was successfully delivered in all types of service—AMHS, CAMHS, Primary Care, Tusla—thereby reflecting a "no wrong door" approach to service provision (25), all sites experienced considerable implementation challenges, and many participants felt that siloed service provision had undermined their capacity to properly support families. The child protection agency, Tusla, in particular, experienced the most implementation difficulties, most likely due to families' fear of social services, although interagency collaboration was shown in this study to support their involvement.

> *"I expected CAMHS to be very family focused. I've been really surprised that it had, like adults, become very much focused on the child is the problem and you fix the child. And you don't look at anything else."* (Manager 4, CAMHS, Site 3)

> *"I think for us to use it to inform our practise but as a package, it probably would work better as a prevention piece on the primary care level."* (Manager 7, AMHS, Site 2)

> *"This is the problem when you fragment service, and they're not integrated. Tusla is a separate agency. Adult and CAMHS are very separate. This shouldn't be. Because children, come out of one family yet the family might be attending three or four different services, which is part of the problem."* (Manager 6, AMHS, Site 1)

Given the complexity of some family cases, it was advised that sustainability of FFP in the RoI would be enhanced if FT was implemented as part of a suite of lower and higher intensity interventions. As indicated, clinicians felt the need to deliver extra sessions to several families, and frequently referred to follow-on services/supports, including individual and couple counselling, family and youth services, parent programmes, men's groups, and dialectical behaviour therapy. It should be noted that 76% of families in the RCT were socially disadvantaged and therefore presented with a high level of need.

> *"Some families probably need longer intervention... And then when the parent can't overcome stigma or family members are resistant, maybe something lower key in just talking with the parent might help also. But FT has been great for the families that come to it."* (Manager 1, Primary care, Site 4)

Longer-Term Sustainability of FFP in Ireland: Systemic Barriers and Roadmap

For longer-term sustainability of FFP in Ireland beyond a small number of committed sites, all participants indicated that FFP is unlikely to flourish within the current medical, individualised, siloed, under-funded, crisis-oriented model of mental health care in Ireland which was perceived as encouraging services to believe it is not their core business to support families with PMI. Other systemic barriers noted by decision makers include a lack of data and accountability of how HSE funding is spent, and initiatives typically being introduced in an *ad hoc* manner with little infrastructural support.

All participants highlighted the need for a multi-level, public health approach to raise service and public awareness on

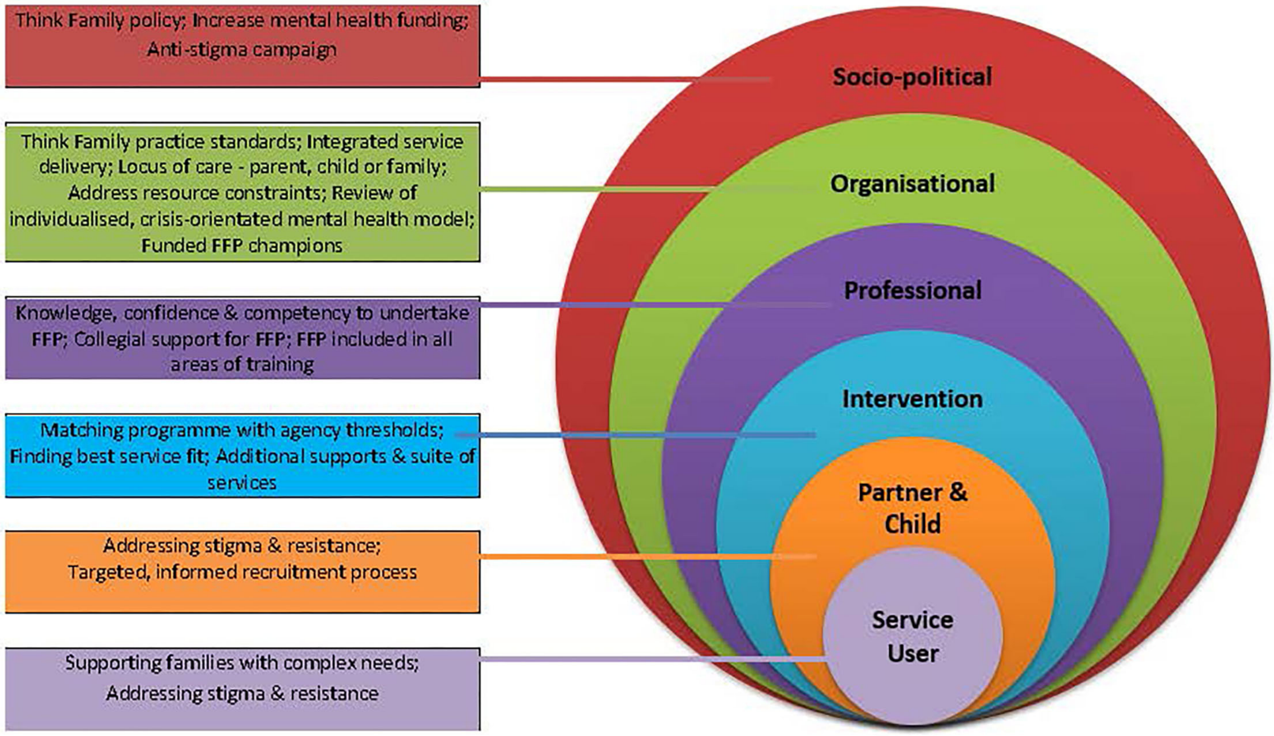


FIGURE 2 | Multi-level approach to embed FFP.

PMI, including: introducing a national "think family" policy initiative/practise guidelines; providing dedicated funding for FFP, and mental health services more generally; launching media campaigns to reduce mental health stigma; addressing systemic/interagency barriers to change (e.g., including FFP within professional training across disciplines, auditing parenting status, and allowing time for FFP within, and across mental health services). In addition, change agents (champions) need access to senior management to effect change at frontline, operational, and strategic levels. Given the movement of personnel within the HSE, multiple FFP positions are needed to ensure sustainable practise (see **Figure 2**).

Interestingly, while all participants agreed that FFP was long overdue in the RoI, there was little consensus on the effectiveness of legislating/mandating FFP or the benefits of introducing standards which may, in practise, be reduced to a meaningless tick-box exercise with little benefits for families or clinical practise. Rather, participants emphasised the benefits of providing training in FFP, such as FT, and having managerial support to deliver FFP to families.

> *"I think if you make this kind of thing mandatory or legislative, it adds a little bit to the scary factor, both for families and us working with them… I think a better investment is to train clinicians in it [FFP/FT] and then support them to do it, allow them time. But you need to move beyond the individual, medical model for that."* (Manager 1, Primary Care, Site 4)

DISCUSSION

Service providers highlighted a number of benefits for the majority of families, while several key facilitators and barriers to implementation and sustainability were also identified. The benefits noted here corroborate those reported by a sample of family members ($n = 45$ from 23 families) who participated in a second qualitative study which is reported in a companion paper (ref withheld for purposes of anonymous review). The findings are also consistent with those of studies of clinicians and families who experienced delivering/attending FT in psychiatric settings in Sweden (30, 47). Perceived benefits for families in this study included: feeling heard and validated, reduced worry and stigma, a greater understanding of mental illness; improved parental confidence; and better family communication. Benefits were reported across different mental health settings (e.g., AMHS/CAMHS) and types of mental disorders and highlighted that FT was capable of being implemented in a country without a "think family" policy or dedicated FFP funding infrastructure.

Within the current study, clinicians/managers identified a number of facilitators and barriers to implementation, which build upon those identified in (the few) previous qualitative studies of FT delivery (28–32, 47), and which should help to inform the future implementation of FT/FFPs across countries. These might also usefully be tested as mediators/moderators within controlled trials. Five of the 15 sites recruited 90% of families (**Table 3**) and participants from these sites provided important insights into key facilitators. These included: the availability and drive of an FFP champion with managerial support; promoting interagency collaboration among AMHS, CAMHS, primary care, and child protection services in the area; engaging in regular awareness-raising and buy-in efforts with management/colleagues (e.g., FT on weekly MDT agenda and offered as part of care plan during initial assessments); encouraging clinicians to participate in FT training; setting up referral and supervision structures, and allowing clinicians sufficient time to engage in FT promotion, recruitment, and delivery activities. The use of multiple modes of recruitment (e.g., brochures, in-person invitations, phone-calls) also appeared to be linked to better family engagement. These findings are important in reinforcing the enablers of successful FFP implementation identified elsewhere, including building community capacity and interagency collaboration (5, 19), as well as targeting management, organisational policy, and professionals' attitudes, skills, and knowledge (5).

Another key facilitator to implementation was the structured, manualised approach of the intervention, and its freely available online training, which greatly increased its accessibility for busy professionals working across different geographical areas. Nevertheless, some clinicians indicated that they would have welcomed supplementary face-to-face training with international FT trainers, and would have liked the online training to show clinicians working with more complex cases (e.g., lone parenting/social disadvantage) and across a range of mental disorders. It should be noted that while clinicians in this study did not receive the 2-day, face-to-face FT training, they were required to undertake the online 'Keeping Families and Children in Mind' training to familiarise themselves in FFP prior to the FT training. They were also invited to several no-cost FFP masterclass/workshop events organised by the research team, whilst an online resource hub was also co-developed to supplement FT training in Ireland (e.g., providing resources on how to work with children, how to engage families) (4).

The level of clinician skill was another important enabler, including their capacity to engage parents in the initial phase, build a partnership with families, and develop a shared, strengths-based, family narrative (27). Most participants in this study linked their confidence and competency in FT delivery to their professional training in systemic approaches (e.g., social workers), and having previously worked within AMHS, CAMHS and child protection settings. Thus, the whole-family approach of FT dovetailed well with their attitudes and experience. Clinicians' self-efficacy beliefs have been indicated elsewhere as a key predictor of provider willingness to conduct FFP (48), and as such, addresses the "*not mine, not trained, too busy, too risky*" mindset that is a common barrier to FFP implementation (49). It is interesting to note that families in our companion study also highlighted the importance of clinician experience/competence and a non-judgemental and hopeful attitude, both of which were seen as helping to reduce stigma and promote family engagement.

A significant barrier to implementation across all sites related to difficulties in engaging families to take part in FT. Likewise, two other FT studies have also noted high refusal rates of up to 60% (13, 47). Clinicians indicated that barriers to engagement presented at family, clinician and organisational levels. Largely similar to FFP barriers noted elsewhere, family barriers included mental health stigma, parental fear and ambivalence about involving children, and families' complex presentations (i.e., PMI is only one of several presenting issues). Clinician/organisational barriers included limited resources/priority given to FFP; ideological differences; fragmented services; no champion to drive implementation; and/or little practical support from colleagues (3, 5, 24, 50). Interestingly, in our companion study of family experiences, several children reported that they were not informed about the purpose of FT and would have appreciated meeting the clinician before commencing sessions. Moreover, it appears that some families may have been approached before they were ready to engage (e.g., symptoms elevated, in denial/unaware of impact of their illness on children). These recruitment difficulties suggest that clinicians may further benefit from the development of FT/FFP training videos/protocols to promote effective engagement strategies and address potential barriers to participation and retention. For instance, addressing issues of stigma, readiness, consent, and confidentiality during the recruitment process and including quotes/videos from previous FT attendees, may help to improve engagement (50). In addition, a child-friendly recruitment approach that uses age-appropriate marketing literature and involved a meet-and-greet session with the facilitating clinician, might help to address children's concerns about attending. Similar protocols might also be usefully developed to promote organisational/clinician commitment to FT/FFP implementation, including, for instance, putting FFP on the weekly agenda and in careplans, discussing ideological concerns (e.g., confidentiality, data protection, service remit), gaining collegial support, and securing dedicated time to undertake FFP. It is important to note that several sites were gaining momentum in recruitment just as the COVID-19 emergency was starting and, for the same reason, those sites which joined the study at a later date, did not have an opportunity to engage families as they had intended (**Table 3**).

Clinicians identified some pressure points when delivering FT. A small number found that facilitating the family sessions was particularly intense (and occasionally volatile) given the range of perspectives and developmental stages of family members. Therefore, it might be useful if the online FT training provided advice on how to tailor the discussion when children of different ages (e.g., 6 vs. 16 year-old) are present. Secondly, most families indicated that they would have liked more child, family and follow-up sessions whereas one third of clinicians indicated that, for complex cases, they had already provided additional sessions beyond the 7-session model and had referred families to further services. The families' perspective most likely reflects their level of need (e.g., 76% were socially disadvantaged), as well as the general unavailability of mental health/family supports in the community, whereas the clinicians' perspective reflects working within a context of limited service resources for FFP. Most previous studies of FT have not mentioned the need for additional sessions or follow-on supports, but this may be due to their participants being largely middle class and relatively high functioning (12–14, 51, 52). There is evidence from two qualitative studies of FT that some parents with low-functioning psychosis and Borderline Personality Disorder may struggle to understand the impact of their illness on their children, and may require additional supports (31, 47). These supports may include extra psycho-educational sessions and/or complementary groups for patients and children, in order to share experiences and learn about their mental illness and its impact on their children (47).

The sustainability of FFP in Ireland was a recurring concern for all participants. Reassuringly, six sites have continued delivering FT beyond the research programme and have established structures to enhance its sustainability, such as integrating FT into organisational procedures and care plans, and providing continued supervision and training of new FT personnel. Therefore, these sites have moved beyond Fixsen's stage of initial implementation, and particularly Site 1, but they have not yet reached full implementation as sustainability is still vulnerable to champions leaving the service (18). The remaining eight sites indicated that they will either: (1) use the FT principles in practise (e.g., "think family" when working with a service user) but not deliver the whole intervention; or (2) deliver FT as individual clinicians, but without receiving much practical support from management/colleagues. Therefore, all sites indicated that implementation of FT has enhanced a "think family" mindset but there is significant variation in terms of embeddedness (18).

These sustainability concerns raise questions about the perceived fit of FT with organisational remit and capacity. In many ways, given the individualised model of care in AMHS/CAMHS, it was a significant paradigm shift for these services to deliver a whole-family intervention, such as FT. While service providers appreciated the benefits gained from the whole-family model, there were nevertheless indications that FT should be implemented as part of a flexible suite of lower and higher intensity interventions, as recommended by international experts in the field (53). Higher intensity interventions may be more suitable for families presenting with complex needs, while lower intensity interventions may appeal to organisations with limited resources/individualised model of care and/or where families have less need or parents are unwilling to involve their children in services. In some jurisdictions, the two-session, parent-only, "Let's talk about the children" (LT) intervention has been implemented in AMHS settings and has been shown to increase understanding of PMI (13). Nevertheless, in our companion study of family experiences, we found that FT allowed children (and partners) to reveal burdens and concerns that would likely have remained concealed with an intervention that only involved interacting with the service-user parent. Furthermore, two head-to-head RCTs of LT and FT found the latter to be more effective in reducing child emotional symptoms and improving the parent–child relationship (12, 13). Therefore, further dialogue is required on whether mental health services should adapt their remit to become less individualised, and more family-focused, and/or whether only lower intensity interventions should be implemented so as to fit in with current service constraints.

Another key sustainability issue is identifying the type of service that is best placed to deliver FT/FFP. While AMHS may appear the most natural fit (given that parents have a diagnosis], our results demonstrate that CAMHS, primary care, and child protection services can effectively deliver FT, thereby reflecting a "no wrong door" approach to FFP provision. Mediating factors in the current study were less related to type of service than to the availability of a champion and local site resources as well as organisational culture, and interagency collaboration. In Australia, where a range of FFP supports have been established for over 20 years, AMHS and primary care are the most common provider settings (24), but in general, there is a consensus that FFP is the responsibility of all services, whether adult- or child-focused (54).

STRENGTHS AND LIMITATIONS

This study is just the second qualitative analysis of practitioner experiences of implementing FT, and the first conducted within the context of an RCT and national programme to introduce FFP for families with PMI across AMHS, CAMHS, primary care and child protection settings (in Ireland). A large and diverse sample of stakeholders ($n = 41$) was interviewed including clinicians and managers across a number of sites, including those that struggled with recruitment. The findings identified a number of barriers and facilitators to implementation and mirror the family experiences of FT reported here in our companion paper.

Limitations include the generalisability of the findings across different cultural contexts and settings. Unlike other jurisdictions where FT was longer established and/or there was prior legislation/FFP practise standards, FT was implemented in Ireland as a catalyst for a paradigm change in mental health provision for families with PMI. In addition, most sites involved AMHS or CAMHS staff so caution is advised, therefore, in generalising to other mental health/family support settings. Furthermore, most of the clinicians/managers were social workers and 80% had previous experience in working within AMHS, CAMHS and/or child protection settings, thereby potentially limiting generalisability to other disciplines and those without cross-agency experience. Importantly, there was some evidence that FT implementation (e.g., site buy in) had taken place because it was the focus of a national research programme funded by the HSE in Ireland. While some clinicians indicated that the RCT timeline also impeded recruitment, all RCTs are time-limited which means that some families were not ready to participate within the timeframe of the study or they did not wish to be part of the control group. Lastly, this is the first study of FT to be undertaken, in part, during a global pandemic. The COVID-19 lockdown restrictions halted recruitment, and seriously affected programme delivery and fidelity which led, in turn, to some family disengagement from services. Service providers were also interviewed during the height of the pandemic restrictions, which may have affected their perspectives given the impact of the pandemic on mental health in the general population (55, 56).

RECOMMENDATIONS FOR PRACTISE, POLICY AND RESEARCH

Benefits were reported for approximately two thirds of families across different diagnoses and mental health settings (AMHS/CAMHS/primary care/child protection), thereby reflecting a "no wrong door" approach to identifying and supporting families. Key implementation facilitators included: acquiring managerial and organisational support through awareness-raising and buy-in activities; building clinician skill in systemic practise; establishing interagency collaboration across AMHS, CAMHS and primary care; setting up referral and supervision structures, and allowing clinicians sufficient time to engage in FT promotion, recruitment and delivery activities. Recruitment difficulties may be targeted by addressing issues of stigma, readiness, consent and confidentiality during the initial engagement process with families and including quotes/videos from previous FT attendees (50). In addition, children's concerns about attending FT may be allayed by using age-appropriate marketing literature and setting up an initial meet-and-greet session with the facilitating clinician before the FT sessions begin. Organisational/clinician commitment to FT/FFP implementation may be enhanced by: putting FFP on the weekly agenda and in careplans, discussing ideological concerns (e.g., confidentiality, data protection, service remit), and securing dedicated time to undertake FFP. In some cases, it may be necessary to signpost families presenting with multiple disadvantage to additional supports following FT. Lastly, where it is difficult to secure organisational support to undertake family work such as FT, it is still important for practitioners to refer relevant families (parents and children) to online resources such as Emerging Minds[1] and to family supports/services in the community.

The longer-term sustainability of FFP in Ireland, and elsewhere, requires a multi-level public-health response to address enduring political, cultural, organisational, and family barriers to change. Such a response would include: "think family" policy/practise standards; dedicated funding for FFP; managerial support to implement FFP; initiatives to reduce mental health stigma and recruitment barriers; and a continuum of FFP to broaden its capacity to identify families (**Figure 2**). "Think Family" policy/practise standards include: mandatory auditing of the parenting status of adult mental health users, balancing the priority given to patient confidentiality with unmet family needs, increased collaboration between traditionally segregated AMHS and CAMHS services, and equipping clinicians with time and resources to undertake FFP (5, 33).

Although FT has been implemented in many countries, this is only the second qualitative analysis of practitioner experiences in implementing the programme. Therefore, further qualitative research of practitioner (and family) experiences is required across different cultural/policy contexts, disciplines and settings. Further research is also needed to identify measures and/or supports that might increase family engagement, including, for example, developing and evaluating training videos that teach recruitment strategies. In addition, the facilitators and barriers to implementation identified in this study (and other qualitative analyses) could be tested as moderator/mediator variables in quantitative research.

CONCLUSION

In order to develop FT, and more broadly FFP, beyond a small number of committed sites, its longer-term sustainability in Ireland (and elsewhere) requires a careful assessment of the perceived fit of interventions with organisational remit and capacity, and the development of a multi-level public-health response to address enduring political, cultural, organisational, and family barriers to change (**Figure 2**). While little is known to date about which specific factors are most effective in promoting FFP, it is likely that change across all levels is required as legislation/standards, or FFP training on their own, are not sufficient (18, 25, 57).

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by four Ethics Committees: the Social Research Ethics Committee in Maynooth University, Ireland (Reference Number SRESC-2018-100), the HSE Research Ethics Committee, Tusla Ethics Review Committee, and the Saint John of God's Research Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

CM, MF, and SMcG conceived and designed the study. CM conducted interviews and coded transcripts, with 25% of transcripts independently coded by MF, SMcGa, and SO'C. CM prepared the initial draft, with subsequent drafts undertaken by MF, with input from CM, SMcG, and SMcGa. All authors contributed to the article and approved the submitted version.

FUNDING

The PRIMERA research programme was funded by the Health Service Executive and the Maynooth University Higher Education Authority COVID-19 Costed Extension Fund.

ACKNOWLEDGMENTS

We are grateful to all participants who generously gave their time to participate in this study, and to all stakeholders involved in introducing and delivering Family Talk as part of the PRIMERA research programme.

---

[1] Emerging Minds website–https://emergingminds.com.au/.

REFERENCES

1. Maybery D, Reupert A, Patrick K. Prevalence of children whose parents have a mental illness. *Psychiatr Bull.* (2009) 33:22–6. doi: 10.1192/pb.bp.107.018861
2. Hosman CMM, van Doesum KMT, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Aust Adv Ment Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
3. Grant A, Reupert A. The impact of organizational factors and government policy on psychiatric nurses' family-focused practice with parents who have mental illness, their dependent children, and families in Ireland. *J Fam Nurs.* (2016) 22:199–223. doi: 10.1177/1074840716643770
4. Mulligan C, Furlong M, McGilloway S. Promoting and implementing family-focused interventions for families with parental mental illness: scoping and installation. *Adv Ment Health.* (2020) 18:202–16. doi: 10.1080/18387357.2019.1614466
5. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psych Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
6. Nicholson J. Parenting and recovery for mothers with mental disorders. In: Levin B, Becker M, editors. *A Public Health Perspective of Women's Mental Health*. New York, NY: Springer (2010). p. 359–72. doi: 10.1007/978-1-4419-1526-9_19
7. Nicholson J, Biebel K, Williams VF, Kkatz-Leavy J. *The Prevalence of Parenthood in Adults With Mental Illness: Implications for State and Federal Policy, Programs, and Providers*. Psychiatry Publication Press (2002) 153p. Available online at: http://escholarship.umassmed.edu/psych_pp/153
8. *United Nations Convention on the Rights of the Child. Treaty no. 27531.* United Nations Treaty Series (1989). p. 3-178. Available online at: https://treaties.un.org/doc/Treaties/1990/09/19900902%2003-14%20AM/Ch_IV_11p.pdf (accessed September, 5 2021).
9. Bee P, Bower P, Byford S, Churchill R, Calam R, Stallard P, et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. *Health Tech Assess.* (2014) 18:1–250. doi: 10.3310/hta18080
10. Siegenthaler E, Munder T, Egger M. Effects of preventive interventions in mentally ill parents on the mental health of the mental health of offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17. doi: 10.1016/j.jaac.2011.10.018
11. Nicholson J, de Girolamo G, Schrank B, eds. *Parents With Mental and/or Substance Use Disorders and Their Children.* Lausanne: Front Media SA (2020). doi: 10.3389/978-2-88963-383-8
12. Beardslee WR, Wright EJ, Gladstone TR, Forbes P. Longterm effects from a randomized trial of two public health preventive interventions for parental depression. *J Fam Psychol.* (2007) 21:703–13. doi: 10.1037/0893-3200.21.4.703
13. Solantaus T, Paavvonen EJ, Toikka S, Punamaki RL. Preventive interventions in families with parental depression. *Eur Child Adolesc Psychiatry.* (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
14. Christiansen H, Anding J, Schrott B, Röhrle B. Children of mentally ill parents—a pilot study of a group intervention program. *Front Psych.* (2015) 6:1494. doi: 10.3389/fpsyg.2015.01494
15. Beardslee WR, Solantaus TS, Morgan BS, Gladstone TR, Kowalenko NM. Preventive interventions for children of parents with depression: international perspectives. *Med J Aust.* (2013) 199 (Suppl. 3):23–5. doi: 10.5694/mja11.11289
16. Breitenstein SM, Gross D, Garvey CA, Hill C, Fogg L, and Resnick B. Implementation fidelity in community-based interventions. *Res Nurs Health.* (2010) 33:164–173. doi: 10.1002/nur.20373
17. Bauer MS, Kirchner J. Implementation science: what is it and why should I care? *Psych Res.* (2020) 283:112376. doi: 10.1016/j.psychres.2019.04.025
18. Fixsen DL, Naoom SF, Blase KA, Friedman RM. *Implementation Research: A Synthesis of the Literature*. Tampa, FL: National Implementation Research Network; Louise de la Parte Florida Mental Health Institute (2005).
19. Biebel K, Nicholson J, Woolsey K, Wolf T. Shifting an agency's paradigm: creating the capacity to intervene with parents with mental illness. *Amer J Psych Rehab.* (2016) 19:315–38. doi: 10.1080/15487768.2016.1231641
20. Gatsou L, Yates S, Goodrich N, Pearson D. The challenges presented by parental mental illness and the potential of a whole-family intervention to improve outcomes for families. *Child Fam Soc Work.* (2017) 22:388–97. doi: 10.1111/cfs.12254
21. Grant A, Reupert A, Maybery D, Goodyear M. Predictors and enablers of mental health nurses' family-focused practice. *Int J Ment Health Nurs.* (2019) 28:140–51. doi: 10.1111/inm.12503
22. Karibi H, Arblaster K. Clinician experiences of "let's talk about children" training and implementation to support families affected by parental mental illness. *J Ment Health Train.* (2019) 14:201–11. doi: 10.1108/JMHTEP-08-2018-0044
23. Ward B, Reupert A, McCormick F, Waller S, Kidd S. Family-focused practice within a recovery framework: practitioners' qualitative perspectives. *BMC Health Serv Res.* (2017) 17:234. doi: 10.1186/s12913-017-2146-y
24. Falkov A, Goodyear M, Hosman CM, Biebel K, Skogøy BE, Kowalenko N, et al. A systems approach to enhance global efforts to implement family-focused mental health interventions. *Child Youth Ser.* (2016) 37:175–93. doi: 10.1080/0145935X.2016.1104104
25. Lauritzen C, Reedtz C, Rognmo K, Nilsen MA, Walstad A. Identification of and support for children of mentally ill parents: a 5 year follow-up study of adult mental health services. *Front Psychiatry.* (2018) 9:507. doi: 10.3389/fpsyt.2018.00507
26. Grant A, Lagson S, Devaney J, Davidson G, Duffy J, Perra O, et al. *A Study of Health and Social Care Professionals' Family Focused Practice With Parents Who Have Mental Illness, Their Children and Families in Northern Ireland.* Final Report. Belfast: Queen's University Belfast (2018) Available online at: https://pureadmin.qub.ac.uk/ws/portalfiles/portal/154075911/Think_Family_Final_Long_Report_27.2.18_Final_Final.pdf (accessed September 20, 2021).
27. Beardslee WR. Master clinician review: parental depression and family health and wellness: what clinicians can do and reflections on opportunities for the future. *J Am Acad Child Adolesc Psychiatry.* (2019) 58:759–67. doi: 10.1016/j.jaac.2019.02.008
28. Pihkala H, Johansson EE. Longing and fearing for dialogue with children—depressed parents' way into Beardslee's preventive family intervention. *Nordic J Psych.* (2008) 62:399–404. doi: 10.1080/08039480801984800
29. Pihkala H, Sandlund M, Cederström A. Children in Beardslee's family intervention: relieved by understanding of parental mental illness. *Int J Soc Psych.* (2012) 58:623–8. doi: 10.1177/0020764011419055
30. Pihkala H, Dimova-Bränström N, Sandlund M. Talking about parental substance abuse with children: eight families' experiences of Beardslee's family intervention. *Nordic J Psych.* (2017) 71:395–401. doi: 10.1080/08039488.2017.1308009
31. Pihkala H, Sandlund M, Cederström A. Initiating communication about parental mental illness in families: an issue of confidence and security. *Int J Soc. Psych.* (2012) 58:258–65. doi: 10.1177/0020764010392088
32. Strand J, Boström P, Grip K. Parents' descriptions of how their psychosis affects parenting. *J Child Fam Stud.* (2020) 29:620–31. doi: 10.1007/s10826-019-01605-3
33. Goodyear M, Hill TL, Allchin B, McCormick F, Hine R, Cuff R, et al. Standards of practice for the adult mental health workforce: meeting the needs of families where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120
34. Solantaus T, Toikka S, Alasuutari M, Beardslee WR, Paavonen EJ. Safety, feasibility and family experiences of preventive interventions for children and families with parental depression. *Int Mental Health Promo.* (2009) 11:15–24. doi: 10.1080/14623730.2009.9721796
35. Tew J, Morris K, White S, Featherstone B, Fenton SJ. *What Has Happened to 'Think Family'–Challenges and Achievements in Implementing Family Inclusive Practice.* Pavilion Publishing and Media (2015). Available online at: http://eprints.hud.ac.uk/id/eprint/26918/ (accessed September 20, 2021).
36. Reedtz C, van Doesum K, Signorini G, Lauritzen C, van Amelsvoort T, Van Santvoort F, et al. Promotion of wellbeing for children of parents with mental illness: a model protocol for research and intervention. *Front Psychiatry.* (2019) 10:606. doi: 10.3389/fpsyt.2019.00606
37. Department of Health. (2020). *Budget 2021: €4 Billion to Protect, Reform and Expand Health and Social Care Services and Implement Universal Healthcare.*

Available online at: https://www.gov.ie/en/press-release/3f618-budget-2021-4-billion-to-protect-reform-and-expand-health-and-social-care-services-and-implement-universal-healthcare/# (accessed September 5, 2021).

38. Department of Health and Children. *'A vision for Change' Report of the Expert Group on Mental Health Policy.* (2006). Available online at: http://hdl.handle.net/10147/43357 (accessed September 5, 2021).
39. Health Research Board. *HRB Statistics Series 40: Irish Psychiatric Units and Hospitals Census 2019 Main Findings*. HRB (2019). Available online at: https://www.hrb.ie/publications/publication/irish-psychiatric-units-and-hospitals-census-2019-main-findings/returnPage/1/ (accessed September 5, 2021).
40. Furlong M, McGilloway S, Mulligan C, McGuinness, C, Whelan, et al. Family talk versus usual services in improving child and family psychosocial functioning in families with parental mental illness (PRIMERA—promoting research and innovation in mental hEalth seRvices for fAmilies and children): study protocol for a randomised controlled trial. *Trials.* (2021) 22:243. doi: 10.1186/s13063-021-05199-4
41. Charmaz K. *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. London: Sage (2006).
42. Moore G, Audrey S, Barker M, Bond L, Bonell C, Hardeman W, et al. *Process Evaluation of Complex Interventions*. London: UK medical Research Council (MRC) guidance (2014).
43. Central Statistics Office, 2021. *Survey on Income and Living Conditions (SILC).* (2019). Available online at: https://www.cso.ie/en/releasesandpublications/ep/p-silc/surveyonincomeandlivingconditionssilc2019/povertyanddeprivation/ (accessed September 5, 2021).
44. VERBI Software. *MAXQDA 2020. Software.* (2019). Available online at: http://www.maxqda.com (accessed March 30, 2020).
45. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care.* (2007) 19:349–57. doi: 10.1093/intqhc/mzm042
46. Furlong M, McGilloway S, Mulligan C, Killion MG, McGarr S, Grant A, et al. Covid-19 and families with parental mental illness: crisis and opportunity. *Front Psychiatry.* (2021) 12:567447. doi: 10.3389/fpsyt.2021.567447
47. Strand J, Rudolfsson L. A qualitative evaluation of professionals' experiences of conducting beardslee's family intervention in families with parental psychosis. *Int J Ment Health Promo.* (2017) 19:289–300. doi: 10.1080/14623730.2017.1345690
48. Maybery D, Goodyear M, Reupert AE, Grant A. Worker, workplace or families: what influences family focused practices in adult mental health? *J Psych Ment Health Nurs.* (2016) 23:163–71. doi: 10.1111/jpm.12294
49. Lauritzen C, Reedtz C, Van Doesum KT, Martinussen M. Implementing new routines in adult mental health care to identify and support children of mentally ill parents. *BMC Health Ser Res.* (2014) 14:58. doi: 10.1186/1472-6963-14-58
50. Van Doesum KT, Riebschleger J, Carroll J, Grové C, Lauritzen C, Mordoch E, et al. Successful recruitment strategies for prevention programs targeting children of parents with mental health challenges: an international study. *Child Youth Ser.* (2016) 37:156–74. doi: 10.1080/0145935X.2016.1104075
51. Beardslee WR, Wright E, Rothberg PC, Salt P, Versage E. Response of families to two preventive intervention strategies: long-term differences in behavior and attitude change. *J Am Academy Child Adol Psych.* (1996) 35:774–82. doi: 10.1097/00004583-199606000-00017
52. Giannakopoulos G, Solantaus T, Tzavara C, Kolaitis G. Mental health promotion and prevention interventions in families with parental depression: a randomized controlled trial. *J Affect Disor.* (2021) 278:114–21. doi: 10.1016/j.jad.2020.09.070
53. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Family options for parents with mental illnesses: a developmental, mixed methods pilot study. *Psychiatr Rehab J.* (2009) 33:106. doi: 10.2975/33.2.2009.106.114
54. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child Youth Ser.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
55. Mental Health Foundation. *The COVID-19 Pandemic, Financial Inequality and Mental Health.* (2020). Available online at: https://www.mentalhealth.org.uk/our-work/research/coronavirus-mental-health-pandemic/covid-19-inequality-briefing (accessed May 28, 2021).
56. Kelly, BD. Impact of Covid-19 on mental health in ireland: evidence to date. *Iran Med J.* (2020) 113:214. Available online at: http://imj.ie/wp-content/uploads/2020/12/Impact-of-Covid-19-on-Mental-Health-in-Ireland-Evidence-to-Date.pdf (accessed September 23:2021).
57. Goodyear M, McDonald M, Von Doussa H, Cuff R, Dunlop B. Meeting the intergenerational needs of families where a parent has a mental illness. *J Parent Fam Ment Health.* (2018) 3:e1011. doi: 10.7191/parentandfamily.1011

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 23 November 2021
doi: 10.3389/fpsyt.2021.760716

Enabling Transdisciplinary Collaboration: Stakeholder Views on Working With "Children With Mentally Ill Parents" Research Groups

Raphaela E. Kaisler and Christiane Grill*

LBG Open Innovation in Science Center, Ludwig Boltzmann Gesellschaft (LBG), Vienna, Austria

Working collaboratively and openly together with stakeholders has become a common phenomenon in research. While previous studies have gathered a clear picture on researchers' attitudes, motivations, and barriers for actively involving stakeholders in transdisciplinary research, the stakeholder perspective is yet unknown. Therefore, this paper sets out to identify how stakeholders perceive transdisciplinary collaborations with researchers. This paper in particular reveals the enablers and barriers for such collaborations from the viewpoint of stakeholders. To do so, we look at how stakeholders, who were actively involved in the governance structure of two "children with mentally ill parents" research groups in Austria, perceived their collaboration with researchers. We used a mixed-method, quantitative-qualitative design. We conducted an online survey and interviews with the members of the advisory board and competence group. These stakeholders reported great satisfaction with the transdisciplinary collaboration and emphasized the value of different expertise. As the most important enablers for successful, transdisciplinary collaboration stakeholders emphasized researchers' open-mindedness toward new perspectives and approaches, flexibility to adapt to the research process along the way, and creativity dealing with diverse backgrounds and skills. Stakeholders further underlined the importance of a person facilitating the collaboration process between researchers and stakeholders to resolve any tensions and insecurities. Concluding, researchers' attitudes, and in particular their understanding of the value of stakeholder involvement in research are key enablers for successful transdisciplinary research collaborations.

Keywords: transdisciplinary collaboration, patient and public involvement, stakeholder engagement, open innovation, children of parents with a mental illness (COPMI), mixed-methods design, transdisciplinary research teams

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Adele Parkinson,
Ara Institute of Canterbury,
New Zealand
Camilla Lauritzen,
Arctic University of Norway, Norway
Mary V. Seeman,
University of Toronto, Canada

****Correspondence:***
Raphaela E. Kaisler
raphaela.kaisler@lbg.ac.at

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *18 August 2021*
Accepted: *01 November 2021*
Published: *23 November 2021*

Citation:
Kaisler RE and Grill C (2021) Enabling Transdisciplinary Collaboration: Stakeholder Views on Working With "Children With Mentally Ill Parents" Research Groups.
Front. Psychiatry 12:760716.
doi: 10.3389/fpsyt.2021.760716

INTRODUCTION

The prevalence of children who live with a parent having a mental illness (COPMI) is about 25% worldwide (1–4). These children are at increased risk of long-term difficulties due to genetic, individual, family, and environmental risk factors (1, 5–8). Specific interventions related to family, social support and community have been shown to make a difference to the selected target groups (children, parents) and settings [psychiatry, community; (9–13)]. Research identified several external factors, governing service practices and the context itself as enablers for a successful implementation of COPMI interventions and services (14–17). Specifically regarding the context, professional influences (i.e., formal and informal norms, rules, policies, standards guiding the professionalization of individuals involved in the implementation) and the social climate (i.e., beliefs, values, customs and practices of the larger community, the system within which the intervention is embedded) are essential. For example, Zeichmeister-Koss et al. (18) recently analyzed the situational context and services of COPMI in the pilot region Tyrol in Austria. The authors found a lack of support processes and standards to meet children's needs and highlighted the gap between research and practice in the Austrian COPMI field.

This gap is not specific to the COPMI field. It generally points to a problem of inter- and transdisciplinary collaborations between researchers, stakeholders, and service user groups [e.g., patients, children and their families; (19)]. Interdisciplinary and transdisciplinary research performance and evaluation are both generative processes of harvesting, capitalizing, and leveraging multiple expertise (20). Here, we distinguish between interdisciplinary research as collaboration between different research disciplines, and transdisciplinary research as work between different research disciplines and stakeholders, such as practitioners, clinicians, patients, people with lived experience in mental illness and health, and family members.

In this article, we now describe and reflect how stakeholders perceive transdisciplinary collaborations with researchers. We analyze the enablers and barriers for such collaborations from the viewpoint of stakeholders. To do so, we look at how stakeholders who were actively involved in the governance structure of two COPMI research groups in Austria perceived their collaboration with researchers.

Perceptions of Transdisciplinary Collaboration

Working collaboratively and openly together with stakeholders across transdisciplinary boundaries has become a common phenomenon in research (21). In the last few years, the importance of involving patients and other stakeholders in health-related research has steadily been growing in the UK (22) as well as worldwide (23–25). Patient and public involvement (PPI) refers to meaningful and active involvement of patients and members of the public in research activities and processes. Consequently, research is carried out "with" or "by" members of the public rather than "to", "about", or "for" them (22, 26). "By involving patients in their research, researchers learn from other people's experience, which then changes their own thinking, values, choices, and actions. This leads to the commonly reported outcomes of involvement—improved research design, delivery, and dissemination—and over time, the wider impacts of a changed research culture and agenda (27)." Public involvement in health-related research has shown that patients and members of the public are indeed able to successfully contribute to specific research problems as well as able to find innovative solutions, for example, via setting research priorities (28), co-producing knowledge (29, 30) or via shaping health care services (31). In line with this, several systematic reviews (32–36) have reported that stakeholder involvement makes a difference to the people affected. However, this type of involvement is also criticized of being weak and anecdotal. Criticism has particularly focused on the lack of empirical data to evaluate impact, the insufficient attention that is paid to the context in which involvement takes place, and the way involvement is actually lived (37). To counteract this criticism and ultimately to avoid tokenistic involvement of stakeholders in research, it is therefore crucial to determine "why" and "who" should be involved at all in research and to acknowledge the experiential knowledge that stakeholders bring to the table. In doing so, active involvement of stakeholders in research may ultimately maximize the opportunities of learning, increase the likelihood of impact, and help to achieve the goal of improved services to the affected community (38).

How researchers perceive transdisciplinary research by involving patients and the public has already been well-studied. Several studies have analyzed researchers' attitudes and motivations for working transdisciplinary with stakeholders. While researchers highlighted the potential benefits of involving the public, they yet expressed strong ambivalence regarding the exact purpose and value of patient and public involvement (19, 39, 40). Furthermore, a few studies have also assessed researchers' viewpoints regarding the barriers that hinder transdisciplinary stakeholder involvement. These studies identified a mix of barriers; particularly, lack of funding, time, and skills, finding the "right" people, organizational and policy barriers, research fatigue, group dynamics (41, 42), researchers' negative attitudes toward PPI (43) and personality characteristics (44). In a recent systematic review of reviews, Ocloo et al. (45) summarized various enablers and barriers of PPI in health and social research from the viewpoint of researchers. These were personal/individual factors, patient/relative involvement and attitudes, health professional relationships with patients, clarity of roles and expectations, knowledge, information and communication, financial compensation and resources, training, general support, power dynamics and organizational constraints, recruitment, and community approach.

How patients and the public perceive transdisciplinary collaboration with researchers is, however, unclear. Thus far, there exists no study analyzing how stakeholders involved in health research view their involvement in research. Therefore, this paper sets out to identify how stakeholders perceive transdisciplinary collaborations with researchers. We are hereby in particular interested in exploring the enablers and barriers of transdisciplinary research collaboration from a stakeholder perspective.

Our Transdisciplinary Collaboration Approach in the Copmi Field

In a first step, the Ludwig Boltzmann Gesellschaft (LBG) launched the crowdsourcing project "Tell Us! What Questions about Mental Illness Should Science Take Up?" (46). The entire health care community in Austria (i.e., patients, family members, and health care professionals) was invited to submit research questions for the field of mental health. After analyzing and thematically collating 400 high-quality submissions, 17 topics were distilled. Out of these 17 topics, a focus on "children of mentally ill parents" (COPMI) emerged as the top research priority. Based on this outcome, 136 PhD students and post-doctoral researchers were invited to an "Ideas Lab": 29 researchers participated in an "Ideas Lab on COPMI" (47). Two people with lived experience were invited to the Ideas Lab to share their experience as children of mentally ill parents, and to ultimately inspire researchers for future research. As an outcome of the Ideas Lab, two research groups "DOT—The Open Door" (48) and "Village—How to Raise a Village to Raise a Child" (49) were established. "DOT" focuses on early adolescents making the difficult leap from primary to secondary school and how supportive relationships between peers help children stay mentally and physically healthy. "Village" aims to strengthen formal and informal support structures around the child through enhancing their village of collaborative care. A relationship manager supported the research groups to establish community and stakeholder interactions, foster patient and public involvement activities, and to accompany them over the 4-year funding period (in total six million Euro).

To ensure transdisciplinary collaboration with stakeholders, the LBG introduced a novel governance structure for the two research groups. Two advisory groups and a competence group consisting of COPMI stakeholders were established for the two research groups. The advisory board each consisted of three scientific experts from different fields (e.g., psychiatry, psychology, implementation science, linguistics, gamification), two adults who lived with a parent with mental illness in their childhood, and an open innovation expert. The advisory boards discussed the research groups' achievements as well as their outlook for the future. In total, the advisory board met six times over the period of 4 years. Due to the COVID-19 pandemic, half of the meetings were held online in 2020 and 2021 via the Zoom video conferencing platform. The competence group consisted of five people (20–30 years old) who lived with a parent with mental illness in their childhood and had various professional backgrounds (e.g., in social work, art, public health, education). The competence group received an honorarium for their contributions and met on average 10 times a year to advice on the research groups' project design, methods, results, and dissemination strategies. The meetings were shifted online in 2020 and 2021 due to the COVID-19 pandemic.

Aim of This Study

In this study, we analyze how stakeholders who were involved as advisory board or competence groups members in the two research groups "DOT—The Open Door" and "Village—How to Raise the Village to Raise the Child" perceive transdisciplinary collaborations with researchers. We are interested in stakeholder views since their perspective on transdisciplinary collaboration has been neglected in health-related research thus far. Furthermore, we identify enablers and barriers for transdisciplinary collaborations between researchers and stakeholders. Therefore, this study sets out to answer the following two research questions: How do stakeholders perceive transdisciplinary collaboration with researchers? What are the enablers and barriers for successful, transdisciplinary research collaborations?

METHODS

To answer the two research questions, we used a mixed-methods, quantitative-qualitative design. First, all advisory board and competence group members were asked to fill in a questionnaire. This first step aimed to reveal the stakeholders' general perceptions of transdisciplinary collaboration. In a subsequent step, we wanted to gain more in-depth insights, thoughts, and reasons of the stakeholders involved in research. Therefore, we conducted semi-structured interviews with purposefully selected advisory board and competence group members.

Survey

Participants

All 13 advisory board members (thereof three males) and all six competence group members (all females) of the two research groups "DOT—The Open Door" and "Village—How to Raise the Village to Raise the Child" were invited to fill in an online survey.

Procedure

The questionnaire was designed with the online survey tool Unipark® (Tivian). An anonymously link to the survey was sent to the members via a personalized email explaining the objective and rationale of the study and asking them to complete a 7-min-long survey. The survey link was open for 6 weeks from April 8 to May 18, 2021. Various reminders were sent via email throughout the 6 weeks. Responses to the survey were then quantitatively analyzed.

Measures

After agreeing to the informed consent, respondents were asked a range of closed-ended questions and one open-ended question. Questions addressed the following themes: the general setup of the meetings (e.g., frequency, preparation material), the structure of the advisory and competence groups (e.g., different expertise), the quality of involvement (e.g., atmosphere, contributions), and the collaboration with researchers. To measure each theme thoroughly, two to six statements were formulated for each theme and respondents were asked to indicate their agreement with each statement along a 5-point Likert scale (1 = do not agree at all −5 = fully agree). Respondents were also asked about their overall satisfaction with the structure of the advisory boards and competence groups and the development of the research group (5-point Likert scale: 1 = not at all satisfied −5 = fully satisfied). Respondents were also asked in how far they would recommend others to participate in such advisory boards and competence groups (5-point Likert scale: 1 = not at all recommended −5 = very much recommended). Lastly, in an open-ended question, respondents were asked about their overall impression of their work.

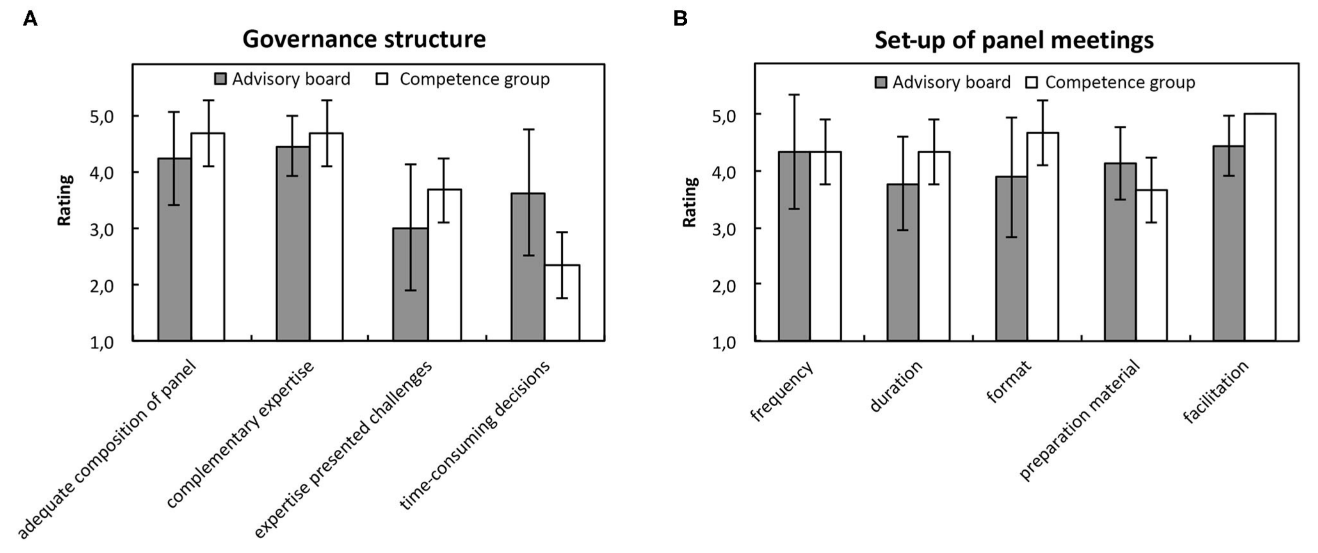


FIGURE 1 | Structure of the panel meetings. **(A)** Shows average ratings of the governance structure and **(B)** the set-up of panel meetings in the online survey. Gray boxes indicate ratings of the advisory board members and white boxes indicate ratings of the competence group members.

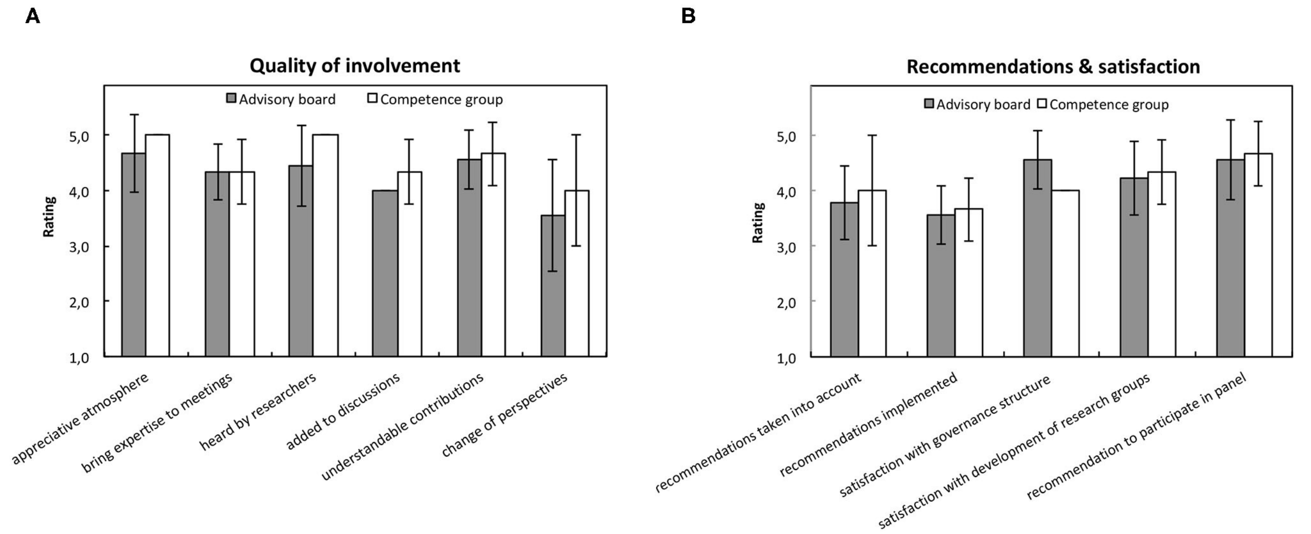


FIGURE 2 | Quality of involvement in the panel meetings. **(A)** Shows average ratings of the quality of involvement and **(B)** recommendations and satisfaction with the panel meetings in the online survey. Gray boxes indicate ratings of the advisory board members and white boxes indicate ratings of the competence group members.

Interviews

Participants

Four advisory board members (one adult who lived with a parent with mental illness in their childhood, one expert in open innovation in science, two experts from the field of psychology) and two competence group members (two adults who lived with a parent with mental illness in their childhood) were asked to be interviewed. Interviewees were selected based on their role in the advisory board and competence group. Among the invited interviewees were two men and four women.

Procedure

Questions for a 1-h long, semi-structured interview were designed and personalized invitation emails explaining the objective and rationale of the interview were sent out. Prior to the interview, the interviewer explained the procedure to the interviewees and obtained written, informed consent in accordance with the ethical guidelines in Austria and the Declaration of Helsinki. All interviewees gave informed consent to be recorded and to publish the data. All interviews were then held online via Zoom and transcribed. The transcripts were then anonymized: all identifying information was removed from the transcripts. The data was then analyzed using thematic analysis.

Interview Guide

The semi-structured interviews covered a range of different topics. These were the interviewee's role in the advisory board or competence group, the collaboration with the researchers (particularly, the joint development of approaches, the integration of different perspective, the challenges for researchers, and differences to rather traditional approaches), and the enablers and barriers for successful, transdisciplinary collaboration.

RESULTS

Survey

Nine advisory board members (response rate: 69%) and three competence group members (response rate: 50%) completed the online survey. Due to the small sample size, we conducted a descriptive, univariate analysis. Here, we report the means (M) and standard deviations (SD) for each survey item (**Supplementary Table 1**).

As to the general setup of the meetings, both, members of the advisory board and members of the competence group, assessed the frequency (M = 4.3, SD = 0.9), duration (M = 3.9, SD = 0.8), format (M = 4.1, SD = 1.0), preparation material (M = 4.0, SD = 0.6), and particularly the facilitation (M = 4.6, SD = 0.05) of the meetings very positively (**Figure 1A**).

Overall, respondents rated the structure of the advisory board and competence group as adequate (**Figure 1B**). Specifically, the composition of these two panels was very adequate for the research groups (M = 4.3, SD = 1.0), and the different expertise on the panels very well-complemented each other (M = 4.5, SD = 0.5). Simultaneously, however, the different expertise among the members of the panels presented challenges (M = 3.2, SD = 1.0) and resulted in more time-consuming decisions (M = 3.3, SD = 1.0). Particularly, the members of the competence group assessed the challenges (M = 3.7, SD = 0.6) and time consumption (M = 3.6, SD = 0.6) of their work due to different expertise slightly more critically than the members of the advisory board (challenges: M = 3.0, SD = 1.1, time: M = 2.3, SD = 0.6).

Moreover, the quality of involvement was rated positively (**Figure 2A**). The atmosphere in the panels was appreciative (M = 4.8, SD = 0.6), members were able to bring their expertise to the meetings (M = 4.3, SD = 0.5), contributions were understandable and comprehensible (M = 4.6, SD = 0.5), the contributions were heard by the researchers (M = 4.6, SD = 0.7), and they added to the discussions (M = 4.1, SD = 0.3). The contributions of the different members also sometimes led to a change of one's own perspective (M = 3.7, SD = 1.0). Overall, members of the competence group assessed all quality aspects of their involvement slightly better than the advisory board members did; especially being heard by the researchers (competence group: M = 5.0, SD = 0.0, advisory group: M = 4.4, SD = 0.7).

The collaboration between the members of the advisory board and the competence group and the researchers was assessed slightly mixed (**Figure 2B**). Respondents were rather indifferent whether their recommendations were considered (M = 3.8, SD = 0.7) and ultimately implemented by the research groups (M = 3.6, SD = 0.5).

Overall, respondents were very satisfied with the structure of the panels (M = 4.4, SD = 0.5) as well as with the development of the research groups (M = 4.3, SD = 0.6), and strongly recommended to participate in such panels (M = 4.6, SD = 0.7).

When asked for their overall impression of their work in the panels in the open-ended question, respondents stated that they learnt a lot and enjoyed being part of the research projects.

Interviews

To gain more in-depth insights, thoughts, and reasons about how stakeholders perceive transdisciplinary collaboration with researchers, we conducted semi-structured interviews. Five interviews were conducted: namely with two competence group members (i.e., children of parents with mental illnesses) and three advisory board members (one adult who lived with a parent with mental illness in the childhood, one expert in open innovation in science, one expert from the field of psychology).

The Role of the Advisory Board and the Competence Group

The various members perceived their roles very differently: For the competence group members, it was clear from the beginning what role they would play. The fact that they were asked to work as children of a mentally ill parent for the two research groups, was a sign of incredible appreciation for them: appreciation not only for a Research Topic that was so dear to their heart but also appreciation of their very personal, lived experience that could enrich research in many multi-faceted ways. The two projects were "*… for someone who is affected a sign of incredible appreciation, because you see the issue that is important to you and with which you, as an affected person identify is being taken up, is seen, money is put into it, something is set in motion.*" The competence group members aimed to shape the research as actively as possible so that on the one hand scarce resources (particularly, money and personnel) were used most effectively and on the other hand that as much research as possible could be done on an under-researched topic. The members aimed to bring as much experiential knowledge to the table as possible and wanted to enrich the project with constructive feedback that often turned out to be quite critical. Among the competence group members, the atmosphere was described as very harmonic and empathetic. They experienced an immediate bond between them right from the beginning due to their background as children of parents with mental illnesses.

For the advisory board members, in contrast, it was not that clear how exactly they could support the research groups. No one had a concrete idea of their role at the beginning. It took some time to figure out what each member could contribute to the research groups. "*You don't necessarily have an idea at the beginning. You enter a new setting, which was developed by the open innovation in science [approach] itself. And you first have to orient yourself, so to speak. You try to find out, so to speak, what my role could be. What can I contribute in* *relation to other participants?*" Over time, however, everyone grew into his/her role. All members aimed to support the researchers as much as possible and to give them constructive and helpful advice—without judging any of their decisions as right or wrong, or good or bad. The fact that the advisory board itself was multidisciplinary was seen as a particular enrichment: The members felt it important to share their perspectives, and to bring their experience and skillset to the table even if it was sometimes quite challenging to funnel the various input and expectations for the projects into specific recommendations. Generally, the disciplinary differences and the different expectations of the advisory board members about what research should achieve resulted in some disagreement among themselves. Nonetheless, these differences gave rise to mutual learning and richness for the whole process.

Collaboration With Researchers

As to the collaboration with the researchers, the competence group members perceived great insecurity on the part of the researchers on how to interact and work with them. Researchers seemed to no have an idea how the collaboration with a competence group should look like. "*Some researchers were like clumsy puppies trying to grasp us. Who are they? How do I deal with them? Also fears, fears of contact.*" Therefore, in the beginning, different forms of collaboration developed. "*Some researchers were very open and very appreciative of the competence members' experiential knowledge from the beginning; they actively asked for feedback and carefully listened to the competence members' feedback. Others were more at loss what to do with the competence group, just told the group what they planned to do without asking for feedback, and overall gave the impression that the competence group members first needed to prove themselves and their value to the project and the research.*" "*Some were able to perceive this more as constructive for themselves and as enrichment. And for others, it was the case that the perception of others, the evaluation and the defense played a greater role and that one then insisted more strongly on emphasizing the autonomy of the researcher.*" "*It really depended on the personality of the researchers to what extent they were able to accept feedback.*"

Competence group members also reported that, over time, both sides started to realize that they could learn a lot from each other due to their various backgrounds, trainings, and skillsets, and that seriously and actively engaging the competence group was an incredible benefit. Particularly for the methodological design and data analysis, the perspectives and the experiential knowledge of the competence group seemed to enrich the projects tremendously. "*We discussed the data analysis and afterwards the researcher was really happy and completely flabbergasted. She said that she had a problem with the data because the data were contradictory for her. And we were all able to say unanimously that's completely logical and gave examples and then she was like: that makes so much sense now. And I thought, yes, that's exactly why I think it's important that we are involved in the data analysis.*" However, the competence group members remained unsure until the end to what extent their feedback was indeed taken seriously and acted upon, and it was not just pure lip service from the researchers that the competence group's advice and feedback were valuable to the projects.

The advisory board members experienced the collaboration with the researchers as a balancing act. While the members always aimed to support the researchers in their plans so that they made progress and those resources were used efficiently and effectively, some researchers interpreted their advice and feedback as interference with their autonomy. It took a period of mutual learning from each other's expertise to reach a shared understanding of the conceptual frameworks and foci of the research projects. Altogether, the collaboration between the advisory board members and the researchers was considered productive and helpful. In retrospect, advisory members yet wished for more time and effort on the part of the researchers to establish and work on their relationships. "*I actually wish that we had been able to be more helpful to the project and I think that would have required more ongoing contact. It would have required the project team to have the time and effort to put into establishing and working in those relationships.*"

Joint Development of Approaches

Altogether, the competence group members were very open to jointly develop research approaches together with the researchers. At the same time, however, they were quite uncertain how much they were allowed to get involved into the project and particularly how much they were allowed to propose alternatives and changes to the planned research course since the basic research agenda was already defined in the Ideas Lab. How the joint development of research approaches exactly looked like, was dependent on the researcher. Competence group members reported that some researchers more strongly thought about deliverables and publications as the ultimate goals and involving the competence group was then more a box ticking exercise. Other researchers seemed to have an authentic interest in getting to know and integrating the perspectives of children of parents with mental illnesses. "*Some researchers thought only of their deliverables. Others went more into interaction and also showed: I am an interested researcher and I want to learn something from you and get something out of you. I want to experience your world.*"

The advisory board members were very open and interested in supporting and giving advice to the researchers regarding research approaches. Bringing the people together despite the internationally different time zones in which the members were located was sometimes quite challenging. In one of their meetings with the researchers, the advisory board members proactively addressed the issue of how they could be more helpful to the researchers and suggested that discussing specific questions that researchers had would be more productive than just being presented with what the researchers had been working on. While the advisory board members overall valued the whole process of being engaged and felt that their meetings were productive, they still had the impression that researchers could have reached out a little more. "*One of the things we brought up in one of our conversations was how can we be more helpful, are there specific issues that need to be addressed or specific questions that you have that we would be able to help you answer. I actually think the project team could have reached out a little more in that regard.*

I wish there had been more and better ways for us to be more helpful." Until the end, advisory board members were never quite certain whether and to which extent researchers followed up on their recommendations: "*I think we gave them good advice. It's not clear to me that they ever took our advice*." The members reported that they never received feedback on which piece of advice worked and which piece did not work. They felt that this would have required more communication, time, and effort on the researchers' part. At the same time, however, the advisory members admitted that they never actively solicited this type of communication.

Integration of Different Perspectives

When it came to the integration of different perspectives, the competence group members always got the feeling that their perspectives and their experiential knowledge were heard, considered, and implemented. The members also reported that actual feedback loops were missing. The members felt that researchers did not update them in the meetings whether and to what extent their feedback was indeed implemented. However, they also admitted that they never actively asked for feedback loops. They also reported that it took them a lot of energy to make themselves heard and to convince researchers that their experiential knowledge also counts and not only the researchers' formally learned knowledge. "*It also took energy to keep pointing the finger and being critical. And I think that also has a lot to do with values. Without a title, I don't really have much of a say in this whole machinery with my expertise, which is 'only' based on personal experience. It doesn't have the same status.*"

The advisory board members felt that openness to integrate different perspectives in their work very much varied among the researchers. Some researchers seemed to present their work as already on track, so no advice was needed from the advisory board. Other researchers seemed to perceive the advice from the advisory board as unjustified criticism of their work and interference with their work and were not willing to engage with the advisory board on a profound level. Other researchers in turn were extremely open toward the feedback of the advisory board and valued their perspective from the outside. The members reported that researchers also opened up about the various challenges (i.e., staffing, budget, administration) that they were facing on a day-to-day basis, which in turn helped the advisory board to better understand specific decisions and approaches from part of the researchers, and ultimately helped the advisory board to give advice that was more helpful. "*I feel like I had respect for and an understanding of people's different perspectives. You need to choose people with diverse expertise but who are open to different perspectives, and who are willing to brainstorm about different the application of different perspectives, and what that suggests in terms of recommendations and outcomes*." Overall, the advisory board members felt that their success was very much dependent on how the researchers perceived the board's role and their advice and how much the researchers themselves were willing to listen and reflect.

Challenges for Researchers

From the perspective of the competence group, the biggest challenge the researchers were facing in their transdisciplinary work was the integration of the various perspectives and skillsets. Not only the researchers brought different trainings, perspectives, and skills to the table. Also, the members of the competence group were just not only children of parents with mental illnesses, but they also brought professional trainings and resources with them. This heterogeneity of trainings, perspectives, and skills was a huge enrichment for the research process but made everything also more complex. "*This transdisciplinarity is the work of the now and the future. These many perspectives that come in. They're a huge enrichment; they also make it more complex, of course. Because I go far beyond the level of content*."

Advisory board members named complexity as the biggest challenge for researchers. Specifically, the biggest obstacles were the complexity to integrate the various perspectives and to agree to a research agenda that everyone could support. It was only when researchers began to communicate these difficulties honestly and openly that the advisory board members felt that they could give good advice. This kind of open and honest communication about research challenges fostered mutual learning on all sides, according to advisory board members. Advisory board members also encouraged the researchers to be courageous, to meet the challenges head on and to not try to do everything perfectly: "*You can simply be courageous. The challenges and the problems that arise, and also to enter into the debate. And not to think that I have to sweep it under the carpet, that everything has to be perfect and so on. That was always my role, to be honest and open, and then others can get on board and learn something from you*." From the advisory board's view, the competence group presented another line of conflict. As research was done on one's own problem, debates about the adequate research process were often highly emotional.

Differences to Traditional Approaches

The competence group members felt that—compared to traditional research approaches—the transdisciplinary collaboration allowed a much more inclusive process. Researchers and competence group members seemed to mutually learn from each other and influence each other so that the research projects could indeed exert enduring and positive impact on the lives of children with mentally ill parents.

The advisory board observed five major differences that uniquely distinguished the transdisciplinary approach from more traditional research approaches: variety of perspectives, flexibility, community work, impact, and boldness of the funder. The members reported that firstly, the variety of perspectives arose not only from the transdisciplinary team of researchers but also the transdisciplinary nature of the competence group and the advisory board. Many different perspectives, trainings and skillsets enriched the whole research process in various ways. Secondly, the transdisciplinary approach allowed a flexibility to reflect on the whole research process and to adapt goals and methods along the way. Advisory board members were certain that such kind of flexibility would not have been possible in traditional research approaches where researchers worked through the work packages as they were described in the research proposal. Thirdly, advisory board members felt that involving children of parents with mental illnesses in the research process laid the foundation for community work. Researchers went out to the communities to involve the various stakeholders and to integrate their perspectives. Researchers themselves seemed to learn from the communities in an iterative process. "*Community work is so much harder and takes so much more time and is so much more challenging. So, the metrics that you use for evaluating success of this initiative need to reflect the fact not only that it's the open innovation business, but also that it's so community-based.*" Fourthly, advisory board members reported that the impact that a research project using a transdisciplinary approach could have, was very different from the impact that traditional research had. Not the number of published papers or the number of citations counted, but how many people had been touched by the research projects mattered: "*you may need to think carefully about things like number of people's lives you've touched, number of kids involved, number of kids who participated in making the project happen, number of families who have been touched in some way, number of other kinds of stakeholders/providers. You may want to think of your social media posts and the volume of likes or shares.*" Fifthly and lastly, the advisory board members mentioned the boldness of the funder to not only provide substantial funding for projects that have never been carried out in this way, but also to provide strong organizational support that accompanied the projects.

Enablers and Barriers for Successful, Transdisciplinary Collaboration

As to successful transdisciplinary collaboration, the competence group members named various enablers and barriers. Firstly, competence group members reported that transdisciplinary collaboration needed regular exchange with the whole group. Sometimes the competence group only met with some researchers but not the whole research group, which led them to focus too much on details and lose sight of the big picture. In connection to this, the competence group recommended children of parents with mental illnesses as co-researchers who were actively involved in the research process. In doing so, these experts would not be seen as some foreign parts loosely attached to the research but as a permanent and equal part of the research team itself. Thirdly, competence group members felt that transdisciplinary collaboration needed a connector—a person positioned between children of parents with mental illnesses and the researchers, who spoke both languages, knew how to mediate the different perspectives, and was convinced that transdisciplinary collaboration benefited research and society. "*For me it was a key person in the process, an excellent link between the structures. And I think the format always needs someone who carries it and who carries the format with him and says, this is so important, I live this authentically and embody this.*" The fourth enabler for successful, transdisciplinary collaboration between researchers and stakeholders that competence group members mentioned was an open mindset. All people involved along the various research stages were asked to have an open mindset. They needed to be open-minded to engage with each other, to learn from each other and to accept that sometimes research does not evolve as planned and approaches need to be adapted. Lastly, transdisciplinary collaboration needed quick wins: rapid results that were tangible for those affected so that they could see that researchers made progress, and that progress positively affected their lives.

The advisory board members also mentioned that having children of parents with mental illnesses as co-researchers in the research team would certainly promote transdisciplinary collaboration. In addition, an open, flexible, and creative mindset contributed to the success of such collaboration according to advisory board members. Everybody involved needed to be open-minded toward new perspectives and approaches, flexible to adapt the research process along the way, and creative in dealing with the different perspectives, trainings, and skills that everyone brings to the table. Additional enablers for successful, transdisciplinary collaboration from the perspective of the advisory board were early involvement, relationship management, and alternative dissemination forms. The advisory members suggested that everybody who needed to be involved in the research project should be involved as early as possible. In fact, already in the Ideas Lab those affected should be involved so that they could gain an understanding and insights into how the idea for the specific project developed.

Furthermore, relationship management was mentioned as an indispensable pillar for transdisciplinary collaboration. As the various members of the research team, the advisory boards, and the competence groups hardly knew each other at the beginning, relationships needed to be built via social events and by sharing information and communicating with each other as much as possible. "*Relationships matter and communication matters and information sharing matters. Some of the biggest challenges have been around this issue of communication and sharing information.*" Lastly, advisory board members reported that research results should not only be disseminated via the traditional ways like publications and conference presentations but also via new and innovative ways that most likely reached those concerned, for instance via community outreach events.

Members also reported that successful, transdisciplinary collaboration started with a clear commitment of the organization to support the transdisciplinary structure accompanied with the boldness to sufficiently fund such research and a dedicated person who managed knowledge and workflows between researchers and competence group and advisory board. Additionally, right from the beginning, everyone involved (i.e., researchers, members of the competence group and the advisory board) needed to be aware of what to expect from each other, and what trainings and skillsets everyone could bring to the table. Regarding the specific collaboration between researchers and competence group and advisory board members, the definition of some ground rules (like, how and when to ask for feedback) might become beneficial for productive, transdisciplinary collaboration. In this way, misunderstandings—particularly when it comes to advising researchers vs. interfering with research plans—can be eliminated right from the start.

TABLE 1 | Summary of enablers for successful, transdisciplinary collaboration.

Domain	Enablers	Examples from interviewees
Governance	Commitment and boldness of funders	…for someone who is affected a sign of incredible appreciation, because you see the issue that is important to you and with which you, as an affected person identify is being taken up, is seen, money is put into it, something is set in motion.
	Supporting interactions	For me it was a key person in the process, an excellent link between the structures. And I think the format always needs someone who carries it and who carries the format with him and says, this is so important, I live this authentically and embody this.
	Openness and flexibility to adaptations	You enter a new setting, which was developed by the OIS itself. And you first have to orient yourself, so to speak. You try to find out, so to speak, what my role could be. What can I contribute in relation to other participants?
Collaboration	Open-minded personality	They went more into interaction and also showed: I am an interested researcher and I want to learn something from you and get something out of you. I want to experience your world.
	Relationships and communication	Relationships matter and communication matters and information sharing matters. Some of the biggest challenges have been around this issue of communication and sharing information.
	Insecurities and tensions	Some researchers were like clumsy puppies trying to grasp us. Who are they? How do I deal with them? Also fears, fears of contact.
	Appreciation of different perspectives	I feel like I had respect for and an understanding of people's different perspectives. You need to choose people with diverse expertise but who are open to different perspectives, and who are willing to brainstorm about different the application of different perspectives, and what that suggests in terms of recommendations and outcomes.
	Feedback loops required	Constant feedback rounds were needed […] I always tried to give very hones feedback […] only positive feedback is often too little, especially in an area where so much has to happen when it comes to involving people who have experience with it.
Challenges	Heterogeneous backgrounds and skills	This interdisciplinarity is the work of the now and the future. These many perspectives that come in. They're a huge enrichment; they also make it more complex, of course. Because I go far beyond the level of content.
	Complexity	You can simply be courageous. The challenges and the problems that arise, and also to enter into the debate. And not to think that I have to sweep it under the carpet, that everything has to be perfect and so on. That was always my role, to be honest and open, and then others can get on board and learn something from you.
Impact	Community work	Community work is so much harder and takes so much more time and is so much more challenging. So, the metrics that you use for evaluating success of this initiative need to reflect the fact not only that it's the open innovation business, but also that it's so community-based. You may need to think carefully about things like number of people's lives you've touched, number of kids involved, number of kids who participated in making the project happen, number of families who have been touched in some way, number of other kinds of stakeholders/providers. You may want to think of your social media posts and the volume of likes or shares.

Overall, stakeholders felt that successful, transdisciplinary collaboration between them and researchers was dependent on the researchers' attitudes. Researchers needed to be open-minded toward new perspectives and approaches, flexible to adapt the research process along the way, and creative in dealing with the different perspectives, trainings, and skills. Additionally, open, honest, and regular communication about day-to-day challenges that researchers were facing fostered mutual learnings and helped competence group and advisory board members to give advice that was more helpful. **Table 1** summarizes the enablers and drivers for successful, transdisciplinary research approaches.

DISCUSSION

Working collaboratively and openly in a transdisciplinary research environment brings a range of challenges. In this study, we reported how stakeholders perceive transdisciplinary collaborations with researchers. Furthermore, we highlighted the enablers and barriers for such collaborations from the viewpoint of stakeholders.

Governance Structure

Overall, the advisory board and competence group perceived the general set up, such as the duration, the frequency, preparation material and the facilitation of the panel meetings, very positively. More importantly, they reported that the structure bringing together different expertise and perspectives caused challenges and resulted in more time-consuming decisions in the panel meetings (**Figures 1A,B**). These aspects well-reflect the considerations of practical support as enablers of PPI (45). The competence group especially emphasized these aspects probably due their (experiential) experience and their limited knowledge of the research process. Similar patterns can also be seen in sandpit approaches, where participants described that "the social dynamics are as interesting as the science" (50). The "language of collaboration" and building trust that makes it easier to challenge different perspective needs to be established before digging into content-related discussions (51).

Competence group and advisory board members rated the quality of involvement interacting with researchers high. This is in line with reports on high levels of consensus among stakeholders regarding the added value and impact of PPI in research (34, 52). However, the collaboration between the members and the researchers and implementation of recommendations was assessed mixed (**Figures 2A,B**). This might be due to the barriers of PPI (45), which could either result in an tokenistic attempt if the PPI principles are not met (30, 53), or in failure to involve the public meaningfully, which may result in an unsuccessful collaboration with the public due to negative attitudes held by researchers (39).

Enablers and Barriers on the Organizational Level

The interviews revealed several enablers for a successful, transdisciplinary research approach on two levels: the organizational (governance) and the individual level (summarized in **Table 1**). This ties to existing research on the principles for stakeholder engagement which can be organized in organizational factors, values and practices (54).

The advisory board emphasized the funders' commitment and boldness as an important factor to enable such a transdisciplinary approach. This is in line with other studies that mentioned financial and general support and resources, and the organizational commitment as key barriers of PPI in health research (41, 42, 45). In fact, LGB invested more than six million Euros in the entire bottom-up approach: from setting the research priority with the community to implementing the innovative research approaches for COPMI where the community defined the Research Topic and stayed an integral part in the research process along the implementation. This transdisciplinary research approach ensures that these areas can and are appropriately funded and staffed by talented individuals who want to dedicate their creative scientific talents to broader issues than their own field in the long term (55).

Furthermore, the interviewees indicated that organizational support structures, such as a person facilitating and supporting the community and stakeholder interactions, links the governance structures and acts as a key player in the process. Similarly, other studies reported the importance of support on an emotional, financial and practical level that is needed for involved people [e.g., see review (45)]: for example, support with the timing of activities, setting and constraints and commitment of public members, providing mentoring and a supportive chair to implement PPI practices. Researchers described the significant additional administrative labor and the lack of practical support for their work, as well as the time and effort diverted from these activities as barrier of PPI (19). Such a key person acts as a contact person for researchers and stakeholders and ensures that support is provided on an organizational, value-based and practice level. For example, the person fosters shared commitment to values and objectives of stakeholder engagement in the project team, recognizes potential tensions between productivity and inclusion, and considers how input from stakeholders can be collated, analyzed and used (54). In line with that, the competence and advisory group members emphasized the importance of such a key player in the process and, in fact, a relationship manager was established for the research groups Village and DOT. However, this person was placed at the LBG headquarters and not at the research groups' local site. Many difficulties arose due to this structure: for example, extensive travel time in setting up stakeholder and community relations at the beginning of the project, not being part of the research team and therefore ongoing negotiation of the roles and tasks as well as less involvement in discussions and decisions. These circumstances led to a change of the role over the years: from a relationship manager (active) to a sparring partner (passive) who discussed the progress of the research groups. One solution—as also indicated by our findings—could be to install a liaison between researchers and people with lived experience who facilitates and supports interactions between the two communities locally. In line with that, the LBG have recently begun to experiment with a new governance structure by embedding a local "stakeholder relationship manager". This manager facilitates the interaction between stakeholder groups and researchers. Another enabler for successful, transdisciplinary collaborations is to embed people with lived experience (in our case COPMIs) as co-researchers in the research team, which has also been suggested by the advisory board and competence group members. The latter even underlined that the involvement as co-researchers would devote the necessary time, commitment, and honorarium of contributions. Further it requires an understanding of the involvement process and to create a "real" position in the research team that had been described previously (19, 30). This addition to the governance structure would involve people with lived experience early right on from the beginning and in each phase of the research process. The advisory board hereby also suggested to involve everybody who needs to be involved as early as possible, in fact, already in the Ideas Lab to gain understanding and insights. These outlined modifications in research teams might ultimately overcome frictions in relationships between researchers and stakeholders and shift power dynamics (42, 45). Working as co-researchers guarantees mutual respect and equality between researchers and the public, and might rebalance the relationship and roles. Eventually, co-researchers might foster active involvement of stakeholders in health research (39).

Enablers and Barriers on the Individual Level

On the individual level, we also identified enablers and barriers for transdisciplinary collaboration between researchers and stakeholders. One major enabler for a successful, transdisciplinary research approach are the researchers' attitudes and values toward patient and public involvement (39, 43). Stakeholders mentioned as a crucial mindset that researchers need to bring to the table: open-mindedness, appreciation for stakeholders, eagerness to learn from other people's perspectives, interest to invest in relationships, continuous communication with stakeholders to address insecurities and tensions arising in the interaction with others, to provide feedback and actions based on the recommendations, respect for heterogeneous backgrounds and skills, and handling of complexity in an honest and open way. Previous studies explored health researchers' attitudes toward PPI and identified the transferring and sharing of power and the misconception of PPI—as participation in clinical trials and dissemination of information and knowledge—as major barriers for successful implementation (39, 40). The latter has been also reported in a recent study (56) that reflects on the limited PPI practices in Austria.

These enablers are also in line with the personal attitudes and values required for participating in the Ideas Lab (51). Based on researchers' attitudes and values captured in the application forms, only researchers describing a positive approach to team work, collaborative working and working with different disciplines and stakeholders were invited to participate in the Ideas Lab. However, these attitudes and values are often not lived and embodied in "real" collaborations with the community. Guimaraes et al. (44) explored the characteristics of inter- and transdisciplinary researchers. The authors found a mix of motivations, attitudes, skills, and behaviors, such as a humble attitude toward the immensity of knowledge, openness to different types of knowledge, tolerance to ideas opposed to one's own view, self-reflectiveness and curiosity, the ability to think in a complex and interlinked manner, and good communication and listening skills. However, these attitudes often do not link to the academic environment and its career paths, where short-term contracts and funding deadlines challenge researchers' ability to involve the public (39). Furthermore, responsibility among researchers is not distributed equally as often female researchers and early career researchers are tasked with stakeholder involvement. Ultimately, these circumstances cause tensions for those who (try to) acknowledge the value of PPI. Not surprisingly, researchers' attitudes toward PPI range from cynical to ambivalent to excited (19). Researchers further reported feelings of concerns when applying PPI practice, which may be due to a natural response to change. They also expressed concerns that PPI undermines professional skills and academic knowledge leading to a sense of de-professionalization (39). Furthermore, in this study, advisory board and competence group members reported indicated that researchers with a positive mindset and values toward PPI dealt with uncertainties and tensions better than researchers who embodied a more traditional scientific approach. To overcome this barrier, the competence group members suggested to organize social events and opportunities to meet outside the research context.

According to our results, it seems that flexibility and creativity are beneficial skills to deal with the challenges and the complexity that arise from transdisciplinary work, to change research approaches and to react to stakeholders' needs. This in turn requires to respect and appreciate heterogeneous backgrounds, different perspectives, professional trainings, and skills that all eventually enrich the discussions and collaborations (19, 44, 45). It therefore is important to carefully reflect on the who and why of involving people with lived experience so that ineffectiveness, tensions, and tokenistic involvement of stakeholders can be avoided (38).

The advisory board also emphasized different dissemination strategies to better highlight the impact that research has on the community, and alternative ways to measure scientific impact (32). Equally important is continuous communication and feedback loops about the implementation of recommendations; a crucial point that has also already been addressed in public involvement guidelines for researchers [e.g., see (57)].

Based on our results, it becomes evident that successful, transdisciplinary collaboration demands specific personality characteristics (44), organizational and financial support structures (45) and highly depends on the peoples' attitudes and values toward PPI (37, 39, 41, 43, 52). Understanding the situational context and the people and the community in which the collaboration takes place (36–38), is crucial; especially for solving complex challenges where multiple stakeholders are involved, such as designing interventions for COPMIs and their families (14–18). Our findings therefore contribute to implementation strategies, in which COPMIs have a key role in recruiting and training researchers with a positive attitude toward PPI and transdisciplinary collaboration, and in identifying tensions in the transdisciplinary collaborations.

Strengths and Limitations of This Study

A strength of this study is that it analyzes for the first time how stakeholders perceive transdisciplinary collaboration; specifically, what enablers and drivers for such collaborations stakeholders can identify. In doing so, our study adds further evidence to previous studies that highlighted how researchers themselves can influence the success of transdisciplinary collaboration. Additionally, and also in line with previous studies, our findings underline the importance of a "neutral" contact person who facilitate the collaboration process between stakeholders and researchers, who addresses uncertainties and tensions, and who mediates among the people involved.

On a methodological level, a limitation of this study concerns the small sample size of the survey. While the competence groups and advisory boards comprised 18 people in total, 11 members responded to the survey. Therefore, we analyzed the data descriptively. To counteract any possible biases, the semi-structured interviews were conducted by a researcher working at LBG, who did not have previous contact or worked with the research groups or advisory board members before. However, it cannot be ruled out that some biased still emerged. Another limitation of this study is that we did not incorporate the researchers' perspective. After careful consideration, we decided not to invite researchers to participate in the survey and the interviews because of the upcoming evaluation of the research groups at the end of 2021 and the already existing literature on researchers' attitudes and vales toward PPI (39, 44, 45, 52). We rather wanted to focus more strongly on the stakeholders' views on transdisciplinary research collaboration.

CONCLUSION

The new governance structures comprising transdisciplinary expertise and children of parents with mental illnesses was highly appreciated among the advisory board and competence group members and added value to the discussions about real life-problems and novel research approaches for COPMI. The transdisciplinary collaboration demanded a thorough understanding of people's perspectives, investment in relationships, and continuous feedback and communication with stakeholders. Furthermore, advisory board and competence group members suggested to continuously invite people with lived experience (in this case, COPMIs) as co-researchers. Open-mindedness toward different perspectives and approaches, flexibility to adapt to the research process along the way, and creativity dealing with other backgrounds and skills were identified as the most important enablers for a successful, transdisciplinary research approach. Consequently, we can conclude that peoples' attitudes and values as well as support structures are key enablers for transdisciplinary research approaches. In our experience, researchers who acknowledge the benefit of PPI practices and have already gained positive experiences working with people with lived experience (COPMI) and stakeholders are more likely to value transdisciplinary collaborations.

Future studies should aim to develop a deeper understanding of attitudes and values work as barriers for transdisciplinary collaborations between researchers and stakeholders. Specifically, future studies should focus on openness as a key enabler for transdisciplinary collaborations and might therefore answer a question that this study has unveiled. To what extent and how is it possible to create awareness and an open mindset among researchers—for instance, via capacity building and trainings—so that transdisciplinary research approaches can successfully be implemented in the future?

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

RK organized the Ideas Lab in 2017 and facilitated the implementation of the LBG Research Group DOT and Village as relationship manager (2018–2021). RK and CG conceived and planned the online survey for the competence group and advisory board members and contributed to the interpretation of survey findings and interviews, conceptualization of this article, and wrote the first manuscript draft. CG designed the online survey in Unipark, which was then administered by RK. Survey results were analyzed by CG. CG also conducted the semi-structured interviews of competence group and advisory board members. All authors critically reviewed the manuscript for intellectual content, approved the final version, and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

FUNDING

The open innovation initiative and following projects are funded by the LBG Open Innovation in Science Center at the Ludwig Boltzmann Gesellschaft, Austria.

ACKNOWLEDGMENTS

We are grateful to the LBG Open Innovation in Science Center at the Ludwig Boltzmann Gesellschaft implementing an open innovation approach and enabling transdisciplinary collaboration in the field of mental health by establishing Research Groups on Children of mentally ill parents. We thank all competence group and advisory board members participating in the online survey and semi-structured interviews. We especially thank Patrick Lehner for providing valuable feedback to the first manuscript draft.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.760716/full#supplementary-material

REFERENCES

1. Maybery D, Reupert A, Goodyear M, Ritchie R, Brann P. Investigating the strengths and difficulties of children from families with a parental mental illness. *Austral E J Adv Mental Health.* (2009) 8:165–74. doi: 10.5172/jamh.8.2.165
2. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry.* (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
3. Ostman M, Hansson L. Children in families with a severely mentally ill member. Prevalence and needs for support. *Soc Psychiatry Psychiatr Epidemiol.* (2002) 37:243–8. doi: 10.1007/s00127-002-0540-0
4. Pretis M, Dimova A. Vulnerable children of mentally ill parents: towards evidence-based support for improving resilience. *Support Learn.* (2008) 23:152–9. doi: 10.1111/j.1467-9604.2008.00386.x
5. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Austral E J Adv Mental Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
6. Power J, Goodyear M, Maybery D, Reupert A, O'Hanlon B, Cuff R, et al. Family resilience in families where a parent has a mental illness. *J Soc Work.* (2016) 16:66–82. doi: 10.1177/1468017314568081
7. Reupert A, Maybery D. Families affected by parental mental illness: a multiperspective account of issues and interventions. *Am J Orthopsychiatry.* (2007) 77:362–9. doi: 10.1037/0002-9432.77.3.362
8. Reupert AE, Cuff R, Drost L, Foster K, van Doesum KT, van Santvoort F. Intervention programs for children whose parents have a mental illness: a review. *Med J Aust.* (2013) 199(3 Suppl.):S18–22. doi: 10.5694/mja11.11145
9. Goodyear M, Hill TL, Allchin B, McCormick F, Hine R, Cuff R, et al. Standards of practice for the adult mental health workforce: meeting the needs of families

where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120

10. Lauritzen C, Reedtz C, Van Doesum KT, Martinussen M. Implementing new routines in adult mental health care to identify and support children of mentally ill parents. *BMC Health Serv Res.* (2014) 14:58. doi: 10.1186/1472-6963-14-58
11. Maybery D, Jones R, Dipnall JF, Berger E, Campbell T, McFarlane A, et al. A mixed-methods study of psychological distress following an environmental catastrophe: the case of the Hazelwood open-cut coalmine fire in Australia. *Anxiety Stress Coping.* (2020) 33:216–30. doi: 10.1080/10615806.2019.1695523
12. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Family options for parents with mental illnesses: a developmental, mixed methods pilot study. *Psychiatr Rehabil J.* (2009) 33:106–14. doi: 10.2975/33.2.2009.106.114
13. Solantaus T, Toikka S. The effective family programme: preventative services for the children of mentally ill parents in Finland. *Int J Mental Health Promot.* (2006) 8:37–44. doi: 10.1080/14623730.2006.9721744
14. Chambers DA, Glasgow RE, Stange KC. The dynamic sustainability framework: addressing the paradox of sustainment amid ongoing change. *Implement Sci.* (2013) 8:117. doi: 10.1186/1748-5908-8-117
15. Greenhalgh T, Wherton J, Papoutsi C, Lynch J, Hughes G, A'Court C, et al. Beyond adoption: a new framework for theorizing and evaluating nonadoption, abandonment, and challenges to the scale-up, spread, and sustainability of health and care technologies. *J Med Internet Res.* (2017) 19:e367. doi: 10.2196/jmir.8775
16. Watson DP, Adams EL, Shue S, Coates H, McGuire A, Chesher J, et al. Defining the external implementation context: an integrative systematic literature review. *BMC Health Serv Res.* (2018) 18:209. doi: 10.1186/s12913-018-3046-5
17. Goodyear M, Obradovic A, Allchin B, Cuff R, McCormick F, Cosgriff C. Building capacity for cross-sectorial approaches to the care of families where a parent has a mental illness. *Adv Mental Health.* (2015) 13:153–64. doi: 10.1080/18387357.2015.1063972
18. Zechmeister-Koss I, Goodyear M, Tuchler H, Paul JL. Supporting children who have a parent with a mental illness in Tyrol: a situational analysis for informing co-development and implementation of practice changes. *BMC Health Serv Res.* (2020) 20:326. doi: 10.1186/s12913-020-05184-8
19. Boylan AM, Locock L, Thomson R, Staniszewska S. "About sixty per cent I want to do it": health researchers' attitudes to, and experiences of, patient and public involvement (PPI)-A qualitative interview study. *Health Expect.* (2019) 22:721–30. doi: 10.1111/hex.12883
20. Klein JT. Evaluation of interdisciplinary and transdisciplinary research: a literature review. *Am J Prev Med.* (2008) 35(Suppl. 2):S116–23. doi: 10.1016/j.amepre.2008.05.010
21. United Nations. *Transforming Our World: The 2030 Agenda for Sustainable Development* (2015). Available online at: https://sustainabledevelopment.un.org/content/documents/21252030%20Agenda%20for%20Sustainable%20Development%20web.pdf (accessed July 26, 2021)
22. National Institute of Health Research (NIHR). *Involve patients*. Available online at: https://www.nihr.ac.uk/health-and-care-professionals/engagement-and-participation-in-research/involve-patients.htm (accessed July 26, 2021).
23. Carman KL, Dardess P, Maurer M, Sofaer S, Adams K, Bechtel C, et al. Patient and family engagement: a framework for understanding the elements and developing interventions and policies. *Health Aff.* (2013) 32:223–31. doi: 10.1377/hlthaff.2012.1133
24. Church J, Saunders D, Wanke M, Pong R, Spooner C, Dorgan M. Citizen participation in health decision-making: past experience and future prospects. *J Public Health Policy.* (2002) 23:12–32. doi: 10.2307/3343116
25. Coultier A. *Engaging Patients in Healthcare*. New York, NY: McGraw Hill Professional (2013).
26. Hayes H, Buckland S, Tarpey M. *Briefing Notes for Researchers: Public Involvement in NHS, Public Health and Social Care Research*. Eastleigh: INVOLVE (2012).
27. Staley K. *The BMJ [Internet]*. BMJ (2018). Available online at: https://blogs.bmj.com/bmj/2018/11/28/researchers-dont-know-what-theyre-missing-the-impact-of-patient-involvement-in-research/ (accessed July 26, 2021).
28. Piil K, Jarden M. Patient involvement in research priorities (PIRE): a study protocol. *BMJ Open.* (2016) 6:e010615. doi: 10.1136/bmjopen-2015-010615
29. Knowles SE, Allen D, Donnelly A, Flynn J, Gallacher K, Lewis A, et al. More than a method: trusting relationships, productive tensions, and two-way learning as mechanisms of authentic co-production. *Res Involv Engagem.* (2021) 7:34. doi: 10.1186/s40900-021-00262-5
30. Kaisler RE, Missbach B. Co-creating a patient and public involvement and engagement 'how to' guide for researchers. *Res Involv Engagem.* (2020) 6:32. doi: 10.1186/s40900-020-00208-3
31. Chudyk AM, Waldman C, Horrill T, Demczuk L, Shimmin C, Stoddard R, et al. Models and frameworks of patient engagement in health services research: a scoping review protocol. *Res Involv Engagem.* (2018) 4:28. doi: 10.1186/s40900-018-0111-5
32. Staley K. *Exploring Impact: Public Involvement in NHS, Public Health and Social Care Research*. Eastleigh: INVOLVE (2009).
33. Shippee ND, Domecq Garces JP, Prutsky Lopez GJ, Wang Z, Elraiyah TA, Nabhan M, et al. Patient and service user engagement in research: a systematic review and synthesized framework. *Health Expect.* (2015) 18:1151–66. doi: 10.1111/hex.12090
34. Brett J, Staniszewska S, Mockford C, Herron-Marx S, Hughes J, Tysall C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. *Health Expect.* (2014) 17:637–50. doi: 10.1111/j.1369-7625.2012.00795.x
35. Domecq JP, Prutsky G, Elraiyah T, Wang Z, Nabhan M, Shippee N, et al. Patient engagement in research: a systematic review. *BMC Health Serv Res.* (2014) 14:89. doi: 10.1186/1472-6963-14-89
36. Brett J, Staniszewska S, Mockford C, Herron-Marx S, Hughes J, Tysall C, et al. A systematic review of the impact of patient and public involvement on service users, researchers and communities. *Patient.* (2014) 7:387–95. doi: 10.1007/s40271-014-0065-0
37. Staley K. "Is it worth doing?" Measuring the impact of patient and public involvement in research. *Res Involv Engagem.* (2015) 1:6. doi: 10.1186/s40900-015-0008-5
38. Staley K, Elliott J, Stewart D, Wilson R. Who should I involve in my research and why? Patients, carers or the public? *Res Involv Engag.* (2021) 7:41. doi: 10.1186/s40900-021-00282-1
39. Thompson J, Barber R, Ward PR, Boote JD, Cooper CL, Armitage CJ, et al. Health researchers' attitudes towards public involvement in health research. *Health Expect.* (2009) 12:209–20. doi: 10.1111/j.1369-7625.2009.00532.x
40. Vale CL, Thompson LC, Murphy C, Forcat S, Hanley B. Involvement of consumers in studies run by the Medical Research Council Clinical Trials Unit: results of a survey. *Trials.* (2012) 13:9. doi: 10.1186/1745-6215-13-9
41. McKenzie A, Bulsara C, Haines H, Hanley B, Alpers K. *Barriers to Community Involvement in Health and Medical Research - Researchers Perspectives on Consumer and Community Involvement in Research: A Qualitative Study*. Nedlands, WA: The University of Western Australia School of Population Health; Telethon Kids; Institute and The University of Notre Dame (2016).
42. Wilson P, Mathie E, Keenan J, McNeilly E, Goodman C, Howe A, et al. Health Services and Delivery Research. In: *Research With Patient and Public Involvement: a RealisT Evaluation – The RAPPORT Study*. Southampton: NIHR Journals Library (2015).
43. Nathan S, Harris E, Kemp L, Harris-Roxas B. Health service staff attitudes to community representatives on committees. *J Health Organ Manag.* (2006) 20:551–9. doi: 10.1108/14777260610702299
44. Guimarães MH, Pohl C, Bina O, Varanda M. Who is doing inter- and transdisciplinary research, and why? An empirical study of motivations, attitudes, skills, and behaviours. *Futures.* (2019) 112:102441. doi: 10.1016/j.futures.2019.102441
45. Ocloo J, Garfield S, Franklin BD, Dawson S. Exploring the theory, barriers and enablers for patient and public involvement across health, social care and patient safety: a systematic review of reviews. *Health Res Policy Syst.* (2021) 19:8. doi: 10.1186/s12961-020-00644-3
46. Ludwig Boltzmann Gesellschaft. *Tell Us! About Mental Health: Ludwig Boltzmann Gesellschaft*. Available online at: https://www.redensiemit.org (accessed July 26, 2021).
47. Ludwig Boltzmann Gesellschaft. *Ideas Lab on, Children of Mentally Ill Parents: Ludwig Boltzmann Gesellschaft*. Available online at: http://www.ideaslab.lbg.ac.at (accessed July 26, 2021).

48. Ludwig Boltzmann Gesellschaft. *DOT - The Open Door: Ludwig Boltzmann Gesellschaft*. Available online at: https://dot.lbg.ac.at/ (accessed July 26, 2021).
49. Ludwig Boltzmann Gesellschaft. *Village - How to Raise a Village to Raise a Child: Ludwig Boltzmann Gesellschaft*. Available online at: http://village.lbg.ac.at/ (accessed July 26, 2021).
50. Giles J. Sandpit initiative digs deep to bring disciplines together. *Nature*. (2004) 427:187. doi: 10.1038/427187a
51. Maxwell K, Benneworth P. The construction of new scientific norms for solving Grand Challenges. *Palgrave Commun.* (2018) 4:52. doi: 10.1057/s41599-018-0105-9
52. Snape D, Kirkham J, Britten N, Froggatt K, Gradinger F, Lobban F, et al. Exploring perceived barriers, drivers, impacts and the need for evaluation of public involvement in health and social care research: a modified Delphi study. *BMJ Open.* (2014) 4:e004943. doi: 10.1136/bmjopen-2014-004943
53. INVOLVE. *Involving Children and Young People in Research: Top Tips and Essential Key Issues for Researchers*. Eastleigh: INVOLVE (2016).
54. Boaz A, Hanney S, Borst R, O'Shea A, Kok M. How to engage stakeholders in research: design principles to support improvement. *Health Res Policy Syst.* (2018) 16:60. doi: 10.1186/s12961-018-0337-6
55. Holm P, Goodsite ME, Cloetingh S, Agnoletti M, Moldan B, Lang DJ, et al. Collaboration between the natural, social and human sciences in Global Change Research. *Environ Sci Policy.* (2013) 28:25–35. doi: 10.1016/j.envsci.2012.11.010
56. Kaisler RE, Kulnik ST, Klager E, Kletecka-Pulker M, Schaden E, Stainer-Hochgatterer A. Introducing patient and public involvement practices to healthcare research in Austria: strategies to promote change at multiple levels. *BMJ Open.* (2021) 11:e045618. doi: 10.1136/bmjopen-2020-045618
57. Kaisler RE, Missbach B. *Patient and Public Involvement and Engagement in Research - A "how to" Guide for Researchers.* Zenodo (2019). doi: 10.5281/zenodo.3578321

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 30 November 2021
doi: 10.3389/fpsyt.2021.783066

A Narrative Evaluation of a Grief Support Camp for Families Affected by a Parent's Suicide

*Anneli Silvén Hagström**

Department of Social Work, Stockholm University, Stockholm, Sweden

Background: Children of parents who suffer mental ill-health and die by suicide are vulnerable to developing psychological and social problems themselves; they also have a severely elevated risk of dying at a young age – particularly through suicide. This highlights the need to design supportive measures that can counteract such negative developments after a parent's suicide.

Aim: This narrative evaluation of a grief support camp for families affected by a parent's suicide arranged by the non-profit organization Children's Rights in Society in Sweden investigates *whether* children [$N = 11$] and parents [$N = 11$] perceived their participation as meaningful and, if so, *in what way*, and the *changes* to which the program was said to have contributed.

Methods: Family members were invited to reflect on their experiences in narratively structured interviews that took place 18 months after participation. Their narrated experiences were analyzed to examine how the program was integrated into their biographies and with what significance. Narratives of change were identified in particular in order to grasp the self-perceived effects of participation.

Results: Both children and parents attributed major significance to their encounters with other suicide bereaved. This led to support exchange and normalization, which countered a perceived "suicide stigma" in everyday life. Help to narratively construct destigmatizing understandings of suicide was also said to have relieved self-blame and shame. Overall, the participants described changes in the form of a better-informed position in grief, increased manageability and enhanced family communication. The parents also reported improved ability to support their children and a more hopeful view of life ahead.

Conclusion: The evaluation showcases how this psychoeducational intervention, at a relatively low cost compared to traditional approaches, has great potential to lessen the negative effects of a suicide in the family by assisting families with psychological processing and de-stigmatization. Parental resources are also strengthened, which can serve as continuing support for the children.

Keywords: bereavement, children, family intervention, grief support, mental health, narrative evaluation, stigmatization, suicide

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
Erin Turbitt,
University of Technology
Sydney, Australia
Jose Luis Ramirez-GarciaLuna,
McGill University Health
Centre, Canada

****Correspondence:***
Anneli Silvén Hagström
anneli.silven.hagstrom@socarb.su.se

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *25 September 2021*
Accepted: *04 November 2021*
Published: *30 November 2021*

Citation:
Silvén Hagström A (2021) A Narrative
Evaluation of a Grief Support Camp
for Families Affected by a Parent's
Suicide. Front. Psychiatry 12:783066.
doi: 10.3389/fpsyt.2021.783066

RATIONALE

The Swedish Health and Medical Services Act (SFS 2017:30, chap 5: 7§) emphasizes the responsibility of health and medical care to provide information, advice and support to a child if her or his parent, or another adult with whom the child lives permanently, suffers from a mental illness or disability, and/or unexpectedly dies (1). However, children who have lost a parent through suicide are a neglected group in Swedish society. In addition, the United Nations Convention on the Rights of the Child has been binding Swedish law since January 2020. This further accentuates the right of parentally suicide-bereaved children to access to age-appropriate information and support. However, neither the national guidelines nor legislation stipulate *the kind of professional support* that should be provided to children who suffer the loss of a parent through suicide. Moreover, although stipulated as mandatory, in practice professional support is only offered exceptionally to such children and their families. One consequence of this failure to routinely offer support is that the remaining parent must be attentive to the child's processing of loss and active in the search for professional support where deemed necessary. This can be a difficult task, however, as parents must manage the effects of their own grief and mourning children may hide their grief to avoid worrying or burdening the grieving parent. In addition, children's access to professional assistance with grief is fully dependent on the local availability of professional bereavement counseling and peer-support groups. These circumstances mean that a considerable number of children must cope without professional grief support after a parent's suicide, due to the remaining parent's lack of initiative or know-how regarding *whether* and *where* to seek professional support, and/or a general shortage of professional grief interventions—especially those which specialize in suicide-bereavement. At the same time, previous studies in the field have established that children who have lost a parent through suicide constitute a risk group for developing complicated grief due to the difficulties of resolving the loss. This has been linked to psychiatric morbidity (e.g., anxiety, depression, PTSD, suicidal ideation) as well as social problems (2). As a result, suicide-bereaved children have a severely elevated risk of dying at a young age—particularly through suicide (3, 4). There is thus a critical need to provide these children with appropriate post-vention measures that can cater for their specific needs in grief and prevent such an accumulation of adverse effects after a parent's suicide.

Since 2013, Children's Rights in Society (Barnens rätt i samhället, BRIS) has organized a grief support program in the format of a weekend camp—known as support weekends—for families affected by a parent's suicide. The primary objectives of this psychoeducational program are to help children and their parents to: (a) identify how life has changed since the suicide loss, and their own responses and needs in grief; (b) develop health-promoting coping strategies; and (c) facilitate open and supportive family communication. A further main aim is to assist parents through dialogue to develop their skills to understand and support their children in grief. This is an exception to the otherwise absence of grief support programs directed at suicide-bereaved children and their families in Sweden.

Knowledge of the perceived meaningfulness and takeaways from similar grief support programs is scant and, to date, few studies have analyzed suicide-bereaved children's and parents' responses to their participation in such programs. This might be explained by the fact that grief support programs that specialize in suicide bereavement are still rare in many countries, and that existing programs have been evaluated first and foremost using quantitative methods, such as questionnaires, that provide pre-printed response options. The few existing studies have importantly concluded that family-based interventions for suicide-bereaved children can lessen suicide-related distress and promote children's emotional and social functioning in grief (5, 6). However, less is known about how these effects are achieved and how the program content has been integrated into suicide bereaved families' grief processes and lives. Hence, children and their parents have only to a limited extent been encouraged to talk freely about their experiences, and to consider how the program was located within their ongoing biographies and with what significance. This is the distinct purpose of this article, in which children's and parents' narrated experiences of their participation in the BRIS grief support program for families affected by a parent's suicide are analyzed to investigate *whether* they perceived the program to be meaningful and, if so, *in what ways*. Of particular interest is to identify so-called *narratives of change* in order to capture how the grief support program is said to have contributed to actual changes in the participants' grieving processes and lives.

PARENTAL SUICIDE-BEREAVED CHILDREN'S GRIEF EXPERIENCES AND NEEDS

Although many children experience the fatal loss of a loved one in childhood, their grief tends to be overlooked by adults who commonly avoid talking with children about death and the deceased, which contributes to a powerlessness in young people's dealing with loss (7). This is particularly evident in cases of unnatural deaths, such as through suicide (8). Following a parent's suicide, in addition to the child's age and maturity/ability to conceptualize death, the supportive role of the remaining parent and an open communication climate in the family have been identified as vital to the ability of children to cope with the loss (9). However, distorted communication commonly occurs due to the remaining parent's efforts to protect the child from the circumstances of the suicide (10). Such concealment may, contrary to its aim, complicate the child's processing of loss and leave the child in a confused and lonely position in grief.

Parentally suicide-bereaved children are also faced with a "double whammy"; in addition to coping with the loss itself, children are left to try to make sense of their parent's suicide [(8), p. 192]. The question, "*Why* did my mother/father *choose* to die?," is central to young mourners' meaning reconstruction after a parent's suicide (11, 12). The search for answers together with the lack of information from adults commonly produce self-blame and shame, as parentally suicide-bereaved children[1] for various reasons tend to take the blame for the suicide on themselves. They may also hold the deceased parent accountable, based on the belief that the parent failed in his or her moral responsibility to care for them, and for selfish reasons chose to leave the child. Both understandings produce a stigmatized identity influenced by anger, shame and blame, either as a "failed" daughter/son or as the offspring of a deeply "immoral person" (12). At the heart of suicide-bereaved children's stigmatization is the sense of having been unloved and/or abandoned by the deceased parent, which ultimately raises questions about the child's self-worth [ibid.; (8)]. This culturally induced "suicide stigma" can be reinforced by non-supportive responses within the children's social network, such as straightforward questions from other children, "is there crazy in your family?" [(8), p. 192], or avoidance and outright rejection (12). Stigmatization has been shown to play a central role in suicide bereavement (13) and in research parentally suicide-bereaved children describe themselves as feeling deviant—and even strange or tainted—by their parent's suicide (14, 15).

Where a parental suicide-bereaved child's and the remaining parent's need for support in grief is substantial and the social support is inadequate, access to professional interventions becomes critical. However, children and families are seldom offered professional support in connection with a family member's suicide (16). Young mourners may also be dissatisfied with the professional support they receive, due to a perceived lack of empathy and knowledge about grief after suicide among professionals (17).

EVALUATION STUDIES IN THE FIELD

A systematic review of the effects of grief support programs for parentally bereaved children shows that when the remaining parent is supported, there is an improvement in parental health in grief and an enhanced capacity to care for the child, leading to positive effects on children's grieving (18). Another advantage of a family-based approach is the opportunity for children and parents to sit down and talk about parental loss together—sometimes for the first time. Grief interventions for a parentally bereaved child together with the remaining parent are therefore generally recommended.

A study examining children's experiences of participation in grief support programs shows that a combination of camp-specific activities and therapeutic conversations/exercises contributed to an improvement in the children's perceived well-being (19). Participation in activities is said to contribute to community, belonging and strengthened self-esteem, and to offer a break from painful emotions. The therapeutic conversations and exercises, in turn, are said to contribute to healing in grief, to understanding and putting words to experiences, to assisting with memory and to forming a continuing bond with the deceased. A review of the effectiveness of bereavement camps for children (20) confirms that this combination of a safe space to express grief in a therapeutic environment in the company of other bereaved children and playful activities is a promising venue to help bereaved children to build resilience.

Nonetheless, the effectiveness of general grief interventions with suicide-bereaved children has been questioned due to weak results (21). It has even been argued by support-group practitioners that suicide-bereaved children may experience reinforced stigmatization in these blended contexts, where they maintain silence about the circumstances of the death (8). Instead, specialized programs led by trained facilitators that take aspects such as the social environment into account yield more promising results (22, 23). An evaluation of a grief support program aimed at suicide-bereaved children and their parents (6), focused on children's reactions to death and suicide, and on strengthening their coping skills, demonstrated a significant reduction in anxiety and depressive symptoms in children. The evaluation of another family-based program focused on children's emotional needs (5) showed a similar reduction in anxiety and depressive symptoms, as well of disruptive behavior in bereaved children. The program also increased knowledge, self-esteem and agency, and led to more successful coping. Although research on suicide-specific support programs for both parentally suicide-bereaved children and their remaining parents is still scant, there are weak but promising indications that family-based programs can help to improve children's coping with parental loss and reduce suicide-specific symptoms of grief connected to complicated grief. There is, however, an urgent need to examine how suicide-bereaved families themselves experience such programs and what they find helpful.

MATERIALS AND METHODS

A Narrative Approach to Program Evaluation

Quantitative methods dominate the evaluation field and a narrative approach to program evaluation is much rarer. Narrative inquiry investigates how people make sense of events, the world they live in and their related identities. Hence, the stories people tell reveal subjective truths about their lives and identities, and offer context-specific knowledge that might not always be discovered using other methods. A main focus of narrative program evaluation is *change*. Baú (24) encourages the researcher to ask people to recognize change when recounting their biography from past to present, including the professional intervention, as this makes it possible to understand how people integrate the program content and how it is applied in their continued living. A narrative approach entails the notion that evaluation is not the endpoint of applied knowledge but a contributor of new culture-specific knowledge. Such knowledge production also comes in the form of a narrative. According to Constantino and Greene [(25), p. 47]: "By telling the program's story, an evaluation may be used to give voice to participants' perspectives, as they and their experiences provide the characters and events of the program's narrative." Like an evaluation story, this article constructs an overall meaning of the participants'

---

[1] In this article, "children" refers not only to biological age, but also to young people's position in relation to the deceased parent. The child participants in the study were both younger children and teenagers.

experiences and takeaways from the program, with the aim of examining the difference made by the program from a wider social and cultural perspective.

Theoretical Basis, Structure and Thematic of the Grief Support Program

The BRIS grief support program for families affected by a parent's suicide comprises two support weekends, Friday to Sunday, 4 months apart. On each occasion 10–12 families participate. This nationwide program is subsidized by the public health authority and located in the middle of Sweden, enabling families from different socio-economic backgrounds and localities to participate. Program information is published on the BRIS website and on social media, and is also distributed to suicide bereavement and mental health organizations. Although the program has a family-based approach, its main objective is to facilitate children's grief. The program is based on a systems theory perspective and the notion that family members' grief is interrelated [see (26, 27)]. Families' post-loss communication and interaction are thus understood as essential to suicide-bereaved children's abilities to cope with grief. The salutogenic perspective 'Sense of coherence' (28) also has a central role in the program. It is based on the notion that honest and age-appropriate information, space and support for expression and reflection, as well as help to develop resilient coping strategies can strengthen children's comprehension and the manageability of parental loss, and contribute to increased meaningfulness in life. The program also draws on theories about childhood grief from an attachment and development perspective (29).

The program is structured around parallel group meetings, where parents and children in parallel but separately process the same themes adapted to age. The composition of the children's groups is based on the current participants and divided according to age. The children in the youngest age group are 4–6 years old and the oldest children are 20 years old. Each group consists of 4–8 children. All the parents are in one group. The main themes processed in the groups are: "Information about suicide and suicide bereavement"; "The family then and now: what happened?"; "What has changed?"; "Grief responses and emotions"; "My grief/others' grief"; "Remembering the deceased parent"; "What helps and how do I take care of myself?"; "Questions I wanted to ask but have not dared"; and "What is my future?" (26). These sessions are combined with grief-oriented family exercises and playful activities, where the latter offer opportunities for relaxation and togetherness within families and between participants (for a fuller description of the program content, see **Supplementary Material**). The psychoeducational components of the program focus on helping the children to express their thoughts and feelings about their parental loss. Participants also learn about why people die by suicide, common grief responses and needs, and strategies for coping with grief, viewed over time [i.e., the oscillation between loss-oriented and restoration-oriented coping, (27)]. In addition, the children are supported to construct a narrative about their deceased parent and identify positive memories and parental attributes with which the child may identify. Psychoeducational components support the parents to understand childhood bereavement, foster their children's expression and emotional and social functioning in grief, and open up space for family conversations about the deceased parent and grief. The parents also ventilate their own grief, but with primary attention on their parenting role.

Study Design and Procedure

The author is a social worker, grief therapist and researcher who specializes in young people's grief after a parent's suicide. She has long clinical experience of talking with children and teenagers about sensitive issues related to family problems and loss, which was gained in child and adolescent psychiatric care. She was asked to conduct an evaluation of the current program without having had any pre-existing relationship with the organizer or any of the personnel. The study was carried out in three steps. First, the author conducted participant observations at the grief support camp on two separate occasions to understand the context and program content, and to observe the knowledge and support exchange through exercises and activities, as well as the social interaction between the professionals (social workers specialized in children's grief) and participating family members—and between the participants themselves. An additional aim was to make contact with the participants, primarily the children and teenagers, in order to build trust, which should have a positive effect on participation in the interviews. Second, all the participants were informed about the study orally by the author and in writing on the first program day, and later invited to take part in the study in an age-appropriate and personally addressed letter followed-up by a telephone contact with the parent of each family. Third, all those who agreed to contribute were interviewed 18 months after the program ended. Two children decided to participate only after meeting the author in connection with interviews with other family members.

Study Participants and Material

All the members of the 14 families that attended the BRIS grief support camp on two different occasions between 2017 and 2018 were invited to participate in the study [$N = 49$]. Of these, 11 children (six girls and five boys), aged between six and 13 with a mean age of nine at the time of their participation, and 11 parents (nine mothers and two fathers) [$N = 22$] from eight families agreed to be interviewed for the study. The time elapsed from the parental loss to program participation varied between 6 months and 5 years, with an average of about 1.5 years. There were variations in urban and rural locations, and socio-economic and cultural backgrounds but ethnic Swedish, middle class families were predominant. The interviews were conducted in-person in the families' homes, and with children and parents separately. A general feature of the interviews, which were adapted according to the children's age and maturity, was that the participants were asked to talk about what life was like before and after participation, and how the program content was perceived and thought to have contributed to grieving and life in general. Special attention was therefore paid to descriptions of daily life, grief reactions, coping strategies and support needs. The material differed in narrative richness and the younger children in particular needed to be more actively supported and reminded of various activities before they could engage in storytelling. The interviews were recorded and transcribed verbatim for analysis. The participant observations mainly contributed to the author being better informed during the interviews but were also used to contextualize the study results.

Analysis of the Participants' Narrated Experiences

The analysis was guided by a narrative methodology for evaluation to investigate *whether* the participating parents and children perceived the program to be meaningful and, if so, *in what ways*. Of particular interest was to identify so-called *narratives of change* in order to capture how the grief support program is said to have contributed to actual changes in the participants' grieving processes and lives. The participants' narrated experiences constructed in research interviews were analyzed using narrative methodology (30), and the concept of "narratives of change" (24). First, the transcriptions were read repeatedly to identify the narrative thematic of the meanings attributed to participation in the BRIS grief support camp. Narratives of change were then delineated and thematically analyzed to grasp the perceived impact of the program on the participants' ongoing grieving processes and lives. The children's and parents' narratives were first analyzed separately and later compared to construct a more complex understanding of each family situation, and find connections and differences in the material. The results have been discussed and validated against the interview material at a research seminar with narrative researchers.

Ethical Considerations

Interviewing children about potentially traumatic and stigmatizing experiences such as the death of a parent through suicide is an ethically sensitive issue. It can stir up unresolved issues and emotions linked to the loss and actualize a need for professional support. The interview situation itself, between an adult interviewer and a child, is also unequal and constitutes an imbalance of power that can incline children to adapt to what they believe is expected of them and ignore their own needs (31). Based on this, the research interviews were conducted with great sensitivity and respect for each child's integrity and personal needs; for example, two sisters chose to be interviewed together and many children chose to make drawings during the interview. The narrative approach facilitated the children to decide for themselves what they wanted to disclose. They were instructed to tell only what they wanted to tell, no matter how much or how little, and to just say "I do not want to talk about it" if they did not want to answer a question by the author. The children usually recognized the author and in conversations before the interview the author discussed memories of the camp to establish contact. Another facilitator for the children to express themselves was that they had all participated in a support group activity and thus to some extent acquired a language for talking about their suicide loss experience. At the end of each interview, the children were asked how they felt after having talked about their loss and grief experiences. Although the interviews brought up painful thoughts and emotions, all the children seemed positive about the interview experience. The children who expressed a continuing need for professional support already had ongoing contacts through school or health care. The research interviews for the study were conducted in accordance with the ethical guidelines for research in the human sciences and with the permission of the Regional Ethical Review Board in Uppsala, Sweden (Id. 2015/504).

RESULTS

The results are structured chronologically from narratives about life before to life after the support program, with meanings and changes highlighted. All the participants have been given fictitious names and any personal details that could reveal identity have been removed or altered in order to maintain confidentiality.

Life Before the Grief Support Program

In the interviews, both children and parents were asked to recall life before their participation in the grief support program. Several children stated in a few words that they did not know anyone else who had lost a parent through suicide, and that they had avoided talking about their parent's suicide outside the family. Sometimes they said they did not feel the need to talk about it within the family either. Most children described how they had tried to live as before with a main strategy being to keep quiet about thoughts and emotions associated with parental loss. The children's more limited narratives were contextualized by the parents' descriptions. Most children were living with both parents at the time of the suicide, although a few parents had separated. In the latter cases, the child either shared accommodation, living every other week with each parent, or lived only with the remaining parent based on an awareness of the reduced caring capacity of the deceased parent due to psychological and/or substance-related problems. All the children in the study told how they had had a valued relationship with their deceased parent, and the parent's suicide had clearly caused a profound loss in their lives.

Most parents had been in contact with the children's schoolteachers to inform them of the parental suicide and the children's classmates were often also informed. Some children explicitly stated that they felt a sense of security knowing that their teachers and peers knew what they had been through, and some also reported that they had been offered professional support from a school counselor or nurse. Two children told of experiences of being bullied before their parent's suicide. In these cases, the information provided to the school seemed to have reinforced a sense of otherness when it did not lead to sympathy and support.

The children's narratives show that the prerequisites for mourning can vary. Many described an active social life on the outside, involving school, peers and spare time interests, but with grief vying for attention on the inside. Others described a situation dominated by grief and loneliness. Vanja, 12 years old, lacked friends and used to go into the school toilet to cry by herself. In retrospect, she reflected on the importance of the grief support camp: "I felt more alone before—that it was just us. Then when you came to the camp, it was like "it's not just us, there are many others as well'."

Suicide as a Traumatic and Stigmatizing Event

In the parents' narratives, a situation of chaos, loneliness and actively seeking professional help dominated their descriptions of life before participation in the grief support program. Kristina is a case in point: "I was a single mom with two children living at home and one that had moved out, and I felt very alone. I started to search on the Internet and found this and felt in my stomach that 'I need help'." Petra depicted the abrupt change in their family life: "Of course it was a shock when it happened and Johanna found him and I wasn't at home and all… my parents moved up to us and stayed the whole summer actually, until school started." A few parents, like Petra, described how they had received emotional and practical support from relatives and friends in their social networks, which was much appreciated in their vulnerable situation. More common, however, was for parents to speak about experiences of stigmatization, and lack of understanding and support. Lisa, the mother of a 6-year old boy, described how she stopped talking about her son's father in their social circle after encountering negative responses to his suicide, such as hurtful comments or avoidant behaviors. She reflected on the social judgements and insecurities surrounding suicide and remembered an incident in childhood, when her mother had talked about a mother who had died by suicide, that had affected her own understanding of suicide.

> "She has destroyed her children's lives," she said. And this mother became a monster in my eyes. It was so awful, you couldn't even touch the subject, that was the feeling I got. Zero sympathy or understanding for the mother, that she could have needed help, or that she maybe was suffering or.… No, it was just… she was demonized, and the children would get hell.

Similarly, Annika, the mother of a 14-year old boy, compared the social responses to suicide to those after more "normal" deaths, a difference that she believed hinders communication and support-seeking after suicide: "They don't know what to say.…If you'd said that 'he was killed in a car accident' oh that would've been 'so tragic' and 'incredibly sad', but when someone *did it to himself* it's another story. That's why it's so hard to talk to someone who has not been through the same thing." Kristina fell ill with a chronic illness after her husband's suicide. She described how she and her teenage children were left alone in this challenging situation.

> We've become alone (deep breath). Now it may be that I've also been ill. That people withdraw for that reason too. So, I don't know if it's been double for us, but friends and acquaintances have just disappeared. You'd think that when something like this happens, relatives might show up to help out with the kids, to support the kids and such, but no.…

In the narrated material as a whole, the suicide stigma and related difficulties of communicating about the parental suicide were a shared experience among the participants. They were also said to affect family interactions. The older children in particular sought to normalize themselves by avoiding talking about the suicide. Petra described how she negotiated between her conviction that children need to talk about the death of their parent to process the loss, and her daughters' resistance to talking about their father's suicide: "I haven't had a hard time saying that Mats took his own life, but I've restrained myself for the children's sake, because they were not ready. I understand that, since there are so many taboos and such about it."

The Decision to Participate

The parents described how they had found out about the BRIS grief support program mainly through local self-help organizations or social networks for suicide-bereaved adults on Facebook. Several children in the study said they were hesitant, or even protested, when their remaining parent had suggested they participate in the camp. Ivar, 14 years old, remembered that his mother had already made up her mind so there was no point in him protesting: "Well *then* I wasn't very into it (laughs). It felt like a really unnecessary and boring thing to do, but mom just went "this is great, let's go!", and we kind of had no choice, we just had to go with her." Other children were positive about going. Anders, 11 years old, for instance, told of his need to meet others and to talk about his parental loss experience: "It's hard to explain, but I thought it'd be fun to go there because you'd get to meet others and talk about *it*." Later he added that he was bullied at school and had never shared this experience with a peer.

All the parents considered that it might be conducive to the grieving process to go away as a family and focus on their suicide loss experience, in addition to meeting other families in a similar situation. Descriptions of some children's reluctance to participate—especially among the teenagers—also appeared in the parents' narratives. Erika, the mother of two teenage daughters, said: "I thought primarily of the girls, that… yes that they'd get to meet other children who have also lost a parent… and exchange experiences and see that it's not just *them*." She convinced her oldest daughter who was unwilling to go that it would be good for the family. Mona, the mother of two boys, found it helpful to be supported in a home visit by the BRIS leaders on how to respond to her teenage son's resistance.

> It was good that the children got to meet some of them as they would meet later, and one of them talked to Ivar and said: "you're not so into this, are you?" (laughs). They said that "well, teenagers are usually a bit negative before, but they're the ones who are the most positive after" (laughs). Then it was easier for me to "force" him to come along.

However, the parents' experiences also differed. Petra took the initiative to participate after her oldest daughter expressed a desire to meet others in the same situation.

> Johanna said that she wanted to meet others who'd experienced the *exact same* so.… Maybe it's difficult to meet someone who has been through the exact same, but here she could meet others who are in a very similar situation. So, then I made up my mind and realized somewhere that this is going to be tough and heavy, but I still wanted us to do it.

The Perceived Meaningfulness of the Grief Support Program

The meanings that the children and parents attributed to their participation in the BRIS grief support program are outlined below. They perceived the encounters with other suicide-bereaved persons, which contributed to support exchange and normalization, to be the most meaningful, but also the help gained to construct destigmatizing understandings of suicide.

The Importance of Connection and Normalization

Many children and parents expressed relief at having had an opportunity to meet other suicide-bereaved. This was described as having a normalizing effect that counteracted the reported suicide stigma in their daily lives. The importance of connection and normalization was mainly stressed in the youngest children's tangible appreciation of and joy at having met other suicide-bereaved children and was more specific in the older children's and parents' narratives. Hugo, 7 years old, just wanted to contribute one thing to the interview. He sat down with his back straight and stated in loud and determined voice: "I think you should get to stay *longer*… and I'd like to come back. In 1 year, there are 12 months and each month I think you should get to go there for 1 week." Agnes, 7 years old, exclaimed: "We got 1 day less than the others because Vanja (her sister) got chickenpox. 1 DAY LESS." She summarized her experience: "I think it's good that there are more who have parents who've died, but it's not so good that they've died." Similarly, 9-year old Mira said: "It's nice in a way that you feel that you're not alone." The children described how they formed new relationships mainly through the playful activities that took place between the grief-oriented group exercises. However, the exercises and conversations in the group meetings represented the backdrop against which this community was created, through a silent awareness of their shared experience of parental suicide. The social parts of the camp, the playful activities such as table tennis, floorball, and crafts with their new-found friends in their spare time, were the main interest, while their narrations about the content of the group meetings were more limited. Anders described his own, and he presumed the other children's, focus of attention during the camp.

> I think it was fun because you got to meet new people and I made new friends. Err that's it really. We kids probably didn't think much about *why* we were there—that it would help us—we didn't really think about that. When we were doing [the exercises] then we thought more, but there was also free time and then you thought of it more as a get together with friends.

When the group meetings were discussed, the children became serious and lowered their voices, which indicated that these were a sensitive subject, probably because they were closely connected to their parent's death—something which most children said they had used to avoid thinking and talking about in everyday life. Elvin, 11 years old, may have been representative of many of the children in terms of how he perceived the more grief-focused conversations: "Yes, it was quite fun, when we didn't talk about what… when we did crafts and stuff… and had juice and biscuits and so on and yes… but it was *really hard* when we talked about what had happened." Not many of the children described what they took from the group exercises and conversations, but Vanja said that it was helpful for her to talk about her own grief experience and listen to others. She said that she recognized herself in another girl's telling but, while she listened, she became aware of the time difference in their loss experience; that is, having lost a parent recently compared to having managed for several years without the deceased parent.

> It was good to get to see how others felt. I don't think there were so many who wanted to talk, but there were some who wanted to tell like *everything*. I recognized myself quite a lot and then there was a girl who said: "I forget my dad more and more and then it feels like I'm letting him down." I feel the same… So, she has managed without her father for one year and I've managed without mine for 5 years—there's a *little* difference.

Vanja likened the grief support camp to a place where broken hearts could heal. She thought back to when they were crafting in her group: "I remember that I painted a broken heart. Then I took glue plus BRIS and glued the heart together. BRIS attracts broken hearts and glues them together."

Kristina believed that it was good for her teenage son and daughter to meet other young people who were affected by a parent's suicide, with the explicit purpose of normalizing them in relation to suicide: "I think it helped a lot to see other *ordinary children*—that they weren't strange in any way. Because that's how you've felt…stared at, everyone was talking about us…and you felt very alone." As she drew on her own experiences, she added that she too found comfort in the meetings with similar people mourning a suicide: "Yes spontaneously, as awful as it may sound, precisely that there are others in the same situation, similar boat, that there are more like *us*." Erika was also grateful to have met other suicide-bereaved families: "I thought it was great to be there, both for me and for the girls, and to see that *we're not alone* in this and just talk to others who are in the same situation and share experiences." Louise, the mother of two boys, stressed the significance of these encounters with reference to her 11-year old son's negative peer experiences. She could see that he was supported in his grief by an awareness that he was not alone in his situation, and his still ongoing relationships with other children from the camp. In fact, all the children said in their interviews that they were pleased to have participated. This account by Louise gives a good description of the development that could be seen among the children during the camp stay, and especially the teenagers who had initially expressed doubts about participating.

> What I remember as the absolute best of all moments on both support weekends was to see these 13-year-olds who had been so quiet and introvert in the beginning. When you had heard their parents' stories about how they…everything they had said and done and enticed to get them there...and on the Sunday, after lunch when we were going home, they ran around and hugged each other and jumped for joy and hugged all the adults and "see you soon," and were so happy. Yes, I get chills.

Finally, Lisa, who had previously described how she had been silenced in her social circle, summed up her experience:

> The community, the warmth, the love and how you didn't feel alone, I took all of this with me. It was very important, how to relate to it all. For us, it feels natural to talk about it, not for everyone, but there you got a space to do it and meet other families. It was sad, but less lonely and isolating. You didn't feel strange or that you should apologize for what had happened... that you should be ashamed. Otherwise, I wouldn't have endured.

As the above shows, the participants were keen to express how much they had appreciated the opportunity to meet other suicide-bereaved families for normalization and support-exchange. In fact, these encounters stood out in the participants' narratives as the most meaningful contribution of the grief support program. However, when people come together based on an expected similarity, such as in the case of suicide bereavement, there is always a risk of disappointment and heightened exclusion if such a sense of belonging does not arise. One father described such a lack of connection. He explained this himself by saying that he is an introverted person who does not like to share emotionally charged topics. In his bereavement story he also positioned himself as different from the others; he said that most parents had struggled with their spouse's mental ill-health before suicide, while he did not consider that his wife was mentally ill. Another potentially negative aspect of this community building is the psychological burden of listening to others' detailed stories about traumatic deaths. One mother told how much she appreciated the group community, but at the same time found engaging with the others' suicide loss experiences emotionally draining.

Support to Construct Destigmatizing Understandings of Suicide

One educational element of the program that drew special attention in the interviews from both children and parents was how they had been assisted in age-appropriate ways to construct destigmatizing understandings of suicide. The notion that suicide is caused by a "thought disease," depression or emotional suffering was introduced and discussed in the groups, adapted to the age of the children and the circumstances of death described. The program theme had been accentuated by research about the negative effects of suicide stigma on mourning families, of which many participants already had lived experience.

In the youngest age group, which was children aged between four and six, the leaders drew a large head on a whiteboard and painted thoughts and emotions in different colors to illustrate the variations in a healthy mind. Gradually, they painted this over in black to show how dark thoughts shaped by a thought disease dominated the mind. Finally, at the time of suicide, only a small light remained in the deceased parent's mind, which was all the love for the child. What the parent may have felt and thought before the suicide was discussed, as well as what the parent could have done instead of dying. The children became involved and told how they thought the parent felt sad and lonely, and had difficulties finding a solution; they concluded with the leaders that it was sad that the parent had not sought help. The youngest children did not recapitulate this meaning construction in their interviews, but several of the older ones did.

In the older age group, Johanna, 13 years old, described how the leaders had likened the depressed mind to a withering garden. The gardener can usually nurture most plants but some are impossible to revive. Eventually, as the illness progresses, the lush garden turns into a withered landscape, and the gardener/depressed individual has difficulties finding new solutions: "I thought it was good that we talked about suicide as a thought disease and that it was nobody's fault. It was that person's *thoughts*... it all came down to that." Her reflection shows how this interpretation of suicide could help to counteract self-blame and stigmatization, since her conclusion opposes the notion that *someone* is to blame for suicide. Similarly, Vanja developed her thoughts on her father's suicide:

> Yes, it was a thought disease. I don't know what it's called... [I: Depression?] Yes. That you only think sad thoughts.... It wasn't *he* who did it, it was the *thoughts*. He couldn't think of anything joyful in life. He just thought that life was wrong and everything.

Through their repeated interpretations of suicide in their interviews, both girls illustrated how they had internalized a destigmatized understanding of their fathers' suicides long after the intervention. Anders used the same knowledge in his meaning construction. He saw his father's suicide as the result of negative thoughts and self-loathing: "He had a thought disease. We heard that he died from a thought disease and when you've got a thought disease you believe that you're bad and can't manage anything. It's like 'It would all be better without me'." He told how he found this explanation reasonable and comforting. The suicide could even be understood as an act of love, since he stated that his father believed he was a burden to his family, and that suicide would thus relieve the family of suffering. These destigmatizing meaning constructions were also attributed meaning in the parents' interviews. Mona told how this explanation of suicide had been recurrently re-established in family conversations by her two sons.

> That was something they could talk about. Then I thought that they [the leaders] must have talked about it in a good way, since they could talk about it (laughs). I think it's so important because it's where I think it's difficult. On the one hand, there are many taboos among the children, that you sit with "I wasn't worthy enough for dad" or something like that. I think this [new information] really came through. Because Elvin recounted it and it's so nice to hear it from him. He told me what he'd realized so he really understood. I think this was one of the most important things for the kids.

Most parents expressed gratitude for this help to find a shared meaning construction of suicide in grief; they described how it reduced feelings of guilt and shame, and became a model for how they could continue to talk with their children about suicide. One mother, however, held on to her resentment toward her former husband who she considered had failed in his parental responsibility to seek help instead of "deciding to leave."

Narratives of Change in the Grieving Process and Life

In their interviews, the children displayed insights about grief as a lifelong process and talked about how they used to cope with it in their daily lives. There was a general perception among the children that thoughts and emotions connected to their parental loss felt to various extents more manageable than before. Most parents, in turn, reflected on what they had learned and how they used this knowledge in family life. Their increased understanding of children's grief was explained as helpful and contributing to more supportive family communication. Many also told how the program had contributed a more hopeful view of life ahead. The main narrated changes are described below.

Children's Strengthened Agency and Management of Grief

In the children's talk about their lives now, they drew on lessons from the program and displayed agency by exemplifying how they had adjusted their coping strategies to grief-related emotions and needs. In the group sessions, the children had shared their loss experiences and strategies in discussions and were normalized and supported in their responses to loss. The children also processed their grief individually. In one exercise, the children had created their own first *aid kit*—a red glittery box in which they put written or drawn tips for themselves about what they could do to manage grief. Several children remembered the advice they had given themselves. Some brought out the saved boxes but declared that they no longer used them. Instead, they described the strategies they now used. Their primary advice to themselves from the program was to engage in different activities such as: "go outdoors and ride a bike," "bake cookies," "build with Lego" or "play with a friend," aimed at distraction to avoid thinking about the deceased parent. These and similar distraction strategies were being used. Ivar is a case in point: "I don't know, I try *not* to think about it (laughs). I do something else like scroll on YouTube or something. Focusing on something else is good." Selma, 9 years old, explained how she tried to activate herself to counteract painful thoughts, but on other occasions allowed herself to be sad.

> Sometimes I just walk around the apartment and: "okay, what can I do?" Then I start watering the flowers or something… and I make drawings and put glitter on and stuff… I want to be alone. Or I go to bed and cry a little bit… then I fix with my mobile phone.

Johanna, described how she had also adjusted to recurring moments of mourning connected to her father's suicide: "I just think about it…and I know that it'll pass. Because I think about it every day and you probably will for the rest of your life." She added: "If I'm really sad I talk to my friends or mum." Anders described how his primary strategy was to talk to someone if he felt sad, although at that time he was not experiencing a need to do so. "I usually talk about it at home, but now I don't do that so much." He had also received support from a school counselor. Agnes, 7 years old, said that she used to seek comfort by cuddling with her hamster, but she said: "Now I run to mom instead." Finally, Vanja described her coping strategy in grief. "When I'm sad I listen to Sofia and Alio." She had earlier explained that it had been helpful to listen to the other children's narrated experiences at the grief support camp and she had continued to listen to others' grief experiences in the form of song texts. In the interview, she played specific songs that she found had a healing effect on her. She reflected: "When I listen so Sofia, I feel like it's me who's singing. It's a beautiful song (she exhales). If something is worse though, like with her, you can really feel 'what a good life I have'."

In general, the children in the study showed an awareness of their emotions and needs in grief, and conveyed a perceived manageability in taking care of these. Grief was discussed as an ongoing process. Intrusive thoughts of the loss were said to come and go but were not considered dangerous or to be avoided at all costs. Instead, the children portrayed how they had created a space for grieving in their daily lives (27). In all the children's narratives, the remaining parent and sometimes other adults and friends were considered available resources that they could turn to for support.

Increased Parental Awareness of Children's Grief Responses and Needs

Most parents repeated pieces of advice that they had received from the group leaders and described them as helpful in interactions with their children. One main lesson that was raised was to strive for open and honest family communication about the suicide in order to support their children's meaning construction of their parent's suicide. At the enrollment interview, the parents were asked to tell the children that their parent had died by suicide, but not all of the children were aware of the detailed circumstances surrounding the death. Thus, the parents told how they had initiated conversations with their children after the camp to ensure that they received at least the basic information. Manuel sought advice regarding *when* and *how* he should tell his two preschool-aged daughters about their mother's suicide.

> For me, it was very important to be able to reach a… new way of dealing with the big issue with the children. Because I didn't really know when to tell… I've always been so busy with… their lives and their primary needs and then came this question: "when should I tell them?" Should I tell them when they're 13 and ask: "dad *how* did mom die?" I didn't know. BRIS had a psychologist who explained why it's important for them to know the truth. For me, that was *the big thing*. Going through this was very important for me, and to get it done the right way. Talking openly with the children feels good and like you're doing the right thing.

Louise was also unsure about how much she should disclose to her 9-year old son about his father's suicide. She described how her son was affected by his participation and the encounters with other suicide-bereaved children in such a way that he had later asked for more information. With guidance, the mother was able to meet his needs.

> Hugo told me that someone in his group had said that his father had shot himself.... Then he told me: "my dad died, and I don't

> know anything." I remember talking to the leaders about it. I told them that "Hugo doesn't know" and I brought it up in the parent group, because I'd initially been advised by a child psychologist that when it comes to such small children one shouldn't tell them about the event. So, I thought I'd handled it correctly but then I realized that it was a huge mistake that he didn't know. It also emerged in conversations we had afterwards that he'd been thinking about horrible, bloody things out in the garage… and that wasn't at all what had happened….

Among other things, the parents were informed that children who lose a parent through suicide may experience feelings of anger, shame and blame, which may be difficult to articulate in grief. Mona had been inspired to help her sons express such complicated dimensions of grief. Despite her efforts, however, her sons did not show much interest in talking about their emotions. She then drew on other advice from the program and changed her position.

> It was a frustration I had that they didn't talk. I needed to pull it out of them. We talked about Jakob in positive terms like "do you remember…?" and such, but not about….But then Lena (one of the leaders) said "you may think that it's a monolog, but it's a dialogue that goes on in the children's heads. You just can't hear it. They'll think on it, but it may not be you who gets to take part in it." Then I felt that's so true. Because if you've got something to say you should say it, even though you don't get a response. They listen and then it continues…and if they feel a need to talk about it, they'll do just that.

Open Communication Within the Family and Social Network

As noted above, the parents became aware through the program of how they could support their children in an open and honest family communication. The program content promoted such communication and the parents also frequently referred to specific exercises to illustrate the changes they had noticed. For example, each family created a collage by cutting out pictures from magazines to portray who the deceased parent was. Once complete, the children and parent presented the deceased parent to the other participants and received positive confirmations. The exercise was intended to help reconnect to the parent as a person separate from the suicide. Lisa described how it motivated her and her preschool-aged son to remember and talk about his deceased father: "The collage was such a good activity; to do it together, but also that the children could explain and present. It became such a good thing, to be reminded of his father, because it felt like we didn't talk enough about him."

Annika reported the changes she saw in her teenaged son in terms of him opening up to her in grief. He was an only child and had not talked about his father's suicide at all before their participation in the program.

> Our lives have really been affected by these weekends. They've been absolutely crucial. They made such a *difference*. Just knowing that there are more. Because it's a huge difference to participate here than going to a regular crisis group. It's not the same *at all*. Lars doesn't talk much, but after this he opened up and he has others to talk to as well.

Sibling relationships were sometimes also said to have improved. Kristina noticed how her teenage son and daughter began to share their grief after their participation and went to their father's grave together. Even communication in the families' social networks was commonly mentioned to have been enriched by the lessons from the grief support program. Louise described how her oldest son had spoken to a few friends about his father's suicide before his participation, which set a rumor in motion. After the program, he started to set boundaries for *when* and with *whom* he wanted to talk about his father's suicide, while her younger son, who had never told anyone that his father died by suicide, started telling the other children and teachers at his preschool and placed a photo of his father on his cloakroom shelf. Overall, participation in the grief support program was said to have contributed to a process of destigmatization, which made both parents and children feel more comfortable about talking about their parental loss in their social networks and less sensitive about the responses of others.

A Changed View of Life Ahead

The children who participated in the study often expressed a positive commitment to leisure activities and friends, and seemed preoccupied with life here and now, while the parents more often reflected on the family's future and expressed a more positive view of life ahead. In addition, the children who had reported previous experiences of bullying and loneliness in grief told of an improved situation after the program through new friendships and increased manageability of grief. Louise exemplified how participation in the grief support program could be described as a turning point in the participants' lives. She described how the more playful family activities had helped her to reconnect with her former self as an active and playful mother, before her husband's suicide 2 years before.

> I thought that the family activities were really great, because I didn't have the strength…I managed quite well to take care of everyday life here at home, but I wasn't….If I think back on myself from *that* time, I don't know if I *ever* laughed, that's how it was. I've always been an inventive person who likes to go outdoors and do things, but it disappeared quite a bit because I had no energy and no desire or anything…all my energy went on just surviving every day. So I was so incredibly grateful to be in a context where someone else organized the activities, where we got to laugh together again, and to do fun things that everyone enjoyed.

She explained how she had been revitalized through the meetings and the positive change she experienced in their family interaction. All in all, this was said to have helped her create a better life for herself and her children.

> Going was a turning point in my life. After that I could *live* again. It's so clear to me that I also began to relate to the children in a more natural way again, as it should be, not in a catastrophic way. Not in worry and such…but that we can trust that maybe we can

> also get to live and have a good time. Although this horrible thing has happened, we can probably actually do just that.

DISCUSSION

Discussion of the Main Results

This narrative evaluation has showcased the significance suicide-bereaved children and parents attributed to their participation in a family-based grief support program. The program is arguably similar to a "compassionate communities approach"[c.f. (32)] given that it aims to educate and support suicide-bereaved families to facilitate their coping with loss as a complement to existing healthcare. In addition, it draws attention to their situation and needs to a general public. First and foremost, both children and parents valued the opportunity to meet other families affected by a parent's suicide; this was said to contribute support exchange and normalization in relation to suicide as a stigmatizing death. An urgent need among suicide-bereaved family members to meet similar grievers has been reported and discussed in several studies, in order to share their experiences, and to learn from others, for example, how to manage the pain and the transition between life before suicide to life after suicide, [e.g., (16, 33–35)]. The community that emerged for most of the participants in this study was based on an overall quest to regain meaning and joy in life after suicide, and this was supported through the program structure and its content.

The result of this study backs up evidence from previous studies (19, 20) that a structure that offers a variation between "grief work" and outdoor recreation or play is particularly appropriate for children, because it supports relationship building and fits with children's developmental need to "go in and out of grief" to avoid suffering overly intense emotions (27, 29). This has also proved suitable for suicide-bereaved families as a whole. The parallel themed sessions in children and parent groups, as well as the family-oriented grief exercises, were said to facilitate a continuing dialogue in the family about sensitive issues related to the parent's suicide [cf. (5, 6)]. Similarly, the playful activities strengthened family interactions and supported a reorientation from the heavy yoke of grief to cheerful escapades in the family. The latter were said to contribute the hope of emotional survival of the suicide and for brighter prospects. This is an important finding, given how bereaved families can lock themselves into grief and tend to do fewer activities together after a parent's suicide (10). The overall empowering social context of the grief support camp stands in stark contrast to the descriptions of the social barriers to support in the participants' daily lives linked to a prevailing suicide stigma. Like so many people mourning a suicide in the family, the narratives in this study echo how both suicide-bereaved children and their parents usually struggled alone before arriving at the grief support camp.

A central element of the program is the help to construct a tolerable meaning of parental suicide that does not stigmatize the bereaved family. Through the meaning reconstruction in the program, the participants learned that the parent suffered from a psychological condition influenced by destructive thoughts, and ultimately sought to escape emotional pain, which clarified that *no person* was to blame. The children in particular voiced release from self-blame and their self-esteem appeared restored through this explanatory model and the specific message that they were not unloved or rejected by the deceased parent (8). From a social constructionist and narrative perspective on loss, grief and trauma (36), such meaning reconstruction in the wake of loss is desirable. It addresses the *crisis of meaning* (11) that arises when suicide challenges previously taken-for-granted beliefs about this life world and the self. It also has the potential to combat stigma and contribute to reconciliation in relationships, including with the deceased, restored identities and even post-traumatic growth (36, 37). The above meaning reconstruction has health benefits too, since feelings of blameworthiness have been associated with grief difficulties, complicated grief, PTSD, depression and other mental health difficulties (38), while the role of self-forgiveness in suicide bereavement has been linked to a decrease in depression and suicidality among suicide loss survivors (39). In addition, the results show that even very young children, can benefit from being included in family communication on and meaning reconstruction of the parent's suicide (40). The open and honest communication that the program encourages between parents and children opposes a more protectionist stance toward children and empowers their position in grief. This is in line with current recommendations that children should preferably be informed of the true circumstances of a death in a developmentally adapted manner (8, 35, 41).

The parents highly valued the educational elements of the program on childhood bereavement after parental suicide and gave several examples of how this knowledge was implemented in family life. In general, the parents expressed increased confidence about their capacity to support their children, which was confirmed in the children's reports on the parent as a resource in their grief. Altogether, this supports the assumption, based on research, that when parents are supported in grief and in their parenting, this has positive effects on children [see (18)].

The narrated changes in the children's grief processes and lives indicated an increased sense of coherence (28). In their telling of experience, the children seemed empowered in their relation to the parental suicide and in their dealing with grief. They appeared to have integrated non-stigmatizing comprehensions of their parent's suicide and performed agency and manageability in grief by recounting their purposefully used coping strategies. They also demonstrated interest in relationships and social activities they found meaningful. This adds to the research on the effects of a family-based approach to work with suicide bereaved children [cf. (5, 6)].

Finally, this study describes an ethical approach to research interviews with children on sensitive subjects. The children's responses showed that even though emotions related to a parent's problems and death can surface in an interview situation, they appreciated being able to contribute their experiences to research in the way they chose.

Limitations

A prerequisite for children's participation in a grief support camp of this kind is the remaining parent's ability to identify such a need. A challenge for research and practice is thus to reach the suicide-bereaved children who do not have a supportive remaining parent—the children who themselves have several risk factors for developing ill-health and suicidality. This study is biased in this regard since all the parents reached out for professional help. In addition, the self-selected sample of participants constituted about half of all the families who participated in the BRIS grief support program. It can be assumed that those who were particularly positive about their participation wanted to "give back" out of gratitude or to help gain the program permanent status. However, those with experiences of a different kind might also be motivated to air their opinions in order to improve program content or prevent such a process gaining legitimacy. In telephone contacts with parents who refrained from participating, their decision was motivated by an overly pressing life situation as a single parent and/or problems with their children's functioning and well-being linked to grief, for which they had sought professional help. All, however, expressed gratitude for their participation in the program. A further limitation of the study was that the children were not invited individually by telephone, but only through an age-adapted and personally addressed letter. The parents were subsequently asked whether they and/or their children wanted to participate. Based on their decision, plans were made for a home visit or the contact was ended. It is possible that more children, especially teenagers, would have been more inclined to participate in the study if they had been in direct contact with the author. Lastly, the long-term follow-up in the study made it possible for the participants to reflect on how their lives had been affected by their participation in the grief support camp 18 months later, but this design made it difficult for the youngest children to remember. From a child perspective, a longitudinal approach with an initial short-term follow-up and further follow-up would have been more appropriate.

CONCLUSION

This family-based grief support program in the format of a weekend camp with a particular focus on children's grief has been shown to have helped to open up family communication and strengthen family resources for coping with a parent's suicide. It has great potential to counteract complications in suicide-bereavement—not least those induced by stigmatizing attitudes and self-imposed blame for suicide—and to promote health and well-being in this vulnerable group. Such a psychoeducational measure is thus considered to be a highly effective intervention with a relatively low cost compared to other traditional approaches. However, although influenced by social relationships and norms, grief is a unique and highly personal process, which means that not everyone will benefit from professionally led support groups. For example, individual preferences can make it difficult to disclose one's own experiences or listen to the stories of others. Therefore, other professional measures should also be considered with the aim of meeting the often-overlooked needs for support after a suicide in the family.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Regional Ethical Review Board in Uppsala, Sweden (Id. 2015/504). Written informed consent to participate in this study was provided by all the children aged 8 and above, and all the children's legal guardians.

AUTHOR CONTRIBUTIONS

The author confirms being the sole contributor of this work and has approved it for publication.

FUNDING

The study was funded by FORTE: Swedish research council for health, working life and welfare (Grants 2018/01052).

ACKNOWLEDGMENTS

The author would like to express her sincere gratitude to the children and parents who provided their experiences for the study, as well as Sofia Grönkvist and Lotta Polfeldt from BRIS for their cooperation. She would also like to thank Ulla Forinder for her involvement in the study and her constructive reading of previous drafts of this article, as well as Stina Michelson and the two reviewers for their valuable comments.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.783066/full#supplementary-material

REFERENCES

1. Swedish Health and Medical Services Act (2017:30, chap 5: 7§). p. 979. Available from: https://www.riksdagen.se/sv/dokument-lagar/dokument/svensk-forfattningssamling/halso--och-sjukvardslag-201730_sfs-2017-30 (accessed March 17, 2021).
2. Cerel J, Aldrich R. The impact of suicide on children and adolescents. In: Jordan JR, McIntosh JL, editors. *Grief After Suicide: Understanding the Consequences and Caring for Survivors*. (2011). p. 81–92. London: Routledge.
3. Guldin M-B, Li J, Søndergaard Pedersen H, Obel C, Agerbo E, Gissler M, et al. Incidence of suicide among persons who had a parent who died during their childhood: a population-based cohort study. *JAMA Psychiatry*. (2015) 72:1227–34. doi: 10.1001/jamapsychiatry.2015.2094
4. Hjern A, Arat A, Rostila M, Berg L, Vinnerljung B. *Hälsa och sociala livsvillkor hos unga vuxna som förlorat en förälder i dödsfall under barndomen* [*Health and Social Living Conditions of Young Adults who Lost a Parent Through Lethal Causes During Childhood*]. Report 2014, 3. Linnaeus University, Växjö, Sweden (2014).
5. Daigle M, Labelle RJ. Pilot evaluation of a group therapy program for children bereaved by suicide. *Crisis*. (2012) 33:350–7. doi: 10.1027/0227-5910/a000147
6. Pfeffer CR, Jiang H, Kakuma T, Hwang J, Metsch M. Group intervention for children bereaved by suicide of a relative. *J Am Acad Child*

Adolesc Psychiatry. (2002) 41:505–13. doi: 10.1097/00004583-200205000-00007

7. Ribbens McCarthy J. "They all look as if they're coping but I'm not": The relational power/lessness of "youth" in responding to experiences of bereavement. *J Youth Stud*. (2007) 10:285–303. doi: 10.1080/13676260701262574
8. Schreiber JK, Sands DC, Jordan JR. The perceived experience of children bereaved by parental suicide. *OMEGA: J Death Dying*. (2017) 75:184–206. doi: 10.1177/0030222815612297
9. Hung N, Rabin L. Comprehending childhood bereavement by parental suicide: a critical review of research on outcomes, grief processes, and interventions. *Death Stud*. (2009) 33:781–814. doi: 10.1080/07481180903142357
10. Cerel MA, Fristad MA, Weller EB, Weller RA. Suicide-bereaved children and adolescents: II. Parental and family functioning. *J Am Acad Child Adolesc Psychiatry*. (2000) 39:437–44. doi: 10.1097/00004583-200004000-00012
11. Neimeyer RA, Sands DC. Meaning reconstruction in bereavement: from principles to practice. In Neimeyer RA, Harris DL, Winouker HR, Thornton GF, editors. *Grief and Bereavement in Contemporary Society: Bridging Research and Practice*. (2011). p. 9–22. New York: Routledge.
12. Silvén Hagström A. Why did he choose to die?: A meaning-searching approach to parental suicide bereavement in youth. *Death Stud*. (2019) 43:185–92. doi: 10.1080/07481187.2018.1457604
13. Jordan JR. Lessons learned: forty years of clinical work with suicide loss survivors. *Front Psychol*. (2020) 11:766. doi: 10.3389/fpsyg.2020.00766
14. Silvén Hagström A. "The stranger inside": suicide-related grief and "othering" among teenage daughters after the loss of a father to suicide. *Nordic Social Work Res*. (2013) 3:37–41. doi: 10.1080/2156857X.2013.801877
15. Sterner Demi A, Howell C. Hiding and healing: resolving the suicide of a parent or sibling. *Arch Psychiatr Nurs*. (1991) 5:350–6. doi: 10.1016/0883-9417(91)90036-5
16. Wilson A, Marshall A. The support needs and experiences of suicidally bereaved family and friends. *Death Stud*. (2010) 34:625–40. doi: 10.1080/07481181003761567
17. Dyregrov K. (2009). How do the young suicide survivors wish to be met by psychologists? A user study. *OMEGA—J Death Dying*. (2009) 59:221–38. doi: 10.2190/OM.59.3.c
18. Bergman A-S, Axberg U, Hanson E. When a parent dies: a systematic review of the effects of support programs for parentally bereaved children and their caregivers. *BMC Palliative Care*. (2017) 16:39. doi: 10.1186/s12904-017-0223-y
19. McClatchey IS, Wimmer JS. Healing components of a bereavement camp: children and adolescents give voice to their experiences. *OMEGA—J Death Dying*. (2012) 65:11–32. doi: 10.2190/OM.65.1.b
20. Clute MA, Kobayashi R. Are children's grief camps effective? *J Social Work End Life Palliat Care*. (2013) 9:43–57. doi: 10.1080/15524256.2013.758927
21. Currier JM, Holland JM, Neimeyer RA. The effectiveness of bereavement interventions with children: A meta-analytic review of controlled outcome research. *J Clin Child Adolesc Res*. (2007) 36:253–9. doi: 10.1080/15374410701279669
22. Andriessen K, Krysinska K, Hill N, Reifels L, Robinson J, Reavley N, et al. Effectiveness of interventions for people bereaved through suicide: a systematic review of controlled studies of grief, psychosocial and suicide related outcomes. *BMC Psychiatry*. (2019) 19. doi: 10.1186/s12888-019-2020-z
23. Linde K, Treml J, Steinig J, Nagl M, Kirsting A. Grief interventions for people bereaved by suicide: a systematic review, *PLoS ONE*. (2017) 12:e0179496. doi: 10.1371/journal.pone.0179496
24. Baú V. A narrative approach in evaluation: "Narratives of change" method. *Qual Res J*. (2016) 16:374–387.
25. Constantino T, Greene J. Reflections on the use of narrative in evaluation. *Am J Eval*. (2003) 24:35–49. doi: 10.1177/109821400302400104
26. Gillispie A, Grönkvist S, Hedrenius S, Polfeldt L. *Stödja Familjer i Kris: Att Arbeta med Parallella Stödgrupper för Barn och Föräldrar [To Support Families in Grief: Working with Parallel Support Groups for Children and Parents]*. Stockholm: Gothia. (2012).
27. Stroebe M, Shut HA. Family matters in bereavement: towards and integrative intra-interpersonal coping model. *Perspect Psychol Sci*. (2015) 10:873–9. doi: 10.1177/1745691615598517
28. Antonovsky A. *Health, Stress and Coping*. San Francisco: Jossie–Bass. (1979).
29. Worden W. *Children and Grief: When a Parent Dies*. New York, NY: Guilford Press. (1996).
30. Riessman CK. *Narrative Methods for the Human Sciences*. London: SAGE. (2008).
31. Punch S. Research with children: The same or different from research with adults? *Childhood*. (2002) 9:321–41. doi: 10.1177/0907568202009003005
32. Zuniga-Villanueva G, Ramirez-GarciaLuna JL, Villafranca-Andino RI. A compassionate communities approach in a grief and bereavement support program: bridging the gap in palliative care. *J Social Work End Life Palliat Care*. (2021) 17:9–18. doi: 10.1080/15524256.2021.1894309
33. Ali F, Lucock M. "It's like getting a group hug and you can cry there and be yourself and they understand": family members' experiences of using a suicide bereavement peer support group. *Bereavement Care*. (2020) 39:51–8. doi: 10.1080/02682621.2020.1771951
34. Ross V, Kõlves K, De Leo D. Exploring the support needs of people bereaved by suicide: a qualitative study. *OMEGA: J Death Dying*. (2021) 82:632–45. doi: 10.1177/0030222819825775
35. Dyregrov K, Dyregrov A. Helping the family following suicide. In: Monroe B, Kraus F, editors. *Brief Interventions with Bereaved Children, (Oxford Scholarship Online)*. (2011). doi: 10.1093/acprof:oso/9780199561643.001.0001
36. Neimeyer RA, Klass D, Dennis MR. A social constructionist account of grief: Loss and the narration of meaning. *Death Stud*. (2014) 38:485–98. doi: 10.1080/07481187.2014.913454
37. Neimeyer RA. Fostering post traumatic growth: A narrative elaboration. *Psychol Inq*. (2004) 15:53–9. doi: 10.1207/s15327965pli1501_02
38. Fiegelman W, Cerel J. Feelings of blameworthiness and their associations with the grieving process in suicide mourning. *Front Psychol*. (2020) 11:610. doi: 10.3389/fpsyg.2020.00610
39. Levi-Belz YC, Gilo T. Emotional distress among suicide survivors: the moderating role of self-forgiveness. *Front Psychiatry*. (2020) 11:341. doi: 10.3389/fpsyt.2020.00341
40. Nadeau J. *Families Making Sense of Death*. Newbury Park, CA: Sage. (1997).
41. Silverman PR. *Never Too Young to Know: Death in Children's Lives*. New York: Oxford University Press. (2000).

Conflict of Interest: The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 13 December 2021
doi: 10.3389/fpsyt.2021.799626

Expressed Emotion and Attributions in Parents With Schizophrenia

Lynsey Gregg[1]*, *Rachel Calam*[1], *Richard J. Drake*[1,2] *and Lauren Wolfenden*[1]

[1] Division of Psychology and Mental Health, School of Health Sciences, Faculty of Biology, Medicine and Health, Manchester Academic Health Science Centre, The University of Manchester, Manchester, United Kingdom, [2] Greater Manchester Mental Health NHS Foundation Trust, The University of Manchester, Manchester, United Kingdom

We examined expressed emotion (EE) and attributions in parents with schizophrenia and compared them to parents without serious mental illness (SMI) in order to better understand the emotional climate of families in which a parent has schizophrenia. Parenting practices and parental reports of child behavior were also compared between the two groups. The relationship of EE to attributions was examined in each group separately. Relationships between parental mental health, EE, and attributions were explored in the parents with schizophrenia only. The Camberwell Family Interview was used to determine both EE and attributions in 20 parents with schizophrenia and 20 parents without SMI. We found that more parents with schizophrenia were rated as high EE than those without (60 and 35%, respectively) although this was not a statistically significant difference. Parents with schizophrenia demonstrated significantly more hostility and criticism toward their children than those without SMI and made more child-blaming attributions. Blame was associated with increased hostility, less warmth, and fewer positive remarks. Parental warmth was related to greater parenting self-efficacy, less harsh parenting practices, better child behavior, and a more positive parent–child relationship. We conclude that EE and attributions are potential explanatory variables to be considered in the development of preventative and early intervention strategies for families with a parent with schizophrenia or other psychotic disorder. Blame and warmth are modifiable factors that could be targeted within family and parenting interventions.

Keywords: serious mental illness (SMI), psychosis, child behavior, family environment, warmth, blame

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
Mary V. Seeman,
University of Toronto, Canada
Hanna Christiansen,
University of Marburg, Germany

****Correspondence:***
Lynsey Gregg
lynsey.gregg@manchester.ac.uk

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *21 October 2021*
Accepted: *19 November 2021*
Published: *13 December 2021*

Citation:
Gregg L, Calam R, Drake RJ and
Wolfenden L (2021) Expressed
Emotion and Attributions in Parents
With Schizophrenia.
Front. Psychiatry 12:799626.
doi: 10.3389/fpsyt.2021.799626

INTRODUCTION

Schizophrenia is a chronic and severe illness, with a high global disease burden and significant economic cost (1). The majority of people with psychotic disorders such as schizophrenia are also parents, and their children have been reported to be at significantly increased risk of poor outcomes, including poorer mental health in adulthood (2, 3). Evidence suggests that both genes and environment contribute to an increased risk of intergenerational transmission of schizophrenia (4) and the family environment could plausibly be considered a modifiable environmental factor contributing to this risk.

The importance of the family environment in the development and maintenance of childhood socioemotional and behavioral problems and wellbeing has been well-established in families without a parent with SMI (5–8). There is evidence that dysfunctional and stressful family environments negatively influence parental executive functioning (9) and parenting and family functioning more broadly (10). In families with a parent with a psychotic disorder, who are additionally disproportionately affected by parental unemployment, isolation and poverty (11) these impacts may be even greater. Psychosis has been found to interfere with the establishment and maintenance of important family routines (12) and stigmatization may serve to prevent help-seeking in families that are struggling (13, 14). Ultimately, children of parents with schizophrenia and other psychotic disorders are more likely to be removed from their parents' care (15, 16) with long term consequences for both parent and children's well-being and at significant economic cost for wider society.

Family environments can be explored using the concept of Expressed Emotion (EE) (17). EE is a well-validated measure of the emotional climate of the family which captures the communication style and attitude of a relative when speaking about another family member. Typically assessed by the Camberwell Family Interview (CFI) (17), EE has been widely studied in the family members of individuals with schizophrenia and other psychotic disorders and has been found to be predictive of relapse and hospitalization: Individuals who reside with families characterized by criticism, hostility and emotional-over involvement ("high-EE environments") are more likely to relapse than those who do not reside in such an environment (18). Conversely, positive affect in the family (characterized by high warmth) has been found to have a protective effect and reduce the likelihood of relapse (19, 20). EE has also been associated with symptoms and functioning in individuals identified as vulnerable to a psychotic disorder (21).

Although, it is plausible that EE may be a potential mechanism of intergenerational transmission in schizophrenia, no research has sought to determine EE in parents living with schizophrenia. This is particularly surprising since parents with mental illness are more likely to have been raised in a family environment characterized by high EE themselves (22). Research to date has primarily focused on families with a depressed parent, finding that EE is typically higher in depressed parents than in those without depression [e.g., (22)] and that in these families, high EE is linked to poorer child behavior [e.g., (23, 24)]. Parental expressed emotion, particularly criticism, has also been associated with the development and maintenance of a range of childhood disorders in families without parental mental illness (25) including increased behavioral problems in individuals with autism (26, 27). Longer terms impacts have also been observed, with parental EE linked to depression, anxiety, and substance misuse in later adulthood (28, 29).

The mechanism of action is not yet established, but the assumption underpinning EE research is that the way parents talk about their relative is indicative of the way they behave toward that relative on a day-to-day basis (30) and causal interpretations of behavior, in the form of attributions, are believed to be the driver of EE (31).

Attributions are beliefs about causality, and are expressions of the way people think about the relationship between an event and a cause (32). Attributional theory suggests that uncovering what people believe about events and their causes is a way to understand and predict their emotional and behavioral responses to those events (33). In the context of parenting, attributions ascribe meaning to children's behavior and guide how the parent relates and responds to their child (34, 35).The key dimensions underlying causal thinking are locus of causality (whether the parent believes the cause of the child's behavior (the "event") to be internal to or external to the child), controllability (whether the parent considers that the child could control (i.e., prevent) the outcome) universality (whether the cause is personal and unique to the child) and stability (is the cause likely to recur?). Attributions that reveal the parent to consider the cause of an event to be both internal and personal to the child, as well as controllable by the child, are considered "blaming."

When parents blame a child for behaviors or events their parental responses tend to be more critical and their parenting harsher (23, 24), further contributing to the development and maintenance of child behavioral problems (34, 36, 37).

Exploration of parental EE and attributions may be an effective way of understanding family dynamics in families affected by parental schizophrenia, and EE could potentially be a useful target for early intervention to improve family functioning and improve long term outcomes in these families. Therefore, we explored EE and attributions in parents experiencing schizophrenia, and compared them to parents without serious mental illness (SMI) in the first study of its kind. In line with research conducted with depressed parents we predicted that parents with schizophrenia would demonstrate greater criticism, greater hostility and less warmth toward their children. We also anticipated that they would make more child-blaming attributions and that the frequency of these blaming attributions would be related to EE (specifically, greater criticism and hostility, and less warmth). Relationships between parental mental health, parenting practices, attributions, and EE were also explored in the parents with schizophrenia in order to ascertain whether mental health was associated with facets of EE or attributions and whether EE and attributions had a directly impact on parenting practices.

METHODS

Ethical Approval

Ethical approval was obtained from Greater Manchester West National Research Ethics Committee.

Sample

Participants were required to be over 18 years old; a parent/primary care-provider, living with and having direct parenting responsibilities for a child aged between 3 and 11 years. Spoken English was required in order to provide informed consent and complete assessments. Families with multiple children nominated an index child with whom they expressed the greatest parenting challenges. Diagnoses for the parents with schizophrenia were corroborated using International Classification of Diseases (ICD-10) checklists and case note review. Those meeting the criteria for schizophrenia

(F20-F29) were eligible. To reduce risk of distress, participants were excluded if they had recently been discharged from inpatient care or if there were known intentions for their child to be removed from their care.

Recruitment

Recruitment to the clinical group was from four NHS Trusts across Greater Manchester, UK. Community Mental Health Teams and Early Intervention Services were approached. Voluntary sector and social services were also utilized, including Local Authority Family Services. Adverts were also placed on online parenting forums and in schools, local authority services, GP surgeries and nurseries to boost recruitment to both groups. Letters were sent to potentially eligible parents registered on a research volunteer database at the University of Manchester. Participants without SMI self-referred and contacted the research team directly.

Measures

The Positive and Negative Syndrome Scale (PANSS) (38) and the Psychotic Symptoms Rating Scales (PSYRATS) (39) were used to determine symptom severity in the parents with schizophrenia. The rater established inter-rater reliability after rating ten "gold standard" video-recorded interviews prior to recruitment taking place, achieving an average intraclass correlation coefficient of 0.85. Parental well-being was assessed in both groups using the Warwick Edinburgh Mental Wellbeing Scale (WEMWBS) (40) and negative emotional states were assessed using the Depression, Anxiety and Stress Short Form Scale (DASS-21) (41). Parenting and child behaviors were explored using a range of measures: Parental self-efficacy was explored using the Parenting Task Checklist (PTC) (42). The Parenting and Family Adjustment Scales (PAFAS) (43) assessed parenting strategies and family dynamics and the Parenting Scale (PS) (44) assessed a range of parenting behaviors including the use of permissive (lax) and harsh (over-reactive) approaches. Child behavior was assessed using the Eyberg Child Behavior Inventory (ECBI) (45) which determines intensity and frequency of problematic child behaviors. Alpha levels were in the good to excellent range ($\alpha = 0.70$–0.95) for all measures except the setting subscale of the parenting task checklist which measures parenting self-efficacy in 14 different settings. Reliability for this subscale was very low at $\alpha = 0.20$.

The Modified Camberwell Family Interview

The original CFI is a standardized semi-structured interview used to assess the emotional attitudes of relatives toward their family member with schizophrenia (17) and is the "gold standard" measure of EE. The modified CFI used in this study was based on previous researchers' adaptations (23, 46). These adaptations focus the CFI on problematic child behaviors as opposed to adult symptom behaviors. Procedures and rating classifications remained unchanged. CFIs were rated for EE by a researcher (LG) who had been formally trained by one of the original developers of the CFI (CV). LG achieved excellent average inter-rater reliability against criterion gold standard raters (0.94). The modified CFI is available from the corresponding author.

The CFI provided ratings of EE on five dimensions: criticism, hostility, EOI, warmth and positive remarks. To rate criticism or "critical comments" statements indicating parental annoyance toward particular behaviors or characteristics are noted and frequency counts collected. Hostility, EOI, and warmth are coded by making conclusions based on information from the entire interview. Hostility is rated when criticism is either generalized or there is rejection and is measured using a four point scale: 0 = no hostility; 1 = generalization only; 2 = rejection only; and 3 = generalization and rejection. EOI uses a six-point-scale with a threshold of 3, and is rated when a parent demonstrates excessive overprotective behaviors or emotional responses toward their child (46). Warmth is an overall rating of sympathy, empathy, interest in and closeness to the child scored from 0 = no warmth to 5 = extreme warmth. Positive remarks reflecting positive parent–child relationships or closeness are noted and frequency counts collected. Parents are classified as "high" EE if there are ≥ 6 critical comments; ≥ 3 EOI ratings or hostility is present.

Causal Attributions: The Leeds Attributional Coding System

In line with previous research [e.g., (23, 46)] spontaneous parental casual attributions regarding child problem behaviors were extracted from the CFIs using the modified Leeds Attributional Coding System (LACS) (47). The LACS was originally modified by White and Barrowclough (24) for parents experiencing depression. A coding manual created by Peters et al. (46) was adapted for the current study to include examples from parents with schizophrenia. A copy is available from the corresponding author.

Following extraction, attributional statements were coded by the second author and an independent rater (AP) along the four key dimensions included in the LACS: internal/external, controllable/ uncontrollable, personal/universal, and stable/unstable (see **Table 1**). Statements were coded in accordance with the instructions given in the LACS, which necessitates one rating on each attributional dimension, using a binary scale for each side of the dimensions. A score of 1 was given for the internal, controllable, personal, and stable ends of the four dimensions and a score of 3 was given for the external, uncontrollable, universal, and unstable ends. A score of 2 was assigned when causes appeared to be a mixture of both ends of the dimension (e.g., a cause that was partly controllable and partly uncontrollable by the child).

Each attributional statement therefore generated four codes, one for internal/external, one for controllable/uncontrollable, one for personal/universal, and one for stable/unstable. A score of 9 was used in rare cases where the cause could not be rated. For each interview, the total number of attributions made that were rated internal, controllable, and personal to the child were counted ("blaming attributions"). Reliability of extraction and rating between raters (LW and AP) was established on a sample of eight randomly selected CFIs.

Proportional attribution scores indicate the general direction of causality on each dimension and were calculated by dividing the number of causes scored as "1" by the number of causes given a score of 1 or 3 (46). They range between 0 and 1 and scores

TABLE 1 | Attribution dimensions.

Dimension	Description
Internal–external	Internal: The cause is a "feature" of the child (e.g., personality traits, physical characteristics, illnesses/symptoms, behavior, thoughts, feelings, knowledge, opinions, and beliefs). External: Factors outside of or imposed on their child. e.g., Actions or traits of other people, the weather, or location.
Personal–universal	Personal: A specific cause leading to an event that would not happen to others (e.g., personality traits, information that identifies that child from others/specific about their child). Universal: Expected or understandable behavior for a child of similar age and/or gender (e.g., typical behavior or reactions, conditions).
Controllable–uncontrollable	Controllable: Belief that behaviors could be changed, influenced or controlled by child (e.g., Tantrums, sulking, aggression, wanting attention, voluntary behaviors, habits, attitudes, laziness, and irritability). Uncontrollable: Belief that that behavior is outside the control of the child, e.g., fear, accidents, illnesses, personality traits/dispositions, characterizes, emotional responses, environmental, or situational factors).
Stable–unstable	Stable: The cause as frequent feature or characteristic of the child (e.g., habits or behavior patterns, not sleeping/tiredness, skills, socio-economic difficulties, or life events). Unstable: The cause is in past tense or infrequent incidents (e.g., moods, ideas, thoughts, single actions/behaviors, luck-fate, or accidents).

>0.50 represent attributions that were predominantly internal, controllable, personal, and stable.

Data Analysis

Data were analyzed using SPSS version 25. EE Criticism and hostility were not normally distributed and were log transformed for analysis. Parents with schizophrenia were compared to parents without SMI using t-tests and Chi squared tests. Pearson's r correlations were used to assess hypothesized relationships between EE and attributions. Exploratory analyses of relationships between EE, attributions, parental mental health, and parenting also used correlation and t-tests. Multiplicity adjustments were not made for these exploratory analyses, despite the large number of tests conducted, in order to avoid accidentally missing true effects (48). Multiple linear regression was used to determine the relative impact of parental mental health status (schizophrenia vs. no SMI) on EE and attributions compared to demographic variables.

RESULTS

Participant Characteristics

Participants in the clinical group had diagnoses of schizophrenia ($n = 11$) or paranoid schizophrenia ($n = 9$). Duration of psychosis was 4–5 years (5%), 5–10 years (50%), 11–20 years (30%), and >20 years (15%). **Table 2** provides an overview of key demographic characteristics and family circumstances for both groups. Significant differences were observed with regards to parental age, household composition and employment. Parents with schizophrenia were younger [$t_{(38)} = 2.72$, $p < 0.05$] and more likely to be single parents [$X^2{}_{(1)} = 14.55$, $p < 0.001$] and unemployed [$X^2{}_{(2)} = 16.04$, $p < 0.001$].

Expressed Emotion: The Modified Camberwell Family Interview

A higher percentage of parents with schizophrenia were rated as high EE overall ($n = 12$, 60%) compared to the non-clinical group ($n = 7$, 35%) although this difference was not found to be statistically significant. They made significantly more critical comments than those without SMI and were more likely to be categorized as highly critical with 50% making 6 or more critical comments compared to 20% of those without SMI [$X^2{}_{(1)} = 3.96$, $p < 0.05$]. Parents with schizophrenia were also more likely to be rated as "hostile" with seven (35%) meeting criteria for this rating compared to just one (5%) in the non-clinical group [$X^2{}_{(1)} = 4.33$, $p < 0.05$]. Six of these seven parents made "rejecting" comments. For the other dimensions of EE (EOI, warmth, and positive remarks) there were no significant differences between the two groups (see **Table 3**).

Spontaneous Parental Causal Attributions

A total of 950 attributions were extracted from the 40 CFIs (567 from the parents with schizophrenia, 383 from the parents without SMI). The mean rate of attributions per minute indicated that the parents with schizophrenia made more attributions than those without. **Table 3** outlines the proportional attribution scores for both groups indicating the predominant direction of causality. Parents with schizophrenia made more attributions that were rated as personal to the child and stable in nature. They also made significantly more "blaming" attributions (attributions rated as internal and personal to, and controllable by, the child).

Relationship of Demographic Variables to Expressed Emotion and Attributions

Since the parents with schizophrenia differed significantly from those without with regard to age, household composition, and employment status (parents with schizophrenia were younger, more likely to be single parents and less likely to be employed) we conducted multiple linear regression analyses to determine the relative impact of SMI status on the EE and attribution variables where differences between the two groups had been observed. In these regressions age, household composition (single vs. not) and employment status (employed vs. not) were entered in the first

TABLE 2 | Participant characteristics.

	Parents with schizophrenia	Parents without SMI
Parent gender, *N* (%)		
Female	19 (95%)	19 (95%)
Male	1 (5%)	1 (5%)
Parent ethnicity, *N* (%)		
White	15 (75%)	16 (80%)
Black	2 (10%)	1 (5%)
Chinese	1 (5%)	0
South Asian	1 (5%)	1 (%%)
Mixed	1 (5%)	2 (10%)
Parent age, mean (SD)	33.9 (7.5)	39.9 (6.4)
Number of children, mean (range)	2 (1–5)	2 (1–6)
Child's gender, *N* (%)		
Female	6 (30%)	7 (35%)
Male	14 (70%)	13 (65%)
Child's ethnicity, *N* (%)		
White	12 (60%)	15 (75%)
Black	1 (%)	1 (5%)
South Asian	1 (5%)	0
Mixed	6 (30%)	4 (20%)
Child's age, *mean (range)*	8 (3–11)	6 (4–10)
Household composition, *N* (%)		
Single parent household	15 (75%)	3 (15%)
Dual parent household	5 (25%)	17 (85%)
Parental employment, *N* (%)		
Unemployed	19 (95%)	7 (35%)
Part time employment	1 (5%)	7 (35%)
Full time employment	0	6 (30%)

TABLE 3 | Differences in expressed emotion, attributions, parenting, and reports of child behavior between groups.

Expressed emotion	Parents with schizophrenia	Parents without SMI	*p*
	Mean (SD)	**Mean (SD)**	
EE Criticism*	8.3 (7.20)	3.3 (2.25)	0.043
EE Hostility*	0.75 (1.21)	0.05 (0.22)	0.021
EE EOI	1.05 (1.05)	1.05 (1.15)	1.00
EE Warmth	1.80 (0.89)	2.25 (0.79)	0.099
EE Positive remarks	2.70 (2.45)	3.30 (1.98)	0.399
Proportional attributions Internal	0.55 (0.09)	0.50 (0.17)	0.242
Personal	0.94 (0.08)	0.76 (0.17)	0.000
Controllable	0.86 (0.12)	0.81 (0.10)	0.159
Stable	0.79 (0.20)	0.65 (0.21)	0.032
"Blaming"	0.46 (0.14)	0.33 (0.18)	0.013
Parenting and child behavior	44.17 (22.69)	85.2 (13.32)	0.000
Behavioral self-efficacy (PTC)			
Setting self-efficacy (PTC)	45.52 (25.04)	85.3 (13.76)	0.000
Parental laxness (PS)	4.50 (1.63)	2.35 (0.77)	0.005
Parental reactivity (PS)	3.63 (1.41)	2.54 (0.70)	0.005
Parental verbosity (PS)	4.56 (1.02)	3.91 (0.54)	0.017
PAFAS consistency	8.70 (2.70)	5.10 (2.44)	0.000
PAFAS coercion	7.65 (3.82)	5.90 (3.70)	0.149
PAFAS positive encouragement	5.55 (3.35)	3.80 (3.27)	0.103
PAFAS parent–child relationship	8.35 (2.51)	4.75 (2.81)	0.025
ECBI Intensity	164.55 (44.56)	106.70 (25.04)	0.000
ECBI Problem	21.15 (10.02)	8.35 (8.13)	0.000

**t-test performed on log transformed variables.*

step and SMI status (schizophrenia vs. no SMI) was entered in the second. We found that SMI status was not a significant predictor of the two EE variables at the 5% significance level adopted for the study, despite large R^2 values (criticism: R^2 change = 0.09, $\beta = 0.417$, $p = 0.056$; hostility: R^2 change = 0.07, $\beta = 0.373$, and $p = 0.096$). SMI status predicted personal attributions (R^2 change = 0.09, $\beta = 0.433$, $p = 0.033$) but not stable (R^2 change = 0.03, $\beta = 0.239$, and $p = 0.286$) or blaming attributions (R^2 change = 0.05, $\beta = 0.320$, and $p = 0.147$).

Parenting and Child Behavior

Table 2 highlights significant differences between the groups with regard to parenting and parental reports of child behavior. Parents with schizophrenia had poorer parenting self-efficacy and were more likely to use harsh (over-reactive), permissive (lax), and overly wordy (verbose) discipline strategies according to their responses to the parenting scale. Parents with schizophrenia were less consistent in their parenting than those without SMI and reported a poorer parent–child relationship. The use of coercion and positive encouragement was not significantly different between groups. Parents with schizophrenia reported significantly more behavior problems in their children (ECBI intensity) and found their children's behavior to be more problematic (ECBI problem).

Relationship Between EE and Attributions

Correlational analyses (Pearson's r) were used to explore hypothesized relationships between EE and attribution variables. Parents with schizophrenia who perceived their children's behavior to be outside of the child's control were warmer about them ($r = -0.47$, $p < 0.05$), and less emotionally over-involved ($r = -0.69$, $p < 0.001$). Parents with schizophrenia who had a tendency to blame the child for the child's negative behaviors were more hostile ($r = 0.47$, $p < 0.05$) and less warm ($r = -0.60$, $p < 0.01$) toward them. However, contrary to our predictions, these parents were not more critical of the child. There were no significant relationships between EE and attributions in the parents without SMI.

TABLE 4 | Correlations between EE, mental health, parenting and child behavior in parents with schizophrenia.

	Criticism	Hostility	EOI	Warmth	Positive Remarks
DASS depression	0.22	0.31	−0.42	−0.71**	−0.62**
DASS anxiety	0.25	0.11	−0.42	−0.38	−0.48*
DASS stress	0.25	0.31	−0.41	−0.63**	−0.62**
WEMWBS total	−0.19	−0.12	0.12	0.67**	0.47*
PANSS positive symptoms	0.23	0.04	−0.20	−0.42	−0.36
PANSS negative symptoms	0.14	0.09	−0.22	−0.53*	−0.24
PANSS general symptoms	0.11	0.01	−0.11	−0.47*	−0.24
PSYRATS hallucinations	0.05	0.04	−0.07	−0.39	−0.33
PSYRATS delusions	0.42	0.26	0.16	−0.60**	−0.43
Setting parental self-efficacy (PTC)	−0.43*	−0.41	0.40	0.74**	0.57**
Behavioral parental self-efficacy (PTC)	−0.23	−0.28	0.24	0.61**	0.52*
Parental laxness (PS)	−0.14	0.09	−0.08	−0.40	−0.06
Parental reactivity (PS)	0.10	0.45*	−0.34	−0.46*	−0.02
Parental verbosity (PS)	−0.01	0.11	0.18	0.05	0.18
PAFAS consistency	−0.12	−0.08	0.15	−0.26	−0.33
PAFAS coercion	−0.01	−0.08	0.16	−0.33	0.09
PAFAS positive encouragement	−0.25	−0.06	0.23	0.13	0.34
PAFAS parent–child relationship	0.18	0.31	−0.35	−0.58**	−0.29
ECBI Intensity	0.49*	0.39	−0.34	−0.62**	−0.64**
ECBI Problem	0.39	0.44	−0.58**	−0.78**	−0.34

$^{*}p < 0.05$; $^{**}p < 0.01$.

DASS, Depression, Anxiety and Stress Short Form Scale; WEMWBS, Warwick Edinburgh Mental Wellbeing Scale; PANSS, Positive and Negative Syndrome Scale; PSYRATS, Psychotic Symptoms Rating Scales; PTC, Parenting Task Checklist; PS, Parenting Scales; PAFAS, Parenting and Family Adjustment Scales; ECBI, Eyberg Child Behavior Inventory.

Relationship of Parental Mental Health to EE and Attributions in Parents With Schizophrenia: Exploratory Analyses

The majority of significant relationships between mental health and EE centered on warmth (see **Table 4**). Higher levels of depression, stress, negative and general symptoms and delusions were each related to decreased warmth. Accordingly, warmth increased as parental subjective wellbeing increased. Higher levels of depression, stress, and anxiety were also related to fewer positive remarks being made. No associations were found between criticism, EOI, hostility and mental health, and well-being. Attribution scores, including blaming attributions, were not related to any mental health variables.

Relationship of Parenting Practices and Child Behavior to EE and Attributions in Parents With Schizophrenia: Exploratory Analyses

Facets of EE were associated with several aspects of parenting (see **Table 4**). Critical comments were related to setting specific self-efficacy and ECBI intensity scores. Hostility was related to the use of harsh (reactive) discipline practices and a t-test confirmed that parents rated as hostile were more reactive than those who were not [means = 4.60 (1.49) and 3.21 (1.19), respectively, $t_{(18)} = 2.23$, $p < 0.05$]. Emotional over-involvement was related to ECBI problem scores but not to self-efficacy or parenting.

As is clear from **Table 4**, warmth was once again the facet of EE with the greatest number of significant associations. Lack of warmth was related to reduced parenting self-efficacy and the use of harsher (over reactive) parenting practices. Higher levels of warmth were related to a better parent–child relationship and better child behavior. A similar pattern of results was observed for positive remarks: Increased frequency of positive remarks was associated with reduced efficacy and lower ECBI intensity scores.

There were markedly fewer relationships between proportional attributions and parenting and child behavior. Internal attributions were not related to any parenting or child behavior measures. A tendency to see problems as personal to the child was linked to decreased parental behavioral self-efficacy ($r = -0.54$, $p < 0.05$); problematic behavior ($r = 0.46$, $p < 0.05$) and a poorer parent–child relationship ($r = 0.46$, $p < 0.05$). The perception that the child could control their behavior was also associated with more problematic behaviors ($r = 0.47$, $p < 0.05$) and a poorer parent–child relationship ($r = 0.57$, $p < 0.01$). A tendency to see the causes of behaviors as stable (and likely to recur) was linked to less consistent parenting ($r = 0.45$, $p < 0.05$). Finally, blaming attributions reflected a poorer parent–child relationship ($r = 0.62$, $p < 0.01$) but were not related to specific parenting behaviors otherwise.

DISCUSSION

This is the first study to examine EE and attributions in parents with schizophrenia, or indeed with any SMI. It confirms that parents with schizophrenia, like other parents, seek to explain their children's behavior and spontaneously make attributions about behaviors they perceive to be negative (24). Furthermore, they do so at a higher rate than parents without SMI. In line with our hypotheses, parents with schizophrenia differed from those without SMI in terms of both expressed emotion and attributions. The finding that parents with schizophrenia were more critical and hostile about their children is an important one, since parental EE has been linked to the development and maintenance of a range of childhood disorders (25) and to mental health difficulties and substance misuse problems in adulthood (28). Our hypothesis that parents with schizophrenia would be less warm than their counterparts without SMI was not supported. This was surprising given the numerous studies reporting poorer parent-infant interactions in mothers with schizophrenia which have highlighted a lack of warmth, sensitivity and responsiveness [e.g., (49–51)]. We did however find that poorer parental mental health was linked to decreased warmth: higher levels of depression, stress and more severe negative and general symptoms and delusions were all associated with lower levels of warmth. It may therefore be the case that the lack of a significant difference between the two groups of parents may in fact reflect symptom variability within the parents with schizophrenia.

Parents with schizophrenia were more likely to attribute their children's behaviors to causes that were more stable and personal to the child and in line with our prediction, they were more likely than the parents without SMI to attribute responsibility for the behavior to the child. This tendency to make more blaming attributions was linked to hostility, which in turn was linked to parenting practices: hostile parents used harsher discipline practices than those who were not hostile. These findings provide support to studies reporting that when parents believe their child's behavior is intentional and unique to the child, they tend to use more coercive and harsh parenting practices (52–54).

Increased blame was also related to a lack of warmth, which in turn was related to reduced parenting self-efficacy and the use of harsher (over reactive) parenting practices. Blame has previously been found to reflect higher levels of parental distress in relation to child behavior (55). Although, we did not assess parents' anger or distress in relation to the child it is indeed likely that this is the driver of a harsher style of parenting. If parents believe that the child is responsible for their behavior, they also believe that the child is capable of modifying it, and parents may therefore engage in more negative feedback (56). The finding that parents with schizophrenia tended to attribute children's behavior to causes that were more stable and personal may additionally indicate a lack of hope for improvements in behavior, further adding to parental distress. It is conceivable that for the parents with the worst mental health, who evidenced the lowest levels of parenting self-efficacy, the tendency to blame children and be less warm toward them may constitute attempts to preserve parental self-esteem and well-being. Future research should aim to further explore the relationship between parental mental health and parental beliefs about children's behavior to elucidate this further.

Owing to the socioeconomic differences between the two groups we considered the possibility that the observed differences in EE and attributions may instead reflect the social isolation and financial stress caused by unemployment and single parenthood. Our regression analyses revealed that this may well be the case. SMI status was not found to independently predict EE although a large effect was observed. This likely reflects the small sample size. A larger sample, with a more closely matched control group would be needed to test this properly. It is likely that the social adversity experienced by parents with schizophrenia is a key feature of the family environment and highly likely to contribute to higher levels of EE toward their children compared to families without SMI who are less likely to be experiencing these stressors. Research shows that families characterized by instability and lacking access to financial resources and social support are families in distress, and this is a key factor in the development of high EE (57).

It must be noted that several unmeasured variables may also have contributed to the observed differences in EE and attributions between the groups, such as comorbid parental and child physical and mental illness.

Despite the study limitations, it is possible to conclude that EE might explain some of the intergenerational risk in families with a parent with schizophrenia. This study highlights opportunities for the development of preventative and early intervention strategies, beyond that of standard family intervention when working with parents with schizophrenia. Parental attributions and EE may offer insight into parenting practices and highlight potential targets for intervention strategies to benefit both parental mental health and longer term outcomes for children.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The study was reviewed and approved by Greater Manchester West National Research Ethics Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

LG: conceptualization, designing the study, data analysis, writing, reviewing, and editing the manuscript. RC and RD: conceptualization, designing the study, and supervision. LW: conceptualization, designing the study, data collection, and writing the manuscript. All authors contributed to the article and approved the submitted version.

FUNDING

LW was supported by a President's Doctoral Scholar award from The University of Manchester.

ACKNOWLEDGMENTS

We gratefully acknowledge the commitment and support of each parent who took part in the study and the staff who referred them. We additionally thank Angharad Polley for her contribution to recruitment and the extraction and rating of attributions.

REFERENCES

1. GBD. Disease and injury incidence and prevalence collaborators. Global, regional, national incidence. prevalence, and years lived with disability for 328 diseases and injuries for 195 countries, 1990-2016: a systematic analysis for the Global Burden of Disease Study 2016. *Lancet.* (2016) 390:1211–59. doi: 10.1016/S0140-6736(18)32279-7
2. Mowbray CT, Bybee D, Oyserman D, MacFarlane P, Bowersox N. Psychosocial outcomes for adult children of parents with severe mental illnesses. Demographic and clinical history predictors. *Health Soc Work.* (2006) 31:99–108. doi: 10.1093/hsw/31.2.99
3. Rasic D, Hajek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder and major depressive disorder: a meta-analysis of family high-risk studies. *Schizophr Bull.* (2014) 40:28–38. doi: 10.1093/schbul/sbt114
4. Tsuang M, Stone W, Faraone S. Genes, environment and schizophrenia. *Br J Psychiatry.* (2001) 178:S18–S24. doi: 10.1192/bjp.178.40.s18
5. Goddard HW, Miller BC. Adding attribution to parenting programs. *Fam Soc J Contemp Soc Serv.* (1993) 74:84–92. doi: 10.1177/104438949307400203
6. Lovejoy MC, Graczyk PA, O'Hare E, Neuman G. Maternal depression and parenting behavior. *Clin Psychol Rev.* (2000) 20:561–92. doi: 10.1016/S0272-7358(98)00100-7
7. Wynne LC, Tienari P, Sorri A, Lahti I, Moring J, Wahlberg K-E, et al. II. Genotype-environment interaction in the schizophrenia spectrum: qualitative observations. *Fam Process.* (2006) 45:435–47. doi: 10.1111/j.1545-5300.2006.00181.x
8. Barlow J, Coren E. The effectiveness of parenting programs. *Res Soc Work Pract.* (2018) 28:99–102. doi: 10.1177/1049731517725184
9. Deater-Deckard K, Chen N, Wang Z, Bell MA. Socioeconomic risk moderates the link between household chaos and maternal executive function. *J Fam Psychol.* (2012) 26:391–9. doi: 10.1037/a0028331
10. Marsh S, Dobson R, Maddison R. The relationship between household chaos and child, parent, and family outcomes: a systematic scoping review. *BMC Public Health.* (2020) 20:513. doi: 10.1186/s12889-020-08587-8
11. Campbell L, Hanlon MC, Poon AW, Paolini S, Stone M, Galletly C, et al. The experiences of Australian parents with psychosis: the second Australian National Survey of Psychosis. *Austr N Zeal J Psychiatry.* (2012) 46:890–900. doi: 10.1177/0004867412455108
12. Strand S, Boström P, Grip K. Parents' descriptions of how their psychosis affects parenting. *J Child Fam Stud.* (2020) 29:620–31. doi: 10.1007/s10826-019-01605-3
13. Park JM, Solomon P, Mandell DS. Involvement in the child welfare system among mothers with serious mental illness. *Psychiatr Serv.* (2006) 57:493–7. doi: 10.1176/ps.2006.57.4.493
14. Reupert A, Gladstone B, Helena Hine R, Yates S, McGaw V, Charles G, et al. Stigma in relation to families living with parental mental illness: an integrative review. *Int J Ment Health Nurs.* (2021) 30:6–26. doi: 10.1111/inm.12820
15. Seeman MV. Intervention to prevent child custody loss in mothers with schizophrenia. *Schizophr Res Treat.* (2012) 2012:796763. doi: 10.1155/2012/796763
16. Powell RM, Parish SL, Mitra M, Nicholson J. Responding to the legal needs of parents with psychiatric disabilities: insights from parent interviews. *Law Inequal.* (2020) 38:69–114.
17. Vaughn C, Leff J. The measurement of expressed emotion in the families of psychiatric patients. *Br J Soc Clin Psychol.* (1976) 15:157–65. doi: 10.1111/j.2044-8260.1976.tb00021.x
18. Butzlaff RL, Hooley JM. Expressed emotion and psychiatric relapse. *Arch Gen Psychiatry.* (1998) 55:547–52. doi: 10.1001/archpsyc.55.6.547
19. Lee G, Barrowclough C, Lobban F. Positive affect in the family environment protects against relapse in first-episode psychosis. *Soc Psychiatry Psychiatr Epidemiol.* (2013) 49:367–76. doi: 10.1007/s00127-013-0768-x
20. Butler R, Berry K, Varese F, Bucci S. Are family warmth and positive remarks related to outcomes in psychosis? A systematic review. *Psychol Med.* (2019) 49:1250–65. doi: 10.1017/S0033291718003768
21. Izon E, Berry K, Law H, French P. Expressed emotion (EE) in families of individuals at-risk of developing psychosis: a systematic review. *Psychiatry Res.* (2018) 270:661–72. doi: 10.1016/j.psychres.2018.10.065
22. Rogosch F, Cicchetti D, Toth S. Expressed emotion in multiple subsystems of the families of toddlers with depressed mothers. *Dev Psychopathol.* (2004) 16:689–709. doi: 10.1017/S0954579404004730
23. Bolton C, Calam R, Barrowclough C, Peters S, Roberts J, Wearden A, et al. Expressed emotion, attributions and depression in mothers of children with problem behaviour. *J Child Psychol Psychiatry.* (2003) 44:242–54. doi: 10.1111/1469-7610.00117
24. White C, Barrowclough C. Depressed and non-depressed mothers with problematic preschoolers: attributions for child behaviours. *Br J Clin Psychol.* (1998) 37:385–98. doi: 10.1111/j.2044-8260.1998.tb01396.x
25. Peris T, Miklowitz D. Parental expressed emotion and youth psychopathology: new directions for an old construct. *Child Psychiatry Hum Dev.* (2015) 46:863–73. doi: 10.1007/s10578-014-0526-7
26. Baker JK, Smith LE, Greenberg JS, Seltzer MM, Taylor JL. Change in maternal criticism and behavior problems in adolescents and adults with autism across a seven-year period. *J Abnorm Psychol.* (2011) 120:465–75. doi: 10.1037/a0021900
27. Romero-Gonzalez M, Chandler S, Simonoff E. The relationship of parental expressed emotion to co-occurring psychopathology in individuals with autism spectrum disorder: a systematic review. *Res Dev Disabil.* (2018) 72:152–65. doi: 10.1016/j.ridd.2017.10.022
28. Schwartz OS, Byrne ML, Simmons JG, Whittle S, Dudgeon P, Yap MBH, et al. Parenting during early adolescence and adolescent-onset major depression: a 6-year prospective longitudinal study. *Clin Psychol Sci.* (2013) 2:272–86. doi: 10.1177/2167702613505531
29. Wilde A, Chan H-N, Rahman B, Meiser B, Mitchell PB, Schofield PR, et al. A meta-analysis of the risk of major affective disorder in relatives of individuals affected by major depressive disorder or bipolar disorder. *J Affect Disord.* (2014) 158:37–47. doi: 10.1016/j.jad.2014.01.014
30. Chambless DL, Bryan AD, Aiken LS, Steketee G, Hooley JM. The structure of expressed emotion: a three-construct representation. *Psychol Assess.* (1999) 11:67–76. doi: 10.1037/1040-3590.11.1.67
31. Barrowclough C, Hooley JM. Attributions and expressed emotion: a review. *Clin Psychol Rev.* (2003) 23:849–80. doi: 10.1016/S0272-7358(03)00075-8
32. Munton A, Silvester J, Stratton P, Hanks H. *Attributions in Action; a Practical Approach to Coding Qualitative Data*. London: Wiley (1999).
33. Weiner B. An attributional theory of achievement motivation and emotion. *Psychol Rev.* (1985) 92:548–73. doi: 10.1037/0033-295X.92.4.548
34. Johnston C, Ohan J. The importance of parental attributions in families of children with attention-deficit/hyperactivity and disruptive behaviour disorders. *Clin Child Fam Psychol Rev.* (2005) 8:167–82. doi: 10.1007/s10567-005-6663-6
35. Sheeber LB, Johnston C, Chen M, Leve C, Hops H, Davis B. Mothers' and fathers' attributions for adolescent behavior: an examination in families of depressed, subdiagnostic, nondepressed youth. *J Fam Psychol.* (2009) 23:871–81. doi: 10.1037/a0016758
36. Sellers R, Harold G, Elam K, Rhoades KA, Poter R, Mars B, et al. Maternal depression and co-occurring antisocial behaviour: testing maternal hostility and warmth as mediators of risk for offspring psychopathology. *J Child Psychol Psychiatry.* (2014) 55:112–20. doi: 10.1111/jcpp.12111
37. Sawrikar V, Dadds M. What role for parental attributions in parenting interventions for child conduct problems? Advances from research into practice. *Clin Child Fam Psychol Rev.* (2018) 21:41–56. doi: 10.1007/s10567-017-0243-4
38. Kay SR, Fiszbein A, Opler LA. The positive and negative syndrome scale (PANSS) for schizophrenia. *Schizophr Bull.* (1987) 13:261–76. doi: 10.1093/schbul/13.2.261
39. Haddock G, McCarron J, Tarrier N, Faragher EB. Scales to measure dimensions of hallucinations and delusions: the psychotic symptom rating scales (PSYRATS). *Psychol Med.* (1999) 29:879–89. doi: 10.1017/S0033291799008661
40. Stewart-Brown S, Tennant A, Tennant R, Platt S, Parkinson J, Weich S. Internal construct validity of the Warwick-Edinburgh Mental Well-being Scale (WEMWBS): a rasch analysis using data from the Scottish health education population survey. *Health Qual Life Outcomes.* (2009) 7:15. doi: 10.1186/1477-7525-7-15

41. Lovibond SH, Lovibond PF. *Manual for the Depression Anxiety Stress Scales*. Sydney: Psychology Foundation Monograph (1995).
42. Sanders MR, Woolley ML. The relationship between maternal self-efficacy and parenting practices: implications for parent training. *Child Care Health Dev.* (2005) 31:65–73. doi: 10.1111/j.1365-2214.2005.00487.x
43. Sanders MR, Morawska A, Haslam DM, Filus A, Fletcher R. Parenting and family adjustment scales (PAFAS): validation of a brief parent-report measure for use in assessment of parenting skills and family relationships. *Child Psychiatry Hum Dev.* (2014) 45:255–72. doi: 10.1007/s10578-013-0397-3
44. Arnold DS, O'Leary SG, Wolff LS, Acker MM. The Parenting Scale: a measure of dysfunctional parenting in discipline situations. *Psychol Assess.* (1993) 5:137–44. doi: 10.1037/1040-3590.5.2.137
45. Eyberg S, Pincus D. *Eyberg Child Behavior Inventory TM (ECBI TM) Sutter-Eyberg Student Behavior Inventory-Revised TM (SESBI-R TM)*. Professional Manual. Odessa, FL: Psychological Assessment Resources (1999).
46. Peters S, Calam R, Harrington R. Maternal attributions and expressed emotion as predictors of attendance at parent management training. *J Child Psychol Psychiatry.* (2005) 46:436–48. doi: 10.1111/j.1469-7610.2004.00365.x
47. Stratton P, Munton T, Hanks H, Heard D, Davidson C. *Leeds Attributional Coding System*. Leeds: Leeds Family Therapy and Research Centre, University of Leeds, Leeds LS2 9JT, England (1988).
48. Bender R, Lange S. Multiple test procedures other than Bonferroni's deserve wider use. *Br Med J.* (1999) 318:600. doi: 10.1136/bmj.318.7183.600a
49. Riordan D, Appleby L, Faragher B. Mother-infant interaction in post-partum women with schizophrenia and affective disorders. *Psychol Med.* (1999) 29:991–5. doi: 10.1017/S0033291798007727
50. Wan M, Salmon MP, Riordan DM, Appleby L, Webb R, Abel KM. What predicts poor mother-infant interaction in schizophrenia? *Psychol Med.* (2007) 37:537–46. doi: 10.1017/S0033291706009172
51. Healy SJ, Lewin J, Butler S, Vaillancourt K, Seth-Smith F. Affect recognition and the quality of mother-infant interaction: understanding parenting difficulties in mothers with schizophrenia. *Arch Womens Mental Health.* (2016) 19:113–24. doi: 10.1007/s00737-015-0530-3
52. Johnston C, Fine S, Weiss M, Weiss J, Weiss G, Freeman WS. Effects of stimulant medication treatment on mothers' and children's attributions for the behavior of children with attention deficit hyperactivity disorder. *J Abnorm Child Psychol.* (2000) 28:371–82. doi: 10.1023/A:1005121110278
53. Park S, Kim B-N, Park M-H. The relationship between parenting attitudes, negative cognition, and the depressive symptoms according to gender in Korean adolescents. *Int J Ment Health Syst.* (2016) 10:35. doi: 10.1186/s13033-016-0069-3
54. Sander JB, McCarty CA. Youth depression in the family context: familial risk factors and models of treatment. *Clin Child Fam Psychol Rev.* (2005) 8:203–19. doi: 10.1007/s10567-005-6666-3
55. Smith A, O'Leary S. Attributions and arousal as predictors of maternal discipline. *Cogn Ther Res.* (1995) 19:459–71. doi: 10.1007/BF02230412
56. Hooley JM, Campbell C. Control and controllability: beliefs and behaviour in high and low expressed emotion relatives. *Psychol Med.* (2002) 32:1091–9. doi: 10.1017/S0033291702005779
57. Boger K. Parental expressed emotion toward children: prediction from early family functioning. *J Fam Psychol.* (2008) 22:784–8. doi: 10.1037/a0013251

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Psychiatry

ORIGINAL RESEARCH
published: 07 January 2022
doi: 10.3389/fpsyt.2021.784022

Do the Child Welfare and Protection Services Involve Children in Cases With Parental Mental Health Problems? A Norwegian Case-File Study

Svein Arild Vis[1]*, *Camilla Lauritzen*[1], *Øivin Christiansen*[2] *and Charlotte Reedtz*[1]

[1] *Regional Centre for Child and Youth Mental Health and Child Welfare, UiT-The Arctic University of Norway, Tromsø, Norway,* [2] *Regional Centre for Child and Youth Mental Health and Child Welfare, Norwegian Research Centre (NORCE), Bergen, Norway*

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Mary V. Seeman,
University of Toronto, Canada
Silvia Krumm,
Universitaetsklinikum Ulm, Germany

***Correspondence:**
Svein Arild Vis
svein.arild.vis@uit.no

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *27 September 2021*
Accepted: *15 December 2021*
Published: *07 January 2022*

Citation:
Vis SA, Lauritzen C, Christiansen Ø and Reedtz C (2022) Do the Child Welfare and Protection Services Involve Children in Cases With Parental Mental Health Problems? A Norwegian Case-File Study.
Front. Psychiatry 12:784022.
doi: 10.3389/fpsyt.2021.784022

Background: Parental mental health problems is a common source of concern reported to child welfare and protection services (CWPS). In this study we explored to what extent the child was invited to participate in the investigation process. We aimed to study: (a) what was the current practice in the child protection service in Norway when the CWPS received a report of concern about children whose parents were affected by mental health problems or substance abuse, (b) to what extent were children involved and consulted, (c) which factors predicted the decision to involve the children, and (d) in cases in which conversations with children were conducted: what was the main content of the conversations.

Method: The study was a cross-sectional case file study ($N = 1{,}123$). Data were collected retrospectively from case records in 16 different child protection agencies. The cases were randomly drawn from all referrals registered in the participating agencies. Differences in how investigations were conducted in cases with and without concerns about parental mental health were analyzed using *t*-tests and chi-square testes. Predictors of child involvement in cases with parental mental health problems ($N = 324$) were estimated by logistic regression analyses.

Results: When the referral to the CWPS contained concerns about parental mental health, there were more consultations with parents, more frequent home visits and the investigation took longer to conclude. The children, however, were less likely to be involved. Children in such cases were consulted in 47.5% of cases. Predictors for involving the children in those cases were child age, concern about the child's emotional problems and if the child was known from previous referrals.

Conclusion: In Norwegian child protection investigations, in which there were concerns about the parent's mental health, conversations with children were conducted to a significantly lower degree compared to cases where the child's problem was the main concern. In such cases, the CWPS workers have to overcome a threshold before they consult with the child. The threshold decreases with child age and when case worker already knows the child.

Keywords: parental mental illness, child involvement, child participation, child welfare and protection, concerns, COPMI

INTRODUCTION

Given the adverse effects of parental mental illness, there is a strong rationale for public health and preventive approaches across services, to safeguard and support the children (1, 2). The risk factors for children of parents with a mental illness (COPMI) have been thoroughly documented in studies across the world (3). In Norway it is mandatory for health care workers who treat parents with mental illness to report concerns to the child welfare and protection services (CWPS) if there is reason to believe that the child is at risk. The Norwegian Health Personnel Act further specifies that health care personnel must consult with patients who are parents, about the children's need for information or support and to provide information, guidance and direct them to relevant interventions for the family (4). Likewise, the CWPS are mandated to involve children in cases concerning their welfare and safety in accordance with age and maturity. The children's right to participate in the CWPS is established by law (The Child Welfare Act, § 1–6). These mandates are the results of increased awareness within social services and the health professional community about the potential risk for children of parents with a mental illness. Consequently, child participation is increasingly seen not only as a legal requirement in case processing but also as a mean to ensure child safety and to improve quality and effectiveness of health care and social services (5).

Despite numerous professional, political, and legislative efforts to strengthen children's participation in health and social practice, there is substantial documentation showing that child involvement is a challenge to practitioners within adult mental health care and the CWPS alike. A five-year follow-up study of identification and support for children of mentally ill parents (6) showed that even though there have been substantial efforts to change practice within adult mental health services in the past decade, children did not receive necessary support from health personnel who were treating their parents.

Intervening early and targeting adverse influences on children and parents may improve outcomes for children (7). Child involvement and child participation is a key ingredient in early intervention. Additionally, psychoeducation is a common component across programs for parents with mental illnesses and their children (8). In the context of parental mental illness, psychoeducation is seen as a tool to reduce feelings of guilt and shame from materializing in the children and their parents. A lack of openness about mental illness is also thought to restrain children from venting emotions such as anger, despair and insecurities about their own life situation and that of their parents. Subsequently, when there is a mental illness in the family, children need accurate mental health information (9, 10). Not receiving information and support may severely affect the lives of these children. Faugli et al. found that children who sought information were often ignored by the health personnel (11).

There is substantial documentation showing that establishing a dialogue with children is a challenge to many adult helpers. Many of the barriers to child involvement seem to be the same across service settings, such as the professional's attitudes and skills (12).

Child welfare and protection workers strive to balance children's right to participate on the one hand, and the right to protection on the other hand. This is especially the case when the case concerns adult's problems, such as parent conflicts, mental health issues, and substance abuse. The workers are worried that they will expose children to such problems because it may be a burden or even harmful to them, which may be avoided by not involving them (13, 14). Age may be another important factor. A study carried out among Norwegian CWPS workers found that the most experienced workers were also the most reluctant to let children participate in child protection processing (15). Other significant explanations for the reluctance to involve children are social workers' and health personnel's lack of professional confidence, skills and tools (14, 16, 17). Previous studies have pointed out that the adult mental health services regarded their competence and knowledge about support for the children of their patients as limited, and that they considered the CWPS to be a more suited service to provide for the children's needs (18). Furthermore, the results showed that adult mental health workers lacked skills in how to approach the family, how to develop trust and confidence, and how to discuss negative consequences of the parental mental illness for the children. Additionally, many reported that they lacked the competence to assess the needs children may have and explained this by their educational background not being child specific (19). On the other hand, little is known about how the CWPS addresses cases of parental mental health problems. We therefore do not know if the CWPS involve the children of parents where there is a reported concern about mental health issues. Studying the CWPS approach to these children may inform us about important issues to be aware of in the overall approach to support COPMI.

AIMS

The main objective of the current study was to explore the child welfare and protection services' approaches to families affected by parental mental illness. Admittedly, child welfare legislation does differ between countries, and some aspects of professional practice may be specific to certain contexts. However, as illustrated by the introductory review there are also aspects of professional practice that is rooted in conceptions about children's abilities and vulnerabilities which transcends borders and traditions. We therefore believe that studying if families where there are concerns about parental mental health are approached differently than families with other types of concerns, is important. The aims of the current study were therefore: (a) to identify who the CWPS in Norway consulted when they received a report of concern about children whose parents are affected by mental health problems, (b) to what extent children were involved and consulted, (c) which factors predicted the decision to involve the children, and (d) in cases where conversations with children were conducted: what was the main content of the conversations.

METHOD

The study is part of a large national research project that was initiated in 2017. The project was approved by the Council for Duty of Confidentiality and the Norwegian Center for Research Data. The researchers were given access to social work records by a decision from the Directorate for Children, and Family Affairs in Norway. This decision allowed the researchers to extract data from case files without seeking informed consent. A license for handling and storage of data were granted by the Norwegian Data Protection Authority on the 29.06.2017 (reference number: 7/00411-2/CDG).

Design and Procedures

The study was designed as a case file study which was carried out retrospectively. A total of 1,365 child welfare and protection cases were randomly drawn from all referrals registered in 16 participating agencies in the period of January 2015 to December 2017. The number of cases from each agency varied between 50 and 150 depending on the size of the agency. The reason why we sampled agencies by size is that we wanted the number of cases drawn from each agency to be about the same proportion of the total available sample from that agency. Data were collected and coded from case records. The researchers were given access to the casefiles and to electronic systems for recordkeeping by the CPS agency. All case files were coded on site at the agency by the use of an electronic web-based data entry form that was developed specifically for this purpose. The data entry form was developed and tested for interrater reliability by independent coding of 20 cases by two researchers. The results showed an average interrater agreement of 86.9%. A total of 13 variables had low reliability (<80% interrater agreement). Three of those were eliminated from the form because it was concluded that reliable information could not be obtained. The remaining 10 variables were reformulated, and the coding manual was revised with better explanation of codes. After this revision the reliability of the instrument was re-tested by independent coding of 42 cases by two researchers. At this second step, interrater agreement was 90.8%. In health research, an interrater agreement over 80% generally are considered acceptable (20). The variables and the codes from the form is available from the corresponding author upon request.

Participants

For the current analyses we included all the cases that were screened in for a child protection investigation ($N = 1{,}123$). Fifty-three percentage of the sample were boys and the mean age was 8.9 years ($SD = 5.1$). In a total of 41.6 % of the referrals, the family had immigrant background. Immigrant background was defined as the child or one of the parents being born in a country other than Norway.

Measures

Referrals to CWPS in Norway is most usually a free text letter submitted by a concerned third party. We coded the concerns in the referral letter as present or absent because this is all that safely can be concluded with high level of reliability. The following types of concerns was coded as present or absent in the referral (i) parental mental health problems or substance abuse problems (ii) child developmental problems, (iii) child externalizing behavior problems (iv) child emotional problems. The main characteristics of the investigation process was registered. This included counting (i) the duration of the investigation measured in number of days before the investigation was concluded, (ii) number of meetings between CWPS and parents, (iii) number of home visitations by the CWPS and (iv) if additional information had been requested from health care services, school, police, social services or other CWPS agencies. Whether or not there had been a consultation with the child as part of the investigation was registered. In instances in which such a consultation had taken place ($N = 680$) the main content of the consultation was coded into seven different pre-determined content Those were (i) exploratory conversation about conditions at home, (ii) information sharing, (iii) conversation to obtain child's opinions, (iv) investigative conversation about episode in the family, (v) supportive conversation, (vi) general conversation without specific aim, (vii) no information about the content. These categories were developed by the researchers based upon the theory of general procedures for the participation of children (21).

Statistical Analyses

Bi-variate differences in the main characteristics of the investigation process, with and without concerns about parental mental health problems were analyzed using t-tests for continuous variables and chi-square testes for categorical variables. Predictors for child consultations in cases with parental mental health problems ($N = 324$) were examined in multivariate analysis using binary logistic regression. In the regression analysis all predictors were entered at together.

RESULTS

Our first aim was to identify who CWPS in Norway consulted in the investigation when they received a report of concern about children whose parents are affected by mental health problems. When receiving a report of concern, the CWPS investigations most commonly consisted of consultations with the child, consultation with a parent, and home visits. In addition, the CWPS obtained information from other services such as health care, school/kindergarten, police, and other social services. We investigated if there were differences in how CWPS investigations were conducted in cases reported with parental mental health issues compared to cases in which such problems were not reported. The results showed that when the report to the CWPS contained concerns about parental mental health there were more consultations with parents, more frequent home visits and the investigation took longer to conclude. The children however were less likely to be consulted. On average children were consulted in 47.5% of those cases (**Table 1**).

We also investigated which other child and case characteristics that may explain whether children were consulted in cases with referrals of suspected parental mental health problems. In the multivariable analysis we identified three statistically significant predictors. The first was child age. For a 5-year difference in child age, the odds ratio for a child consultation were 3.18. This means that a 12-year-old child were more about three times more likely to be consulted than a 7-year-old child. The second predictor was if a concern about the child's emotional problems had been raised in the report. Then the child was about 2.8 times more likely to be consulted. The third predictor was if the child was known by the agency from previous reports, then the chance of a child consultation was increased by a magnitude of about 1.3 for each previous report (**Table 2**).

A final aim was to study the cases where children were consulted and identify what the content of conversations with children was. The most common form of conversation in our sample was exploratory conversations about conditions in the home (72.6%). In such conversations, the child was encouraged to talk about how it is at home without it being related to episodes or specific events. In this category, there may also be exploratory conversations about the child's everyday life, for example how the child is doing at school.

Furthermore, 36.9% of the child conversations took the form of an informative conversation. An informative conversation is characterized by the child receiving information about the case and/or what will happen in the future. In 31.5% of the cases in our sample, the focus in the children's conversation was on obtaining the child's point of view or opinions. This was more frequently the topic in cases with suspected parental mental health problems. More frequent in those types of cases were also consultations which had a supportive rather than an investigatory purpose (**Table 3**).

DISCUSSION

The results showed that when the CWPS initiated an investigation based on a report of concern about parental mental health, the investigation was significantly more extensive in time and efforts compared to investigations of other types of concerns. In particular, the CWPS workers carried out more consultations with parents and more frequently made home visits. A reasonable interpretation of these findings is that such reports were considered more serious by the CWPS and hence that they conduct more thorough investigations.

TABLE 2 | Predictors for child consultation in CWPS cases with parental mental health problems and/or parental substance abuse problems ($N = 324$).

	B	OR (95% CI)
Number of previous referrals	0.23	1.26 (1.07–1.48)**
Child sex = male	0.38	1.46 (0.86–2.47)
Child age	0.23	1.25 (1.18–1.33)***
Child immigrant background = no	0.03	1.03 (0.56–1.87)
Concern about child developmental problem = no	−0.06	0.94 (0.24–3.78)
Concern about child externalizing problems = no	−0.01	1.0 (0.42–2.36)
Concern about child emotional problem = no	1.02	2.76 (1.004–7.58)*

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.*

TABLE 1 | CWPS investigations in cases referred for parental mental health problems and/or parental substance abuse vs. other problems ($N = 1{,}059$–$1{,}123$).

Investigation activity	Mental health/substance abuse problem in family *N* (%)	Other problem *N* (%)	χ^2 (df)
Consultations with the child = yes	154 (47.5%)	526 (65.8%)	32.3 (1)***
Information from health care = yes	232 (71.6%)	485 (60.7%)	11.9 (1)**
Information from school/child care = yes	177 (54.6%)	549 (68.7%)	20.0 (1)***
Information from police = yes	111 (34.3%)	253 (31.7%)	0.71 (1)
Information from social services = yes	78 (24.1%)	117 (14.6%)	14.3 (1)***
Information from other CPS agency = yes	21 (6.5%)	52 (6.5%)	0.0001 (1)
	M (SD)	**M (SD)**	**t (df)**
Duration of the investigation (days)	72.6 (58.6)	64.6 (53.9)	−2.19 (1118)*
Consultations with a parent (number of times)	3.18 (2.85)	2.77 (2.08)	−2.34 (469[a])*
Home visits (number of times)	0.95 (1.44)	0.70 (0.85)	−2.90 (418[a])**

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$. [a]Adjusted for non-equal variance.*

TABLE 3 | Differences in content of CWPS conversations with children ($N = 680$).

Content	Referrals with mental health problems ($n = 154$)		Referrals with other problems ($n = 526$)		X^2 (df) *P* for the difference
	n	%	*n*	%	
Exploratory conversation about conditions at home	114	71.1	374	71.1	0.50 (1)
Informational conversation	51	33.1	199	37.8	1.14 (1)
Conversation to obtain child's opinions	61	39.6	152	28.9	6.35 (1)*
Investigative conversation about episode in family	20	13.0	126	24.0	8.50 (1)**
Supportive conversation	17	11.0	31	5.9	4.81 (1)*
General conversation without specific aim	8	5.2	31	5.9	0.11 (1)
No information about the content	19	12.3	34	6.5	5.72 (1)*

*$p < 0.05$, **$p < 0.01$.

Another interpretation is that the CWPS workers, who are social workers and not health personnel, feel less competent and more insecure about how to evaluate the seriousness of mental health problems. This assumption might explain why these investigations became more extensive with respect to time spent and the number of consultations with parents. Parents' fear of the CWPS and subsequent resistance to inform CWPS workers about parental mental illness may also result in more complicated and time-consuming investigations compared to other types of problems. Establishing a trusting and cooperative relation with the parents, particularly when there are concerns about alcohol and/or substance abuse can be more difficult and time consuming.

When examining to what extent children were involved in cases of parental mental illness, we found that the child was less involved when the report concerned parental mental health or substance abuse. It is hard to understand what explains this practice, especially since the CWPS workers spend more time investigating these cases compared to other cases. However, this finding resonates with previous research which shows that one of the most important reason why caseworkers do not talk to children about difficult topics is that the belief that it may be a burden for children to become involved in adults' problems (14). There is nevertheless no reason to believe that resistance to talking to children in such situations is specific only to the child welfare and protection services. The same belief has been identified among health personnel (18).

In terms of which factors may predict the decision to consult the children, we found that child consultations were more likely to take place with increasing child age. Age is however not solely a predictor for involvement in cases with reported parental mental health concerns, but in all cases within the CWPS. This finding is an expected one, in line with many other studies (22–24). The main reason for this is that children are increasingly able, and perhaps also willing, to talk about family problems as they mature, or that the CWPS considers them to be less vulnerable compared to younger children and therefore more frequently initiate consultations. Although child age should not automatically disqualify children from an opportunity to talk to the social worker, we do recognize that there are limits to what can reasonably be expected from the youngest children. However, as younger children are more dependent of developmental and social support from their parents, they are also more vulnerable to lack of proper care, and hence social workers should acknowledge this in their work with younger children.

Another result of interest is that previous referrals increased the likelihood of children being involved. One plausible explanation may be that when there are previous referrals the CWPS already has knowledge about and may be acquainted with the family and the child. When the caseworker already has established a relationship with the child, this may contribute to reduce the fear that reaching out to the child will be disruptive or harmful for the child. It is known from previous studies (25) that many previous referrals are used as an indication that there is increased risk of child abuse or neglect. Increased perception of risk for the child due to the conditions at home may offset the fear a case worker has of disrupting the child by consultations.

There are good reasons to consult with children when there are concerns about the child's emotional problems. First and foremost because internalized mental health problems cannot readily be assessed without the contribution from the person in question, some form of self-report is usually required and recommended (26).

In terms of identifying what the main content of the conversations with children were when such conversations had been conducted, we found that nearly two-fifths of the conversations were aimed at giving the child information about the ongoing investigation. Usually, the social worker will have to explain to the child what the reason for the investigation is and thus disclose some information about the parents' problems in order to initiate a conversation with the child. As discussed above reluctancy to disclose such information may explain why consultations are less likely to occur. However, when they do occur this provides an opportunity to not only seek information from the child but also to provide some basic psycho-educative support. This is of great importance given the high risk these children have for developing mental health issues themselves (1, 27–29). This is particularly important when we take into consideration the relatively high chance that the case will ultimately be dismissed without any further service provision for the child or the family (28). It is positive that conversations with an aim to support the child were more often recorded in cases with reported parental mental health problems, albeit the frequency of this types of conversations were overall very low. We are quite certain that more than 11 % of the children in such cases are in need of support given the high prevalence of mental health issues among COPMI (30, 31). Admittedly, for practical and legal reasons there are limitations to how comprehensive support measures can be at this stage in CWPS case processing. However, as a minimum it could be expected that (i) the situation is explained to the child with emphasis upon the reason for contact with the CWPS, (ii) that the child is informed about what is going to happen and eventually that (iii) the reasons for subsequent decision are clearly explained. Interview studies have indicated that this is expected by children (32).

In overall 31.5% of the cases in our sample, the focus in the conversations with the child was on obtaining the child's point of view or opinions. In relation to children's right to be heard, this may seem to be a somewhat low number. The child's point of wiew was, however, more often part of the conversation if the concern was about parental mental health (39.6%). It is possible that the CWPS has talked to the child about the child's opinions and wishes without recording it. Nevertheless, the child's voice and what the child thinks about the case should emerge in a larger proportion of cases. We therefore call upon all professional partners to collaborate and to keep pushing the participation agenda forward. It is our belief that it is helpful for the development and health of COPMI children.

IMPLICATIONS FOR PRACTICE

The shortcomings in current practice in terms of involving children in cases where a parent has mental health problems have not previously been documented. However, previous studies in Norway have shown how the CWPS investigates their cases and their process from concern to decision-making (33). It was concluded that there is a need for a quality system to achieve quality assurance in practice. Studies have documented that the professionals would prefer to have more guidance and a framework to assist the assessment of risk (28). Furthermore, research has shown that the way investigations are carried out also differs between agencies (28). A national knowledge-based system and focus on the child's needs, can contribute to better documentation and a CWPS practice that to a larger extent involves the children. In January 2022, several changes will be made to the Norwegian Child Welfare Act. The intention is to strengthen prevention of child maltreatment. Children's right to participate will also be strengthened. It is, however, unclear how the amendments will be implemented in practice. There is no reason to believe that amendments in legislation will take place without an operationalized system to support a new practice. The findings of this study highlight the need for national guidelines that makes it mandatory to include children in all child protection cases.

LIMITATIONS AND STRENGTHS

This analysis is based upon what was recorded in case files. Not everything a social worker does during case processing goes into written records. It is therefore possible and likely that the numbers presented here slightly underestimates the extent and type of contact between social workers and children. That being said, the findings do not deviate substantially from what has been reported by others. The case files included in this study were solely from child welfare and protection services, and not from adult mental health services. Subsequently, we do not have information about the parents' diagnosis. We do not know if the type of mental health problems the parent had may have influenced the decision to not involve children in some cases.

It is a substantial strength that our data were randomly drawn and represents a large and representative sample.

CONCLUSIONS

In child protection cases in which the concern is mainly about the parent's mental health or substance abuse, conversations with children are conducted to a significantly lower degree compared to other cases. The CWPS are more likely to consult with older children and if the child has been referred before. The findings indicates that social workers perceptions about child vulnerability is a major obstacle for child inclusion and participation in child protection investigations. More children should be consulted in cases with reported concerns about parental mental health. Knowing that a substantial proportion of these children have or will develop problems themselves we cannot maintain a high threshold for consulting them. In our view, a child consultation should not only seek to extract information from children but should also seek to utilize the potential preventive effects that lies in basic psychoeducation. The CWPS workers are in the best position to make sure the child is involved and receives information. Child involvement is a goal that can be achieved.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because our license from the data protection authority does not allow sharing of raw data. Requests to access the datasets should be directed to Svein Arild Vis, svein.arild.vis@uit.no.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Norwegian Data Protection Authority NSD-Norwegian center for research data. Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

SV designed the study and conducted the analyses. SV, ØC, and CL collected the data. SV, CL, ØC, and CR contributed to the writing of the article manuscript. All authors agree to be accountable for the content of the work.

FUNDING

The study was funded by the Norwegian Directorate for Children, Youth and Family Affairs (Bufdir) and UiT–The Arctic University of Norway.

ACKNOWLEDGMENTS

This study is part of a larger project deigned to research the decision-making process in Norwegian Child Protection Services, and many have contributed to the overall project. The authors of this article would like to acknowledge the contribution of the following researchers to the overall project: SV (PI), ØC, CL, Karen J. Skaale Havnen, Anette Iversen, Toril Tjelflaat, Sturla Fossum, Marte Knag Fylkesnes, Veronika Paulsen, Gro Ulset, Eirinn Ljones, Kirsten Buck Rustad, Reidun H. Nygård, Frederikke Jarlby, Jannike Kaasbøl, and Stein Erik Ulvund. Additionally, the authors wish to thank the Research Support Team at RKBU/UiT-The Arctic University of Norway.

REFERENCES

1. Pierce M, Hope HF, Kolade A, Gellatly J, Osarn CS, Perchard R, et al. Effects of parental mental illness on children's physical health: systematic review and meta-analysis. *Br J Psychiatry*. (2020) 217:354–63. doi: 10.1192/bjp.2019.216
2. Nicholson J, de Girolamo G, Schrank B. Editorial: Parents with Mental and/or Substance Use Disorders and Their Children. *Front Psychiatry*. (2019) 10:915. doi: 10.3389/fpsyt.2019.00915
3. Reupert D, Maybery J, Nicholson, Göpfert M, Seeman MV. (editors) *Parental psychiatric disorder: Distressed parents and their families*. Cambridge University Press (2015). p. 238–47.
4. *Norwegian Ministry of Health and Care Services*. Changes in the law. (2009). Available online at: http://www.regjeringen.no/pages/2188713/PDFS/OTP200820090084000DDDPDFS.pdf (accessed September 9, 2021).
5. Vis SA, Strandbu A, Holtan A, Thomas N. Participation and health–a research review of child participation in planning and decision-making. *Child Fam Soc Work*. (2011) 16:325–35. doi: 10.1111/j.1365-2206.2010.00743.x
6. Lauritzen C, Reedtz C, Rognmo K, Nilsen MA, Walstad A. Identification of and support for children of mentally ill parents: a 5 year follow-up study of adult mental health services. *Front Psychiatry*. (2018) 9:507. doi: 10.3389/fpsyt.2018.00507
7. Kowalenko NM, Mares SP, Newman LK, Williams AES, Powrie RM, Van Doesum KTM. Family matters: infants, toddlers and preschoolers of parents affected by mental illness. early interventions targeting adverse influences on young children and their parents can improve children's outcomes. *Med J Aust*. (2012) 1:14–17. doi: 10.5694/mjao11.11285
8. Reupert AE, Cuff R, Drost L, Foster K, van Doesum KTM, van Santvoort F. Intervention programs for children whose parents have a mental illness: a review. *Med J Aust*. (2012) 1:7–9. doi: 10.5694/mjao11.11145
9. Mevik K, Trymbo BE. *Når foreldre er psykisk syke*. [When Parents are mentally ill]. Oslo: Universitetsforlaget (2002).
10. Grove CJ, Riebschleger J, Bosch A, Cavanaugh D, van der Ende C. Expert views of children's knowledge needs regarding parental mental illness. *Child. Youth Serv. Rev.*. (2017) 79:249–55. doi: 10.1016/j.childyouth.2017.06.026
11. Faugli A, Kufås E, Haukland M, Kallander EK, Ruud T, Weimand BM. I have cried a lot: a qualitative study on children experiencing severe parental illness. *Scand J Caring Sci*. (2020) 35:1196–206. doi: 10.1111/scs.12938
12. Lauritzen C, Reedtz C, Van Doesum K, Martinussen M. Factors that may facilitate or hinder a family-focus in the treatment of parents with a mental illness. *J Child Fam Stud*. (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
13. Van Bijleveld GG, Dedding CW, Bunders-Aelen JF. Children's and young people's participation within child welfare and child protection services: a state-of-the-art review. *Child Fam Soc Work*. (2015) 20:129–38. doi: 10.1111/cfs.12082
14. Vis SA, Holtan A, Thomas N. Obstacles for child participation in care and protection cases-why Norwegian social workers find it difficult. *Child Abus Rev*. (2012) 21:7–23. doi: 10.1002/car.1155
15. Vis SA, Fossum S. Organizational factors and child participation in decision-making: Differences between two child welfare organizations. *Child Fam Soc Work*. (2015) 20:277–87. doi: 10.1111/cfs.12076
16. Tchernegovski P, Hine R, Reupert AE, Maybery DJ. Adult mental health clinicians' perspectives of parents with a mental illness and their children: single and dual focus approaches. *BMC Health Serv Res*. (2018) 18:611. doi: 10.1186/s12913-018-3428-8
17. Kristensen KB, Lauritzen C, Reedtz C. Child-focused practice in social services for adults in Norway. *J Soc Work*. (2021) 1–21. doi: 10.1177/14680173211029725
18. Lauritzen C, Reedtz C. (2016). Child responsible personnel in adult mental health services. *Int J Ment Health Syst*. 10:64. doi: 10.1186/s13033-016-0098-y
19. Lauritzen C, Reedtz C. Support for children of mental health service users in Norway. *Ment Health Pract*. (2013) 16:12–8. doi: 10.7748/mhp2013.07.16.10.12.e875
20. McHugh ML. Interrater reliability: The kappa statistic. *Biochemia medica*. (2012) 22:276–82. doi: 10.11613/BM.2012.031
21. Skivenes M, Strandbu A. A child perspective and children's participation. *Child Youth Environ*. (2006) 16:10–27. Available online at: https://www.jstor.org/stable/10.7721/chilyoutenvi.16.2.0010
22. McDowall JJ. *Experiencing out-of-home care in Australia: The views of children and young people*. (2013). Available online at: https://apo.org.au/sites/default/files/resource-files/2013-04/apo-nid33738.pdf (accessed October 22, 2021).
23. Cashmore J. Children's participation in family law decision-making: Theoretical approaches to understanding children's views. *Child Youth Serv Rev*. (2011) 33:515–20. doi: 10.1016/j.childyouth.2010.05.008
24. Thomas N, O'Kane C. Children's participation in reviews and planning meetings when they arelooked after'in middle childhood'. *Child Fam Soc Work*. (1999) 4:221–30. doi: 10.1046/j.1365-2206.1999.00112.x
25. Fluke JD, Harlaar N, Brown B, Heisler K, Merkel-Holguin L, Darnell A. Differential response and children re-reported to child protective services: County data from the National Child Abuse and Neglect Data System (NCANDS). *Child Maltreat*. (2019) 24:127–36. doi: 10.1177/1077559518816381
26. Sun S, Wang S. The children's depression inventory in worldwide child development research: a reliability generalization study. *J Child Fam Stud*. (2015) 24:2352–63. doi: 10.1007/s10826-014-0038-x
27. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Australian e-Journal for the Advancement of Mental health*. (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
28. Christiansen Ø, Havnen KJS, Iversen AC, Fylkesnes MK, Lauritzen C, Nygård RH, et al. Barnevernets undersøkelsesarbeid–fra bekymring til beslutning. [Child protection investigations in Norway] Tromsø: UiT The arctic University of Norway. (2019). Available online at: https://bufdir.no/globalassets/global/nbbf/barnevern/a_medvirke_nar_barnevernet_undersoker_en_studie_av_barn_og_foreldres_medvirkning_i_barnevernets_undersokelsesarbeid.pdf (accessed September 25, 2021)
29. Thanhäuser M, Lemmer G, de Girolamo G, and Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. *Curr Opin Psychiatry*. (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342

30. Bifulco A, Moran P, Ball C, Jacobs C, Baines R, Bunn A. Childhood adversity, parental vulnerability and disorder: examining inter-generational transmission of risk. *J Child Psychol Psychiatry*. (2002) 43:1075–86. doi: 10.1111/1469-7610.00234
31. Kamis C. The long-term impact of parental mental health on children's distress trajectories in adulthood. *SMH*. (2021) 11:54–68. doi: 10.1177/2156869320912520
32. Havnen KJS, Christiansen Ø, Ljones EH, Lauritzen C, Paulsen V, Jarlby F, et al. Å medvirke når barnevernet undersøker. [Participation in child protection investigations] Tromsø: UiT The arctic University of Norway (2020). Available online at: https://bufdir.no/Bibliotek/Dokumentside/?docId=BUF00005143 (accessed September 25, 2021)
33. Vis SA, Christiansen Ø, Havnen KJS, Lauritzen C, Iversen AC, Tjelflaat T. Barnevernets undersøkelsesarbeid – fra bekymring til beslutning. Samlede resultater og anbefalinger. *[The child welfare's investigation work – from concern to decision. [Final results and recommendations for practice]. Barnevernets undersøkelsesarbeid: Publikasjoner fra prosjektet* Tromsø: UiT (2020).

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's Note: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

NOMENCLATURE

COPMI: Children of parents with a mental illness CWPS: Child Welfare and Protection Services CRC: Convention of the Rights of the Child

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 13 January 2022
doi: 10.3389/fpsyt.2021.793344

Perceived Support and Sense of Social Belonging in Young Adults Who Have a Parent With a Mental Illness

Aude Villatte [1,2,3,4†], ***Geneviève Piché*** [1,2,3,4*†] ***and Sylvie Benjamin*** [1,2]

[1] Laboratory LaPProche, Department of psychoeducation and psychology, Universite du Quebec en Outaouais, Saint-Jerome, QC, Canada, [2] Centre de recherche universitaire sur les jeunes et les familles (CRUJEF), Quebec, QC, Canada, [3] Réseau de recherche en santé des populations du Québec (RRSPQ), Montreal, QC, Canada, [4] Réseau Intersectoriel de Recherche en Santé de l'Université du Québec (RISUQ), Quebec, QC, Canada

OPEN ACCESS

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
Abigail Lucy Dunn,
University of Sussex, United Kingdom
Herman Hay-ming Lo,
Hong Kong Polytechnic University,
Hong Kong SAR, China
Campbell Paul,
Royal Dental Hospital of
Melbourne, Australia

****Correspondence:***
Geneviève Piché
genevieve.piche@uqo.ca

[†] These authors have contributed equally to this work and share first authorship

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *12 October 2021*
Accepted: *21 December 2021*
Published: *13 January 2022*

Citation:
Villatte A, Piché G and Benjamin S (2022) Perceived Support and Sense of Social Belonging in Young Adults Who Have a Parent With a Mental Illness. Front. Psychiatry 12:793344. doi: 10.3389/fpsyt.2021.793344

This participatory action research explores the perceived social support of youth whose parents have a mental illness during their transition to adulthood. Social support is an important protection factor during this developmental period, but few studies have explored how these young adults perceive their social support. Nor has any study assessed whether participation in a group-based participatory action research project could improve these youth's sense of support.

Purpose: (1) identify which aspects of social support these youth spontaneously address when talking about their experiences in Photovoice workshops; (2) explore how participants view these types of workshops as a good way to improve their sense of social support and belonging.

Methodology: Ten young adults (nine women and one man) between the ages of 18 and 25 who have at least one parent with a mental illness participated in *Photovoice* meetings in 2019. These group meetings aimed to explore and share their experiences as young adults whose parents have a mental illness. The testimonies were combined with data obtained from the abbreviated version of the *Social Provisions Scale* and the *Scale of Social Belonging*.

Results: The quantitative results suggest that participants consider their social support levels to be high, but their qualitative statements highlight low level or absence of parental support in terms of emotional, informative or instrumental levels. They see themselves as an important source of support for their parent and discuss the importance of having other supports figures (romantic partner, employer, friends, sibling, etc.). Conversely, they have difficulty asking for help for various reasons (including fear of stigma). They consider that their participation in this *Photovoice* project allowed them to feel heard, supported and to develop a sense of belonging to a group.

Discussion: To conclude, clinical issues to be considered for psychosocial intervention with young adults of parents with a mental illness are discussed.

Keywords: young adults, parent with a mental illness, social support, social belonging, young carers

INTRODUCTION

The transition from adolescence to adulthood is a pivotal period of development occurring approximately between the ages of 18 and 25 (1), sometimes as early as age 16 (2, 3). This period is recognized as being fraught with many significant challenges (e.g., important choices to be made, autonomy to be acquired, professional domain or post-secondary education to be discovered, maintenance or adoption of healthy lifestyle habits, etc.) (1, 4, 5). Because of these challenges, this transition is particularly conducive to the emergence or worsening of mental health problems (6–8) that could then impact the entire adult life (9, 10). Findings from several studies suggest that the transition to adulthood may be particularly challenging for youth who have a parent with a mental disorder, who represent 12 to 37% of youth (11–13). Compared to their peers, these youth are more likely to have a mental illness, report more psychological distress, both internalizing and externalizing symptoms, feelings of isolation and powerlessness, relationship, academic, and professional difficulties, substance use problems, and delinquent behavior (14–22). Knowing that unaddressed difficulties at this age of life could jeopardize the entire adult trajectory, in addition to generating significant economic and human costs (23), it is urgent to identify the levers likely to promote a successful transition to adulthood for these youth.

Social Support as a Determining Variable During the Transition to Adulthood

For young people in the general population, informal (e.g., parental availability and supervision, friendships, presence of meaningful adults) and formal (e.g., program, community, policies, recreation, meaningful stakeholders) social support is one of the key predictors of the ability to cope with the challenges of transitioning to adulthood (2, 6, 10, 24–26). In particular, social support promotes the establishment of stable relationships and improves school perseverance during this period (10, 27).

Perceived social support, which refers to a person's belief in and evaluation of their connections with others, is particularly important. One meta-analysis has highlighted the link between youth's perceived social support and well-being (28), while another has exposed an inverse link between perceived social support and depressive symptoms in youth (29). Also, results from both reviews have suggested that the quality of social support is more strongly associated with well-being and depression than the size of the support network. Similarly, results of five longitudinal studies have found that social support acts as a protective factor against depression in youth during their transition to adulthood (26, 30–33). Notably, in Scardera et al. (26) longitudinal study of 1,174 young adults, those who perceived high levels of social support at 19 years old were less likely to report mental health problems such as depressive and anxiety symptoms, at 20 years old.

The social support network of young people transitioning to adulthood, in addition to being crucial for their psychosocial adaptation, undergoes profound transformations during this period. Young people going through this developmental period face major changes in their social interactions and new challenges, such as the development of autonomy toward their parents and the emergence of stability and intimacy in their social and romantic relationships (32). To varying degrees across youth in this age group (25), parental support tends to decline while peer and romantic partner support increases (34). The relative influence of parental vs. peer or romantic partner support is still a source of debate in the scientific literature. Findings from several studies have suggested that parental support is a central protective factor in the transition to adulthood (6, 25, 29), particularly in reducing depressive symptoms (35). Instead, other studies have highlighted the role of peers, explaining that they can, in some cases, mitigate more difficult family dynamics or even compensate for poor parental support (36). Specifically, at this age, youth tend to associate more closely with people who share their interests (e.g., sports, community, peer group, volunteering) and with whom they feel a connection (37). Social interactions can greatly influence their sense of social belonging, through regular encounters with their friend group, the affects generated (38), the ability for the individual to name their expectations and fears, and the development of a shared language (39). The role of the romantic partner should also be a factor to consider, at a time when love life is gradually becoming more important to many youth (36, 40). In particular, the emerging young adult gradually perceives their romantic relationship to be more reliable. A shared intimacy accentuates mutual support, and their partner provides a significant contribution by meeting various needs (36). For many young people transitioning to adulthood, their romantic partner becomes a primary source of support, which can strengthen their resilience and reduce their stress (36).

Social Support for Young People With a Parent With a Mental Illness

Some literature points out that the role of social support in promoting well-being among youth transitioning to adulthood may be even more significant for youth considered more vulnerable during the transition to adulthood, including those from disadvantaged backgrounds or vulnerable families (29, 41), as is the case with youth that have a parent with a mental illness (8). This idea is in line with results of studies highlighting the buffer effect of social support against the negative effects of parental mental illness on child and adolescent mental health (42–44) and, more broadly, in line with the *stress-buffer model*, which suggests that the positive effect of social support should be greater in a context of adversity or significant stress (45).

The successful transition to adulthood for youth that have a parent with a mental illness may be intimately linked to the presence of support from those around them (24, 46). The presence of a positive relationship with the parent (46) and reciprocity within that relationship (24), as well as commitment and cohesion among family members (46) seem to have an influence on the young adult's well-being and resilience. In addition, the presence of positive support from peers, significant others or competent stakeholders, could act as a cross-domain buffering by helping youth that have a parent with a mental illness cope with what they are experiencing at the family level

(47). In general, friendly support could temper psychological (e.g., irrational thoughts, isolation) and emotional processes (e.g., decreases shame and fear, reassurance of one's worth), especially through the guidance and assistance of a significant peer (36, 48). Moreover, several support programs targeting these young adults and enabling them to connect with their peers, especially through professionally supervised online forums, have made it possible to offer social support (8) and have shown positive effects (49, 50). In contrast, a lack of social support (e.g., parental, friendship, romantic partner, mental health professional) is an additional vulnerability factor for youth that have a parent with a mental illness at the dawn of their transition to adulthood (24, 46).

To date, the few empirical studies that have explored how youth transiting to adulthood who have a parent with a mental illness perceive the social support they receive and offer, illustrate that they generally receive little parental support in terms of emotional, instrumental or cognitive support (14) and are also often important providers of support for their parents (46, 51). Many of these young people provide emotional, financial or even instrumental support to parents from childhood onwards, through various roles and responsibilities related to the demands of daily life. The child might then fulfill various responsibilities which become more pronounced with age, which can weaken their adaptation and lead to a process of parentification (14). This process occurs as a result of parenting gaps, but can become a burden for children of all ages, including emerging adults (14, 51). Additionally, some literature points to barriers in accessing formal resources for youth that have a parent with a mental illness of all ages (51), including the lack of knowledge among mental health professionals and parents with a mental illness regarding the impacts of parental mental illness on their child's experience, as well as their support needs (8, 52, 53). The paths of youth that have a parent with a mental illness are also negatively colored by associative stigma related to their parent's mental disorder (54). Associative stigma represents social disapproval and negative reactions toward young people due to their proximity to someone with a mental illness, often reported by youth that have a parent with a mental illness (55) and hampering their willingness to seek informal or formal support (54).

In addition to being few in number, studies that have considered the perspectives of youth of parents with a mental illness transitioning to adulthood on the social support they provide and receive all adopt "traditional" research designs by interviewing youth using questionnaires or interviews, with the limitation of restricting their responses by pre-established questions. The value of using participatory research methods that focus on artistic mediums-such as the Photovoice method that relies on photography and storytelling (56)-has been underlined with other clienteles, particularly in the mental health field (57, 58). Participating in this type of research stimulates participants' reflections and expression while allowing them to become more aware of the recurring issues they encounter, to consider solutions that make sense collectively, and to feel that they are contributing to social change by producing data that will be brought to the attention of decision-makers. Furthermore, no research has evaluated the extent to which participating in a group action research project can improve the perceived social support of these youth. This question is of major interest given that this type of project could be put in place as part of prevention and intervention programs for youths that have a parent with a mental illness.

OBJECTIVES

The present study pursues two main objectives: (1) identify which aspects of social support youth whose parents have a mental illness during their transition to adulthood spontaneously address when talking about their experiences in Photovoice workshops; (2) explore how participants view these types of workshops as a good way to improve their sense of social support and belonging. As the results of studies that have highlighted the relevance of using the Photovoice methodology with emerging adults (58), it is possible to formulate the hypothesis that proposing a Photovoice project to children of parents with a mental illness transiting into adulthood could have a beneficial effect on their feeling of belonging to a group and of feeling recognized, valued and supported.

In the present study, social support is defined as a multidimensional construct, which corresponds to a person's perception of caring or helping behaviors from people in their network (45). These behaviors can be categorized into several dimensions, including emotional (e.g., affection, empathy), instrumental (e.g., transportation) and informational (e.g., counseling) support (59). The present study will focus on self-reported perceptions of social support.

METHODS

This study is based on a secondary analysis of data from a participatory action research conducted with youth whose parents have a mental illness, using a Photovoice approach. This initial study, conducted in 2019, aimed to: (a) explore the challenges faced by youth living in such a family context during their transition to adulthood; (b) co-create with participants a recommendations report and tools that could support the transition to adulthood of youth living with a parent with a mental illness; (c) identify the benefits of participating in such a project, from the perspective of the participants themselves. It was conducted using a concurrent nested design (60) mainly focused on qualitative data (oral and written testimonies of the participants during the Photovoice meetings) and considering certain quantitative data to complete the portrait produced.

To reach the objectives of the present study, all qualitative and quantitative data shared by participants during the initial study that were specifically related to the theme of social support (e.g., group sharing around the issue of social support, evaluation of benefits in terms of perceived social support) were extracted from the initial corpus and have been analyzed. The next subsections present the methodology of the initial study and describe how the social support data were collected, specifically.

Recruitment Procedures

A promotional poster was installed in strategic locations of two targeted regions (Gatineau, Saint-Jerome, in Quebec,

TABLE 1 | Family characteristics of participants.

	Affected parent	Diagnosis	Chronicity
Participant 1	Mother and father	Depressive/substance use disorder	Since about 10 years
Participant 2	Mother	Depressive/substance use disorder	Since about 7 years
Participant 3	Mother	Anxiety/depressive disorder	Since about 11 years
Participant 4	Mother	Borderline, depressive and anxiety disorders	Since about 12 years
Participant 5	Mother	No diagnosis but anxiety/depression	Since about 20 years
Participant 6	Mother and father	Anxiety/depressive disorder	Since about 11 years
Participant 7	Mother and father	Anxiety/depressive disorder	Since about 10 years
Participant 8	Mother	Anxiety disorder	Since about 7 years
Participant 9	Mother	Anxiety/bipolar disorder	Since about 10 years
Participant 10	Mother and father	No diagnosis but anxiety/depression	Since "always"

Canada). Also, the advertisement was distributed widely through the mailing lists of educational institutions and community mental health organizations, as well as on various social media groups and pages. Interested potential participants were directed to the website of the research laboratory. After viewing a detailed presentation of the project, they could complete the information and consent form. This online registration was a preamble to a telephone interview which validated the eligibility as well as the informed consent of the potential participant.

Sample

The inclusion criteria for this study were: (a) to be between 16 and 25 years of age; (b) to have a parent with mental health problems that significantly impaired their functioning in the past 18 months, as perceived by the youth; (c) that the parent's primary disorder not be substance abuse; (d) to reside within a 50 km radius of either of the two main campuses of the Université du Quebec en Outaouais (Gatineau and Saint-Jerome); and (e) to be able to speak and understand the French language.

Eighteen youth responded to the recruitment advertisement but eight of them ultimately did not follow through to meet with the research assistant for a pre-project interview or chose not to enroll in the Photovoice workshops following the interview due to a scheduling conflict. Therefore, ten participants (including nine young women) from two administrative regions of Quebec, Canada, took part in the project. It should be noted that one participant stopped coming to the meetings after the first workshop and a second stopped after the third Photovoice workshop, both for unknown reasons.

Of these ten participants, 70% lived exclusively with their parent(s), 20% had moved out of the family home and 10% stayed with their parent(s) occasionally. These young people reported a wide variability in the frequency of contact with their parents, ranging from never to several times a day. The youngest participant was 18 years old and the oldest was 25 years old (average age 21.6 years of age). The majority (90%) have brothers and/or sisters. Eighty percent of the participants were attending a post-secondary institution while also having a part-time job. In addition, half of the sample felt they were in a precarious financial situation.

All of the youth reported having a mother with a mental illness and 40% of them reported the presence of a disorder on their father's side as well (see **Table 1**). Reported parental mental illnesses were various: major depressive disorder, borderline personality disorder, and anxiety disorder, sometimes with comorbid substance use problems. All participants indicated that their parent's mental disorders had been present for several years.

As for the mental health of the participants, 40% of them indicated that they had received a diagnosis, ranging from anxiety disorders, mood disorders or borderline personality disorder.

Data Collection

The conduct of the Photovoice project, the number of meetings and the number of participants were planned according to the recommendations of Wang and Burris (56). The participants were divided into two groups, according to their location and the meetings were conducted in presence spanning April through June 2019. Two facilitators (one male and one female), trained in the method and supervised by the research team throughout the project, accompanied each group by providing information and stimulating participation and discussion (e.g., by inviting youth to elaborate on their comments). In the first meeting, participants were first asked to answer the pre-participation questionnaire allowing, among other things, to evaluate their level of perceived social support. Then, the facilitators introduced participants to the Photovoice methodology, to the ethical stakes of such an approach and to the theme around which they would be led to express themselves using photos. before workshops. Over the course of the four next 2-h meetings, each participant had the opportunity to present his or her photos taken to testify to the experience of having a parent with a mental disorder to the group. The group members could react to each other's photos by naming how they related to their own experiences, for example. The participants' comments during these group meetings were recorded in audio format and a verbatim transcription was made. In addition to oral comments on the photos, participants were asked to write a caption and a title summarizing the message conveyed by each of their own photos. These written texts were given to the facilitators at the end of each meeting and were transcribed. The last three meetings allowed the youth to create awareness tools for different audiences, in order to convey the major issues that emerged from their discussions (see https://lapproche.uqo.ca/projets/photovoice1625/ for an overview of the tools co-created with the participants). During the last meeting, participants were asked to respond to the post participation questionnaire (level of perceived social support and their sense of social belonging to the project). Then, 6 months later, an online questionnaire was sent to participants, concerning the perceived benefits of their participation in the Photovoice group meetings.

Variables and Measures

Perceived Social Support

The verbatim excerpts and written captions of the photos addressing content related to the concept of perceived social support were combined to form the qualitative data corpus. This selection was carried out by the principal researcher and a master's student.

In order to enrich this qualitative information, the social support variable was also examined pre- and post-project using Caron's (61) abbreviated version of the *Social Provision Scale* (SPS-10). This 10-item self-report questionnaire assesses the youth's perception on five social support dimensions (Attachment, Social integration, Reassurance of worth, Reliable alliance, Guidance and Opportunity for nurturance), that correspond to the social support functions identified in Boucher and Laprise's (59) theoretical model. Thus, the SPS-10 proposes two items for each of the dimensions, for example: "I feel I am part of a group of people who share my attitudes and beliefs" (social integration); "I have people close to me who provide me with a sense of emotional security and well-being" (attachment); "There is someone with whom I could discuss important decisions that affect my life" (guidance); "I have relationships where my competence and expertise are recognized" (reassurance of worth); "There are people I can count on in case of emergency" (reliable alliance). Respondents were asked to rate themselves on each of these items using a four-point Likert-type scale ranging from Strongly Disagree (=1) to Strongly Agree (=4). The possible individual score on the SPS-10 ranges from 10 (very low social support) to 40 (very high social support), with a score of 30 and above meaning that the individual has high social support, either accessible, available, and satisfactory (62). The psychometric properties of this scale demonstrate its suitability, including excellent concurrent validity (61, 63). In addition, the internal consistency of the SPS-10 is excellent (alpha = 0.88).

Sense of Belonging

The sense of belonging to the project variable was documented in a post-project measurement with the acceptance subscale of the *Sense of Social Belonging Scale* (64). This subscale has 5 items, for example: "In my relations with the other participants, I felt supported," on which the participants must position themselves using a seven-point Likert scale ranging from Don't agree at all (=1) to Very strongly agree (=7). The score obtained can vary from 5 (very low sense of social belonging) to a maximum score of 35 (very strong sense of social belonging). The questionnaire presents very satisfactory psychometric qualities (64): internal consistency is excellent (Alpha = 0.90), criterion validity is robust with scales evaluating social support (65) and temporal stability is satisfactory (ICC = 0.70).

Perceived Benefits of Project Participation

An online questionnaire, sent to participants 6 months after the end of the project, assessed the perceived benefits in terms of support and social belonging. The items in this questionnaire are open-ended, such as "How would you describe the climate and exchanges that took place within your group," "What elements or factors led you to initially participate in the project?," "What elements or factors invited you to maintain your participation?"

Data Analyses

To respond to the first objective of the present study, Paillé and Mucchielli's (66) technique was used to make sense of the qualitative data (verbatim of the meetings and the photo captions). Hence, the PI conducted a content analysis of the transcribed data using an inductive method. As the data were read, codes were assigned to each new concept and then grouped into categories to create a thematic tree using NVivo12 software. After presentation of the coding tree to the research team and necessary adjustments, a final interpretation of the results was proposed. Descriptive analyses of the data collected during the pre-measure of the Social Provision Scale were analyzed using the Statistical Package for Social Sciences (SPSS, Standard edition 25.0). As stated by Creswell and Plano Clark (60), the combination of data provides a more accurate picture of the explored phenomenon.

To reach the second objective of the study, a content analysis was carried out based on the responses to the open-ended questions (post 6-month online questionnaire) using the method proposed by Thomas (67). Also, statistical analyses of the pre and post measure on the *Social Provision Scale*, as well as the post measure of the *Social Belonging Scale* were conducted, using SPSS 25.0. Paired sample t-tests were conducted to identify any changes in participant's perceived social support and frequency statistics were used to outline the participant's perception of social belonging at the end of the project.

RESULTS

Participant's Perceptions of the Social Support Offered and Received

Thematic analysis of verbatims and photo captions addressing perceived social support allowed us to distinguish four main dimensions: the social support offered and received in the youth-parent relationship; the impact of this family dynamic on the youth; the importance of social support from people other than the parent; the challenges associated with seeking outside help. The following subsections describe what participants say about each of these dimensions and provide additional insight from the quantitative data obtained with the pre measure of the *Social Provision Scale*.

A Caregiver Role With the Parent: "Giving Without Expecting to Receive in Return"

During the group discussions, the participants addressed a major point in their journey, namely the fact that they embodied one of the main sources of support within their families, being at the same time supervisors, friends, guardians, and providers for their parents: "*Well, I call her...always...Sometimes I would clean her house completely. I would make her meals*" (Claudia[1]), "*I clearly gave all the money I made to my parents so we wouldn't lose the house*" (Daniel), "*My mother drinks a lot, so I told her 'I promise not to ask you any questions or judge you, but when you drink, you call me so I can go get you*" (M.C.). This role of support provider seemed to be adopted even outside the relationship with the parents. Participants emphasized how much they contributed to supporting their siblings, friends, or romantic partners, for example: "*With my friends, I'm the one who listens (…) and I take care of my brother a lot.*" (Daniel).

The participants seemed to be aware that this support was beyond what a young person is usually expected to provide to his or her parent: "*Act as a parent instead of the parent.*" (Joany); "*… As a child, you're not supposed to have that role with your parents*" (Bianca).

In addition, they point out the contrast with the little support they receive from the parent. Indeed, all of the participants mentioned the absence or low level of parental support, particularly in terms of emotional, instrumental and informative functions but also in terms of supervision: "*There are never any congratulations*" (Claudia).

> "*Making my lunch, taking my bath, going to bed on time when I was little, it wasn't necessarily my mother who would tell me to do it or who would do it for me (…). I knew there was something wrong, something different about my mother, but it wasn't named, it wasn't presented to me, it wasn't explained to me*" (Marianne).

Through their comments, they expressed needs that were unmet by their parents, such as those for comfort, being listened to and feeling loved. They also pointed to the fact that they did not feel free to express themselves: "*I can't tell my mom I'm not well*" (M.C.).

According to the participants, weak parental support could be explained in part by the manifestations specific to their parent's disorder, which in turn lead to a lessening of the interactions between the young adult and their parent. A parent with a mental illness may be less emotionally available and demonstrate more anger leading to rejection: *"They can't take it, they already have enough of their own to deal with, so, you have to find someone else* (…). *When she (my mother) is like that, I can't talk to her. She just screams and cries. I have to do it on my own*" (Marie-Pier). The stigma surrounding mental health problems would also help explain why parents do not discuss their difficulties with their children: *"It's taboo, we don't talk about it*" (Marianne).

FIGURE 1 | *Hand-in-Hand*: Being a caregiver for a relative with a mental disorder means offering support, encouragement and guidance without expecting to receive anything in return. It is a team effort. It means offering support, encouragement, accompaniment and help to the other person, but also forgetting oneself and risking being dragged down when the parent relapses. It also means playing the role of parent to your parent and becoming a tool for their well-being. It tinges your emotions: it's both satisfying and disappointing. But it does make you grow in any case. (Bianca).

Impact of This Youth-Parent Dynamic: "Both Satisfying and Disappointing"

The role of caregiver to the parent is discussed ambivalently by participants, who report both positive impacts and more negative issues about it. **Figure 1** illustrates this.

Specifically, participants report a number of "positive" impacts as a result of providing strong support to their parent. The numerous tasks carried out in the perspective of supporting their parent, even their family, have contributed, according to several young people, to the development of their autonomy (e.g., ability to accomplish household tasks, set goals and maintain a budget) and social-emotional qualities (empathy, maturity, etc.) and thereby to their self-esteem: "*By always practicing this on a daily basis (supporting the parent), I consider myself a better person: more empathetic, more open to the problems of others. Sometimes I find that people are too superficial and self-centered*" (Daniel). The satisfaction of being able to help someone and the feeling of being connected to the parent were also mentioned: "*It's like being a team, doing everything together*" (Claudia).

The vocational identity of the young people also seems to be strongly influenced by their experiences as caregivers. Three of the young people in the sample chose to pursue a career path based on helping others: "*…I'm not interested in it, but it's something that's part of me too. I was born into it and it's going to follow me all my life*" (Victoria). The two participants who were already involved in a helping profession underlined the risk of overload that such a position can entail and the importance of setting limits: "*I can't intervene on what is too close to me, I can't intervene with people who have an alcohol problem*" (M.C.).

---

[1] Some participants chose to keep their first name, others chose a pseudonym.

While providing support to their parents might have had some positive effects, participants noted that this role had also had significant negative repercussions. The amount of time, money and energy spent on family support may make it difficult for the young person to meet his or her own needs (e.g., thinking about what he or she would like to do with his or her life, having time to see friends.) Some participants were concerned about leaving their parents alone or leaving the family nest, while others tried to preserve the emotional well-being of their parents first, sometimes at the expense of their own well-being.

> *"… I'm going to be stressed out about, like, going out at night because I know that she, she doesn't feel well. So then I start to make scenarios in my head… I'm afraid all the time… is she going to fall back into alcohol? Is she going to have suicidal thoughts because of me?... I try to comfort her in everything".* (Marie-Pier).

These repercussions of the caregiving role can lead to cognitive and emotional fatigue, which was mentioned by six of the participants, using the following terms: "feeling worn out and old" and "really tired." The following comments illustrate these repercussions:

> *"Sometimes I tend to carry all his emotions on my shoulders, in addition to all that I'm going through... It's like I go through everything twice... there are responsibilities, that yes a child has because deep down we love our parent, but I think it can become heavy…"* (Marie-Pier).
> *"… it got me down. In the sense that you know yes, you give a lot, you give a lot, but at one point it takes your energy away, it takes your time away, it brings you down".* (Marianne).

The lack of access to information and advice can, on the other hand, generate a lot of misunderstanding and frustration toward their perception of an optimal parental support. A chain reaction can then arise, between the unspoken words, the lack of access to information, the altered communication and several negative repercussions for youth that have a parent with a mental illness (i.e., frustration, guilt, sadness, as well as psychological distress or mental health symptoms).

> "*It made me angry not to understand, not knowing what was going on… and also angry toward myself, toward my mother, toward my family for not explaining it to me. It's scary to go and talk to someone, to get help".* (Marianne).

Finally, the lack of parental support may be particularly detrimental during the transition to adulthood, when young people need a positive role model to build their future adult identity (see **Figure 2**).

In the face of all of these impacts, many participants emphasized that limiting the support to be given to the parent and mourn the long-awaited parental support are important strategies for protecting oneself.

> *"I have come to understand that you can't help someone who doesn't want to help themselves. You have to learn to understand that you can't do more than the person wants" (Bianca).*

FIGURE 2 | *Growing Up Small*: As we transition into adulthood, we may feel small and insecure because of the lack of role models growing up. Perhaps our role models had difficulty holding on to life themselves because of their difficulties. So, we don't feel equipped to deal with this big world and still see ourselves as small. (Victoria).

> *"At some point, in a tiredness that might have made sense, I felt compelled to set my limits. So I said to her: "You know, Mom, something really needs to happen here. This can't go on" (M.C.).*
> *"It's mourning the loss of one's parents, although you still have hope that things will get better, of course, you always have hope, but when it's continuous, consecutive through time… well, at a given moment there's this letting go, this resignation a little bit also, because of the fact that well, I don't want to have too much hope because… it's always like this*" (Daniel).

More than half of the participants mentioned that they sometimes had to distance themselves from their parent in order to refocus on themselves or to self-soothe. They attempt to recharge their batteries, as an adaptive strategy that allows them to self-regulate and let go in the face of repercussions generated by their parent's mental illness: "*I started to think more about myself than about others. I only went to see my mom on the weekends. Doing things just for me*" (Marianne).

Social Support as a Protective Factor: "What Helps in Dealing With the Challenges of Having a Parent With a Mental Disorder Is to Be Supported"

Having the support of one or more people around you is considered to be one of the most protective factors in dealing

FIGURE 3 | *Together*: What helps us feel better about the challenges of having a parent with a mental disorder is social support. It allows us to confide in each other during difficult times, to share our happiness and to dream together. My girlfriend and some of my friends offer me a lot of support in my daily life. (Victoria).

with the above-mentioned impacts. Victoria's words and photo (see **Figure 3**) testify to this.

> *"You are looking out for your parent's well-being, but you are looking out for your own well-being elsewhere, by seeking support elsewhere because you are not necessarily going to get it from your parent"* (Victoria).

Almost all participants (except one) mentioned the presence of other people in their environment who played a major role in their lives and quantitative data indicate that they report a social support score that is considered high (62). In the original sample ($n = 10$), the mean score on the *Social Provisions Scale* (pre measure) was 35.2 (26.00–40.00). Only one participant fell below the threshold of 30 with a score of 26. This result is consistent with results of studies conducted on samples of emerging adults from the general population, which highlight that 10% of the sample typically does not meet the threshold for high social support (62). The results show that the dimension of social integration (e.g., "There are people who enjoy the same social activities as I do") has the lowest mean score ($x = 3.25$), while tangible help (e.g., "There are people I can count on to help me in times of real need") is the dimension with the highest mean score ($x = 3.7$). It thus seems that these young people better perceived the help from those around them in cases of real need or emergency (items concerning reliable alliance) vs. from participating in social activities or being part of a group (items concerning social integration). **Table 2** shows the average score obtained on the SPS-10 by dimension in the pre-project measure.

Siblings, extended family members and friends are examples of support figures mentioned. For example, one participant recounted how fortunate she was to have a strong bond with her sisters which helped her cope with and normalize daily life while facing parental mental illness. This form of sibling support endured, and her strong positive assessment of it referred to mutual feedback and to the development of her own self-esteem. In the same vein, the notions of non-judgment and recognition were valued and raised when it came to proximal relationships:

> *"My uncles were there and they encouraged me more than my mother. And when they saw me… "Congratulations, we are proud of you!""* (Bianca).

The period of transition to adulthood, with the opportunities for encounters that it offers, allows the appearance of new support figures. The discussions highlighted that romantic partners become an important resource in terms of social support. Six participants explained that their romantic partners fulfilled many functions and that the latter enabled them to develop in a healthier way. This type of social support responded to all their support needs, such as emotional, instrumental, and informational. For example, participants indicated that their romantic partners "*have their back*" (comforting, loving and being loved), that part of their success was "*because of their partner*" (advice, guidance, acquiring skills), that he or she helped them overcome "*obstacles*" (meeting the demands of everyday life or dealing with problems) and that he or she contributed to their self-esteem. The great importance given to their romantic partners was mentioned several times during the meetings: "*Stability within my relationship is something… as unstable as my life has been, it's the opposite for my couple.*" (Bianca).

> *"And I'm proud of what I am, vs. what I could have been if, for example, my boyfriend had not been in my life, if he had not advised me"*. (Joany).

In-laws and employers can also act as new support figures during this period: "*It's been about two years since I've started having the impression that I have a role model in my life and it is my boss who is very warm and very maternal too"*. (Victoria).

Challenges Encountered in the Search for External Support: "It's Not Always Easy to Get Help"

Participants in this study mentioned that the support of people close to them was sometimes no longer sufficient to meet their needs. The dimension in which they seem to consider the support of their relatives as sometimes insufficient concerns the space they would need to be able to talk about what they are experiencing with their parent. The participants emphasize that talking about what they are experiencing with their parent could be a good coping strategy ("*It helps a lot to be able to talk about it,*" Claudia), but indicate that they rarely find people to whom they can talk about the parental mental disorder.

Participants highlighted the impact the lack of information and the stigma surrounding mental health issues can have on the support they received. Notably, 80% of participants felt that most people did not understand their situation, are uncomfortable hearing what the youth has to say or would be judgmental of the youth and family: "*In my circle, that's how it's perceived: "Oh, when you talk about your mother... Oh, change the subject…, I never have the opportunity to talk about it. I*

TABLE 2 | Social provision scale mean score (pre-project results).

	Subscales					
	Attachment	Reliable alliance	Social integration	Guidance and opportunity for nurturance	Reassurance of worth	Total score
Participant 1	4	4	3.5	4	4	39
Participant 2	4	4	4	4	4	40
Participant 3	4	4	4	4	3.5	39
Participant 4	3.25	3.5	3	5	2.75	30
Participant 5	4	4	3.5	4	4	39
Participant 6	3.5	3.5	2	4	4	34
Participant 7	3	3	3	1	3	26
Participant 8	4	4	4	4	4	40
Participant 9	3.5	4	2.5	3.5	3	33
Participant 10	3	3	3	3	4	32
	x = 3.63	x= 3.7	x= 3.25	x = 3.65	x = 3.63	Mean score = 35.2

always feel like I'm making them feel uncomfortable" (Joany). Two participants reported never being comfortable discussing their parent's mental illness with anyone (friend, colleague, mental health professional, teacher, close family member). The fear of associative stigmatization (disapproval and negative reactions toward them) and auto-stigmatization (internalization of the stigma) were present in the discourse of participants and it is important to specify that they developed in an environment where they themselves had little or no information about mental illness: "*I'm so suspicious of people… to be told… that I'm not normal because of my parent.*" (Joany).

The feeling of being the only young person around them living with a parent with a mental disorder fuels their feelings of guilt, shame and ambivalence about seeking help and support, as raised in this excerpt:

> "*You're isolated in this because you don't know that there are other people like you. You don't want to talk about it because you're afraid of other people's judgement, you're afraid that other people will tell you: we don't care*". (Daniel).

While some named the challenge of verbalizing a request for help, other participants report that sometimes, they did not know where to turn for help:

> "*When I was crying because I felt guilty... I didn't know who to cry to, I didn't know who to call... You go through your contact list three times, you don't call anyone after all because like, who am I going to tell this to*". (Victoria).
> "*It's not that I was alone. I had lots of people, but I couldn't talk to them.*" (Daniel).

These feelings of not being able to turn to others for fear of misunderstanding or fear of being stigmatized can lead to voluntary social withdrawal. The following example illustrates this point:

> "*After that you isolate yourself because you don't want to tell others about it, you want to get better. Well, feel better*". (Marianne).

Participants also mentioned that the felt emotional load (e.g., guilt, fear, doubt) sometimes became too high, forcing them to seek external professional help. Thus, by themselves, in a self-taught way and as a last resort, Five participants said that they had consulted professionals such as psychiatrists, doctors or psychologists in recent years, that is, during their transition to adulthood. Personal development and the urgency to take care of oneself were their main motivations.

> "*I've tried a lot, but I'm at the point where I need to see a psychologist... which I didn't want to do at first, but now I'm at that point*". (Marie-Pier).

Again, there are several fears that appear to be barriers to seeking formal help, including fear of being stigmatized and fear of harming the family system: "*You're afraid to go and see them (social workers), to tell them what you're going through, and then they call the Youth Protection and you leave. It ends up that you don't go see anyone*" (Bianca), "*You don't want the other students to see you coming out of the social worker office, because they know that there's something wrong with you or your family*" (Joany).

The Perceived Effects of Their Participation in the Project on Social Support and the Feeling of Belonging

Perceived Effect of Their Participation on Sense of Support and Social Belonging

First, participants rated the *Photovoice* experience as positive: excellent ($n = 3$), very good ($n = 4$) and good ($n = 1$). Also, all youth say they would recommend participation in an equivalent project to all other young adults that have a parent with a mental illness.

Second, all participants spoke of the normalization felt and conveyed through the project as a result of countering their feelings of social isolation: "*It normalizes a lot to see that we are* *not alone.*[2]" Quickly, they recognized each other through their own experiences and, for the first time, they met young people with a similar background. In addition, several excerpts from the group discussions made it possible to identify a link between participation in meetings, the perception of social support and the feeling of belonging generated. The young adults took a look at their journey together and were unanimous in expressing the fact that they felt listened to, as evoked in the following extract: *"We needed to speak and we found this space… it's like a first source of support".*

Moreover, an additional effect noted by the participants was that the exchanges allowed them to underline their respective resilience as well as to offer a discourse that conveyed hope.

> *"It's true that this normalizes things and it takes us away from this view of our experience, which is a bit victimizing, where we feel alone in what we are going through, and that it's very sad...when we can just change our perspective".*

Participants point out that their continued participation and mobilization are due to the enriching interactions between members, the desire to support other youth living this reality, and the positive repercussions on their sense of well-being.

> *"What motivated me was that I felt I didn't have a space outside to talk about my parent's mental health issue and I thought I could find that space and availability, while building something bigger with our experience."*
> *"The group discussions allowing to understand each other and have strong emotional exchanges, introspection and awareness."*

In addition, the collected responses highlight two important characteristics of this study that allowed the experience to be considered positive. Firstly, participants described the climate within the group as respectful, offering an atmosphere of openness and listening, which led to mutual trust between them. Second, they said that the support in their respective groups was understanding, empathetic and non-judgmental. One participant explained that her feeling of being supported allowed her to feel comfortable to name her personal challenges to the group.

Complementary Insights From Quantitative Data About Perceived Social Support and Sense of Belonging

The average score of participants on the *Sense of Belonging Scale* following their participation is 31.12 (25.00–35.00), which is considered high, considering that the maximum score is 35 (64). The results highlight that the item "In my relationships with other participants, I felt listened to" is the one that obtains the highest score. Moreover, all the answers are at the high end of the proposed Likert scale, which validates a high level of agreement toward the feeling of belonging generated within the group by the collective project.

TABLE 3 | Social provision scale mean score (by dimension; pre and post-project results).

Subscales	Pre	Post
Attachment	3.63	3.80
Reliable alliance	3.71	3.68
Social integration	3.25	3.56
Guidance and opportunity for nurturance	3.42	3.69
Reassurance of worth	3.38	3.75
Total	35.2	37.0

Furthermore, the descriptive statistics from the pre- and post-measures with the SPS-10 reveal that the total mean score increased from 35.2 to 37 out of 40 (see **Table 3**), as did the mean scores for each dimension, with the exception of the reliable alliance dimension (e.g., "I have people I can count on in an emergency"). The greatest increase in the average score is granted to the dimension measuring the reassurance of worth (e.g., "I have relationships where my knowledge and competence are recognized"). The young people therefore evaluated having a high sense of social belonging within their group and their perception of social support improved following their participation in the project. The following verbatim illustrates the results obtained:

> *"We sympathized with each other a lot, I think, with our lifestyles...Finally yes, it's true, I'm not alone. And, I have proof because we are very similar."*

DISCUSSION

This participatory action research first documented how emerging adults whose parent has a mental illness talk about the social support they provide to their loved ones, as well as the support they receive from those around them. According to them, young people provide a great deal of support to their mentally ill parent, in all areas, in accordance with several previous studies (24, 51). Also, all youth rated their parental support as low and some as non-existent. This finding is consistent with the literature on children of parents with a mental illness of all ages, who report receiving little parental support (46, 55, 68, 69).

However, the comments exchanged during the meetings and the average score obtained by the participants on the *Social Provisions Scale*-compared to that reported in similar studies (62) show that they feel they receive a high level of social support. If their parents are perceived as unavailable and unapproachable to meet their needs, due to their mental health problems (e.g., fatigue, consumption, irritability), the youth participants perceive positive support coming from their broader social network: romantic partners, close or extended family members, bosses, friends, teachers or mental health professionals. All these sources of support remind one that the transition to adulthood, rich with opportunities to meet new people and to free oneself from parental supervision, is a period conducive to resilience

---

[2] The questionnaires completed six months after participation in Photovoice meetings were completed anonymously. It is therefore not possible to identify which participant made the comment.

(1, 2, 70, 71). Results thus suggests that because they are faced with low parental support, young adults of parents with a mental illness seek support elsewhere to meet their needs, especially emotional ones. Indeed, these different sources of support fulfill various functions, such as allowing young people to feel loved, recognized and encouraged, to receive advice and to express themselves. Consistent with work that highlights how critical these are to the development of young people in transition to adulthood in the general population (6, 25, 36), the young people who have a parent with a mental illness in our study considered them to be "vital" support, mitigating the impact of inadequate support from their parents. This qualitative finding, however, diverges significantly from the results of a large longitudinal study conducted among young people in transition to adulthood with a depressed parent, in which quality of social functioning was not identified as playing a significant protective role in resilience among youth with a depressed parent, in comparison with the parent-child relationship or the youth's intelligence quotient (46). More research is needed to clarify the nature and strength of the links between the perception of positive non-parental social support and the development of resilience in young adults with a parent with a mental illness.

Our results also made it possible to observe the little informational support that young people receive about parental mental illness, which is in line with the results of some previous studies (13, 51, 69), as well as their difficulties in mobilizing their support network and in seeking help (54). In fact, this study highlights that a range of factors create difficulties in seeking help or social support in general, such as the taboo surrounding parental illness, which seems to persist despite efforts made in recent years to reduce the stigma surrounding mental health problems (54). This finding is consistent with what several authors have noted about the impact of stigma (54) and developmental issues (such as the search for autonomy) (72) which complicate help-seeking during the transition to adulthood. Considering that participants say they want advice on mental health and the development of skills to better manage the family situation, and that half of them had asked for the advice of a mental health professional in recent years, it seems crucial to promote their access to informational social support and formal help regarding parental mental health (8, 69).

Finally, our results highlight that children of parents with a mental illness transiting to adulthood tend to isolate themselves on purpose, particularly in times when they encounter challenges. Indeed, many participants reported consciously distancing themselves from their parent or friends to regulate their negative emotions and protect themselves from the judgment of others. As well, some of them said that they had given so much to their parent that they perceived solitary withdrawal as a time of respite. Studies of other groups of youth facing bullying or stigmatization have shown that social withdrawal can be used as a coping strategy (73, 74), but the present study is the first to highlight this practice among young adults of parents with a mental illness. However, although in the short term this strategy allows youth to avoid stress and cut themselves off from the source of discomfort or suffering, it carries a risk of isolation and low social reciprocity (73).

Regarding the second research question related to improving perceived social support, notably through the feeling of belonging generated by participating in a common project, our results are encouraging. First, perceived social support increased for all the youth following their participation and their felt social belonging in this co-construction project turned out to be high. Also, at the 6-month post-project measurement, effects as well as social and relational gains from their participation were still mentioned. For some, it was about normalizing what they experienced or reducing their feeling of isolation, while others stated a better understanding of their relationship with the parent. Our findings are consistent with other studies from general population that have found beneficial influence of participation in participatory action research using Photovoice on emerging young adults (57, 58), particularly in terms of perceived support (75). The participatory and group-based approach seems to be a means of fostering a sense of belonging, particularly through the recognition of experiential knowledge (25, 39), and subsequently improving the perception of social support through exchanges between group members (38).

Clinical Implications

Results of this study makes it possible to highlight certain specific clinical implications for mental health and education professionals who work with young adults of parents with a mental illness. First, it is suggested that professionals advocate for the development of resources specific to these vulnerable youth in schools, communities and care institutions, as well as promote the resources that are available to the latter, as young adults may not be aware of them. Because of the taboos that still surround mental health issues, their self-directedness in terms of seeking social support but also a tendency toward self-sufficiency specific to the transition to adulthood, these youth may have difficulty seeking help despite recognizing that it could be helpful. Recommendations for facilitating access to services and interventions are highlighted, based on the reflections of young adults of parents with mental illness who participated in a complementary component of this study (76). They suggested, among other things, greater visibility and diversity of mental health resources, adaptation of communication channels for youth their age (e.g., online, anonymous, interactive, social via networks), and explanation of rights and confidentiality rules as part of appropriate support. In addition, participants recommended that mental health and education professionals be open in their approach, advocate for an egalitarian relationship and provide a discreet environment. In particular, professionals need to consider the difficulties these youth may face, such as fear of being judged on their experiences and their lack of knowledge about parental illness due to poor communication.

Second, results of this study underline the relevance of systematically evaluating the quality of social support from various sources, both intrafamilial (parents, siblings) and extrafamilial (friends, romantic partners, colleagues, employers), as well as both informal and formal (e.g., intervener, support group) among these youths. Apart from standardized evaluation instruments, the proximity circle, an interpersonal psychotherapy (IPT) clinical tool, could be used to help youths identify important people and resources in their social network, in a more informal way (77). In addition to evaluating this aspect of the young adult's experience, results of this study, in accordance with recommendations from recent studies, advise that professionals support youth of parents with a mental illness in their social needs (78), including by helping them find strategies to maintain or improve their support network. This can be achieved through various manualized individual, group-based, or online interventions (79), or through the use of an informational booklet (80). In the same vein, it is also suggested that professionals experiment with various intervention modalities (e.g., discussion groups, digital platforms) to transmit information and offer support to young people whose parent has a mental illness, and use various mediums (e.g., photography, writing, art) to reduce the barriers to help-seeking and promote participant introspection. The development of a participatory group project may be an interesting strategy to facilitate a sense of being heard and sharing with others (peers and supportive adults). It would be beneficial to the resilience of these youth who are used to dealing with stigma and rejection within their usual peer group and who report lacking support from a parental figure (81).

In addition to providing psychosocial services and ensuring that they are accessible (e.g., by offering them online and in the environments frequented by young people on a daily basis) and visible, particularly through attractive promotion on social networks, it is important to provide a variety of concrete assistance measures (e.g., financial support, logistical assistance such as meal deliveries, academic accommodations to facilitate school-work-family balance) so that young people feel supported. The recent implementation of a Policy for Caregivers in Quebec (82), which aims to raise awareness among different audiences of what young caregivers may experience, while recognizing and valuing their role more and offering them assistance measures (e.g., respite), seems to be a promising avenue for young people who play a caregiver role to feel recognized and supported.

Support for families is also an important avenue. Improving the adult care system and providing support to parents could also relieve the youth of a sense of responsibility. Working with families to support parents in becoming more stable, in offering a secure and positive home for their children, and having a sufficient safety net around them would probably make it easier for the youth to become independent and to project themselves positively into their adult lives.

Continued efforts to reduce the stigma surrounding mental health problems and to promote positive mental health and support-seeking as an effective strategy for living a fulfilling life seems, at last, essential (76). The more young people who have parents with mental health problems are exposed to people who are aware of what they may be experiencing and the right strategies for individual and collective self-care, the more likely they are to feel supported.

Strengths and Limits

One of the strengths of this study is that it allowed the youth participants to have an initial space to talk about what they were experiencing and to explore their reality with other young people living in similar situations. This first step brought a sense of normalization and provided a stepping stone to further define themselves and find meaning in their difficult family situations (52). By recognizing their lived experience, the Photovoice method contributes to the involvement and commitment of young adults (83). In fact, the participants stated that they would recommend participation in a similar project to all young adults of parents with a mental illness like themselves.

However, the study has some limits that need to be clarified. First, the sample size is small, which provides unrepresentative results; therefore, it is not possible to infer or generalize the results to all young adults of parents with a mental illness at the outset. The research design also does not make it possible to determine causal links. Furthermore, the scope of the results is limited since the sample is mainly composed of women and youth whose parents have anxiety-depressive disorders. On another note, some dimensions of social support were not examined in this study (59). Although the SPS-10 has excellent psychometric qualities (63), it would be beneficial to combine its use with instruments assessing the diversity and size of the network, as well as the quality of relationships (84) in order to capture the multiple facets of the concept of social support in future studies.

CONCLUSION

In conclusion, this participatory action research study documented the perception of social support among young people transitioning to adulthood whose parent has a mental illness. The results highlight low levels of felt parental support, as noted by all the participants, and their great difficulty in soliciting support from their social network, even though they considered it essential, even vital. Participants pointed to the major importance of other significant people in their environment, such as their romantic partners, friends or mental health professionals, as a source of formal social support. In general, our results underline the relevance of providing young adults whose parent has a mental illness with resources that meet their needs and recognizing the specific issues related to their transition to adulthood, especially in a context where they frequently provide major support to their parent.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Comité d'Éthique de la recherche de l'Université du Québec en Outaouais (UQO) (2019-191). The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

AV undertook and led the development of the research design and methodology, with contributions from GP. AV and SB undertook the literature review and led the data collection, with contributions from GP. SB led the data analysis and interpretation of findings, with contributions from AV and GP. SB and GP wrote the manuscript with editing/contributions from AV. All authors contributed to the article and approved the submitted version.

FUNDING

Data collection for this study was funded by the Social Sciences and Humanities Research Council (CRSH 892-2018-2024).

ACKNOWLEDGMENTS

The authors wish to thank the young adults that agreed to take part in this study. Their generosity in sharing their lived experience has helped us better understand the challenges they face and how we might better respond to their needs. Another special thanks to Camille Ballick for her linguistic review of the manuscript.

REFERENCES

1. Arnett JJ. *Emerging Adulthood: The Winding Road From The Late Teens Through The Twenties.* Oxford University Press (2014). doi: 10.1093/acprof:oso/9780199929382.001.0001
2. Masten AS, Burt KB, Roisman GI, Obradović J, Long JD, Tellegen A. Resources and resilience in the transition to adulthood: continuity and change. *Dev Psychopathol.* (2004) 16:1071–94. doi: 10.1017/S0954579404040143
3. Roberge MC, Déplanche F. Synthèse des connaissances sur les champs d'action pertinents en promotion de la santé mentale chez les jeunes adultes. *Institut national de santé publique du Québec (INSPQ).* (2017) 73.
4. Domene JF, Nee JJ, Cavanaugh AK, McLelland S, Stewart B, Stephenson M, et al. Young adult couples transitioning to work: the intersection of career and relationship. *J Vocat Behav.* (2012) 81:17–25. doi: 10.1016/j.jvb.2012.03.005
5. Eskritt M, Doucette J, Robitaille L. Does future-oriented thinking predict adolescent decision making? *J Genet Psychol.* (2014) 175:163–79. doi: 10.1080/00221325.2013.875886
6. Boden JM, Sanders J, Munford R, Liebenberg L. The same but different? applicability of a general resilience model to understand a population of vulnerable youth. *Child Indic Res.* (2018) 11:79–96. doi: 10.1007/s12187-016-9422-y
7. Bruffaerts R, Mortier P, Auerbach RP, Alonso J, Hermosillo De. la Torre AE, Cuijpers P, et al. Lifetime and 12-month treatment for mental disorders and suicidal thoughts and behaviors among first year college students. *Int J Methods Psychiatr Res.* (2019) 28:e1764. doi: 10.1002/mpr.1764
8. Drost LM, van der Krieke L, Iedema-den Boer Z, Sytema S, Schippers GM. Social support in chat sessions for adolescents and young adults living with a family member with mental illness. *Int J Ment Health Nurs.* (2018) 27:683–92. doi: 10.1111/inm.12354
9. Asselmann E, Wittchen H-U, Lieb R, Beesdo-Baum K. Sociodemographic, clinical, and functional long-term outcomes in adolescents and young adults with mental disorders. *Acta Psychiatr Scand.* (2018) 137:6–17. doi: 10.1111/acps.12792
10. Yuan R, Ngai SS. Agentic personality as mediator of social capital on developmental outcomes in the transition to adulthood: evidence from Shanghai, China. *J Adolesc.* (2016) 46:1–13. doi: 10.1016/j.adolescence.2015.10.013
11. Bassani DG, Padoin CV, Philipp D, Veldhuizen S. Estimating the number of children exposed to parental psychiatric disorders through a national health survey. *Child Adolesc Psychiatry Ment Health.* (2009) 3:1–7. doi: 10.1186/1753-2000-3-6
12. Reedtz C, van Doesum K, Signorini G, Lauritzen C, van Amelsvoort T, Van Santvoort F, et al. Promotion of wellbeing for children of parents with mental illness: a model protocol for research and intervention. *Front Psychiatry.* (2019) 10:606. doi: 10.3389/fpsyt.2019.00606
13. Reupert A, Maybery D. What do we know about families where parents have a mental illness? a systematic review. *Child Youth Serv.* (2016) 37:98–111. doi: 10.1080/0145935X.2016.1104037
14. Mitchell JM, Abraham KM. Parental mental illness and the transition to college: coping, psychological adjustment, and parent–child relationships. *J Child Fam Stud.* (2018) 27:2966–77. doi: 10.1007/s10826-018-1133-1
15. van Santvoort F, Hosman CM, Janssens JM, van Doesum KT, Reupert A, van Loon LM. The impact of various parental mental disorders on children's diagnoses: a systematic review. *Clin Child Fam Psychol Rev.* (2015) 18:281–99. doi: 10.1007/s10567-015-0191-9
16. Apter G, Bobin A, Genet M-C, Gratier M, Devouche E. Update on mental health of infants and children of parents affected with mental health issues. *Curr Psychiatry Rep.* (2017) 19:1–7. doi: 10.1007/s11920-017-0820-8
17. Duncan G, Browning J. Adult attachment in children raised by parents with schizophrenia. *J Adult Dev.* (2009) 16:76–86. doi: 10.1007/s10804-009-9054-2
18. Ensminger ME, Hanson SG, Riley AW, Juon H-S. Maternal psychological distress: adult sons' and daughters' mental health and educational attainment. *J Am Acad Child Adolesc Psychiatry.* (2003) 42:1108–15. doi: 10.1097/01.CHI.0000070261.24125.F8
19. Hameed MA, Lewis AJ. Offspring of parents with schizophrenia: a systematic review of developmental features across childhood. *Harv Rev Psychiatry.* (2016) 24:104–17. doi: 10.1097/HRP.0000000000000076
20. Weissman MM, Wickramaratne P, Nomura Y, Warner V, Pilowsky D, Verdeli H. Offspring of depressed parents: 20 years later. *Am J Psychiatry.* (2006) 163:1001–8. doi: 10.1176/ajp.2006.163.6.1001
21. Weissman MM, Wickramaratne P, Gameroff MJ, Warner V, Pilowsky D, Kohad RG, et al. Offspring of depressed parents: 30 years later. *Am J Psychiatry.* (2016) 173:1024–32. doi: 10.1176/appi.ajp.2016.15101327
22. Mowbray CT, Bybee D, Oyserman D, MacFarlane P, Bowersox N. Psychosocial outcomes for adult children of parents with severe mental illnesses: demographic and clinical history predictors. *Health Soc Work.* (2006) 31:99–108. doi: 10.1093/hsw/31.2.99
23. Whiteford HA, Ferrari AJ, Degenhardt L, Feigin V, Vos T. The global burden of mental, neurological and substance use disorders: an analysis from the Global Burden of Disease Study 2010. *PLoS ONE.* (2015) 10:e0116820. doi: 10.1371/journal.pone.0116820
24. Abraham KM, Stein CH. Staying connected: young adults' felt obligation toward parents with and without mental illness. *J Fam Psychol.* (2010) 24:125. doi: 10.1037/a0018973
25. Howard Sharp KM, Schwartz LE, Barnes SE, Jamison LE, Miller-Graff LE, Howell KH. Differential influence of social support in emerging adulthood across sources of support and profiles of interpersonal and non-interpersonal potentially traumatic experiences. *J Aggress Maltreatment Trauma.* (2017) 26:736–55. doi: 10.1080/10926771.2017.1289999
26. Scardera S, Perret LC, Ouellet-Morin I, Gariépy G, Juster R-P, Boivin M, et al. Association of social support during adolescence with depression, anxiety, and suicidal ideation in young adults. *JAMA Netw Open.* (2020) 3:e2027491. doi: 10.1001/jamanetworkopen.2020.27491
27. Bussières E-L, St-Germain A, Dubé M, Richard M-C. Efficacité et efficience des programmes de transition à la vie adulte: Une revue systématique. *Can Psychol Can.* (2017) 58:354. doi: 10.1037/cap0000104

28. Chu PS, Saucier DA, Hafner E. Meta-analysis of the relationships between social support and well-being in children and adolescents. *J Soc Clin Psychol.* (2010) 29:624–45. doi: 10.1521/jscp.2010.29.6.624
29. Rueger SY, Malecki CK, Pyun Y, Aycock C, Coyle S. A meta-analytic review of the association between perceived social support and depression in childhood and adolescence. *Psychol Bull.* (2016) 142:1017. doi: 10.1037/bul0000058
30. Colman I, Zeng Y, McMartin SE, Naicker K, Ataullahjan A, Weeks M, et al. Protective factors against depression during the transition from adolescence to adulthood: findings from a national Canadian cohort. *Prev Med.* (2014) 65:28–32. doi: 10.1016/j.ypmed.2014.04.008
31. Galambos NL, Barker ET, Krahn HJ. Depression, self-esteem, and anger in emerging adulthood: seven-year trajectories. *Dev Psychol.* (2006) 42:350. doi: 10.1037/0012-1649.42.2.350
32. Pettit JW, Roberts RE, Lewinsohn PM, Seeley JR, Yaroslavsky I. Developmental relations between perceived social support and depressive symptoms through emerging adulthood: blood is thicker than water. *J Fam Psychol.* (2011) 25:127. doi: 10.1037/a0022320
33. Macalli M, Côté S, Tzourio C. Perceived parental support in childhood and adolescence as a tool for mental health screening in students: a longitudinal study in the i-Share cohort. *J Affect Disord.* (2020) 266:512–9. doi: 10.1016/j.jad.2020.02.009
34. Soenens B, Park S-Y, Vansteenkiste M, Mouratidis A. Perceived parental psychological control and adolescent depressive experiences: a cross-cultural study with Belgian and South-Korean adolescents. *J Adolesc.* (2012) 35:261–72. doi: 10.1016/j.adolescence.2011.05.001
35. Shulman S, Kalnitzki E, Shahar G. Meeting developmental challenges during emerging adulthood: the role of personality and social resources. *J Adolesc Res.* (2009) 24:242–67. doi: 10.1177/0743558408329303
36. Szwedo DE, Hessel ET, Loeb EL, Hafen CA, Allen JP. Adolescent support seeking as a path to adult functional independence. *Dev Psychol.* (2017) 53:949. doi: 10.1037/dev0000277
37. Collins A, van Dulmen M. Friendships and romance in emerging adulthood: assessing distinctiveness in close relationships. In: *Emerging adults in America: Coming of age in the 21st century.* Washington, DC: American Psychological Association (2006). p. 219–34. doi: 10.1037/11381-009
38. Lo Coco G, Gullo S, Profita G, Pazzagli C, Mazzeschi C, Kivlighan Jr DM. The codevelopment of group relationships: the role of individual group member's and other group members' mutual influence and shared group environment. *J Couns Psychol.* (2019) 66:640. doi: 10.1037/cou0000349
39. Nizet I, Monod Ansaldi R. Construction de bénéfices mutuels en contexte collaboratif: pistes théoriques et méthodologiques. *Phronesis.* (2017) 6:140–52. doi: 10.7202/1040224ar
40. Furman WD, Winkles J. Transformations in heterosexual romantic relationships across the transition into adulthood: "meet me at the bleachers … i mean the bar". In: Laursen B, Collins A, editors. *Relationship Pathways: From Adolescence to Young Adulthood*. (2012). Available online at: http://dx.doi.org/10.4135/9781452240565.n9.
41. Gutowski E, White AE, Liang B, Diamonti A-J, Berado D. How stress influences purpose development: the importance of social support. *J Adolesc Res.* (2018) 33:571–97. doi: 10.1177/0743558417737754
42. Hoefnagels C, Meesters C, Simenon J. Social support as predictor of psychopathology in the adolescent offspring of psychiatric patients. *J Child Fam Stud.* (2007) 16:87. doi: 10.1007/s10826-006-9070-9
43. Daches S, Vine V, Layendecker KM, George CJ, Kovacs M. Family functioning as perceived by parents and young offspring at high and low risk for depression. *J Affect Disord.* (2018) 226:355–60. doi: 10.1016/j.jad.2017.09.031
44. Sell M, Barkmann C, Adema B, Daubmann A, Kilian R, Stiawa M, et al. Associations of family functioning and social support with psychopathology in children of mentally ill parents: multilevel analyses from different rating perspectives. *Front Psychol.* (2021) 12:705400. doi: 10.3389/fpsyg.2021.705400
45. Cohen S, Wills TA. Stress, social support, and the buffering hypothesis. *Psychol Bull.* (1985) 98:310. doi: 10.1037/0033-2909.98.2.310
46. Pargas RCM, Brennan PA, Hammen C, Le Brocque R. Resilience to maternal depression in young adulthood. *Dev Psychol.* (2010) 46:805. doi: 10.1037/a0019817
47. Grove C, Reupert A. Moving the field forward: developing online interventions for children of parents with a mental illness. *Child Youth Serv Rev.* (2017) 82:354–8. doi: 10.1016/j.childyouth.2017.10.003
48. Caron J, Guay S. Soutien social et santé mentale: concept, mesures, recherches récentes et implications pour les cliniciens. *Santé Ment Au Qué.* (2005) 30:15–41. doi: 10.7202/012137ar
49. Woolderink M, Bindels JA, Evers SM, Paulus AT, van Asselt AD, van Schayck OC. An online health prevention intervention for youth with addicted or mentally ill parents: experiences and perspectives of participants and providers from a randomized controlled trial. *J Med Internet Res.* (2015) 17:e274. doi: 10.2196/jmir.4817
50. Reupert AE, Cuff R, Drost L, Foster K, Van Doesum KT, Van Santvoort F. Intervention programs for children whose parents have a mental illness: a review. *Med J Aust.* (2013) 199:S18–22. doi: 10.5694/mja11.11145
51. Ali L, Ahlström BH, Krevers B, Skärsäter I. Daily life for young adults who care for a person with mental illness: a qualitative study. *J Psychiatr Ment Health Nurs.* (2012) 19:610–7. doi: 10.1111/j.1365-2850.2011.01829.x
52. Gladstone BM, Boydell KM, Seeman MV, McKeever PD. Children's experiences of parental mental illness: a literature review. *Early Interv Psychiatry.* (2011) 5:271–89. doi: 10.1111/j.1751-7893.2011.00287.x
53. Maybery D, Goodyear M, O'Hanlon B, Cuff R, Reupert A. Profession differences in family focused practice in the adult mental health system. *Fam Process.* (2014) 53:608–17. doi: 10.1111/famp.12082
54. Reupert A, Gladstone B, Helena Hine R, Yates S, McGaw V, Charles G, et al. Stigma in relation to families living with parental mental illness: an integrative review. *Int J Ment Health Nurs.* (2021) 30:6–26. doi: 10.1111/inm.12820
55. Grové C, Reupert A, Maybery D. The perspectives of young people of parents with a mental illness regarding preferred interventions and supports. *J Child Fam Stud.* (2016) 25:3056–65. doi: 10.1007/s10826-016-0468-8
56. Wang C, Burris MA. Photovoice: concept, methodology, and use for participatory needs assessment. *Health Educ Behav.* (1997) 24:369–87. doi: 10.1177/109019819702400309
57. Lorenz LS, Kolb B. Involving the public through participatory visual research methods. *Health Expect.* (2009) 12:262–74. doi: 10.1111/j.1369-7625.2009.00560.x
58. Marcotte J, Villatte A, Vrakas G, Laliberté A. L'identité narrative de jeunes dits ≪vulnérables≫ s' apprêtant à transiter vers l'âge adulte au Québec. *Psychol Fr.* (2019) 64:241–55. doi: 10.1016/j.psfr.2018.01.002
59. Boucher K, Laprise R. Le soutien social selon une perspective communautaire. In: Dufort F, Guay J, editors. *Agir au coeur des communautés. La psychologie communautaire et le changement social*. Québec: Les Presses de l'Université Laval (2001) p. 117–56.
60. Creswell JW, Clark VLP. *Designing And Conducting Mixed Methods Research*. Los Angeles: Sage publications (2017).
61. Caron J. Une validation de la forme abrégée de l'Échelle de provisions sociales: l'ÉPS-10 items. *Santé Ment Au Qué.* (2013) 38:297–318. doi: 10.7202/1019198ar
62. Orpana HM, Lang JJ, Yurkowski K. Recherche quantitative originale Validation d'une version réduite de l'Échelle de provisions sociales au moyen de données d'enquêtes nationales canadiennes. *Promot Santé Prév Mal Chron Au Can.* (2019) 39:11. doi: 10.24095/hpcdp.39.12.02f
63. Gottlieb BH, Bergen AE. Social support concepts and measures. *J Psychosom Res.* (2010) 69:511–20. doi: 10.1016/j.jpsychores.2009.10.001
64. Richer SF, Vallerand RJ. Construction et validation de l'échelle du sentiment d'appartenance sociale (ÉSAS). *Eur Rev Appl Psychol.* (1998) 48:129–38.
65. Russell DW, UCLA. Loneliness scale (version 3): reliability, validity, and factor structure. *J Pers Assess.* (1996) 66:20–40. doi: 10.1207/s15327752jpa6601_2
66. Paillé P, Mucchielli A. *L'analyse Qualitative En Sciences Humaines Et Sociales-5e éd.* Paris: Armand Colin (2021).
67. Thomas DR. A general inductive approach for analyzing qualitative evaluation data. *Am J Eval.* (2006) 27:237–46. doi: 10.1177/1098214005283748
68. Kelley ML, Bravo AJ, Hamrick HC, Braitman AL, White TD, Jenkins J. Parents' reports of children's internalizing symptoms: associations with parents' mental health symptoms and substance use disorder. *J Child Fam Stud.* (2017) 26:1646–54. doi: 10.1007/s10826-017-0677-9
69. Sherman MD, Hooker SA. Supporting families managing parental mental illness: Challenges and resources. *Int J Psychiatry Med.* (2018) 53:361–70. doi: 10.1177/0091217418791444
70. Sussman S, Arnett JJ. Emerging adulthood: developmental period facilitative of the addictions. *Eval Health Prof.* (2014) 37:147–55. doi: 10.1177/0163278714521812

71. Mendonça M, Fontaine AM. Perception mutuelle des relations entre les jeunes adultes et leurs parents: distance, appui et conflit. *Rev Int Léducation Fam.* (2013) 1:59–84. doi: 10.3917/rief.033.0059
72. Wilson CJ, Rickwood DJ, Bushnell JA, Caputi P, Thomas SJ. The effects of need for autonomy and preference for seeking help from informal sources on emerging adults' intentions to access mental health services for common mental disorders and suicidal thoughts. *Adv Ment Health.* (2011) 10:29–38. doi: 10.5172/jamh.2011.10.1.29
73. Galland O. Youth: discrimination based on appearance. *Econ Stat.* (2006) 393:151–84.
74. Salmon N. 'We just stick together': How disabled teens negotiate stigma to create lasting friendship. *J Intellect Disabil Res.* (2013) 57:347–58. doi: 10.1111/j.1365-2788.2012.01541.x
75. Yi J, Zebrack B. Self-portraits of families with young adult cancer survivors: Using photovoice. *J Psychosoc Oncol.* (2010) 28:219–43. doi: 10.1080/07347331003678329
76. Villatte A, Piché G, Habib R. Comment soutenir la résilience des jeunes en transition vers l'âge adulte qui ont un parent atteint d'un trouble mental? ce qu'en pensent les jeunes concernés. *Int J Child Adolesc.* (2021) 8:1–18. doi: 10.7202/1077723ar
77. Dietz LJ, Weinberg R, Mufson L, Weinberg RB. *Family-based interpersonal psychotherapy for depressed preadolescents.* Oxford University Press (2018). doi: 10.1093/med-psych/9780190640033.001.0001
78. Reupert A, bee P, Hosman CM, Van Doesum KT, Drost LM, falkov A, et al. Prato research collaborative for change in parent and child mental health: principles and recommendations for working with children and parents living with parental mental illness. *J Child Psychol Psychiatry*. (2021) 62:1132–9. doi: 10.1111/jcpp.13521
79. Marston N, Stavnes K, Van Loon LM, Drost LM, Maybery D, Mosek A, et al. A content analysis of intervention key elements and assessments (IKEA): what's in the black box in the interventions directed to families where a parent has a mental illness? *Child Youth Serv.* (2016) 37:112–28. doi: 10.1080/0145935X.2016.1104041
80. Grové C, Melrose H, Reupert A, Maybery D, Morgan B. When your parent has a mental illness: children's experiences of a psycho-educational intervention. *Adv Ment Health.* (2015) 13:127–38. doi: 10.1080/18387357.2015.1063637
81. Parkinson A, Keddell E, Walker P. Exploring peer support as a strategy to reduce self-stigma for marginalised children of parents with mental illness (COPMI). *Br J Soc Work.* (2021) 51:849–68. doi: 10.1093/bjsw/bcaa161
82. Ministère de. la Santé et des Services Sociaux (MSSS). *Politique nationale pour les personnes proches aidantes* (2021).
83. Becquet V, Goyette M. *L'engagement des jeunes en difficulté*. Sociétés Jeun En Diffic Rev Pluridiscip Rech. (2014) (14).
84. Flores Jr LE, Cyranowski JM, Amole M, Swartz HA. Prospective assessment of social network quality among depressed mothers treated with brief psychotherapy: the Social Network Quality (SNQ) scales. *Compr Psychiatry.* (2017) 78:98–106. doi: 10.1016/j.comppsych.2017.06.012

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 14 January 2022
doi: 10.3389/fpsyt.2021.815526

Identification of Children of Mentally Ill Patients and Provision of Support According to the Norwegian Health Legislation: A 11-Year Review

Charlotte Reedtz[1]*, *Eva Jensaas*[2], *Trine Storjord*[2], *Kjersti Bergum Kristensen*[1] *and Camilla Lauritzen*[1]

[1] *Faculty of Health Science, Regional Centre for Child and Adolescent Mental Health and Child Welfare (RKBU North), Arctic University of Norway, Tromsø, Norway,* [2] *University Hospital of North Norway, Tromsø, Norway*

Background: According to amended legislation implemented in Norway in 2010, personnel in healthcare services for adults are obligated to identify patients' minor children and to assess the family situation. Health personnel is also obligated to contribute to adequate support to families affected by parental mental illness or substance use disorders. The intention behind the amendment was to support and protect children of mentally ill parents, as they are at risk of developing problems of their own. The aim of the present study was to evaluate health personnel's practice during the years 2010-2020, more specifically; (a) to what extent children of patients with mental illness and substance use disorders are registered in patient records, and (b) to what extent activities relating to the assessment and support of patients' minor children are documented in patient records.

Method: The participants in the study are patients admitted to Division for Mental Health and Substance Use at the University Hospital of North Norway in the years 2010–2020. The data was drawn from patient records during October 2021.

Results: The registration of patients' minor children is considerably strengthened since the introduction of the new Norwegian Health Personnel Act in 2010, and estimates show that 56% of patients' minor children are identified. However, only 31% of cases where patients have identified minor children this result in health personnel performing activities to support the children.

Discussion: Based on the rising proportion of identified minor children throughout the 10-year period, it seems evident that the dissemination efforts have contributed to the development of some new skills among health personnel. However, compared with the national estimation that 35% of mentally ill and substance abusing patients have minor children, a large proportion of children remains unidentified. After identification, there seem to still be a long way to go before minor children are systematically offered support. Different solutions to strengthen the implementation of new skills in clinical practice, to ensure the identification of minor children and provision of necessary support for them is discussed.

Keywords: change of clinical practice, children of mentally ill parents, provision of support, prevention of socio-emotional problems, identification of risk status

OPEN ACCESS

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Frances Louise Dark,
Metro South Addiction and Mental
Health Services, Australia
Susanne Jaeger,
ZfP Südwürttemberg, Germany

****Correspondence:***
Charlotte Reedtz
charlotte.reedtz@uit.no

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *15 November 2021*
Accepted: *07 December 2021*
Published: *14 January 2022*

Citation:
Reedtz C, Jensaas E, Storjord T,
Kristensen KB and Lauritzen C (2022)
Identification of Children of Mentally Ill
Patients and Provision of Support
According to the Norwegian Health
Legislation: A 11-Year Review.
Front. Psychiatry 12:815526.
doi: 10.3389/fpsyt.2021.815526

TRANSGENERATIONAL TRANSMISSION OF MENTAL DISORDERS

The transgenerational transmission of mental disorders is a significant cause of mental illness, and children of parents with mental illness or substance use disorders (COPMI) are at risk of developing mental illness themselves (1). Elevated risk for COPMI has been demonstrated across the diagnostic spectrum and is both diagnosis-specific and general (2). In addition, parents' symptomatology also have an impact on their social functioning and may in turn shape the way parents interact with their children. Impairment of parenting skills, reduced quality of care parents provides and problems in the parent-child interactions, is often a result of psychopathology in parents (3, 4). Furthermore, such impairments may in turn lead to insensitivity, hostility directed at the child, rejection and neglect (3), with possible subsequent insecure attachment (5, 6), emotional dysregulation, negative emotionality and pathological coping strategies as well as psychopathology in childhood, adolescence and adulthood (1, 3). As a consequence of hereditary, social and parent-child interaction factors, COPMI are very likely to constitute the next generation of mentally ill persons and parents (7).

Many children live in families with parental mental health problems and one in five has a parent with mental illness (8). In Norway, it has been estimated that 450,000 minor children (41.5 % of all children) have parents with mental illness or alcohol use disorder (9). The National Institute of Public Health (NIPH) has calculated this based on prevalence studies of the number of adults who qualify for a psychiatric diagnosis or alcohol dependency in 1 year. The numbers are adjusted for the fact that people with diagnosable mental illnesses have children to a smaller degree compared to healthy people, and that they often find partners with diagnosable mental illnesses. Since the estimate represent a cross section of the data throughout a year, consequently the number of children with parents with diagnosable mental illnesses throughout their childhood is even higher. Other researchers (10) have also estimated that one third of the patients at Norwegian hospitals have care responsibilities for minor children. Even though there is a solid evidence base for the many risk factors related to the transgenerational transmission of mental illnesses, research shows that it has been very difficult for professionals working with mentally ill patients and substance use problems to identify and support their children (2, 11, 12). In Norway, these children have not traditionally been registered in their parents' records and hence have not been identified. Without routines to assess whether patients have children, it is impossible to safeguard children who are affected by parental mental illness and related family problems. For these reasons, Norwegian authorities made legislative amendments in 2010 to safeguard minor children (0–18 years) of patients with mental illnesses and substance use disorders by adding new paragraphs to the Norwegian Health Personnel Act (13). Health personnel have since 2010 been required to provide minor children with information and necessarily follow up related to parental mental illness and substance use. The new regulations require all health professionals to; (1) register dependent children in the patient's record, (2) inform the parent about children's need for information and support, (3) assist in providing the children in the family with information, (4) provide the children with information about the opportunities to visit parents at the hospital, (5) assess children's and the family's needs, and (6) obtain parents' consent to cooperate with other services in establishing necessary support (14).

IMPLEMENTATION OF PRACTICE CHANGES

Implementation is defined as targeted effort to carry out plans, decisions or interventions in a municipality, organization or general practice (15). This definition implies that implementation processes are targeted, managed and described in great enough details for independent observers to be able to observe the process and evaluate it. A model for degrees of implementation was developed by Fixsen et al. (15). The model categorizes goals and results of an implementation process as either paper implementation, process implementation or skills implementation. In paper implementation, decisions about innovations are rooted in formal resolutions. In process implementation, procedures and systems are changed to make it possible to materialize the innovations, and relevant participants are provided with necessary training. In skills implementation, relevant participants are conducting the innovation in such a way that new skills are manifested in clinical practice and can be measured. The legislation related to COPMI represent paper implementation according to Fixsen et al.'s model, as the planned innovation was rooted in new paragraphs in existing legislation and regulations related to these. The legislative authorities also described how the new laws and regulations should be operationalized, thus representing the next level in the implementation model, process implementation.

Changing human behavior is however challenging (16). This is also true for changing health personnel's practice related to patient's minor children (11, 17). Implementation science focuses on studying methods for promoting the uptake of consolidated research findings into routine healthcare practice and health policy, and many researchers have studied which factors have an impact on the implementation of new practices (16, 18, 19). A recent scoping review conducted by Fakha et al. (18), identified an interplay of 25 main factors that acted as barriers and facilitators during the implementation of diverse health care innovations (18). There is a wide range of interrelated factors existing at multiple levels that determine the success of the implementation of innovations (18), which explains why changing clinical practice is challenging, time consuming and needs to be monitored over time.

In a previous study at the University Hospital in North Norway (UNN), which is the largest hospital in the region, results showed that only 4–7% of patients were registered with minor children, even though 35.3% of patients were estimated to have minor children (20). Given the speed of implementation in year 2015, it was calculated that it would take ∼19 years before patients' children were registered, and hence identified the way they should according to the amended legislation from 2010. There may be many reasons to why health personnel do not register or identify patients' children in patient records. First, they may have low professional awareness related to the fact that many patients are caring for minor children, and that these children are at risk of developing social and/or mental health problems themselves (21). Second, health personnel who work with adult patients may feel insecure in discussions about childcare and in including patients' children in the treatment, because they are not trained or educated to do this (12). Third, the financial structure of health services in Norway is based on client contacts and as patients' children are not clients, contact with COPMI does not result in any financial support or refund. Fourth, it may be unclear whose responsibility it is to register patients' children in the journal. Fifth, time constraints in clinical work at hospitals may result in health personnel not prioritizing assessment of whether the patient has children or the needs of these children (22). A recent study showed that the rate of registering patients' minor children was higher in university hospitals compared to smaller hospitals in the country, and that Norwegian hospitals had implemented change in clinical practice related to COPMI at a medium level (13).

SPECIFIC ACTIONS TO SUPPORT IMPLEMENTATION OF THE AMENDED LEGISLATION IN NORTH NORWAY

A crucial instrument to change the clinical practice related to patients' minor children, was to make it mandatory for all hospitals in Norway to appoint child responsible personnel (CRP) in wards, clinics and institutions. The intention was that CRP should be responsible for promoting and coordinating support for patients' minor children (23). The University Hospital in North Norway (UNN) also chose to establish a new function named CRP-coordinator in each clinic and these served as managers of all CRP in their clinic. Furthermore, The Northern Norway Regional Health Authority made guidelines to describe the mandated clinical practice to identify and support COPMI, and these were effective from 2012. The guidelines described which information about COPMI should be registered, who should register, how to document the information in the electronic patient records (EPR), as well as where in the EPR this information belonged. The EPR utilized by the hospitals in North Norway is called DIPS, which is the largest supplier of eHealth systems to Norwegian hospitals. DIPS provides a software package for EPR, which in turn provide health care workers with an integrated and unified electronic presentation of all important and relevant clinical information about patients, including patients' minor children. In DIPS patients' minor children should be registered at the front page in the EPR, among central patient administrative information.

In addition to the provision of specific guidelines and procedures related to the process implementation of the innovation, The Northern Norway Regional Health Authority provided health personnel with opportunities to participate in training programs related to service provision for COPMI. From 2013 they financed even larger parts of the implementation process, and a considerable sum were used to train health personnel and to support implementation activities. In 2014, UNN was also provided with a 50% position as CRP coordinator managing all CRP and COPMI related activities in the hospital, in order to support implementation activities related to the innovation. In 2016 a National Professional Procedure for patients with minor children was implemented and approved by the National Health Library. This procedure and the guidelines from the regional health authority coincided and put even greater pressure to change clinical practice accordingly. Furthermore, from 2017, UNN decided to change the terms for CRP-coordinators in each clinic and chose to pay health personnel in 20% positions to support the implementation process. In sum, The Northern Norway Regional Health Authority have taken control over a variety of implementation activities, and it seems safe to say that the legislative changes related to COPMI in UNN has been followed by implementation support at both the local, regional and national level. However, until now, it remains unclear whether the implementation of the innovation represents what could be characterized as changed clinical practice through acquirement of new skills related to COPMI.

THE PRACTICE CHANGES TO BE MONITORED OVER TIME

In this particular study, the practice changes to be monitored over time is linked to the amended legislation (the new paragraphs in the Norwegian Health Personnel Act from 2010), where the registration of COPMI was the core intention, as well as documenting activities related to provision of support for patients' minor children. All COPMI activities should be documented in the EPR by a COPMI report. The COPMI report should include information about identification and assessment of the child, conversations about the child and family with the patient, conversations with the child and family, consent or no consent to cooperate with other services, evaluations of the situation of the child, as well as further follow-up. The COPMI reports have changed throughout the ten-year period from several separate documents to five documents which can be utilized in the EPR in 2020. As of now, the main COPMI report is designed as a form which could be continued with new entries as new activities are performed in clinical practice. The intention with this report is to provide an easily accessible overview of relevant information regarding the patient's minor children and how they have been informed and supported. According to the mandatory guidelines, all patients with mental illnesses or substance use disorders who are registered with minor children should have at least one COPMI related documents in EPR and this is a COPMI report.

AIMS OF THE CURRENT STUDY

The present study is part of a longitudinal COPMI project at the Arctic University of Norway, in which the goal was to support the implementation of new routines arising from legislative amendments, as well as to evaluate the process of change (20). The aim of the present study was to evaluate (a) to what extent health personnel registered children of patients with mental illness and substance use in electronic patient records (EPR) during the years 2010–2020, and (b) to what extent activities relating to the assessment and support of COPMI are documented in EPR according to the mandatory guidelines.

METHODS

Participants

Participants in this study were all patients in the Division for Mental Health and Substance Use disorders in the largest hospital in North Norway. The University Hospital in North Norway (UNN) is responsible for the delivery of mental health care services at the specialist level in the two most northern counties in Norway (see **Figure 1**). Northern Norway is an area with large geographical spread, covering an area two times larger than Denmark. UNN is one of four public health undertakings in the region, all part of The Northern Norway Regional Health Authority.

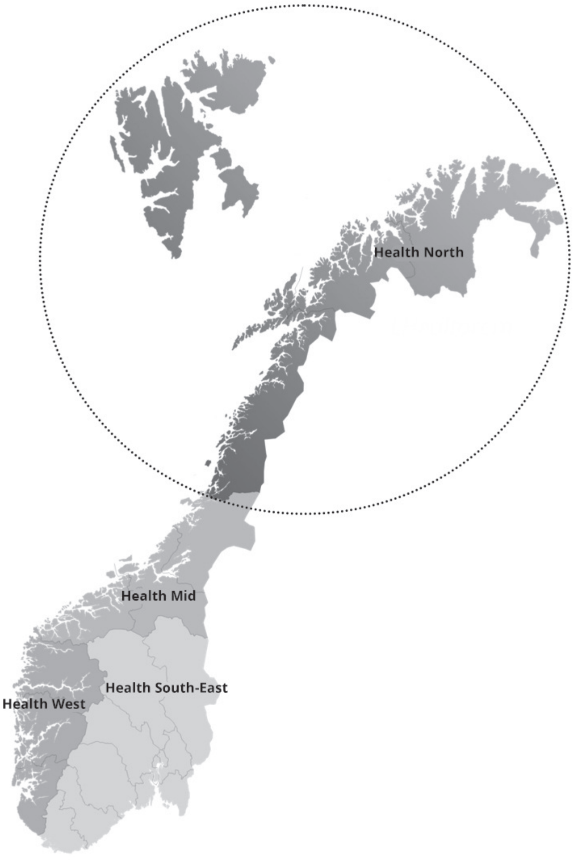


FIGURE 1 | Norwegian Regional Health Authorities. The Northern Norway Regional Health Authority in circle.

Design and Procedure

This study is a retrospective registry study based on anonymized data from EPR. Data from all patients were drawn from DIPS EPR in October 2021 and consisted of:

- Information regarding the patients' children, registered in the administrative front page of the EPR.
- Information regarding COPMI related activities, registered in COPMI reports in the EPR.

Recruitment

UNN's participation in this quality assurance study is rooted in a collaboration between The Arctic University of Norway - UiT and a project manager at UNN.

Ethical Considerations

Quality assurance studies in UNN do not require patient consent. All analyzed data was anonymized. The project has been approved by the Data protection officer at UNN.

Statistical Analyses

Statistical analyses were computed using SPSS (version 19) and Excel® for Microsoft 365 MSO.

Based on data from Statistics Norway (SSB) we have estimated the probability of an adult person having children between the age of 0 and 18 years old. Using Bayes' formula (24) we calculated the probability of mentally ill adults in Norway having children. These calculations are based on statistical information from SSB and the Norwegian Institute of Public Health (NIPH). SSB provides data on the number of households in Norway, the portion of married, co-habitants and single adults, as well as to what degree people in these groups have children. In a study from NIPH (9) researchers estimated the relative risk of being mentally ill when you have children vs. being mentally ill in the general population. We included this information in our calculations and calculated the probability for the degree to which mentally ill patients in Norway have children. Our analyses showed that the chance that mentally ill adults in Norway have children is 33.5%. In addition to that the Norwegian Institute of Public Health has estimated that there is a 5.4% elevated risk to have mental illness when one is a parent, compared to the risk of this in the general adult population (9). We added this risk into our calculations and the result showed that there is a 35.3% chance to have children when adults are mentally ill (25).

Time series modeling in SPSS was used to predict how many years it would take to adhere to the amended legislation related to COPMI.

RESULTS

Patients Registered With Minor Children in EPR

A total of 28,906 unique patients received mental health care at Division for Mental Health and Substance Use Disorders at UNN during the years 2010–2020, in outpatient and inpatient units/wards at several locations (see **Table 1**). The mean age of patients ranged from 39 (median 38) in 2010 to 38 (median 35) in 2020. The results show that the number of patients with

TABLE 1 | Total number of patients per year, estimated number of patients with minor children, actual number of patients with registered minor children, and actual number of patients with registered minor children and at least one COPMI report documenting mandated COPMI activities in the EPR.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020
Total number of patients*	6,244	6,563	6,695	6,579	6,612	6,950	6,952	7,080	7,150	6,981	6,507
Estimated number of patients with minor children	2,204	2,317	2,363	2,322	2,334	2,453	2,454	2,499	2,524	2,464	2,297
Number of patients with minor children**	2	51	181	280	484	682	844	977	1,088	1,188	1,286
Number of patients with minor children and at least on registered document related to COPMI***	20	230	181	204	189	244	279	268	343	369	401

*Data from HN LIS available from The Northern Norway Regional Health Authority.
**Data collected by using DIPS Report 2531765.
*** Data collected by using DIPS Report 2531754.

registered children in the years 2010–2020 increased rapidly from the first year with new legislation and onward. Based on the estimated probability of 35.3% that mentally ill patients in Norway have children, results show that the number of patients with minor children vary from 2,204 to 2,524 during the period from 2010 to 2020. The percentage of registered COPMI, based on these estimates, have increased from 0.1% in 2010, to 27.8% in 2015 and to almost 56% in 2020.

Many patients are however patients over a longer period of time than 1 year or have been admitted more than one time during the 11-year period. In such cases they are counted as unique patients every year they were admitted as a patient in the Division for Mental Health and Substance Use Disorders, and hence possibly more than one time. Subsequently, minor children of these patients may also be registered every year they were admitted. **Table 2** presents patients registered with minor children for the first time per year and the actual number of minor children these patients were registered with. Of the 455 children that were registered in 2019, a total of 238 of them were younger than 6 years of age. Results show that a total of 3,476 unique minor children have been identified during the 10-year period.

Based on the speed of changes related to the identification of patients' minor children from 2010 and onward the results show that it will take a total of 18 years until all minor children are identified, and hence this could be a reality in year 2028.

COPMI Related Documents in EPR for Patients With Registered Minor Children

The results show that COPMI related documents are registered in the EPR of 35.5% of patients with registered minor children or lower, during the 11-year period (see **Figure 2**).

Figure 2 shows that not every patient with registered minor children had at least one COPMI report in the EPR as mandated. In 2020 only 17.5% of patients estimated to have minor children had documented activities related to COPMI in EPR. Results show that during the last 3 years the number of patients with registered minor children and at least one document in EPR have stabilized at around 31% (31.5% in 2018, 31.1% in 2019 and 31.2% in 2020), indicating that more than two thirds of patients with registered minor children did not have mandated documents related to COPMI in the EPR. More specifically, the results show that in 2020 a total of 1,286 patients were registered with minor children (56%), whereas only 401 of these (31.2%) had registered documents related to COPMI in the EPR (see **Figure 3**). **Figure 2** presents the estimated number of patients with unregistered minor children, actual number of patients with registered minor children and number of patients with registered minor children and at least one document related to mandated COPMI activities in 2020. The estimated number of patients with unregistered minor children in 2020 is 1,011.

Based on the speed of changes concerning documentation of COPMI related activities for patients with registered minor children the results show that it will take a total of 82 years from 2010 until this clinical practice is implemented, and hence this could be a reality in year 2092.

DISCUSSION

The first aim of the present study was to evaluate to what extent health personnel registered children of patients with mental illness and/or substance use disorders (COPMI) according to the Norwegian Health Personnel Act in electronic patient records (EPR) during the years 2010–2020. The result from this study shows that the identification of COPMI has improved considerably since the introduction of the new Norwegian Health Personnel Act in 2010. It is very positive that more children are identified, as this is a necessity to provide COPMI with the information and support they are entitled to. According to the straightforward procedures on how to register patients' children in the EPR, it seems that this is not time-consuming and is feasible for most health personnel.

Based on the rising proportion of identified COPMI throughout the 11-year period, it seems evident that the implementation steps and activities that have been utilized at UNN have contributed to the development of new skills among health personnel. The willingness to organize the implementation process, finance core activities and positions, as well as providing the workforce with training opportunities represents important steps to support development of COPMI related skills such as identifying patients' minor children.

However, compared with the national estimations that 35.3% of patients with a mental illness and/or substance use disorders have minor children, a large proportion of children still remains

TABLE 2 | Total number of patients registered with minor children for the first time per year, and the total number of children they were registered with in the EPR.

	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020
Total number of patients registered with minor children first time*	4	54	125	128	285	295	318	309	329	316	263
Total number of minor children registered first time*	4	60	166	177	391	405	443	442	466	455	467

Data collected by using DIPS Report 2531765.

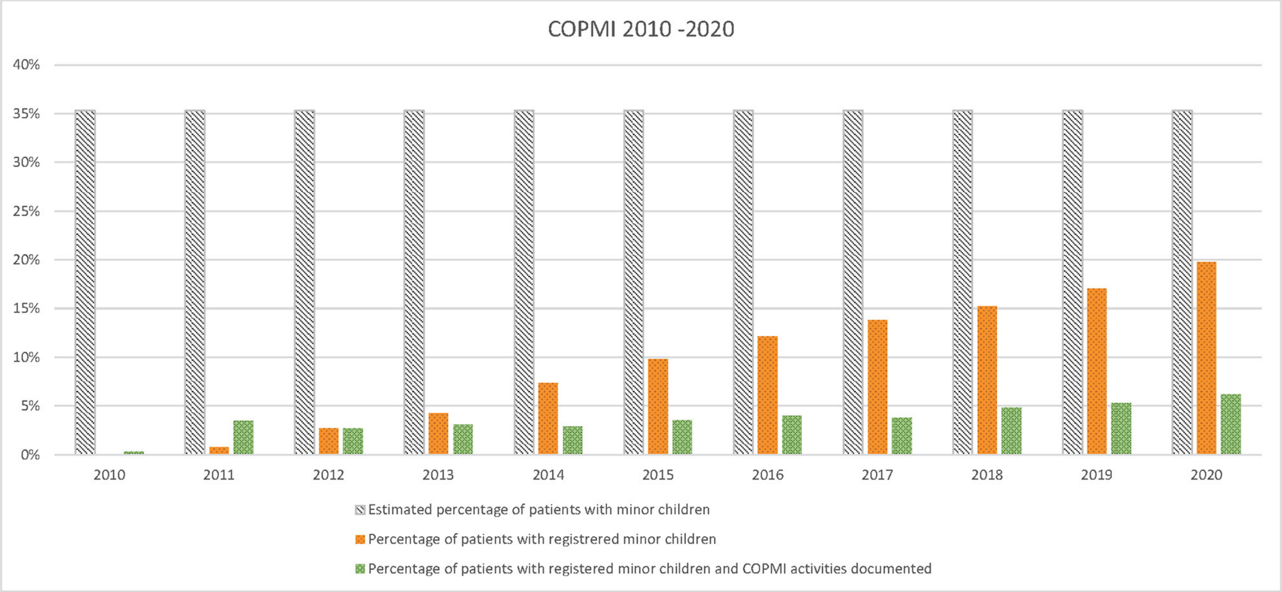


FIGURE 2 | Estimated percentage of patients with minor children, actual percentages of patients with registered minor children and percentages of percentages of patients with registered and one document related to mandated COPMI activities in 2010–2020.

unidentified. To our knowledge, there are no reasons to believe that patients in the Division for Mental Health and Substance Use Disorders at UNN have children to a lower extent than the national estimates. Furthermore, there are no available data to support that the population of the two northernmost counties in Norway are parents to a lower extent than the rest of the country.

The actual age distribution among patients may also be relevant in this context, if the patients were too old to have children aged 0–18. However, our data shows that the mean age of patients in the Division for Mental Health and Substance Use disorders at UNN are in line with national reports on patient data where 70% of all patients in Norwegian mental health care is between 18 and 49 years. The national figures for age distribution among these patients are overlapping with our results, and hence most patients receiving mental health care services are in the age where the probability of having children aged 0–18 is very high. In addition, very few contacts or brief stays in the hospital per patient, could also explain the lack of registered minor children, because of the reduced time frame for doing this among health personnel. However, the mean amount of contacts per patient were five, and hence personnel had several chances to register patients‘ minor children.

Based on our results, it seems safe to conclude that not all health personnel have developed the skills to identify COPMI, and hence that the implementation has not moved beyond paper and procedural implementation for all. One implication of the result that an estimated number of 44% of patients with minor children are not registered with children, is that thousands of COPMI during the 11-year period are unidentified. These children may still be invisible to public services and are at even higher risk of developing social, emotional and mental health problem themselves, since they cannot be reached with effective support and/or interventions. It has taken 11 years to fill in about half of the gap between existing and identified COPMI. New steps are warranted to increase the number of identified children until all COPMI are identified according to the law.

Several changes in the implementation process may contribute to further improvement of the identification of COPMI. In previous studies researchers have pointed to changes in the software of the EPR as a source of strengthening the identification of COPMI (24). One suggested solution was that health personnel should not be able to make entries in EPR unless they had registered if the patient had minor children and had entered the names and birth dates of these children in the front

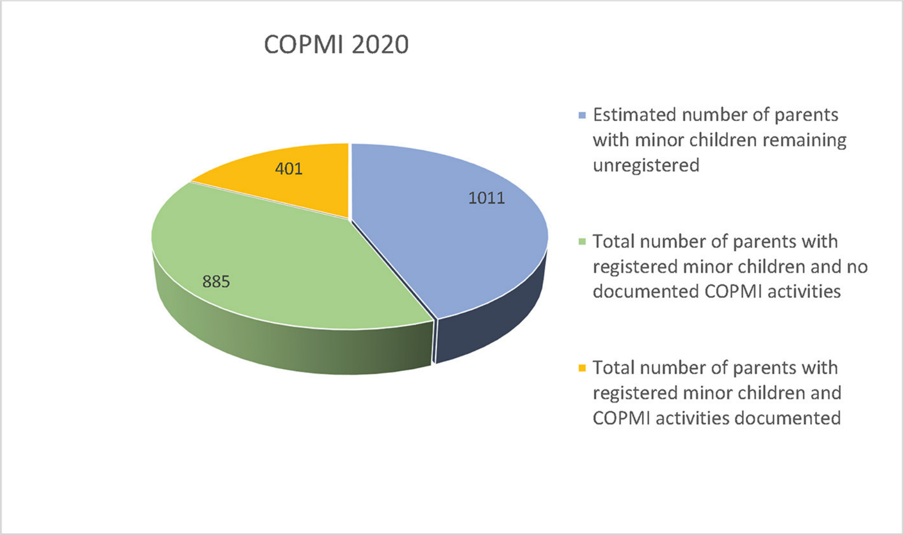


FIGURE 3 | Total number of patients with minor children and patients with registered minor children and one document related to mandated COPMI activities in 2020.

administrative page of EPR. This would automatize identification skills among health personnel and result in full identification of COPMI. Another suggested solution was that patients' minor children were imported directly *via* the link between the EPR and the National Population Register, as for other patient variables such as id number. However, DIPS EPR is a complex software package, and changes like this have never been made.

Researchers have also suggested that the identification of patients' minor children and documenting COPMI related activities in EPR should be included as national quality indicators (13, 25). As such quality indicators also constitute the basis for the funding of the five regional health authorities in the country this could reinforce the adherence to the law and related guidelines for health personnel. The most recent recommendations included the establishment of national, regional and local implementation teams to strengthen the implementation support in all health care institutions (13). Skogøy et al. (13) have characterized the Norwegian process to implement legislation to protect COPMI as diverse and separate dissemination efforts, rather than a coherent implementation strategy (13). It is widely agreed that interventions to change professionals' practice need to be clearly specified (26). A coherent implementation strategy in this context should involve (a) defining the actions to be taken by health personnel, (b) an operationalization of the new practice, and (c) defining the mechanisms that are thought to result in change. In our view, the participating hospital has come a long way in terms of a and b but seem to lack a clear definition of the skills needed to fully implement the new practice as intended in the legislative amendments. According to Fixsen and colleagues' model of degrees of implementation, it is the skills level that represent the active mechanism for change (15). We believe there is still important work to do to define the skills needed in all health personnel in terms of identifying COPMI.

The second aim of the present study was to evaluate to what extent health personnel performed activities or interventions for minor children that was documented in the EPR according to the mandatory guidelines during the years 2010–2020. When a document concerning the patient's minor children is created in the EPR, it shows that a measurable activity related to provision of information and support to COPMI has been documented. The creation of such documents does not inform about the quality of activities, only the fact that it has been created. Therefore, such documents do not represent any form of quality assurance that the child is provided with the support they are entitled to. In order to evaluate that, one would have to enter each document and assess the quantity of the work documented in the reports. However, every patient in the Division for Mental Health and Substance Use Disorders at UNN with identified minor children should have at least one document concerning their minor children in their EPR. Lack of such documents, as the result in this study shows for the large majority of registered children, indicate that the mandatory guideline has not been followed, and hence that the implementation process has not reached the skills level for health personnel in this aspect either.

These finding are not unique, and in a study on the content of conversations with patients who are parents and conversations to support minor children (27), researchers explored data from EPR in 2010–2015. Results showed that very few patients registered with minor children received any type of documented parenting support, and that only a tiny fraction of registered children were included in conversations about their parents' mental health. Along with the results from the present study this clearly shows that the implementation and documentation of COPMI related activities lacks behind the identification of COPMI. After identification, there seem to still be a long way to go before COPMI are systematically offered support. This may be due to the lack of clearness around what this kind of activity should entail. Regardless of the reasons for the gap between registered minor children and the provision of support for these children, identifying COPMI and not offering support is ethically questionable in light of the existing knowledge about transgenerational transmission of mental disorders.

On the positive side, documentation in DIPS EPR is currently being developed so that procedural coding can be used to quantify different clinical activities at UNN. It is The Norwegian Directorate for Health and Social Affairs that are developing the codes, and DIPS implement them into EPR. Examples of such codes are family assessment of patients with minor children, conversations with patients about COPMI, conversations with COPMI, and collaboration with municipal services such as schools, day care centers, public health nurses, child welfare and protection services and so on. A total of 10 codes have been developed for COPMI so far and these activities can be coded in DIPS EPR.

However, even though these codes exist, health personnel are not mandated to use them yet. This means that health personnel may or may not code, and that whether they do or not have no consequences. A practice where health personnel utilize these codes for every patient with minor children will provide information about the quantity of all COPMI related activities in the future. Such practice will thus inform hospitals about to which degree they follow the law and provide children with necessary support and follow up. It is not a necessity that all patients with minor children have reports for each code, because some activities are based on consent from patients. This implies that only one or two coded activities may reflect a clinical practice in accordance with the law, if the patient did not consent for all possible activities. Furthermore, every patient and their children may not need all the same interventions. Many families struggling with mental health issues may have been identified in the health and social services in the communities where they live, and if this is the case many of them may receive support and interventions locally.

Coding registration of patients' minor children and activities related to COPMI in EPR may be experienced as an extra workload for health care personnel. It may however also motivate them. An example of how coding could be used as a motivational tool for COPMI, is how some diagnoses directly provides more funding for some wards. Serious malnutrition is one such diagnosis, and coding this diagnose provide wards with a specific amount of money per patient. To make such coding a mandatory part of the activity-based funding for hospitals seems like a reasonable step to prevent COPMI from being the next generation of mentally ill persons and parents. According to recently published principles and recommendations for working with children and parents living with parental mental illness (28), three take-home messages seem especially important for the adult mental health care services, even in Norway where legislative amendments have been made to protect minor children. These are:

- At intake, identify parenting status including pregnancy.
- Engage with clients in their parenting role and responsibilities.
- Engage with clients' children to identify, and respond to, their needs and/or initiate and coordinate agency referrals for children.

It seems that the participating hospital in this study has come a long way in terms of identification of parenting status, whereas engaging with parents about parenting issues and providing support for the children and families still needs to be developed.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

CR and EJ have contributed to the conceptualization of the study design. EJ, TS, and CR have retrieved and analysed the data. CR have put the paper in writing. CL, EJ, and KK contributed to commenting on and improving the manuscript text. All authors have approved of the final version of the paper.

REFERENCES

1. Thanhäuser M, Lemmer G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. *Curr Opin Psychiatry.* (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342
2. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness: a necessity to provide relevant support. *Front Psychiatry.* (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
3. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *AeJAMH Austral J Advanc. Mental Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
4. Morawska A, Winter L, Sanders MR. Parenting knowledge and its role in the prediction of dysfunctional parenting and disruptive child behaviour. *Child Care Health Dev.* (2009) 35:217–26. doi: 10.1111/j.1365-2214.2008.00929.x
5. Cicchetti D, Rogosch FA, Toth SL. Maternal depressive disorder and contextual risk: contributions to the development of attachment insecurity and behavior problems in toddlerhood. *Dev Psychopathol.* (1998) 10:283–300. doi: 10.1017/S0954579498001618
6. Hipwell AE, Goossens FA, Melhuish EC, Kumar R. *Severe Maternal Psychopathology and Infant–Mother Attachment*. New York, NY: Development and Psychopathology (2000). p. 157–75. doi: 10.1017/S0954579400002030

7. Christiansen H, Reck C, Zietlow A-L, Otto K, Steinmayr R, Wirthwein L, et al. Children of Mentally Ill Parents at Risk Evaluation (COMPARE): design and methods of a randomized controlled multicenter study-part I. *Front Psychiatry.* (2019) 10:128. doi: 10.3389/fpsyt.2019.00128
8. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry.* (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
9. Torvik FA, Rognmo K. *Barn av foreldre med psykiske lidelser eller alkoholmisbruk: Omfang og konsekvenser*. Rapport 4. Oslo: Folkehelseinstituttet (2011).
10. Lilleng S, Ose SO, Hjort H, Bremnes R, Pettersen I, Kalseth J. *Polikliniske pasienter i psykisk helsevern for voksne 2008*. Rapport 3. Oslo: SINTEF (2009).
11. Lauritzen C, Reedtz C, Rognmo K, Nilsen MA, Walstad A. Identification of and support for children of mentally ill parents: a 5 year follow-up study of adult mental health services. *Front Psychiatry.* (2018) 9:507. doi: 10.3389/fpsyt.2018.00507
12. Lauritzen C, Reedtz C, Van Doesum KT, Martinussen M. Factors that may facilitate or hinder a family-focus in the treatment of parents with a mental illness. *J Child Fam Stud.* (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
13. Skogøy BE, Sørgaard K, Maybery D, Ruud T, Stavnes K, Kufås E, et al. Hospitals implementing changes in law to protect children of ill parents: a cross-sectional study. *BMC Health Serv Res.* (2018) 18:609. doi: 10.1186/s12913-018-3393-2
14. Ministry of Health and Care Services. *Ot.prp. nr. 84. Om lov om endringar i helsepersonell loven m.m. (oppfølging av born som pårørande) (About changes in the Health Personnel Act, children as next of kin)*. Oslo: Ministry of Health and Care Services (2008–2009). p. 44.
15. Fixsen DL, Naoom SF, Blasé KA, Friedman RM, Wallace F. *Implementation Research: A Synthesis of the Literature*. Tampa, FL: University of South Florida (2005).
16. Chaudoir SR, Dugan AG, Barr CHI. Measuring factors affecting implementation of health innovations: a systematic review of structural, organizational, provider, patient, and innovation level measures. *Implement Sci.* (2013) 8:22. doi: 10.1186/1748-5908-8-22
17. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatry Mental Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
18. Fakha A, Groenvynck L, de Boer B, van Achterberg T, Hamers J, Verbeek H. A myriad of factors influencing the implementation of transitional care innovations: a scoping review. *Implement Sci.* (2021) 16:21. doi: 10.1186/s13012-021-01087-2
19. Rasmussen LMP, Patras J, Neumer SP, Adolfsen F, Martinsen KD, Holen S, et al. Facilitators and barriers to the implementation of EMOTION: an indicated intervention for young schoolchildren. *Scand J Educ Res.* (2020) 64:728–43. doi: 10.1080/00313831.2019.1596976
20. Reedtz C, Lauritzen C, van Doesum KT. Evaluating workforce developments to support children of mentally ill parents: implementing new interventions in the adult mental healthcare in Northern Norway. *BMJ Open.* (2012) 2:e000709. doi: 10.1136/bmjopen-2011-000709
21. Maybery D, Reupert AE. workforce capacity to respond to children whose parents have a mental illness. *Austral New Zeal J Psychiatry.* (2006) 40:657–64. doi: 10.1080/j.1440-1614.2006.01865.x
22. Skogøy BE, Ogden T, Weimand B, Ruud T, Sørgaard K, Maybery D. Predictors of family focused practice: organisation, profession, or the role as child responsible personnel? *BMC Health Serv Res.* (2019) 19:793. doi: 10.1186/s12913-019-4553-8
23. Lauritzen C, Reedtz C. Child responsible personnel in adult mental health services. *Int J Ment Health Syst.* (2016) 10:64. doi: 10.1186/s13033-016-0098-y
24. Aalen OO. *Innføring i statistikk med medisinske eksempler*. Oslo: Ad Notam Gyldendal (1998).
25. Reedtz C, Mørch L, Lauritzen C. *Registreres psykiatriske pasienters barn i elektronisk pasientjournal.* Nordisk: Sygeplejeforskning (2015) p. 36–45. doi: 10.18261/ISSN1892-2686-2015-01-04
26. Kolehmainen N, Francis JJ. Specifying content and mechanisms of change in interventions to change professionals' practice: an illustration from the Good Goals study in occupational therapy. *Implement Sci.* (2012) 7:100. doi: 10.1186/1748-5908-7-100
27. Kristensen KB, Lauritzen C, Reedtz C. (2022). Support for Children of Parents with Mental Illness. An Analysis of Patients' Health Records. (in press).
28. Reupert AE, Bee P, Hosman CM, van Doesum K, Drost LM, Falkov A, et al. Editorial perspective: prato Research Collaborative for change in parent and child mental health–principles and recommendations for working with children and parents living with parental mental illness. *J Child Psychol Psychiatry.* (2021). doi: 10.1111/jcpp.13521

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

PERSPECTIVE
published: 18 January 2022
doi: 10.3389/fpsyt.2021.761889

A Sustainability Model for Family-Focused Practice in Adult Mental Health Services

Becca Allchin[1,2], Bente M. Weimand[3,4], Brendan O'Hanlon[5] and Melinda Goodyear[1,6]*

[1] School of Rural Health, Monash University, Melbourne, VIC, Australia, [2] Eastern Health, Mental Health Program, Melbourne, VIC, Australia, [3] Division Mental Health Services, Akershus University Hospital, Lørenskog, Norway, [4] Faculty of Health and Social Sciences, Department of Health, Social and Welfare Studies, University of South-Eastern Norway, Drammen, Norway, [5] The Bouverie Centre, La Trobe University, Melbourne, VIC, Australia, [6] Emerging Minds, Hilton, SA, Australia

Background: Translating evidence-based practice to routine care is known to take significant time and effort. While many evidenced-based family-focused practices have been developed and piloted in the last 30 years, there is little evidence of sustained practice in Adult Mental Health Services. Moreover, many barriers have been identified at both the practitioner and organizational level, however sustainability of practice change is little understood. What is clear, is that sustained use of a new practice is dependent on more than individual practitioners' practice.

Design and Method: Drawing on research on sustaining Let's Talk about Children in adult mental health services and in the field of implementation science, this article proposes a model for sustaining family focused practice in adult mental health services.

Sustainability Model for Family-Focused Practice: An operational model developed from key elements for sustaining Let's Talk about Children identifies six action points for Adult Mental Health Services and their contexts to support the sustainability of family-focused practices. The model aims to support Services to take action in the complexity of real-world sustainability, providing action points for engaging with service users and practitioners, aligning intra-organizational activities, and the wider context.

Conclusion: The model for sustaining family-focused practice draws attention to the importance of sustainability in this field. It provides a practical framework for program developers, implementers, adult mental health services and policy-makers to consider both the components that support the sustainability and their interconnection. The model could be built on to develop implementation guides and measures to support its application.

Keywords: sustainability, family-focused practice, mental health promotion, parents, mental ill-health, Let's Talk about Children intervention

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
Lynsey Gregg,
The University of Manchester,
United Kingdom
Stephanie Best,
Macquarie University, Australia
Alexandra Harrison,
Harvard University, United States

****Correspondence:***
Becca Allchin
rebecca.allchin@monash.edu

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *20 August 2021*
Accepted: *21 December 2021*
Published: *18 January 2022*

Citation:
Allchin B, Weimand BM, O'Hanlon B and Goodyear M (2022) A Sustainability Model for Family-Focused Practice in Adult Mental Health Services.
Front. Psychiatry 12:761889.
doi: 10.3389/fpsyt.2021.761889

INTRODUCTION

Research in the past 30 years has explored the impact of a parent's mental ill-health on family life, raising awareness of the importance of family-focused practices for parents and their children (1–6). Such work in mental health services identifies a dual focus (i) improving the outcome for the person with the mental illness and (ii) reducing distress in family members while building their resilience and well-being (7–9). In Adult Mental Health Services (AMHS), family-focused practices encompass approaches, programs, interventions, models and frameworks that acknowledge the whole family context of the person receiving services (2, 10). These take into account the relational nature of recovery and therefore attend to the person's parenting role and family relationships and provide support to the parent in the context of their children and family, while also attend to the intergenerational mental health needs (10–12). Components of effective interventions include psychoeducation directed at both parents and children, adapting parenting behavior through increasing parent agency and skill building, and improving family communication particularly about mental illness (13).

There is now established evidence that these family-focused practices have an impact on supporting the parent in their parenting role and their mental health recovery (10, 14–17) and on protecting children and promoting their resilience (18–21). There are now many evidence-based family-focused practices or programs and documentation of ongoing delivery of programs (22–24). There is, however, little evidence of the use family-focused practices in routine care within AMHS (25–30).

To understand the lack of use of evidence-based family-focused practice in AMHS, research efforts have explored barriers at the practitioner and organization level. Inadequate family-focused training has been identified at the practitioner level, as has a lack of the necessary knowledge, skills and confidence in family-focused practice, limiting their ability to identify and support the parenting role of their clients while also holding their clients' children in mind (31–39). These barriers are reinforced by organizational contexts that do not routinely identify their client's parental status (29, 40–42) and are funded to work with individuals within a biomedical professional-centered approach that is focused on treatment in acute episodic care (10, 11, 20, 43). The formalized, centralized organizational structures common in AMHS are also known to foster the continuation of existing cultures, making innovation and change more difficult (44). These shape the work and the workforce to make it difficult to prioritize working with whole families with the preventive and early intervention approach inherent in family-focused practices in under-funded settings (2, 43, 45). Additionally, a lack of government and organizational structures such as policies and directives, create an authorizing void for the promotion of family-focused practices and impede leadership support for translating such practices into practitioner's everyday work (45–48).

In recent years, greater attention to the process of implementing family-focused practices has resulted in developments to address these barriers. These include practice guidelines and frameworks for family-focused practice in AMHS (19, 49, 50), integrated training, implementation and research programs (51–53) and international collaboration supporting the integration of policy and research (54–56). While these significantly contribute to the understanding of what is needed to sustain family-focused practice in AMHS, there is a need to draw this knowledge together to consider the multiple components in combination to assist AMHS to implement and sustain family-focused practice. This article proposes a model for sustaining family-focused practice in AMHS.

DESIGN AND METHOD

The barriers to family-focused practice noted above illustrate the multi layered factors that impact sustainability and show it to be intimately linked with implementation. While sustainability is focused on the degree to which the intervention can continue to deliver its planned benefits, it relies on practitioners who are able to faithfully deliver it, who in turn need support from their organizations to enable them to deliver its core elements (57).

The field of implementation science studies strategies and structures to support implementation of research into practice and has developed a growing body of frameworks, models and theories (58, 59). It has been posited, however, that much of the work developed in implementation science is used to support other researchers but is not yet common knowledge within the practice world (60). Acknowledging healthcare settings as complex entities, has additionally led to a call for integrating complexity science with implementation science to enable a more dynamic approach to implementation research and practice that fits the reality of change in healthcare setting (61–63).

Sustaining family-focused practice is the work of the healthcare setting. While researchers, purveyors or innovators may develop, trial, pilot, or even implement a family-focused practice, the ongoing work of sustainability is dependent on those within the healthcare setting making the ongoing adjustments necessary for the practice to be ongoingly delivered (57, 64, 65). Equipping healthcare services to apply implementation science knowledge could assist them with evidence-based strategies for applying the necessary adjustments locally. This, however, requires the development and application of implementation tools, described by Westerlund et al. (60) as user- or practice-friendly tools, that are suitable for the context and flexible and able to be adapted to fit settings.

A model is an intentional simplification that can provide an accessible description to guide an implementation process or investigation and so can be applying theory to practice (58). Building on what is known about practitioner and organizational barriers to family-focused practice and frameworks from implementation science, this article proposes a model for actions to support the sustaining a family-focused practice in AMHS.

The model is drawn from a series of five mixed method studies exploring the sustainability of the family-focused practice, Let's Talk about Children (LTC) in eight AMHS in Victoria, Australia, involved in a RCT of LTC (52). The series of studies documented practitioner use and organizational capacity in the eight AMHS and developed an explanatory model of factors enabling sustainability in one AMHS (45, 66–69). The research series used a participatory research approach working in partnership with change agents within AMHS across Victoria. This helped to ground the model in practice wisdom and supporting it to be what Westerlund et al. (60) describes as

TABLE 1 | Key elements influencing sustainability of LTC.

Cluster of influences	Influencing elements
Parent	Parent identification data Parent trust/connection with practitioner
Practitioner	Access to parents on caseload Adapt LTC to parent-consumers needs and working model of team Use of practice support where available Team's workload Characteristics (gender, profession, prior experience) Practitioner's use of LTC monitored Practitioner connection with parent
Organizational	Organizational ownership of implementation Senior leadership communicating priority Middle management enabling fit to everyday work Feedback loops connecting data collections Organizational structures • Allocation system accounting for parenting role • Practitioner training and support infrastructure • Data collection systems -parent numbers, trained practitioners, practitioner's application post training • Reporting systems that consider parent, child and family well-being • Organizational adjustments to fit LTC
Wider Context	Introduction of recovery-oriented policy Parent, child and family focused Mental Health Act Government funded family-focused service development positions in AMHS
Implementation context	Research trial with trusted organizations Supported localized implementation Internal implementers within AMHS Parallel innovations - free online training and resources package

"practice-friendly." The outcomes of these studies were clustered deductively using sustainability and implementation models and frameworks (65, 70, 71). Five clusters of key elements were identified as influencing LTC's sustainability (69). These clusters related to (1) the parent, (2) the practitioner, (3) the organization, (4) the wider context and (5) the implementation context (see **Table 1**). While these elements can be considered individually, the studies' outcomes highlight the intersectionality between these elements as an important contributor to sustainability.

For example, a parent cannot be offered the family-focused practice if the practitioner allocated to them is not equipped with the skill and confidence to use it. Without a system to identify clients as parents, skilled practitioners may not be allocated parents. A skilled practitioner will find it difficult to maintain confidence if they are only rarely allocated a parent. Without a monitoring system, there will be no way of knowing if a practitioner is applying their skills, and if parents are being offered the family-focused practice to know if is being sustained. Additionally, without monitoring there is nothing to inform decision making and provide input for troubleshooting difficulties. If the wider systems do not fund AMHS to work with families or prioritize preventative mental health, an organization may find it difficult to integrate the family-focused practice into their model of care.

Conversely, a training program does not ensure sustainability, as trained practitioners may not be able to implement their new skills in practice. A system for identifying the parental status of clients will, in itself, not ensure that they are allocated for their care to trained practitioners, or have practitioners who are endorsed with the time and scope to use their skills. Data collected without feedback loops to adjust implementation cannot inform policy, training, support and allocation structures. These are each part of the picture of sustainability but on their own will not enable sustainability. They are required to be applied in combination.

SUSTAINABILITY MODEL FOR FAMILY-FOCUSED PRACTICE

Working from these known key elements influencing sustainability of the family-focused practice of LTC, the following model was developed to operationalize the action points for AMHS and their external contexts to support family focused practice practice more broadly (See **Figure 1**: Sustainability model for family-focused practice). Framed in outcomes focused language to help operationalize action and reflecting the interconnecting nature of the elements, the model identifies six points of meso (intra organizational) and macro (broader contextual) level action, each incorporating multiple elements. Designed as an intentional simplification for a practical purpose, this model aims to support AMHS to hold in mind the complexity of sustainability and the requirement of simultaneous actions while providing actionable starting places. The first two actions points relate to how the AMHS engages with its service users and their practitioners. The next three action points focus on internal organizational activities important for implementation and sustainability. The last action point articulates important actions in the wider context.

Recognize, Allocate, and Measure Outcomes for Parents, Children, and Families

Recognition of a client's parental status can allow for service delivery to be tailored to address their, their children's and their family's needs. Knowledge of prevalence of parenting amongst the organization's clients can be used to drive the number and location of skilled practitioners needed to adequately enable parents, children and families to access family-focused practice. Organizations can support parents by allocating them to practitioners with the skills and confidence to deliver family-focused practice. Flexible allocation systems that can attend to the match between parent and practitioner readiness can support the therapeutic alliance and enable family-focused practice to be delivered. Recognition of parenting status also can support the organization's capacity to apply appropriate measures that assist them in monitoring both what services are delivered and if they give the expected benefits for parents, children and families.

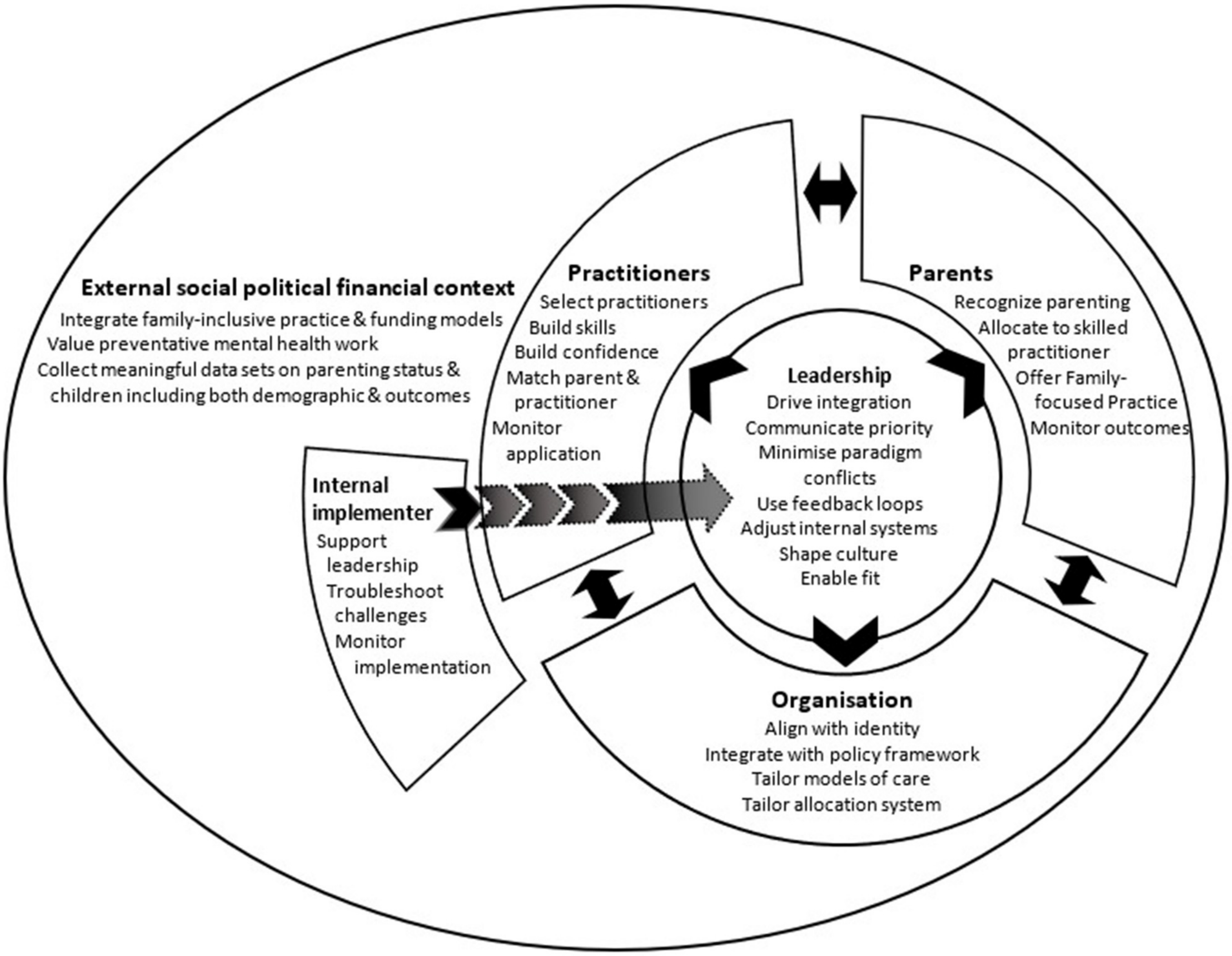


FIGURE 1 | Sustainability model for family-focused practice.

Select, Support, and Monitor Practitioners

The selection of practitioners needs to take into account factors such as access to parents on their caseload, the practitioners' skills and knowledge of the impact of mental illness on parents, children and families, as well as their ability to hold a dual perspective while working with an individual. Building practitioners' skills and confidence to use family-focused practice requires flexible practice support that facilitates their capacity to reflect on and monitor their own practice against expected outcomes. Such support structures need to be co-developed so as to be tailored to fit practitioners' specific needs. Developing systems to monitor practitioners' application of family-focused practice provides a feedback loop that can help to identify support needs, communicate priority and address fidelity issues.

Integrate Within Organization Identity and Structures

Aligning family-focused practice within an organization's identity and integrating it into policy structures, can enable models of care to be tailored to fit family-focused practice, and support the incorporation of its core competencies into position descriptions and recruitment processes. Embedding family-focused practice into organizational policy also supports the development of infrastructure to enable practice, such as practitioner training, support and monitoring systems, and parent recognition and allocation systems. Organizational policy can additionally provide an anchor for family-focused practice in times of personnel or structural change that can facilitate its continued use. Furthermore, integrating outcome measures and reporting structures that incorporate whole-of-family well-being can help to reinforce a preventative mental health focus that is foundational to family-focused practice.

Leadership to Drive Sustainability

Organizational ownership is needed to support the internal adjustments required for the integration and sustainability of family-focused practice in AMHS. Adjustments to complex, internal structures need whole-of-organization commitment that requires leadership at multiple levels within the organization. At a higher level this includes communicating this work as a priority, developing training and support infrastructure, creating feedback loops and reporting systems. At the level of middle management this includes building cultures that promote recovery-focused family-inclusive mental health practice, facilitating the translation of family-focused practice into everyday practice and utilizing the feedback loops to support practice. Held together, the multiple levels of leadership and the structures they provide can help to minimize paradigm conflicts that exist for family-focused practice in AMHS.

Local Support for Implementation and Sustainability

Having an internal implementer to support leadership in the implementation process can help support sustainability. The presence of the internal implementer can be an anchor to the priority of the work and provide resources for leadership to build practitioners' skills and confidence. Working with leadership, they can assist in monitoring implementation through feedback loops that can enable ongoing adaptation of implementation processes to support sustainability.

Incorporate Family-Inclusive Preventative Mental Health Care in the Wider Context

Incorporating a family-inclusive, preventative lens within the funding and political context within which AMHS operates, creates a foundation for sustaining family focused practice. Integrating these lenses into recovery-focused mental health practice can support shifts in the funding models from an individual to whole-of-family perspective and the valuing of preventative mental health work that underpins family-focused practice. These shifts create an authorizing environment for AMHS leadership to give priority for delivering family-focus practice and the integration of family-focused practice into AMHS models of practice. These shifts also reinforce the need for reporting measures that account for parent, children and family outcomes and that emphasize resilience and well-being rather than risk.

IMPLICATIONS/APPLICATION

This model for sustaining family-focused practice in AMHS provides points of action for AMHS and their external contexts. The model extends existing peer reviewed work that identifies barriers and facilitators of implementation and models that explain sustainability, through drawing these together to provide actionable points of focus for those within an adult mental health system. It is intended to provide a practical framework for integrating the evidence in implementation science as applied to family-focused practice. The model is envisioned to be a tool for program developers, implementers, AMHS and policy-makers to consider both the components that support the sustainability and their interconnection.

As noted here, there is a need for ongoing attention to the complexity and importance of sustainability in the field of parents with mental ill-health, and their children and families. As AMHS are complex and changing entities, ongoing attention to the interconnection between practice, and the organisation's capacity to support practice, is required to enable continued quality of care. Sustaining family-focused practice, shifts the focus from the program, innovation or practice being implemented, to the mechanisms that enable them to be able to be utilized beyond the focused implementation or research trial. As sustainability happens within the work of the health service, equipping AMHS to not only implement but also sustain family-focused practice is pivotal for the field to promote better outcomes for parents, children and families.

This model goes some way to assist this process by identifying points of actions for AMHS and their external contexts, that are articulated as part of a whole, in order to address the complexity and work toward sustainability.

Further work is required to develop practice-friendly tools to support the application of this model. Practical implementation guides could operationalize each of the points of action. Monitoring and measuring tools could provide feedback loops on sustainability for AMHS. Coproduction of these application tools would support their usability by AMHS for their specific contexts. Additionally, this model provides a framework for developers of innovations, practices or interventions to build practice-friendly tools to support their sustained use in AMHS.

CONCLUSION

The model showcases the importance of actions that need operationalization at the organizational and wider context level to be able to influence the multiple systems involved in creating sustained family focus practice. This level of complexity can be overwhelming and difficult for program developers, implementers, AMHS and policy-makers to hold in mind, leading to a focus on the actions or elements in isolation. The model, however, highlights the inadequacy of an isolated view of actions or elements if the aim is to build sustainability at the local level that fit their context.

AUTHOR'S NOTE

The content in this manuscript has been adapted from BA's thesis available online https://doi.org/10.26180/14214686.v1.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

BA conceived the article, developed the concept of the sustainability model, and drafted the article. MG, BO'H, and BW contributed to the interpretation of the analysis, the refining of the manuscript, reviewed drafts, and contributed to the write up. All authors were contributors to each of the five studies that underpin this work. All contributed to the analysis and drawing together of their combined outcomes and refining the sustainability model.

FUNDING

An Australian Government Research Training Program Scholarship supported BA in her doctorate of philosophy, during which she developed the body of work underpinning the model.

ACKNOWLEDGMENTS

The multiphase participatory doctorate of philosophy research was additionally supported by an advisory group giving invaluable input into the design and applicability. Membership comprised of Cheree Cosgriff (FaPMI coordinator and internal implementer), Georgia Cripps (FaPMI coordinator with Let's Talk practitioner experience), Helen Fernandes (Participatory research practitioner), Jane Shamrock (Qualitative Participatory Research Academic) and Brad Wynne (AMHS manager).

REFERENCES

1. Rutter M, Quinton D. Parental psychiatric disorder: effects on children. *Psychol Med.* (1984) 14:853–80. doi: 10.1017/S0033291700019838
2. Maybery DJ, Foster K, Goodyear MJ, Grant A, Tungpunkom P, SkogØy BE, et al. How can we make the psychiatric workforce more family focused? In: Reupert AE, Maybery DJ, Nicholson J, Gopfert M, Seeman MV, editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. 3rd ed. New York, NY: New York Cambridge University Press (2015). p. 301–11.
3. Nicholson J, Biebel K, Hinden B, Henry A, Stier L. *Critical Issues for Parents with Mental Illness and their Families*. Rockville, MD: Substance Abuse and Mental Health Services Administration Center for Mental Health Services (2001).
4. Gopfert M, Webster J, Seeman MV. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. 1st ed. New York, NY: Cambridge University Press (1996).
5. Ballal D, Navaneetham J. Talking to children about parental mental illness: the experiences of well parents. *Int J Soc Psychiatry.* (2018) 64:367–73. doi: 10.1177/0020764018763687
6. Brockington I, Chandra P, Dubowitz H, Jones D, Moussa S, Nakku J, et al. WPA guidance on the protection and promotion of mental health in children of persons with severe mental disorders. *World Psychiatry.* (2011) 10:93–102. doi: 10.1002/j.2051-5545.2011.tb00023.x
7. Dixon L, McFarlane WR, Lefley H, Lucksted A, Cohen M, Falloon I, et al. Evidence-based practices for services to families of people with psychiatric disabilities. *Psychiatr Serv.* (2001) 52:903–10. doi: 10.1176/appi.ps.52.7.903
8. Mottaghipour Y, Bickerton A. The pyramid of family care: a framework for family involvement with adult mental health services. *Aust E J Adv Mental Health.* (2005) 4:210–7. doi: 10.5172/jamh.4.3.210
9. Wyder M, Bland R. The recovery framework as a way of understanding families' responses to mental illness: balancing different needs and recovery journeys. *Aust Soc Work.* (2014) 67:179–96. doi: 10.1080/0312407X.2013.875580
10. Foster KN, Maybery DJ, Reupert AE, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child Youth Serv.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
11. Price-Robertson R, Obradovic A, Morgan B. Relational recovery: beyond individualism in the recovery approach. *Adv Ment Health.* (2016) 15:108–20. doi: 10.1080/18387357.2016.1243014
12. Price-Robertson R, Olsen G, Francis H, Obradovic A, Morgan B. *Supporting Recovery in Families Affected by Parental Mental Illness* (CFCA Practitioner Resource). Melbourne, VIC: Child Family Community Australia information exchange, Australian Institute of Family Studies (2016). Available online at: https://aifs.gov.au/cfca/publications/supporting-recovery-families-affected-parental-mental-illness
13. Marston N, Stavnes K, Van Loon LMA, Drost LM, Maybery DJ, Mosek A, et al. A content analysis of intervention key elements and assessments (IKEA): what's in the black box in the interventions directed to families where a parent has a mental illness? *Child Youth Serv.* (2016) 37:112–28. doi: 10.1080/0145935X.2016.1104041
14. Awram R, Hancock N, Honey A. Balancing mothering and mental health recovery: the voices of mothers living with mental illness. *Adv Ment Health.* (2017) 15:147–60. doi: 10.1080/18387357.2016.1255149
15. Beardslee WR, Wright E, Gladstone T, Forbes P. Long-term effects from a randomized trial of two public health preventive interventions for parental depression. *J Fam Psychol.* (2007) 21:703. doi: 10.1037/0893-3200.21.4.703
16. McKay EA. Mothers with mental illness: an occupation interrupted. In: Esdaile SA, Olson JA, editors. *Mothering Occupations: Challenge, Agency, and Participation*. Philadelphia: FA Davis Company (2004). p. 238–58.
17. Reupert AE, Maybery DJ, Kowalenko NM. Children whose parents have a mental illness: prevalence, need and treatment. *Med J Aust Open.* (2012) 1:7–9. doi: 10.5694/mjao11.11200
18. Goodyear MJ, Obradovic A, Allchin B, Cuff R, McCormick F, Cosgriff C. Building capacity for cross-sectorial approaches to the care of families where a parent has a mental illness. *Adv Ment Health.* (2015) 13:153–64. doi: 10.1080/18387357.2015.1063972
19. Goodyear MJ, Hill T-L, Allchin B, McCormick F, Hine RH, Cuff R, et al. Standards of practice for the adult mental health workforce: Meeting the needs of families where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120
20. Maybery DJ, Reupert AE. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
21. Siegenthaler E, Munder T, Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17. doi: 10.1016/j.jaac.2011.10.018
22. Rimehaug T. The ecology of sustainable implementation. *Z Psychol.* (2014) 222:58–66. doi: 10.1027/2151-2604/a000166
23. Cooklin A, Bishop P, Francis D, Fagin L, Asen E. *The Kidstime Workshops. A Multi-Family Social Intervention for the Effects of Parental Mental Illness. Manual*. London: CAMHS Press (2012).
24. Wolpert M, Hoffman J, Martin A, Fagin L, Cooklin A. An exploration of the experience of attending the Kidstime programme for children with parents with enduring mental health issues: parents' and young people's views. *Clin Child Psychol Psychiatry.* (2015) 20:406–18. doi: 10.1177/1359104514520759
25. Leenman G, Arblaster K. Navigating rocky terrain: a thematic analysis of mental health clinician experiences of family-focused practice. *J Mental Health Train Educ Pract.* (2020) 15:71–83. doi: 10.1108/JMHTEP-04-2019-0022
26. Doesum K, Maia T, Pereira C, Loureiro M, Marau J, Toscano L, et al. The impact of the "semente" program on the family-focused practice of mental health professionals in Portugal. *Front Psychiatry.* (2019) 10:305. doi: 10.3389/fpsyt.2019.00305
27. Lauritzen C, Reedtz C, Rognmo K, Nilsen MA, Walstad A. Identification of and support for children of mentally ill parents: a 5 year follow-up study of adult mental health services. *Front Psychiatry.* (2018) 9:507. doi: 10.3389/fpsyt.2018.00507
28. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness: a necessity to provide relevant support. *Front Psychiatry.* (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
29. Ruud T, Maybery D, Reupert A, Weimand B, Foster K, Grant A, et al. Adult mental health outpatients who have minor children: prevalence of parents, referrals of their children, and patient characteristics. *Front Psychiatry.* (2019) 10:163. doi: 10.3389/fpsyt.2019.00163

30. Potijk MR, Drost LM, Havinga PJ, Hartman CA, Schoevers RA. "…and how are the kids?" Psychoeducation for adult patients with depressive and/or anxiety disorders: a pilot study. *Front Psychiatry*. (2019) 10:4. doi: 10.3389/fpsyt.2019.00004
31. Jessop ME, De Bondt N. A consultation service for adult mental health service clients who are parents and their families. *Adv Ment Health.* (2012) 10:149–56. doi: 10.5172/jamh.2011.10.2.149
32. Goodyear MJ, Maybery DJ, Reupert AE, Allchin R, Fraser C, Fernbacher S, et al. Thinking families: a study of the characteristics of the workforce that delivers family-focussed practice. *Int J Ment Health Nurs.* (2017) 26:238–48. doi: 10.1111/inm.12293
33. Grant A, Reupert A, Maybery D, Goodyear M. Predictors and enablers of mental health nurses' family-focused practice. *Int J Ment Health Nurs.* (2019) 28:140–51. doi: 10.1111/inm.12503
34. Tchernegovski P, Hine R, Reupert AE, Maybery DJ. Adult mental health clinicians' perspectives of parents with a mental illness and their children: single and dual focus approaches. *BMC Health Serv Res.* (2018) 18:611. doi: 10.1186/s12913-018-3428-8
35. Maybery DJ, Goodyear MJ, Reupert AE, Grant A. Worker, workplace or families: what influences family focused practices in adult mental health? *J Psychiatr Ment Health Nurs.* (2016) 23:163–71. doi: 10.1111/jpm.12294
36. Skogøy BE, Ogden T, Weimand B, Ruud T, Sørgaard K, Maybery D. Predictors of family focused practice: organisation, profession, or the role as child responsible personnel? *BMC Health Serv Res.* (2019) 19:1–13. doi: 10.1186/s12913-019-4553-8
37. Maybery DJ, Goodyear MJ, O'Hanlon B, Cuff R, Reupert AE. Profession differences in family focused practice in the adult mental health system. *Fam Process.* (2014) 53:608–17. doi: 10.1111/famp.12082
38. Grant A, Goodyear MJ, Maybery DJ, Reupert AE. Differences between Irish and Australian psychiatric nurses' family-focused practice in adult mental health services. *Arch Psychiatr Nurs.* (2016) 30:132–7. doi: 10.1016/j.apnu.2015.07.005
39. Gregg L, Adderley H, Calam R, Wittkowski A. The implementation of family-focused practice in adult mental health services: a systematic review exploring the influence of practitioner and workplace factors. *Int J Ment Health Nurs.* (2021) 30:885–906. doi: 10.1111/inm.12837
40. Maybery DJ, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry*. (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
41. Ballal D, Navaneetham J, Chandra PS. Children of parents with mental illness: the need for family focussed interventions in India. *Indian J Psychol Med.* (2019) 41:228–34. doi: 10.4103/IJPSYM.IJPSYM_430_18
42. Dunn A, Startup H, Cartwright-Hatton S. Adult mental health service engagement with patients who are parents: Evidence from 15 English mental health trusts. *Br J Clin Psychol.* (2022). doi: 10.1111/bjc.12330
43. Isobel S, Allchin B, Goodyear M, Gladstone BM. A narrative inquiry into global systems change to support families when a parent has a mental illness. *Front Psychiatry*. (2019) 10:310. doi: 10.3389/fpsyt.2019.00310
44. Damanpour F. Organizational innovation: a meta-analysis of effects of determinants and moderators. *Acad Manage J.* (1991) 34:555–90. doi: 10.5465/256406
45. Allchin B, Goodyear MJ, O'Hanlon B, Weimand BM. Leadership perspectives on key elements influencing implementing a family-focused intervention in mental health services. *J Psychiatr Ment Health Nurs.* (2020) 27:616–27. doi: 10.1111/jpm.12615
46. Grant A, Reupert AE. The impact of organizational factors and government policy on psychiatric nurses' family-focused practice with parents who have mental illness, their dependent children, and families in Ireland. *J Fam Nurs.* (2016) 22:199–223. doi: 10.1177/1074840716643770
47. Biebel K, Nicholson J, Williams V, Hinden BR. The responsiveness of state mental health authorities to parents with mental illness. *Adm Policy Ment Health.* (2004) 32:31–48. doi: 10.1023/B:APIH.0000039661.54974.ce
48. Hinden B, Wolf T, Biebel K, Nicholson J. Supporting clubhouse members in their role as parents: necessary conditions for policy and practice initiatives. *Psychiatr Rehabil J Fall.* (2009) 33:98–105. doi: 10.2975/33.2.2009.98.105
49. Foster KN, Goodyear MJ, Grant A, Weimand B, Nicholson J. Family-focused practice with EASE: a practice framework for strengthening recovery when mental health consumers are parents. *Int J Ment Health Nurs.* (2019) 28:351–60. doi: 10.1111/inm.12535
50. Nicholson J, English K, Heyman M. The parenting well learning collaborative feasibility study: training adult mental health service practitioners in a family-focused practice approach. *Community Ment Health J.* (2021). doi: 10.1007/s10597-021-00818-5
51. Solantaus T, Toikka S. The effective family programme: preventative services for the children of mentally ill parents in Finland. *Int J Mental Health Promot.* (2006) 8:37–44. doi: 10.1080/14623730.2006.9721744
52. Maybery DJ, Goodyear MJ, Reupert AE, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry.* (2017) 17:198. doi: 10.1186/s12888-017-1357-4
53. Yates S, Gatsou L. Undertaking family-focused interventions when a parent has a mental illness - possibilities and challenges. *Practice.* (2020) 33:1–16. doi: 10.1080/09503153.2020.1760814
54. Nicholson J, Reupert AE, Maybery DJ, Grant A, Lees R, Mordoch E, et al. The policy context and change for families living with parental mental illness. In: Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman MV, editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. 3rd ed. New York, NY: Cambridge University Press (2015). p. 354–64.
55. Ruud T. Routine outcome measures in Norway: only partly implemented. *Int Rev Psychiatry.* (2015) 27:338–44. doi: 10.3109/09540261.2015.1054268
56. Skogøy BE, Sørgaard K, Maybery D, Ruud T, Stavnes K, Kufås E, et al. Hospitals implementing changes in law to protect children of ill parents: a cross-sectional study. *BMC Health Serv Res.* (2018) 18:1–11. doi: 10.1186/s12913-018-3393-2
57. Stirman SW, Kimberly J, Cook N, Calloway A, Castro F, Charns M. The sustainability of new programs and innovations: a review of the empirical literature and recommendations for future research. *Implement Sci.* (2012) 7:1–19. doi: 10.1186/1748-5908-7-17
58. Nilsen P. Making sense of implementation theories, models and frameworks. *Implement Sci.* (2015) 10:1–13. doi: 10.1186/s13012-015-0242-0
59. Moullin JC, Sabater-Hernández D, Fernandez-Llimos F, Benrimoj SI. A systematic review of implementation frameworks of innovations in healthcare and resulting generic implementation framework. *Health Res Policy Syst.* (2015) 13:11. doi: 10.1186/s12961-015-0005-z
60. Westerlund A, Nilsen P, Sundberg L. Implementation of implementation science knowledge: the research-practice gap paradox. *Worldviews Evid Based Nurs.* (2019) 16:332–4. doi: 10.1111/wvn.12403
61. Greenhalgh T, Papoutsi C. Studying complexity in health services research: desperately seeking an overdue paradigm shift. *BMC Med.* (2018) 16:95. doi: 10.1186/s12916-018-1089-4
62. Braithwaite J, Churruca K, Long JC, Ellis LA, Herkes J. When complexity science meets implementation science: a theoretical and empirical analysis of systems change. *BMC Med.* (2018) 16:63. doi: 10.1186/s12916-018-1057-z
63. Long KM, McDermott F, Meadows GN. Being pragmatic about healthcare complexity: our experiences applying complexity theory and pragmatism to health services research. *BMC Med.* (2018) 16:94. doi: 10.1186/s12916-018-1087-6
64. Dearing JW. Evolution of diffusion and dissemination theory. *J Public Health Manag Pract.* (2008) 14:99–108. doi: 10.1097/01.PHH.0000311886.98627.b7
65. Scheirer MA, Dearing JW. An agenda for research on the sustainability of public health programs. *Am J Public Health.* (2011) 101:2059–67. doi: 10.2105/AJPH.2011.300193
66. Allchin B, O'Hanlon B, Weimand BM, Boyer F, Cripps G, Gill L, et al. An explanatory model of factors enabling sustainability of let's talk in an adult mental health service: a participatory case study. *Int J Ment Health Syst.* (2020) 14:48. doi: 10.1186/s13033-020-00380-9
67. Allchin B, O'Hanlon B, Weimand BM, Goodyear MJ. Practitioners' application of Let's Talk about Children intervention in adult mental health

services. *Int J Ment Health Nurs.* (2020) 29:899–907. doi: 10.1111/inm.12724

68. Allchin B, Weimand BM, O'Hanlon B, Goodyear MJ. Continued capacity: factors of importance for organizations to support continued Let's Talk practice – a mixed-methods study. *Int J Ment Health Nurs.* (2020) 29:1131–43. doi: 10.1111/inm.12754
69. Allchin R. *Exploring the Implementation and Sustainability of Let's Talk About Children – A Model for Family-Focused Practice in Adult Mental Health Services*. Clayton, VIC: Monash University (2020).
70. Damschroder LJ, Lowery JC. Evaluation of a large-scale weight management program using the consolidated framework for implementation research (CFIR). *Implement Sci.* (2013) 8:51. doi: 10.1186/1748-5908-8-51
71. Maher L, Gustafson D, Evans A. *Sustainability Model and Guide*. Coventry: NHS Institute for Innovation and Improvement (2007).

Author Disclaimer: This article represents the authors' original work and has not been submitted for publication elsewhere.

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers
in Psychiatry

ORIGINAL RESEARCH
published: 27 January 2022
doi: 10.3389/fpsyt.2021.787166

A Scoping Review of Interventions Designed to Support Parents With Mental Illness That Would Be Appropriate for Parents With Psychosis

Jessica Radley[1*], *Nithura Sivarajah*[2], *Bettina Moltrecht*[1], *Marie-Louise Klampe*[3], *Felicity Hudson*[4], *Rachel Delahay*[3], *Jane Barlow*[5†] *and Louise C. Johns*[1,2†]

[1] Department of Psychiatry, University of Oxford, Oxford, United Kingdom, [2] Oxford Health NHS Foundation Trust, Oxford, United Kingdom, [3] Department of Experimental Psychology, University of Oxford, Oxford, United Kingdom, [4] School of Psychological Science, Faculty of Life Sciences, University of Bristol, Bristol, United Kingdom, [5] Department of Social Policy and Intervention, University of Oxford, Oxford, United Kingdom

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Lynsey Gregg,
The University of Manchester,
United Kingdom
Jennifer Strand,
University of Gothenburg, Sweden

***Correspondence:**
Jessica Radley
jessica.radley@psych.ox.ac.uk

[†]These authors have contributed equally to this work and share last authorship

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *30 September 2021*
Accepted: *03 December 2021*
Published: *27 January 2022*

Citation:
Radley J, Sivarajah N, Moltrecht B, Klampe M-L, Hudson F, Delahay R, Barlow J and Johns LC (2022) A Scoping Review of Interventions Designed to Support Parents With Mental Illness That Would Be Appropriate for Parents With Psychosis. Front. Psychiatry 12:787166. doi: 10.3389/fpsyt.2021.787166

The experience of psychosis can present additional difficulties for parents, over and above the normal challenges of parenting. Although there is evidence about parenting interventions specifically targeted at parents with affective disorders, anxiety, and borderline personality disorder, there is currently limited evidence for parents with psychotic disorders. It is not yet known what, if any, interventions exist for this population, or what kinds of evaluations have been conducted. To address this, we conducted a scoping review to determine (1) what parenting interventions have been developed for parents with psychosis (either specifically for, or accessible by, this client group), (2) what components these interventions contain, and (3) what kinds of evaluations have been conducted. The eligibility criteria were broad; we included any report of an intervention for parents with a mental health diagnosis, in which parents with psychosis were eligible to take part, that had been published within the last 20 years. Two reviewers screened reports and extracted the data from the included reports. Thirty-eight studies of 34 interventions were included. The findings show that most interventions have been designed either for parents with any mental illness or parents with severe mental illness, and only two interventions were trialed with a group of parents with psychosis. After noting clusters of intervention components, five groups were formed focused on: (1) talking about parental mental illness, (2) improving parenting skills, (3) long-term tailored support for the whole family, (4) groups for parents with mental illness, and (5) family therapy. Twenty-three quantitative evaluations and 13 qualitative evaluations had been conducted but only eight interventions have or are being evaluated using a randomized controlled trial (RCT). More RCTs of these interventions are needed, in addition to further analysis of the components that are the most effective in changing outcomes for both the parent and their children, in order to support parents with psychosis and their families.

Keywords: review, intervention, psychosis, parenting, children, mental health

INTRODUCTION

Parenting can be challenging for parents who experience psychosis. Psychotic symptoms include positive symptoms, such as hallucinations and delusions, and negative symptoms, such as apathy and blunted affect (1). Psychosis has other associated difficulties, including memory and concentration problems, co-morbid affective conditions, difficulties in understanding the mental states of others, and sensitivity to stress and poor sleep (2). Individuals who experience psychosis also often have to cope with side-effects from anti-psychotic medication, particularly sedation (3). These symptoms and side-effects can make it more difficult for parents to empathize with their children and communicate clearly, and to offer the consistent, responsive care required for healthy child development (4–6). A diagnosis of psychosis is also associated with adverse childhood experiences, such as sexual, physical, and emotional abuse (7, 8), which may affect parents forming stable attachments with their own children (9). During an acute episode of psychosis, parents may find it difficult to care for their children at all (10) and family life can be disrupted if the parent is hospitalized (11).

Although not all parents with psychosis experience problems with their parenting, those who report more severe symptoms and a longer duration of illness are more likely to show such problems (12). However, it is not only symptom severity that makes parenting challenging; a diagnosis of psychosis is associated with many environmental factors that can precipitate further difficulties, including being a single parent, (13), poor social support (14), financial instability (15, 16), and unemployment (17). These socioeconomic factors, in turn, are associated with more frequent experiences of psychiatric symptoms (18), and predict a poorer quality of parenting (14). This social adversity may even be more detrimental to parenting than the direct effects of parental mental illness (19).

Intervening with these families could lead to positive outcomes for both the parent and their child. Elements of a successful intervention may include crisis management in anticipation of future relapses (20), links to other services to provide parents with practical support (21), as well as help with parenting skills (22). Custody loss is experienced by parents with serious mental illness more often than parents without mental health problems (23, 24). It is a fear of many of these parents (25), which can mean some parents are reluctant to seek help and take part in parenting interventions (26). Therefore, appropriate interventions should acknowledge the parenting role as an important part of recovery (27, 28), which could then help to prevent custody loss (29), while also reducing the risk of the children developing mental health problems themselves (30). Research with children of parents with mental illness has shown that they want to understand their parent's mental illness (21), and explanation about their parent's illness may be protective for these children (31).

Parenting interventions aim to improve parenting skills and relationships within the family (32) by providing parents with skills focused on encouraging positive behavior and education about child development (33, 34). Parenting interventions often have a focus on parents whose children are demonstrating behavioral difficulties (35) and there is good evidence that they can reduce emotional and behavioral difficulties for these children (34). More recently some of these interventions have been amended to support parents with mental health problems [e.g., (36)] or the intervention has been used in its original form with a group of parents with a mental health diagnosis, like Triple P (37) and Tuning into Kids (38). Parenting interventions that are tailored toward parents with mental health difficulties were initially designed for parents with affective disorders (39), and this client group is still the focus of many such programs (40, 41). Specific programs have, however, also been developed for parents with other types of mental health diagnoses, such as anxiety (42), and personality disorders (43). However, the availability of interventions for parents with psychosis is limited, with the majority focusing on mothers experiencing postpartum psychosis (44), leaving a significant gap with regard to interventions for parents with psychosis who have older children. To address this gap, we need to know which interventions exist, as well as what elements these interventions contain in order to address the needs of families with parental psychosis. Ways in which these needs may be addressed include planning for periods of hospitalization (20) and improving parents' ability to understand their child's mental states (45).

This review is the sequel to a Cochrane systematic review (46) in which a search was undertaken to identify the evidence for parenting interventions designed to improve parenting skills or the parent-child relationship in parents with psychosis. However, only one study was identified, which was published almost 40 years ago. Other similar reviews include Schrank et al. (47) and Suarez et al. (48). Schrank et al. (47) conducted a systematic review of interventions that reported quantitative findings, in which at least 50% of the participants were parents with severe mental illness (which they defined as psychotic or bipolar disorders) and identified 15 interventions. Suarez et al. (48) conducted a scoping review for interventions for mothers with any kind of mental illness that had described some kind of outcome for the study participants, and identified nine interventions.

The aim of this review is to identify what interventions are available for parents with psychosis, to describe the content of these interventions, and provide a narrative synthesis about existing evaluations and what they have found.

Research Questions

1. What parenting interventions have been developed for parents with psychosis (either specifically for, or accessible by, this client group)?
2. What are the components of these interventions?
3. What kinds of evaluations have been conducted to determine their acceptability and effectiveness, and what do the findings show?

METHODS

The current scoping review systematically searched all relevant databases, trial registries and gray literature with the aim of mapping current research about parenting interventions for parents with psychosis. In contrast to Radley et al. (46), Schrank et al. (47), and Suarez et al. (48), it treated as eligible any report of an intervention regardless of the level of evaluation to which it has been subjected. The inclusion criteria were also broader in that any intervention for parents with mental health problems was included. Interventions for parents with specific mental health diagnoses in which parents with psychosis were not eligible to take part were excluded from this review since these interventions may not be appropriately designed to address the needs of parents with psychosis. In order to address the gap that exists around interventions for parents with psychosis with older children, we only included studies in which the children were older than 2 years. This review was also limited to papers published within the last 20 years in order to describe what may be currently available for these parents.

This manuscript is written in accordance with the PRISMA guidance for reporting scoping reviews (49).

Protocol and Registration

The protocol was uploaded to the Open Science Framework (https://osf.io/3d7t9/) in May 2021.

Eligibility Criteria

This review followed the scoping review framework by Arksey and O'Malley (50). It included peer-reviewed papers, trial registries, and gray literature including Ph.D. theses, websites, and preprints. To be included, reports had to be written in the last 20 years and include an evaluation or description of an intervention for parents with a mental health diagnosis, in which parents with psychosis were eligible to take part. The intervention could be child-focused, parent-focused, or family-focused as long as there was a specific component for the parent.

The following were excluded:

- Reviews.
- Interventions designed for the children of parents with a mental health diagnosis with no parenting component.
- Interventions designed to improve service-response or healthcare professional knowledge of parental mental illness with no parenting component.
- Interventions that excluded parents with psychosis.
- Interventions that targeted parents with children under the age of 2 years.

Records were also excluded if they were written in any language apart from English. However, it became clear that a large number of potentially eligible German papers were being excluded. It was decided the review would be incomplete without consideration of these papers, and therefore a German-speaking author, BM, reviewed all of these records at full-text stage.

Information Sources

Eight databases were searched on January 11th 2021, and updated on November 6th 2021, for records published since January 2001 in PsycINFO, Embase, MEDLINE, CINAHL, ASSIA, Scopus, Web of Science, and ProQuest Dissertations and Theses. The search strategy was designed in collaboration with an experienced librarian and altered to suit the requirements of each database. The records found in each database were deduplicated after importing them into EndNote. The ICTRP was searched for trial registries.

Once the included reports had been identified, their reference lists were searched for further eligible reports. Finally, titles of included reports were entered into Google Scholar to find more recent published work that had cited these reports. This was done in April 2021, and updated in November 2021.

JR searched the reference lists of any similar reviews known to the authors or any reviews found during the search for any additional eligible reports in April 2021.

Search Strategy

An original search strategy was created in collaboration with a librarian. After trialing this, it was clear that more general words for "mental health" needed to be added to retrieve papers in which parents with psychosis might have been involved, but where psychosis was not mentioned in the title or abstract. It also became clear that searches using index subject headings were not as effective as searches using key terms. Therefore, only searches using key terms were used for the final search strategy. The full electronic search strategy for MEDLINE was as following:

1. ((schizophreni* or smi or "serious mental illness" or "severe mental illness" or psychosis or paranoi* or "mental health" or "mental* ill*" or "mental* disorder*" or "mental* impair*" or "psychiatric") adj4 (parent* or mother* or father* or maternal* or paternal*)).ab,ti.
2. (psychotherap* or therap* or intervention* or train* or education* or program*).ab,ti.
3. limit 1 to yr = "2001-Current"
4. limit 2 to yr = "2001-Current"
5. 3 and 4

A similar search strategy was adapted for other databases, trial registries, preprint servers and websites. Websites were searched using Google Advanced, by limiting the domain to org.uk, gov, gov.uk, com.au, nhs.uk, or org.

Selection of Sources of Evidence

After the records obtained from the database search were deduplicated using EndNote, they were imported onto Rayyan, which is an online platform designed for multiple reviewers to work on systematic reviews (51). Reviewers are kept blind to each other's decisions, and are able to mark records as "include," "exclude," or "maybe" and can also mark exclusion reasons or add notes. This process was used to determine which records would be brought forward to full text review. All records were reviewed by JR, then FH and MLK each screened 50% of records, such that each record was screened twice. Every record that was deemed to be eligible by at least one researcher was brought forward to full text review (i.e., if there was a disagreement, this record was brought forward to full text review).

Full text review was completed using Excel. JR retrieved the full texts for every paper. NS reviewed a random sample of 25% of the records, and a Cohen's kappa of 0.90 was achieved (52).

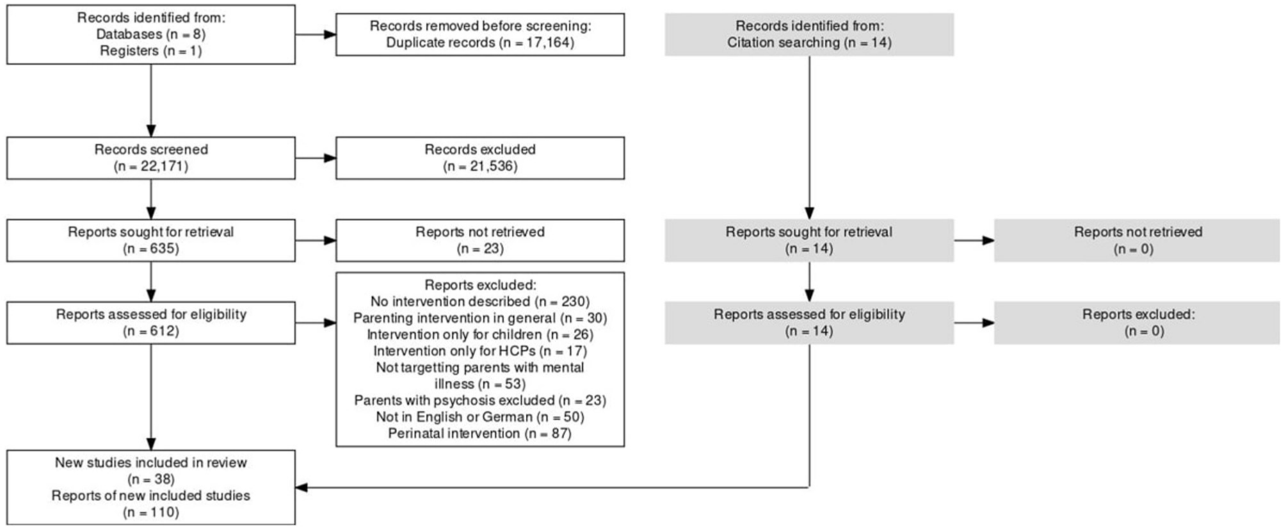


FIGURE 1 | Flow diagram of identification of reports.

The German records were screened at full-text stage by BM only. Reasons for exclusion are detailed in **Figure 1**.

Trial registries, preprint servers and websites were reviewed by JR only.

Data Charting Process

JR, LJ, and JB discussed the included papers and decided which details to extract from each report in order to satisfy the research questions. An excel form was created to capture this data with limits in terms of what values could be entered under each section. JR extracted data from all reports, then NS and RD extracted data from 50% of the papers each, such that each included paper underwent double data extraction. Where information was not available in the paper, the relevant field in the data extraction form was left blank. Disagreements were resolved through discussion. BM extracted data from the included reports which were written in German.

Data Items

Each data item was a study of an intervention. Data were extracted from each report on (1) location of the intervention, (2) who the intervention was intended for, (3) who delivers the intervention and how much training they receive, and (4) the format of the intervention. When an intervention provided separate components for the parent and the child, only components relevant to the parent intervention were reported.

Details about the components of each intervention were extracted e.g., explaining mental illness to children, psychoeducation, parenting skills, case management. Where the same intervention had been trialed by different teams but no adjustments had been made to the components, it was collapsed into one item.

If an evaluation had been completed, or registered as a protocol, participants' demographic details, and the design and results of the evaluation were extracted.

Qualitative evaluations were only included when participants were given the opportunity to answer open-ended questions, as part of a survey or an interview. When available, the themes produced from a qualitative analysis were extracted, otherwise the most salient elements from the qualitative research were extracted. If multiple intervention members were interviewed (e.g., parent, child, facilitator), only the data produced by the parents that were specific to the parenting intervention were extracted.

For quantitative evaluations, outcome measures related to the parent or child were extracted, and classified into "child behavior," "child psychosocial," "child quality of life," "parenting," "parent psychosocial," or "parent quality of life," and any significant differences obtained on these measures were indicated.

The final data charting form can be found in **Supplementary File 1**.

Synthesis of Results

Once the data charting form was completed, frequency data on the interventions was reported. After charting the components of each intervention, interventions with similar components were grouped into five categories. After inspection of the clusters of components in these similar interventions, these categories were named: (1) Talking about parental mental illness, (2) Improving parenting skills, (3) Long-term tailored support for the whole family, (4) Groups for parents with mental illness, and (5) Family therapy. A narrative summary was provided for the qualitative and quantitative evaluations of interventions.

RESULTS

Selection of Records

After duplicates were removed, a total of 22,171 records were screened by at least two reviewers at the title and abstract stage.

If at least one reviewer decided a record should be included to full text stage, it was brought forward, which was the case for 635 records. Of these, 23 could not be retrieved from library journal databases, and the remainder were assessed for eligibility at full text stage. The main reason for exclusion ($n = 502$) was that the report did not describe an intervention (see **Figure 1** for further detail). A total of 96 reports were included in the review. After looking at their reference lists as well as using Google scholar to search for more recent reports that had cited them, 14 more reports were found, making a total of 110. Most interventions had multiple reports describing them, such that the 110 reports described 38 studies of interventions, which accounted for 34 interventions in total. Three reports were written in German. All reports that were included can be found in **Supplementary File 2**.

Records were identified from database searches and trial registries. No additional records were identified through organizational websites, preprint servers or through searching the reference lists of other similar reviews.

Characteristics of Interventions

Many interventions had been delivered in more than one country. The country that had developed the most interventions was Australia ($n = 7$), followed by the UK ($n = 6$), Germany ($n = 5$), the Netherlands ($n = 5$), and the USA ($n = 5$). There was also a report of an intervention from each of the Scandinavian countries: Sweden ($n = 3$), Finland ($n = 2$), Denmark ($n = 1$), and Norway ($n = 1$). Switzerland and Israel had two interventions each and Portugal and Ireland had one each. **Table 1** presents the data extracted from each of the included studies.

Most interventions were designed either for parents with any mental illness or parents with severe mental illness, as defined by the study authors. Only two interventions were trialed with a group of parents with psychosis—*Triple P* (93) and *Family Talk* (68)—neither of which had been adapted from their original format. Eighteen interventions were designed for the whole family, six were for the affected parent and their child(ren) and 13 were for the affected parent only. Most interventions were led by a mental health professional or a social worker, or were in the form of self-help except for *Family Options* which is led by a graduate in psychology (63) and the *Godparents programme* which is led by a non-professional (71).

Many interventions were designed to be delivered in an outpatient community setting ($n = 13$), seven in a home setting, and eight interventions either in a community and home setting, or involved both a community and a home element. Three interventions were provided online, and it wasn't possible to determine the location of five interventions. Most interventions were delivered on a one-to-one basis ($n = 22$), a smaller number having been designed to be delivered using a group format ($n = 8$), or using both individual and group components ($n = 7$). Group interventions were more likely to be for the parent only or for both the parent and the child with a parent group and a child group being held separately.

The shortest intervention was *Let's Talk about Children* in either the meeting format, with two to three sessions (75), or via a self-help booklet (77). Some interventions were open-ended, meaning the parents could attend for as long as they liked [e.g., (67)] and the *Godparents programme* lasted for at least 3 years (71).

Intervention Components

Out of the 38 studies included in this review, four described *Beardslee's Family Talk* (65–68) and two described the *Triple P self-help workbook* (92, 93). Therefore, these 38 studies described 34 unique interventions. Of the 34 interventions listed in **Table 2**, most covered parenting skills ($n = 21$), aimed to strengthen the parent–child relationship ($n = 18$) or contained psychoeducation on child development ($n = 17$). Many interventions also had a focus on the child by including psychoeducation for the parent either on how their illness might impact upon their child ($n = 16$) or explaining mental illness to the child ($n = 16$). The intervention that comprised the most components was *VIA Family*, which contained 12 out of the 20 total components. Interventions were grouped into the following five categories depending on their focus.

Talking About Parental Mental Illness

Eight interventions focused on explaining parental mental illness to the child[ren] in the family and giving family members the space to talk about their experiences of parental mental illness. *Family Talk* was originally designed in the USA to target families with affective disorders (39) and has since been used with parents with any mental illness. Depending on its adaptation, it usually involves six to eight sessions, includes separate meetings for the parents and the children, and concludes with whole family meetings. *Let's Talk about Children* is a similar, but much shorter intervention in which the children are not invited to the meetings, and instead the parents are given advice on how to talk about their mental illness to their child (76). *Let's Talk about Children* also exists in a booklet form (77). The *Effective Child and Family Program* (61) offers either *Family Talk* or *Let's Talk about Children*, as well as self-help material with the potential for a multiagency meeting for the family, if any problems are identified. The *CHIMPS intervention* in Germany (59) has adapted *Family Talk* by including psychodynamic elements. *Child Talks+* (57) aims to enable the parents to explain mental illness to their children and for family members to get a chance to talk about their experiences. It consists of four meetings, with the first two being only with the parents, and the children attending the final two. The *Child and Family Inclusive Program* (54) has a similar focus but allows families to choose whether children are seen together with the parents, or separately. *KidsTime* (73) is an intervention that both children and parents attend, in which children take part in a drama group and parents take part in a parent group. Everyone meets at the end of the session to watch the children perform, and the content of these performance often centers on the parent's mental illness.

Improving Parenting Skills

Eleven of the interventions had a focus on improving parenting skills. Four interventions (36, 37, 91, 93) were based on the *Triple P*, originally designed for the parents of children with behavioral difficulties (99). *Triple P* teaches parents about enhancing their relationship with their children, encouraging certain behaviors, discouraging others, and setting clear boundaries (99). In this

TABLE 1 | Characteristics of 38 studies of interventions.

Intervention and authors of primary report(s)	Country	Parent diagnosis and child age	Who takes part in intervention	Who delivers intervention and training	Setting of intervention	Referral method	Group or Individual	Length of intervention	Manualized
BROSH program (53)	Israel	MI, 0–18	Whole family	Mental health professional or social worker	Home	Adult mental health or child services	Individual	3 h weekly meeting for 2 years	No
Child and family inclusive programme (54, 55)	Australia	MI, 4–18	Whole family	Mental health professional or social worker	Community or home	Adult mental health or self-referral	Individual	3–8 60–90 min sessions	No
Child resilience programme (56)	USA (Indiana)	SMI, 8–18	Whole family	*Unknown*	Community	Adult mental health	Both	7–8 weekly individual family sessions 2+ monthly group therapy	No
Child Talks+ (57, 58)	Norway, Portugal, the Netherlands	MI, 0–18	Whole family	Mental health professional or social worker Two days	Community or home	Adult mental health	Individual	Four weekly or biweekly 1 h sessions	Yes
CHIMPS intervention (59)	Germany and Switzerland	MI, 3–19	Whole family	Mental health professional Two days	Community	Adult mental health	Individual	8 × 60–90 min sessions over a period of 6 months	Yes
Counseling and support service (60)	Germany	MI	Whole family	Mental health professional	Community	Adult mental health	Individual		No
Effective child and family program (61, 62)	Finland	MI	Whole family	Mental health professional or social worker Seventeen days	Community or home	Adult mental health	Individual	6–8 sessions for Family Talk OR 2–3 for Let's Talk 1 family meeting	Yes
Family options (63, 64)	USA (Massachusetts)	SMI, 18 months–16	Whole family	Psychology graduates	Home	Adult mental health or child services	Individual	Weekly meetings for 12 months	No
Family Talk (65)	Germany	MI	Whole family	*Unknown*	Community	Self-referral	Both	2 × 90 min group sessions for parents 5 group sessions for children One individual family session Over 3 months	Yes
Family Talk (66)	Ireland	MI, 5–18	Whole family	Mental health professional or social worker Online training–15 h and monthly supervision	Community or home	Adult mental health	Individual	7 weekly 60–90 min sessions	Yes

(Continued)

TABLE 1 | Continued

Intervention and authors of primary report(s)	Country	Parent diagnosis and child age	Who takes part in intervention	Who delivers intervention and training	Setting of intervention	Referral method	Group or Individual	Length of intervention	Manualized
Family Talk (67)	Sweden	MI, 8–18	Whole family	Mental health professional or social worker 5 days of theory, 5 days of supervision in a year	*Unknown*	Adult mental health	Individual	6 or 7 sessions	Yes
Family Talk (68, 69)	Sweden	Psychosis, 8–17	Whole family	Mental health professional or social worker	*Unknown*	*Unknown*	Individual	6 or 7 sessions	Yes
FWA Newpin service (70)	UK (London)	MI, 0–5	Parent and child	Social worker	Community	*Unknown*	Both	Meetings held twice a week	No
Godparents programme (71)	Switzerland	MI, 0–18	Whole family	Non-professionals Introductory event, regular peer supervision, two-four supervisions with coordinator a year	Home	Adult mental health or child services	Individual	Regular meetings for at least 3 years	No
Integrated family treatment (72)	USA (New Hampshire)	SMI	Parent and child	Mental health professional	Home	Adult mental health	Individual	1–5 years of sessions	No
Invisible children's project (20)	USA (New York)	MI	*Unknown*	Social worker	*Unknown*	Child services referral	*Unknown*	*Unknown*	No
KidsTime (73, 74)	UK, Germany, Spain	MI	Parent and child	Mental health professional or social worker Two days	Community	Adult mental health or child services or self-referral	Group	Monthly meetings lasting 2.5 h	Yes
KopOpOuders (22)	The Netherlands	MI, 1–21	Parent	Mental health professional	Online	Adult mental health or child services or self-referral	Group	8 weekly 90 min sessions	Yes
Let's talk about children (75, 76), ACTRN12616000460404	Finland, Sweden, Australia	MI, 0–18	Parent	Mental health professional Two days online and 4 h face to face	*Unknown*	Adult mental health	Individual	2 or 3 weekly 60 min sessions	Yes
Let's talk about children booklet (77)	Australia	MI, 0–18	Parent	Self-help	Community or home	Adult mental health	Individual	Open-ended	No
Living with under fives (78, 79)	Australia	SMI, 0–5	Parent and child	Occupational therapist	Community	Adult mental health or child services	Group	Weekly meetings lasting 2 h	No
Parenting internet intervention (80)	USA (Pennsylvania)	SMI, 0–18	Parent	Self-help	Online	Self-referral	Individual	12 weekly 30 min sessions	Yes
Parenting with success and satisfaction workbooks (81–83)	The Netherlands	SMI, 0–21	Parent	Self-help with option of Mental health professional Four days	Community or home	Adult mental health	Both	Weekly meetings for a year	Yes

(Continued)

TABLE 1 | Continued

Intervention and authors of primary report(s)	Country	Parent diagnosis and child age	Who takes part in intervention	Who delivers intervention and training	Setting of intervention	Referral method	Group or Individual	Length of intervention	Manualized
Preventive basic care management (PBCM) (84, 85)	The Netherlands	MI, 3–10	Whole family	*Unknown*	Home	Adult mental health	Individual	18 months	No
SEEK (86)*	Germany	SMI	Parent and child	Mental health professional	Child inpatient unit	Adult mental health or child services	Group	6 × 90 min sessions over 5 weeks	Yes
Strengths based parenting programme (87)	Australia	MI	Parent	Mental health professional	Community	Adult mental health or self-referral	Group	5 weekly 2 h sessions	No
The lighthouse (leuchtturm) parenting programme (88)*	Germany	SMI, 0–14	Parent	Psychologist, social worker, psychiatrist, nurses	Adult inpatient unit	Adult mental health or self-referral	Both	5 individual sessions (2 with video feedback) One session with care worker 4 group sessions Weekly over 12 weeks	Yes
Therapeutic group (89)	Israel	MI	Parent	Mental health professional or social worker	Community	Adult mental health or child services	Group	Weekly meetings for 21 months	No
Think family whole family programme (90)	UK (Leicester)	MI	Whole family	Mental health professional or social worker Two days	*Unknown*	*Unknown*	Individual	8 sessions	Yes
Triple P (91)*	Germany	SMI, 2–10	Parent	Mental health professional 10 sessions of training	Community	Adult mental health or child services	Individual	8–10 weekly 50–60 min sessions	Yes
Triple P – every parent's self-help workbook (92)	UK (Manchester)	MI, 2–12	Parent	Self-help with option of mental health professional 45–60 min	Home	Child services referral	Individual	Booklet is completed over 10 weeks	Yes
Triple P – every parent's self-help workbook (93)	UK (Manchester)	Psychosis, 3–10	Parent	Self-help with option of mental health professional	Home	Adult mental health or child services	Individual	10–14 weekly visits for 1.5 h	Yes

(Continued)

TABLE 1 | Continued

Intervention and authors of primary report(s)	Country	Parent diagnosis and child age	Who takes part in intervention	Who delivers intervention and training	Setting of intervention	Referral method	Group or Individual	Length of intervention	Manualized
Triple P + CBT (37)	Germany	MI, 1.5–16	Whole family	Mental health professional	Community	Adult mental health	Both	25–45 sessions CBT 8–10 sessions Triple P Weekly or bi-weekly sessions for 6–12 months	Yes
Triple P + mental health components (36)	Australia	MI, 2–12	Parent	Mental health professional or social worker	Community or home	Adult mental health or child services or self-referral	Both	6 weekly 2.5–3 h group + four individual visits	Yes
Tuning into kids (38)	Australia	MI, 3–12	Parent	Mental health professional	Community	Adult mental health	Group	6 weekly 2 h sessions	Yes
VIA family (94)	Denmark	SMI, 6–12	Whole family	Child psychiatrist, child psychologist, adult mental health nurse social worker, and a family counselor	Community or home	Adult mental health	Individual	1–2 sessions introduction 2–4 sessions lifeline and history 6–8 sessions psychoeducation 3–10 sessions Triple P 8 sessions groups for children and parents All over 18 months	No
You are okay (95, 96)	The Netherlands	MI, 10–20 with mild individual disability	Parent and child	Self-help with option of support from social worker	Online	Child services referral	Individual	5 sessions online for parents + 10 weekly support group sessions for children	Yes
Young SMILES (97, 98)	UK (Manchester)	SMI, 6–16	Whole family	Mental health professional or social worker Three days	Community	Adult mental health or child services	Group	5 weekly 2 h sessions	Yes

MI, mental illness; SMI, severe mental illness.
**Indicates paper written in German.*

TABLE 2 | Components of 34 interventions, separated into five categories.

Intervention and Primary report(s)	Explaining mental illness to child(ren)	Psycho-education on how PMI impacts on child	Psycho-education on mental health	Psycho-education on child development	Chance for family to talk about experiences of PMI	Parent-child relationship	Parenting skills	Parent well-being or self-care	Parent social support	Parent emotional support	Peer support	Money management	Goal setting	Crisis planning for periods of poor MH	Family therapy	Case management	Interagency or multi team collaboration	Signposting to other supportive agencies	Mentalizing component	Separate child element
TALKING ABOUT PARENTAL MENTAL ILLNESS																				
Family Talk (65–69)	X	X	X		X															X
Let's Talk about children (75, 76), ACTRN12616000460404	X	X			X															
Let's Talk about Children booklet (77)	X	X			X															
Effective Child and Family Program (61, 62)	X	X			X												X	X		
CHIMPS intervention (59)	X	X	X		X	X		X	X						X					
Child Talks+ (57, 58)	X	X	X		X		X			X										
Child and family inclusive programme (54, 55)	X				X															
KidsTime (73, 74)	X	X	X		X				X		X						X	X		X
IMPROVING PARENTING SKILLS																				
Triple P self-help workbook (92, 93)				X		X	X						X							
Triple P + CBT (37)							X													
Triple P + mental health components (36)		X		X		X	X							X						
Triple P (91)*	X	X	X	X		X	X	X					X	X						
Tuning into kids (38)				X		X	X			X	X								X	
The lighthouse (leuchtturm) parenting programme (88)*				X		X	X		X				X				X	X	X	
Strengths based parenting programme (87)	X	X	X	X		X	X	X	X					X				X		
KopOpOuders (22)				X		X	X		X					X						
You are okay (95, 96)	X	X					X		X											X
Parenting internet intervention (80)		X		X		X	X	X			X									
Parenting with success and satisfaction workbooks (81–83)							X	X					X					X		

(Continued)

TABLE 2 | Continued

Intervention and Primary report(s)	Explaining mental illness to child(ren)	Psycho-education on how PMI impacts on child	Psycho-education on mental health	Psycho-education on child develop-ment	Chance for family to talk about experiences of PMI	Parent-child relationship	Parenting skills	Parent well-being or self-care	Parent social support	Parent emotional support	Peer support	Money manage-ment	Goal setting	Crisis planning for periods of poor MH	Family therapy	Case manage-ment	Interagency or multi team collaboration	Signposting to other supportive agencies	Mentalizing component	Separate child element
LONG-TERM TAILORED SUPPORT FOR THE WHOLE FAMILY																				
Invisible children's project (20)							X		X	X				X		X	X	X		
Family options (63, 64)										X	X	X	X	X		X	X	X		
Integrated family treatment (72)				X		X	X		X	X							X	X		
VIA family (94)		X	X	X		X	X		X			X		X		X	X	X		X
Preventive basic care management (PBCM) (84, 85)				X					X							X	X	X		X
Godparents programme (71)					X				X	X										X
BROSH program (53)	X			X		X	X	X				X			X	X	X		X	X
GROUPS FOR PARENTS WITH MENTAL ILLNESS																				
Living with under fives (78, 79)				X		X	X		X					X			X	X		X
FWA newpin service (70)						X		X	X		X								X	
Therapeutic group (89)	X			X		X	X			X	X						X	X		
Young SMILES (97, 98)	X			X		X	X													X
SEEK (86)*	X		X	X	X	X	X	X		X				X			X	X		X
FAMILY THERAPY																				
Child resilience programme (56)	X	X	X		X	X	X		X				X	X	X					X
Think family whole family programme (90)		X	X		X								X	X	X			X		
Counseling and support service (60)		X		X											X	X				
Total	16	16	10	17	12	18	21	8	14	8	6	3	7	11	5	6	12	14	4	11

PMI, parental mental illness.

TABLE 3 | Participant characteristics in 23 completed evaluations of included interventions.

Intervention name	No. of parent participants	Percentage with psychotic diagnosis	Age of parents (mean, standard deviation or range)	Percentage of mothers	Ethnicity of parents	Marital or living status of parents	Education of parents	Employment of parents	Age of children (mean, standard deviation or range)	Percentage of daughters	Number of children in family	Percentage of children living with parents
BROSH program (53)	11	36.4%	Mean = 39.2 Range = 32–57	*Unknown*	*Unknown*	27.3% single 27.3% divorced 45.4% married	*Unknown*	57% unemployed	Range = 2 months–11.5 years	*Unknown*	*Unknown*	*Unknown*
Family options (63)	22	4.6%	Mean = 36 SD = 8.3	100%	77.2% White 9.1% Black 9.1% Hispanic 4.6% Asian	36.4% lived with a significant other	More than 80% completed high school	18% part or full-time employed	*Unknown*	52%	Mean = between 2 and 3 SD = 1.3 Range = 1–5	88.5% of children lived with parents
Family Talk (67)	66	13.6%	*Unknown*	80.3%	*Unknown*	32% single	*Unknown*	*Unknown*	Median = 12	*Unknown*	*Unknown*	*Unknown*
Family Talk (68)	8	100%	*Unknown*	75%	*Unknown*	*Unknown*	*Unknown*	100% unemployed and unable to work	Range = 8–15	57.1%	*Unknown*	86% lived with at least one parent 14% placed in foster care
Family Talk (65)	37	0%	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	Mean = 10.41 SD = 2.66	*Unknown*	*Unknown*	*Unknown*
Integrated family treatment (72)	8	*Unknown*	Range = 20–41	100%	100% Caucasian	37.5% not living with partner 62.5% married or living with partner	62.5% at least high school education	*Unknown*	*Unknown*	*Unknown*	Range = 1–4	*Unknown*
KidsTime (74)	5	*Unknown*	*Unknown*	100%	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*
KopOpOuders (22)	48	6.3%	Mean = 37 SD = 6.8	85.4%	90% Dutch 10% Belgian, Turkish or Danish	58% dual parent families 56% married	42% intermediate education 27% higher education	52% employed	Mean = 6.7 SD = 5.3	*Unknown*	83% of parents had 1 or 2 children	*Unknown*
Let's talk about children (75)	39	42.5%	Mean = 39.9 Range = 26–62	94.9%	*Unknown*	51.2% single parent household	*Unknown*	*Unknown*	Mean = 9.5 Range = 6 months–18 years	*Unknown*	Mean = 1.8 Range = 1–5	*Unknown*

(Continued)

TABLE 3 | Continued

Intervention name	No. of parent participants	Percentage with psychotic diagnosis	Age of parents (mean, standard deviation or range)	Percentage of mothers	Ethnicity of parents	Marital or living status of parents	Education of parents	Employment of parents	Age of children (mean, standard deviation or range)	Percentage of daughters	Number of children in family	Percentage of children living with parents
Let's talk about Children booklet (77)	19	0%	Mean = 42.9 Range = 34–60	89.5%	94.7% born in Australia 5.3% born overseas	26.3% single 57.9% married or living together 15.8% separated or divorced	5.3% primary education 42% intermediate education 52.7% higher education	*Unknown*	*Unknown*	*Unknown*	Mean = 1.8	84.2% lived full time with children 10.6% lived with children more than half the time 5.2% lived with children less than half the time
Parenting internet intervention (80)	60	13.3%	Mean = 37 SD = 7	100%	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*
Parenting with success and satisfaction workbooks (82)	26	7.7%	Range = 21–52	76.9%	*Unknown*	42% unmarried 19% married 39% divorced/widowed	54% primary education 42% intermediate education 4% higher education	42% employed	*Unknown*	*Unknown*	35% had 1 child 65% had 2–4 children	69% were legally responsible for their child 12% were legally responsible with a foster poster 19% were not legally responsible for their child
Preventive basic care management (PBCM) (85)	99	*Unknown*	*Unknown*	87.9%	33% Dutch 19% Moroccan 15% Turkish 14% Surinamese 7% Netherland Antilles 12% other	46% single parent family	*Unknown*	*Unknown*	Mean = 6.08	45%	Mean = 2.13	*Unknown*

(Continued)

TABLE 3 | Continued

Intervention name	No. of parent participants	Percentage with psychotic diagnosis	Age of parents (mean, standard deviation or range)	Percentage of mothers	Ethnicity of parents	Marital or living status of parents	Education of parents	Employment of parents	Age of children (mean, standard deviation or range)	Percentage of daughters	Number of children in family	Percentage of children living with parents
SEEK (86)*	26	*Unknown*	Mean = 37.1	92.3%	*Unknown*	34.6% single 53.8% married 11.6% divorced/separated 65.4% living with a partner	3.4% primary education 65.3% intermediate education 30.7% higher education	*Unknown*	Mean = 5.92	46.2%	*Unknown*	*Unknown*
Strengths based parenting programme (unnamed) (87)	4	25%	Mean = 36.75 Range = 23–48	75%	100% Anglo-Australian	*Unknown*	*Unknown*	*Unknown*	Mean = 9.6 Range = 2–21	*Unknown*	50% had 1 child 25% had 2 children 25% had 8 children	*Unknown*
The lighthouse (leuchtturm) parenting programme (88)*	5	0%	*Unknown*	100%	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*
Therapeutic group (unnamed) (89)	35	14.3%	Mean = 43	45.7%	*Unknown*	50% divorced or separated	*Unknown*	*Unknown*	Mean = 2.7 Range = 1–9	*Unknown*	*Unknown*	*Unknown*
Triple P (91)*	42	0%	Mean = 37 SD = 5.1	83.3%	*Unknown*	70% married or living with partner 17% single/separated/divorced 13% Unknown	*Unknown*	*Unknown*	Mean = 6 SD = 2.7	43%	61.5% had one child 27% had two children 11.5% had three children	*Unknown*
Triple P + mental health components (36, 103)	86	4.7%	Mean = 32.6 SD = 6.4	90.7%	93% Not aboriginal or Torres Strait 7% Aboriginal or Torres Strait	38% single 62% married or living with partner	*Unknown*	*Unknown*	Mean = 4.9	38%	*Unknown*	*Unknown*

(Continued)

TABLE 3 | Continued

Intervention name	No. of parent participants	Percentage with psychotic diagnosis	Age of parents (mean, standard deviation or range)	Percentage of mothers	Ethnicity of parents	Marital or living status of parents	Education of parents	Employment of parents	Age of children (mean, standard deviation or range)	Percentage of daughters	Number of children in family	Percentage of children living with parents
Triple P self-help workbook (93)	10	100%	Mean = 33 Range = 26–48	100%	80% White British 10% Black other 10% Chinese	90% sole parent household 10% cohabiting	30% primary education 10% intermediate education 60% higher education	10% employed part-time 90% unemployed and not able to work	Mean = 8 Range = 4–10	40%	Mean = 2 Range = 1–5	*Unknown*
Tuning into kids (38)	8	12.5%	*Unknown*	87.5	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*	*Unknown*
You are okay (95)	41	*Unknown*	Mean = 43.9	85.4%	87.8% born in the Netherlands	51.2% single parent family	26.8% primary education 63.4% intermediate education 9.8% higher education	53.7% unemployed	Mean = 14.1	38.2%	*Unknown*	*Unknown*
Young SMILES (97)	33	9.1%	*Unknown*	90.9%	91% White British 6% Asian 3% *Unknown*	81.8% unmarried	81.8% intermediate education 12.2% higher education 3% *Unknown*	96.9% unemployed	Mean = 10.6	60%	*Unknown*	100% of children lived with parents

TABLE 4 | Design and results of 28 completed evaluations or protocols for evaluations of included interventions.

Intervention name	**QUANTITATIVE EVALUATION**		**QUALITATIVE EVALUATION**	
	Design	**Quantitative results**	**Data collection and analysis**	**Qualitative results**
Studies with both quantitative and qualitative evaluations				
Let's talk about children (75)	Quasi-experimental	– Parenting ○ Parenting stress scale – Parent psychosocial ○ General functioning index of MFAD	Semi-structured interviews Interpretative phenomenological analysis and thematic analysis	• Insight ○ Parents commented they focused on their child more after LT ○ Parents felt they family was more connected after LT • Normalizing ○ LT gave parents more confidence in their own parenting • Family communication ○ Families talked about PMI more after LT • Clinician support for the parenting role ○ One parent said her case manager now better sees her in the context of her family • Additional support required • Parents saw LT as the start of a conversation and identified the next stages including helping their children to regulate their emotions
Let's talk about children booklet (77)	Within group pre-post analysis	– Parenting ○ Parenting self-agency measures, Parenting and mental illness scale *No significance testing*	Semi-structured interviews Thematic analysis	• General feedback regarding the resource ○ Parents felt they could relate to the resource ○ Some parents felt the resource could be upsetting ○ The booklet helped with asking for support • How the parents used the resource ○ The resource helped parents feel they could start a conversation with their child about PMI ○ One parent questioned whether it was important to have conversations about PMI • Recommendations for dissemination ○ The resource is useful for parents at all stages of their illness • One parent suggested that it would only work for those who had accepted their diagnosis
The lighthouse (leuchtturm) parenting programme (88)	Within group pre-post analysis	– Parenting ○ EBI *No significance testing*	*Unknown*	• Parents enjoyed the mentalization metaphors • Parents enjoyed the group format and speaking to other parents with mental illness • Some parents asked for longer and more sessions • Parents reported their stress levels decreasing • Parents reported their parenting self-efficacy increasing
Parenting with success and satisfaction workbooks (83)	Non-randomized controlled trial	– Parenting ○ TOPSE – Parent psychosocial ○ PES – Parent quality of life ○ WHOQOL-BREF, EUROQOL-VAS*	Semi-structured interviews *Unknown analysis*	• Parents could identify relevant support systems following intervention • One parent said she felt she had made progress in her role as a mother
Triple P + mental health components (36, 103)	Within group pre-post analysis	– Child behavior ○ ECBI* – Parenting ○ Parenting scale*	Semi-structured interviews Thematic analysis	• Being in a group with others with mental illness ○ Knowing others also had a mental illness reduced anxiety ○ Parents felt they had similar experiences to others in the group and felt understood • Focus on child development and parenting with a mental illness ○ Parents felt they learnt techniques on how to handle their child's behaviors ○ Parents could identify their own triggers so felt more in control ○ Parents felt they understood their children more after Triple P • The home visits • Parents felt the home visits at the end of the intervention helped embed the learning from Triple P

(Continued)

TABLE 4 | Continued

Intervention name	**QUANTITATIVE EVALUATION**		**QUALITATIVE EVALUATION**	
	Design	**Quantitative results**	**Data collection and analysis**	**Qualitative results**
Triple P self-help workbook (93)	Within group pre-post analysis	– Parenting ○ Parenting tasks checklist, Parenting scale*, Parenting and family adjustment scales* – Parent psychosocial ○ Psyrats*, DASS-21, PANSS, Calgary Depression Scale, PSP*, WEMBWBS* – Child behavior ○ ECBI*, SDQ*	Semi-structured interviews Interpretative phenomenological analysis	• The discovery of self and lost possibilities ○ Parents felt positive about taking part in Triple P ○ Parents spoke about the relationship between mental health and parenting ○ Parents felt they were more in control after Triple P • The transition to appropriate parenting ○ Parents felt their parenting had improvement after Triple P e.g., less screaming and more open communication with their child ○ Parents thought their children were happier after Triple P and that family life was better • Parents took more pride from their role as a parent after Triple P
Tuning into kids (38)	Within group pre-post analysis	– Parenting ○ Parents concerns questionnaire* – Parent psychosocial ○ K10, DERS, PESQ	Open-ended questionnaire Conventional content analysis	• Parents felt comfortable in the group format • Some parents felt they were more skilled in their parenting at Tuning into Kids • Some parents identified communication with their child was better • One parent said she felt she could help her daughter with her anxiety more
Young SMILES (97)	Feasibility RCT	– Child quality of life ○ PedsQL, KIDSCREEN, CHU9D – Child psychosocial ○ RCADS – Child behavior ○ SDQ – Parenting ○ Mental health literacy questionnaire, Parenting Scale, PSI *No significance testing*	Semi-structured interviews Thematic analysis	• Intervention coherence ○ Some parents felt there was not enough focus on them as a parent • Affective attitude ○ Parents were keen for their child to understand PMI ○ Parents felt hopeful for the future after attending Young SMILES ○ Some parents felt comfortable with the group approach and some didn't like it • Burden ○ Parents felt anxious about going to the group ○ Some parents felt pressured to attend the group • Ethnicity ○ Some parents valued separate parent and child groups and some wished they had been with their children ○ Parents enjoyed the setting of the Young SMILES intervention • Opportunity costs ○ One parent interpreted Young SMILES as claiming her mental illness was damaging her child ○ One parent said the assessment was too invasive and her mental health declined as a result • Perceived effectiveness ○ Parents felt their children were coping better after Young SMILES and that the family environment was more relaxed ○ Parents enjoyed being in a group with others who had similar experiences • Self-efficacy ○ Parents spoke highly of the facilitator and the non-judgmental nature of the group • Parents felt respected in the group
Studies with only a quantitative evaluation				
BROSH Program (53)	Within group pre-post analysis	– Parent psychosocial ○ CANS subscale—impact on caregiver – Child psychosocial ○ CANS subscale—affect regulation *No significance testing*		

(Continued)

TABLE 4 | Continued

Intervention name	QUANTITATIVE EVALUATION		QUALITATIVE EVALUATION	
	Design	**Quantitative results**	**Data collection and analysis**	**Qualitative results**
Child talks+ (57)	Protocol Full RCT	– Child quality of life ○ KIDSCREEN-27, PEDS – Child psychosocial ○ READ, GSQ-APMI, Children's mental health literacy scale – Child behavior ○ SDQ – Parenting ○ Parent-child communication scale, PSCS		
CHIMPS intervention (59)	Protocol Full RCT	– Child psychosocial ○ Schedule for affective disorders and schizophrenia for school aged children, Youth self-report, Children global assessment scale – Child behavior ○ CBCL – Child quality of life ○ KIDSCREEN – Parent psychosocial ○ BSI, Health questionnaire, Global assessment of relative functioning, Oslo social support questionnaire – Parent quality of life ○ EQ-5D – Parenting ○ FB-A		
Family options (63)	Within group pre-post analysis	– Parent psychosocial ○ Global Severity Index of BSI*, Posttraumatic Stress Symptom Scale, SF-8, MOS-SSS		
Family Talk (65)	Non-randomized controlled trial with healthy control group	– Child behavior ○ CBCL**, SDQ** – Parenting ○ Knowledge about mental illness questionnaire**		
Family Talk (66)	Protocol Full RCT	– Child behavior ○ SDQ – Child psychosocial ○ RCADS, SCARED-5, CYRM-12 – Parent psychosocial ○ BASIS-24, CSE		
Integrated family treatment (72)	Within group pre-post analysis	– Parenting ○ HOME, Parent Stress Inventory – Parent psychosocial ○ BSI – Child quality of life ○ Lehman Quality of Life interview *No significance testing*		
KopOpOuders (22)	Within group pre-post analysis	– Parenting ○ Parenting Scale*, OOO* – Child behavior ○ SDQ		
Parenting internet intervention (80)	Full RCT	– Parenting ○ PSCS**, HFPI**, MOS-SSS, Family Coping Inventory		

(Continued)

TABLE 4 | Continued

Intervention name	QUANTITATIVE EVALUATION		QUALITATIVE EVALUATION	
	Design	**Quantitative results**	**Data collection and analysis**	**Qualitative results**
Preventive basic care management (PBCM) (85)	Full RCT	– Parenting ○ HOME, Parenting skill subscale of FFQ**, Parenting Daily Hassles – Child behavior ○ SDQ		
SEEK (86)*	Non-randomized controlled trial	– Parenting ○ EBI* – Parent psychosocial ○ HSCL-25 – Child behavior ○ CBCL		
Triple P (91)*	Non-randomized controlled trial with healthy control group	– Parent psychosocial ○ DASS-21** – Parenting ○ EFB-K ○ PEV – Child behavior ○ SDQ**		
Triple P + CBT (37)	Protocol Full RCT	– Child behavior ○ CBCL – Child psychosocial ○ Kinder-DIPS – Parent psychosocial ○ DIPS, BSI, PID-5-BF – Parenting ○ EFB, ESF, Child knowledge about mental disorders – Child quality of life ○ KIDSCREEN-10 – Parent quality of life ○ EUROQOL, AQoL-8D		
VIA family (94)	Protocol Full RCT	– Child behavior ○ CBCL - Child psychosocial ○ CGAS, Days absent from school – Parenting ○ FAD, HOME		
You are okay (95)	Quasi-experimental	– Child behavior ○ SDQ* – Child psychosocial ○ Self-perception profile for adolescents, COMPI specific cognitions, NRI-BSV – Parent psychosocial ○ SSL-12-I – Parenting ○ Perceived parental competence, Parental involvement with child's treatment, Parenting Scale		
Studies with only a qualitative evaluation				
Family Talk (67)			Open-ended questionnaire *Unknown analysis*	• Important for parent's recovery that the children understood how they had experienced their illness • Relationship with partner strengthened post Family Talk • Communication was easier post Family Talk • Parents felt they learned to focus on children more

(Continued)

TABLE 4 | Continued

Intervention name	QUANTITATIVE EVALUATION		QUALITATIVE EVALUATION	
	Design	**Quantitative results**	**Data collection and analysis**	**Qualitative results**
Family Talk (68)			Semi-structured interviews Qualitative content analysis	• Information ○ Family Talk improved family members' knowledge about PMI ○ FT meant the child knew who to turn to if their parent became ill • General parenting and child support ○ Some parents felt they had received good advice on parenting ○ Some parents felt that FT had not given them any specific support or made any concrete changes • Communication ○ Before FT, parents hesitated to talk about PMI ○ Some parents felt FT allowed them to communicate with their child about PMI, and others still found it too difficult to talk about • Understanding ○ Family members felt their understood each other's experiences better after FT ○ Parents who did not have custody of their children felt FT gave them an insight in their children's daily lives • Structure ○ Parents appreciated that their child was able to talk to the professional delivering the intervention ○ Parents appreciated the structure of the intervention and that the professional followed a manual ○ Some parents asked for a more holistic structure, where their illness wasn't the focus, and other family problems could be discussed
KidsTime (74)			Semi-structured interviews Thematic analysis	• Aims and impact ○ Parents felt they could communicate about PMI to their child ○ Parents gained more awareness about how PMI affected their child ○ Parents enjoyed being in a group of others with similar experiences ○ Parents felt their relationship with their child has improved, and that they feel more confident in their parenting role • Nature of referral process ○ Parents appreciated that they were referred by the school in contrast to being referred by a health or social care system • Need for extended support ○ Parents wanted more support for their children in schools
Strengths based parenting programme (unnamed) (87)			Written reflections and semi-structured interviews Thematic analysis	• Parents felt the programme helped them communicate effectively with their child • Parents felt they could relax a bit more during difficult parenting moments • Parents felt their understood their emotions better and could help their children to do so too
Therapeutic group (unnamed) (89)			Open-ended questionnaire Grounded theory	• Overcoming difficulties to connect to the children and maintain relationships with them ○ Parents provided suggestions to each other on how to maintain contact with their child ○ Parents felt comfortable in the group to share these difficulties

(Continued)

TABLE 4 | Continued

Intervention name	QUANTITATIVE EVALUATION		QUALITATIVE EVALUATION	
	Design	**Quantitative results**	**Data collection and analysis**	**Qualitative results**
				• Speaking with the child about the mental illness ○ Group members discussed whether or not to tell their child about their mental illness and how to do this in an age appropriate way • Improving parenting skills and developing the role of a parent ○ Parents expressed insecurities in their own parenting ○ Group members gave each other advice on setting boundaries and discipline • Hopes and fears regarding parenting ○ Parents spoke about their goals which including meeting child more often, developing a good relationship with their child, and taking more responsibility for their child

AQoL-8D, assessment of quality of life; BASIS-24, behavior and symptom identification scale 24; BSI, brief symptom inventory; CANS, child and adolescent needs and strengths; CBCL, child behavior checklist; CGAS, children's global assessment scale; CHU9D, child healthy utility 9D; CSE, coping self-efficacy questionnaire; CYRM-12, child and youth resilience measure 12; DASS-21, depression anxiety and stress scales short form; DERS, difficulties in emotional regulation scale; DIPS, diagnostic interview of mental disorders for parents and children; EBI, Eltern-Belastungs-Inventar; ECBI, Eyberg child behavior inventory; EFB, erziehungsfragebogen; ESF, elternstressfragebogen; FAD, family assessment device; FB-A, allgemeiner familienfragebogen; FFQ, family functioning questionnaire; GSQ-APMI, guilt and shame questionnaire for adolescents of parents with mental illness; HFPI, healthy families parenting inventory; HOME, home observation for measurement of the environment; HSCL-25, Hopkins symptom checklist-25; K10, Kessler psychological distress scale; MOS-SSS, medical outcomes study, social support survey; NRI-BSV, network of relationships inventory-behavioral systems version; OOO, Ouderlijke Opvattingen over Opvoeding; PANSS, positive and negative syndrome scale; PEDS, parents' evaluations of developmental status; PES, psychological empowerment scale; PESQ, parents emotional style questionnaire; PEV, positives elternverhalten; PID-5-BF, personality inventory for DSM-5-brief form; PSI, parent stress index; PSCS, parenting sense of competence scale; PSOC, parenting sense of competence; PSP, personal and social performance scale; PSYRATS, psychotic symptom rating scales; RCADS, revised child anxiety and depression scale; READ, resilience scale for adolescent; SCARED-5, screen for child anxiety related disorders; SCORE-15, systematic clinical outcome and routine evaluation; SDQ, strengths and difficulties questionnaire; SF-8, short form-8; SSL-12-I, Dutch social support list-interactions; TOPSE, tool to measure parenting self-efficacy; WEMBWBS, Warwick Edinburgh mental well-being.

**For sig. improvement with intervention group pre vs. post.*

***For sig. improvement between intervention and control group post intervention.*

review, the *Triple P Every Parents' Self-Help Workbook* (92, 93) was used for parents with mental illness, and Stracke et al. (37) combined *Triple P* with cognitive behavioral therapy. Both Phelan et al. (100) and Kuschel et al. (91) add two additional components about parental mental health to the *Triple P* syllabus. Two interventions were based on mentalization. The *Lighthouse (Leuchtturm) Parenting Programme* (88) is rooted in mentalization-based therapy, and aids parents in better understanding their child's mental states, and teaches behavioral management skills. *Tuning into Kids* focuses on teaching parents how to recognize and respond to their child's emotions (101), and Isobel et al. (38) trialed it with parents with mental illness. McFarland et al.'s (87) strengths based parenting programme took elements from *Triple P* and *Tuning into Kids*, and also had a focus on talking about parental mental illness to the child. *KopOpOuders* (22) is an online course which covers boundary setting, communicating, child development and emergency planning. *You are Okay* (95) is an intervention for parents with mental illness whose children have an intellectual disability. It has a support group for the children as well as an online course for parents which is based on the content of *KopOpOuders*. The *Parenting Internet Intervention* designed by Kaplan et al. (80) contained modules on child development, stress management, the effects of parental mental illness, and setting boundaries. *Parenting with Success and Satisfaction (PARSS)* (81) is a series of three workbooks, and has a focus on parenting skills. One of the workbooks is designed for parents not currently living with their children.

Long-Term Tailored Support for the Whole Family

Seven interventions offered longer-term support (at least 1 year long) for families with parental mental illness, and often involved case management and collaboration with other agencies. The *Invisible Children's Project* (20) is mandated as part of a child welfare plan in the U.S. and involves case management for the whole family. *Family Options* (64) is an intervention in the U.S. where Family Coaches are assigned to a family to provide many types of support, including emotional support, advocacy, and goal setting. These Family Coaches can be contacted 24 h a day in the case of an emergency. *Integrated Family Treatment* (72) in the U.S. offers a range of home-based services to families including psychoeducation and signposting to other forms of support. *VIA family* (94) in Denmark assigns families a case manager, and offers a range of supports including psychoeducation, *Triple P* (99), advocacy, social support, and liaison with schools. *Preventative Basic Care Management (PBCM)* (84) in the Netherlands also assigns families a case manager and coordinates the services involved in the families' care. The *BROSH program* (53) lasts 2 years and is a collaboration from child welfare, child mental health and adult mental health services is Israel. It consists of weekly home meetings either with the parent or the whole family where parents learn about child development, mentalizing skills, and can get help with financial issues. The children are also offered individual psychotherapy. The *Godparents programme* (71) takes a different approach, in which lay people are trained to perform the godparent role in Switzerland. They are assigned to a family for at least 3 years and act as another adult figure for the child and social support for the parent.

Groups for Parents With Mental Illness

Five interventions were designed as groups for parents with mental illness. *Living with Under Fives* (78) and *FWA Newpin* (70) are both designed for parents with children up to 5 years old and provide a space for the parent and child to play together alongside other families. *Living with Under Fives* also offers components on psychoeducation, parenting skills, budgeting, and links parents with other agencies. Shor et al. (89) describe a long-term therapeutic group for parents where they can raise parenting issues and give each other advice. The primary aim of *Young SMILES* (97) is to improve the quality of life of children affected by parental mental illness by teaching children about mental illness, recognizing stress, and accessing support networks. It includes a parent group that has components on supporting their children and successful family communication. *SEEK* (86) was developed as a compulsory part of treatment for parents with mental illness whose children are currently in inpatient treatment. It involves psychoeducation on mental illness, talking to children about mental illness, and family stress.

Family Therapy

Three interventions were focused on providing family therapy. The *Think Family Whole Family Programme* (90) is based on the Meriden Family Programme (102), which is a behavioral family intervention that teaches communication and problem-solving skills. The *Think Family Whole Family Programme* adds further elements about parental mental illness. The *Child Resilience Program* (56) provides family therapy with separate parent and child groups, as well as sessions on psychoeducation, parenting skills, and building resilience. Becker et al. (60) briefly describes a counseling and support service for the whole family.

Evaluations of Interventions

Twenty-three out of the 38 included studies of interventions had some kind of quantitative evaluation of parent or child outcomes, and 13 studies involved a qualitative evaluation of acceptability from the parents. Eight studies had both a quantitative and qualitative evaluation.

Table 3 lists the demographic details of participants. All interventions had more female participants than male. In all studies apart from Wolfenden (93) and Strand and Meyersson (68), in which every participant had a psychotic diagnosis, the proportion of participants with a psychotic diagnosis ranged between 0 and 42.5%, or was unknown. There were in total at least 53 participants with a psychotic diagnosis in the studies with a quantitative evaluation, and at least 60 in the studies with a qualitative evaluation.

Table 4 lists the studies that contained completed evaluations or protocols for evaluations, and reports their design, outcome measures used, and qualitative results.

Quantitative Evaluations

Out of the 23 quantitative evaluations, 11 had a control group and only eight randomly assigned the participants to the control or intervention group. Out of these eight randomized control trials (RCTs), five were protocols. The three completed RCTs evaluated *PBCM*, (85), the *Parenting Internet Intervention* (80), and *Young SMILES* (97). The number of participants in completed studies ranged from eight to 99.

Most interventions had an outcome measure for both the parent and the child. The interventions that only involved the use of a measure for the parent included *Family Options* (63), *Let's Talk about Children* in both the face-to-face and booklet format (75, 77), *Parenting with Success and Satisfaction* (82), *Tuning into Kids* (38), The *Lighthouse (Leuchtturm) Parenting Programme* (88), and the *Parenting Internet Intervention* (80). There was very little consistency in terms of which outcome measures were used. For example, while both *Child Talks+* and *Let's Talk about Children* aimed to enable the parent to explain their mental illness to their child, *Child Talks+* included six child outcome measures and two parent measures on communication and self-efficacy (57) while *Let's Talk about Children* only used measures on parenting stress and family functioning (75). There was also variation in which measure each study had seen an improvement. For example, *You are Okay* (95) and *Family Talk* (65) appeared to have an impact on child behavior, whilst *KopOpOuders* and *Mental Health Triple P* appeared to have improved parenting skills.

Randomized controlled trials are the gold standard for the assessment of effectiveness, with non-randomized trials or trials without a control group being susceptible to a range of sources of bias (104). Three RCTs were included in this review. *Young SMILES* did not conduct significance testing or report effect sizes as it was a feasibility trial. The other two RCTs, *Preventative Basic Care Management* and the *Parenting Internet Intervention* both showed improvement on parenting measures of skills and self-efficacy (80, 85). *Preventative Basic Care Management* reported improvement on the parenting subscale of the Family Functioning Questionnaire (85). The *Parenting Internet Intervention* showed improvement on two measures of parenting: Healthy Families Parenting Inventory and Parenting Sense of Competence Scale, but not on the Medical Outcomes Study—Social Support Survey (80). The *Parenting Internet Intervention* did not include any child outcome measures (80). *Preventative Basic Care Management* measured child behavior using the Strengths and Difficulties Questionnaire, but did not find any significant differences between the intervention and control group following the intervention (85).

Qualitative Evaluations of Acceptability

Table 4 provides a narrative summary of the qualitative results of the included reports. Thirteen studies involved a qualitative evaluation with eight reporting themes. Parents reported in eight out of 13 studies that they felt they could communicate more easily with their children about parental mental illness after receiving the intervention. This included two studies reporting on the *Family Talk* intervention (67, 68), both studies on *Let's Talk About Children* (75, 77) and *KidsTime* (74), in which the aim of the intervention is to enhance communication. Parents in five out of 13 studies felt their parenting had improved following the intervention, which includes four studies in which the aim was to enhance parenting skills, two *Triple P* studies (93, 103), *Tuning into Kids* (38), the *Lighthouse (Leuchtturm) Parenting Programme* (88), as well as Shor et al.'s (89) therapeutic group. Parents in seven out of 13 studies reported that they understood, and could focus, on their children's needs more. Parents in one evaluation of *Family Talk* said that the intervention played an important part in their recovery (67).

For the six interventions that were held in a group format, parents all commented on how they enjoyed being in a group with other parents who have experienced similar difficulties, although some of the parents who took part in *Young SMILES* reported they felt anxious and pressured about attending. The parents in *Mental Health Triple P* also commented that they enjoyed the home visits (103).

These results suggest that most interventions have a good level of acceptability to parents, and there was also appreciation for different intervention formats including groups and home visiting.

Parents in four studies highlighted potential improvements on structure of the intervention. In the *Family Talk* intervention for parents with psychosis, parents said they would have preferred an intervention where their illness was not the focus (68). Some parents who received the *Let's Talk about Children* booklet found it upsetting (77). In *Young SMILES*, parents felt there was too much emphasis on their child and not enough on them as a parent, and one parent reported that the focus on her mental illness felt damaging (97). Parents in the *Lighthouse (Leuchtturm) Parenting Programme* stated they wanted a higher number of sessions which were longer in duration (88). In two out of 13 studies, parents spoke about the next stages, which included wanting more support for their children in schools (74) and wanting to help their child regulate emotions better (75).

DISCUSSION

Summary of Evidence

This scoping review involved a systematic search of relevant databases and other sources to establish what a parenting intervention for parents with psychosis might look like. The three aims of this review were to determine (1) what parenting interventions were available for parents with psychosis, (2) what components these interventions provided, and (3) what kinds of evaluations had been undertaken, and what they showed in terms of outcomes. Thirty-eight studies were included which described 34 interventions.

What Parenting Interventions Are Available for Parents With Psychosis?

Thirty-four interventions were described, of which most were designed for either parents with mental illness or parents with severe mental illness. When parents with psychotic diagnoses were included in these interventions, there were often in the minority compared to parents with other diagnoses. Both researchers (105) and parents diagnosed with mental illness (106) have recommended the use of diagnostic-specific groups, and recently, RCTs of parenting interventions for parents with anxiety (42) and with borderline personality disorder (43) have been conducted, and report promising results. In this review, only two interventions focused solely on parents with a psychotic diagnosis, and both had a sample size of 10 participants or fewer. These were *Family Talk* (68) and *Triple P* (93), both of which were unchanged from their usual delivery format. It may be the case that parents with psychosis would benefit from specific additions to parenting interventions, like safety planning for acute episodes (107), or a focus on regaining self-confidence during periods of stabilization (108).

Parents with mental illness often want their family to be involved in their treatment (21), and parenting can be a valued part of one's personal recovery (27). Reflecting this desire, most interventions in this review were designed either for the parent with a mental illness and their child, or for the whole family, which typically included the parent with a mental illness, their children, their partner, and sometimes additional family members. When interventions were designed solely for the parent, they were often delivered in a group format. Parents with mental illness can often face social isolation (14), and an intervention in a group setting could be one way of alleviating this. Parents with psychosis, specifically asked for a group intervention in order to be able to meet others in a similar situation, share parenting tips, and find social support (109). However, parents in the *Young SMILES* intervention found that attending a group can also be anxiety provoking (97).

Despite the fact that these parents can face poor social and emotional support, only a few interventions incorporated peer support, where someone who has also experienced poor mental health is involved in delivering the intervention (110). Having parent peers involved in delivering parenting interventions may help alleviate the lack of social support, and could also help to reduce the stigma felt by parents (111).

When considering the availability of interventions, it is important to note that geography is one of the biggest limiting factors in terms of which interventions parents can access. The 38 studies included in this review came from 14 countries, the majority of which were from Australia, who have also been a leader in policy advancement for parents with mental illness and their children for the last 20 years (112). As well as integrating interventions in mental health and social care services, the parenthood status of patients must be identified. This has been done well in Norway where, alongside the *Child Talks+* intervention, an assessment form has also been implemented to improve recording and identification of patients' dependants (113). It is not enough for these interventions to be developed and tested, they need to be recommended in policy and made available to the parents who would benefit from them.

What Are the Components of These Interventions?

The interventions identified in this review were grouped into five categories, depending on the cluster of their components. It is important to consider which of these five categories of interventions best address the needs of parents with psychosis.

The largest group, which consisted of 11 interventions, had a focus on improving parenting skills, and the one RCT, Kaplan et al.'s (80) *Parenting Internet Intervention*, demonstrated improvement on measures of parenting satisfaction and coping skills. Parents with psychosis have demonstrated difficulties in reflective functioning and parental sensitivity (6, 14, 114), and this is particularly true for individuals with a higher severity of illness (12, 115). However, parents with psychosis and their families may need more support that goes beyond just addressing parenting skills.

The children affected by parental mental illness have expressed a desire for their parent's symptoms to be explained to them (19, 21), and the second largest group of interventions was developed in response to this need. Eight interventions had a focus on explaining mental illness to the children. Often, they also included psychoeducation about the effects of parental mental illness on the child. Additionally, these interventions provided an opportunity for the children and, sometimes, the parent's partner, to talk about their experiences of parental mental illness. However, psychoeducation about parental mental illness alone may not be sufficient to bring about positive change for the parent or for their child (116). Parents with psychosis who participated in *Family Talk* stated that they wanted less focus on the effects of their illness (68), and parents who had participated in *Young SMILES* stated they wanted more parenting components, and not solely a focus on their children (97).

The third largest group consisted of seven long-term whole family interventions, which typically lasted longer than the other interventions, and were more holistic. These often involve case management, whereby the family receives continuous care from one individual, interagency collaboration and links with other supportive agencies. Often crisis planning for potential relapses is also incorporated, as well as help with other difficulties that affect these families, such as financial issues. An example of one of these interventions is *VIA Family*, which had multiple stages. First the family is introduced to the intervention, then a life history is taken, and the family received psychoeducation. Then *Triple P* is offered and, finally parent and children groups are provided. Throughout the intervention, there are many optional extras, such as psychological treatment for the child's mental health difficulties, advice on finances, and social support for the parent (94).

The needs of parents with psychosis are often complex and diverse. Parents with severe mental illnesses have reported difficulties with practical issues such as finances and household tasks as well as fears about custody loss (25). Parents with psychosis and their families additionally struggle with parenting skills (6, 14, 117), self-confidence (109), and relapse of symptoms and subsequent hospitalization (117). Furthermore, these needs may be different during acute episodes of psychosis and periods of stability (108, 117). Therefore, interventions that solely address parenting skills or aim to explain mental illness to the children of these parents are likely to be insufficient, and more holistic long-term interventions may be the most suitable to address the needs of this group of parents. However, a more complex intervention will come with higher costs. Only *Preventative Basic Care Management* has been subjected to a cost-effectiveness evaluation (118). The authors stated that the intervention was more costly than care as usual, but could not conclude whether it was cost-effective or not (118). Identification of the essential components needed to enhance the well-being of these parents and their families is needed to enable us to implement effective interventions both in terms of psychosocial and economic outcomes.

It is also necessary to note that inpatient facilities in Germany often provide many components described in this study, such as selfcare, peer support, and signposting, as part of routine inpatient treatment (119) and that those receiving the *SEEK* intervention (86) and the *Lighthouse Parenting Programme* (88) will have also benefitted from these elements.

What Kinds of Evaluations Have Been Conducted to Determine the Acceptability and Effectiveness of Interventions for Parents With Psychosis and What Do They Show?

Parenting interventions for parents with mental illness are relatively new, and as such have an emerging evidence base. Around two-thirds of the interventions described in this review had been evaluated in some way, and only eight of these evaluations were RCTs, with only three having results available. One of these RCTs, *Young SMILES* (97), did not conduct significance testing since it was a feasibility trial. The other two, *Preventative Basic Care Management* (85) and Kaplan et al.'s (80) *Parenting Internet Intervention*, demonstrated significant differences between the parents in the intervention and control groups on measures of parenting. Therefore, it seems there is initial evidence that parenting interventions for parents with mental illness can improve aspects of parenting, such as skills and self-efficacy.

Children of parents with any kind of mental health diagnosis are more likely than children without parental mental illness to exhibit internalizing and externalizing problems (16) and are at risk of developing a mental health problem (120, 121). While, in theory, enhancing parenting skills should improve the child's quality of life and later psychosocial health (122), it is nevertheless still important to assess changes in children's functioning following such intervention. The RCT with the longest follow-up in this review was *Preventative Basic Care Management* (85), and did not report any difference in child behavior between the intervention and control group after 18 months of intervention. There is therefore, currently a lack of evidence demonstrating the effectiveness of parenting interventions in producing positive outcomes for the children of parents with mental illness. The longest two RCTs that are currently taking place are *VIA Family* (94) and *Triple P* combined with CBT (37), and it will be noteworthy to see if these interventions have any impact on children's functioning at follow-up.

Thirteen studies involved a qualitative evaluation of a parenting intervention. Most studies reported positive comments made by parents on intervention content and format, indicating that most interventions have a good level of acceptability. However, some parents who received the *Let's Talk about Children booklet* found it upsetting (77), which highlights the importance of parents with mental illness being supported by a professional during the delivery of parenting interventions. Parents in the *Family Talk* intervention and *Young SMILES* wanted less focus on their mental health (68, 97), and parents in *Young SMILES* also wanted more focus on them as a parent rather than solely on their child (97). These results suggest that interventions should be careful not to stigmatize or blame parents, and should recognize the centrality of their identity as a parent (27).

STRENGTHS AND LIMITATIONS

This review has updated the results from the reviews conducted by Schrank et al. (47) and Suarez et al. (48), which identified fifteen and nine interventions, respectively. In contrast to Schrank et al. (47) and Suarez et al. (48), this review did not set a limit for what proportion of the study sample needed a psychotic diagnosis, and included interventions that had not yet been evaluated. Additionally, many of the interventions included in this review have been published in the 5 years since Schrank et al. (47) and Suarez et al. (48) conducted their reviews. Since this review did not solely include interventions which had been tested with a certain proportion of parents with a psychotic disorder, it identified many interventions that could be helpful for parents with psychosis and their families.

Scoping reviews do not necessarily need a quality assessment (50). However, one limitation of this review is that the lack of quality assessment means the results of the studies included in this review are not contextualized alongside an assessment of their risk of bias. The main limitation of this review is that it only included papers that are published in English or German. Fifty reports were rejected at full-text review due to being written in another language, and it is likely that some would have been eligible for inclusion in this review. Another limitation relates to how we identified the components of each intervention, in which we only extracted the components that had been described in the report of each study, some of which did not always contain much detail. It may well be the case, therefore, that some interventions included more components than indicated in **Table 2**.

FUTURE DIRECTIONS

Future research needs to investigate which components are the most effective in improving outcomes for both the parent and the child. The needs of parents with psychosis and their families are complex, and it is not sufficient for interventions to aim solely to enhance parenting skills or explain mental illness to their children. Only two interventions in this review were conducted exclusively with parent participants with a diagnosis of psychosis (68, 93), and yet they had been unchanged from their usual delivery format and therefore not tailored toward the needs of parents with psychosis. Interventions must attempt to address practical issues, periods of unplanned hospitalization, and parents' own self-confidence and self-efficacy.

When addressing parenting skills, a psychotic diagnosis does predict deficits in social cognitive abilities (45), which affects parents' ability to understand their child's mental states (114). Therefore, parents with psychosis would likely benefit from interventions with a mentalizing component, which was the case in four interventions included in this review (38, 53, 70, 88).

When interventions did include parents with a psychotic diagnosis in their evaluation, they were often in the minority compared to parents with other mental health conditions. Interventions which are designed for parents with any kind of mental illness should endeavor to include more parents with a psychotic diagnosis when evaluating the intervention in order to determine whether these interventions are indeed effective for those with more severe mental illnesses, like psychosis.

It is promising that some of the interventions in this review are currently being tested in an RCT. As well as testing interventions, we must investigate what types of interventions are most effective, in order to produce evidence-based and cost-effective programs.

CONCLUSIONS

Many parenting interventions exist for parents who have experienced mental illness, from which parents with a diagnosis of psychosis and their families may benefit, however no intervention has been developed and evaluated to specifically support parents with psychosis and their families. Five categories of intervention were identified, reflecting their key components. The two largest categories were "talking about parental mental illness" and "improving parenting skills." The third category described holistic long-term interventions targeting the whole family, and which often involved the provision of a wide range of components, with implications in terms of cost. Of the 34 studies included in this review, only two RCTs provided evidence for the potential effectiveness of the parenting interventions, thereby highlighting the significant evidence gap. In order to help parents who have experienced psychosis and their families, we need to know which components are effective in improving outcomes for both the parent and their children, and whether any psychosis-specific components would benefit these families.

DATA AVAILABILITY STATEMENT

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found below: https://osf.io/z4rpn.

AUTHOR CONTRIBUTIONS

JR: conceptualization, designing the study, selection of sources of evidence, data extraction, data analysis, and writing the manuscript. NS: selection of sources of evidence and data extraction. BM: selection of sources of evidence, data extraction, reviewing, and editing the manuscript. M-LK and FH: selection of sources of evidence. RD: data extraction. LJ and JB: conceptualization, designing the study, supervision, reviewing, and editing manuscript. All authors contributed to the article and approved the submitted version.

FUNDING

JR is a D.Phil. student and is funded by Mental Health Research UK.

ACKNOWLEDGMENTS

The authors thank Karine Barker, lead librarian for non-medical sciences at the University of Oxford, for her guidance with searching journal databases and searching for gray literature.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.787166/full#supplementary-material

REFERENCES

1. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders (DSM-5)*. American Psychiatric Pub (2013). doi: 10.1176/appi.books.9780890425596
2. Myin-Germeys I, Delespaul PA, Van Os J. Behavioral sensitization to daily life stress in psychosis. *Psychol Med.* (2005) 35:733–41. doi: 10.1017/S0033291704004179
3. Lally J, MacCabe JH. Antipsychotic medication in schizophrenia: a review. *Br Med Bull.* (2015) 114:169–79. doi: 10.1093/bmb/ldv017
4. Seeman M V. Schizophrenia and motherhood. In: Reupert AE, Maybery DM, Nicholson J, Gopfert M, Seeman M V, editors. *Parental Psychiatric Disorder Distressed Parents and Their Families. 3rd ed.* Cambridge: Cambridge University Press (2015). p. 107–16. doi: 10.1017/CBO9781107707559.012
5. Campbell LE, Poon AWC. Parenting challenges for persons with a serious mental illness. *Ment Heal Soc Work.* (2020). p. 457–74. doi: 10.1007/978-981-13-6975-9_16
6. Wan MW, Salmon MP, Riordan DM, Appleby L, Webb R, Abel KM. What predicts poor mother-infant interaction in schizophrenia? *Psychol Med.* (2007) 37:537–46. doi: 10.1017/S0033291706009172
7. Bebbington P. Childhood sexual abuse and psychosis: aetiology and mechanism. *Epidemiol Psichiatr Soc.* (2009) 18:284–93. doi: 10.1017/S1121189X00000233
8. Varese F, Smeets F, Drukker M, Lieverse R, Lataster T, Viechtbauer W, et al. Childhood adversities increase the risk of psychosis: a meta-analysis of patient-control, prospective-and cross-sectional cohort studies. *Schizophr Bull.* (2012) 38:661–71. doi: 10.1093/schbul/sbs050
9. Van Wert M, Anreiter I, Fallon BA, Sokolowski MB. Intergenerational transmission of child abuse and neglect: a transdisciplinary analysis. *Gend Genome.* (2019) 3:247028971982610. doi: 10.1177/2470289719826101
10. Jungbauer J, Stelling K, Kuhn J, Lenz A. How do mothers and fathers suffering from schizophrenia experience their parenthood? Results from an in-depth interview study. *Psychiatr Prax.* (2010) 37:233–9. doi: 10.1055/s-0029-1223535
11. Abel KM, Hope H, Faulds A, Pierce M. Promoting resilience in children and adolescents living with parental mental illness (CAPRI): children are key to identifying solutions. *Br J Psychiatry.* (2019) 215:513–5. doi: 10.1192/bjp.2019.118
12. Campbell LE, Hanlon MC, Galletly CA, Harvey C, Stain H, Cohen M, et al. Severity of illness and adaptive functioning predict quality of care of children among parents with psychosis: a confirmatory factor analysis. *Aust N Z J Psychiatry.* (2018) 52:435–45. doi: 10.1177/0004867417731526
13. Ranning A, Laursen TM, Thorup AAE, Hjorthøj C, Nordentoft M. Children of parents with serious mental illness: with whom do they grow up? A prospective, population-based study. *J Am Acad Child Adolesc Psychiatry.* (2016) 55:953–61. doi: 10.1016/j.jaac.2016.07.776
14. Abel KM, Webb RT, Salmon MP, Wan MW, Appleby L. Prevalence and predictors of parenting outcomes in a cohort of mothers with schizophrenia admitted for joint mother and baby psychiatric care in England. *J Clin Psychiatry.* (2005) 66:781–9. doi: 10.4088/JCP.v66n0618
15. Topor A, Andersson G, Denhov A, Holmqvist S, Mattsson M, Stefansson CG, et al. Psychosis and poverty: coping with poverty and severe mental illness in everyday life. *Psychosis.* (2014) 6:117–27. doi: 10.1080/17522439.2013.790070
16. Johnson SE, Lawrence D, Perales F, Baxter J, Zubrick SR. Prevalence of mental disorders among children and adolescents of parents with self-reported mental health problems. *Community Ment Health J.* (2018) 54:884–97. doi: 10.1007/s10597-017-0217-5
17. Luciano A, Nicholson J, Meara E. The economic status of parents with serious mental illness in the United States. *Psychiatr Rehabil J.* (2014) 37:242–50. doi: 10.1037/prj0000087
18. Kahng SK, Oyserman D, Bybee D, Mowbray CT. Mothers with serious mental illness: when symptoms decline does parenting improve? *J Fam Psychol.* (2008) 22:162–6. doi: 10.1037/0893-3200.22.1.162
19. Gladstone BM, Boydell KM, Seeman MV, Mckeever PD. Children's experiences of parental mental illness: a literature review. *Early Interv Psychiatry.* (2011) 5:271–89. doi: 10.1111/j.1751-7893.2011.00287.x
20. Hinden BR, Biebel K, Nicholson J, Mehnert L. The invisible children' s project: key ingredients of an intervention for parents with mental illness. *J Behav Health Serv Res.* (2005) 32:393–408. doi: 10.1097/00075484-200510000-00006
21. Wahl P, Bruland D, Bauer U, Okan O, Lenz A. What are the family needs when a parent has mental health problems? Evidence from a systematic literature review. *J Child Adolesc Psychiatr Nurs.* (2017). 30:54–66. doi: 10.1111/jcap.12171
22. Van Der Zanden RAP, Speetjens PAM, Arntz KSE, Onrust SA. Online group course for parents with mental illness: development and pilot study. *J Med Internet Res.* (2010). 12:e50. doi: 10.2196/jmir.1394
23. Park JM, Solomon P, Mandell DS. Involvement in the child welfare system among mothers with serious mental illness. *Psychiatr Serv.* (2006) 57:493–7. doi: 10.1176/ps.2006.57.4.493
24. Kaplan K, Brusilovskiy E, O'Shea AM, Salzer MS. Child protective service disparities and serious mental illnesses: results from a national survey. *Psychiatr Serv.* (2019) 70:202–8. doi: 10.1176/appi.ps.201800277
25. Dolman C, Jones I, Howard LM. Pre-conception to parenting: a systematic review and meta-synthesis of the qualitative literature on motherhood for women with severe mental illness. *Arch Womens Ment Health.* (2013) 16:173–96. doi: 10.1007/s00737-013-0336-0
26. Khalifeh H, Murgatroyd C, Freeman M, Johnson S, Killaspy H. Home treatment as an alternative to hospital admission for mothers in a mental health crisis : a qualitative study. *Psychiatr Serv.* (2009) 60:634–9. doi: 10.1176/ps.2009.60.5.634
27. Price-Robertson R, Obradovic A, Morgan B. Relational recovery: beyond individualism in the recovery approach. *Adv Ment Heal.* (2017) 15:108–20. doi: 10.1080/18387357.2016.1243014
28. Wyder M, Bland R. The recovery framework as a way of understanding families' responses to mental illness: balancing different needs and recovery journeys. *Aust Soc Work.* (2014) 67:179–96. doi: 10.1080/0312407X.2013.875580
29. Krumm S, Becker T, Wiegand-Grefe S. Mental health services for parents affected by mental illness. *Curr Opin Psychiatry.* (2013) 26:362–8. doi: 10.1097/YCO.0b013e328361e580
30. Reedtz C, Lauritzen C, Stover Y V, Freili JL, Rognmo K. Identification of children of parents with mental illness: a necessity to provide relevant support. *Front Psychiatry.* (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
31. Cooklin A. Promoting children's resilience to parental mental illness: engaging the child's thinking. *Adv Psychiatr Treat.* (2013) 19:229–40. doi: 10.1192/apt.bp.111.009050
32. Butler J, Gregg L, Calam R, Wittkowski A. Parents' perceptions and experiences of parenting programmes: a systematic review and metasynthesis of the qualitative literature. *Clin Child Fam Psychol Rev.* (2020) 23:176–204. doi: 10.1007/s10567-019-00307-y
33. Mytton J, Ingram J, Manns S, Thomas J. Facilitators and barriers to engagement in parenting programs: a qualitative systematic review. *Heal Educ Behav.* (2014) 41:127–37. doi: 10.1177/1090198113485755

34. Barlow J, Coren E. The effectiveness of parenting programs: a review of campbell reviews. *Res Soc Work Pract.* (2018) 28:99–102. doi: 10.1177/1049731517725184

35. Pidano AE, Allen AR. The incredible years series: a review of the independent research base. *J Child Fam Stud.* (2015) 24:1898–916. doi: 10.1007/s10826-014-9991-7

36. Phelan R, Howe DJ, Cashman EL, Batchelor SH. Enhancing parenting skills for parents with mental illness: the Mental Health Positive Parenting Program. *Med J Aust.* (2012) 199:S30–3. doi: 10.5694/mja11.11181

37. Stracke M, Gilbert K, Kieser M, Klose C, Krisam J, Ebert DD, et al. CoMPARE family (Children of mentally ill parents at risk evaluation): a study protocol for a preventive intervention for children of mentally ill parents (Triple P, evidence-based program that enhances parentings skills, in addition to gold-standard CBT CBT with the mentally ill parent) in a multicenter RCT-Part II. *Front Psychiatry.* (2019). 10:54. doi: 10.3389/fpsyt.2019.00054

38. Isobel S, Meehan F, Pretty D. An emotional awareness based parenting group for parents with mental illness: a mixed methods feasibility study of community mental health nurse facilitation. *Arch Psychiatr Nurs.* (2016) 30:35–40. doi: 10.1016/j.apnu.2015.10.007

39. Beardslee WR, Hoke L, Wheelock I, Rothberg PC, Van de Velde P, Sawtling S. Initial findings on preventive intervention for families with parental-affective disorders. *Am J Psychiatry.* (1992). 149:1335–1340. doi: 10.1176/ajp.149.10.1335

40. Bee P, Bower P, Byford S, Churchill R, Calam R, Stallard P, et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. *Health Technol Assess.* (2014). 18:1–250. doi: 10.3310/hta18080

41. Reupert AE, Maybery DJ. What do we know about families where parents have a mental illness? A systematic review. *Child Youth Serv.* (2016) 37:98–111. doi: 10.1080/0145935X.2016.1104037

42. Cartwright-Hatton S, Ewing D, Dash S, Hughes Z, Thompson EJ, Hazell CM, et al. Preventing family transmission of anxiety: feasibility RCT of a brief intervention for parents. *Br J Clin Psychol.* (2018) 57:351–66. doi: 10.1111/bjc.12177

43. Day C, Briskman J, Crawford MJ, Foote L, Harris L, Boadu J, et al. Randomised feasibility trial of the helping families programme-modified: an intensive parenting intervention for parents affected by severe personality difficulties. *BMJ Open.* (2020) 10:1–12. doi: 10.1136/bmjopen-2019-033637

44. Gearing RE, Alonzo D, Marinelli C. Maternal schizophrenia: psychosocial treatment for mothers and their children. *Clin Schizophr Relat Psychoses.* (2012). 6:27–33B. doi: 10.3371/CSRP.6.1.4

45. Achim AM, Ouellet R, Roy MA, Jackson PL. Mentalizing in first-episode psychosis. *Psychiatry Res.* (2012) 196:207–13. doi: 10.1016/j.psychres.2011.10.011

46. Radley J, Grant C, Barlow J, Johns L. Parenting interventions for people with schizophrenia or related serious mental illness (Review). *Cochrane Database Syst Rev.* 2020:CD013536. doi: 10.1002/14651858.CD013536.pub2

47. Schrank B, Moran K, Borghi C, Priebe S. How to support patients with severe mental illness in their parenting role with children aged over 1 year? A systematic review of interventions. *Soc Psychiatry Psychiatr Epidemiol.* (2015) 50:1765–83. doi: 10.1007/s00127-015-1069-3

48. Suarez EB, Lafrenière G, Harrison J. Scoping review of interventions supporting mothers with mental illness: key outcomes and challenges. *Community Ment Health J.* (2016) 52:927–36. doi: 10.1007/s10597-016-0037-z

49. Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation. *Ann Intern Med.* (2018) 169:467–73. doi: 10.7326/M18-0850

50. Arksey H, O'Malley L. Scoping studies: towards a methodological framework. *Int J Soc Res Methodol Theory Pract.* (2005) 8:19–32. doi: 10.1080/1364557032000119616

51. Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan-a web and mobile app for systematic reviews. *Syst Rev.* (2016) 5:1–10. doi: 10.1186/s13643-016-0384-4

52. Cohen J, A. coefficient of agreement for nominal scales. *Educ Psychol Meas.* (1960) 20:37–46. doi: 10.1177/001316446002000104

53. Oppenheim-Weller S, Shtark T, Aldor R. Families with parental mental illness: studying a home-based intervention program. *Child Fam Soc Work.* (2021) 26:617–28. doi: 10.1111/cfs.12843

54. Cowling V, Garrett M. Child and family inclusive practice: a pilot program in a community adult mental health service. *Australas Psychiatry.* (2009) 17:279–82. doi: 10.1080/10398560902840232

55. Cowling V, Garrett M. A child-inclusive family intervention in a community adult mental health service. *Aust New Zeal J Fam Ther.* (2012) 33:101–13. doi: 10.1017/aft.2012.13

56. Gutjahr A. *Child Resilience Program an Intervention for Children of Chronically Mentally Ill Parents*. Ph.D. thesis, Spalding University (2007).

57. Reedtz C, van Doesum KT, Signorini G, Lauritzen C, van Amelsvoort T, van Santvoort F, et al. Promotion of wellbeing for children of parents with mental illness: a model protocol for research and intervention. *Front Psychiatry.* (2019). 10:606. doi: 10.3389/fpsyt.2019.00606

58. Van Doesum KT, Lauritzen C, Reedtz C. *Child Talks+ Manual. Regional Centre for Child and Youth Mental Health and Child Welfare.* Tromso: The Arctic University of Norway (2020). Available online at: https://uit.no/Content/713118/cache=20210401114628/UiT_RKBU-Nord_A4_manual_ENG_web.pdf (accesesed April 27, 2021).

59. Wiegand-Grefe S, Filter B, Busmann M, Kilian R, Kronmüller KT, Lambert M, et al. Evaluation of a family-based intervention program for children of mentally ill parents: study protocol for a randomized controlled multicenter trial. *Front Psychiatry.* (2021). 11:561790. doi: 10.3389/fpsyt.2020.561790

60. Becker T, Kilian S, Killian R, Lahmeyer C, Krumm S. Family needs, children and parenthood in people with mental illness. *Eur Psychiatry.* (2009) 24:S48. doi: 10.1016/S0924-9338(09)70281-0

61. Solantaus T, Toikka S. The effective family programme: preventative services for the children of mentally ill parents in Finland. *Int J Ment Health Promot.* (2006) 8:37–44. doi: 10.1080/14623730.2006.9721744

62. Solantaus T, Reupert AE, Maybery DJ. Working with parents who have a psychiatric disorder. In: Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman M V, editors. *Parental Psychiatric Disorder Distressed Parents and Their Families*. 3rd ed. Cambridge: Cambridge University Press (2015). p. 238–47. doi: 10.1017/CBO9781107707559.023

63. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Developing family options: outcomes for mothers with severe mental illness at twelve months of participation. *Am J Psychiatr Rehabil.* (2016) 19:353–69. doi: 10.1080/15487768.2016.1231639

64. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Family options for parents with mental illnesses: a developmental, mixed methods pilot study. *Psychiatr Rehabil J.* (2009) 33:106–14. doi: 10.2975/33.2.2009.106.114

65. Christiansen H, Anding J, Schrott B, Röhrle B. Children of mentally ill parents - a pilot study of a group intervention program. *Front Psychol.* (2015). 6:1494. doi: 10.3389/fpsyg.2015.01494

66. Furlong M, McGilloway S, Mulligan C, McGuinness C, Whelan N. Family Talk versus usual services in improving child and family psychosocial functioning in families with parental mental illness (PRIMERA—Promoting Research and Innovation in Mental hEalth seRvices for fAmilies and children): study protocol for a randomi. *Trials.* (2021) 22:1–18. doi: 10.1186/s13063-021-05199-4

67. Pihkala H, Cederström A, Sandlund M. Beardslee's preventive family intervention for children of mentally ill parents: a swedish national survey. *Int J Ment Health Promot.* (2010) 12:29–38. doi: 10.1080/14623730.2010.9721804

68. Strand J, Meyersson N. Parents with psychosis and their children: experiences of Beardslee's intervention. *Int J Ment Health Nurs.* (2020) 26:908–20. doi: 10.1111/inm.12725

69. Strand J, Rudolfsson L, A. qualitative evaluation of professionals' experiences of conducting Beardslee's family intervention in families with parental psychosis. *Int J Ment Health Promot.* (2017) 19:289–300. doi: 10.1080/14623730.2017.1345690

70. Lederer J, McHugh M, FWA. Newpin - working with parents with mental health problems and their young children. *Ment Heal Rev J.* (2006) 11:23–7. doi: 10.1108/13619322200600038

71. Mueller B, Fellmann L. Supporting children of parents with mental health problems through professionally assisted lay support–the

"godparents" program. *Child Youth Serv.* (2019) 40:23–42. doi: 10.1080/0145935X.2018.1526071

72. Brunette MF, Richardson F, White L, Bemis G, Eelkema RE. Integrated family treatment for parents with severe psychiatric disabilities. *Psychiatr Rehabil J.* (2004) 28:177–80. doi: 10.2975/28.2004.177.180
73. OurTime. *KidsTime Workshop Manual*. London: OurTime (2020).
74. Ford DM. *The 'Kidstime' Intervention for Children of Parents With Mental Illness: An Exploration of the Experience of the 'Kidstime' Workshops and Relevant School-Based Support*. Ph.D. thesis. (2019).
75. Maybery DJ, Goodyear M, Reupert AE, Sheen J, Cann W, O'Hanlon B, et al. A mixed method evaluation of an intervention for parents with mental illness. *Clin Child Psychol Psychiatry.* (2019) 24:717–27. doi: 10.1177/1359104518822676
76. Maybery DJ, Goodyear M, Reupert AE, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry.* (2017). 17:198. doi: 10.1186/s12888-017-1357-4
77. Cooper V, Reupert AE. "Let's talk about children" resource: a parallel mixed method evaluation. *Soc Work Ment Health.* (2017) 15:47–65. doi: 10.1080/15332985.2016.1170090
78. Bassett H, Lampe J, Lloyd C. Living with under-fives: a programme for parents with a mental illness. *Br J Occup Ther.* (2001) 64:23–8. doi: 10.1177/030802260106400105
79. Bassett H, Lloyd C. At-risk families with mental illness: partnerships in practice. *New Zeal J Occup Ther.* (2005) 52:31–7.
80. Kaplan K, Solomon P, Salzer MS, Brusilovskiy E. Assessing an Internet-based parenting intervention for mothers with a serious mental illness: a randomized controlled trial. *Psychiatr Rehabil J.* (2014) 37:222–31. doi: 10.1037/prj0000080
81. van der Ende PC, Venderink MM, van Busschbach JT. Parenting with success and satisfaction among parents with severe mental illness. *Psychiatr Serv.* (2010) 61:416. doi: 10.1176/ps.2010.61.4.416
82. van der Ende PC. *Vulnerable Parenting, A Study on Parents With Mental Health Problems: Strategies and Support*. Groningen: Hanze University of Applied Sciences Groningen. (2016).
83. van der Ende PC, van Busschbach JT, Nicholson J, Korevaar EL, van Weeghel J. Parenting and psychiatric rehabilitation: can parents with severe mental illness benefit from a new approach? *Psychiatr Rehabil J.* (2014) 37:201–8. doi: 10.1037/prj0000067
84. Wansink HJ, Hosman CMH, Janssens JMAM, Hoencamp E, Willems WJCT. Preventive family service coordination for parents with a mental illness in the Netherlands. *Psychiatr Rehabil J.* (2014) 37:216–21. doi: 10.1037/prj0000073
85. Wansink HJ, Janssens JMAM, Hoencamp E, Middelkoop BJC, Hosman CMH. Effects of preventive family service coordination for parents with mental illnesses and their children, a RCT. *Fam Syst Heal.* (2015) 33:110–9. doi: 10.1037/fsh0000105
86. Fritz L-M, Domin S, Thies A, Yang J, Stolle M, Fricke C, et al. Profitieren psychisch erkrankte Eltern und psychisch belastete Kinder von einer gemeinsamen Eltern-Kind-Behandlung? *Kindheit und Entwicklung.* (2018) 27:253–67. doi: 10.1026/0942-5403/a000264
87. McFarland L, Fenton A. Unfogging the future: investigating a strengths-based program to build capacity and resilience in parents with mental illness. *Adv Ment Heal.* (2019) 17:21–32. doi: 10.1080/18387357.2018.1476065
88. Volkert J, Georg A, Hauschild S, Herpertz SC, Neukel C, Byrne G, et al. Bindungskompetenzen psychisch kranker Eltern stärken: adaptation und pilottestung des mentalisierungsbasierten leuchtturm-elternprogramms. *Prax Kinderpsychol Kinderpsychiatr.* (2019) 68:27–42. doi: 10.13109/prkk.2019.68.1.27
89. Shor R, Kalivatz Z, Amir Y, Aldor R, Lipot M. Therapeutic factors in a group for parents with mental illness. *Community Ment Health J.* (2015) 51:79–84. doi: 10.1007/s10597-014-9739-2
90. Gatsou L, Yates S, Goodrich N, Pearson D. The challenges presented by parental mental illness and the potential of a whole-family intervention to improve outcomes for families. *Child Fam Soc Work.* (2017) 22:388–97. doi: 10.1111/cfs.12254
91. Kuschel A, Granic M, Hahlweg K, Hartung D. ≪Nicht von schlechten Eltern!≫ Effekte einer therapieintegrierten Familienintervention. *Verhaltenstherapie.* (2016) 26:83–91. doi: 10.1159/000446170
92. Butler J, Gregg L, Calam R, Wittkowski A. Exploring staff implementation of a self-directed parenting intervention for parents with mental health difficulties. *Community Ment Health J.* (2021) 57:247–61. doi: 10.1007/s10597-020-00642-3
93. Wolfenden LL. *Parental Psychosis: Exploring Emotional and Cognitive Processes and the Feasibility of a Parenting Intervention*. Ph.D. thesis, University of Manchester (2018).
94. Müller AD, Gjøde ICT, Eigil MS, Busck H, Bonne M, Nordentoft M, et al. VIA family - a family-based early intervention versus treatment as usual for familial high-risk children: a study protocol for a randomized clinical trial. *Trials.* (2019) 20:1–17. doi: 10.1186/s13063-019-3191-0
95. Riemersma I, van Santvoort F, Van Doesum KT., Hosman CM., Janssens JMA, Van Der Zanden RAP, et al. 'You are Okay': effects of a support and educational program for children with mild intellectual disability and their parents with mental health concerns. *J Intellect Disabil.* (2020). 2020:174462952095376. doi: 10.1177/1744629520953765
96. Riemersma I, van Santvoort F, Janssens JMA, Hosman CM, van Doesum KT. "You are Okay": a support and educational program for children with mild intellectual disability and their parents with a mental illness: study protocol of a quasiexperimental design. *BMC Psychiatry.* (2015) 15:318. doi: 10.1186/s12888-015-0698-0
97. Abel KM, Bee P, Gega L, Gellatly J, Kolade A, Hunter D, et al. An intervention to improve the quality of life in children of parents with serious mental illness: the Young SMILES feasibility RCT. *Health Technol Assess.* (2020) 24:1–136. doi: 10.3310/hta24590
98. Gellatly J, Bee P, Gega L, Bower P, Hunter D, Stewart P, et al. A community-based intervention (Young SMILES) to improve the health-related quality of life of children and young people of parents with serious mental illness: randomised feasibility protocol. *Trials.* (2018) 19:550. doi: 10.1186/s13063-018-2935-6
99. Sanders MR. Triple P-positive parenting program as a public health approach to strengthening parenting. *J Fam Psychol.* (2008) 22:506–17. doi: 10.1037/0893-3200.22.3.506
100. Phelan R, Lee L, Howe D, Walter G. Parenting and mental illness: a pilot group programme for parents. *Aust Psychiatry.* (2006) 14:399–402. doi: 10.1080/j.1440-1665.2006.02312.x
101. Havighurst SS, Harley AE, Prior MR. Tuning into kids: an emotion-focused parenting program - initial findings from a community trial. *J Community Psychol.* (2009) 37:1008–23. doi: 10.1002/jcop.20345
102. Fadden G, Heelis R. The Meriden Family Programme: lessons learned over 10 years. *J Ment Heal.* (2011) 20:79–88. doi: 10.3109/09638237.2010.492413
103. Coates D, Phelan R, Heap J, Howe D. "Being in a group with others who have mental illness makes all the difference": the views and experiences of parents who attended a mental health parenting program. *Child Youth Serv Rev.* (2017). 78:104–11. doi: 10.1016/j.childyouth.2017.05.015
104. Higgins JP, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, et al. *Cochrane Handbook for Systematic Reviews of Interventions*. Chichester UK: John Wiley & Sons (2019). doi: 10.1002/9781119536604
105. Hosman CM, van Doesum KT, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: II. *Intervent. Aust. e-J. Adv Ment Heal.* (2009) 8:264–76. doi: 10.5172/jamh.8.3.264
106. Venkataraman MJ, Ackerson B. Parenting among mothers with bipolar disorder: strengths, challenges, and service needs. *J Fam Soc Work.* (2008) 11:389–408. doi: 10.1080/10522150802441825
107. Birchwood M, Spencer E, McGovern D. Schizophrenia: early warning signs. *Adv Psychiatr Treat.* (2000) 6:93–101. doi: 10.1192/apt.6.2.93
108. Radley J, Barlow J, Johns L. Mental health professionals' experiences of working with parents with psychosis and their families : a qualitative study. *BMC Health Serv Res.* (2021) 1:1–11. doi: 10.1186/s12913-021-06416-1
109. Radley J, Barlow J, Johns L. The needs and experiences of parents with psychosis: a qualitative interview study (2021). Submitted.

110. Repper J, Carter T, A. review of the literature on peer support in mental health services. *J Ment Heal.* (2011) 20:392–411. doi: 10.3109/09638237.2011.583947
111. Nicholson J, Valentine A. Key informants specify core elements of peer supports for parents with serious mental illness. *Front Psychiatry.* (2019) 10:106. doi: 10.3389/fpsyt.2019.00106
112. Nicholson J, Reupert AE, Grant A, Lees R, Maybery DJ, Mordoch E, et al. The policy context and change for families living with parental mental illness. In: Reupert AE, Maybery DJ, Joanne N, Gopfert M, Seeman MV, editors. *Parental Psychiatric Disorder Distressed Parents and Their Families*. Cambridge: Cambridge University Press (2015). p. 354–64. doi: 10.1017/CBO9781107707559.034
113. Lauritzen C, Reedtz C, Rognmo K, Nilsen MA, Walstad A. Identification of and support for children of mentally ill parents: a 5 year follow-up study of adult mental health services. *Front Psychiatry.* (2018) 9:507. doi: 10.3389/fpsyt.2018.00507
114. Mehta UM, Bhagyavathi HD, Kumar CN, Thirthalli J, Gangadhar BN. Cognitive deconstruction of parenting in schizophrenia: the role of theory of mind. *Aust N Z J Psychiatry.* (2014) 48:249–58. doi: 10.1177/0004867413500350
115. Røhder K, Nyström-Hansen M, MacBeth A, Davidsen KA, Gumley A, Brennan J, et al. Antenatal caregiving representations among expectant mothers with severe mental illness: a cross-sectional study. *J Reprod Infant Psychol.* (2019) 37:370–83. doi: 10.1080/02646838.2019.1578868
116. Marston N, Stavnes K, Van Loon LMA, Drost LM, Maybery DJ, Mosek A, et al. A content analysis of Intervention Key Elements and Assessments (IKEA): what's in the black box in the interventions directed to families where a parent has a mental illness? *Child Youth Serv.* (2016). 37:112–28. doi: 10.1080/0145935X.2016.1104041
117. Strand J, Boström PK, Grip K. Parents' descriptions of how their psychosis affects parenting. *J Child Fam Stud.* (2020) 29:620–31. doi: 10.1007/s10826-019-01605-3
118. Wansink HJ, Drost RMWA, Paulus, Aggie TG, Ruwaard D, Hosman CMH, et al. Cost-effectiveness of preventive case management for parents with a mental illness: a randomized controlled trial from three economic perspectives. *BMC Health Serv Res.* (2016). 16:228. doi: 10.1186/s12913-016-1498-z
119. Kivelkitz L, Dirmaier J, Harter M. Entscheidungshilfe - ambulante oder stationare behandlungs - moglichkeiten. *Psychenet.* (2020). Available online at: https://psychenet.de/de/entscheidungshilfen/ambulante-oder-stationaere-behandlungsmoeglichkeiten/das-versorgungssystem-behandlungsmoeglichkeiten/das-versorgungssystem-behandlungsmoeglichkeiten.html (accessed September 28, 2021).
120. Fekadu W, Mihiretu A, Craig TKJ, Fekadu A. Multidimensional impact of severe mental illness on family members: systematic review. *BMJ Open.* (2019) 9:1–12. doi: 10.1136/bmjopen-2019-032391
121. Dean K, Stevens H, Mortensen PB, Murray RM, Walsh E, Pedersen CB. Full spectrum of psychiatric outcomes among offspring with parental history of mental disorder. *Arch Gen Psychiatry.* (2010) 67:822–9. doi: 10.1001/archgenpsychiatry.2010.86
122. Waldmann T, Stiawa M, Dinc Ü, Saglam G, Busmann M, Daubmann A, et al. Costs of health and social services use in children of parents with mental illness. *Child Adolesc Psychiatry Ment Health.* (2021) 15:1–11. doi: 10.1186/s13034-021-00360-y

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 31 January 2022
doi: 10.3389/fpsyt.2021.806884

Development of an Evidence-Informed and Codesigned Model of Support for Children of Parents With a Mental Illness— "It Takes a Village" Approach

Melinda Goodyear[1,2]*, Ingrid Zechmeister-Koss[3], Annette Bauer[4], Hanna Christiansen[5], Martina Glatz-Grugger[6] and Jean Lillian Paul[6,7]

[1] School of Rural Health, Monash University, Melbourne, VIC, Australia, [2] Emerging Minds, National Workforce Centre for Child Mental Health, Hilton, SA, Australia, [3] Austrian Institute of Health Technology Assessment, Vienna, Austria, [4] Department of Health Policy, Care Policy and Evaluation Centre, London School of Economics and Political Science, London, United Kingdom, [5] Department of Psychology, Clinical Child and Adolescent Psychology, Philipps University Marburg, Marburg, Germany, [6] Mental Health Research Program, The Village, Ludwig Boltzmann Gesellschaft, Innsbruck, Austria, [7] Division of Psychiatry I, Department of Psychiatry, Psychotherapy and Psychosomatics, Medical University Innsbruck, Innsbruck, Austria

OPEN ACCESS

Edited by:
Domenico De Berardis,
Azienda Usl Teramo, Italy

Reviewed by:
Frances Louise Dark,
Metro South Addiction and Mental Health Services, Australia
Julio Torales,
National University of Asunción, Paraguay

***Correspondence:**
Melinda Goodyear
melinda.goodyear@monash.edu

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *15 November 2021*
Accepted: *22 December 2021*
Published: *31 January 2022*

Citation:
Goodyear M, Zechmeister-Koss I, Bauer A, Christiansen H, Glatz-Grugger M and Paul JL (2022) Development of an Evidence-Informed and Codesigned Model of Support for Children of Parents With a Mental Illness— "It Takes a Village" Approach. Front. Psychiatry 12:806884. doi: 10.3389/fpsyt.2021.806884

Providing support to parents and their children to help address the cycle of intergenerational impacts of mental illness and reduce the negative consequences for children is a key focus of selective prevention approaches in public mental health. However, a key issue for children of parents with a mental illness is the lack of access to early intervention and prevention support when needed. They are not easily identifiable (until presenting with significant mental health issues of their own) and not easily accessing the necessary support that address the complex interplay of parental mental illness within families. There are significant barriers to the early identification of these children, particularly for mental health care. Furthermore, there is a lack of collaborative care that might enhance identification as well as offer services and support for these families. The "It takes a Village" project seeks to improve mental health outcomes for children through the co-development, implementation and evaluation of an approach to collaborative practice concerned with the identification of families where a parent has a mental illness, and establishing a service model to promote child-focused support networks in Austria. Here we describe the development of service delivery approach for the "It takes a Village" project that aims to improve identification and support of these children within enhancements of the existing service systems and informal supports. The paper describes the use of codesign and other implementation strategies, applied to a research setting, with the aim of impacting the sustainability of workforce reform to achieve lasting social impact. Results highlight the steps involved in translating evidence-based components, local practice wisdom and lived experience into the "It takes a Village" practice model for Tyrol, Austria. We highlight through this paper how regional context-specific solutions are essential in the redesign of care models that meet the complex needs of children of parents with a mental illness. Service system and policy formation with local and experienced stakeholders are also vital to ensure the solutions are implementation-ready, particularly when introducing new practice models that rely on organizational change and new ways of practice with vulnerable families. This also creates a solid foundation for the evaluation of the "It take a Village" approach for children of parents with a mental illness in Austria.

Keywords: codesign, family focused, strength-based, children, parents with mental illness, family intervention, prevention, early intervention

INTRODUCTION

International studies estimate that one in four children currently grows up with a parent with mental illness worldwide (1). Children whose parents have mental illness have an increased risk of developing behavioral, academic, and/or mental health problems due to a range of genetic, environmental, and psychosocial factors (2). A key issue for these children is that they are often considered "invisible" from view of the existing service system in accessing early intervention support (3). Increased engagement with these children from services that may come into contact with their families can help provide supports to promote the healthy development of these children (4).

Mental illness typically occurs within families, impacting parents, children, and the whole family situation (5). Population estimates indicate that over 50% of people with a lifetime diagnosis of mental illness are parents (6), and worldwide, between 12 and 45% of adults attending adult mental health services are parents (7). These parents face similar parenting issues as all parents and while not all parents with a mental illness struggle, there are many that do, often due to issues such as poverty and social isolation usually associated with mental illness (8). Furthermore, because of the increased likelihood of stigmatization and discrimination accompanying a mental illness, these families may face greater challenges accessing support. This, in combination with a lack of visibility for early intervention support, may explain why less than one in six children are currently receiving support for emerging mental health issues at any one time (9, 10).

Family-focused service delivery in mental health services is a model that views the person with the mental illness in the context of their family relationships (e.g., being a parent) (4, 11). Family focused practice, targeting support toward supporting parenting and child well-being, has been a promising selective prevention strategy as a way to enhance public mental health at the population level (12). This type of approach focusses on supporting families to buffer against the impacts of mental illness on all family members, including children (13, 14). However, this type of service delivery is not as common in services who might be coming into contact with these parents and their families (15). In adult mental health, for example, a change to this type of service delivery is slow, as it is in conflict with the predominant medicalised individual client care model, and enhanced by limiting supportive administrative structures to encourage family focused practice (16–19).

Providing targeted intervention support to parents and their children can help break the cycle of intergenerational transmission of mental illness and improve outcomes for children of parents with a mental illness (20). Several approaches to address the intergenerational impacts have been outlined worldwide (21). Early intervention programs targeting parents with a mental illness have been shown to be effective in reducing vulnerability of young people to mental illness or negative social outcomes (22). A meta-analysis has shown that intervention with families can reduce the incidence of children developing similar mental health issues by up to 40% (22). Analysis of randomized control trials found that individual, group and family-based interventions were effective in reducing internalizing behavior and, to a lesser extent, externalizing behavior in children of parents with a mental illness (22, 23).

Interventions targeting parental behavior or parent-child interactions have typically shown small but significant positive outcomes on sensitivity and responsiveness between parents and children (24). Parenting support models have been developed as an early intervention approach addressing parenting behavior and understanding of child development through social learning models (25). Adaptation of parent support programs is commonplace though, to respond to the fear of negative judgement and stigma and shame that can accompany mental illness and/or co-occurring substance misuse for parents (26).

As mentioned, there are challenges for children experiencing a vulnerability to their mental health in accessing early intervention support (2, 4). They are not easily identifiable (until in significant need of their own treatment) and do not easily access the necessary support to address the impact of mental illness within families (3). Furthermore, support in adult focused services has typically been focused on engaging the parent in the care of the child, with limited consultation regarding the nature of that support that addresses the needs and listens to the "voice" of the child (27).

A need still exists for systemic change which emphasizes the early identification and prevention of risk factors for children's mental health (28–31). Making these children visible involves both direct support to children and parents focussing on improving behavioral outcomes; as well as a need to draw on strategies to promote motivation to change for those families deeply affected by systemic disempowerment (4, 5, 32, 33). Whole of family approaches and integrated models need to be considered moving forward to address the multiple and complex needs of these families, and addresses the influences that affect generations with mental health challenges (13, 34–37).

There is emerging evidence for the role of collective impact and integrated models supporting parents with mental health challenges and their children. In Finland, a brief intervention model such as Let's Talk About Children (38), helping parents with mental illness to support the everyday life of the child, has shown effectiveness when implemented across adult, child, and family focused services in a region. The program has been shown to improve outcomes for children and parents in terms of emotional symptoms, parental self-efficacy, and result in a significant decline in child protection referrals when the intervention is implemented across all service systems interacting with families (39).

We have recently developed a similar integrated early intervention model using codesign and open innovation in science methods in Austria (40). Our international, multi-disciplinary-led initiative takes the concept of "raising the village to raise the child" and applied it to an early identification and collaborative support approach to improve outcomes for children of parents with a mental illness and their families ("It takes a Village" practice approach) (3). The approach is aimed at improving early identification of children and adolescents whose parents have a mental illness (sensitive identification; SENSE) and enhancing the support networks around the child and their family by increasing their informal and formal resources (Collaborative Village Approach; CVA). The project aims to focus on the children's perspective (of their support network) and design an approach that is collaborative, strength-based, and offers support to the family in the region of Tyrol, Austria (8).

This paper describes this early intervention model in detail. In doing so, we describe the process of development of the practice approaches—SENSE and CVA through an extensive scoping and codesign process to develop evidence informed practice approaches that not only draw on practice wisdom and local context knowledge, but also draw on the international research on interventions for these children and their families. The co-design approach utilized in the Village project is influenced by the notion of participatory research, whereby researchers work together with key stakeholders with a good understanding of the local system, to use their collective experiences and creativity to co-create a new product, practice or new way of addressing a local issue (41, 42). This approach benefits from the value it places on sharing the production of knowledge across disciplines or across contexts, as a way to enhance the usability and social relevance of the knowledge generated, particularly for community-based or health-based services (41, 43).

Based on the premise of participatory design, the development of knowledge in this way in partnership with those who will use it, is believed to facilitate knowledge translation and support the integration of the practice approaches in the real world setting for evaluation. The translation of evidence into the routine delivery of family focused practice supporting families where a parent has a mental illness continues to be a significant challenge in this field (44). Here we invited community stakeholders including people living with a mental illness or their children and professionals, to participate in a creative group process with the goal of designing new practice approaches for adult mental health and other support services to provide support for children of parent with mental illness (3). The rationale behind the idea of designing practice approaches in a participatory manner is that the approaches better suit the context, are accepted and valued by stakeholders and are more sustainable than producer-push approaches (45).

MATERIALS AND METHODS

This paper continues on from the protocol paper published in Volume 1 in this special interest topic (3) by showcasing the results of the codesign process, in which the development of the practice approaches was formed. Here, we describe and present findings from the participatory process to understand:

1) The contextual needs, what is currently working and not working for children of parents with a mental illness (drawing on data from the scoping stage),
2) The key elements and a conceptual understanding of best practice for COPMI (evidence review findings),
3) The desired practice elements of the approach to develop a model (codesign workshop findings), and
4) The conditions necessary to implement and trial the practice approaches (implementation design).

Scoping Data Sources

A number of research activities were conducted in preparation of the co-design process aimed at understanding the local context and understanding international best practice. The following data were used and is now published: (a) a situational analysis of Tyrolean societal and service provision context in relation to families (46), (b) an analysis mental health care service uptake (47), (c) a mapping of mental health service usage in Tyrol (47), (d) a synthesis of the knowledge from the literature and international experts about what works, for whom, and in which context (48). These secondary data sources were narratively summarized to give an overview of the results of the scoping phase.

Co-design Process Data

A series of six codesign workshops were conducted locally in 2018–2019, with live-video conferencing as needed, to develop the key design concept (49). The findings of the codesign workshops were documented in the workshop planning documents, transcripts from audio recordings of the workshops, as well as workshop materials such as slides and outcome documents. These documents described the aims and activities of the workshops, presented content delivered during the workshop, results and summaries of the decisions made, transcribed and translated audio recordings of the workshop discussions, and observations and reflections from the researchers participating in the workshops. Content analysis (50) was used to examine the key decision-making steps that led to the development of the practice model throughout the series of workshops.

While the workshops were mainly held in German, some aspects of the workshops were conducted in English to accommodate participation and delivery of content from non-native German speaking researchers (JP, MGo). All documented material, including audio transcripts, were translated to English, and examined by both a German speaker (IZK) and an English speaker (MGo) for accuracy and consensus in the content analysis (50).

RESULTS

The practice approaches were developed through a series of stages: (1) scoping; (2) co-design; (3) acceptance of the design; (4) aspects of feasibility and suitability of practice approaches to local context. Those stages are now described in detail below.

Stage 1: Scoping—Identifying the Existing Context and Service System, Needs of Families, and International Best Practice

Firstly, scoping was conducted to understand the local context (46). The region of Tyrol is in the Western part of Austria, and geographically consists of many mountains and valleys. The population size is roughly 750,000 from which 140,000 (19%) are dependent children (0–18 years). Just over 85% of Tyroleans are Austrian citizens. Catholic religion plays the most important role regarding religious communities. With respect to economic structure, 50% of the population is actively working in paid employment, the remainder is either retired (20%), in education, or in other forms of activity (parental leave, household leading only, military service). Tourism industry accounts for 20% of the Tyrolean gross domestic product (46).

Data about the existing practices, barriers, and facilitators to support for children of parents with a mental illness in the existing service in Tyrol, Austria, were drawn from the scoping stage. Essentially this stage determined the scope of the unmet need and gaps in the existing service system.

Defining the Unmet Needs and Gaps

Service usage data indicated that the most parents in Tyrol were seeking treatment within the primary health system (e.g., medication prescription from a family doctor, GP), but the majority of severely ill parents were seeking treatment in the adult mental health inpatient hospital system (47). Support services, however, directly targeting children of parents with a mental illness were extremely limited in the region. Publicly funded mental health care or psychotherapy for children and adolescents were also limited, although privately funded outpatient psychotherapy/psychiatry was available for those families who could finance this themselves.

During the scoping stage, it was clear that there was an identified awareness and need to support children of parents with mental illness in the region. Stakeholder interviews identified many existing practice challenges in care for children of parents with a mental illness in Tyrol (46). These included:

- A lack of standardized identification and recording of parents with mental illness accessing treatment services. This included little or no documentation on the children of parents seeking treatment.
- A lack of standardized documentation, training, and education for professionals regarding identifying children who are living with a parent with a mental illness, particularly in talking with parents who are presenting to treatment services.
- A lack of awareness and practice guidelines in how to support children of parents with a mental illness and where to seek support for a family.

In terms of existing services provided, there was a recognition of the need to ask about a child's welfare if the parent presented to hospital or emergency services as unwell. However, there were little formalized processes of support services to refer children and their families for support, unless detrimental issues were identified. The main approach taken by adult mental health professionals involved contacting the child and youth welfare system or social worker within the hospital to address the crises needs of the family. This process of accessing support could lead to installing family support services, however, the system of support was triggered by referral due to an identified risk issue for the child (referral to child and youth welfare) (46).

Some social services were available including youth centers, parenting support programs, and mental health self-help groups for adults. Some voluntary support offers were also available such as "host grandmothers" and volunteers for tutoring in educational needs. One potentially relevant service was identified ("Kinderleicht"), specifically addressing the support needs of children of parents with addictive disorders. However, this service was small and only servicing one region of Tyrol. Issues were also identified across the region with equity of access to programs and support, with more service options available in urban areas compared to some of the rural regions of Tyrol (46, 47).

International Best Practice

Interviews with international experts in the implementation of family focused practice for these families found a number of key themes to understand more about the nature of the challenges and also enablers to practice in order to produce desired outcomes for children of parents with a mental illness. As described in (48), core components of programs included a focus on building strengths of parents in their parenting skills and helping children to understand parents' mental illness. The interviews also highlighted the interplay between practitioner, parent, and child outcomes; and the need for sufficient resources, such as training and supervision and organization support for family focused practice [see (48) for more detail].

Bringing in the Evidence Base

Brief scoping reviews were conducted in between the workshops to understand the core practice elements of the codesigned practice approaches, and to bring in international evidence for local adaptation. Key peer reviewed research articles were reviewed that covered practice guidelines and recommendations for practice and were expanded to using key literature searches in Medline, PsycINFO and Google Scholar for the terms *"identification", "social support," "collaborative practice," "practice guidelines" "family intervention" "parents with a mental illness"* and "*children*" or *"children of parents with a mental illness"* or *"COPMI."* In addition, the Village team of international researchers were each asked to review and explore known family focused interventions from their own and similar countries of origin to contribute to the existing approaches determined in the literature review. Core elements from the literature and selected best practice approaches were presented to workshop participants with a series of activities that enabled participants to select and discuss how to transfer the international evidence into the local context. See **Appendix 1** for an example.

Stage 2: A Series of Codesign Workshops With Local Community Stakeholders to Develop the Design of the Practice Approaches

Following a review of the key components of participatory codesign methodology (43, 45, 51–55), a series of workshops were designed by the Village Project team. The overall aim of the series of workshops was to develop practice approaches that were evidence-informed, suited to the context, are acceptable to local stakeholders, and feasible and ready for dissemination. As part of this process, it was anticipated that designated networks amongst stakeholders could be built to support the translation of the practices into local services, and a commitment and authorization by stakeholders managing local services could be gained to implement the codesigned practices in their own environment. The practice approaches and tools were developed to increase the identification of children and to support them in everyday life by strengthening networks of formal and informal support systems of the child and their family in Tyrol. A key focus of the design included a focus on including the "child's voice" in exploring and designing their "village" of support.

Participants

Key stakeholders were identified to participate in the workshops and included a representation from a variety of fields. The aim was to include a maximum of perspectives based on the findings from the scoping stages on identified potentially relevant organizations and professionals who may come into contact with these families (46, 47). Participants were then selected based on a number of criteria including field (practice, policy); sector; profession; target group; function (management etc) and gender. Another important consideration was also to ensure the number of participants did not exceed 18 per workshop to maintain a productive working atmosphere.

A total of 26 individuals representing 14 different local organizations participated across the six workshops. In addition to this, a total of 13 persons from the interdisciplinary research-partner team attended across the six workshops. On average, 16 community representatives and 4 research team members attended each workshop. There was higher representation from the health care sector, practice-focused professionals, and participants were more likely to work in the medical profession compared with other professionals (see **Table 1** below). Adult mental health services were more strongly represented than others, more females than males and more participants were in middle management roles. Not all participants attended all workshops, but attending participants could nominate a proxy in their place if they wished.

TABLE 1 | Characteristics of workshop participants.

Sector (*n*)		**Field (*n*)[a]**	
Health care	11	Practice	14
Social care	6	Research	4
Education	2	Policy/Payer	4
Informal/voluntary	2		
Other	(1)		
Profession (*n*)		**Service sector (*n*)**	
Medical doctor	*4*	*Primary care*	*2*
- Psychiatrist	4	*Adult mental health*	*7*
- General practitioner	1	- inpatient	7
Nurse	1	- outpatient	5
Social worker	2	*Child mental health*	*3*
Psychologist	3	- inpatient	3
Pedagogue	1	- outpatient	3
Public health specialist	(1)	Children's service	3
Social scientist	(1)	Parental service	2
Peer worker	(1)	Service for families	3
Other	(1)	Not applicable	2
Sex (*n*)[b]		**Function within organization (*n*)**	
Female	13–15	Top management	4–6
Male	6–7	Middle management	7–11
		Front line staff	3–6
		Not applicable	2

[a] some stakeholders represented more than one field; [b] number dependent on proxies that attended; brackets indicate that these categories were not represented in each workshop.

The Workshops—Designing the Practice Approaches

The workshops included both presentations and group work facilitation exercises to develop up the concepts of the "It takes a Village" practice approach. Key decisions were made at each of the workshops to focus and consensus was sought on the design concept. Several facilitation techniques were used and these are described in (56).

The aims and key decisions of each workshop are described in **Table 2** below. The workshops involved presenting international best practice examples and evidence on effective approaches. Workshop participants then identified options on how those might be implemented locally in Tyrol. The aim was to find a balance between evidence-based practice and feasibility within the local context and constraints (57, 58).

The Workshop Results

Key Decisions

Each codesign workshop was constructed to make key decisions about the development of the practice model, the evaluation, and the implementation to be delivered as part of the Village project, as outlined above.

TABLE 2 | Co-design workshop aims and key decisions in the development of the "It takes a Village" practice approach.

Workshop (no. attendees)	Aims	Key decisions that resulted
1 ($n = 15$)	Awareness of group participants and their relationship with the issue of COPMI. Development of a sense of identity as a codesign group. Understanding of the principles of open innovation and codesign and their role in this process. Introduction to the Village Project and a background introduction to the needs of COPMI from research and local scoping results. Presentation of three case vignettes outlining case journeys for COPMI within the region (information elaborated on from scoping) to identify areas of change.	Agreement on terms of reference. Agreement on rules for communication. Identification of key areas for change from the presentation of case vignettes of child focused care found in the scoping stage.
2 ($n = 15$)	Development of a shared vision. Familiarizing with a theory of change. Prioritizing areas for change.	Agreement made on the roadmap for the design of the practice approaches in the codesign workshop series. Agreement on common vision, assumptions and priority goals. Selection of max. nine prioritized areas for change.
3 ($n = 17$)	Identifying practice options for four prioritized areas for change around improving identification of COPMIs in adult mental health in Tyrol; based on proposed practice approaches in the literature and international expert interviews.	Agreement on options for transferring Phase 1—SENSE practices on identification to Tyrol (e.g., who should be asking about parent status, which questions to be asked, options on how parents admitted to hospitals can stay in contact with their child, options on how/where/when to talk with the child about the parental mental illness, options on how to address social resources around the child/family for the first time); agreeing on the stance (e.g., strength-based, acknowledging privacy, empathic and respectful, non-judgemental).
4 ($n = 18$)	Identifying practice options for the remaining five prioritized areas for change around improving support of children via a collaborative village approach (CVA); based on proposed practice approaches in the literature and in the international expert interviews.	Agreement on options for transferring Phase two—CVA practices on activating support around these children in Tyrol (e.g., how to refer the child/family to support program, which organizations could host the "facilitators" who would work with the child/family, which practice steps are involved in working with the children/families to activate support, which qualifications are required.
5 ($n = 16$)	Finalizing the practice concepts on identification and collaborative support from previous workshop. Identifying key aspects of the evaluation design (How to evaluate the change process as well as its results).	Agreement on the first point of identification, referral pathway and key practices of Village Facilitators in working with children/family as well as options for hosting the facilitator based on previous workshops. Agreement on inclusion/exclusion criteria, study design, options for outcome indicators. [see (48) for more detail of the outcomes]
6 ($n = 13$)	Defining feasibility, commitments and next implementation steps.	An agreed approach to practice, implementation and evaluation procedure is available including a commitment of organizations and persons to implement changes in their every-day practice.

In workshop 1, areas for change were selected from reviewing several case vignettes of existing practice drawn together from the scoping data [see more in (49)]. Areas for change from reviewing these vignettes were identified by the workshop participants. These were:

- Improving family communication about mental illness (parents and children).
- Improving education to families about mental illness.
- Asking parents about their children when seeking treatment.
- Providing psychoeducation to children in schools.
- Establishing adequate infrastructure for children to visit parents in adult psychiatry.
- Support contact between parents and children when parents are unwell and in treatment.
- Begin a conversation with families as early as possible when a parent is unwell:
    - Inform children of their parent's mental ill-health.
    - Develop standardized processes to identify social resources around the child.
    - Develop guidance and knowledge of 'good enough' parenting as an orientation for adult mental health professionals.
    - Include development of a crisis plan in standard process of care.
    - Include family members and children in the development of crisis plans and decisions.
- Primary health care to actively work with families of parents with a mental illness (provided the GP is aware of the parent's illness).
- Educate families/relatives about the importance of children needing support, as with physical illnesses in parents.
- Increase availability of social workers in adult mental health for family/child coordination.
- Adult Mental Health to refer families for support outside of psychiatry.

- Schools to provide supports for children of parents with a mental illness.
- Improve communication between organizations.
- Raise awareness in the community that children of parents may need support as much as children of parents with a physical illness.
- Organize mental health care earlier in a way that avoids the need for acute crisis care (avoid trauma for children).
- Improve information on available support in families, adult mental health, other relevant organizations and communities.

In Workshop 2, a consensus was reached about the common goal for the design:

> *The Village approach promotes the healthy development and mental health of COPMI.*

Several preconditions and assumptions were agreed on for the design of the practice approaches. These included: the service provider has information of the parent's mental illness; there is increased help-seeking from families through better information and understanding of mental illness; and knowledge and awareness of mental health needs to increase in the community.

Workshop participants prioritized the areas identified in workshop 1 in terms of what is the best way forward to achieve the agreed vision. These were selected as follows:

1) All providers in adult psychiatry (for example psychosocial services) actively ask patients about their children/family situation.
2) Healthy caregivers and children are (kindly) informed about parental mental illness; talks take place as early as possible without hierarchy (child focused).
3) There are standardized procedures for identifying social resources around the child; caregivers are informed.
4) Contact between the affected parent and children is actively supported in the acute phase.
5) At each visit, a family contingency plan is prepared—mobilizing existing resources; caregivers. Children are involved in an age appropriate way. Decisions included; talks take place "at eye level."
6) Families are actively invited by family doctors, supports if a parent is mentally ill.
7) Psychoeducation is developed and implemented for schools.
8) All providers know existing offers and their contents (for example, are better informed about child and youth welfare).
9) Support for children is actively organized and families are cared for continuously, while "normalization" is respected.

An agreement was made that the focus would be for activities within adult mental health—but other areas such as primary health and school system would be beneficial to include at a later stage. It was deemed that prioritized areas 1–3 were to be designed as part of Phase 1—sensitive identification (SENSE) and areas 4–9 were seen as steps within Phase 2—the collaborative village approach (CVA).

The Design Concept (Results From Workshops 3–4)

The product at the end of the workshop series included two practice models: (1) a visualization of a pathway for the identification of children of parents with a mental illness—a standardized and systematic SENSE process in selected hospital adult mental health and primary care institutions (**Figure 1**); and (2) a visualization of the process of establishing both informal and formal support (the Village) for children of parents with mental illness and their families—the Collaborative Village Approach (CVA) (**Figure 2**). These draft concepts were agreed to by the workshop participants as the primary design outcome, that would be implemented and evaluated in the next stage of the research project. Some details (e.g., with regard to coordination responsibilities) remained unsolved at that point in the design process (indicated by question marks in the figure). For some points within the pathway, options were specified.

The process steps were unpacked separately as part of the workshop process, and are shown in **Figures 3–6**.

Key Steps in the practice model that were agreed to were as follows:

1. Identifying parenting status and child and family characteristics and responsibilities (SENSE 1; **Figure 3**).
2. Exploring with a parent about the child's adjustment—strengths and challenges (SENSE 2; **Figure 4**).
3. Developing a shared understanding with parents and children on the day to day life of the child and the supports in place and needs to strengthen these supports (CVA 1; **Figure 5**).
4. Develop a support plan to strengthen and maintain the child's supports through a network meeting (CVA 2; **Figure 6**).
5. Review the support plan, troubleshooting and addressing issues for sustainability into the future (CVA 2; **Figure 6**).

Sensitive Screening/Identification of Children Living With a Parent Who Has a Mental Illness

SENSE 1: Identifying Information and Building Trust

The goal of phase 1 of the SENSE approach is to identify whether a patient with a mental illness has children and is therefore a father or mother (**Figure 3**). Identification questions are intended to be used during admission or during a visit of the treating physician or during a visit to the general practice.

One initial outcome required from this SENSE approach is the recording of the **parenting status** of the adult patient, their **family caring roles**, and their **children's gender** and **age and living situation**.

SENSE 2: Short, Goal-Focused Conversation About Parenting and the Daily Life of the Child

The outcome of phase 2 of the SENSE approach (**Figure 4**: Practical elements of SENSE phase 2) is a more in-depth conversation with parents about their parenting **strengths and challenges**, strengths and vulnerabilities for their **child's adjustment**, and a brief understanding of the existing **child's social support network**. The parent could also be asked about any immediate needs and wishes they may have for enhancing the strengths of their child, or in relation to their parenting

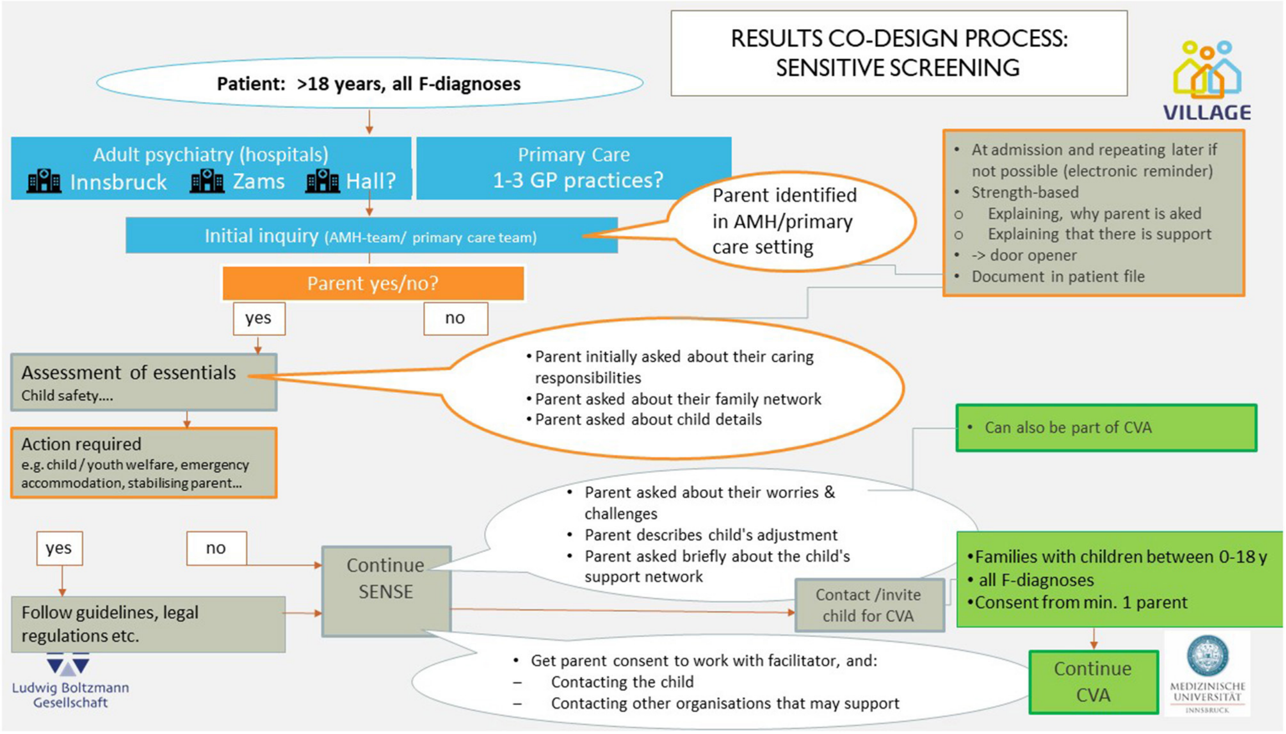


FIGURE 1 | SENSE (Identification) pathway. CVA, collaborative village approach; SENSE, sensitive screening; GP, general practitioner.

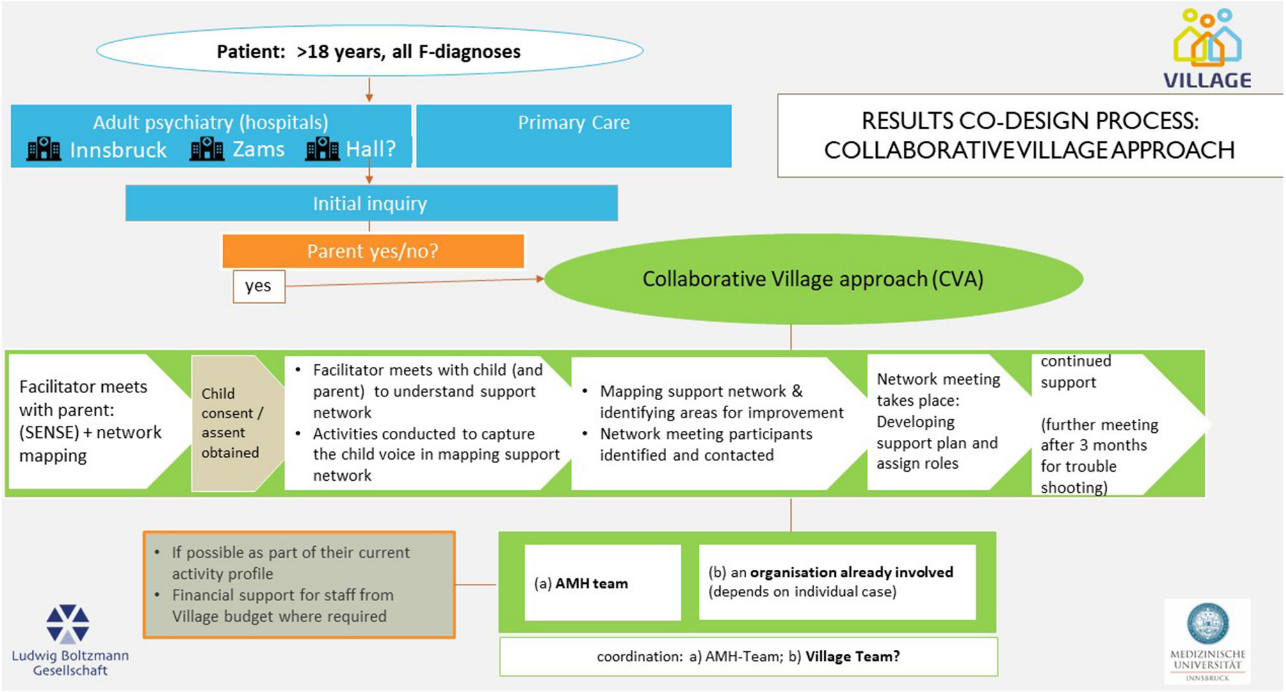


FIGURE 2 | Pathway for the Collaborative Village Approach (CVA).

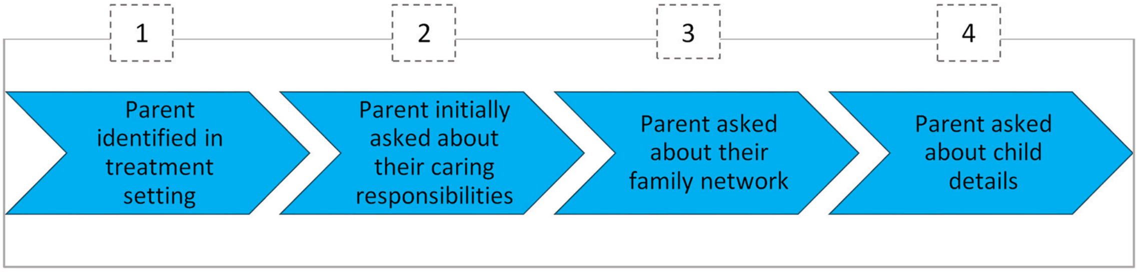


FIGURE 3 | Practical elements of SENSE phase 1.

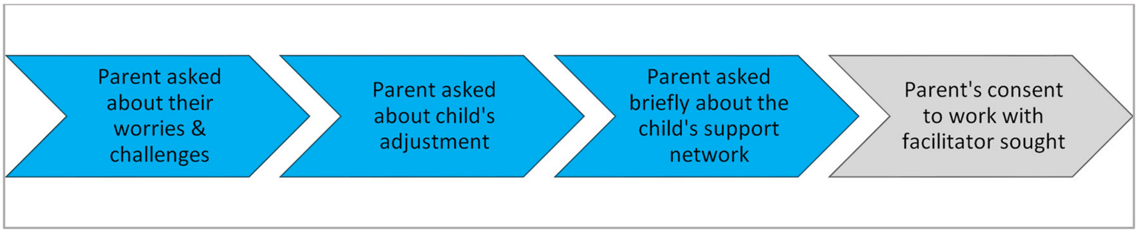


FIGURE 4 | Practical elements of SENSE phase 2.

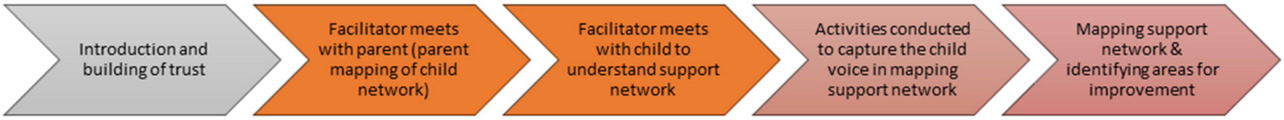


FIGURE 5 | Practical elements of CVA Phase 1.

FIGURE 6 | Practical elements of CVA Phase 2.

strengths and challenges. Sensitive, open questions to understand the current living situation of the parents and the child are important here, for example, *Can you tell us a little about your parenting and caring roles at the moment in your life?*

Enhancing the Social Network: The Collaborative Village Approach (CVA)

The idea of CVA is to help build a day-to-day life that ensures the best possible support for the child/youth in their local support network, or "village" and thereby promote the healthy development of children of parents with a mental illness. This should be driven by parent and child in partnership and supported through conversations with the Village Facilitator. In this part the village facilitator takes on a key capacity building and curious role. The Village Facilitator works together with the family and their social network to strengthen the social support for the child. Working directly with the child and seeking their perspective is a key component of the CVA.

CVA 1—A Shared Understanding the Support Network of the Child

The first step of the CVA approach involves the Village Facilitator engaging with a parent for the first time in the role of the Village Facilitator (**Figure 5**). The primary focus of this first interaction is to build engagement and the beginnings of a collaborative relationship to promote the well-being of the parent's child. As part of this interaction, a series of activities and questions are asked to learn about the child's social network from the parents' perspective. The aim is to develop a common understanding of the child's everyday life, existing support and possible gaps and potential for improvement between the parent and Village Facilitator. The role of the Village Facilitator is to identify the parent's view of the child support network and ask specific questions when needed to understand missing persons or institutions who are capable to close potential network gaps for the child.

The next step in this phase is for the facilitator to support the children to contribute their understanding of their social network. This step helps create a common understanding with parents and children about the child's everyday life and existing support, as well as to identify what is needed to improve the situation. The activities described are aimed for children from 4 years of age. The aim of the activities is to hear directly from the children about their support. This activity visualizes the existing networks and identifies gaps in support. Ultimately, the child should help define what their "village" looks like and this network should be made visible.

Following an analysis of the parent and child support network results, a family meeting is conducted to help develop a shared understanding of the support network and identify areas for enhancement or improvement. The idea of this step is to develop a common idea of how the subsequent network meeting in CVA 2 should be organized.

CVA 2—The Network Meeting and Support Plan Is Developed and Reviewed

The concept of the network meeting (**Figure 6**) is derived from the "Family Group Conferencing" practice (other common names: Social Network Conference, Family Group Conference, Family Council, Relatives Council). The concept for our CVA network meeting was informed by early developments in family conferencing in New Zealand and has since then been applied in a wide range of fields (e.g., child protection, domestic violence, youth justice) including mental health (59–61).

Through an independent coordinator (in our case "Village Facilitator") informal and formal support systems are brought together, while at the same time the family and especially the children are encouraged to take responsibility for decision-making. In other words, the Village Facilitator is responsible for the process, but not for the outcome of those meetings. The underlying ethos of the network meeting is based on the principle that the family and its social network are capable of finding their own solutions to support children with mentally ill parents (they have control over the solutions and are recognized as experts in their own lives).

The role of professional service providers and community members is to facilitate and resource plans and decisions that are consistent with securing and supporting a child's resilience in their daily life. The focus is to support the child's day to day activities, and the provision of practical, emotional, and social support. At the end of the network meeting, there should be an agreed support plan which enhances the daily life of the child, both from formal and informal support providers for the child. Following 3 months of implementation of the support plan, the plan is reviewed for any future refinements. At the end of a 6 months phase of engagement, the idea is that the work of the village facilitator is handed fully over to families and support personnel to lead and maintain the network of support where needed.

Theoretical Basis of the Approach

As discussed in the workshops the theoretical approach and stance is a core part of family interventions for families where a parent has a mental illness. It was agreed that the practice approaches are delivered with the following theoretical perspectives in mind:

1. Motivational interviewing
2. Capacity building approaches for families and practitioners
3. Consideration of the social determinants of health
4. Working within an understanding of the sociology of childhood
5. Focusing on building self-regulation skills and promoting self-determination and choice in families

Core Practice Principles

The Practice Approach is built on the following principles of practice:

- Orientation on strengths of the family (members) instead of weaknesses
- Recognition of the decision-making competence of parents and children (building self-determination)
- Trauma Sensitivity: Awareness of the effects of traumatic events in families and children and creating an atmosphere in which all persons feel safe, welcome, and supported

The aim of the described practices is to develop a sense of trust and a feeling of confidence for the concerned parents and children. All elements of the described practice approach open up the possibility that the parenting experience with a mental illness and growing up in a family where one parent is mentally ill will be normalized (with the experience of not being alone) and recognized. The focus is also on a non-judgmental, interested stance toward families, which helps to create a trustful and supportive atmosphere for parents and children and which helps mental health professionals, general practitioners, or village facilitators to have a meaningful conversation with parents and children.

Another central principle of all the practice steps described above is that, whenever possible, the viewpoint of the children and the parents is integrated into all processes and decisions. The perspective of the families concerned serves as an essential basis for understanding their needs and developing a common social support for the children.

Stage 3: Acceptance of the Design

Commitments for participating in the different practice steps were sought and documented in Workshop 6. A commitment for the SENSE approach was obtained and these sites would serve as the pilot sites—one hospital inpatient based (Innsbruck), 1 day clinic based (Zams). The possibility of identifying parents in primary health through GP practices was also suggested. The CVA approach was seen to be a process outside of psychiatry, in community services; with the exception of the day clinic (Zams) that proposed a model where CVA process could be delivered as part of the routine treatment team.

Stage 4: Aspects of Feasibility and Suitability of Practice Approaches to Local Context

At the completion of workshop 6, participants undertook an activity to identify barriers and enablers for the implementation of the codesigned practice approaches. In terms of where the practice approaches could take place, participants deemed SENSE could feasibly be delivered in adult mental health services or general practice clinics, without the need for additional resources or costs. Participants stressed that it would require, however, equipping existing staff with the procedures and supportive structures to undertake SENSE and refer parents with mental illness to the Village Project. Structured documentation and leadership were deemed to be important to support staff to undertake SENSE. Primary health care needed structured questions, and Adult Mental Health needed question prompts documentation to ask questions that identify children of adult patients as part of routine practice.

For the CVA process, workshop participants indicated that in most situations this process was outside adult psychiatry and would need to be resourced through additional funding. Although one adult mental health service identified they could embed a village facilitator within the treating team, if they were commissioned and funded to do so (the hospital in the village of Zams, Tyrol). Clear referral pathways to CVA were needed, as well as knowledge of the possible support network options available in the region needs to be clearly documented.

Several uncertainties to the implementation were identified by the workshop participants. Concerns were raised regarding a lack of time, money and staffing resources to deliver the practices; lack of willingness from informal care providers; lack of suitable physical resources and infrastructure available; lack of organizational support for village facilitation role; difficulty co-ordinating attendees for network meetings; difficulties seeking informed consent from families; language and communication barriers; and skills in talking sensitively with parents and children. Several options were discussed as part of the activity that might help overcome these situations (see **Appendix 2** for more information).

- Finally, workshop participants indicated the willingness of their organizations to implement the practice approaches.
- Commitments were made to implement the SENSE in two Adult Mental Health settings and potentially 1–3 primary health care settings.
- Commitment to take part in CVA network meetings in 11 out of 14 participating organizations.
- Expression of interests to provide staff for village facilitator role in 4 represented organizations.
- Commitment to participate in the implementation check-ins (local implementation team, multi-agency implementation team, advisory board) by organizations that will implement practice changes.

DISCUSSION

This paper showcases a process of intervention design to address a gap in service delivery for children of parents with a mental illness in Tyrol, Austria. The intervention "It takes a Village" approach consists of evidence-informed and codesigned practice elements, developed with people with lived experience in practice and also with those living with the challenges of mental illness in the region (3). The approach includes elements of practice that assist adult treatment providers to sensitively identify parents of dependent children who may be seeking treatment for their mental health challenges (SENSE). The second component consists of practices and steps for facilitators to enhance the "village" of support for a child living with a parent with mental health issues, and includes a focus on informal and formal support structures as well as understanding the parent and child's perspective on the child's daily life (Collaborative Village Approach, CVA).

The "It takes a Village" practice model, as outlined in this paper, is built around a participatory process from all areas of the project, including in understanding (1) the contextual needs, what is currently working and not working for children of parents with a mental illness (scoping), (2) key elements and a conceptual understanding of best practice for families where a parent has a mental illness (evidence review), (3) practice elements of the approach to develop a model (codesign), and (4) understanding the conditions necessary to implement and trial the practice approaches (implementation design). Alongside this, was the development of an evaluation logic and realist approach framework to design the outcome measures of the evaluation of the village approach (48). From this process, we achieved a high agreement from stakeholders to trial the practice approaches, where relevant, in their organizational setting.

The process draws on approaches outlined in the implementation science field. Here we have applied best practice from implementation science in applying principles of codesign and a series of structures and strategies to help integrate best practice evidence into "real world" settings (62). We have utilized a participatory design approach where those involved in delivering the intervention or using services shape the evidence of what works into a practice approach suitable for their contextual setting. These approaches are becoming fundamental to the transfer of innovation that when applied involve changes to practice, particularly in mental health settings (42). A paradigm shift toward recovery-oriented practice, from a predominantly bio-medical focused one has encompassed a strong focus on consumer involvement in service design and resulted in a range of successes in service delivery approaches. This shift in service delivery has been found to occur more successfully when there is a whole of organization approach involving organizational leadership as well as consumers with lived experience in the design and support for the delivery of these new methods of practice (63). We expect the process described for development of the practice approaches in Tyrol will show similar ease in the transfer to practice and service delivery for families locally.

As shown in the workshop series, the codesigned practice approaches were developed on the evidence base for interventions and supports for families where a parent has a mental illness. Steps outlined for the practice approach in this paper have similarities with practice elements outlined in other well-known evidence-based interventions such as Let's Talk About Children (38, 64), Family Talk (65), Parenting Well (66, 67), and Social Network Conferences (38, 68), and other evidence-based practice elements described in the research (4, 11, 69).

A fundamental basis to the "It takes a Village" practice model is drawn from the use of motivational interviewing techniques to assist in outlining rapid engagement techniques that can support practitioners in talking with parents and their children. Motivational interviewing skills uses various communication techniques to improve a person's self-efficacy or sense of their own capability, and enhances their motivation for changes through a focus on a person's desired behaviors (70–72). Because of this, motivational interviewing has parallels with the promotion of self-determination and self-regulation in a person (73), two areas of change that has more recently been linked as core elements for families benefitting from family interventions (5). Motivational interviewing skills also prove useful in managing parent ambivalence or engagement issues in child and family social work (74). Similarly, self-regulation skills are also proving useful in working with parents with mental illness for engagement in parenting support programs (75). Interestingly, they are also now being considered as a strategy in supporting practitioners in the change process to implement new practice approaches themselves (76).

A common criticism of selection prevention approaches for children of parents with a mental illness has been a lack of theory or conceptual framework in the evidence base of family interventions for these families (77, 78). This presents a problem for not only the design of evaluation or outcome studies, but also in understanding the assumptions underlying the mechanisms of change associated with mental health, family functioning, and child development that selective prevention programs are usually targeted toward (79). Some family evidence-based interventions in this area though report strong theoretical foundations associated with strengths-based, recovery-focused or resiliency frameworks (65, 80, 81). Drawing on the international evidence, the codesign workshop series described in this paper was able to explore the theoretical perspectives and evidence base to formulate practice approaches built on concepts of being strengths-focused and trauma-informed; built on theories of self-determination and self-regulation; and integrating an understanding of social determinants of health, and the sociology of childhood in its design. These perspectives are operationalized in the designed practice approaches through the stance and curiosity lens of the approach. This encompasses a focus on the "how" a practice is delivered as well as the "what" in terms of components of the practice approach.

This essential aspect of the designed practice approaches is reinforced through the questioning and engagement stance adopted by those working with these families in the delivery of the practice approaches. The stance highlights the values that underlie the practical action and determines *why* a professional may do something in a certain way when working with parents and their children. The principles of practice outlined in the stance include: (1) An orientation on strengths of the family (members) instead of weaknesses; (2) Recognition of the decision-making competence of parents; (3) Integration of the child's voice and perspective as a fundamental basis to the support plan design; and finally, (4) cultural and trauma sensitivity in practice. The focus is also on a non-judgmental, interested stance toward families, which helps to create a trustful and supportive atmosphere for parents and children and which helps a clinician, general practitioner, or village facilitator to have meaningful conversations with parents and their children. This focus is not new though to family interventions for children of parents with a mental illness. These are reported components of interventions such as Family Talk (65), Let's Talk about Children (5, 38, 64) and Family Options (80, 81).

A core part of the participatory process of the design of the Village approach was in designing a practice model that is acceptable and feasible for implementation in the local context. Participants were able to prioritize areas for change based on a thorough scoping stage, and also adapt the evidence base to the local setting of what might work within the region of Tyrol, Austria. The stakeholders with decision making abilities or policy influences were also able to indicate an agreement and willingness to implement the practice approaches at the completion of this codesign process, securing the beginnings of the next stage of the research project for "real world" implementation and evaluation. This participatory process has many advantages but is particularly encouraged in the development of innovations to help address the lag in efficiencies to translation to practice of evidence of what works, particularly for the reduction in burden of disease in public health approaches (42, 82).

Community stakeholders, in this study, however, identified that even with a process of codesign, there still remained challenges and uncertainties to the implementation of the practice approaches in the local setting. These barriers were believed to require organizational support to be overcome in the relevant practice settings. They included an allocation of time, resources, and funding to support the practice approaches to be delivered; alongside various skill-based training supports, policies and procedures to undertake the identification process (SENSE); and a flexible approach to the delivery of network meetings and requirements of informal and formal support providers. While not new, the application of family focused practice in mental health care settings continues to be accompanied by significant challenges in its implementation (21, 83). The integration of implementation science principles that aid in creating drivers to support practice change is becoming an important vehicle for effective translation to practice of evidence-based interventions in this area (44, 84, 85), as well as working in partnership between researchers, policy makers and service providers as part of support and sustaining change (86).

Selective prevention strategies, such as parenting or child focused interventions for families with mental health challenges, remain an effective public mental health strategy to improve child outcomes for children of parents with a mental illness (12, 87, 88). Such interventions have been shown to reduce the relative risk of a child developing the mental illness as its parent by about 40% (22). It is expected the "It takes a Village" model, which draws heavily on other effective interventions, will also improve outcomes for children (48). Interventions such as this—that focus on a two-generational approach (a parent and the child)—and on drawing together or improving elements within a child's daily life or ecology of influence—have also been shown to be effective in other selective prevention programs such as in child welfare with multi-stressed families (44, 89–91). Core to positive outcomes in research in this area, however, rely on program fidelity and implementation support strategies to that ensure program elements are delivered as intended (92, 93).

Equity of access to mental health-care, particularly for selective prevention approaches, remains a significant global challenge (21). Of note, in the design of the "It takes a Village" approach, implementation of the practice approaches were designed for primary health as well as adult psychiatry, a decision we expect will improve access for a number of parents who might be seeking medication support from their general practitioner only. We know from the scoping analysis that this will contain a significant number of Austrian families (47). While providing options for improving reach of the research study, this implementation approach is also in line with a focus on more community based and stepped care model of mental health care, whereby people have access to treatment outside hospital based mental health services (12).

In terms of limitations, it must be noted that this is not a study of the effectiveness of the practice approaches. While it broadly is expected to produce the desired outcomes for children of parents with a mental illness, there is evidence to suggest that family interventions in Austria for vulnerable children can have poor uptake (94). Therefore, there are significant unknowns about how these practice approaches may work in socio-cultural norms of Tyrol, and an understanding of the impact on child outcomes is yet to be determined in the project. We anticipate though, that with thorough consultation and design with local stakeholders as well as an understanding of best practice and implementation from the international literature, we are positioning the "It takes a Village" approach with solid foundations for achieving positive outcomes for families where a parent has a mental illness in Austria. A realist framework is being utilized in the evaluation of the practice approaches in Tyrol (3, 48) and it is anticipated that this selective prevention approach will be effective in improving the social and emotional well-being of children and their parents with mental health challenges.

In conclusion, the paper outlines a key process to developing evidence informed changes to practice and service delivery in mental health care for families. The participatory process itself, with key stakeholders, is a vital element in developing the translation to practice to suit local contextual needs. This is necessary to ensure effective elements of service redesign can meet and address existing gaps in care to intervene in addressing the intergenerational transmission of mental illness within families. Future studies in this project, however, will ultimately determine the direct benefits for families, practitioners, and the service system in Austria.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Monash University Human Research Ethics Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

MGo led the analysis with IZ-K but all authors contributed to the analysis. MGo led the manuscript preparation and all authors were involved in editing the manuscript. All authors contributed to the design of the study and the collection of data.

FUNDING

The Village is a research project funded by the Austrian Federal Ministry of Health—Science and Research through the Open Innovation in Science Center at the Ludwig Boltzmann Gesellschaft GmbH, hosted at the Medical University of Innsbruck, with collaboration of Co-Investigator institutions.

ACKNOWLEDGMENTS

We would like to thank the local village research team in Innsbruck, as well as interviewees and participants of the codesign workshops who kindly provided their vision and enthusiasm toward this shared goal.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.806884/full#supplementary-material

REFERENCES

1. Maybery D, Reupert A, Patrick K, Goodyear M, Crase L. Prevalence of parental mental illness in Australian families. *Psychiatric Bull.* (2009) 33:22–6. doi: 10.1192/pb.bp.107.018861
2. Goodyear M, McDonald M, von Doussa H, Cuff R, Dunlop B. Meeting the intergenerational needs of families where a parent has a mental illness [English and Spanish versions]. *J Parent Family Mental Health.* (2018) 3:1. doi: 10.7191/parentandfamily.1011
3. Christiansen H, Bauer A, Fatima B, Goodyear M, Lund IO, Zechmeister-Koss I, et al. Improving identification and child-focused collaborative care for children of parents with a mental illness in Tyrol, Austria. *Front Psychiatry.* (2019) 10:233. doi: 10.3389/fpsyt.2019.00233
4. Goodyear M, Hill T-L, Allchin B, McCormick F, Hine R, Cuff R, et al. Standards of practice for the adult mental health workforce: meeting the needs of families where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120
5. Goodyear M, Allchin B, Burn M, von Doussa H, Reupert A, Tchernagovski P, et al. Promoting self-determination in parents with mental illness in adult mental health settings. *J Family Nurs*. (2022). doi: 10.1177/10748407211067308
6. Nicholson J, Biebel K, Katz-Leavy J, Williams V. The Prevalence of parenthood in adults with mental illness: implications for state and federal policymakers, programs, and providers. In: Manderscheid RW. Henderson MJ, editors. *Mental Health, United States, 2002, DHHS Pub No (SMA) 3938*. Rockville, MD: Substance Abuse and Mental Health Services Administration, Center for Mental Health Services (2004). p. 120–37.
7. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry*. (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
8. Carpenter-Song EA, Holcombe BD, Torrey J, Hipolito MMS, Peterson LD. Recovery in a family context: experiences of mothers with serious mental illnesses. *Psychiatr Rehabil J.* (2014) 37:162–9. doi: 10.1037/prj0000041
9. Lawrence D, Johnson S, Hafekost J, Boterhoven de Haan K, Sawyer M, Ainley J, et al. *The Mental Health of Children and Adolescents: Report on the Second Australian Child and Adolescent Survey of Mental Health and Wellbeing*. Canberra, ACT: Department of Health (2015).
10. Segal L, Guy S, Furber G. What is the current level of mental health service delivery and expenditure on infants, children, adolescents, and young people in Australia? *Austral N Zealand J Psychiatry*. (2017) 52:163–72. doi: 10.1177/0004867417717796
11. Foster K, Goodyear M, Grant A, Weimand B, Nicholson J. Family-focused practice with EASE: a practice framework for strengthening recovery when mental health consumers are parents. *Int J Ment Health Nurs.* (2019) 28:351–60. doi: 10.1111/inm.12535
12. Wahlbeck K. Public mental health: the time is ripe for translation of evidence into practice. *World Psychiatry.* (2015) 14:36–42. doi: 10.1002/wps.20178
13. Nicholson J, Friesen BJ. Developing the evidence base for families living with parental psychiatric disabilities: crossing the bridge while we’re building it. *Psychiatr Rehabil J.* (2014) 37:157–61. doi: 10.1037/prj0000092
14. Walsh F. *Strengthening Family Resilience, Third Edition*. New York, NY: Guilford Publications (2016).
15. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatric Mental Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
16. Biebel K, Nicholson J, Woolsey K. Implementing an intervention for parents with mental illness: building workforce capacity. *Psychiatr Rehabil J.* (2014) 37:209–15. doi: 10.1037/prj0000046
17. Allchin B, Goodyear M, O’Hanlon B, Weimand BM. Leadership perspectives on key elements influencing implementing a family-focused intervention in mental health services. *J Psychiatr Ment Health Nurs.* (2020) 27:616–27. doi: 10.1111/jpm.12615
18. Allchin B, Weimand BM, O’Hanlon B, Goodyear M. Continued capacity: factors of importance for organizations to support continued Let’s Talk practice–a mixed-methods study. *Int J Ment Health Nurs.* (2020) 29:1131–43. doi: 10.1111/inm.12754
19. Allchin B, O’Hanlon B, Weimand BM, Boyer F, Cripps G, Gill L, et al. An explanatory model of factors enabling sustainability of let’s talk in an adult mental health service: a participatory case study. *Int J Ment Health Syst.* (2020) 14:48. doi: 10.1186/s13033-020-00380-9
20. Wainberg ML, Helpman L, Duarte CS, Vermund SH, Mootz JJ, Gouveia L, et al. Curtailing the communicability of psychiatric disorders. *Lancet Psychiatry*. (2018) 5:940–4. doi: 10.1016/S2215-0366(18)30342-0
21. Falkov A, Goodyear M, Hosman CMH, Biebel K, Skogøy BE, Kowalenko N, et al. A systems approach to enhance global efforts to implement family-focused mental health interventions. *Child Youth Serv*. (2016) 37:175–93. doi: 10.1080/0145935X.2016.1104104
22. Siegenthaler E, Munder T, Egger M. Effect of preventative interventions in mentally ill parents on the mental health of the offspring: systematic review and meta analysis. *J Am Acad Child Adolescent Psychiatry*. (2012) 51:8–17. doi: 10.1016/j.jaac.2011.10.018
23. Lannes A, Bui E, Arnaud C, Raynaud JP, Revet A. Preventive interventions in offspring of parents with mental illness: a systematic review and meta-analysis of randomized controlled trials. *Psychol Med.* (2021) 51:2321–36. doi: 10.1017/S0033291721003366
24. Thanhäuser M, Lemmer G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. *Curr Opin Psychiatry.* (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342
25. Phelan RF, Howe DJ, Cashman EL, Batchelor SH. Enhancing parenting skills for parents with mental illness: the Mental Health Positive Parenting Program. *Med J Aust.* (2013) 199:S30–S3. doi: 10.5694/mja11.11181
26. Coates D, Phelan R, Heap J, Howe D. “Being in a group with others who have mental illness makes all the difference”: the views and experiences of parents who attended a mental health parenting program. *Children Youth Serv Rev.* (2017) 78:104–11. doi: 10.1016/j.childyouth.2017.05.015
27. Gladstone BM, Boydell KM, McKeever P. Recasting research into children’s experience of parental mental illness: beyond risk and resilience. *Soc Sci Med.* (2006) 62:2540–50. doi: 10.1016/j.socscimed.2005.10.038
28. Guy S, Furber G, Leach M, Segal L. How many children in Australia are at risk of adult mental illness? *Austral N Zealand J Psychiatry.* (2016) 50:1146–60. doi: 10.1177/0004867416640098
29. Furber G, Leach M, Guy S, Segal L. Developing a broad categorisation scheme to describe risk factors for mental illness, for use in prevention policy and planning. *Aust N Z J Psychiatry.* (2016) 51:230–40. doi: 10.1177/0004867416642844
30. Price-Robertson R, Smart D, Bromfield L. *Family Is for Life Connections between Childhood Family Experiences and Wellbeing in Early Adulthood*. Melbourne, VIC: Australian Institute of Family Studies. (2010) 7–17 p.
31. Moore TG, McDonald M, Carlon L, O’Rourke K. Early childhood development and the social determinants of health inequities. *Health Promot Int.* (2015) 30:ii102–15. doi: 10.1093/heapro/dav031
32. Power J, Goodyear M, Maybery D, Reupert A, O’Hanlon B, Cuff R, et al. Family resilience in families where a parent has a mental illness. *J Soc Work.* (2016) 16:66–82. doi: 10.1177/1468017314568081
33. Ingoldsby EM. Review of interventions to improve family engagement and retention in parent and child mental health programs. *J Child Fam Stud.* (2010) 19:629–45. doi: 10.1007/s10826-009-9350-2
34. Foster K. Advancing the prevention of intergenerational mental illness: where are we now, where do we go from here? *Adv Mental Health.* (2015) 13:97–9. doi: 10.1080/18387357.2015.1068470
35. Tapias E, Coromina M, Grases N, Ochoa S. Psychological Treatments with children of parents with mental illness: a systematic review. *Child Youth Care Forum.* (2021) 50:1107–30. doi: 10.1007/s10566-021-09608-2
36. Campbell TCH, Reupert A, Sutton K, Basu S, Davidson G, Middeldorp CM, et al. Prevalence of mental illness among parents of children receiving treatment within child and adolescent mental health services (CAMHS): a scoping review. *Eur Child Adolesc Psychiatry.* (2021) 30:997–1012. doi: 10.1007/s00787-020-01502-x
37. Dopp AR, Lantz PM. Moving upstream to improve children’s mental health through community and policy change. *Adm Policy Ment Health.* (2020) 47:779–87. doi: 10.1007/s10488-019-01001-5
38. Solantaus T, Toikka S, Alasuutari M, Beardslee W, Paavonen E. Safety, feasibility and family experiences of preventive interventions for children and families with parental depression. *Int J Ment Health Promot.* (2009) 11:15–24. doi: 10.1080/14623730.2009.9721796

39. Niemelä M, Kallunki H, Jokinen J, Räsänen S, Ala-Aho B, Hakko H, et al. Collective impact on prevention: let's talk about children service model and decrease in referrals to child protection services. *Front Psychiatry.* (2019) 10:64. doi: 10.3389/fpsyt.2019.00064
40. Kaisler R, Paul J. Evidence-based practice and polices for impact on mental health of children and adolescents. *J Res Technol Policy Eval.* (2019) 48:114–9. doi: 10.22163/fteval.2019.377
41. Bird M, McGillion M, Chambers EM, Dix J, Fajardo CJ, Gilmour M, et al. A generative co-design framework for healthcare innovation: development and application of an end-user engagement framework. *Res Involve Engage.* (2021) 7:12. doi: 10.1186/s40900-021-00252-7
42. Ammerman A, Smith TW, Calancie L. Practice-based evidence in public health: improving reach, relevance, and results. *Annu Rev Public Health.* (2014) 35:47–63. doi: 10.1146/annurev-publhealth-032013-182458
43. Kleinsmann M, Valkenburg R. Barriers and enablers for creating shared understanding in co-design projects. *Design Stud.* (2008) 29:369–86. doi: 10.1016/j.destud.2008.03.003
44. Yates S, Gatsou L. Undertaking family-focused interventions when a parent has a mental illness - possibilities and challenges. *Practice.* (2021) 33:103–18. doi: 10.1080/09503153.2020.1760814
45. Greenhalgh T, Jackson C, Shaw S, Janamian T. Achieving research impact through co-creation in community-based health services: literature review and case study. *Milbank Q.* (2016) 94:392–429. doi: 10.1111/1468-0009.12197
46. Zechmeister-Koss I, Goodyear M, Tüchler H, Paul JL. Supporting children who have a parent with a mental illness in Tyrol: a situational analysis for informing co-development and implementation of practice changes. *BMC Health Serv Res.* (2020) 20:326. doi: 10.1186/s12913-020-05184-8
47. Zechmeister-Koss I, Tüchler H, Goodyear M, Lund IO, Paul JL. Reaching families where a parent has a mental disorder: using big data to plan early interventions. *Neuropsychiatrie.* (2020) 34:39–47. doi: 10.1007/s40211-019-00323-y
48. Bauer A, Best S, Malley J, Christiansen H, Goodyear M, Zechmeister-Koss I, et al. Towards a program theory for family-focused practice in adult mental health care settings: an international interview study with program leaders. *Front Psychiatry.* (2021) 12:741225. doi: 10.3389/fpsyt.2021.741225
49. Zechmeister-Koss I, Goodyear M, Bauer A, Paul J. Involving stakeholders in developing and evaluating evidence-informed support practices for children who have a parent with a mental illness: results from a co-development process in Tyrol. *Das Gesundheitswesen.* (2020) 82:447. doi: 10.1055/s-0040-1708944
50. Elo S, Kyngäs H. The qualitative content analysis process. *J Adv Nurs.* (2008) 62:107–15. doi: 10.1111/j.1365-2648.2007.04569.x
51. Goodyear-Smith F, Jackson C, Greenhalgh T. Co-design and implementation research: challenges and solutions for ethics committees. *BMC Med Ethics.* (2015) 16:78. doi: 10.1186/s12910-015-0072-2
52. Hinchcliff R, Greenfield D, Braithwaite J. Is it worth engaging in multi-stakeholder health services research collaborations? Reflections on key benefits, challenges and enabling mechanisms. *Int J Qual Health Care.* (2014) 26:124–8. doi: 10.1093/intqhc/mzu009
53. Jagosh J, Macaulay AC, Pluye P, Salsberg J, Bush PL, Henderson J, et al. Uncovering the benefits of participatory research: implications of a realist review for health research and practice. *Milbank Q.* (2012) 90:311–46. doi: 10.1111/j.1468-0009.2012.00665.x
54. Park MM, Zafran H, Stewart J, Salsberg J, Ells C, Rouleau S, et al. Transforming mental health services: a participatory mixed methods study to promote and evaluate the implementation of recovery-oriented services. *Implement Sci.* (2014) 9:119. doi: 10.1186/s13012-014-0119-7
55. Sanders EBN, Stappers PJ. Co-creation and the new landscapes of design. *CoDesign.* (2008) 4:5–18. doi: 10.1080/15710880701875068
56. Zechmeister-Koss I, Aufhammer S, Bachler H, Bauer A, Bechter P, Buchheim A, et al. *Practices to Support Co-Design Processes: A Case-study of Codesigning a Program for Children with Parents with a Mental Health Problem in the Austrian Region of Tyrol* (2020) (Unpublished).
57. Castro FG, Barrera M, Martinez CR. The cultural adaptation of prevention interventions: resolving tensions between fidelity and fit. *Prevent Sci.* (2004) 5:41–5. doi: 10.1023/B:PREV.0000013980.12412.cd
58. Bopp M, Saunders RP, Lattimore D. The tug-of-war: fidelity versus adaptation throughout the health promotion program life cycle. *J Prim Prev.* (2013) 34:193–207. doi: 10.1007/s10935-013-0299-y
59. Conference FIFG. *Resource Materials for Awareness and Training on Family Group Conference Practice.* (2011). Available online at: https://www.fgcni.org/cmsfiles/Publications/RMText-Version.pdf (accessed September 30, 2019).
60. Früchtel F, Roth E. *Familienrat und inklusive, versammelte Methoden des Helfens.* Heidelberg: Carl-Auer-Systeme Verlag (2017).
61. Hansbauer P, Hensen G, von Spiegel H. *Familiengruppenkonferenz: Eine Einführung.* Weinheim und München: Juventa Verlag (2009).
62. Kirchner JE, Smith JL, Powell BJ, Waltz TJ, Proctor EK. Getting a clinical innovation into practice: an introduction to implementation strategies. *Psychiatry Res.* (2020) 283:112467. doi: 10.1016/j.psychres.2019.06.042
63. Lorien L, Blunden S, Madsen W. Implementation of recovery-oriented practice in hospital-based mental health services: a systematic review. *Int J Ment Health Nurs.* (2020) 29:1035–48. doi: 10.1111/inm.12794
64. Solantaus T, Toikka S. The effective family programme: preventative services for the children of mentally ill parents in Finland. *Int J Ment Health Promot.* (2006) 8:37–44. doi: 10.1080/14623730.2006.9721744
65. Beardslee WR, Wright EJ, Gladstone TRG, Forbes P. Long-term effects from a randomized trial of two public health preventive interventions for parental depression. *J Family Psychol.* (2007) 21:703–13. doi: 10.1037/0893-3200.21.4.703
66. Biebel K, Nicholson J, English K, Guyer-Deason M, Wixted S. Let's talk about parenting: recovery for parents living with mental illnesses. *Psychiatry Inform Brief.* (2016) 13:1100. doi: 10.7191/pib.1100
67. Nicholson J, English K, Heyman M. The parentingwell learning collaborative feasibility study: training adult mental health service practitioners in a family-focused practice approach. *Community Ment Health J.* (2021). doi: 10.1007/s10597-021-00818-5. [Epub ahead of print].
68. Solantaus T, Puras D. Caring for children of parents with mental health problems — a venture into historical and cultural processes in Europe. *Int J Mental Health Promot.* (2010) 12:27–36. doi: 10.1080/14623730.2010.9721823
69. Gladstone B, Exenberger S, Weimand B, Lui V, Haid-Stecher N, Geretsegger M. The capability approach in research about children and childhood: a scoping review. *Child Indic Res.* (2021) 14:453–75. doi: 10.1007/s12187-020-09766-0
70. Taveras EM, Gortmaker SL, Hohman KH, Horan CM, Kleinman KP, Mitchell K, et al. Randomized controlled trial to improve primary care to prevent and manage childhood obesity: the high five for kids study. *Arch Pediatr Adolesc Med.* (2011) 165:714–22. doi: 10.1001/archpediatrics.2011.44
71. Miller WR, Rollnick S. *Motivational Interviewing: Helping People Change.* New York, NY: Guilford Press (2012).
72. Lundahl BW, Kunz C, Brownell C, Tollefson D, Burke BL. A meta-analysis of motivational interviewing: twenty-five years of empirical studies. *Res Soc Work Pract.* (2010) 20:137–60. doi: 10.1177/1049731509347850
73. Miller WR, Rollnick S. Meeting in the middle: motivational interviewing and self-determination theory. *Int J Behav Nutr Phys Activ.* (2012) 9:25. doi: 10.1186/1479-5868-9-25
74. Forrester D, Westlake D, Glynn G. Parental resistance and social worker skills: towards a theory of motivational social work. *Child Family Soc Work.* (2012) 17:118–29. doi: 10.1111/j.1365-2206.2012.00837.x
75. Sanders MR, Mazzucchelli TG. The promotion of self-regulation through parenting interventions. *Clin Child Fam Psychol Rev.* (2013) 16:1–17. doi: 10.1007/s10567-013-0129-z
76. Mazzucchelli TG, Ralph A. Self-regulation approach to training child and family practitioners. *Clin Child Fam Psychol Rev.* (2019) 22:129–45. doi: 10.1007/s10567-019-00284-2
77. Fraser C, James EL, Anderson K, Lloyd D, Judd F. Intervention programs for children of parents with a mental illness: a critical review. *Int J Mental Health Promot.* (2006) 8:9–20. doi: 10.1080/14623730.2006.9721897
78. Reupert A, Goodyear M, Eddy K, Alliston C, Mason P, Maybery D, et al. Australian programs and workforce initiatives for children and their families where a parent has a mental illness. *Adv Mental Health.* (2009) 8:277–285. doi: 10.5172/jamh.8.3.277
79. Suarez EB, Lafrenière G, Harrison J. Scoping review of interventions supporting mothers with mental illness: key outcomes and challenges. *Community Ment Health J.* (2016) 52:927–36. doi: 10.1007/s10597-016-0037-z
80. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Family options for parents with mental illnesses: a developmental, mixed methods pilot study. *Psychiatr Rehabil J.* (2009) 33:106–14. doi: 10.2975/33.2.2009.106.114

81. Nicholson J, Albert K, Gershenson B, Williams V, Biebel K. Developing Family Options: outcomes for mothers with severe mental illness at twelve months of participation. *Am J Psychiatr Rehabil.* (2016) 19:353–69. doi: 10.1080/15487768.2016.1231639
82. Green LW. Making research relevant: if it is an evidence-based practice, where's the practice-based evidence? *Fam Pract.* (2008) 25:i20–i4. doi: 10.1093/fampra/cmn055
83. Isobel S, Allchin B, Goodyear M, Gladstone BM. A narrative inquiry into global systems change to support families when a parent has a mental illness. *Front Psychiatry.* (2019) 10:310. doi: 10.3389/fpsyt.2019.00310
84. Mulligan C, Furlong M, McGilloway S. Promoting and implementing family-focused interventions for families with parental mental illness: scoping and installation. *Adv Mental Health.* (2020) 18:202–16. doi: 10.1080/18387357.2019.1614466
85. Skogøy BE, Maybery D, Ruud T, Sørgaard K, Peck GC, Kufås E, et al. Differences in implementation of family focused practice in hospitals: a cross-sectional study. *Int J Ment Health Syst.* (2018) 12:77. doi: 10.1186/s13033-018-0256-5
86. Nicholson J, Reupert A, Grant A, Lees R, Maybery D, Mordoch E, et al. The policy context and change for families living with parental mental illness. In: Reupert A, Maybery D, Nicholson J, Göpfert M, Seeman MV, editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families. Vol.3.* Cambridge, CA: Cambridge University Press (2015). p. 354–64. doi: 10.1017/CBO9781107707559.034
87. Christiansen H, Reck C, Zietlow A-L, Otto K, Steinmayr R, Wirthwein L, et al. Children of Mentally III Parents at Risk Evaluation (COMPARE): design and methods of a randomized controlled multicenter study—part I. *Front Psychiatry.* (2019) 10:128. doi: 10.3389/fpsyt.2019.00128
88. Stracke M, Gilbert K, Kieser M, Klose C, Krisam J, Ebert DD, et al. COMPARE Family (Children of Mentally Ill Parents at Risk Evaluation): a study protocol for a preventive intervention for children of mentally ill parents (Triple P, Evidence-Based Program That Enhances Parentings Skills, in Addition to Gold-Standard CBT With the Mentally Ill Parent) in a multicenter RCT—Part II. *Front Psychiatry.* (2019) 10:54. doi: 10.3389/fpsyt.2019.00054
89. Hoagwood K, Koretz D. Embedding prevention services within systems of care: strengthening the nexus for children. *Appl Prevent Psychol.* (1996) 5:225–34. doi: 10.1016/S0962-1849(96)80014-X
90. Lee BR, Ebesutani C, Kolivoski KM, Becker KD, Lindsey MA, Brandt NE, et al. Program and practice elements for placement prevention: a review of interventions and their effectiveness in promoting home-based care. *Am J Orthopsychiatry.* (2014) 84:244–56. doi: 10.1037/h0099811
91. Turner CW, Robbins MS, Rowlands S, Weaver LR. Summary of comparison between FFT-CW® and usual care sample from administration for children's services. *Child Abuse Negl.* (2017) 69:85–95. doi: 10.1016/j.chiabu.2017.04.005
92. Palinkas LA, Spear SE, Mendon SJ, Villamar J, Reynolds C, Green CD, et al. Conceptualizing and measuring sustainability of prevention programs, policies, and practices. *Transl Behav Med.* (2019) 10:136–45. doi: 10.1093/tbm/ibz170
93. Bezeczky Z, El-Banna A, Petrou S, Kemp A, Scourfield J, Forrester D, et al. Intensive Family Preservation Services to prevent out-of-home placement of children: a systematic review and meta-analysis. *Child Abuse Negl.* (2020) 102:104394. doi: 10.1016/j.chiabu.2020.104394
94. Nagl-Cupal M, Hauprich J. Invisible population: understanding recruitment barriers of a nurse-led support programme for families with caregiving children in Austria. *Nursing Open.* (2020) 7:1164–72. doi: 10.1002/nop2.491

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

SYSTEMATIC REVIEW
published: 01 February 2022
doi: 10.3389/fpsyt.2021.721796

Expressed Emotion in the Family: A Meta-Analytic Review of Expressed Emotion as a Mechanism of the Transgenerational Transmission of Mental Disorders

***Julia Fahrer*[1,2]*, *Nathalie Brill*[1], *Lisa Marie Dobener*[1], *Julia Asbrand*[3] and *Hanna Christiansen*[1]**

[1] Department of Clinical Child and Adolescent Psychology, Faculty of Psychology, Philipps University Marburg, Marburg, Germany, [2] Department of Special Needs Educational and Clinical Child and Adolescent Psychology, Faculty of Psychology and Sports Science, Justus-Liebig-University Gießen, Gießen, Germany, [3] Institute of Psychology, Humboldt-Universität zu Berlin, Berlin, Germany

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Victoria E. Cosgrove,
Stanford University, United States
Renee Rienecke,
Eating Recovery Center (ERC),
United States
Lynsey Gregg,
The University of Manchester,
United Kingdom

****Correspondence:***
Julia Fahrer
julia.fahrer@staff.uni-marburg.de

Specialty section:
This article was submitted to Child and Adolescent Psychiatry, a section of the journal Frontiers in Psychiatry

Received: *07 June 2021*
Accepted: *14 December 2021*
Published: *01 February 2022*

Citation:
Fahrer J, Brill N, Dobener LM, Asbrand J and Christiansen H (2022) Expressed Emotion in the Family: A Meta-Analytic Review of Expressed Emotion as a Mechanism of the Transgenerational Transmission of Mental Disorders. Front. Psychiatry 12:721796. doi: 10.3389/fpsyt.2021.721796

Background: High Expressed Emotion (HEE) has been identified as a risk factor for the exacerbation and course of mental illness. EE has been investigated as a caregiver's response to an offspring's problem behavior and pathology. The present meta-analysis regards EE from a transgenerational perspective and as one mechanism that might explain the transgenerational transmission of mental disorders.

Method: We identified a total of 13 studies relying on 16 independent samples of parent-child dyads of parents with a mental illness and healthy controls; these were included in our analysis. Results were synthesized into one effect size per sample; meta-regression on additional effects of parental diagnostic category, child mental illness, and child age were also applied.

Results: Parents with a mental illness are classified as HEE significantly more often. Effects were established for high criticism, albeit of small size (OR = 1.45), although they become stronger whenever offspring exhibit mental illness themselves (OR = 2.82).

Conclusion: The current study highlights the dearth of studies on EE in families in which a parent has a mental illness and its effects on their children. Our findings highlight EE as a potential mechanism for attributing the transgenerational transmission of mental disorders, especially for the EE-variable criticism, indicating dysfunctional parent-child interactions.

Systematic Review Registration: http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42019117609, identifier: CRD42019117609.

Keywords: Expressed Emotion, children of parents with a mental illness, COPMI, transgenerational transmission, mental disorders, risk factor, parent child interaction

INTRODUCTION

Worldwide, about 12.1–38.5% of children and adolescents are living with a parent who experiences a Mental illness[1] (1–4). A parent's mental illness is a powerful risk factor (OR 2.4) for their offspring to develop mental health problems (5), and about one third of the children of parents with a mental illness experience serious mental illness later in life (6). Many studies have shown adverse outcomes in children of parents with a mental illness, including children's attachment problems, internalizing, and externalizing behavior problems as well as social, cognitive, physical, and mental illness (6–10).

The Transgenerational Transmission of Mental Disorders system was developed and advanced to provide a comprehensive model to explain such transgenerational transmission of disorders in children of parents with a mental illness (9). This model identifies four major domains (i.e., 1. parent, 2. family, 3. child, 4. social environment) that interact with their respective systems and are influenced by five transmission mechanisms (i.e., 1. genetics, 2. prenatal factors, 3. parent-child-interaction, 4. Family, and 5. social factors) (11). Child development over its life span is considered, as are the concepts of multi- and equifinality, concordance, and specificity (9). Specifically parent-child-interaction is considered to be a core mechanism contributing to the heightened risk of children of parents with a mental illness for developing a serious mental illness (9, 12, 13) (see **Figure 1**).

Parent-Child-Interaction

Parenting behaviors are influenced by parental psychopathology, attitudes, attributional styles as well as the child's characteristics on a dyadic level (14, 15). Interaction patterns in families of parents with a mental illness are characterized by elevated rates of insensitive, for example, intrusive, hostile, and critical parental behaviors, the lack of parental warmth and the shortage of acknowledgment of children's developmental, emotional, and attachment needs (7, 10). However, different mental disorders have a different impact on parental behavior and its manifestation (6). Disrupted parental behavior poses a risk for child development and usually is studied with restriction to one diagnosis, but not comparing multiple diagnosis within one study. For instance, mothers with postnatal depression (for example, lower amount of affectionate touch, sensitivity, reciprocity) show a different relational behavioral profile when interacting with their child than mothers with anxiety disorders (e.g., maternal overstimulation of the child, high maternal intrusiveness, parent led interaction) (16). Traditionally, parent, child and dyadic behavior is assessed with standardized, videotaped paradigms and coded with behavior observation schemes (17). Besides these standardized behavior observation schemes, Expressed Emotion (EE) appears to be indicative of dysfunctional parent-child interactions (18), and thus an assessment of interest in this context.

Over the past five decades, EE has been a concept of interest in the field of family relations, and is regarded as an indirect measure of the emotional family climate. Developed for parents of adult patients with schizophrenia, EE was identified to play an important role in the course and relapse of mental illness (19). After controlling for patient variables, such as severe behavior or work impairment, EE still appears to be indicative of negative interactions within a family (20). EE reflects a person's affective attitude toward a close relative and is believed to play an important role in the development and perpetuation of mental disorders in offspring (20–22). It is differentiated in High (HEE) or Low Expressed Emotion (LEE). HEE reflects a high amount of criticism, hostility (CRIT), and/or Emotional Overinvolvement (EOI), whereas LEE is characterized by positive or neutral remarks, low hostility, criticism or emotional overinvolvement toward a close relative and in relationships within families (22, 23). High CRIT levels are linked to negative parental behaviors, such as more parental antagonism, harshness, negativity, and disgust. Low levels of CRIT are associated with more responsive and supportive parenting behavior (18). A current meta-analysis by Rea et al. (24) on the Five-minute Speech Sample (FMSS) in children and youths with internalizing and externalizing symptomatology supports the overall validity of HEE especially with CRIT in the context of child and adolescent health, while the EOI measure appears less robust in such contexts. Nevertheless, the analysis identifies a very small but significant effect between parental EOI and child internalizing symptoms however this result should be interpreted with caution, as the authors point out, that the effect may be caused by specific EOI criteria rather than the construct as a whole (25) and EOI may require more clarification and adaptation (24). Therefore, HEE, and predominantly CRIT, can be perceived as one mechanism of disrupted parent-child interactions in the Transgenerational Transmission of Mental Disorders. EE can be assessed via the Camberwell Family Interview (21, 26, CFI), the FMSS; (27), the Preschool Five-Minute Speech Sample (28, PFMSS), and questionnaires such as the Family Attitude Scale (29, FAS) or Family Questionnaire (30, FQ). Despite the incorporation of hostility in the CFI, it is not captured within the FMSS coding guidelines, as it shows a great overlap with CRIT (31) and does not appear with enough frequency (27). Therefore, the hostility rating is not included in the present analysis and this paper focuses on CRIT and EOI.

Parents With a Mental Illness and EE

EE traditionally was developed to assess caregiver's attitudes on adult patients with schizophrenia. Attributions that perceive the cause of problem behavior as internally controllable by the patient/offspring result in more negative emotional responses (32, 33), and there is a strong link between attribution theory and EE. There appears to be an attribution-negative affect link in HEE relatives linking hostility and CRIT to negative affect (15). CRIT is assumed to be a correlate of the typical cognitive and attributional style of mothers with depressive disorders (8) and has been identified as a possible moderator of the association between maternal depression and a child's

---

[1] Mental illness as diagnosed by a validated, standardized clinical assessment tool (e.g., clinical interview).

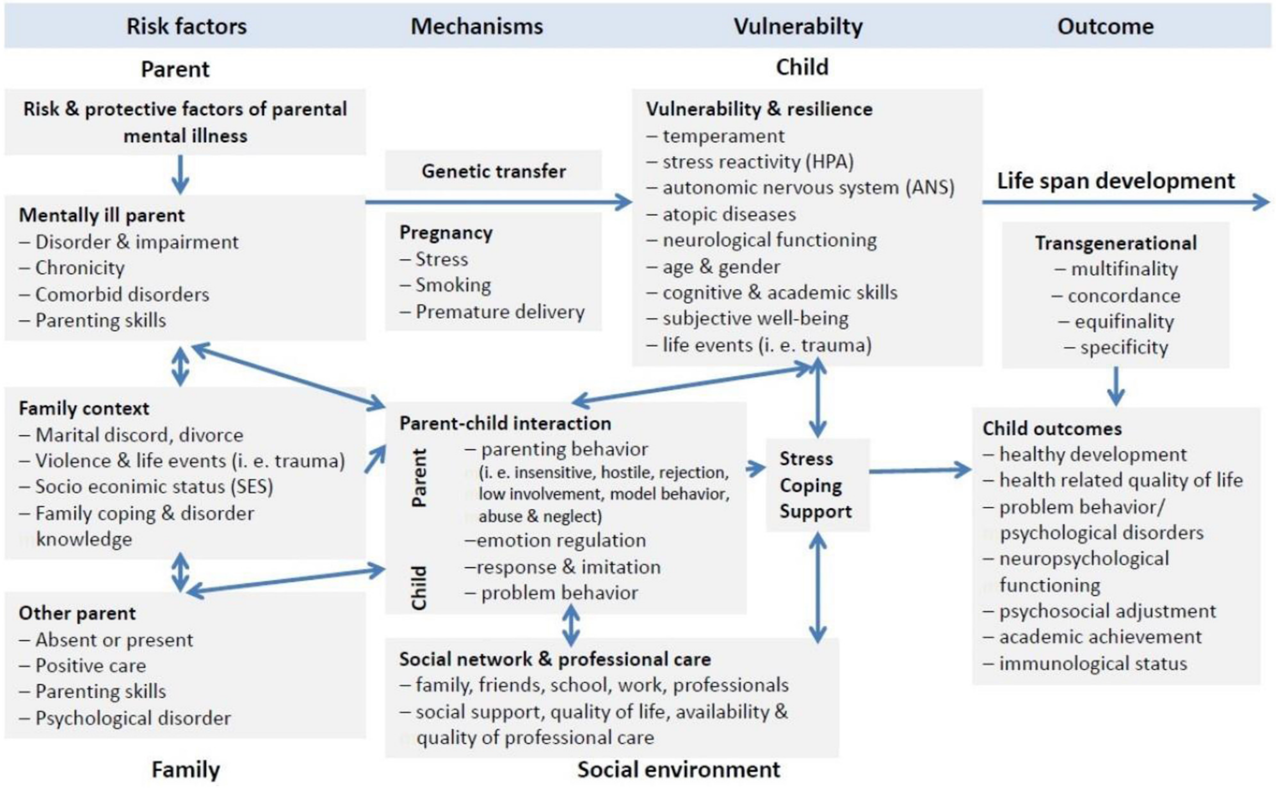


FIGURE 1 | Model of transgenerational transmission of mental illness adapted from Hosman et al. (9) and Christiansen et al. (11).

internalizing and externalizing symptoms (34, 35). In contrast to this, fathers with depression do not present with higher levels of CRIT, but do make fewer warm and positive remarks than healthy controls (36), although this is no component of the traditional HEE component. Based on such finding, a sex difference regarding CRIT and parental depression might be assumed. Mothers and fathers with a mental illness are five to nine times more likely to be classified as HEE than parents without any mental health condition (37). Parental EE status seems to be relatively stable over time (38) creating a challenge for vulnerable, genetically predisposed children, and it therefore has the potential to promote a self-perpetuating cycle of children's problem behavior and HEE within a family (39). Given that parents with a mental illness may have been exposed to parental HEE themselves, they may be prone to reacting more negatively, hostilely, and improperly when facing their children's challenges and problem behavior and therefore exhibit HEE, especially CRIT (40, 41). While there are indications that HEE is more prevalent in families in which a parent has a mental illness, predominantly depression (42, 43), generalizable evidence is lacking. Previous research on EE in the field of child and adolescent psychology has been focusing on clinically referred children (44) or the emotional family climate within families of children with internalizing and externalizing symptoms, respectively (24).

EE in the Field of Child Psychology

In the field of clinical child and adolescent psychology, HEE is regarded as an indicator of the quality of the parent-child-relationship (45). As EE reflects parental attitudes (38, 39) and HEE is a correlate of negative parental behaviors (18), it is not clear whether negative parental attitudes result in more negative parental behaviors, or vice versa. Parental HEE is linked to difficult child temperament (38, 46), and is a correlate of disruptive attachment patterns (47). Parental EE is considered a stable predictor for the course of mental illness and treatment response in children and adolescents (37, 44, 48–50). Low levels of warmth, increased hostility and critical comments have been associated with children's behavioral problems (44, 45, 51–53) and antisocial behavior (49). Parents of children with one axis I diagnosis are significantly more likely to be classified as HEE than parents of healthy controls (37, 54, 55). They appear even more critical when children carry an additional axis I diagnosis to depression (56, 57). Moreover, HEE has been positively identified in predicting the onset of attention deficit hyperactivity disorder (ADHD) (38), comorbid oppositional defiant disorder (ODD) (58), the clinical course of childhood anxiety, bipolar and depressive disorders (49, 59, 60), as well as the treatment response of adolescents suffering from eating disorders (61–63). Neither an offspring's sex nor a family's socio-economic status (SES) are associated with the parental EE status (37, 64) and the assessment of parental psychopathology or burden has achieved little attention when studying EE and child development. Therefore, EE has been and remains a risk factor of interest in the field of clinical psychology and a potential mechanism for explaining the transgenerational transmission of mental disorders. While most articles claim EE to be stable over time (38, 65–67), other methods (e.g., clinical interviews) show some evidence that EE might be somewhat able to change (68). This warrants further research because it might be an interaction with changes in offspring's behavior due to developmental steps, especially at the time before school entry.

Aims

Although extensive research has been carried out on EE and offspring's psychopathology, comparatively little is known about EE in families in which a parent had a mental illness. The following meta-analysis aimed to investigate HEE as a typical cognitive and affective style of parents with a mental illness.

Moreover, we focused on children of parents with a mental illness and parental EE, assuming EE to be a transgenerational mechanism facilitating the development of mental illness in children of parents with a mental illness (39). The current study aimed to contribute to the current literature by first presenting a comprehensive, quantitative report on the prevalence of HEE in families in which a parent has a mental illness and control families. Secondly, we aimed to identify moderators of the relationship between parental psychopathology and HEE to compute a meta-regression. Therefore, we predicted that parents of younger children tend to show less HEE. Parental diagnosis, sex, and presence of youth psychopathology may account for additional effects on the parental EE status.

METHODS

Data Sources and Searches

This meta-analysis was performed according to the "Preferred Reporting Items for Systematic Reviews and Meta-Analyses" (PRISMA) statement (69). We conducted our search in the following databases: PubMed, The Cochrane Library, PsycInfo, Web of Science, ERIC, and PubPSYC (see **Appendix A** for search terms). We restricted our search to experimental and observational studies and meta-analyses published in the English or German language until November 2021. Search criteria included parents of minor children as the population addressed, all mental disorders, and the standardized assessment and report of EE or employing a shared measure of EE (see **Appendix A**). The review protocol is registered on PROSPERO (http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42019117609; registration number CRD42019117609). In total, 1,159 studies were identified. **Figure 2** shows the flowchart with all study extraction stages.

Inclusion Criteria

Abstracts of all studies identified from the initial search were screened based on specific inclusion criteria. Studies were included if they reported (a) an experimental or observational design based on (b) a sample of parents of children aged 18 years or younger, (c) a standardized assessment or screening of parental psychopathology and a diagnosis according to DSM or ICD, and (d) a valid measure of parental EE such as the CFI (21, 26), FMSS (27, 70), PFMSS (28), FAS (29), or FQ (30), or results of common parental EE scales, such as HEE or LEE, EOI, or Criticism.

For our meta-analysis of parental psychopathology and expressed emotion, parents had to be classified as the index patient to ensure an estimation of predictive effects. Control conditions had to be no intervention or an internal comparison in case of cohort studies. Furthermore, parents and children had to be free of pervasive developmental disorders to avoid adverse factors caused by these. Studies comparing an active intervention with medication (i.e., psychotherapy vs. medication only) were excluded. All studies included had to provide sufficient information to calculate effect sizes (e.g., means and SDs, *T*-tests for independent samples, n per subgroup, r, Odds Ratio).

Study Selection

Studies, titles, and abstracts were screened by one reviewer (JF) and relevant studies were extracted that matched our aim and inclusion and exclusion criteria specified for this review. In this respect, a systematic two-stage screening process to identify relevant studies was applied and two authors (JF and LMD) carried out full text screening independently. Discrepancies were resolved through discussion with a third author (HC).

Study Characteristics/Data Collection Process

Each study was coded on several different domains including publication year, country, primary study aims, study design (e.g., control group), setting, recruitment method, length of follow up, inclusion and exclusion criteria, study participants (i.e., age, sex, diagnosis of parent and children) characteristics of the applied diagnostic instruments or screenings, assessment measure and report of EE status, and the blinding of EE raters. Furthermore, parental diagnostic category was dummy coded, differentiating on a superordinate level of diagnosis, for example, depressive disorders, anxiety disorders. Since information on the children's diagnoses was lacking, only the presence or absence of a diagnosed mental illness was coded. As studies reported inconsistent EE outcome categories, presence or absence of HEE/LEE, CRIT, and EOI was coded in the first step. As CRIT was reported predominantly and information on EOI was lacking, only data on CRIT were extracted. Presence or absence of statistical parameters (M, SD, SE, CI, correlation and regression coefficient, β, χ^2, B) and N per subgroup was coded. As not all studies reported children‘s mean age but rather age ranges, we coded age categories as well (infants $\leq$ 20 months, preschoolers $>$ 20 months and $\leq$ 6 years, school age children $>$ 6 years and $\leq$ 12 years, and adolescents $>$ 12 years and $\leq$ 18 years). When defining the age range for the school aged children, we followed the typical age of school entry in Germany even though this might deviate from school entry ages in other countries, as that is where the authors of the study are based.

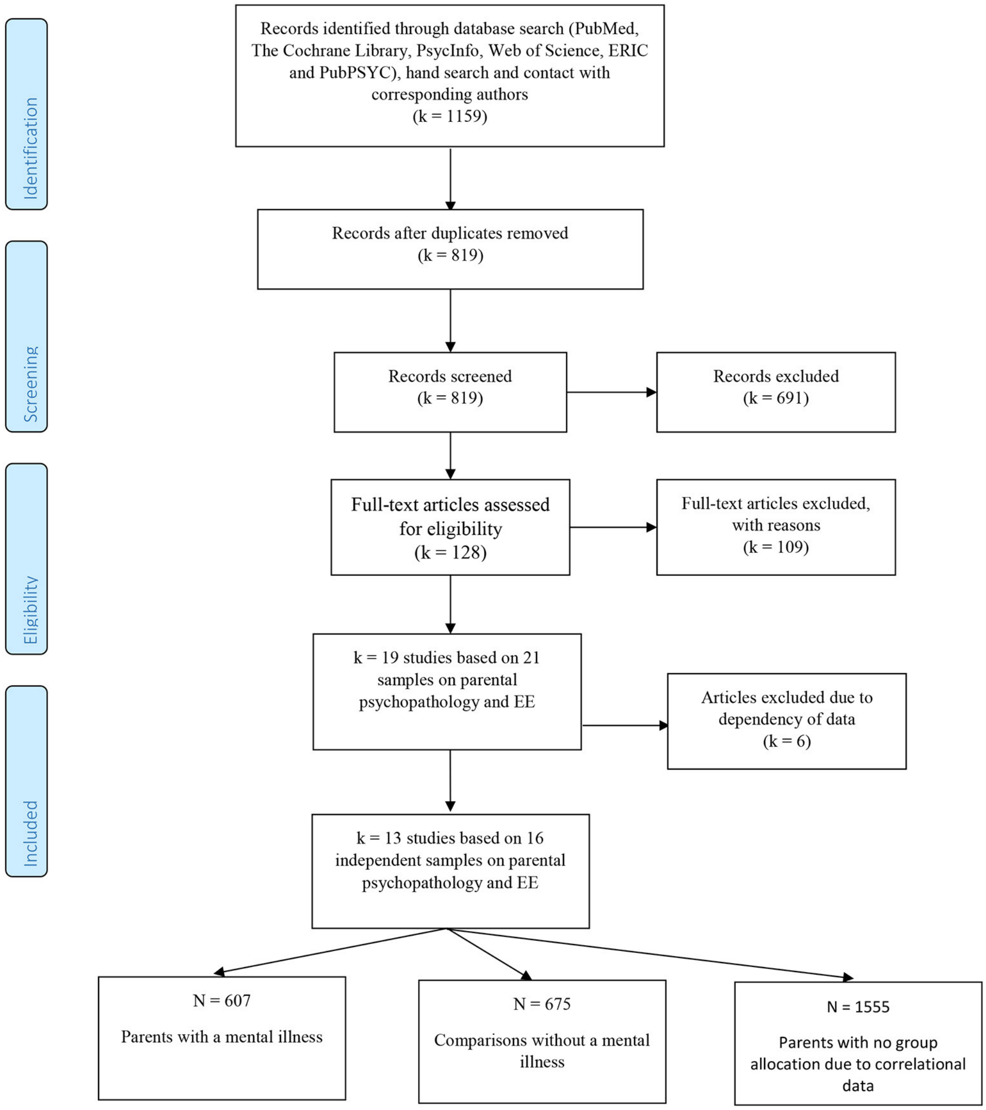


FIGURE 2 | Study flowchart.

A subset of study data was extracted by two raters independently, and inter-rater reliability was calculated for each variable. Inter-rater agreement for the coded study characteristics was $k = 1.00$ indicating perfect agreement, except for the variable type of comparison with $k = 0.43$ that resulted in moderate agreement (71).

Interrater-agreement for the coded moderator variables was between $k = 0.57$–1.00, indicating substantial agreement. Study quality was coded with *the Newcastle Ottawa rating scale for observational studies* NOS (72) by two independent raters (JF and NB). The coders completed a standardized form for each study independently to compute inter-rater reliability. An overall quality score was calculated by adding up all the criteria resulting in a maximum score of 9 for each study described in the **Supplementary Material**. Inter-rater agreement was $k = 0.49$ indicating moderate agreement (71). This is in line with results from recent research, indicating poor to medium inter-rater agreements on the NOS rating scale (73).

Effect Size Calculation

For the meta-analysis reporting on parental psychopathology and EE, correlational data were transformed into Fisher's *z*, studies reporting continuous data/means and standard deviation were transformed into Cohen's d and binary/dichotomized data into log odds ratios (74). Conversion of Effect sizes into Log Odds Ratios and variance was performed with esc Version 0.5.0 for R (version 3.6.1). Log Odds Ratio was used as the common index for meta-analysis to analyze the odds of being classified as HEE in groups of parents with a mental illness and healthy controls, and transformed back into Odds Ratios (OR) afterwards for improved intelligibility (74). In the case of studies comparing parents with a history of mental illness with current mental illness and to healthy controls, we chose the group currently suffering from symptoms. In case of different articles reporting on the same study but referring to a subsample's different sample sizes (e.g., the subsample of mothers or fathers with incomprehensible dropouts), the paper with the highest quality rating was chosen for meta-analysis to ensure one effect size per sample entering the analysis (75).

Meta-analysis was performed with Metafor Version 2.1-0 for R (Version 3.6.1) using the random effect model (REM) with DerSimonian-Laird method estimator for effect variance τ^2 (76, 77). Furthermore, heterogeneity of the estimated true effect was calculated using Cochran's *Q*-test for homogeneity and the I^2-statistic (78). Moderator analysis followed heterogeneous effects to identify influencing variables. For continuous variables, moderator analysis was performed with metafor using univariate metaregression models (MEM). Categorical moderator analyses were performed with metafor, recoding categorical variables into dichotomous dummy variables. For all estimated true effects, sensitivity analyses were performed using fixed effect models (FEM) as implemented in Metafor to examine biases due to the choice of the meta-analytic model. Additionally, the influence of potential outliers was examined by using studentized deleted residuals (79). Furthermore, publication bias was examined by funnel plot inspection and test of asymmetry with a rank correlation (80) and regression test (81).

RESULTS

Data on 16 independent comparisons (parents with a mental illness vs. without) derived from 13 studies with a total of $N = 2815$ parents ($n =$ 2,254 mothers, $n = 561$ fathers) were available for our analyses. The sample consisted of 607 parents with a diagnosis of a mental disorder, 675 healthy parents. Further, 1,533 parents could not be assigned to the group of parents with or without a mental illness because only correlational data on the association between EE and parental mental illness was reported from both parents with and without a mental illness within the same group (see **Table 1**). Nevertheless, OR were computed for the correlation based data. Studies were conducted in the USA, UK, and Australia.

For details on the studies as well as parental disorders, see **Table 2**. Study quality (**Table 2**; **Supplementary Material**) was generally medium with 6.25 on the NOS (min. 2, max. 9). Studies predominantly reported CRIT and lacked information on EOI. Because of this, the future analysis only refers to data on the CRIT specification of the HEE construct. Across 13 studies with k = 16 independent samples and unique effects, overall parental psychopathology was positively associated with CRIT ($\hat{\mu}_0$ =1.34 [95% CI = 1.01–1.77] $p < 0.05$). Cochran's *Q*-Test suggests variability among true effects (Q = 35.28, df = 15, $p = 0.022$). The variance in the true effect is estimated to be $\tau^2 = 0.15$. The amount of total variability between the observed effect sizes due to heterogeneity is estimated to be $I^2 = 57.49\%$, and was "moderate" (78).

TABLE 1 | Characteristics of parental psychopathology and EE studies with means, SDs, percentages.

Parental disorder	Maternal depression: 11 studies Maternal and paternal depression: 2 studies Anxiety disorder: 1 study Affective disorder, not further specified: 1 study
Family composition	80% mothers-child dyad 20% father-child dyad
Parental age	36.68 years (SD = 6.68 years)
Children's age	≤ 20 months (infants): 3 studies ≤ 6 years (pre-schoolers): 3 samples ≤ 12 years (school age kids): 7 samples ≤ 18 years (adolescents): 3 samples
Children's sex	54.7% female
Study design	Observational: 0 Longitudinal studies: 9 Experimental: 0 Controlled trials: 3 Randomized controlled trial: 1
Assessment setting	Clinic: 1 study (8%) Home: 4 studies (31%) Home and centre based: 3 studies (23%) Not reported: 5 studies (38%)
EE assessment tool	FMSS: 10 studies (79%) Preschool Five Minute Speech Sample: 1 study: (7 %) Camberwell Family Interview: 1 study (7%) Family Attitude Scale: 1 study (7%)
Grouping	Parents with a diagnosis of mental illness: *N* = 607 (*n* = 184 male, *n* = 423 female) Parents without any diagnosis of a mental illness: *N* = 675 (*n* = 219 male, *n* = 456 female) Group Membership n.A. due to correlational data: 1,533

Sensitivity Analyses

Two samples of one study (91) were identified as outliers by using externally standardized residuals. Controlling for those samples did result in marked differences; thus those samples were excluded from further analysis. Reiterating the analysis for the reduced sample under the REM revealed a small effect (92) between parental mental illness and CRIT ($\hat{\mu}_0$ =1.45 [95% CI = 1.19–1.76] $p < 0.001$) (see **Table 3**, **Figure 3**).

With the reduced sample, we tested the data under the FEM. The common true effect of the included studies is estimated to be $\hat{\theta} = 1.43$ [95% CI = 1.23–1.68], $z = 4.51$, $p <$ 0.0001). These findings are almost identical to those obtained applying the REM, and results seem to be robust for choosing a meta-analytic model.

Publication Bias

In terms of potential publication bias, a funnel plot inspection revealed no asymmetrical distribution of the observed effects around the average true effect (see **Figure 4**). The visual inspection is supported by the rank correlation test (Kendall's $\tau = -0.09$, $p = 0.67$) and the regression test ($z = -0.15$, $p = 0.88$) indicating no funnel plot asymmetry.

Performing Cochrane's *Q*-Test with the reduced sample, heterogeneity remained insignificant (Q = 16.58, df =

TABLE 2 | Studies included for meta-analysis with the dependent variable parental EE.

Study	Country	Disorder studied in parents	Clinical assessment tool	Setting	Female rate	Parent's age in years (M)	Child's age (M)	EE assessment	Study quality	*N*	OR
Psychogiou et al. (82)	UK	Depression	SCID, PHQ-9	Home	48%		3.9 years (SD = 0.8)	PFMSS	9	302	
					Mothers	36				144	1.55
					Fathers	39				158	1.34
Mellick et al. (83)	USA	Depression	SCID-I	N.A.	100%	40	10–16 months	FAS	6	81	2.08
Gravener Davis (84)	USA	Depression	DIS-IV, BDI	Home and centre based	100%	N.A.	24 months	FMSS	4	205	1.73
Gravener et al. (34)	USA	Depression	DIS-IV, BDI	Home and centre based	100%	31.68	20 months	FMSS	8	198	1.87
Burkhouse et al. (85)	USA	Depression	SADS-L, BDI-II	N.A.	100%	38.56	9.97 years	FMSS	7	100	0.87
Tompson et al. (86)	USA	Depression	SCID, BDI	Home and centre based	100%	42.2	8–12 years	FMSS	0	171	2.82
Gibb et al. (87)	USA	Depression, anxiety disorder	SADS-L, BDI	N.A.	100%	38.56	9.97 years (SD = 1.32)	FMSS	6	100	0.86
Netsi (36)	UK	Depression	SCID-I, EPDS	Home	50%	33.11	12 months	FMSS	7	103	0.70
Frye and Garber (42)	USA	Depression	SCID	N.A.	100%	38.56	11.88 years (SD = 0.55)	FMSS	7	194	2.36
Nelson et al. (88)	USA	Depression	SCID-IV, BDI	Home	100%	41	15.2 years	FMSS	9	739	1.31
Brennan et al. (89)	AUS	Depression	SCID	Home	0%	25.58 (at time of birth)	15.2 years	FMSS	8	300	0.68
Hirshfeld et al. (90)	USA	Depression, anxiety disorder	DIS	Clinic	100%	N.A.	11 years	FMSS	8	70	3.00
Schwartz et al. (91)	USA	Affective disorders	SADS-L	N.A.	100%	N.A.		CFI	4	252	
							1–9 years			25	4.8
							10–14 years			104	0.06
							15–19 years			123	1.25

Annotation: Studies are ordered by publication year with the most recent being at the top. N.A., not available; SCID, Structured Clinical Interview for DSM Disorders; SCID-IV, Structured Clinical Interview for DSM IV Disorders; PHQ-9, Patient Health Questionnaire-9; DIS-IV, Diagnostic Interview Schedule for DSM-IV; BDI, Beck's Depression Inventory; BDI-II, Beck's Depression Inventory Revision; SADS-L, Schedule for Affective Disorders and Schizophrenia- Lifetime version; EPDS, Edinburgh Postnatal Depression Scale; EE Assessment, Expressed Emotion Assessment; FMSS, Five Minute Speech Sample; PFMSS, Preschool Five Minute Speech Sample; CFI, Camberwell Family Interview; FAS, Family Attitude Scale; N, total number of participants; OR, Odds Ratio.

TABLE 3 | Summary statistics regarding parental psychopathology and CRIT.

Study sample	Mean ES (OR)	95% CI	*z* score	Q	τ^2	I^2	*k*	*N*
Initial sample	1.34	[1.01–1.77]	2.06	35.28**	0.15	57.49	16	2,815
Reduced sample	1.45***	[1.19–1.76]	3.71	16.58	0.02	21.58	14	2,686

Initial sample, all studies included in analysis; Reduced sample, sample after outlier removal; CI, confidence Interval; Q, Homogeneity statistic; τ^2, variance between true effects; I^2 amount of true variance among total variance; k, number of comparison- controls; N, total number of participants.
$^*p < 0.05$; $^{**}p < 0.01$; $^{***}p < 0.001$.

13, $p = 0.22$). The variance of the true effect is estimated to be $\tau^2 = 0.0277$ (SE = 0.0514). The amount of total variability between the observed effect sizes due to heterogeneity is estimated to be $I^2 = 21.58\%$, and was overall low (93). Nevertheless, the moderator analysis seemed appropriate due to the low sample numbers (k) included (see **Table 4**).

Meta-Regression

We performed the meta-regression with the reduced set of samples. Moderator analysis revealed child age as not significant when entered as a continuous variable ($\hat{\beta}_i = -0.001$, $Q_{\text{Model}} = 0.002$, df = 1, $p = 0.96$). Child age was a significant moderator for the strength of the association between parental mental illness and Crit, when entered as a categorical variable into the model

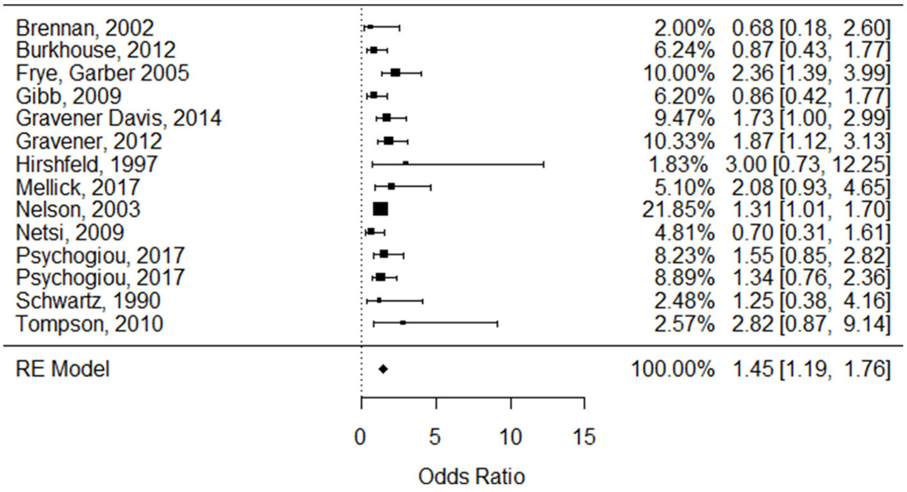


FIGURE 3 | Forest plot for the odds ratio based on the log scale of the association between parental psychopathology and high Expressed Emotion derived from 13 studies (based on 14 independent samples).

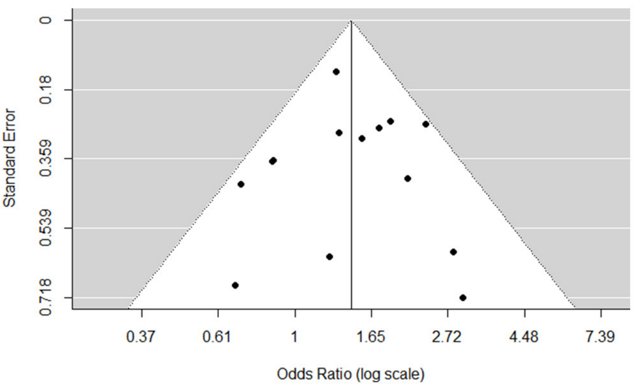


FIGURE 4 | Funnel plot for the odds of the association between parental mental illness and CRIT after removal of outliers.

(infants: $\hat{\beta}_i = 0.40$, $Q_{\text{Model}} = 12.36$, df $= 2$, $p = 0.0021$; pre-schoolers: $\hat{\beta}_i = 0.36$, $Q_{\text{Model}} = 12.23$, df $= 2$, $p = 0.0022$; school-age children: $\hat{\beta}_i = 0.36$, $Q_{\text{Model}} = 12.25$, df $= 2$, $p = 0.0022$; adolescents: $\hat{\beta}_i = 0.30$, $Q_{\text{Model}} = 12.30$, df $= 2$, $p = 0.0021$). In accordance with those results, the absence of a child's diagnosis was a significant moderator when analysed as a dichotomous moderator ($\hat{\beta}_i = 0.43$, $p < 0.0001$, $Q_{\text{Model}} = 1.86$, df $= 2$, $p = 0.17$) as τ^2 was reduced, thus explaining the variance through the moderator.

Study quality proved to be a non-significant moderator ($\hat{\beta}_i = -0.06$, $p = 0.31$) ($Q_{\text{Model}} = 1.03$, df $= 1$, $p = 0.31$), as did parental diagnostic category ($Q_{\text{Model}} = 1.02$, df $= 2$, $p = 0.60$). When parental sex was examined ($Q_{\text{Model}} = 2.49$, df $= 1$, $p = 0.11$), we observed significant associations with the female ($\hat{\beta}_i = 0.43$, $p < 0.0001$) but not male sex ($\hat{\beta}_i = -0.42$, $p = 0.11$).

TABLE 4 | Results of meta-regression including hypothesized moderators.

Moderator	$\hat{\beta}_i$	SE	95% CI
Intercept	1.83**	0.67	[0.51, 0.316]
Infant	−1.31	0.70	[−2.68, 0.06]
Pre-schooler	−1.19	0.72	[−2.6, 0.23]
School age	−1.06	0.65	[−2.33, 0.21]
Adolescent	−1.50*	0.68	[−2.83, −0.16]
Child diagnosis	−0.80*	0.31	[−1.40, −0.18]
Male gender	−0.54	2.9	[−1.10, 0.01]

$\hat{\beta}_i$, estimate of regression parameters; SE, adjusted standard error of regression parameter; CI, confidence interval.
*$p < 0.1$; *$p < 0.05$; **$p < 0.01$; ***$p < 0.001$.*

When entering parental sex, presence of the child's diagnosis and child-age category as predictors into the regression model (see **Table 4**) ($Q_{\text{Model}} = 11.87$, df $= 6$, $p = 0.06$), the amount of unaccounted variability decreased ($I^2 = 0\%$) and the moderators accounted for $R^2 = 100$ % of the heterogeneity. The child-age category adolescence ($\hat{\beta}_i = -1.495$, $p = 0.02$) and presence of the child's diagnosis ($\hat{\beta}_i = -0.80$, $p = 0.01$) remained significant predictors in the multiple meta-regression with adolescents producing small effects ($\hat{\mu} = 1.40$) and children with a diagnosis producing medium effects ($\hat{\mu} = 2.82$) (92) on EE when living with a parent suffering from a mental illness.

DISCUSSION

The aim of the present meta-analysis was to estimate the overall effects of a parental mental illness on EE compared to controls without any mental illness within the literature. With respect to existing studies on EE and youth psychopathology, we were able to depict a small but significant overall effect (OR = 1.45) between parental mental illness and CRIT. This finding provides support for our assumption that parental CRIT is a specific reactional style of parents with a mental illness and more frequent in parent-child relations within their families. Parents with a mental illness tend to react more critically and make more critical statements when asked about their relationship with their child. For this reason, HEE cannot be regarded as only a reaction to children's psychopathology (44), but as an interactional style in families in which a parent has a mental illness. The existence of a critical, negative family climate and harsh, intrusive parenting behavior thus proves to be a robust risk factor for a child's socio-emotional development (94), and as a stressor that may interact with a child's vulnerability (39) and stress reactivity (95). Thus, parental criticism may act as one mechanism in the transgenerational transmission of mental illness (9, 10). This effect proved to be stable toward the choice of the meta-analytic model and without significant publication bias.

Our analysis was limited to the coding of CRIT and lacks information on EOI, because data on EOI was neither sufficiently available nor reported, and the present studies mainly reported on CRIT. This is not very surprising as the use of EOI in studies on children lacks validity and is under discussion (24). An adaption of the EOI construct has already been demanded elsewhere and suggestions have been made to only incorporate self-sacrifice and overprotection, as those appear developmentally salient. Statements of attitude, excessive detail, and emotional displays within EOI do not appear striking when made by a parent about a minor child (24, 25, 96).

Surprisingly, only one (91) out of 13 studies used the CFI to assess EE. There appears to be a trend in studies published after 1997 using the shorter FMSS rather than the CFI, which initially was considered as gold standard tool to assess EE (21, 26).

Unfortunately, our sample only consisted of parents with depression and anxiety disorders, and our findings are limited to that diagnosis spectrum. CRIT can be regarded as reflecting the attributional and cognitive style typical of depression (97). However, information on EE in the families of parents with mental disorders other than depression and anxiety is urgently required to improve our understanding of family interactions, especially EE, as a mechanism of transmission.

Our sample consisted predominantly of mothers with a mental illness, and female controls. Unfortunately, 1,533 dyads could not be allocated to the clinical or control group due to correlational data from the studies included. Future studies should aim for a more balanced sex relation and be clear about group allocation. Female sex functioned as a significant moderator. Nonetheless, we cannot draw any conclusions about fathers and CRIT based on our data. This finding is congruent with the literature, because fathers have been neglected in the research on parents with a mental illness (98). There are indications that fathers with depression do not present with higher levels of HEE or CRIT, but that they do make fewer warm, positive remarks than healthy controls (36). This indicates a potential sex difference in the reactional and interactional style of parents with a mental illness, but it is a difference that needs clarification. The presence or absence of paternal warmth should come to the fore when studying fathers with a mental illness in the future, because that factor is not automatically included in the HEE/CRIT code and only is depicted indirectly within the LEE rating as it is one component that is rated and conglomerated into LEE/HEE. Parental sensitivity and warmth appear to be strong behavioral protective factors for children's development and pathology in the preschool age in the Transgenerational Transmission of Mental Disorders (99). Based on this consideration, a sex difference in the EE of preschoolers' parents and especially of the positive component warmth, is particularly important. Future studies should consider to report the level of parental warmth in addition to the HEE/LEE rating.

Implementing adolescent age into the regression resulted in small effects (OR = 1.40), but we can make no statement about younger ages. We were able to show a significant increase in overall effects (OR = 2.82) when children were presenting with a mental health problem themselves, providing support for EE CRIT acting as a mechanism in the transgenerational transmission of mental disorders. Considering HEE's prognostic power in predicting treatment response in adolescents (61–63), this finding appears fundamental. Adolescents with eating disorders show worse treatment response when living with a HEE parent. But the parental attitude about the relationship to the child does not only seem to be influenced by the burden provoked by the child's mental illness but the parent's mental health as well. Parents with a mental illness make more CRIT statements than healthy controls. These results support that children of parents with a mental illness are exposed to more CRIT in their home environment and, as they develop symptoms themselves, face even more parental CRIT and therefore are exposed to greater challenges in responding to treatment. Assessing parental psychopathology should be implemented in future studies observing EE and child treatment response.

One additional possible explanation is that genetically vulnerable children who may have a difficult temperament are exposed to overly critical parents, develop problem behavior and psychopathology over time. The children's problem behaviors provoke even more negativity and criticism from already burdened parents, leading to an internal vicious circle of mental illness, critical cognitions, attitudes, and reactions the children might adapt while growing up that appear on the level of family interactions in the Transgenerational Transmission of Mental Disorders system.

To our knowledge, this meta-analysis is the first to assess the overall effects of the presence or absence of a parental mental illness on CRIT, and to integrate the concept of CRIT within the Transgenerational Transmission of Mental Disorders system. It is important to identify CRIT's wider prevalence in parents with a mental illness, because future therapeutic interventions may identify and target parental CRIT as a specific component of parent-child-relations and reflection of the family climate in clinical assessments. As behavioral observations of parent-child-interactions are so time-consuming, costly, and require extensive training of observers, EE carries the potential to detect disrupted intra-family interactions within families of parents with a mental illness in everyday therapeutic interventions.

Limitations

We were unable to differentiate the children's mental illnesses, nor whether they were suffering from either internalizing or externalizing disorders. There was also a lack of specific information on children's age in the studies included. It is important to clearly differentiate children's age, and not just age categories, because the exposure to CRIT at an early age predicts the development of problem behavior later in life (38). This is essential, as during the first 3 years of life, children are especially vulnerable to dysfunctional, insensitive parent-child-interactions (100, 101) and the risk for psychopathology in offspring rises when a child is exposed to a stressful, critical home environment (51) and HEE parental attitudes. In future studies it would be worthwhile to focus on particularly vulnerable ages and insensitive parenting, in particular CRIT, to be able to adapt and implement preventive programs at an early stage.

Study quality did not function as a moderator, and inter-rater-agreement was medium despite the extensive training of coders. Furthermore, inter-rater agreement of the study variable *type of comparison* was moderate due to the difficulty of rating comparisons in population-based studies. Our search was restricted to articles in English and German, which may have precluded the identification of other relevant studies, although we included the gray literature to avoid the "file drawer-problem", as published studies most often report significant findings that disturb the overall balance of results. Furthermore, data was exclusively descended from English speaking countries within the Organization for Economic Co-operation and Development (OECD).

The systematic literature search was updated last in November 2021, thus potentially new articles published after November 2021 are not included in this review. However, in order to be able to complete a review and meta-analysis, one has to come to a decision of when to stop and we believe that we were able to arrive at significant results with the studies included, especially in light of the fact that results of the publication bias analysis do not indicate a distortion and according to fail safe n analysis $k = 36$ studies would need to be included to change the result to non-significance.

CONCLUSIONS

The current study highlights the dearth of studies on EE in families of mentally ill parents and their children, who already carry a higher risk of developing mental illnesses themselves. Established effects of CRIT and parental mental illness are significant, although generally small, and become stronger as offspring develop mental disorders themselves. These results support the importance of HEE/CRIT as a mechanism in the Transgenerational Transmission of Mental Disorders and as a firm component of dysfunctional parent-child-interactions. Future studies are needed to deepen our understanding of EE and particularly of EOI and warmth in families in which parents suffer from a mental illness. The research on EE in families of children of parents with a mental illness has the potential to guide future preventive interventions and may be exploited in interventions especially developed to improve parent-child-relations.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Materials**, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

JF designed the study, performed and updated the search strategy, completed the statistical analysis, and wrote the protocol. LMD did the independent full text screening as second rater and was involved in the inclusion process of studies. NB did the quality assessment and coding of study variables as second rater. JA gave methodological advice and supported the drafting of the manuscript. HC functions as PHD advisor and supervised the study and preparation of manuscript and commented on the whole manuscript. All authors contributed to the article and approved the submitted version.

ACKNOWLEDGMENTS

We would like to thank our student assistant Michelle Marschall for her support in the preparation of this manuscript.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2021.721796/full#supplementary-material

REFERENCES

1. Bassani DG, Padoin CV, Veldhuizen S. Counting children at risk: exploring a method to estimate the number of children exposed to parental mental illness using adult health survey data. *Soc Psychiatry Psychiatr Epidemiol.* (2008) 43:927–35. doi: 10.1007/s00127-008-0376-3
2. Maybery DJ, Reupert AE, Patrick K, Goodyear M, Crase L. Prevalence of parental mental illness in Australian families. *Psychiatri Bull.* (2009) 33:22–6. doi: 10.1192/pb.bp.107.018861
3. Gravel R, Béland Y. The Canadian Community Health Survey: mental health and well-being. *Can J Psychiatry.* (2005) 50:573–9. doi: 10.1177/070674370505001002
4. Reupert AE, J Maybery D, Kowalenko NM. Children whose parents have a mental illness: prevalence, need and treatment. *Med J Aust.* (2013) 199:S7–9. doi: 10.5694/mja11.11200
5. Wille N, Bettge S, Ravens-Sieberer U. Risk and protective factors for children's and adolescents' mental health: results of the BELLA study. *Eur Child Adolesc Psychiatr.* (2008) 17:133–47. doi: 10.1007/s00787-008-1015-y

6. van Santvoort F, Hosman CM, Janssens JM, van Doesum KT, Reupert A, van Loon LM. The impact of various parental mental disorders on children's diagnoses: a systematic review. *Clin Child Fam Psychol Rev.* (2015) 18:281–99. doi: 10.1007/s10567-015-0191-9
7. Christiansen H, Röhrle B, Fahrer J, Stracke M, Dobener L-M. *Kinder von Eltern mit psychischen Erkrankungen: State of the Art für Psychotherapeutinnen, Pädiaterinnen, Pädagoginnen*. Wiesbaden: Springer Fachmedien Wiesbaden (2020). doi: 10.1007/978-3-658-30519-2
8. Goodman SH, Gotlib, IH. Transmission of risk to children of depressed parents: Integration and conclusions. In: Goodman SH, Gotlib IH, editors. *Children of depressed parents: Mechanisms of risk and implications for treatment.* Washington, DC: American Psychological Association (2002). p. 307–26. doi: 10.1037/10449-012
9. Hosman CM, van Doesum KT, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. the scientific basis to a comprehensive approach. *Austr J Adv Mental Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
10. Mattejat F, Remschmidt H. The children of mentally ill parents. *Dtsch Arztebl Int.* (2008) 105:413. doi: 10.3238/arztebl.2008.0413
11. Christiansen H, Reck C, Zietlow A-L, Otto K, Steinmayr R, Wirthwein L, et al. Children of Mentally III Parents at Risk Evaluation (COMPARE): design and methods of a randomized controlled multicenter study—part I. *Front Psychiatry.* (2019) 10:128. doi: 10.3389/fpsyt.2019.00128
12. Goodman SH, Gotlib IH. Risk for psychopathology in the children of depressed mothers: a developmental model for understanding mechanisms of transmission. *Psychol Rev.* (1999) 106:458. doi: 10.1037/0033-295X.106.3.458
13. Stracke M, Gilbert K, Kieser M, Klose C, Krisam J, Ebert DD, et al. COMPARE family (Children of mentally ill parents at risk evaluation): a study protocol for a preventive intervention for children of mentally ill parents (Triple P, evidence-based program that enhances parentings skills, in addition to gold-standard CBT with the mentally ill parent) in a multicenter RCT—Part II. *Front Psychiatry.* (2019) 10:54. doi: 10.3389/fpsyt.2019.00054
14. Bugental DB, Johnston C. Parental and child cognitions in the context of the family. *Annu Rev Psychol.* (2000) 51:315–44. doi: 10.1146/annurev.psych.51.1.315
15. Barrowclough C, Hooley JM. Attributions and expressed emotion: a review. *Clin Psychol Rev.* (2003) 23:849–80. doi: 10.1016/S0272-7358(03)00075-8
16. Feldman R. Parenting behavior as the environment where children grow. In: Mayes LC, Lewis M, editors. *The Cambridge Handbook of Environment in Human Development.* New York, NY: Cambridge University Press (2012). p. 535–67.
17. Feldman R. *Coding Interactive Behavior Manual. Unpublished Manual.* Ramat Gan: Bar-Ilan University, Israel (1998).
18. McCarty CA, Lau AS, Valeri SM, Weisz JR. Parent-child interactions in relation to critical and emotionally overinvolved expressed emotion (EE): is EE a proxy for behavior? *J Abnorm Child Psychol.* (2004) 32:83–93. doi: 10.1023/B:JACP.0000007582.61879.6f
19. Brown GW, Monck EM, Carstairs GM, Wing JK. Influence of family life on the course of schizophrenic illness. *Br J Prev Soc Med.* (1962) 16:55. doi: 10.1136/jech.16.2.55
20. Hooley JM. Expressed emotion and relapse of psychopathology. *Annu Rev Clin Psychol.* (2007) 3:329–52. doi: 10.1146/annurev.clinpsy.2.022305.095236
21. Brown GW, Rutter M. The measurement of family activities and relationships: a methodological study. *Human Relat.* (1966) 19:241–63. doi: 10.1177/001872676601900301
22. Leeb B. *Manual zur Kodierung der 'Expressed Emotion'aus der Fünf-Minuten-Sprechstichprobe (FMSS).* Heidelberg.
23. Brown GW. *The Discovery of Expressed Emotion: Induction or Deduction.* New York, NY: Guilford press (1985).
24. Rea HM, Factor RS, Kao W, Shaffer A. A meta-analytic review of the five minute speech sample as a measure of family emotional climate for youth: relations with internalizing and externalizing symptomatology. *Child Psychiatry Hum Dev.* (2020) 51:656–69. doi: 10.1007/s10578-020-00964-z
25. Gar NS, Hudson JL. An examination of the interactions between mothers and children with anxiety disorders. *Behav Res Ther.* (2008) 46:1266–74. doi: 10.1016/j.brat.2008.08.006
26. Rutter M, Brown GW. The reliability and validity of measures of family life and relationships in families containing a psychiatric patient. *Soc Psychiatry.* (1966) 1:38–53. doi: 10.1007/BF00583828
27. Magaña AB, Goldstein MJ, Karno M, Miklowitz DJ, Jenkins J, Falloon IR. A brief method for assessing expressed emotion in relatives of psychiatric patients. *Psychiatry Res.* (1986) 17:203–12. doi: 10.1016/0165-1781(86)90049-1
28. Daley D. *Coding manual for the Pre-School Five Minute Speech Sample.* (2001). [Epub ahead of print].
29. Kavanagh DJ, O'Halloran P, Manicavasagar V, Clark D, Piatkowska O, Tennant C, et al. The Family Attitude Scale: reliability and validity of a new scale for measuring the emotional climate of families. *Psychiatry Res.* (1997) 70:185–95. doi: 10.1016/S0165-1781(97)00033-4
30. Wiedemann G, Rayki O, Feinstein E, Hahlweg K. The Family Questionnaire: development and validation of a new self-report scale for assessing expressed emotion. *Psychiatry Res.* (2002) 109:265–79. doi: 10.1016/S0165-1781(02)00023-9
31. Vaughn CE, Leff JP. The influence of family and social factors on the course of psychiatric illness: a comparison of schizophrenic and depressed neurotic patients. *Br J Psychiatry.* (1976) 129:125–37. doi: 10.1192/bjp.129.2.125
32. Hooley JM. Expressed emotion: a review of the critical literature. *Clin Psychol Rev.* (1985) 5:119–39. doi: 10.1016/0272-7358(85)90018-2
33. Brewin CR, MacCarthy B, Duda K, Vaughn CE. Attribution and expressed emotion in the relatives of patients with schizophrenia. *J Abnorm Psychol.* (1991) 100:546. doi: 10.1037/0021-843X.100.4.546
34. Gravener JA, Rogosch FA, Oshri A, Narayan AJ, Cicchetti D, Toth SL. The relations among maternal depressive disorder, maternal expressed emotion, and toddler behavior problems and attachment. *J Abnorm Child Psychol.* (2012) 40:803–13. doi: 10.1007/s10802-011-9598-z
35. Campbell SB, Shaw DS, Gilliom M. Early externalizing behavior problems: toddlers and preschoolers at risk for later maladjustment. *Dev Psychopathol.* (2000) 12:467–88. doi: 10.1017/S0954579400003114
36. Netsi E. *Paternal Depression and Infant Outcomes: The Role of Expressed Emotion.* Oxford (2009).
37. Hibbs ED, Hamburger SD, Lenane M, Rapoport JL, Kruesi MJ, Keysor CS, et al. Determinants of expressed emotion in families of disturbed and normal children. *J Child Psychol Psychiatry.* (1991) 32:757–70. doi: 10.1111/j.1469-7610.1991.tb01900.x
38. Peris TS, Baker BL. Applications of the expressed emotion construct to young children with externalizing behavior: stability and prediction over time. *J Child Psychol Psychiatry.* (2000) 41:457–62. doi: 10.1111/1469-7610.00630
39. Peris TS, Miklowitz DJ. Parental expressed emotion and youth psychopathology: new directions for an old construct. *Child Psychiatry Hum Dev.* (2015) 46:863–73. doi: 10.1007/s10578-014-0526-7
40. Rogosch FA, Cicchetti D, Toth SL. Expressed emotion in multiple subsystems of the families of toddlers with depressed mothers. *Dev Psychopathol.* (2004) 16:689–706. doi: 10.1017/S0954579404004730
41. Bolton C, Calam R, Barrowclough C, Peters S, Roberts J, Wearden A, et al. Expressed emotion, attributions and depression in mothers of children with problem behaviour. *J Child Psychol Psychiatry.* (2003) 44:242–54. doi: 10.1111/1469-7610.00117
42. Frye AA, Garber J. The relations among maternal depression, maternal criticism, and adolescents? Externalizing and internalizing symptoms. *J Abnorm Child Psychol.* (2005) 33:1–11. doi: 10.1007/s10802-005-0929-9
43. Beardslee WR, Gladstone TR, O'Connor EE. Transmission and prevention of mood disorders among children of affectively ill parents: a review. *J Am Acad Child Adolesc Psychiatry.* (2011) 50:1098–109. doi: 10.1016/j.jaac.2011.07.020
44. Schimmelmann BG, Meng H, Koch E, Karle M, Preuss U, Schulte-Markwort M. Expressed Emotion im Verlauf kinder-und jugendpsychiatrischer Störungen. *Fortschr Neurol Psychiatr.* (2003) 71:517–26. doi: 10.1055/s-2003-42870
45. Daley D, Sonuga-Barke EJ, Thompson M. Assessing expressed emotion in mothers of preschool AD/HD children: psychometric properties of a modified speech sample. *Br J Clin Psychol.* (2003) 42:53–67. doi: 10.1348/014466503762842011

46. Miklowitz DJ. The role of family systems in severe and recurrent psychiatric disorders: a developmental psychopathology view. *Dev Psychopathol.* (2004) 16:667–88. doi: 10.1017/S0954579404004729
47. Jacobsen T, Hibbs E, Ziegenhain U. Maternal expressed emotion related to attachment disorganization in early childhood: a preliminary report. *J Child Psychol Psychiatry.* (2000) 41:899–906. doi: 10.1111/1469-7610.00677
48. Schloss S, Schramm M, Christiansen H, Scholz K-K, Schuh LC, Doepfner M, et al. Expressed emotion, mother-child relationship, and ADHD symptoms in preschool-a study on the validity of the German Preschool Five Minute Speech Sample. *Z Kinder Jugendpsychiatr Psychother.* (2015) 43:425–31. doi: 10.1024/1422-4917/a000384
49. Asarnow JR, Goldstein MJ, Tompson M, Guthrie D. One-year outcomes of depressive disorders in child psychiatric in-patients: evaluation of the prognostic power of a brief measure of expressed emotion. *J Child Psychol Psychiatry.* (1993) 34:129–37. doi: 10.1111/j.1469-7610.1993.tb00975.x
50. Peris TS, Yadegar M, Asarnow JR, Piacentini J. Pediatric obsessive compulsive disorder: family climate as a predictor of treatment outcome. *J Obs.-Compuls Relat Disord.* (2013) 1:267–73. doi: 10.1016/j.jocrd.2012.07.003
51. Caspi A, Moffitt TE, Morgan J, Rutter M, Taylor A, Arseneault L, et al. Maternal expressed emotion predicts children's antisocial behavior problems: using monozygotic-twin differences to identify environmental effects on behavioral development. *Dev Psychol.* (2004) 40:149. doi: 10.1037/0012-1649.40.2.149
52. Rutter M, Yule B, Quinton D, Rowlands O, Yule W, Berger M. Attainment and adjustment in two geographical areas: III—Some factors accounting for area differences. *Br J Psychiatry.* (1975) 126:520–33. doi: 10.1192/bjp.126.6.520
53. Vostanis P, Nicholls J, Harrington R. Maternal expressed emotion in conduct and emotional disorders of childhood. *J Child Psychol Psychiatry.* (1994) 35:365–76. doi: 10.1111/j.1469-7610.1994.tb01168.x
54. Hibbs ED, Zahn TP, Hamburger SD, Kruesi MM, Rapoport JL. Parental expressed emotion and psychophysiological reactivity in disturbed and normal children. *Br J Psychiatry.* (1992) 160:504–10. doi: 10.1192/bjp.160.4.504
55. Hibbs ED, Hamburger SD, Kruesi MJ, Lenane M. Factors affecting expressed emotion in parents of ill and normal children. *Am J Orthopsychiatry.* (1993) 63:103–12. doi: 10.1037/h0079395
56. Asarnow JR, Tompson M, Hamilton EB, Goldstein MJ, Guthrie D. Family-expressed emotion, childhood-onset depression, and childhood-onset schizophrenia spectrum disorders: is expressed emotion a nonspecific correlate of child psychopathology or a specific risk factor for depression? *J Abnorm Child Psychol.* (1994) 22:129–46. doi: 10.1007/BF02167896
57. Asarnow JR, Tompson M, Woo S, Cantwell DP. Is expressed emotion a specific risk factor for depression or a nonspecific correlate of psychopathology? *J Abnorm Child Psychol.* (2001) 29:573–83. doi: 10.1023/a:1012237411007
58. Christiansen H, Oades RD, Psychogiou L, Hauffa BP, Sonuga-Barke EJ. Does the cortisol response to stress mediate the link between expressed emotion and oppositional behavior in Attention-Deficit/Hyperactivity-Disorder (ADHD)? *Behav Brain Funct.* (2010) 6:45. doi: 10.1186/1744-9081-6-45
59. Miklowitz DJ, Biuckians A, Richards JA. Early-onset bipolar disorder: a family treatment perspective. *Dev Psychopathol.* (2006) 18:1247–65. doi: 10.1017/S0954579406060603
60. Millman Z. *Expressed Emotion and Levels of Family Psychopathology in Parents of Bipolar Adolescents*. Boulder (2011).
61. Le Grange D, Eisler I, Dare C, Hodes M. Family criticism and self-starvation: a study of expressed emotion. *J Fam Ther.* (1992) 14:177–92. doi: 10.1046/j.1992.00451.x
62. Szmukler GI, Eisler I, Russell GF, Dare C. Anorexia nervosa, parental 'expressed emotion'and dropping out of treatment. *Br J Psychiatry.* (1985) 147:265–71. doi: 10.1192/bjp.147.3.265
63. van Furth EF, van Strien DC, Martina LM, van Son MJ, Hendrickx JJ, van Engeland H. Expressed emotion and the prediction of outcome in adolescent eating disorders. *Int J Eat Disord.* (1996) 20:19–31. doi: 10.1002/(SICI)1098-108X(199607)20:1<19::AID-EAT3>3.0.CO;2-7
64. Parker G, Johnson P. Parenting and schizophrenia: an Australian study of expressed emotion. *Aust N Z J Psychiatry.* (1987) 21:60–6. doi: 10.3109/00048678709160900
65. McGuire JB, Earls F. Research note: the test-re test stability of the five minute speech sample in parents of disadvantaged, minority children. *J Child Psychol Psychiatry.* (1994) 35:971–9. doi: 10.1111/j.1469-7610.1994.tb02306.x
66. Sandberg S, Rutter M, Järvi J. Brief measure of expressed emotion: internal consistency and stability over time. *Int J Methods Psychiatr Res.* (2003) 12:182–91. doi: 10.1002/mpr.155
67. Hastings RP, Daley D, Burns C, Beck A. Maternal distress and expressed emotion: cross-sectional and longitudinal relationships with behavior problems of children with intellectual disabilities. *Am J Ment Retard.* (2006) 111:48–61. doi: 10.1352/0895-8017(2006)111[48:MDAEEC]2.0.CO;2
68. Richards JS, Vásquez AA, Rommelse NN, Oosterlaan J, Hoekstra PJ, Franke B, et al. A follow-up study of maternal expressed emotion toward children with attention-deficit/hyperactivity disorder (ADHD): relation with severity and persistence of ADHD and comorbidity. *J Am Acad Child Adolesc Psychiatry.* (2014) 53:311–9. e1. doi: 10.1016/j.jaac.2013.11.011
69. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Ann Intern Med.* (2009) 151:264–9. doi: 10.7326/0003-4819-151-4-200908180-00135
70. Leeb B, Hahlweg K, Goldstein MJ, Feinstein E, Mueller U, Dose M, et al. Cross-national reliability, concurrent validity, and stability of a brief method for assessing expressed emotion. *Psychiatry Res.* (1991) 39:25–31. doi: 10.1016/0165-1781(91)90005-A
71. Landis JR, Koch GG. The measurement of observer agreement for categorical data. *Biometrics.* (1977) 33:159–74. doi: 10.2307/2529310
72. Wells GA, Tugwell P, O'Connell D, Welch V, Peterson J, Shea B, et al. *The Newcastle-Ottawa Scale (NOS) for Assessing the Quality of Nonrandomized Studies in Meta-Analyses*. (2015). Available online at: http://www3.med.unipmn.it/dispense_ebm/2009-2010/Corso%20Perfezionamento%20EBM_Faggiano/NOS_oxford.pdf (accessed August 12, 2019).
73. Hartling L, Milne A, Hamm MP, Vandermeer B, Ansari M, Tsertsvadze A, et al. Testing the Newcastle Ottawa Scale showed low reliability between individual reviewers. *J Clin Epidemiol.* (2013) 66:982–93. doi: 10.1016/j.jclinepi.2013.03.003
74. Borenstein M, Hedges LV, Higgins JP, Rothstein HR. *Introduction to Meta-Analysis*. Chichester: John Wiley & Sons. (2009) doi: 10.1002/9780470743386
75. Rebar AL, Stanton R, Geard D, Short C, Duncan MJ, Vandelanotte C. A meta-meta-analysis of the effect of physical activity on depression and anxiety in non-clinical adult populations. *Health Psychol Rev.* (2015) 9:366–78. doi: 10.1080/17437199.2015.1022901
76. Deeks JJ, Higgins JP, Altman DG. Analysing data and undertaking meta-analyses. In: Higgins JP, Green S, editors. *Cochrane handbook for systematic reviews of interventions.* Chichester, NH: John Wiley & Sons (2019). p. 243–84.
77. DerSimonian R, Laird N. Meta-analysis in clinical trials. *Control Clin Trials.* (1986) 7:177–88. doi: 10.1016/0197-2456(86)90046-2
78. Higgins JP, Thompson SG. Quantifying heterogeneity in a meta-analysis. *Stat Med.* (2002) 21:1539–58. doi: 10.1002/sim.1186
79. Viechtbauer W, Cheung MW-L. Outlier and influence diagnostics for meta-analysis. *Res Synth Methods.* (2010) 1:112–25. doi: 10.1002/jrsm.11
80. Begg CB, Mazumdar M. Operating characteristics of a rank correlation test for publication bias. *Biometrics.* (1994) 50:1088–101. doi: 10.2307/2533446
81. Egger M, Smith GD, Schneider M, Minder C. Bias in meta-analysis detected by a simple, graphical test. *BMJ.* (1997) 315:629–34. doi: 10.1136/bmj.315.7109.629
82. Psychogiou L, Moberly NJ, Parry E, Selina N, Kallitsoglou A, Russell G. Parental depressive symptoms, children's emotional and behavioural problems, and parents' expressed emotion—Critical and positive comments. *PLoS One.* (2017) 12:e0183546. doi: 10.1371/journal.pone.0183546
83. Mellick W, Sharp C, Wilkinson A. The moderating role of maternal depression in the relation between adolescent behavioral inhibition and maternal critical expressed emotion. *Child Psychiatry Hum Dev.* (2017) 48:546–53. doi: 10.1007/s10578-016-0680-1
84. Gravener Davis JA. *Representations of Toddler, Self, and Caregiver in Mothers With and Without Major Depressive Disorder and Their Relation to Child*

Socioemotional Functioning Over a One-Year Follow-Up Period. New York, NY: (2014).

85. Burkhouse K, Uhrlass D, Stone L, Knopik V, Gibb B. Expressed emotion-criticism and risk of depression onset in children. *J Clin Child Adolesc Psychol.* (2012) 41:771–7. doi: 10.1080/15374416.2012.703122
86. Tompson MC, Pierre CB, Boger KD, McKowen JW, Chan PT, Freed RD. Maternal depression, maternal expressed emotion, and youth psychopathology. *J Abnorm Child Psychol.* (2010) 38:105–17. doi: 10.1007/s10802-009-9349-6
87. Gibb BE, Uhrlass DJ, Grassia M, Benas JS, McGeary J. Children's inferential styles, 5-HTTLPR genotype, and maternal expressed emotion-criticism: an integrated model for the intergenerational transmission of depression. *J Abnorm Psychol.* (2009) 118:734–45. doi: 10.1037/a0016765
88. Nelson DR, Hammen C, Brennan PA, Ullman JB. The impact of maternal depression on adolescent adjustment: the role of expressed emotion. *J Consult Clin Psychol.* (2003) 71:935–44. doi: 10.1037/0022-006X.71.5.935
89. Brennan PA, Hammen C, Katz AR, Le Brocque RM. Maternal depression, paternal psychopathology, and adolescent diagnostic outcomes. *J Consult Clin Psychol.* (2002) 70:1075–85. doi: 10.1037/0022-006X.70.5.1075
90. Hirshfeld DR, Biederman J, Brody L, Faraone SV, Rosenbaum JF. Expressed emotion toward children with behavioral inhibition: associations with maternal anxiety disorder. *J Am Acad Child Adolesc Psychiatry.* (1997) 36:910–7. doi: 10.1097/00004583-199707000-00012
91. Schwartz CE, Dorer DJ, Beardslee WR, Lavori PW, Keller MB. Maternal expressed emotion and parental affective disorder: risk for childhood depressive disorder, substance abuse, or conduct disorder. *J Psychiatr Res.* (1990) 24:231–50. doi: 10.1016/0022-3956(90)90013-G
92. Cohen J. *Statistical Power Analysis for the Behavioral Sciences*. London: Routledge. (2013) doi: 10.4324/9780203771587
93. Higgins JP, Thompson SG, Deeks JJ, Altman DG. Measuring inconsistency in meta-analyses. *BMJ.* (2003) 327:557–60. doi: 10.1136/bmj.327.7414.557
94. Seifer R. Young children with mentally ill parents: Resilient developmental systems. In: Luthar SS, editor. *Resilience and Vulnerability: Adaptation in the Context of Childhood Adversities.* Cambridge: Cambridge University Press (2003). p. 29–49.
95. Sapolsky RM. The possibility of neurotoxicity in the hippocampus in major depression: a primer on neuron death. *Biol Psychiatry.* (2000) 48:755–65. doi: 10.1016/S0006-3223(00)00971-9
96. McCarty CA, Weisz JR. Correlates of expressed emotion in mothers of clinically-referred youth: an examination of the five-minute speech sample. *J Child Psychol Psychiatry*. (2002) 43:759–68. doi: 10.1111/1469-7610.00090
97. Beck AT. *Cognitive Therapy of Depression*. New York, NY: Guilford press (1979).
98. Reupert A, Maybery D. Fathers' experience of parenting with a mental illness. *Fam Soc.* (2009) 90:61–8. doi: 10.1606/1044-3894.3846
99. Priel A, Djalovski A, Zagoory-Sharon O, Feldman R. Maternal depression impacts child psychopathology across the first decade of life: oxytocin and synchrony as markers of resilience. *J Child Psychol Psychiatry*. (2019) 60:30–42. doi: 10.1111/jcpp.12880
100. Rutter M. Clinical implications of attachment concepts: retrospect and prospect. *J Child Psychol Psychiatry*. (1995) 36:549–71. doi: 10.1111/j.1469-7610.1995.tb01311.x
101. Cohn JF, Tronick E. Specificity of infants' response to mothers' affective behavior. *J Am Acad. Child Adolesc Psychiatry*. (1989) 28:242–8. doi: 10.1097/00004583-198903000-00016

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

APPENDIX A

Table A1 | Full search strategy for databases; terms within categories were combined with OR, between categories with AND.

Population	Disorder	Intervention
• Parent* • Child • Maternal • Parental • "Mentally ill parents"	• Mental* • Mental illness • Mentally ill • Mental disorder* • Mental health problems • Mental disease • Affective disorder* • Bipolar disorder • Anxiety disorder • Phobia • Specific phobia • Generalized anxiety disorder • Depression* • Panic disorder* • Substance abuse • Addictive disorder • Addiction* • Eating disorder* • Anorexia nervosa • Bulimia nervosa • Insomnia • Obsessive compulsive disorder* (OCD) • Impulse control disorder* • Adjustment disorder* • Post traumatic stress disorder* (PTBS) • Personality disorder* • Borderline • Somatoform disorder* • Hypochondria • Psychososmatic disorder*	• Expressed Emotion • EE • Five minute speech sample • Camberwell family interview • Family questionnaire • Family attitude scale

**truncation or wildcard.*

frontiers in Psychiatry

BRIEF RESEARCH REPORT
published: 14 February 2022
doi: 10.3389/fpsyt.2022.824241

An Evidence-Based Practice Developed *in-situ*: Let's Talk About Children and a Consolidation of Its Evidence Base

Becca Allchin[1,2]* *and Tytti Solantaus*[3]

[1] *Eastern Health, Mental Health Program, Melbourne, VIC, Australia,* [2] *School of Rural Health, Monash University, Melbourne, VIC, Australia,* [3] *Department of Public Health and Welfare, Finnish Institute for Health and Welfare, Helsinki, Finland*

Background: Traditional models of evidence-based practice assume knowledge is developed in research settings before being installed in practice settings. The role practice settings can play in enhancing effectiveness and enabling sustainability is not therefore acknowledged. Developing interventions *in-situ* alongside developing their evidence base, provides another pathway to evidence-based practice. One example is Let's Talk about Children (LTC), a brief family-focused intervention that promotes parent, family and child wellbeing. Let's Talk about Children has been developed and adapted to respond to the context into which it has been established, leading to different descriptions reported in its 20 year collection of evidence. Collating the diverse literature on LTC, this paper showcases an evidence-based practice developed *in-situ* in order to guide future innovation.

Method: Using an integrative review, key literature using LTC were identified through electronic databases and snowballing techniques. Constant comparison analysis synthesized the data to develop patterns and themes.

Findings: From the 26 records, three forms of LTC were identified and outcomes related to parents, family and child wellbeing, implementation and sustainability were collated. Consolidated outcomes show overall agreement in effectiveness and acceptability outcomes across different settings and populations. Implementation and sustainability impacts are entwined with the context, and influenced by its development *in-situ*.

Conclusions: The study documents that the *in-situ* model is effective at developing sustainable evidence-based practice. In consolidating the evidence, the review clarified LTC's forms and outcomes, and draws attention to the importance of research on mechanisms of change.

Keywords: Evidence-based practice, Let's Talk about Children, family-focused practice, parental mental health, mental health promotion, child wellbeing, prevention in child mental health

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Violette McGaw,
Lifespan Psychology, Australia
Polina Anang,
University of Manitoba, Canada

Correspondence:
Becca Allchin
rebecca.allchin@monash.edu

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *29 November 2021*
Accepted: *13 January 2022*
Published: *14 February 2022*

Citation:
Allchin B and Solantaus T (2022) An Evidence-Based Practice Developed in-situ: Let's Talk About Children and a Consolidation of Its Evidence Base.
Front. Psychiatry 13:824241.
doi: 10.3389/fpsyt.2022.824241

INTRODUCTION

Evidence-based practice emerged in the concept of evidence-based medicine in the 1990's (1) which stressed applying evidence from relevant research to clinical decision making rather than relying on intuition. Evidence-based practice's endorsement led to the rise of interventions created and tested under research conditions that would then be disseminated to practice. This has been described as a 'pipeline' process; developing *efficacy*–testing if a practice could work under tightly controlled conditions, then *effectiveness*–testing if it does work in less controlled conditions, before *disseminating*–getting the practice to be utilized in service settings (2, 3). Emphasis on each of those phases has shifted, with efficacy dominating the early years of evidence-based practice, moving to effectiveness to build more generalizability and then to dissemination to improve uptake in routine practice (3, 4). More recently, this latter phase has shifted to a focus on implementation, seen as a more active process of equipping services to adopt and sustain such practices (4).

The concept of applying evidence to practice is hard to argue with, however, debate about what constitutes as evidence, and how it is applied has raised questions about the concept and development of evidence-based practices (5–7). The pipeline approach to developing interventions has resulted in interventions that may appear successful but not continue to provide benefits to end-users due to difficulties in implementing or sustaining them in practice (8). Additionally, the 15–20-year process can result in the implemented practice being already outdated by new evidence (4). Hawe (7) indicated that the pipeline process assumes a unidirectional pathway from research to practice, with knowledge developed in research domains before being "installed" into practice domains. Such a unidirectional process of knowledge development does not recognize the role practice settings can have in shaping evidenced-based practices in general, and especially where local-level adaptations may be important for enhancing effectiveness or driving sustainability (7, 9–11).

Another pathway to evidence-based practice has placed a greater value on the practice setting, by developing and adapting interventions *in-situ* while building evidence. One example of this is the Finnish, *Lapset puheeksi,* or in English, Let's Talk about Children (LTC), a family-focused practice with a specific emphasis on the parenting role and the needs of their children (12). The second author developed the first version in 2001, as a component of the Effective Child and Family (ECF) program [*Toimiva lapsi* & *perhe -työ],* a promotive and preventative approach to child wellbeing which included a suite of tools as documented in **Table 1** (12, 20, 21). A large ongoing government-supported nation-wide initiative, the ECF program included training, implementation and research. It aimed to equip health and social services to meet the minimum standards of the Finnish Child Welfare Act to address dependent children's need for care and support (12, 20, 22).

LTC served as a control group intervention to a more resource intensive preventative family intervention, 'Family Talk Intervention' (FTI) in the ECF program (18, 22). LTC was created to fit a health system with limited capacity to provide intensive family treatment for all consumers who were parents (12). So as to be used in adult-focused services, LTC was designed to be delivered by professionals with no experience or training in child development and assessment in the course of their ordinary work (12).

The purpose of LTC is to promote family mental wellbeing while also mitigating and/or preventing mental health issues for both parents and children (12). LTC takes an ecological understanding of child development, resilience and wellbeing that sees the child in the context of their relationships with their environment (23). Central to LTC is engaging parents in the support of their children. It works from the assumption that families are key resources for supporting child wellbeing and that everyday interactions are the stage on which child development plays out (22). Along with research and clinical experience, LTC's development was informed by international interventions for families where a parent has a mental illness including a Dutch mini-intervention and the US-originated FTI (12).

LTC is described as a "low threshold public health intervention" (23) because it is brief, low resource-intensive and has been applied in different settings and with different populations (12, 24). It has been translated, adapted and utilized across a range of countries and cultures including Estonia, Norway Sweden, Greece (25), Japan (26), Australia (27–29) and the USA (30, 31).

Drawing together the evidence for a practice developed *in-situ* can pose unique complications. As it is adapted and developed to fit the practice settings and the population, the way it is described in the literature can vary and its focus audience differ. As a consequence, a clear understanding of the evidence-base can be challenging.

Using LTC as an example, this paper showcases an evidence-based practice developed *in-situ* in order to guide future innovation. The study collates the diverse literature on LTC, to identify its forms and outcomes, and explore the implementation and sustainability impact of developing evidence-base practice *in-situ*. This study used the following questions:

- What was the context of the study (country, population, study type)?
- How was LTC described?
- Was LTC studied alone or with other interventions?
- What outcomes, implementation and sustainability impacts were documented?

METHODS

An integrative review method which permits reviewing qualitative and quantitative literature was used to consolidate what was known about LTC based on the research questions (32).

Search Strategy and Eligibility Criteria

Key literature on LTC published from 2001-July 2021 was sourced through six health and social databases (Medline, APA PsychArticles, PsychInfo, Embase & Embase plus, Emcare, CINAHL, Scopus) using the search terms of Let* Talk about the Children and Let* Talk about Children. Additional peer-reviewed and gray literature was found through "snowballing techniques" (33) and direct contact with developers and implementers. Given the limited articles published, no exclusion criteria were applied except being published in English and that it met the criteria of documenting outcomes for LTC.

Screening, Selection, and Data Extraction

A total of 149 records were identified via the database search with an additional 7 records via snowballing. After duplicates were removed, 89 records were screened at title and abstract removing

TABLE 1 | Effective child and family program's suite of tools.

ECF suite of tools[a]	Purpose	Details
Let's Talk about Children Discussion (LT-D)	Map child's life & develop an action plan to promote child's wellbeing	2–3 structured conversations between parent & practitioner. These include an invitation, and two structured conversations using an age-appropriate log and providing parents with the guidebooks (13)
Let's Talk about Children Network meeting (LT-N) also known as Effective Family Network meeting (EFN)	Build a network around the child & family	Parent & practitioner identify people to help facilitate wellbeing of the child i.e., family's own network of supports & services such as child psychiatry, school, housing (14).
Information booklets for parents & young people (12).	Self-guided psychoeducational material	How can I help my children? A guidebook for parents with mental health problems or issues (15) How can I care for my children? A guidebook for parents struggling with drug or alcohol use (16) What's up with our parents? A guidebook for young people whose parents have a mental health problem (17)
Family Talk Intervention (FTI) also known as The Effective Child & Family Intervention (ECFI)/Beardslee Family Intervention, Family Intervention, Preventive Family Intervention (PFI) or Let's Talk Family intervention	Facilitated family conversations by practitioner	A 6–8 session practitioner-led intervention that facilitates conversations between parents and children about the impact of the mental illness on family life (18)
Vertti child and parent group activities[b]	Peer support group program	A 10-week parallel peer support psychoeducation group for children and their parents (19)

[a] *information and links to training can be found at https://mieli.fi/en/development-projects/effective-child-and-family-work.*
[b] *ECF training does not include training in this program.*

an additional 31 records. The remaining records' full text were then assessed for documenting outcomes for LTC, resulting in 26 records included in review (see **Figure 1**).

Analysis

Data was extracted by the primary author and entered into a matrix according to review questions. Constant comparison was used to group extracted data into systematic categories to enable data synthesis through the identification of patterns and themes consistent with integrative review methods (32).

FINDINGS

Records

The study identified 26 records documenting outcomes for LTC; five of randomized control trials (RCT), three Quantitative papers, five Qualitative papers, seven of mixed method studies and six descriptive and commentary records (see **Supplementary Materials 1, 2**). The records highlight that the emerging evidence-base of LTC is derived from a set of discrete research endeavors in diverse settings beginning in Finland and now including Greece, Japan, Australia and USA. The variety of settings included adult mental health settings both clinical and Non-government, general hospital psychiatry, child and family services and universal settings (12, 20, 21, 25, 27, 34–36). The range of populations studied included families where a parent has depression, bipolar disorder, life threatening cancer, schizophrenia, schizoaffective disorder, borderline personality disorder, anxiety, Post-traumatic stress disorder, gambling and other co-occurring issues. The early studies were of the version of LTC developed for the RCT (LT-1) and later have been on the manualised intervention of two or three sessions (LT-D) designed for either treatment or universal settings. In some studies, LTC has been included as part of a suite of interventions (12, 20, 25, 35, 37). RCT's on LTC have compared it to a more intensive family intervention (20, 22, 25, 37) as well as to usual practice (36).

The research endeavors in regards to LTC have included a focus on its effectiveness, safety and acceptability in its different forms, in different settings and in different populations (20, 25–27, 38). There is also documentation of its implementation (12, 21, 29, 30, 34, 39) and on its sustainability (21, 40–42).

Different Forms of LTC

The records document LTC as evolving to fit its context. While the controlled adaptations to LTC resulted in changes in the way it was described in the literature, its different forms are recognized as developments of the same intervention as noted in **Table 2**.

Initially, LTC (LT-1) was described as a conversation with parents about their children, and included the provision of guidebooks (15–17) and development of an action plan to address the strengths and vulnerabilities identified in the discussion (12, 20). LTC Network meeting (LT-N), was developed to further address the strengths and vulnerabilities through linking the child and family to support (12, 14, 21). While the LT-N was recommended, at first it was not officially part of LTC.

After the data collection for the ECF RCT ended in 2006, LTC was described as a series of structured conversations including an introduction invitation and a set of two discussions which used a structured log and provided parents with guidebooks (LT-D) (13, 21, 26). The structured log was developed at the request of adult mental health practitioners who, with no training in child mental health, needed more detailed support for conversations about children's strengths, vulnerability and need for further support. Systematically mapping the child's life,

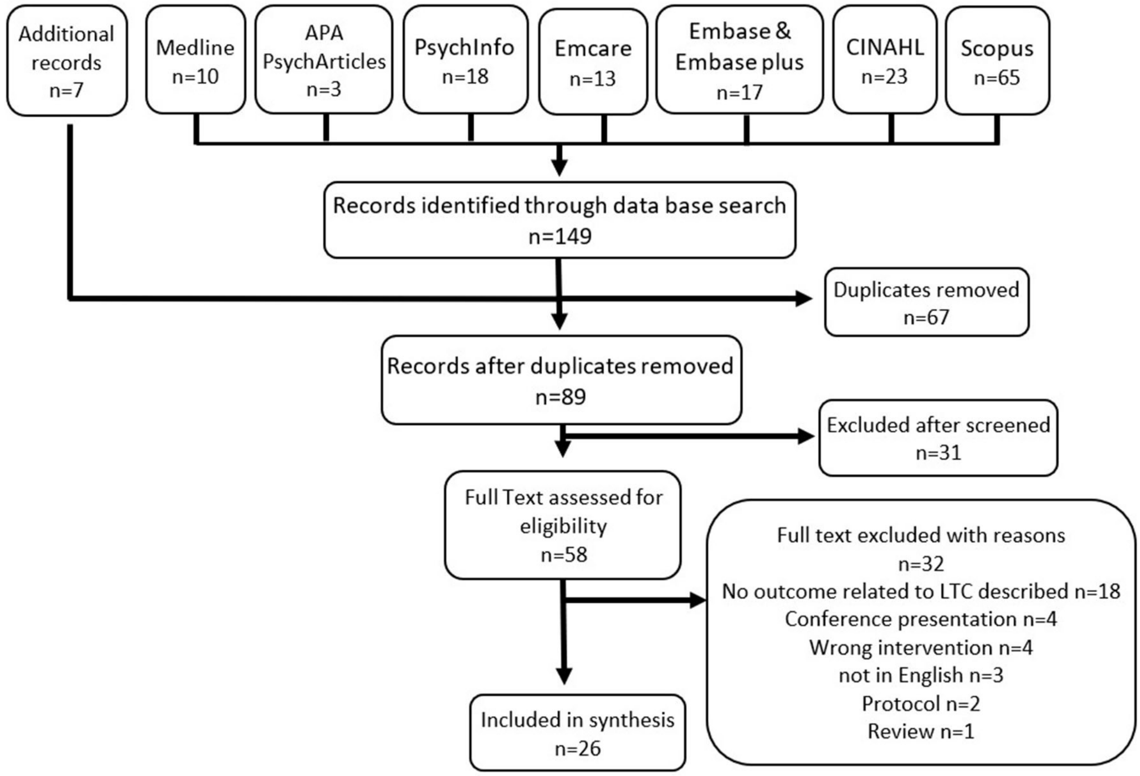


FIGURE 1 | Search and screening.

it provided a comprehensive picture of the child and family's life and wellbeing.

Subsequently, LTC was adapted to a Finnish public health intervention delivered to the general population without an underlying risk or problem. The motto being "Every child is worth a discussion" (mieli.fi/letstalk). This incorporated a whole-of-region approach with education settings and services working together as part of the national strategy (35, 44). New versions of the log were developed to facilitate the parent, teacher and child (as appropriate) to jointly map the child's life with the aim of creating concrete support for the everyday life of the child also at school and in daycare. LT-N was incorporated into LTC making it a two-step intervention with LT-D, with municipalities made responsible to organize relevant services and support people to come together for the network meeting (35). This LTC approach, called the Let's Talk about Children Service Model (LT-SM), facilitates systematic promotion of child wellbeing and development in universal settings (35).

Evidence Base

Summary of Evidence Base

The first RCT on LTC, carried out in Finland, was based on LT-1 (20–22, 37), with the rest of the studies using the structured LT-D with or without LT-N. LT-N was used in the first RCT but was not officially part of LTC, while in the Greek RCT it was.

As the outcomes of LTC's different versions are in agreement, the following documentation does not differentiate by version. LTC has been found to be acceptable for parents (20, 26, 27) and for mental health practitioners (13, 25, 29, 38). The records make connections between parent, family and child wellbeing outcomes, which are explored in detail below. Additionally, implementation and sustainability outcomes and impacts have been documented both in focused studies (21, 40–42) and from the context of other studies.

Parent and Family Outcomes

Studies on LTC document improvements in parents' mental health and wellbeing, in their parenting skills and confidence, and in their relationship with their children. Mental health and wellbeing improvements included decreased anxiety and depression (20, 25–27, 36), increased motivation for mental health treatment (20, 26), improvements in their own social support (25) and a greater future orientation with increased confidence in the child's and family's future (20, 26).

Shifts in parenting included improved confidence in parenting and greater self-acceptance (20, 22, 26) with more parenting ideas (20) and a decrease in parenting stress (27). Parents also reported less guilt, shame and worries about their children (20, 22, 26). Improved parent-child connection was documented through an increased understanding of their children (20, 25). The family outcomes are in line with the parent outcomes, with improved

TABLE 2 | Descriptions of the versions of LTC.

Let's Talk about Children Versions	Details
Let's Talk about Children Discussion-One (LT-1)	Early version of LTC used in the RCT with a conversation guide but without the structured log. Documented as conversations with parents about their children and providing parents with the guidebooks taking between one 15 min or two 45 min sessions. All practitioners, however, used more than 15 min (20) with 75% using one full session and 24% using two sessions (22).
Let's Talk about Children Discussion (LT-D)	Structured version of LTC using a series of 2–3 structured conversations including an introduction invitation and set of two discussions (LT-D) and providing parents with the guidebooks (13, 21, 23, 26). Discussion 1 uses an age-appropriate structured log to assist the parent to map the strengths and vulnerabilities within the everyday encounters and routines in the child's life (23). Discussion 2 builds on the previous discussion exploring how the parents can promote the child's wellbeing through building resilience in the systems around the child. Utilized in two different settings: 1. Child development & education. Early childhood, primary schools & high schools each have own log. 2. Service settings including both in treatment or care settings (i.e., psychiatric services, palliative care units, consultation psychiatry, child protection) and in promotive settings (i.e., maternal child health, community health). Six age-appropriate logs.
Let's Talk about Children Network meeting (LT-N) also known as Effective Family Network meeting (EFN)	An extension to LT-1 and LT-D that facilitates linking the child and family to support by building a network around the child. Used after LT-1 and LT-D as required, the parent identifies people including the family's own network of supports, schools, as well as services such as child psychiatry, housing etc. that may be able to help facilitate the wellbeing of the child (12, 14, 43). This became the second step of the two-step model of LTC.
Let's Talk about Children Service Model (LT-SM)	Use of LTC for collective impact through connecting systems across whole regions. Regional implementation strategy starts with community engagement and includes establishing a regional senior management group to enable service coordination and collaboration, as well as local management groups to oversee local implementation (35). Includes the two-step model of LTC: the parent and worker first use LT-D to chart the child's everyday life and develop an action plan to enhance strengths and support vulnerability. If a second step is needed, the LT-N is used to broaden the network of support for the child and family (35).

family connection and communication, confidence to talk about mental illness (20, 27), leading to mutual understanding in the family (20, 25, 27).

Child Outcomes

LTC studies document a decrease in negative outcomes and an increase in promotive factors for children. Improvements included a decrease in child depression (25), anxiety and behavioral problems (22, 25). Increases were documented for the prosocial behavior needed to solve interpersonal conflicts and promote relationships (22, 25), as well as their subjectively perceived social support and health-related quality of life (25). An increase was also seen in children's positive and functional thinking, which was associated with improvement in children's symptoms of depression and anxiety (37). In two studies, these outcomes were seen later than the parent and family improvements, at 10–18 months after LTC was delivered (22, 37).

Child outcomes were significantly associated with improvements in family functioning (25). Child prosocial behavior, emotional/behavioral problems, anxiety and health related quality of life were also associated with improvements in parenting and the parent's social support (22, 25). The linking of child outcomes to shifts in family processes highlighted LTC as a preventative and promotive intervention for child mental health for families where parental anxiety and depression is present (22, 25, 37). The study by Niemelä et al. (35) documents a significant reduction in children being referred to child protection in the region in which LT-SM was implemented.

Implementation and Sustainability Impacts

The records document different contexts impacting implementation and sustainability of LTC. These include diverse approaches to building workforce capacity, adaptations made or required, and commonalities of organizational capacity to support practice.

LTC was developed and implemented to meet a need to develop adult mental health practice that satisfied Finnish minimum standards in welfare and health care legislation (12, 20, 21). Consequently, its implementation in Finland was embedded within a broader government initiative to incorporate promotive and preventative approaches to child wellbeing. LTC was documented as being feasible for use in Finnish general psychiatry and adult mental health settings (13, 20). The ECF approaches, including LTC, were recorded as being in use in two thirds of the health districts in Finland after 5 years and endorsed in national recommendations for Finnish health services (12, 13, 36).

Records note implementation outside Finland as piloting or trialing a cost-effective evidence-based practice and/or testing its applicability to the different cultures or populations (26, 27). In Australia, implementation is documented as part of localized pilots and trials of LTC in adult focused mental health settings in the context of national government supported online training and resource development (38, 45). Time constraints, high caseloads and tension between child protection concerns and the therapeutic relationship are noted as challenges for the fit of family-focused practice including LTC, in clinical mental health services (29, 38, 39). In Greece, implementation was part of a multiphase government-funded 3-year child mental health promotion program that first tested the ECF's fit to the context within the RCT, finding it feasible for use and family culture in Greece. It was then scaled up to 90 mental health services where 529 families received an intervention and a majority of practitioners chose LTC (25). Implementation in Japan is recorded as testing LTC's fit to context, finding it safe and feasible to be used in parents with mood disorders in Japanese culture (26). In the USA, implementation was in the context of a statewide initiative incorporating a research-service collaboration to adapt LTC to their service delivery context (30). The brevity of LTC was identified as promoting its ability to build to scale in public health in Finland (13, 20, 21, 36) and in Greece (25).

To enable LTC to fit these cultures and populations, self-help booklets and the log were translated (25, 26, 38, 45), and handouts were tailored for different settings (cancer, gambling) to guide discussions with children (13, 34). The adapted material was documented as acceptable to parents in Australia and Japan (26, 45).

Different approaches were used for building workforce capacity to deliver LTC. Where implementation was embedded within broader shifts, such as in Finland, building workforce capacity included an initial broad awareness-raising process prior to the method training. Regional campaigns aimed at the public and professionals in health, social services and education sectors, ran open seminars and media coverage. These built awareness on family and child experiences of parental mental disorder, the importance of prevention and promotion in child and family mental health and the basics of preventive interventions (12). The subsequent method training in the new approaches included training and supporting master trainers from within organizations and the provision of practice supervision (12, 13, 21). Training infrastructure for LTC's sustainability in Finland is presumed from documentation of master trainers training others, a pool of trainers, large numbers of trained practitioners (12, 21, 35) and its use in routine practice with families affected by parental cancer (13, 36).

Where implementation was piloting or trialing a cost-effective evidence-based practice, such as Australia and Japan, specific LTC method training is documented as the focus of workforce capacity building. In Australia, this took a variety of approaches; a train the trainer model (27, 46), online training modules only (38) or online with face-to-face training (29). The studies of the latter two, identified a need for support to apply the training to practice and suggested incorporating opportunity for practice into training, observing others' use LTC and Post-training follow-up (29, 38).

In the USA, where implementation was embedded within an adaptation process, a comprehensive change process using a learning collaborative was documented that incorporated in-person training, virtual hubs, coaching and debriefing (30). While costly, this approach was noted to have multifaceted impacts to support implementation (30).

Overall, the training of LTC was identified as effective for increasing practitioners' skill and knowledge about the impact of mental illness on parents, children and families (21, 30, 38, 46) and on supporting families (21, 29, 38). Change in practice is noted with parents reporting having ongoing discussions about family and children after delivery of LT-1 (20) and improvements in practitioners' ability to gauge a parent's understanding of their children, reflect on the impacts on children and work together with the parent to address impacts and provide resources/referrals where necessary (29, 36, 38). Practitioners indicated that using the practice increased their enjoyment and motivation at work (21). All Finnish trained practitioners were noted as using the suite of ECF interventions, implying the use of LTC (21). Documentation specifically of practitioner's delivery of LTC is only documented in one Australian record which noted over half offering and less than half delivering it (41). Adaptations to delivery were also documented, with practitioners delivering it in less sessions or without the structured log to enable LTC to fit everyday practice (29, 41). Such adaptations were not accompanied with monitoring parent, child and family outcomes and core mechanisms of change for LTC are not clearly articulated.

Common organizational capacities important for implementing and sustaining LTC are identified in the records. Organizational ownership with multiple levels of leadership and internal implementers is noted as vital for sustainability (35, 39, 40, 42). Senior leadership in particular was identified as giving or needed to give authority and vision (34, 35, 40, 42) while other leadership was important for integrating into practitioners' everyday work (39, 42). Infrastructure identified as supporting implementation and LTC's continued use included training structures, data collection and feedback systems on parent numbers, training gaps and practitioners use, and integration into committee structures and policy (40, 42). Municipal cross-sector collaboration with multilevel implementation support and regular data collection was seen as important for LT-SM sustained use (35).

DISCUSSION

We studied the consolidated evidence of LTC as an example of an evidence-based intervention developed *in-situ*, focusing on parent and family, child and implementation and sustainability outcomes. It demonstrates the relevance of this approach to developing evidence-based interventions.

Development *in-situ* means that intervention development is influenced by context and knowledge from evolving experience over time (9). This contrasts to evidence-based practices developed via the pipeline approach which are often understood as finished by the time of implementing into practice settings and universally applicable via an implementation process (7). While adaptations can be seen as threats to fidelity and the lack of sustained practice as an implementation issue, development *in-situ* allows the practice context to influence the shaping of the intervention (47).

Our study demonstrates how intervention development *in-situ* enabled a rapid response to an acknowledged problem, rather than waiting for a fully-developed intervention with a research base. LTC's initial practice was able to immediately address a known need while continuing to evolve, based on knowledge of the practice setting and the needs and experiences of practitioners and family members.

The current review of LTC highlights that developing an intervention to fit the setting while simultaneously developing its evidence-base may also be advantageous for building interventions that can be sustained in real world settings. The alignment between the setting and LTC, vital for sustainability (8), can be seen in this study with increased structure built within the intervention and organizational support as it evolved. Building interventions *in-situ* brings the work on effectiveness and implementation together. Under these circumstances, it is less likely that an intervention that does not fit the practice setting could be deemed effective and the suitability of an intervention is measured in the light of adjustments made within the organization. The documentation of the spread and sustainability of LTC within Finland (12, 21, 35, 36) suggests this approach is a useful pathway to evidence-based practices that fit settings and can be sustained.

A challenge for developing evidence-based interventions *in-situ*, however, is the ability to consolidate the evidence base and draw together a clear understanding of the practice. As seen in this study, the intervention's description shifted as it was adapted to context, culture and population, and outcomes were published in different fields over many years. The three forms of LTC identified in this review are thus consistent with a practice shaped by the setting, with an evolving body of evidence. Regardless of LTC version, the studies document similar outcomes for children, parents and families. The referenced studies in this review, however, lack detailed descriptions of LTC's adjustments and analysis for subsets of families, limiting clear understanding for who it is and is not a good fit for. This remains a task for future studies. Collectively, the evidence also draws attention to possible core mechanism of change for LTC. Having these clearly articulated could promote adaptations that result in the same expected outcomes and provide guidance for its implementation and evaluation.

The example of LTC invites different ways to consider evidence-based practice. Rather than the action of an intervention being defined and manualized, the evidence-base for the core mechanisms of change could be clearly articulated, to enable practices to be fitted to settings. The focus shifts then from fidelity of a manualized intervention, to measuring how the core mechanisms are enacted within practice. As seen for LTC, it is uncommon, however, for these core mechanisms of evidence-based practices to be articulated and have measures identified (48), or for a practice logic or underlying theory to be incorporated into manuals. These will be important to enable consistent evaluation that can build a body of evidence as it is adapted.

CONCLUSION

This study set out to showcase an evidence-based practice developed *in-situ* through an integrative review of literature on LTC. In consolidating the evidence, the review clarified how the three forms of LTC reported in the literature, document similar outcomes for children, parents and families, and provide a window into its spread and sustainability. The results suggest that intervention development influenced by the practice context provides benefits for implementation and does not compromise the evidence-base. Intervention development *in-situ* is a relevant developmental pathway for evidence-based practices. Clear articulation of the core mechanisms of change is important for consistent evaluation and reporting the adjustments made in the intervention in different settings will support future *in-situ* evidence-based practice development.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Materials**, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

BA designed and led the data collection and early analysis in consultation with TS. Both authors analyzed the data, contributed and edited the manuscript, and approved the final manuscript.

ACKNOWLEDGMENTS

This paper builds on and updates a review of the literature on LTC carried out during research supported by an Australian Government Research Training Program Scholarship undertaken by BA for a doctorate of philosophy supervised by Dr. Melinda Goodyear, Dr. Brendan O'Hanlon and Prof. Bente M. Weimand.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.824241/full#supplementary-material

REFERENCES

1. Guyatt G, Cairns J, Churchill D, Cook D, Haynes B, Hirsh J, et al. Evidence-based medicine: a new approach to teaching the practice of medicine. *JAMA*. (1992) 268:2420–5. doi: 10.1001/jama.268.17.2420
2. Brown CH, Curran G, Palinkas LA, Aarons GA, Wells KB, Jones L, et al. An overview of research and evaluation designs for dissemination and implementation. *Annu Rev Public Health*. (2017) 38:1–22. doi: 10.1146/annurev-publhealth-031816-044215

3. Bauer MS, Kirchner J. Implementation science: What is it and why should I care? *Psychiatry Res*. (2020) 283:112376. doi: 10.1016/j.psychres.2019.04.025
4. Proctor EK, Landsverk J, Aarons GA, Chambers D, Glisson C, Mittman B. Implementation research in mental health services: an emerging science with conceptual, methodological, and training challenges. *Adm Policy Ment Health Serv Res*. (2009) 36:24–34. doi: 10.1007/s10488-008-0197-4
5. Rubin A. Improving the teaching of evidence-based practice: introduction to the special issue. *Res Soc Work Pract*. (2007) 17:541–7. doi: 10.1177/1049731507300145
6. Nevo I, Slonim-Nevo V. The myth of evidence-based practice: towards evidence-informed practice. *Br J Soc Work*. (2011) 41:1176–97. doi: 10.1093/bjsw/bcq149
7. Hawe P. Lessons from complex interventions to improve health. *Annu Rev Public Health*. (2015) 36:307–23. doi: 10.1146/annurev-publhealth-031912-114421
8. Scheirer MA, Dearing JW. An agenda for research on the sustainability of public health programs. *Am J Public Health*. (2011) 101:2059–67. doi: 10.2105/AJPH.2011.300193
9. Chambers DA, Glasgow RE, Stange KC. The dynamic sustainability framework: addressing the paradox of sustainment amid ongoing change. *Implement Sci*. (2013) 8:117. doi: 10.1186/1748-5908-8-117
10. Chambers DA, Norton WE. The adaptome: advancing the science of intervention adaptation. *Am J Prev Med*. (2016) 51(4, Supplement 2):S124–31. doi: 10.1016/j.amepre.2016.05.011
11. Scheirer MA. Is sustainability possible? A review and commentary on empirical studies of program sustainability. *Am J Eval*. (2005) 26:320–47. doi: 10.1177/1098214005278752
12. Solantaus T, Toikka S. The effective family programme: preventative services for the children of mentally ill parents in Finland. *Int J Ment Health Promot*. (2006) 8:37–44. doi: 10.1080/14623730.2006.9721744
13. Niemelä M, Väisänen L, Marshall C, Hakko H, Räsänen S. The Experiences of mental health professionals using structured family-centered interventions to support children of cancer patients. *Cancer Nurs*. (2010) 33:E18–27. doi: 10.1097/NCC.0b013e3181ddfcb5
14. Väisänen L, Niemelä M. Vanhemman mielenterveyden häiriö ja lapset. Lapsikeskeinen näkökulma psykiatrisessa sairaalassa - Osa 2. [Parental mental disorder and childen - a child-centred standpoint in a psychiatric hospital]. *Suomen Lääkärilehti*. [The Finnish Medical Journal] (2005) 39:3889–93.
15. Solantaus T. *How Can I Help My Children? A Guide to Parents Who Have Mental Health Problems*. Mieli: The Finnish Association for Mental Health (2002).
16. Solantaus T. *How Can I Care For My Children? A Handbook for Parents Struggling With Drug or Alcohol Use*. Mieli: The Finnish Association for Mental Health (2010).
17. Solantaus T. *What's up With My Parents? A Handbook for Older Children and Adolscents Whose Mother or Father Have Mental Health Problems*. Mieli: The Finnish Association for Mental Health (2005).
18. Beardslee WR, Salt P, Porterfield K, Rothberg PC, Van De Velde P, Swatling S, et al. Comparison of preventive interventions for families with parental affective disorder. *J Am Acad Child Adolesc Psychiatry*. (1993) 32:254–63. doi: 10.1097/00004583-199303000-00004
19. Söderblom B, Inkinen M. *Vertti Käsikirja Ryhmäohjaajille: Uudistettu painos 2018* [Vertti handbook for group leaders; Revised Edition 2018], profami oy (2018).
20. Solantaus T, Toikka S, Alasuutari M, Beardslee WR, Paavonen EJ. Safety, feasibility and family experiences of preventive interventions for children and families with parental depression. *Int J Ment Health Promot*. (2009) 11:15–24. doi: 10.1080/14623730.2009.9721796
21. Toikka S, Solantaus T. The effective family programme II: clinicians' experiences of training in promotive and preventative child mental health methods. *Int J Ment Health Promot*. (2006) 8:4–10. doi: 10.1080/14623730.2006.9721746
22. Solantaus T, Paavonen JE, Toikka S, Punamäki R-L. Preventive interventions in families with parental depression: children's psychosocial symptoms and prosocial behaviour. *Eur Child Adolesc Psychiatry*. (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
23. Solantaus T, Reupert AE, Maybery DJ. Working with parents who have a psychiatric disorder. In: Reupert AE, Maybery DJ, Nicholson J, Gopfert M, Seeman MV, editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. 3rd ed. New York, NY: Cambridge University Press (2015). p. 238–47. doi: 10.1017/CBO9781107707559.023
24. Beardslee WR, Solantaus T, Morgan BS, Gladstone TR, Kowalenko NM. Preventive interventions for children of parents with depression: international perspectives. *Med J Aust*. (2012) 199(3 Suppl):S23–5. doi: 10.5694/mja11.11289
25. Giannakopoulos G, Solantaus T, Tzavara C, Kolaitis G. Mental health promotion and prevention interventions in families with parental depression: a randomized controlled trial. *J Affect Disord*. (2021) 278:114–21. doi: 10.1016/j.jad.2020.09.070
26. Ueno R, Osada H, Solantaus T, Murakoshi A, Inoue T. Safety, feasibility, fidelity, and perceived benefits of an intervention for parents with mood disorders and their children — "Let's Talk About Children" in Japan. *J Fam Psychother*. (2019) 30:272–91. doi: 10.1080/08975353.2019.1678092
27. Maybery DJ, Goodyear MJ, Reupert AE, Sheen J, Cann W, O'Hanlon B, et al. A mixed method evaluation of an intervention for parents with mental illness. *Clin Child Psychol Psychiatry*. (2019) 24:717–27. doi: 10.1177/1359104518822676
28. Maybery DJ, Goodyear MJ, Reupert AE, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry*. (2017) 17:198. doi: 10.1186/s12888-017-1357-4
29. Karibi H, Arblaster K. Clinician experiences of "Let's Talk about Children" training and implementation to support families affected by parental mental illness. *J Ment Health Train Educ Pract*. (2019) 14:201–11. doi: 10.1108/JMHTEP-08-2018-0044
30. Nicholson J, English K, Heyman M. The parenting well learning collaborative feasibility study: Training adult mental health service practitioners in a family-focused practice approach. *Commun Ment Health J*. (2022) 58:261–76. doi: 10.1007/s10597-021-00818-5
31. Nicholson J, English K. *Parenting Well Practice Profile*. National Research Center for Parents with Disabilities, Brandeis University and the Massachusetts Department of Mental Health, United States. (2019). Available online at: https://heller.brandeis.edu/parents-with-disabilities/work/parenting-well.html (accessed February 01, 2020).
32. Whittemore R, Knafl K. The integrative review: updated methodology. *J Adv Nurs*. (2005) 52:546–53. doi: 10.1111/j.1365-2648.2005.03621.x
33. Greenhalgh T, Peacock R. Effectiveness and efficiency of search methods in systematic reviews of complex evidence: audit of primary sources. *BMJ*. (2005) 331:1064–5. doi: 10.1136/bmj.38636.593461.68
34. von Doussa H, Sundbery J, Cuff R, Jones S, Goodyear M. 'Let's Talk About Children': investigating the use of a family-focused intervention in the gambling support services sector. *Aust N Z J Fam Ther*. (2017) 38:482–95. doi: 10.1002/anzf.1233
35. Niemelä M, Kallunki H, Jokinen J, Räsänen S, Ala-Aho B, Hakko H, et al. Collective impact on prevention: Let's talk about children service model and decrease in referrals to child protection services. *Front Psychiatry*. (2019) 10:64. doi: 10.3389/fpsyt.2019.00064
36. Niemel M, Repo J, Wahlberg KE, Hakko H, Räsänen S. Pilot evaluation of the impact of structured child-centered interventions on psychiatric symptom profile of parents with serious somatic illness: struggle for life trial. *J Psychosoc Oncol*. (2012) 30:316–30. doi: 10.1080/07347332.2012.664258
37. Punamäki R-L, Paavonen J, Toikka S, Solantaus T. Effectiveness of preventive family intervention in improving cognitive attributions among children of depressed parents: a randomized study. *J Fam Psychol*. (2013) 27:683–90. doi: 10.1037/a0033466
38. Tchernegovski P, Reupert AE, Maybery DJ. "Let's Talk about Children": a pilot evaluation of an e-learning resource for mental health clinicians. *Clin Psychol*. (2015) 19:49–58. doi: 10.1111/cp.12050
39. Allchin B, Goodyear MJ, O'Hanlon B, Weimand BM. Leadership perspectives on key elements influencing implementing a family-focused intervention in mental health services. *J Psychiatr Ment Health Nurs*. (2020) 27:616–27. doi: 10.1111/jpm.12615
40. Allchin B, O'Hanlon B, Weimand BM, Boyer F, Cripps G, Gill L, et al. An explanatory model of factors enabling sustainability of let's talk in an adult mental health service: a participatory case study. *Int J Ment Health Syst*. (2020) 14:48. doi: 10.1186/s13033-020-00380-9

41. Allchin B, O'Hanlon B, Weimand BM, Goodyear MJ. Practitioners' application of Let's Talk about children intervention in adult mental health services. *Int J Ment Health Nurs*. (2020) 29:899–907. doi: 10.1111/inm.12724
42. Allchin B, Weimand BM, O'Hanlon B, Goodyear MJ. Continued capacity: factors of importance for organizations to support continued Let's Talk practice – a mixed-methods study. *Int J Ment Health Nurs*. (2020) 29:1131–43. doi: 10.1111/inm.12754
43. Solantaus T. Commentary: 'Let's talk about children': Investigating the use of a family focused intervention in the gambling support services sector. *Aust N Z J Fam Ther*. (2017) 38:496–7. doi: 10.1002/anzf.1239
44. Solantaus T, Niemelä M. Arki kantaa – jos se pannaan kantamaan. *Perheterapia [Family Therapy]*. (2016). Available online at: https://mieli.fi/sites/default/files/inline/arki_kantamaan_solantaus_niemela_2016.pdf (accessed November 20, 2018).
45. Cooper V, Reupert AE. "Let's Talk About Children" resource: a parallel mixed method evaluation. *Soc Work Ment Health*. (2017) 15:47–65. doi: 10.1080/15332985.2016.1170090
46. The Bouverie Centre. *Mental Health Beacon: Implementing family inclusive practices in Victorian Mental Health Services*. The Bouverie Centre (2015).
47. Dearing JW. Evolution of diffusion and dissemination theory. *J Public Health Manag Pract*. (2008) 14:99–108. doi: 10.1097/01.PHH.0000311886.98627.b7
48. Kirk MA, Haines ER, Rokoske FS, Powell BJ, Weinberger M, Hanson LC, et al. A case study of a theory-based method for identifying and reporting core functions and forms of evidence-based interventions. *Transl Behav Med*. (2021) 11:21–33. doi: 10.1093/tbm/ibz178

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 17 February 2022
doi: 10.3389/fpsyt.2022.782939

Association of Paternal Workplace and Community Social Capital With Paternal Postnatal Depression and Anxiety: A Prospective Study

Satomi Doi [1,2]*, *Aya Isumi* [1,2] *and Takeo Fujiwara* [1]*

[1] *Department of Global Health Promotion, Tokyo Medical and Dental University (TMDU), Tokyo, Japan,* [2] *Japan Society for the Promotion of Science, Tokyo, Japan*

Objective: The aim of this study is to examine the association between workplace and community social capital and fathers' postpartum depression and anxiety at 3 months after delivery in Japan.

Methods: Fathers who had babies delivered in two obstetric clinics in Tokyo, Japan were recruited to take part in the study (response rate = 76.2%). Participants completed questionnaires measuring workplace and community social capital, depression, and anxiety at 1 week and a follow-up at 3 months post-delivery (N = 398). Multiple linear regression analyses were performed with multiple imputation for missing data (at most, N = 60, 15.1%).

Results: Community social capital was inversely associated with both depressive symptoms (β = −0.21, 95%CI = −0.33 to −0.08) and anxiety (β = −0.38, 95%CI = −0.66 to −0.11) at 3 months, after adjusting for covariates. No association was found between workplace social capital and depressive symptoms and anxiety.

Conclusion: Paternal community social capital, but not workplace social capital, was shown to be a preventive factor for paternal depression and anxiety up to 3 months post-delivery. To prevent paternal mental health problems during the postpartum period, an intervention to promote paternal community, rather than workplace, social capital may be warranted.

Keywords: father, community social capital, workplace social capital, depression, anxiety, postnatal mental health

Edited by:
Joanne Nicholson,
Brandeis University, United States

Reviewed by:
Ariane Göbel,
University of Hamburg, Germany
Dominik A. Moser,
Centre Hospitalier Universitaire
Vaudois (CHUV), Switzerland

Correspondence:
Satomi Doi
doi.hlth@tmd.ac.jp
Takeo Fujiwara
fujiwara.hlth@tmd.ac.jp

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *25 September 2021*
Accepted: *24 January 2022*
Published: *17 February 2022*

Citation:
Doi S, Isumi A and Fujiwara T (2022)
Association of Paternal Workplace and
Community Social Capital With
Paternal Postnatal Depression and
Anxiety: A Prospective Study.
Front. Psychiatry 13:782939.
doi: 10.3389/fpsyt.2022.782939

INTRODUCTION

Paternal mental health problems, such as depression and anxiety during the perinatal period, are common issues (1, 2) presenting a public health concern, in addition to maternal perinatal mental health. Previous studies estimated that 8.4% of fathers across North and South America, the United Kingdom, Asia, and Australia/Oceania (3, 4), and 8.8% of those in Japan (5) showed paternal depression within 1-year postnatal. Postnatal paternal depression is associated with poor satisfaction with the marital relationship (6, 7), increased maternal depression (8), poor father-infant interaction (9, 10), poor child development (10), and increased child maltreatment (5). Furthermore, a previous study found that paternal postnatal depression is associated with depression in offspring (only female) at 18 years of age, which was mediated by maternal depression (11).

In terms of paternal anxiety, systematic reviews found that from 2.4 to 51.0% of fathers show some anxiety disorders during the postnatal period (12, 13). Paternal anxiety, which may raise the risk of paternal depression (14), is associated with poor paternal parenting self-efficacy (15) and poor father-infant interaction (16). A previous study suggests that, in addition to treatment for depression, assessment and treatment of paternal anxiety are needed (13).

In order to prevent paternal postnatal depression and anxiety, preventive factors must be identified. In general, it is well-established that social capital plays an important role in preventing depression and anxiety (17–19). Social capital is defined as resources that are available *via* civic participation in voluntary organizations, norms of mutual aid and reciprocity, and a level of interpersonal trust (20). Though there is still debate regarding its precise definition, social capital can be categorized as individual level and contextual level, such as community, school, or workplace (21). Perceived social support can be considered as one aspect of individual level social capital (22, 23). For example, people with a high individual level of social capital are more likely to have access to psychosocial resources to cope with their mental distress, considered as perceived social support (24). Further, social capital induces structural aspects, such as belonging to a group (25), which improve mental health (26). Among mothers during the postnatal period, a higher level of social capital was associated with lower levels of depression (27–29). However, little is known about the association between social capital and paternal depression and anxiety during the postnatal period.

For fathers, not only community social capital but also workplace social capital may play an important role in preventing depression and anxiety because, compared to mothers, fathers tend to continue working. In this study, community social capital represents trust in neighbors and reciprocity in the neighborhood. The association of community social capital on mental health is well-established (30). Besides the neighborhood, the workplace is considered a major social organization, in which there is both formal and informal face-to-face communication and many sources of social capital (31). Especially in Japan, fathers are less likely to take paternity leave (32), although the rate of paternity leave in Japan has increased slowly (3.2% in 2016, 6.2% in 2018, and 12.7% in 2020) (33). Furthermore, it may be helpful to consider an intervention designed to promote paternal social capital to address community and workplace social capital independently.

In previous studies which examined the association between social capital and mental health problems, participants' socio-economic status—including education level, annual household income, and employment—and history of psychiatric disorders were adjusted for as confounders (34). Further, adverse childhood experience is an important confounder because it is associated with both social capital and mental health problems (35). Additionally, covariates regarding delivery such as paramipara, feelings when pregnancy was confirmed, and paternal childcare leave are adjusted for to examine the association among parents (36). As for possible mediators, paternal postpartum depression and anxiety at 1 week after delivery, maternal depressive symptoms and anxiety at 1 week after delivery, and the number of people who can be consulted about parenting were also adjusted for in our analyses. Thus, the aim of this study is to examine the association between workplace and community social capital and postpartum depression and anxiety among fathers in Japan, at 3 months after delivery, adjusted for possible covariates.

METHODS

Participants

We approached 548 couples who delivered their babies in two obstetrics hospitals in Tokyo, Japan. A total of 350 couples were approached from obstetric hospital A, which is a hospital for high-risk and emergency pregnancies, and 198 couples from hospital B, a general obstetric hospital. During their hospital stay (within 1 week after delivery), the couples completed and returned anonymous questionnaires after written informed consent was acquired. The participants who completed the questionnaire 1 week after delivery included 418 couples (response rate: 76.2%): 250 in hospital A (71.4%) and 168 in hospital B (84.8%). In this study, we excluded fathers who did not work and did not report exposure (i.e., workplace and community social capital) ($N = 20$); thus, the analytic sample totaled 398 couples. These couples then received follow-up questionnaires *via* mail 3 months after delivery, and 363 questionnaires of the analytical sample were returned completed (follow-up rate: 91.2%): 212 from hospital A (89.8%) and 151 from hospital B (93.2%) (**Figure 1**). As missing data at 3 months after delivery were imputed using multiple imputations, the analytical sample was 398 couples.

Measures

Community and workplace social capital were assessed via the father's questionnaire when the baby and the mother were discharged from the hospital (1 week after delivery). Community social capital included the following four questions, used in previous studies (37–39), rated on a scale of 1 to 5: (1) "Do you think that people in your neighborhood can be trusted?"; (2) "Do you think that people in your neighborhood are willing to help their neighbors?"; (3) "Do you think that people in your neighborhood trust each other?"; (4) "Do you think that people in your neighborhood help each other?" A higher total score, ranging from 4 to 20, denotes a higher level of community social capital. Cronbach's α in this study was 0.89. Workplace social capital was assessed using the Japanese version of the workplace social capital scale (40), which was developed from the original version (41). The scale has eight items (e.g., "Our supervisor treats us with kindness and consideration," "People feel understood and accepted by each other," and "We can trust our supervisor."), rated on a scale of 1 to 5. The mean score ranges from 1 to 5, where a higher means score denotes a higher level of workplace social capital. The Japanese version scale has good internal consistency and reliability (40). In this study, Cronbach's α was 0.92.

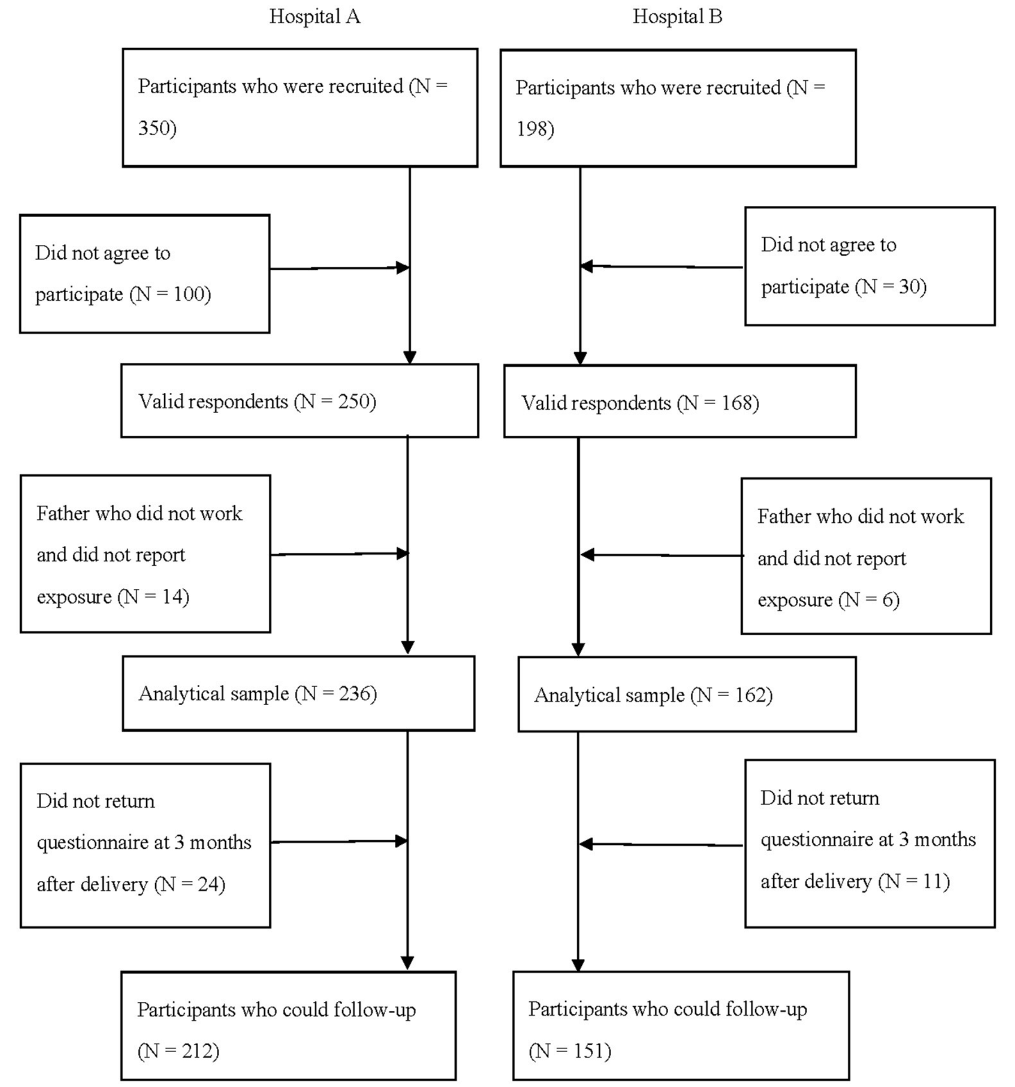


FIGURE 1 | Participant flow chart.

Paternal depression was assessed at 1 week and 3 months after delivery. We used the Japanese version of the Edinburgh Postnatal Depression Scale (EPDS) (42), which is a 10-item self-report questionnaire with a scale of 0 to 3. A higher total score, ranging from 0 to 30, denotes a higher level of depressive symptoms. In this study, the Cronbach's α of depression at 3 months after delivery was 0.80. Paternal anxiety was also assessed at 1 week and 3 months after delivery. We used the Japanese version of the State-Trait Anxiety Inventory (STAI) (43), which is a 40-item (20-item trait and 20-item state anxiety) self-report questionnaire with a scale of 1 ("not at all") to 4 ("very much so"). In this study, we used the score of state anxiety, where a higher total score, ranging from 20 to 80, denotes a higher level of state anxiety. In this study, the Cronbach's α of anxiety at 3 months after delivery was 0.92.

Regarding covariates, in addition to the obstetric hospital where mothers gave birth, paternal basic demographics, job, history of psychiatric disorders, adverse childhood experiences, history of delivery, paternal feelings when pregnancy was confirmed, maternal mental health, and the number of people who can be consulted about parenting, which were assessed within 1 week after delivery, were included. Fathers were asked about their age, education level ("junior high school," "high school," "technical college or junior college," "college education," "graduate college," or "unknown"), employment ("full-time job," "part-time job," "self-employed," homeworker," or "other"), paternal childcare leave ("yes," "planning to take," or "no")—assessed at 3 months after delivery—history of psychiatric disorders ("yes" or "no"), paternal adverse childhood experiences—including 8-items, such as parental loss, abuse, and neglect from parents—("yes" or "no"), and feelings when pregnancy was confirmed ("happy," "unexpected but happy," "unexpected and confused," "did not know what to do," "no feelings," or "other"). Mothers were asked about annual household income (JPY 2,000,000 or less, JPY 2,010,000–4,000,000, JPY 4,010,000–6,000,000, JPY 6,010,000–8,000,000, JPY 8,010,000–10,000,000, JPY 10,010,000–15,000,000, JPY 15,010,000 or more," or "unknown") and history of delivery ("primipara" or "multipara"). Normal pregnancy was assessed as whether the mother had any major complications during the pregnancy using one item: "Did you have a normal pregnancy?" ("yes" or "no"). Maternal depressive symptoms were assessed using the EPDS, and maternal anxiety was assessed using the STAI.

Ethics

This study was approved by the Institutional Review Board of the National Center for Child Health and Development (730) and Tokyo Medical and Dental University (M2017-078).

Statistical Analysis

We performed multiple linear regression analyses to examine the associations of community and workplace social capital within 1 week after delivery with paternal depressive symptoms (i.e., EPDS total score) and anxiety (i.e., STAI state total score) at 3 months after delivery. After estimating the crude model, Model 1 added paternal education, age, annual household income, employment, childcare leave, adverse childhood experiences, paternal history of psychiatric disorders, history of delivery, normal pregnancy, and paternal feelings when pregnancy was confirmed. In addition to the covariates in Model 1, Model 2 further added paternal depressive symptoms (only for depressive symptoms as the outcome) or anxiety (only for anxiety as the outcome) at 1 week after delivery and maternal depressive symptoms and anxiety at 1 week after delivery. Finally, Model 3 further added the number of people who can be consulted about parenting and both types of social capital into Model 2. We conducted the power calculation (power = 0.90, alpha = 0.01) to compute sample size because a high number of covariates were included in the final model. We found that a sample of at least 161 subjects is required in our analysis, indicating that our sample size is sufficient.

We performed multiple imputations (100 imputed datasets) based on Rubin's rule (44, 45) to address missing values. Missing data were found in outcomes including paternal EPDS and STAI scores at three months after delivery (N = 35, 8.8%), and in covariates including annual household income (N = 22, 5.5%), paternal childcare leave (N = 60, 15.1%), and the number of people who can be consulted about parenting (N = 28, 7.0%). There were no differences in characteristics, workplace social capital, community social capital, depressive symptoms, and anxiety at 1 week after delivery (data not shown). The following variables were used for the multiple imputation: paternal symptoms of attention-deficit/hyperactivity disorder (ADHD) assessed using the Japanese version of the 6-item Adult ADHD Self-Report Scale (ASRS-J-6) (46), autism traits assessed using the Japanese version of the Autism-Spectrum Quotient (AQ) (47), domestic violence from their partner, belonging to an organization, attendance at delivery, and health condition were measured via responses from the father's questionnaire. In addition, maternal adverse childhood experiences, feelings toward her baby (i.e., mother-infant bonding)—assessed using the Japanese version of Mother-to-Infant Bonding Scale (MIBS-J) (48)—type of housing, and child's sex were measured via the mother's questionnaire and used for multiple imputation. All analyses were conducted using STATA version 15.0 SE.

RESULTS

The distribution of characteristics among participants is shown in **Table 1**. Approximately 10% of fathers were <30 years old, 80% were college graduates or higher, 20% experienced some adverse childhood experiences, 90% had a full-time job, 10% took or planned to take childcare leave, 2% had a history of psychiatric disorders, and 80% felt happy when the pregnancy was confirmed. The percentage of low-income households (i.e., JPY 4,000,000 or less) was 9%. Furthermore, about 20% of fathers participated in group activities in the last year before delivery. On average, fathers had nine (SD = 8.1) people with whom they could consult about parenting.

Table 2 shows the distribution of paternal workplace and community social capital at 1 week after delivery, and EPDS and STAI scores at 1 week and 3 months after delivery. The paternal workplace and community social capital means were 3.70 (SD = 0.80) and 13.30 (SD = 2.82), respectively. The EPDS score means were 3.11 (SD = 3.17) at 1 week after delivery and 3.92 (SD = 3.77) at three months after delivery. The STAI score means were 34.12 (SD = 8.68) at 1 week after delivery and 34.49 (SD = 8.89) at 3 months after delivery. We also show the correlations among workplace social capital, community social capital, social participation in the last year before delivery, and number of people who can be consulted about parenting in **Supplementary Table 1**. A significant but weak correlation was found between workplace social capital and community social capital (r = 0.17, $p < 0.01$).

Tables 3, **4** shows the results of the linear regression analyses examining the association of social capital with depressive symptoms and anxiety in fathers. In terms of depressive symptoms at 3 months after delivery, community social capital was inversely associated with depressive symptoms (β = −0.21, 95%CI = −0.33 to −0.08) after adjusting for covariates and workplace social capital. Despite workplace social capital being inversely associated with depressive symptoms in Model 1 (β = −0.63, 95%CI = −1.13 to −0.14), the coefficient became non-significant after adjusting for paternal depressive symptoms and maternal mental health problems at 1 week after delivery. Community social capital, paternal part-time job, and paternal depressive symptoms at 1 week after delivery showed significant coefficients among covariates. In terms of anxiety at 3 months after delivery, community social capital was inversely associated with anxiety (β = −0.38, 95%CI = −0.66 to −0.11) at 3 months in Model 3. However, the association between workplace social capital and anxiety remained non-significant after adjusting for paternal anxiety and maternal mental health problems at 1 week after delivery. Community social capital, annual household income (JPY 4,010,000–6,000,000) and anxiety at 1 week after delivery showed significant coefficients among covariates.

TABLE 1 | Characteristics of sample in this study (N = 398).

	Total (N = 398)	
	N or mean	**% or SD**
Obstetric hospital		
A	236	59.6
B	162	40.4
Paternal age		
<30	43	10.8
30–<40	252	63.3
40–<50	96	24.1
50+	7	1.8
Paternal education		
High school or less	36	9.0
Some college	36	9.0
College or more	324	81.4
Unknown	2	0.5
Annual household income (JPN yen)		
≦4,000,000	36	9.0
4,010,000–8,000,000	141	35.4
8,010,000–15,000,000	161	40.5
15,001,000+	38	9.5
Unknown	17	4.3
Missing	5	1.3
Paternal ACE total score (0–8)		
0	323	81.2
1	60	15.1
2+	15	3.8
Paternal employment		
Full-time job	364	91.5
Part-time job	4	1.0
Self-employed	24	6.0
Other	6	1.5
Paternal childcare leave		
Yes or Planning to take	46	11.6
No	292	73.4
Missing	60	15.1
Paternal history of psychiatric disorders		
No	390	98.0
Yes	8	2.0
History of delivery		
First birth	273	68.6
Multiparity	124	31.2
Missing	1	0.3
Normal pregnancy		
Yes	327	82.2
No	69	17.3
Missing	2	0.5
Paternal feelings when pregnancy was confirmed		
Happy	334	83.9
Unexpected but happy	48	12.1
Unexpected and confused/did not know what to do/no feelings/other	13	3.3
Missing	3	0.8
Social participation in the last year before delivery		
Yes	74	18.6
No	324	81.4
Number of people who can be consulted about parenting	9.47	8.10

TABLE 2 | Distribution of exposure and outcome.

Variables	N	Mean	SD	Minimum	Max
Workplace social capital	398	3.70	0.80	1	5
Community social capital	398	13.30	2.82	4	20
Depressive symptoms at 1 week after delivery	398	3.11	3.17	0	22
Depressive symptoms at 3 months after delivery	363	3.92	3.77	0	22
Anxiety at 1 week after delivery	398	34.12	8.68	20	68
Anxiety at 3 months after delivery	363	34.49	8.89	19	70

The results of these analyses using complete data were shown in the **Supplementary Tables 2, 3**. Most of the associations were similar to the data employing multiple imputations, although effect sizes were slightly higher in the complete data analysis.

DISCUSSION

This study showed that paternal community social capital, but not workplace social capital, was associated with lower levels of paternal depression and anxiety up to three months after delivery. Thus, we suggest that paternal community social capital may be a preventive factor for paternal depression and anxiety, rather than workplace social capital.

This is the first study to examine the impact of paternal community and workplace social capital on postnatal depression and anxiety. Our findings, in which only community but not workplace social capital was associated with paternal postpartum depression and anxiety, are partially consistent with those of previous studies. In terms of community social capital, it was shown to be associated with mental disorders in a prospective study (49) and a review (18). Moreover, a higher level of maternal social capital during pregnancy was associated with a lower EPDS score at 6–8 weeks after delivery (27). This association can be explained as follows. First, community social capital may alleviate paternal concern about bothering the neighbors due to infant crying. In the Japanese context, caregivers are more likely to be concerned about bothering cohabitants such as grandparents and neighbors due to their infant crying (50). Second, fathers with a higher level of community social capital may be able to access information and receive parenting-related

TABLE 3 | Association between paternal social capital and paternal depressive symptoms at 3 months after delivery after multiple imputations.

		Workplace social capital			**Community social capital**			**Model 3 β (95%CI)**
		Crude β (95%CI)	**Model 1 β (95%CI)**	**Model 2 β (95%CI)**	**Crude β (95%CI)**	**Model 1 β (95%CI)**	**Model 2 β (95%CI)**	
Workplace social capital		**−0.71** **(−1.17 to −0.24)**	**−0.63** **(−1.13 to −0.14)**	−0.15 (−0.60 to 0.30)				−0.01 (−0.47 to 0.45)
Community social capital					**−0.25** **(−0.38 to −0.12)**	**−0.26** **(−0.40 to −0.12)**	**−0.22** **(−0.34 to −0.09)**	**−0.21** **(−0.33 to −0.08)**
Obstetrics hospital	A		Ref	Ref		Ref	Ref	Ref
	B		−0.58 (−1.45 to 0.29)	0.07 (−0.72 to 0.86)		−0.55 (−1.41 to 0.32)	0.11 (−0.67 to 0.88)	0.11 (−0.66 to 0.89)
Paternal age			−0.06 (−3.07 to 0.57)	−0.03 (−0.10 to 0.04)		−0.37 (−0.11 to 0.04)	−0.02 (−0.08 to 0.05)	−0.02 (−0.09 to 0.05)
Paternal education	High school or less		0.05 (−1.35 to 1.45)	−0.14 (−1.39 to 1.11)		Ref	Ref	Ref
	Some college		−1.20 (−2.59 to 0.19)	**−1.37** **(−2.60 to −0.14)**		−1.63 (−3.44 to 0.18)	−1.48 (−3.08 to 0.13)	−1.41 (−3.03 to 0.20)
	College or more		Ref	Ref		Ref	Ref	Ref
	Unknown		−1.84 (−8.03 to 4.35)	−0.42 (−6.09 to 5.26)		−1.17 (−7.44 to 5.10)	0.50 (−5.22 to 6.22)	0.41 (−5.33 to 6.15)
Annual household income (JPN yen)	≦2,000,000		6.30 (−1.92 to 14.51)	3.00 (−4.22 to 10.22)		Ref	Ref	Ref
	2,010,000–4,000,000		**2.16** **(0.51 to 3.81)**	**1.48** **(0.0002 to 2.95)**		−4.83 (−13.11 to 3.45)	−2.15 (−9.35 to 5.05)	−2.11 (−9.32 to 5.10)
	4,010,000–6,000,000		**1.46** **(0.17 to 2.75)**	0.84 (−0.31 to 1.99)		−5.32 (−13.49 to 2.86)	−2.60 (−9.72 to 4.51)	−2.54 (−9.67 to 4.59)
	6,010,000–8,000,000		Ref	Ref		Ref	Ref	Ref
	8,010,000–10,000,000		**1.32** **(0.03 to 2.60)**	**1.34** **(0.19 to 2.49)**		−5.61 (−13.84 to 2.63)	−2.10 (−9.27 to 5.07)	−2.07 (−9.24 to 5.11)
	10,000,000–15,000,000		**1.45** **(0.25 to 2.65)**	**1.42** **(0.35 to 2.49)**		−5.67 (−13.89 to 2.56)	−2.15 (−9.31 to 5.00)	−2.10 (−9.26 to 5.07)
	15,001,000+		0.35 (−1.18 to 1.88)	0.56 (−0.82 to 1.93)		−6.81 (−15.05 to 1.22)	−3.13 (−10.31 to 4.05)	−3.07 (−10.26 to 4.13)
Paternal ACE total score	0		Ref	Ref		Ref	Ref	Ref
	1		0.31 (−0.77 to 1.49)	0.38 (−0.58 to 1.35)		0.32 (−0.75 to 1.40)	0.38 (−0.57 to 1.33)	0.38 (−0.57 to 1.33)
	2+		1.24 (−0.79 to 3.27)	0.30 (−1.51 to 2.11)		1.21 (−0.80 to 3.22)	0.14 (−1.65 to 1.92)	0.11 (−1.68 to 1.90)

(Continued)

TABLE 3 | Continued

		Workplace social capital			Community social capital			Model 3 β (95%CI)
		Crude β (95%CI)	Model 1 β (95%CI)	Model 2 β (95%CI)	Crude β (95%CI)	Model 1 β (95%CI)	Model 2 β (95%CI)	
Paternal employment	Full-time job		Ref	Ref		Ref	Ref	Ref
	Part-time job		**5.21 (1.47 to 8.94)**	**4.83 (1.50 to 8.16)**		**5.17 (1.47 to 8.87)**	**4.74 (1.46 to 8.02)**	**4.68 (1.40 to 7.97)**
	Self-employed		0.65 (−1.02 to 2.32)	0.44 (−1.05 to 1.92)		0.40 (−1.24 to 2.03)	0.45 (−0.99 to 1.89)	0.43 (−1.05 to 1.91)
	Other		−0.32 (−3.95 to 3.32)	0.69 (−2.59 to 3.96)		−0.15 (−3.77 to 3.46)	0.96 (−2.28 to 4.19)	0.99 (−2.25 to 4.23)
Paternal childcare leave	Yes or Planning to take		Ref	Ref		Ref	Ref	Ref
	No		−0.52 (−1.71 to 0.66)	−0.46 (−1.52 to 0.60)		−0.72 (−189 to 0.45)	−0.56 (−1.60 to 0.48)	−0.51 (−1.55 to 0.54)
Paternal history of psychiatric disorders	No		Ref	Ref		Ref	Ref	Ref
	Yes		2.02 (−0.94 to 4.97)	−1.53 (−4.24 to 1.19)		2.21 (−0.70 to 5.13)	−1.61 (−4.28 to 1.07)	−1.63 (−4.31 to 1.06)
History of delivery	First birth		Ref	Ref		Ref	Ref	Ref
	Multiparity		0.11 (−0.73 to 0.94)	−0.04 (−0.96 to 0.88)		0.38 (−0.47 to 1.22)	0.55 (−0.22 to 1.32)	0.51 (−0.27 to 1.28)
Normal pregnancy	Yes		Ref	Ref		Ref	Ref	Ref
	No		−0.11 (−1.13 to 0.91)	−0.04 (−0.96 to 0.88)		−0.04 (−1.04 to 0.98)	−0.06 (−0.96 to 0.85)	−0.05 (−0.96 to 0.86)
Paternal feelings when pregnancy was confirmed	Happy		Ref	Ref		Ref	Ref	Ref
	Unexpected but happy/unexpected and confused/did not know what to do/no feelings/other		0.69 (−0.39 to 1.78)	0.39 (−0.58 to 1.36)		0.71 (−1.89 to 0.45)	0.38 (−0.58 to 1.33)	0.39 (−0.57 to 1.35)
Paternal depressive symptoms at 1 week after delivery				**0.60 (0.48 to 0.72)**			**0.59 (0.48 to 0.71)**	**0.59 (0.47 to 0.71)**
Maternal depressive symptoms at 1 week after delivery				0.05 (−0.07 to 0.16)			0.05 (−0.07 to 0.16)	0.04 (−0.07 to 0.16)
Maternal anxiety at 1 week after delivery				−0.01 (−0.06 to 0.04)			−0.01 (−0.06 to 0.04)	−0.01 (−0.06 to 0.04)
Number of people who can be consulted about parenting								−0.02 (−0.07 to 0.03)

95%CI, 95% Confidence Interval. Boldface means statistical significant ($p < 0.05$).

Model 1 adjusted for paternal education, paternal age, annual household income, paternal employment, paternal childcare leave, paternal adverse childhood experiences, paternal history of psychiatric disorders, history of delivery, obstetrics hospital, normal pregnancy, and paternal feelings when pregnancy was confirmed.

Model 2 added paternal depressive symptoms at 1 week after delivery, maternal depressive symptoms, and anxiety at 1 week after delivery into Model 1.

Model 3 included number of people who can be consulted about parenting and both types of social capital into Model 2.

TABLE 4 | Association between paternal social capital and paternal anxiety at 3 months after delivery after multiple imputations.

		Workplace social capital			**Community social capital**			**Model 3 β (95%CI)**
		Crude β (95%CI)	**Model 1 β (95%CI)**	**Model 2 β (95%CI)**	**Crude β (95%CI)**	**Model 1 β (95%CI)**	**Model 2 β (95%CI)**	
Workplace social capital		**−3.07** **(−4.14 to −2.00)**	**−2.94** **(−4.09 to −1.80)**	−0.72 (−1.74 to 0.31)				−0.55 (−1.59 to 0.49)
Community social capital					**−0.49** **(−0.80 to −0.17)**	**−0.53** **(−0.86 to −0.20)**	**−0.40** **(−0.66 to −0.13)**	**−0.38** **(−0.66 to −0.11)**
Obstetrics hospital	A		Ref	Ref		Ref	Ref	Ref
	B		**−2.82** **(−4.83 to −0.81)**	−0.98 (−2.69 to 0.73)		**−2.82** **(−4.87 to −0.76)**	−0.88 (−0.90 to 2.96)	−0.91 (−2.61 to 0.79)
Paternal age			−0.02 (−0.20 to 0.15)	−0.04 (−0.18 to 0.11)		0.03 (−0.14 to 0.21)	−0.01 (−0.15 to 0.14)	−0.01 (−0.15 to 0.14)
Paternal education	High school or less		−0.85 (−4.09 to 2.38)	−0.21 (−2.91 to 2.50)		−0.31 (−3.60 to 2.99)	−0.16 (−2.84 to 2.51)	−0.22 (−2.91 to 2.48)
	Some college		−0.70 (−3.91 to 2.52)	−0.45 (−3.12 to2.22)		−1.17 (−4.49 to 2.15)	−0.91 (−3.58 to 1.76)	−0.93 (−3.60 to 1.75)
	College or more		Ref	Ref		Ref	Ref	Ref
	Unknown		−4.98 (−18.87 to 8.91)	−2.69 (−14.75 to 9.37)		−3.71 (−17.90 to 10.48)	−1.32 (−13.33 to 10.70)	−1.24 (−13.26 to 10.78)
Annual household income (JPN yen)	≦2,000,000		0.73 (−18.38 to 19.85)	3.38 (−12.21 to 18.96)		2.28 (−17.44 to 22.00)	4.48 (−11.00 to 19.95)	4.41 (−11.08 to 19.90)
	2,010,000–4,000,000		1.97 (−1.85 to 5.80)	0.26 (−2.93 to 3.46)		2.30 (−1.63 to 6.23)	0.22 (−2.96 to 3.40)	0.20 (−2.99 to 3.38)
	4,010,000–6,000,000		**3.53** **(0.54 to 6.54)**	**2.96** **(0.50 to 5.41)**		**4.27** **(1.19 to 7.35)**	**3.27** **(0.82 to 5.72)**	**2.20** **(0.75 to 5.65)**
	6,010,000–8,000,000		Ref	Ref		Ref	Ref	Ref
	8,010,000–10,000,000		2.81 (−0.13 to 5.75)	1.59 (−0.88 to 4.06)		2.69 (−0.34 to 5.71)	1.75 (−0.71 to 4.20)	1.88 (−0.59 to 4.36)
	10,000,000–15,000,000		1.92 (−0.86 to 4.71)	1.29 (−1.01 to 3.58)		1.28 (−1.55 to 4.11)	1.19 (−1.07 to 3.45)	1.34 (−0.95 to 3.63)
	15,001,000+		1.18 (−2.37 to 4.73)	1.43 (−1.52 to 4.37)		0.79 (−2.86 to 4.44)	1.23 (−1.71 to 4.16)	1.27 (−1.67 to 4.20)
Paternal ACE total score	0		Ref	Ref		Ref	Ref	Ref
	1		0.02 (−2.48 to 2.51)	0.38 (−1.70 to 2.46)		0.14 (−2.41 to 2.69)	0.41 (−1.66 to 2.47)	0.37 (−1.70 to 2.43)
	2+		2.67 (−2.00 to 7.35)	−1.89 (−5.85 to 2.06)		3.28 (−1.50 to 8.05)	−2.16 (−6.09 to 4.36)	−2.21 (−6.14 to 1.73)

(Continued)

TABLE 4 | Continued

		Workplace social capital			Community social capital			Model 3 β (95%CI)
		Crude β (95%CI)	**Model 1 β (95%CI)**	**Model 2 β (95%CI)**	**Crude β (95%CI)**	**Model 1 β (95%CI)**	**Model 2 β (95%CI)**	
Paternal employment	Full-time job		Ref	Ref		Ref	Ref	Ref
	Part-time job		3.89 (−4.74 to 12.52)	2.53 (−4.64 to 9.70)		4.18 (−4.65 to 13.00)	2.42 (−4.69 to 9.53)	2.40 (−4.72 to 9.52)
	Self-employed		0.34 (−3.52 to 4.19)	−0.32 (−3.54 to 2.90)		−1.14 (−5.04 to 2.76)	−0.54 (−3.70 to 4.36)	−0.22 (−3.43 to 2.99)
	Other		0.51 (−7.97 to 8.98)	−0.10 (−7.30 to 7.10)		0.21 (−8.42 to 8.84)	0.27 (−6.90 to 4.36)	0.39 (−6.78 to 7.56)
Paternal childcare leave	Yes or Planning to take		Ref	Ref		Ref	Ref	Ref
	No		−0.96 (−3.69 to 1.77)	−1.33 (−3.61 to 0.95)		−1.65 (−4.43 to 1.13)	−1.59 (−3.85 to 0.67)	−1.52 (−3.79 to 0.76)
Paternal history of psychiatric disorders	No		Ref	Ref		Ref	Ref	Ref
	Yes		0.70 (−6.00 to 7.40)	−1.37 (−8.08 to 4.35)		2.30 (−4.48 to 9.09)	−1.27 (−6.90 to 4.36)	−1.57 (−7.26 to 4.11)
History of delivery	First birth		Ref	Ref		Ref	Ref	Ref
	Multiparity		0.48 (−1.46 to 2.43)	0.20 (−1.48 to 1.88)		1.00 (−1.02 to 3.02)	0.59 (−1.11 to 2.29)	0.63 (−1.07 to 2.33)
Normal pregnancy	Yes		Ref	Ref		Ref	Ref	Ref
	No		0.71 (−1.62 to 3.05)	0.95 (−1.01 to 2.91)		1.28 (−1.10 to 3.65)	1.03 (−0.90 to 2.96)	0.89 (−1.05 to 2.84)
Paternal feelings when pregnancy was confirmed	Happy		Ref	Ref		Ref	Ref	Ref
	Unexpected but happy/unexpected and confused/did not know what to do/no feelings/other		1.54 (−0.96 to 4.04)	−0.16 (−2.25 to 1.94)		1.72 (−0.82 to 4.26)	−0.19 (−2.26 to 1.89)	−0.20 (−2.28 to 1.88)
Paternal anxiety at 1 week after delivery				**0.62** **(0.52 to 0.71)**			**0.63** **(0.54 to 0.72)**	**0.61** **(0.52 to 0.71)**
Maternal depressive symptoms at 1 week after delivery				0.06 (−0.19 to 0.32)			0.05 (−0.20 to 0.30)	0.06 (−0.19 to 0.32)
Maternal anxiety at 1 week after delivery				0.01 (−0.09 to 0.12)			0.01 (−0.09 to 2.96)	0.01 (−0.10 to 0.11)
Number of people who can be consulted about parenting								0.02 (−0.09 to 0.12)

95%CI, 95% Confidence Interval. Boldface means statistical significant ($p < 0.05$).

Model 1 adjusted for paternal education, paternal age, annual household income, paternal employment, paternal childcare leave, paternal adverse childhood experiences, paternal history of psychiatric disorders, history of delivery, obstetrics hospital, normal pregnancy, and paternal feelings when pregnancy was confirmed.

Model 2 added paternal anxiety at 1 week after delivery and maternal depressive symptoms and anxiety at 1 week after delivery into Model 1.

Model 3 included number of people who can be consulted about parenting and both types of social capital into Model 2.

care, as mothers with a higher level of community social capital can access better prenatal care and delivery care (51). Further studies to identify the mechanism of association between community social capital and paternal postnatal depression and anxiety should be conducted. Nonetheless, community social capital plays a significant role in preventing postnatal depression and anxiety among not only mothers but also fathers, indicating that promoting community social capital for fathers may be effective in preventing paternal postnatal depression and anxiety.

In terms of workplace social capital, we found no association, although previous studies reported that workers with a lower level of workplace social capital showed the onset of depression in Finland (male: 20%) (52), Germany (male: 74.4%) (53), and Japan (male: 77.6%) (54). This discrepancy can be explained by differences in the target population (i.e., age of bearing a child) and the assessment period (i.e., right after delivery). For workers, assuming the family status is stable, the workplace is considered a major social context (55). Thus, employment status, job stress, working hours, and job insecurity have huge impacts on the mental health of workers (56). Workers with a higher level of workplace social capital are more likely to be able to cope with their stress (57), which may lead to lower levels of mental health problems. However, in the case of changing family status, such as bearing a child, fathers may face further stress related to parenting and the relationship with their partner, in addition to job stress during the transition to fatherhood. Therefore, fathers may not be able to cope with the stress related to fatherhood transition through workplace social capital. The type of support and information fathers receive from community and workplace social capital during the perinatal period needs to be identified.

Though there was no association between workplace social capital and mental health problems, the significant coefficient was shown in Model 1 in which depressive symptoms and anxiety at 1 week after delivery was not adjusted. Paternal depressive symptoms and anxiety at 1 week after delivery are considered not only confounders but also mediators on a time-series basis, which indicates overadjustment. Additionally, participants who reported higher levels of depressive symptoms and anxiety might be more likely to perceive social capital negatively. This negative perception due to mental health problems might influence workplace social capital more strongly than community social capital because people spend more time and have greater social relations at the workplace than the community (58). In the current study, the correlation between workplace and community social capital was small ($r = 0.17$). Thus, the impact of mental health problems on workplace and community social capital might differ. Despite adjusting for a history of psychiatric disorders in our analysis, further studies to assess paternal depressive symptoms and anxiety before and during pregnancy, and a longitudinal study with a larger sample size that excludes fathers with depressive symptoms and anxiety at the baseline, are needed.

The current study has several limitations. First, our findings are limited in generalizability due to the participant recruitment method and paternal characteristics. Study participants were recruited from two obstetrics hospitals in Tokyo, Japan. Furthermore, we found that the annual household income of our participants was higher than in another study that targeted families living in Tokyo (59). Second, both exposure (i.e., paternal social capital) and outcomes (paternal depression and anxiety) were self-reported, which may lead to common method bias for causal inference on the association between paternal social capital and their mental health problems. Further research needs to be conducted to assess mental health using objective measures such as interviews by professionals. Third, although the follow-up rate in this study was high and multiple imputations were performed, it is possible that fathers with lower levels of social capital and severe mental health problems dropped out of our survey. Fourth, there are unmeasured confounders, such as severe obstetric complications during pregnancy, although we could assess normal pregnancy in a subjective way.

CONCLUSIONS

Our findings indicate that a higher level of paternal community social capital at birth, but not workplace social capital, prevents paternal depression and anxiety at 3 months after delivery. Even though fathers show a similar level of postnatal depression as mothers, they are less likely to have social support compared to mothers (60). To date, there are some programs designed to promote social capital for parents, nearly all of which target mothers (61, 62). To prevent paternal mental health problems during the postnatal period, an intervention to promote paternal community, rather than workplace, social capital may be warranted.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

This study was approved by the Institutional Review Board of the National Center for Child Health and Development (730) and Tokyo Medical and Dental University (M2017-078). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

TF designed the study. TF and AI managed administration of the study, including the ethical review process and provided critical comments on the manuscript related to intellectual content. SD analyzed data and drafted the manuscript. All authors have read and approved the final manuscript.

FUNDING

This study was supported by Grants-in-Aid for Scientific Research from the Japan Society for the Promotion of Science (JSPS KAKENHI Grant Numbers 15K12735 and 18K13318), the Mitsubishi Foundation, Grants for Social Welfare Activities, a Research Development Grant for Child Health and Development from the National Center for Child Health and Development, and the Ministry of Health, Labour and Welfare (21DA1004 and 19AA100).

ACKNOWLEDGMENTS

The authors are grateful to Manami Ochi, Michiko Sekiguchi, Yuri Hiramoto, and Hiromi Nakano who recruited the participants and managed the survey.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.782939/full#supplementary-material

REFERENCES

1. Bandelow B, Michaelis S. Epidemiology of anxiety disorders in the 21st century. *Dialogues Clin Neurosci.* (2015) 17:327. doi: 10.31887/DCNS.2015.17.3/bbandelow
2. Wynter K, Rowe H, Fisher J. Common mental disorders in women and men in the first six months after the birth of their first infant: a community study in Victoria, Australia. *J Affect Disord.* (2013) 151:980–5. doi: 10.1016/j.jad.2013.08.021
3. Cameron EE, Sedov ID, Tomfohr-Madsen LM. Prevalence of paternal depression in pregnancy and the postpartum: an updated meta-analysis. *J Affect Disord.* (2016) 206:189–203. doi: 10.1016/j.jad.2016.07.044
4. Rao WW, Zhu XM, Zong QQ, Zhang Q, Hall BJ, Ungvari GS, et al. Prevalence of prenatal and postpartum depression in fathers: a comprehensive meta-analysis of observational surveys. *J Affect Disord.* (2020) 263:491–9. doi: 10.1016/j.jad.2019.10.030
5. Takehara K, Suto M, Kakee N, Tachibana Y, Mori R. Prenatal and Early postnatal depression and child maltreatment among Japanese fathers. *Child Abuse Negl.* (2017) 70:231–9. doi: 10.1016/j.chiabu.2017.06.011
6. Pinto TM, Samorinha C, Tendais I, Figueiredo B. Depression and paternal adjustment and attitudes during the transition to parenthood. *J Reprod Infant Psychol.* (2020) 38:281–96. doi: 10.1080/02646838.2019.1652256
7. Johansson M, Svensson I, Stenström U, Massoudi P. Depressive symptoms and parental stress in mothers and fathers 25 months after birth. *J Child Health Care.* (2017) 21:65–73. doi: 10.1177/1367493516679015
8. Paulson JF, Bazemore SD. Prenatal and postpartum depression in fathers and its association with maternal depression: a meta-analysis. *JAMA.* (2010) 303:1961–9. doi: 10.1001/jama.2010.605
9. Koch S, De Pascalis L, Vivian F, Meurer Renner A, Murray L, Arteche A. Effects of male postpartum depression on father–infant interaction: the mediating role of face processing. *Infant Ment Health J.* (2019) 40:263–76. doi: 10.1002/imhj.21769
10. Ip P, Li TM, Chan KL, Ting AYY, Chan CY, Koh YW, et al. Associations of paternal postpartum depressive symptoms and infant development in a chinese longitudinal study. *Infant Behav Dev.* (2018) 53:81–9. doi: 10.1016/j.infbeh.2018.08.002
11. Gutierrez-Galve L, Stein A, Hanington L, Heron J, Lewis G, O'Farrelly C, et al. Association of maternal and paternal depression in the postnatal period with offspring depression at age 18 years. *JAMA Psychiatry.* (2019) 76:290–6. doi: 10.1001/jamapsychiatry.2018.3667
12. Philpott LF, Savage E, FitzGerald S, Leahy-Warren P. Anxiety in fathers in the perinatal period: a systematic review. *Midwifery.* (2019) 76:54–101. doi: 10.1016/j.midw.2019.05.013
13. Leach LS, Poyser C, Cooklin AR, Giallo R. Prevalence and course of anxiety disorders (and symptom levels) in men across the perinatal period: a systematic review. *J Affect Disord.* (2016) 190:675–86. doi: 10.1016/j.jad.2015.09.063
14. Vismara L, Rollè L, Agostini F, Sechi C, Fenaroli V, Molgora S, et al. Perinatal parenting stress, anxiety, and depression outcomes in first-time mothers and fathers: a 3-to 6-months postpartum follow-up study. *Front Psychol.* (2016) 7:938. doi: 10.3389/fpsyg.2016.00938
15. Pinto TM, Figueiredo B, Pinheiro LL, Canário C. Fathers' parenting self-efficacy during the transition to parenthood. *J Reprod Infant Psychol.* (2016) 34:343–55. doi: 10.1080/02646838.2016.1178853
16. Parfitt Y, Pike A, Ayers S. The impact of parents' mental health on parent–baby interaction: a prospective study. *Infant Behav Dev.* (2013) 36:599–608. doi: 10.1016/j.infbeh.2013.06.003
17. Xue X, Reed WR, Menclova A. Social capital and health: a meta-analysis. *J Health Econ.* (2020) 72:102317. doi: 10.1016/j.jhealeco.2020.102317
18. Ehsan AM, De Silva MJ. Social capital and common mental disorder: a systematic review. *J Epidemiol Community Health.* (2015) 69:1021–8. doi: 10.1136/jech-2015-205868
19. Borgonovi F, A. Life-cycle approach to the analysis of the relationship between social capital and health in Britain. *Soc Sci Med.* (2010) 71:1927–34. doi: 10.1016/j.socscimed.2010.09.018
20. Lochner K, Kawachi I, Kennedy BP. Social capital: a guide to its measurement. *Health Place.* (1999) 5:259–70. doi: 10.1016/S1353-8292(99)00016-7
21. Berry HL, Welsh JA. Social capital and health in Australia: an overview from the household, income and labor dynamics in Australia survey. *Soc Sci Med.* (2010) 70:588–96. doi: 10.1016/j.socscimed.2009.10.012
22. Fowler K, Etchegary H. Economic crisis and social capital: the story of two rural fishing communities. *J Occup Organ Psychol.* (2008) 81:319–41. doi: 10.1348/096317907X226972
23. Brehm J, Rahn W. Individual-Level Evidence for the Causes and Consequences of Social Capital. *Am J Polit Sci.* (1997) 1997:999–1023. doi: 10.2307/2111684
24. Han Y, Chung RY-N. Are both individual-level and county-level social capital associated with individual health? a serial cross-sectional analysis in China, 2010–2015. *BMJ Open.* (2021) 11:e044616. doi: 10.1136/bmjopen-2020-044616
25. Jetten J, Haslam SA, Cruwys T, Greenaway KH, Haslam C, Steffens NK. Advancing the social identity approach to health and well-being: progressing the social cure research agenda. *Eur J Soc Psychol.* (2017) 47:789–802. doi: 10.1002/ejsp.2333
26. Cruwys T, Dingle GA, Haslam C, Haslam SA, Jetten J, Morton TA. Social group memberships protect against future depression, alleviate depression symptoms and prevent depression relapse. *Soc Sci Med.* (2013) 98:179–86. doi: 10.1016/j.socscimed.2013.09.013
27. Kritsotakis G, Vassilaki M, Melaki V, Georgiou V, Philalithis AE, Bitsios P, et al. Social capital in pregnancy and postpartum depressive symptoms: a prospective mother–child cohort study (the Rhea Study). *Int J Nurs Stud.* (2013) 50:63–72. doi: 10.1016/j.ijnurstu.2012.08.012
28. Nagy E, Moore S, Silveira PP, Meaney MJ, Levitan RD, Dubé L. Low socioeconomic status, parental stress, depression, and the buffering role of network social capital in mothers. *Journal of Mental Health.* (2020) 2020:1–8. doi: 10.1080/09638237.2020.1793118
29. Eastwood JG, Kemp LA, Jalaludin BB, Phung HN. Neighborhood adversity, ethnic diversity, and weak social cohesion and social networks predict high rates of maternal depressive symptoms: a critical realist ecological study in South Western Sydney, Australia. *Int J Health Serv.* (2013) 43:241–66. doi: 10.2190/HS.43.2.d

30. Ehsan A, Klaas HS, Bastianen A, Spini D. Social capital and health: a systematic review of systematic reviews. *SSM-Popul Health.* (2019) 8:100425. doi: 10.1016/j.ssmph.2019.100425
31. Firouzbakht M, Tirgar A, Ebadi A, Nia HS, Oksanen T, Kouvonen A, et al. Psychometric properties of persian version of the short-form workplace social capital questionnaire for female health workers. *Int J Occup Environ Med.* (2018) 9:184. doi: 10.15171/ijoem.2018.1264
32. Miyajima T, Yamaguchi H, I. Want to but I Won't: pluralistic ignorance inhibits intentions to take paternity leave in Japan. *Front Psychol.* (2017) 8:1508. doi: 10.3389/fpsyg.2017.01508
33. Ministry of Health LaW. *Basic Survey of Gender Equality in Employment Management 2020 (2021)*. Available from: https://www.mhlw.go.jp/toukei/list/71-r02.html (accessed 15 December, 2022).
34. Lofors J. Sundquist K. Low-linking social capital as a predictor of mental disorders: a cohort study of 45 million. *Swedes Soc Sci Med.* (2007) 64:21–34. doi: 10.1016/j.socscimed.2006.08.024
35. Lu W, Xiao Y. Adverse childhood experiences and adolescent mental disorders: protective mechanisms of family functioning, social capital, and civic engagement. *Health Behav Res.* (2019) 2:e1035-e. doi: 10.4148/2572-1836.1035
36. Zhou C, Zheng W, Yuan Q, Zhang B, Chen H, Wang W, et al. Associations between social capital and maternal depression: results from a follow-up study in China. *BMC Preg Childbirth.* (2018) 18:1–9. doi: 10.1186/s12884-018-1673-9
37. Kaneko N, Ishigaki K, Agawa K. Positive child-rearing feelings of mothers and regional cultural aspect of social capital. *J Cult Nurs Stud.* (2019) 11:12–21. doi: 10.24658/bunkakango.11.1_1_12
38. Yagi J, Fujiwara T, Yambe T, Okuyama M, Kawachi I, Sakai A. Does social capital reduce child behavior problems? Results from the Great East Japan earthquake follow-up for children study. *Social Psychiat Psychiatr Epidemiol.* (2016) 51:1117–23. doi: 10.1007/s00127-016-1227-2
39. Nawa N, Isumi A, Fujiwara T. Community-level social capital, parental psychological distress, and child physical abuse: a multilevel mediation analysis. *Soc Psychiatry Psychiatr Epidemiol.* (2018) 53:1221–9. doi: 10.1007/s00127-018-1547-5
40. Odagiri Y, Ohya Y, Inoue S, Hayashi T, Uchiyama A, Takamiya T, et al. Reliablity and validation of the japanese version of the measure of workplace social capital scale (in Japanese). *Sangyo Eiseigaku Zasshi.* (2010) 83:631.
41. Kouvonen A, Kivimäki M, Vahtera J, Oksanen T, Elovainio M, Cox T, et al. Psychometric evaluation of a short measure of social capital at work. *BMC Public Health.* (2006) 6:1–10. doi: 10.1186/1471-2458-6-251
42. Okano T. Validation and reliability of a Japanese version of the epds. *Arch Psychiatric Diag Clin Eval.* (1996) 7:523–33.
43. Nakazato K, Mizuguchi K. Studies on psychometric characteristics of depression in the field of internal medicine. *Jpn J Psychosomc Med.* (1982) 22:107–12.
44. Rubin DB. *Multiple Imputation for Nonresponse in Surveys.* New York: Wiley (2004).
45. Sterne JA, White IR, Carlin JB, Spratt M, Royston P, Kenward MG, et al. Multiple imputation for missing data in epidemiological and clinical research: potential and pitfalls. *BMJ.* (2009) 338:157–60. doi: 10.1136/bmj.b2393
46. Takeda T, Tsuji Y, Kurita H. Psychometric properties of the Japanese Version of the adult attention-deficit hyperactivity disorder (ADHD) self-report scale (ASRS-J) and its short scale in accordance with DSM-5 diagnostic criteria. *Res Dev Disabil.* (2017) 63:59–66. doi: 10.1016/j.ridd.2017.02.011
47. Kurita H, Koyama T, Osada H. Autism-spectrum quotient-Japanese version and its short forms for screening normally intelligent persons with pervasive developmental disorders. *Psychiatry Clin Neurosci.* (2005) 59:490–6. doi: 10.1111/j.1440-1819.2005.01403.x
48. Yoshida K, Yamashita H, Conroy S, Marks M, Kumar C, A. Japanese version of mother-to-infant bonding scale: factor structure, longitudinal changes and links with maternal mood during the early postnatal period in Japanese mothers. *Arch Womens Ment Health.* (2012) 15:343–52. doi: 10.1007/s00737-012-0291-1
49. Fujiwara T, Kawachi I, A. Prospective study of individual-level social capital and major depression in the United States. *J Epidemiol Commun Health.* (2008) 62:627–33. doi: 10.1136/jech.2007.064261
50. Fujiwara T, Yamada F, Okuyama M, Kamimaki I, Shikoro N, Barr RG. Effectiveness of educational materials designed to change knowledge and behavior about crying and shaken baby syndrome: a replication of a randomized controlled trial in Japan. *Child Abuse Negl.* (2012) 36:613–20. doi: 10.1016/j.chiabu.2012.07.003
51. Lamarca GA. do C Leal M, Sheiham A, Vettore MV. The association of neighbourhood and individual social capital with consistent self-rated health: a longitudinal study in brazilian pregnant and postpartum women. *BMC Preg Childbirth.* (2013) 13:1–17. doi: 10.1186/1471-2393-13-1
52. Kouvonen A, Oksanen T, Vahtera J, Stafford M, Wilkinson R, Schneider J, et al. Low workplace social capital as a predictor of depression: the finnish public sector study. *Am J Epidemiol.* (2008) 167:1143–51. doi: 10.1093/aje/kwn067
53. Woo JM, Okusaga O, Postolache TT. Seasonality of suicidal behavior. *Int J Environ Res Public Health.* (2012) 9:531–47. doi: 10.3390/ijerph9020531
54. Sakuraya A, Imamura K, Inoue A, Tsutsumi A, Shimazu A, Takahashi M, et al. Workplace social capital and the onset of major depressive episode among workers in japan: a 3-year prospective cohort study. *J Epidemiol Community Health.* (2017) 71:606–12. doi: 10.1136/jech-2016-208561
55. Kawachi I, Kennedy BP, Glass R. Social capital and self-rated health: a contextual analysis. *Am J Public Health.* (1999) 89:1187–93. doi: 10.2105/AJPH.89.8.1187
56. Harnois G, Gabriel P. *Mental Health and Work: Impact, Issues and Good Practices*. (2000). New York: Cornell University.
57. Firouzbakht M, Tirgar A, Oksanen T, Kawachi I, Hajian-Tilaki K, Nikpour M, et al. Workplace social capital and mental health: a cross-sectional study among iranian workers. *BMC Public Health.* (2018) 18:794. doi: 10.1186/s12889-018-5659-3
58. Gao J, Weaver SR Dai J, Jia Y, Liu X, Jin K, et al. Workplace social capital and mental health among chinese employees: a multi-level, cross-sectional study. *PLoS ONE.* (2014) 9:e85005. doi: 10.1371/journal.pone.0085005
59. Ando S, Nishida A, Yamasaki S, Koike S, Morimoto Y, Hoshino A, et al. Cohort profile: the Tokyo teen cohort study (Ttc). *Int J Epidemiol.* (2019) 48:1414-g. doi: 10.1093/ije/dyz033
60. Mao Q, Zhu LX, Su XY. A comparison of postnatal depression and related factors between Chinese new mothers and fathers. *J Clin Nurs.* (2011) 20:645–52. doi: 10.1111/j.1365-2702.2010.03542.x
61. Sommer TE, Sabol TJ, Chase-Lansdale PL, Small M, Wilde H, Brown S, et al. Promoting parents' social capital to increase children's attendance in head start: evidence from an experimental intervention. *J Res Educ Eff.* (2017) 10:732–66. doi: 10.1080/19345747.2016.1258099
62. Fielden JM, Gallagher LM. Building social capital in first-time parents through a group-parenting program: a questionnaire survey. *Int J Nurs Stud.* (2008) 45:406–17. doi: 10.1016/j.ijnurstu.2006.09.008

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

SYSTEMATIC REVIEW
published: 18 February 2022
doi: 10.3389/fpsyt.2022.813519

How Do Children of Parents With Mental Illness Experience Stigma? A Systematic Mixed Studies Review

Lisa-Marie Dobener [1,2*†], *Julia Fahrer* [1†], *Daniel Purtscheller* [2,3†], *Annette Bauer* [2,4†], *Jean Lillian Paul* [2,3†] and *Hanna Christiansen* [1,2†]

[1] Department of Clinical Child and Adolescent Psychology, Philipps University Marburg, Marburg, Germany, [2] Mental Health Research Program, The Village, Ludwig Boltzmann Gesellschaft, Innsbruck, Austria, [3] Department of Psychiatry, Psychotherapy and Psychosomatics, Division of Psychiatry I, Medical University Innsbruck, Innsbruck, Austria, [4] Care Policy and Evaluation Centre (CPEC), London School of Economics and Political Science, London, United Kingdom

Edited by:
Francisco José Eiroa-Orosa,
University of Barcelona, Spain

Reviewed by:
Joanna Fox,
Anglia Ruskin University,
United Kingdom
Sami Richa,
Saint Joseph University, Lebanon

***Correspondence:**
Lisa-Marie Dobener
lisa-marie.dobener@uni-marburg.de

†ORCID:
Lisa-Marie Dobener
orcid.org/0000-0001-8901-2312
Julia Fahrer
orcid.org/0000-0002-1661-9313
Daniel Purtscheller
orcid.org/0000-0002-9993-262X
Annette Bauer
orcid.org/0000-0001-5156-1631
Jean Lillian Paul
orcid.org/0000-0003-0891-7035
Hanna Christiansen
orcid.org/0000-0002-8104-0711

Specialty section:
This article was submitted to
Child and Adolescent Psychiatry,
a section of the journal
Frontiers in Psychiatry

Received: 11 November 2021
Accepted: 10 January 2022
Published: 18 February 2022

Citation:
Dobener L-M, Fahrer J, Purtscheller D, Bauer A, Paul JL and Christiansen H (2022) How Do Children of Parents With Mental Illness Experience Stigma? A Systematic Mixed Studies Review. Front. Psychiatry 13:813519. doi: 10.3389/fpsyt.2022.813519

Stigma can have devastating health and wellbeing impacts, not just on people with mental health problems, but on people associated with the stigmatized person. This is called stigma-by-association. Children whose parents have mental health problems are a particularly vulnerable group, and stigma acts as a mechanism, contributing to the transgenerational transmission of mental disorders. The current study is a systematic mixed studies review, synthesizing knowledge about how this group of children experience stigma-by-association. Overall, 32 studies were included, after a systematic search including quantitative, qualitatative, and mixed methods studies. The methodological quality was assessed and qualitative content analysis undertaken. We grouped children's stigma experiences into four dimensions, i.e., experienced stigma, anticipated stigma, internalized stigma, and structural discrimination. Results show that stigma is an important factor in those children's lives, and needs further investigation in qualitative and quantitative research. The current study emphasizes the importance of anti-stigma interventions and campaigns.

Keywords: children of parents with mental illness, stigma, child mental health, systematic review, qualitative content analysis

INTRODUCTION

Children of parents with a mental illness[1] have received increasing attention over recent years, especially as it is estimated that one in five children world-wide has a parent with a mental illness (1–3). Those children often face specific challenges that are associated with reduced mental health, poorer academic achievement, and impaired social well-being and quality of life (1, 4, 5). Social adversities associated with mental illness, including poverty, as well as genetic predisposition and family dysfunction, act as mechanisms contributing to these risks (6). Those mechanisms have been integrated into a comprehensive model: the transgenerational transmission of mental disorders (TTMD) model identifies four major domains (1. parent, 2. family, 3. child, 4. social environment) that interact with their respective systems, and are influenced by five transmission mechanisms

---

[1] Throughout our paper we mainly use the term "children" when referring to the children of people with a mental illness as this is the population of interest. This can also mean that we are talking about the adult children of parents with a mental illness—depending on the study population in the original papers—by using the term 'children', referring to children of parents with a mental illness.

(1. genetics, 2. prenatal factors, 3. parent-child-interaction, 4. family, and 5. social factors) (1, 7). According to this model, stigma can be seen as a component of social factors, acting as a mechanism for the transmission of mental disorders that leads to multiple challenges and negative outcomes.

As captured in the concepts of stigma-by-association (SBA) (8), family stigma [e.g., (9)], or courtesy stigma (10, 11), stigma can affect not only the person with a mental health problem, but also people connected to the person, such as family or friends (12). The conceptual thinking of SBA by Pryor et al. (8) focuses on the general public and potential mechanisms that might contribute to the emergence of SBA, but lacks the perspective and experiences of those affected. Current research mostly describes SBA as guilt, blame, and contamination, ascribed to and experienced by family members (13, 14). Children are most frequently described as experiencing such "contamination" stigma, i.e., the general public tends to see them as being contaminated by the parental mental illness [e.g., (13, 14)]. The theoretical model of SBA by Philipps and Gates (15) focuses on various stigma dimensions and facets that could affect children, but for the very special group with incarcerated parents. On an experiential level, perceived stigma, internalized stigma, discrimination, and differences in social, cultural, economic, and political power are identified for this group. Another qualitative study classified stigma dimensions described by family members of people with schiziophrenia, finding structural elements of discrimination, and interpersonal interaction elements, such as social exclusion (16, 17).

Goffman has defined experiencing "courtesy stigma" as 'an individual who is related through the social structure to the stigmatized (…) leads the wider society to treat both individuals in some respect as one' (11), and research so far has not conceived of the stigma dimensions for relatives, but rather solely named the stigma facets to be important for relatives of people with a stigmatized condition. Given this, we assume that SBA for children can potentially contain all of the stigma facets described for the primary recipients: *Experienced SBA* describes personally experienced prejudice and discrimination (18, 19); *perceived SBA* explains the "belief that most people will devalue, discriminate the stigmatized" (19); *anticipated SBA* includes expectations that others will devalue and discriminate against them (18); *affiliate stigma* describes the self-stigma of associates of people with a mental illness, i.e., the "internalization of stigma among associates of targeted individuals" on the affective, cognitive and behavioral level (20); and *structural discrimination* targets societal and policy structures that reproduce existing social inequalities (21).

The experience of SBA and discrimination, the anticipation of what others may think about them, and accommpaning self-stigmatization, can have a deep impact on individuals. There is evidence that 8–22 % of family members of people with a mental illness experienced stigma's negative impacts on themselves, e.g., ruined self-esteem (21%), disrupted family relationships with other family members (22%), and with their ill family members (20%) (16). It has also been reported that family members of someone with a mental illness may avoid social situations and events, reduce or break contact with family and friends, spend energy on hiding the secret, and experience discrimination within employment and/or housing situations (14, 22). Although a growing body of research focuses on SBA, very few studies investigated how these stigma experiences affect individual family members (23); systematic studies, especially those targeting children, are missing that would shed light on their SBA experiences while classifying their experiences into the various stigma dimensions. The same is true for the concept of affiliate stigma (20): while children of parents with a mental illness often do care for their parents [parentification, e.g., (24)], they have not been the focus in studies on affiliate stigma so far.

A recent systematic review on the evidence of stigma concepts for children of parents with a mental illness aimed to identify stigma-related experiences and outcomes as reported by parents and children (25). Their findings summarize stigma concepts from the primary literature, all of which describe different individual facets of children's and their parents' experiences of stigma. This review highlights the lack of uniform definitions for such stigma experiences, as well as the lack of an all-encompassing concept that includes the various dimensions of stigma experiences of children whose parents have a mental illness. The main finding of this review is that affected children report feelings of embarrassment, shame, and the need to hide their parental mental illness, though those findings are not integrated into an overall framework of different stigma dimensions.

The aim of the present study is therefore (1) to gain knowledge about how the children of parents with a mental illness experience stigma, (2) to synthesize this knowledge into a primary model of SBA on an experienced level for this population. In so doing, we (3) hope to contribute to SBA's theoretical model, that is currently insubstantial and not focused on children.

To achieve this, evidence from qualitative, quantitative, and mixed-method studies regarding children's stigma experiences related to their parents' mental illness is synthesized and collated.

METHODS

Protocol and Registration

The review is registered and approved by PROSPERO, registration number CRD42019112838 (www.crd.york.ac.uk/PROSPERO).

Design

This review follows the PRISMA statement for conducting systematic reviews (26). Considering stigmatization of mental illness and its impact upon children is a complex and multifaceted problem. We thus included qualitative, quantitative, and mixed-methods studies to synthesize knowledge from different methodological perspectives, and diverse evidence.

Search Strategy

Five electronic databases (PubMed, Cochrane Library, PsycINFO, PSYNDEX, Web of Sciences) were searched (updated April 2020), using a detailed search strategy developed for PubMed and adapted for other databases, see **Figure 1**. The four different search term combinations (illustrated in the figure by the colors blue, green, orange and yellow) were combined with the OR function. Additional references from current reviews and theoretical articles were also reviewed to identify additional citations. Our search strategy consisted of five aspects and was developed with the Village research team (https://village.lbg.ac.at/about), an expert librarian, and an expert in stigma theories.

Eligibility Criteria

Studies were eligible for inclusion if they met the following criteria: (a) original primary peer-reviewed research; (b) published in English or German, (c) investigation of stigma experiences, or stigma as a relevant issue for children, and (d) children of parents with a mental illness were a (sub-)sample in the study. We did not use a filter for publication dates.

Exclusion Criteria

Studies were excluded if they: (a) focused on the general public as study population, i.e., we were not interested in the stigma that public stigma carries against children, but on the experience-level of the affected individuals, or (b) their stigma measures did not allow for a clear distinction between the different stigma dimensions, or if their only informative value for children was whether they experienced more or less stigma than other family members, or (c) did not differentiate enough between children and other family members regarding their stigma experiences, i.e., if the original paper did not state clearly if their results reflect the experiences of all family members, including children, we excluded them as we could not ensure whether those findings applied to this population. In qualitative studies with different subsamples, we used only those statements made by the children. In quantitative studies, we only used the analysis results of the subgroup of children.

Study Selection

After conducting the standard search as outlined above, all titles and abstracts were screened by independent reviewers (L-MD, FS). They were eligible for full text screening if children of parents with mental illness were participants in the study, and if the abstract either directly highlighted stigma or stigma-related concepts (see search strategy, aspect 5) or included children's specific experiences that may include stigma experiences. Full texts of potentially eligible studies were retrieved and independently assessed for eligibility by two review authors (L-MD, JF). Any disagreement was resolved through discussion with a third review author (HC).

Quality Appraisal

Three review authors (L-MD, AB, JLP) independently assessed the risk of bias in the included studies. Two authors independently appraised the quality and looked for convergence. Due to heterogeneity in methodological approaches in the included studies, i.e., qualitative, quantitative as well as mixed methods methodology, the "Mixed Methods Appraisal Tool" [MMAT; (27)] was used. The quality of each study was assessed applying two general criteria for all the studies, and five criteria adapted to each specific methodology requirement. As recommended by the authors of the MMAT tool, we did not exclude any studies due to low methodological quality, but used their information to estimate potential biases. Nevertheless, to provide an overview of study quality, we calculated an overall score for each study in terms of study quality, ranging from 0 to 100%, in which we allocated one star for each 20% of the criteria met.

Data Extraction

Various information was extracted from the original studies addressing our research question. In addition to general information on data collection, analysis methods, and study population, all text passages related to stigma experiences were extracted. This means that the results and discussion sections of the included studies were systematically searched for stigma-related terms and content. Once the data analysis was completed, we determined which stigma dimensions could be allocated to which studies. All the extracted data are summarized in **Supplementary Table S1**. Two authors (L-MD, DP) extracted data independently, discrepancies were identified and resolved through discussion with a third author (HC).

Analysis

Qualitative content analysis was conducted for comprehensive description and interpretation. A qualitative data-based convergent synthesis design was adopted (28). We used Kuckartz' (29) guidance for conducting qualitative content analyses, using the same synthesis method for both qualitative and quantitative studies. Thus, quantitative data were transformed into qualitative categories in order to explore similarities and/or differences between studies. The quantitative data were also examined in terms of their stigma results, and narratively described results were coded. Any evidence found was summarized correspondingly. We used the software MAXQDA (Version 2018) to manage the storage of analysis. Extracted results were read several times, and memos were made to become immersed in the meaning. Sub-categories were compiled inductively and material-driven. Out of the formulated sub-categories, generic categories were abstracted, describing themes that emerged. Those generic categories were grouped into main categories with the help of comprehensive stigma frameworks (18, 19). The complete data (i.e., extracted results of interest) was coded with those main categories.[2] Coding was done independently by two researchers (L-MD, FS) to ensure the reliability of data analysis. Discrepancies were resolved through discussion with HC.

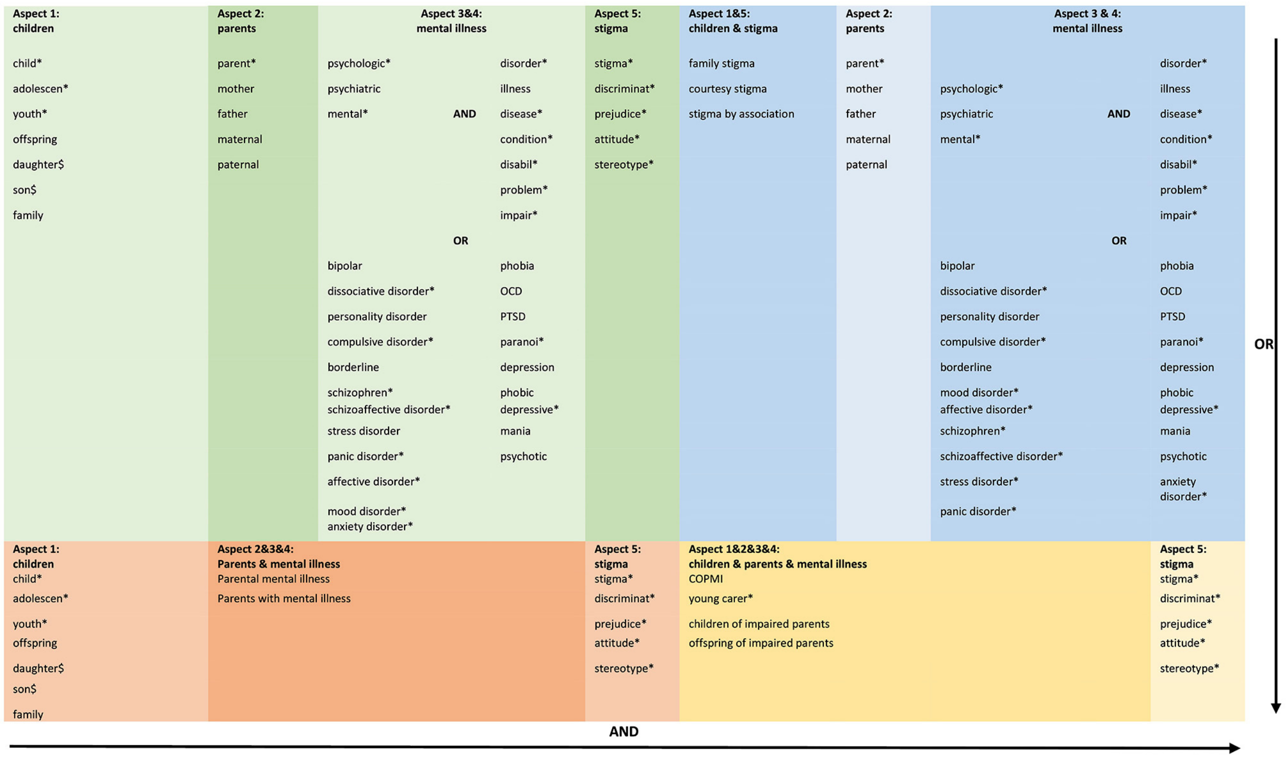
RESULTS

Search Outcome

Most studies were excluded at the title/abstract screening stage (see **Figure 2**), leaving 208 for full-text screening. Most of the studies were excluded at this stage because we could not differentiate their findings to other family members well enough, or because studies were not empirical or not peer-reviewed. A few quantitative studies also had to be excluded in the

---

[2] The coding guideline is available upon request.



Aspect 1: children
child*
adolescen*
youth*
offspring
daughter$
son$
family

Aspect 2: parents
parent*
mother
father
maternal
paternal

Aspect 3&4: mental illness
psychologic*
psychiatric
mental*

AND

disorder*
illness
disease*
condition*
disabil*
problem*
impair*

OR

bipolar
dissociative disorder*
personality disorder
compulsive disorder*
borderline
schizophren*
schizoaffective disorder*
stress disorder
panic disorder*
affective disorder*
mood disorder*
anxiety disorder*
phobia
OCD
PTSD
paranoi*
depression
phobic
depressive*
mania
psychotic

Aspect 5: stigma
stigma*
discriminat*
prejudice*
attitude*
stereotype*

Aspect 1&5: children & stigma
family stigma
courtesy stigma
stigma by association

Aspect 2: parents
parent*
mother
father
maternal
paternal

Aspect 3 & 4: mental illness
psychologic*
psychiatric
mental*

AND

disorder*
illness
disease*
condition*
disabil*
problem*
impair*

OR

bipolar
dissociative disorder*
personality disorder
compulsive disorder*
borderline
mood disorder*
affective disorder*
schizophren*
schizoaffective disorder*
stress disorder*
panic disorder*
phobia
OCD
PTSD
paranoi*
depression
phobic
depressive*
mania
psychotic
anxiety disorder*

Aspect 1: children
child*
adolescen*
youth*
offspring
daughter$
son$
family

Aspect 2&3&4: Parents & mental illness
Parental mental illness
Parents with mental illness

Aspect 5: stigma
stigma*
discriminat*
prejudice*
attitude*
stereotype*

Aspect 1&2&3&4: children & parents & mental illness
COPMI
young carer*
children of impaired parents
offspring of impaired parents

Aspect 5: stigma
stigma*
discriminat*
prejudice*
attitude*
stereotype*

OR

AND

FIGURE 1 | Search strategy.

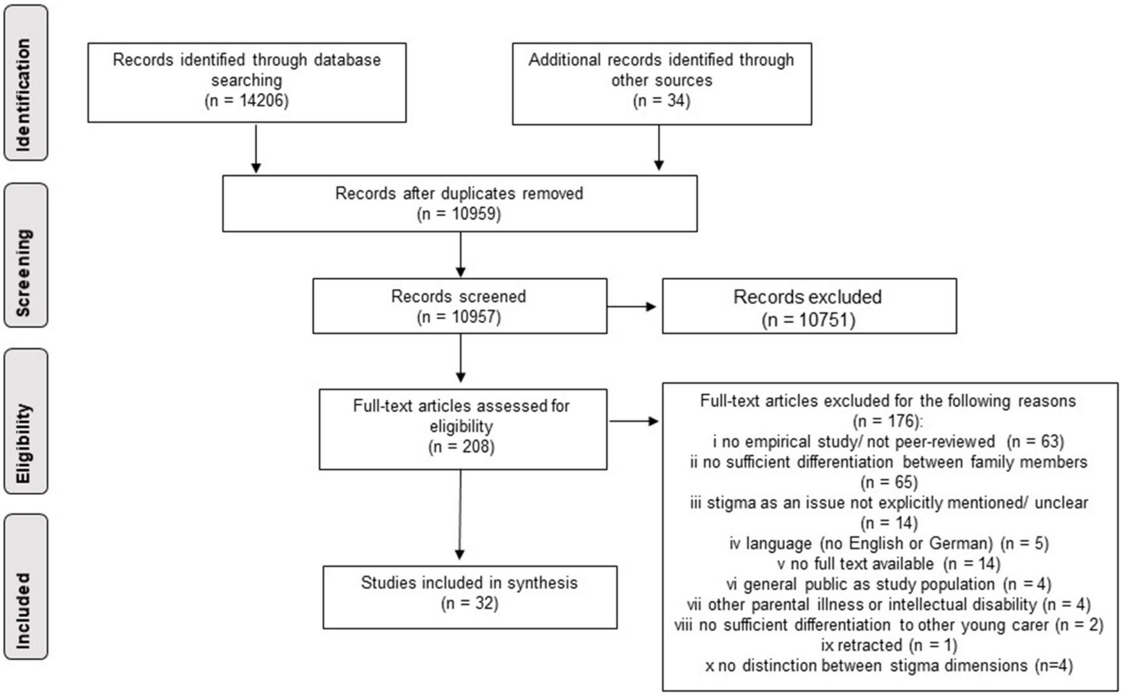


FIGURE 2 | PRISMA flow chart demonstrating study selection.

last stage because they measured unclear and mixed-up stigma dimensions, making it impossible to assign them to individual dimensions (see **Figure 1**). We were ultimately able to include a total of 32 articles from 30 studies[3].

Study and Population Characteristics

The majority of studies (30 ≙ 93.8 %) were qualitative; only one quantitative and one mixed method study were included, **Table 1**. Most of the studies were conducted in Europe (20 ≙ 62.5 %). Half of them had minor children as participants, the other half had adult children; one study combined the two. We ended up including more than half of the studies that had not aimed to explore stigma in this population, but aimed to capture their (everyday) experiences and burden. Stigma then emerged as one of the central themes in these studies. However, this also meant that most of the studies lacked a clear definition of stigma. **Supplementary Table S1** provides a detailed description of the included studies.

Quality Appraisal

The quality of the studies was very heterogeneous and overall, 13 of the included studies met 60% or more criteria (for details see **Table 2**). They ranged from zero to five in their score, meaning that they ranged from 0 to 100 % of the criteria met. In qualitative studies, that were assessed as being of lower quality, often a clear rationale regarding the data collection method, the method of analysis or interview guidelines was missing, or the study population was not sufficiently defined. Quantitative studies of lower quality did not include a representative sample for their target population and had a low risk of non-response bias. The mixed-method study especially lacked in contrasting the results from the two study approaches and bringing the results together.

Identified Aspects of Stigma Related to Parental Mental Illness

Following our analysis, we identified four different stigma dimensions described by or regarding children that are well-known from the stigma experiences of people who have a stigmatized condition themselves: *experienced SBA*, *anticipated SBA*, *affiliate (internalized) stigma,* as well as *structural discrimination*. Within these main categories, we identified various subcategories that are described in the following sections. **Table 3** reports the category system for stigma dimensions, their frequencies, and associated studies.

Experienced SBA—Having Experienced Unpleasant (Re)Actions of Others

We identified two generic categories of SBA in this section: (1) having unmet emotional needs, and (2) experiencing the hostile behaviors of others. Both revealed various subcategories and are now described in more detail.

TABLE 1 | Study characteristics.

Characteristics	*n* (% of 32 articles)
Publication date	
2013–2019	21 (65.6)
2007–2012	7 (21.9)
2001–2006	4 (12.5)
Country of origin	
Europe	20 (62.5)
North America	5 (15.6)
Australia	4 (12.5)
Asia	2 (6.3)
Africa	1 (3.1)
Study design	
Qualitative	30 (93.8)
Quantitative	1 (3.1)
Mixed Methods	1 (3.1)
Study population	
COPMI	18 (56.2)
Children, including COPMI	2 (6.3)
Relatives, including COPMI	6 (18.7)
Children, parents and professionals	4 (12.5)
Young carer, including COPMI	2 (6.3)
Age of COPMI	
Aged above 18/21	14 (43.8)
6–22 years	14 (43.8)
Not reported /applicable	3 (9.3)
Both	1 (3.1)
Parental mental illness	
Diverse Disorders combined	11 (34.4)
Mental illness, not specified	7 (21.8)
Affective Disorder	4 (12.5)
Schizophrenia	4 (12.5)
Alcohol or Drug Dependence	4 (12.5)
Obsessive Compulsive Disorder (OCD)	2 (6.3)

Having Unmet Emotional Needs

Being Confronted With Inappropriate Language and Contents About Mental Illness. Some children reported that they were unhappy about being classified in a way that they would not describe themselves, e.g., being called a "young carer", and were unhappy about not being asked whether they identified with that designation (47). Some participants reported that others gave them unwanted advice, such as recommending them to use birth control to avoid passing on mental illness to their children (42). One child remembers that her boyfriend's family reacted to her parent's mental illness saying "*What if you two got married and you have children and they have a mental illness?*" (42).

Experiencing Others Who Cannot Understand Them or Cope With Their Situation. Often, children reported that people did not know how to act and cope with the information of parental illness [e.g., (48)]. One participant stated that they wished there was

---

[3] Van der Sanden (22) and van der Sanden (30) stem from one study with different research questions but the same data; Cogan et al. (31) and Cogan et al. (32) also stem from one study.

TABLE 2 | Quality assessment (MMAT).

Criterion	**Clear research questions?**	**Data address research questions?**	**Qualitative approach appropriate?**	**Data collection methods adequate?**	**Findings adequately derived?**	**Interpretation of results sufficiently substantiated?**	**Coherence between qualitative data sources, collection, analysis and interpretation**	**Overall quality of the study[a]**
QUALITATIVE STUDIES								
Blakeman et al. (33)	Yes	Can't tell	Yes	Can't tell	Yes	Yes	Can't tell	***/80%
Bolas et al. (34)	Yes	Yes	Yes	Can't tell	Can't tell	Can't tell	Can't tell	*/20%
Carroll and Tuason (35)	Yes	Yes	Yes	Yes	Yes	Can't tell	Yes	****/80%
Cogan et al. 2005 (31)	Yes	Yes	Yes	Can't tell	Yes	Yes	Yes	****/80%
Dam et al. 2018 (36)	Yes	Can't tell	Can't tell	Yes	Can't tell	Yes	Can't tell	**/40%
Davison and Scott (37)	Can't tell	Can't tell	Yes	Yes	Can't tell	Can't tell	Can't tell	*/20%
Haug Fjone et al. (38)	Yes	Yes	Yes	Yes	Can't tell	No	Can't tell	**/40%
Fudge and Mason (39)	Can't tell	Yes	Yes	Can't tell	Can't tell	No	Can't tell	*/20%
Griffiths et al. (40)	Yes	Yes	Can't tell	Can't tell	Can't tell	Yes	Yes	**/40%
Kadish (41)	Can't tell	Can't tell	Yes	Can't tell	Can't tell	Yes	Can't tell	**/40%
Karnieli-Miller et al. (42)	Yes	Yes	Yes	Can't tell	Yes	Yes	Yes	****/ 80%
Krupchanka et al. (43)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	*****/ 100%
Leahy (44)	Yes	Can't tell	Yes	Can't tell	Can't tell	Can't tell	Can't tell	*/ 20%
Leinonen et al. (45)	Yes	Yes	Yes	Yes	Yes	Can't tell	Yes	****/ 80%
McCormack et al. (46)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	*****/ 100%
Moore et al. (47)	Yes	Yes	Yes	Can't tell	Can't tell	Can't tell	Yes	**/ 40%
Mordoch and Hall (48)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	*****/ 100%
Murphy et al. (49)	Yes	Can't tell	Yes	Yes	No	Yes	Can't tell	***/ 60%
Nieto-Rucian and Furness (50)	Yes	Yes	Yes	Yes	Can't tell	Can't tell	Yes	***/ 60%
Oskouie et al. (51)	Can't tell	Can't tell	Yes	No	Yes	Can't tell	Can't tell	*/20%
Östman (52)	Yes	Yes	Yes	Can't tell	No	No	No	*/ 20%
Rezayat et al. (53)	Yes	Yes	Yes	Can't tell	Yes	Yes	Yes	****/80%
Stengler-Wenzke et al. (54)	Yes	Yes	Yes	Can't tell	Yes	Yes	Yes	****/ 80%
Tabak et al. (55)	Yes	Can't tell	Can't tell	Can't tell	Can't tell	Yes	Yes	**/ 40%
Tamutiene and Jogaite (57)	Yes	Yes	Yes	Can't tell	Yes	Can't tell	Can't tell	**/ 40%
Trondsen and Tjora (58)	Yes	Yes	Yes	Yes	Can't tell	Yes	Yes	****/ 80%
van der Sanden et al. (22)	Yes	Yes	Can't tell	Yes	Yes	Can't tell	No	**/40%
van der Sanden et al. (30)	Yes	Yes	Yes	Can't tell	Yes	Yes	Yes	****/80%
Wahl et al. (59)	Can't tell	Yes	Can't tell	Can't tell	Can't tell	No	No	-/0%
Widemalm and Hjärthag (60)	Yes	Yes	Can't tell	Yes	Can't tell	Can't tell	Yes	**/40%
Criterion	**Clear research questions?**	**Data adress research questions?**	**Sampling strategy relevant to address research question?**	**Sample representative of the target population?**	**Measurements appropriate?**	**Risk of nonresponse bias low?**	**Statistical analysis appropriate to answer the research question?**	**Quality**
QUANTITATIVE STUDIES								
Haverfield and Theiss (61)	Yes	Yes	Yes	No	Yes	No	Can't tell	**/40%

(Continued)

TABLE 2 | Continued

Criterion	Clear research questions?	Data address research questions?	Adequate rationale for using mixed methods design?	Different study components effectively integrated?	Outputs of integration adequately interpreted?	Divergences between quantitative and qualitative results adequately addressed?	Different study components adhere to the quality criteria of each tradition of the methods involved?	
MIXED METHODS STUDY								
Cogan et al. (32)	Yes	Can't tell	Can't tell	Can't tell	Can't tell	No	Can't tell	-/0%

*[a] The quality score was calculated according to the official advices of the authors of the MMAT tool (http://mixedmethodsappraisaltoolpublic.pbworks.com/w/file/fetch/140056890/Reporting%20the%20results%20of%20the%20MMAT.pdf). *20% of the criteria met; **40% of the criteria met; ***60% of the criteria met; ****80% of the criteria met; *****100% of the criteria met.*

greater community awareness so "*that they know there's nothing wrong—anyone could get it if they get stressed out—they could get it*" (39). Some also reported that others failed to intervene, could make things worse (35), or that friends did not understand their experiences and they were disappointed in how friends responded (48). Some children emphasized that they wanted to talk to family members, but their parents and grandparents refused, and they felt unable to talk openly about such family issues (49, 50). A few children described having nobody to talk to and ask for help. They believed that this was due to taboos and others' ignorant behaviors [e.g., (35, 36)], having direct influences on their social interactions.

Experiencing Withdrawal and Rejection. In addition, children perceived that others actively avoided them: for example, an adult child in Dam's study described, respectively (36) *"When I met someone, they ignored me; they went over to the other side of the road"*, as a result of having lived in a small community with a mentally-ill parent. Moreover, participants reported family members breaking off their relationship with them after finding out about their parent's illness (65), or experiencing the loss of friendships: *"...my friend's mother called me over and told me that I couldn't be friends with her, because I'll be the same as my mother"* (57).

Experiences of Hostile Behaviors of Others

Being the Victim of Bullying and Laughter. Bullying (e.g., "*I was bullied, the others laughed at me.*") (36) as one form of overt hostile behavior, was frequently mentioned: Children reported being: teased, laughed at, drawn into fights, and treated like a "*leper*" (36). A participant in Mordoch and Hall's (48) study stated that they *"Sometimes [I] get in fights by accident"* because other people labeled them as different. In another study, one participant wished that others would "*stop teasing*" (39).

Being Confronted With Hurtful Words. Furthermore, children reported that their peers used stigmatizing words when talking about their ill parents, like 'crazy' (36), 'mad' (51), or their living situation as a 'crazy house' (59).

Anticipated SBA—Fearing Reactions of Others in the Future

Regarding anticipated stigma in the future, similar topics that we identified in the experienced SBA emerged for anticipated SBA: Children feared overt hostile behaviors from others, as well as negative attitudes and ascriptions. They were also worried that others would not be able to understand and would reject them. Those generic categories are now presented together with their sub-categories.

Fearing Hostile Behaviors of Others

Fearing Ridicule. When children were asked why they hesitate to tell others about their parent's mental illness, they described being afraid others would laugh at them (32, 57), and being afraid of being teased or bullied (33, 49).

Fearing Gossip. Furthermore they feared that other people would share this information and gossip about them (33, 48). One participant describes her tendency to isolate herself due to fearing gossip and accompanying discrimination: "*I was not able to develop close friendships at school because I had this fear of them coming home with me and seeing what it was like and then telling everybody at school and I would be a laughing stock*" (33).

Fear of Negative Attitudes and Ascriptions

Fear of Being Labeled as "Different". Children frequently feared being labeled as "*different from others*" if people discovered their parent's mental illness. They thought people might believe their mothers, for example, might not act like other mothers [e.g., (60, 64)], or that they are "*berserk*" (33).

Fear of Other People's Negative Attitudes. They also feared that others might be scared or have *negative attitudes* toward them or their family (52). One participant stated that they do not invite friends home because they are scared that others might think something bad about their family (33). Another child reported being scared of being judged or criticized for having a "*whacko*" father (46).

Fear for Parents to be Described as Bad. Some children worried others might think their parents could not take care of them because of their mental illness and that they might be taken away (39, 57). One participant stated: "(…) *It's just that I'm scared they'll put it down to her being a bad mum... she's not a bad mum, she loves us*" (32).

TABLE 3 | Identified aspects of stigma related to parental mental illness.

Main category	Generic category	Sub-category	References
Experienced SBA (29)	Having unmet emotional needs (18)[a]	• Experiencing withdrawal and rejection (6)	• (33, 36, 57, 62, 63)
		• Experiencing others who cannot understand or cope with parental mental illness (7)	• (35, 39, 48–50)
		• Being confronted with inappropriate language and statements about mental illness (5)	• (30, 42, 47, 57)
	Experiencing hostile behaviours of others (11)	• Being the victim of bullying and laughter (8)	• (36, 39, 45, 48, 62)
		• Being confronted with hurtful words (3)	• (36, 51, 59)
Anticipated SBA (29)	Fearing hostile behaviors of others (6)	• Fearing gossip (2)	• (33, 48)
		• Fearing ridicule (4)	• (32, 33, 49, 57)
	Fearing of negative attitudes and ascriptions (15)	• Fear of other people's negative attitudes (5)	• (33, 40, 52, 59)
		• Fear of being labeled as “different“ (7)	• (30, 46, 48, 50, 52, 60, 64)
		• Fear for parents to be described as bad (3)	• (39, 57)
	Fearing others' lack of understanding and rejection (8)	• Fearing others' lack of knowledge and understanding (4)	• (37, 57, 59, 62)
		• Fearing withdrawal and rejection (4)	• (22, 33, 34, 41)
Affiliate stigma (51)	Perceiving themselves as being contaminated (9)	• Struggling to avoid being contaminated (8)	• (30, 35–37, 54, 58, 64)
		• Fearful of passing it on (1)	• (30)
	Perceiving themselves as being inferior (42)	• Feeling ashamed and embarrassed (30)	• (22, 30–36, 40, 41, 44, 46, 49–51, 54, 55, 57–61, 64)
		• Perceiving themselves as different from others (9)	• (30, 33, 41, 46, 49, 50, 58, 64)
		• Self-blaming (3)	• (31, 64)
Structural discrimination (35)	Perceiving discrimination within the mental health system (10)	• Not receiving information in hospital (3)	• (32, 36)
		• Experiencing cold furniture and atmosphere in the hospital (1)	• (36)
		• Experiencing insensitive treatment by professionals (3)	• (36, 50)
		• Perceiving a lack of care provided for the parent (3)	• (43, 60)
	Perceiving discrimination within the education system (10)	• Needing more education in school about mental illness (1)	• (32)
		• Feeling teachers ignore their parents' mental illness (7)	• (33, 36, 57, 62)
		• Disadvantages (2)	• (39, 62)
	Perceiving a lack of knowledge provided/societal taboo (8)	• Needing more information and openness from society (8)	• (36, 37, 39, 49, 50, 58)
	Perceiving discrimination within the police (2)	• Experiencing discrimination during contacts with the people (1)	• (43)
		• Feeling ignored by the police (1)	• (57)
	Perceiving discrimination within media (2)	• Perceiving media representations of people with mental illness as bad (2)	• (32)
	Perceiving discrimination within social work (3)	• Feeling unseen by social workers (1)	• (57)
		• Perceiving social workers as a source of control rather than help (2)	• (57)

[a] *Numbers in brackets show how many codes we identified for those categories.*

Fearing Others' Lack of Understanding and Rejection

Fearing Others' Lack of Knowledge About Mental Illness and Lack of Understanding. Moreover, children worried that others have little knowledge of mental illness (59) and that their situation and experiences will not be understood (57, 62). In some instances, they anticipated others' misconceptions and did not get any help, or tell anybody about it *"because some children just do not know what this [mental illness] is and then just tell people that it is something really bad, that is stupid for you"* (59). On the other hand, two participants in the study from Davison and Scott (37) anticipated that personal support interventions could even make things worse for children, in case other people try too hard to help them, leading to more worries and fear: "*People sort of trying to help them too much when they don't have a problem could make people overthink things… It might increase the fear factor…*".

Fearing Withdrawal and Rejection. They also feared others might withdraw from, reject (34, 41), or exclude them (33). Some children also feared losing contacts, such as relationships when their partner discovers their parent's mental illness: "*I've got a girlfriend, should I tell her… I've been going out with her for 3 or 4 months…but she's never met my mother. And I do have a particular reason, for, like, putting it off as long as possible. And that, oh I don't know, feeling embarrassed, it's just the idea, I don't know actually. Let her get to know me first and if she likes me enough, then it won't make any difference any more*" (22).

Affiliate Stigma—Internalizing the Stigma

The self-stigmatization of children manifested in two different ways, namely the two generic categories we identified: (1) perceiving themselves as being contaminated, and (2) perceiving themselves as being inferior. Both are now presented in more detail together with their subcategories.

Perceiving Themselves as Being Contaminated

Struggling to Avoid Being Contaminated. van der Sanden et al. (30) found that adult children feared contamination. The fear of being contaminated relied on one of two components: Either (1) children knew about their status of being a member of an at-risk group for developing a mental illness, which could result in fear and self-reflexive sensitivity related to possible symptoms (58); or (2) they feared being seen as connected to their ill parent so closely that they would also be considered "*crazy*" (35): "*My Mom is crazy, so they are going to think I am crazy". One* child reported that parental mental illness is something that cannot be discussed because it is like a "*an infected tumor never to heal*" (P9) (36). An online self-help group for adolescents with a mentally ill parent reduced these fears, as one participant described: *"They seemed quite normal; after all, it was just that they had a terrible situation. Then I thought, "Oh my God, they are never going to be mentally ill like that."... I felt it was less likely that I would also become like my mom.... Because [one of the participants] had a boyfriend, I felt that I too had a chance to get a boyfriend, friends, and live a completely normal life […]"* (58).

Fearful of Passing It on. One child reported fearing stigmatization being passed on to the next generation when they have children themselves (30), and thus characterize themselves as a possible source for transmitting mental illness.

Perceiving Themselves as Being Inferior

Feeling Ashamed and Embarrassed. Beliefs of being inferior were mainly characterized by statements related to shame and embarrassment: Children often viewed their parents' mental illness as a "*secret*" (35, 44) that must be kept "*behind closed doors*" (31). One child described doing this to protect their parent from other people's stigmatizing behaviors (44). Often, children mentioned that they did not want anybody to know about their parent's mental health issues, but sometimes were unsure why they were ashamed, or did not want anybody to know: "*And I do have a particular reason, for, like, putting it off as long as possible. And that, oh I don't know, feeling embarrassed, it's just the idea, I don't know actually*" (22). Sometimes participants even reflected that this secrecy would keep them from establishing deep relationships, for example one participant said: *"I avoid entering deep relations with others due to fear of explaining my situation"* (51). One son of a father with a mental illness stated: *"I wish my father had another disease and didn't suffer from mental illness. Father's illness is very bad for me. I am ashamed to speak about it and to communicate with others"* (51). In the quantitative study by Haverfield and Theiss (61), intrafamilial topic avoidance regarding parental illness was also associated with magnified avoidance of disclosing the topic to others—this was evident in the male participants at least.

Perceiving Themselves as Different to Others. Frequently, this shame, embarrassment, and disclosure was described as a result of being seen as "different", "abnormal", or "wrong". A commonly mentioned experience for children was that they felt different from their peers (41, 49, 50), and the feeling of being different led them to think dichotomously, i.e., believing other families were "*right*", while their own family was "*wrong*": *"Our family was wrong…we were just a dysfunctional, broken family, we just weren't normal like other families"* (46).

Self-Blaming. Self-blame was also identified in children: They were more likely to blame themselves for their parents' difficult situation than were children whose parents have no mental illness (31). One child explained: *"I sometimes think it's my fault, I blame myself for the way my dad is feeling, even though he tells me it's not"* (31).

Structural Discrimination—Perceiving Inequalities in Institutions and Within the Society

We identified three generic categories within the structural discrimination facet now discussed in more detail, namely perceived discrimination within (1) the mental health system; (2) the education system, and (3) other sources of discrimination

Perceiving Discrimination Within the Mental Health System.

Most of the references for structural discrimination related to interactions with the healthcare system: Children described interactions at the interpersonal level within the health care system, such as being treated insensitively, and perceived that doctors were afraid of them and maintained an emotional distance (50): *"To visit mum in the hospital was like coming to an office. It was a nonexistent relationship, you know. Hello, Goodbye, You must go in that direction (P4)"* (36). Furthermore, they felt ignored by staff (36). Due to feeling fear and insecure when visiting their parents in hospital, they wished that someone would stay with them and talk to them about their situation (36). On a broader institutional level, children strongly requested more general information about mental illness from mental health workers (32). Children remarked that the cold furniture in the hospitals made them feel uncomfortable (36). They also mention a general lack of care for their parents. They had the impression that their parents were getting inadequate healthcare, and were discharged due to inadequate bed capacity, leaving the children in charge of their parents: "*After 2 weeks she was sent home, even though she didn't feel ready. I and my sister had to move home and take care of her ourselves. I will never forgive the psychiatric institution for what they did to me and my sister. We literally had to act like 'extra mothers' to our own mother*" (60).

Perceiving Discrimination Within the Education System

The education system was also identified as a source of structural discrimination: children complained of a lack of education about mental illness at school (32). They criticized a lack of understanding about students who have to look after their parents (39), with teachers ignoring their situation instead of talking with them "*Well it's a very stereotypical town where you would never want to admit that anything is wrong... so a lot of it was hush hush. So, like, we'll give you a little extension on your homework. If you need someone to talk to you can go and talk to somebody. But we're not going to like, raise any ags*" (33); see also (36, 62).

Other Sources of Structural Discrimination

Other sources of structural discrimination were the police (43, 57), media (31), and social work (57). Events experienced there or unclarified mandates from the institutions led to a mistrust in the children, who felt abandoned or did not seek help for fear of losing their parents (57). Above all, the general taboo around this topic was identified, which is not inherent in a specific organization, but rather within society in general, with people not talking to children about these problems, which might hinder children obtaining appropriate and useful information (49, 50, 58). Not getting any information about parental mental illness and its heritability or about their risks in general left children to overestimate their risk of developing a bipolar disorder themselves (37). One participant said: "*I think knowing about the risk would be really helpful—I've always worried that I would get it (BD)*" (37).

DISCUSSION

We identified 32 studies for this review that investigated or addressed the stigma experiences of children with parents with a mental illness. The studies are heterogeneous in their design and methodological quality. Low-quality papers failed to state a comprehensible rationale for their data collection or analysis, or failed to sufficiently describe (or even recruit) a representative sample. More than half of the studies did not investigate stigma primarily, rather, they only identified stigma as an important problem for this population. Due to a mixed-up and vague measurement of stigma dimensions, we were only able to include one quantitative study and one mixed-method study. Qualitative content analysis was done inductively, and we were able to cluster our results in four stigma dimensions that match those absorbed in stigma theories (18, 19, 66): experienced SBA, anticipated SBA, affiliate stigma, and structural discrimination. We were able to fill those dimensions with content that is specific for the stigma experiences that the children of parents with a mental illness suffer, and we have thus closed the gap in showing that these theoretical categories were confirmed with qualitative content analyses, and that the stigma experiences that the children expressed align with theory. The affiliate stigma dimensions we identified highlight the close connections between stigma and feelings of guilt, isolation, and secrecy in affected children, supporting results of the review by Reupert et al. (25).

While this review does not enable causal conclusions about the health risks after stigmatizing experiences, our results demonstrate that such experiences are indeed stressful and likely to affect the health and well-being of those stigmatized, as conceptualized in the TTMD model (1, 7). Stigma is therefore no mere isolated mechanism that impairs a child's well-being—it is a mechanism interwoven and manifold on different levels in families with mental illness.

Dimensions of Stigma

Experienced SBA

Our findings regarding *experienced SBA* reveal that children do have experiences that are very specific to their role as the offspring of a parent with a mental illness. At school and in other areas of social life, they are bullied and teased. They often feel incapable of making strong connections with other people in a satisfying way, as they often sense the need to keep their parent's mental illness a secret. In their early years, children can experience—depending on the severity and symptoms of their parent's mental illness and the existence of reliable other family members—a lack of emotional support within their family (55, 56). For them therefore, relationships functioning as a secure base outside the family might be even more important than for other children. In adolescence, this can be problematic, as it is a life phase characterized by various developmental steps that include the establishment of new and meaningful relationships outside the primary family, and developing a strong sense of identity (70).

Anticipated SBA

As far as we know, *anticipated stigma* has not been described in the literature to characterize family members of people with a mental illness. This SBA dimension was identified in this review and was closely connected to feelings of fear and worry. This could be an important aspect of hiding the parent's mental illness and failing to seek help, and it supports the evidence from studies on the primary recipients of mental illness stigma, which showed that anticipated stigma and discrimination are key reasons for not seeking help [e.g.,(67)].

Affiliate Stigma

This review sheds light on how children might *internalize the stigma*. While some quantitative studies claimed that children are more apt to experience contamination, and that blame and shame play more important roles for the parents and spouses of people with a mental illness (13, 68), our review's results show that blame and shame are also important for children. This is especially true regarding the negative perception of themselves as being different from others—thereby perceiving themselves as inferior. This review reveals this as a specific and frequently mentioned self-stigma for children. This finding is in line with general stigma theories for primary recipients, which posit that a core component of stigma is the mark of otherness, which is then followed by negative evaluation (69). Children seem to internalize this *otherness* by feeling a sense of being "abnormal" or "wrong". Park and Park's (71) family-stigma concepts could explain the frequent mention of feeling "different" identified in this review. According to their theory, general family stigma arises for two reasons: (1) it might be due to negative events (like the parental illness) or (2) to exceptional family structures. As structures in families with a parental illness differ in many ways [e.g., (53)] from those of other families, we can assume that such children are perceived and perceive themselves differently because of both labels. This may provide an explanation for the perceived family otherness so frequently cited in this review. Often, this otherness was associated with inferiority, which was considered a reason that this must be kept a secret and possibly keeping children and their families from seeking help. As previous research has shown for relatives of people with a mental illness, internalized stigmatization is especially associated with psychological problems such as reduced self-esteem—causing both psychological distress and lower quality of life (72, 73).

Structural Discrimination

Furthermore, our review demonstrates that *structural discrimination* is a key stigma dimension not only for those suffering from mental illness, but also for their children. A recently published systematic review (25) described structural discrimination being detected within law, medicine and education: in legal terms, for example, the greater likelihood of parents losing custody of their child, as well as being accused of having an "unsound mind" in all matters for people with mental illness. These are undoubtedly important findings for families and especially parents with a mental illness that probably affect the entire family system including children. Nevertheless, our review suggests that there is structural discrimination surpassing those descriptions that is described by the children themselves and which we clustered as an element of structural discrimination, as it occurs within institutions or characterizes a societal attitude that differs from individual attitudes. The results of the current review show that there is an inherent stigmatizing structure in different areas of society, e.g., schools and hospitals. For children, structural discrimination includes their parents' experiences, which in turn have consequences for themselves. This form of stigma also entails disadvantages personally experienced through various institutions. There is evidence that healthcare systems are a main source of children's structural discrimination. Children are affected by the lack of care offered or provided to their parents: They are often put in charge of taking care of their parents when hospitals/adult mental health professionals turn them away. Generally, the most significant themes within structural discrimination is the lack of education and information about mental illness, and society's tendency to ignore the subject. This prevents children from getting the necessary information and support to establish a healthy way of coping with their situation, like talking to others. Mental health literacy and positive family communication about the disorder are both known to be major protective factors for children (74, 75). However, the results of this review reveal a societal structure in which they do not get enough information at all.

Strengths and Limitations

This is the first systematic mixed-method review to explore and collate different facets of stigma for children of parents with a mental illness, focusing on the experience level of children themselves and grouping those facets into dimensions to inform how we conceive of affiliate stigma on the experience level. Having included both qualitative and quantitative study designs is a strength of this review—the topic is thus covered from different perspectives and with various measures. Furthermore, including both under-age and adult children deepens our knowledge of their stigma experiences: Views from children still living with their parent (as in about half of the included studies) provide information on how they are assessing their current situation. Retrospective views from adult children add important information, especially as some thoughts cannot be expressed and understood by children of younger ages, as they might not be aware of stigmatizing structures. For instance, children cannot anticipate that some stigmatizing experiences might influence their future in a specific way.

However, retrospective views can also lead to bias. Memories can change, and ascriptions made more easily [recall-bias (76)]. Secondly, we only included studies written in English or German, as well as peer-reviewed publications, and no gray literature. Thus, it is possible some important literature is missing. In addition, the studies mostly came from Western countries, restricting our results and conclusions to those cultures. As stigma is the result of the social context and therefore differs over time and cultures, the present review provides an overview of possible topics relevant for children, but to ensure generalizability, a wider range of cultures would need to be addressed. As most of the studies came from Europe, this review mainly contributes to understanding how stigma affects children of parents with a mental illness in European countries, which might well differ in other countries. Third, the quality of studies we included ranged from very low to very high quality. In many qualitative studies, data collection methods, analyses methods, as well as interview guidelines were not specified, which made it difficult to assess their quality and adequacy. In addition, we mainly included qualitative studies as we could only include one quantitative and one mixed-method study. The other quantitative studies of potential relevance had to be excluded because of their low relevance for this specific population, i.e., they only assessed which kinship status experienced more or less stigma by association, or they mixed up different stigma measures which made it impossible to classify the stigma.

Implications for Policy, Practice, and Future Research Directions

We propose several policy, practice, and research implications as a result of our review. Our findings highlight that much more education about mental illnesses for the general public, and especially children, is essential. Enhanced education about the risks and potential support networks that can lower those risks could lead to addressing some of these misconceptions and fears children may have: It could pave the way for a realistic evaluation of their own risks, but also various means of dealing with these risks. According to the literature on the manifold effects of stigma on relatives of people with mental illness [e.g., (14, 16, 22)] and our study results, we can assume that stigma is a major factor contributing to the transgenerational transmission of mental disorders. Thus, at the level of social factors contributing to mental illness, stigma represents a target for prevention and intervention programs for the children of parents with a mental illness. This should be highlighted when considering stigma for this population, and could make an important difference in developing strategies to reduce stigma. Overall, stigma-reducing interventions, in terms of minimizing the public stigma of people with a mental illness, as well as encouraging more openness and acceptance by "normalizing" mental health problems, and empowering people to talk openly about mental illness, would be a further promising step to escape the vicious circle of mental illness stigma that contributes to the transgenerational transmission of mental illness.

The review has shown that adult health care services seem to be an important source of discrimination: It is not just the fact that children feel so ignored and insensitively treated by healthcare staff, but also the atmospheres that keep kids from feeling comfortable that should be addressed. Hospital and psychiatric staff for adult health care should undergo training in engaging with the children of parents with a mental illness [see also (77)]. Other support structures need to be established to lessen the pressure children feel that it is up to them to look after their parents.

Our findings emphasize the need for individual investigations to determine whether different aspects are important for specific family members. They might indeed differ more than previous research suggests, considering the homogeneity of the questionnaires used for all family members. As very few studies have provided evidence about which aspects influence how strongly children experience stigma, there is a need for researchers to investigate additional factors in more detail and across more types of parental mental illnesses. Future research should focus on a deeper differentiation of stigma dimensions and how they interact, and on their impact on the health and well-being of this specific population, which is worldwide, large, and carries such a high risk.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

L-MD designed and completed the study, wrote the protocol, was involved as the main person in charge in all analyses steps, and wrote the manuscript. JF did the independent full text screening as the second rater and was involved in the studies' inclusion process. DP did the data extraction as second rater and was involved in the studies' inclusion process. JLP did the quality assessment for qualitative studies as a second rater, supported the intersubjective validity by discussing the categories, and supported the writing of the manuscript by reviewing and editing. AB did the quality assessment for quantitative studies and the mixed method study as a second rater, supported the intersubjective validity by discussing the categories, and supported the writing of the manuscript by reviewing and editing. HC functions as PhD advisor and supervised the study and preparation of manuscript and supported finding agreement between two independent raters by discussing ambiguous study inclusions. All authors contributed to the article and approved the submitted version.

FUNDING

The paper was written as part of a 4-year research project the Village funded by the Austrian Federal Ministry of Health—Science and Research through the Open Innovation in Science Center at the Ludwig Boltzmann Gesellschaft GmbH, hosted at the Medical University of Innsbruck, with collaboration of Co-Investigator institutions. The funders did not influence the collection, analysis, or interpretation of data, and played no role in writing the manuscript.

ACKNOWLEDGMENTS

Authors would like to thank the whole Village team for their support during the entire process of creating and conducting this systematic review. Furthermore, authors would like to acknowledge the support of the librarian Susanne Waldmann of the Philipps University of Marburg in the systematic literature search. We would also like to acknowledge the contribution of Sara Evans-Lacko from the London School of Economics and Political Science in the compilation of the search strategy, as well as Feline Schelhammer, who supported the data management of the review within her student assistant job at the Philipps University of Marburg.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.813519/full#supplementary-material

REFERENCES

1. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands. I The scientific basis to a comprehensive approach. *Aust e-J Adv Mental Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
2. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin*

Psychiatry. (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427

3. Maybery DJ, Reupert AE, Patrick K, Goodyear M, Crase L. Prevalence of parental mental illness in Australian families. *Psychiatr bull*. (2009) 33:22–6. doi: 10.1192/pb.bp.107.018861
4. Radicke A, Barkmann C, Adema B, Daubmann A, Wegscheider K, Wiegand-Grefe S. Children of parents with a mental illness. Predictors of health-related quality of life and determinants of child-parent agreement. *Int J Environ Res Public Health*. (2021) 18:379. doi: 10.3390/ijerph18020379
5. Rasic D, Hajek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder, and major depressive disorder. A meta-analysis of family high-risk studies. *Schizophrenia Bull*. (2014) 40:28–38. doi: 10.1093/schbul/sbt114
6. Reupert A, Maybery D. What do we know about families where parents have a mental illness? *A systematic review. Child Youth Serv*. (2016) 37:98–111. doi: 10.1080/0145935X.2016.1104037
7. Christiansen H, Reck C, Zietlow A-L, Otto K, Steinmayr R, Wirthwein L, et al. Children of Mentally III Parents at Risk Evaluation (COMPARE): Design and Methods of a Randomized Controlled Multicenter Study-Part I. *Front Psychiatry*. (2019) 10:128. doi: 10.3389/fpsyt.2019.00128
8. Pryor JB, Reeder GD, Monroe AE. The infection of bad company. Stigma by association. *J Pers Social Psychol*. (2012) 102:224–41. doi: 10.1037/a0026270
9. Phelan JC, Bromet EJ, Link BG. Psychiatric illness and family stigma. *Schizophrenia Bull*. (1998) 24:115–26. doi: 10.1093/oxfordjournals.schbul.a033304
10. DeFleur ML. Stigma: notes on the management of spoiled identity. In: Erving Goffman, Englewood Cliffs. New Jersey: Prentice-Hall (1963). 147 pp. Cloth, $4.50; paper, $1.95. In: *Social Forces* 43:127–128. doi: 10.1093/sf/43.1.127
11. Goffman E. *Stigma. Notes on the management of spoiled identity. 26th pr*. New York: Simon and Schuster (A Touchstone Book) (1986).
12. Goffman E. *Stigma. Über Techniken der Bewältigung beschädigter Identität. 1.* Aufl. Frankfurt/M.: Suhrkamp (Suhrkamp Taschenbuch, 235) (1967).
13. Corrigan P, Watson AC, Miller FE. Blame, shame, and contamination. The impact of mental illness and drug dependence stigma on family members. *J Family Psychol*. (2006) 20:239–46. doi: 10.1037/0893-3200.20.2.239
14. Larson JE, Corrigan P. *The Stigma of Families with Mental* Illness (2008). doi: 10.1176/appi.ap.32.2.87
15. Phillips SD, Gates T. A conceptual framework for anderstanding the stigmatization of children of incarcerated parents. *J Child Fam Stud*. (2011) 20:286–94. doi: 10.1007/s10826-010-9391-6
16. Angermeyer MC, Schulze B, Dietrich S. Courtesy stigma—a focus group study of relatives of schizophrenia patients. *Soc Psychiatry Psychiatr Epidemiol*. (2003) 38:593–602. doi: 10.1007/s00127-003-0680-x
17. Wahl OF, Harman CR. Family views of stigma. *Schizophr Bull*. (1989) 15:131–9. doi: 10.1093/schbul/15.1.131
18. Fox AB, Earnshaw VA, Taverna EC, Vogt D. Conceptualizing and Measuring Mental Illness Stigma. The mental illness stigma framework and critical review of measures. *Stigma Health*. (2018) 3:348–76. doi: 10.1037/sah0000104
19. Pescosolido BA, Martin JK. The stigma complex. *Annu Rev Sociol*. (2015). 41:87–116. doi: 10.1146/annurev-soc-071312-145702
20. Mak WWS, Cheung RYM. Affiliate stigma among caregivers of people with intellectual disability or mental illness. *J Appl Res Intell Disabilities*. (2008) 21:532–45. doi: 10.1111/j.1468-3148.2008.00426.x
21. Bos AER, Pryor JB, Reeder GD, Stutterheim SE. Stigma. *Adv Theor Res Basic Appl Social Psychol*. (2013) 35:1–9. doi: 10.1080/01973533.2012.746147
22. van der Sanden RLM, Stutterheim SE, Pryor JB, Kok G, Bos AER. Coping with stigma by association and family burden among family members of people with mental illness. *J Nerv Ment Dis*. (2014) 202:710–7. doi: 10.1097/NMD.0000000000000189
23. Corrigan P, Kerr A, Knudsen L. The stigma of mental illness. Explanatory models and methods for change. *Appl Prevent Psychol*. (2005) 11:179–90. doi: 10.1016/j.appsy.2005.07.001
24. van Loon LMA, van de Ven MOM, van Doesum KTM, Hosman CMH, Witteman CLM. Parentification, stress, and problem behavior of adolescents who have a parent with mental health problems. *Fam Process*. (2017) 56:141–53. doi: 10.1111/famp.12165
25. Reupert A, Gladstone B, Helena Hine R, Yates S, McGaw V, Charles G, et al. Stigma in relation to families living with parental mental illness. An integrative review. *Int J Mental Health Nurs*. (2021) 30:6–26. doi: 10.1111/inm.12820
26. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferring reporting items for systematic reviews and meta analyses: the PRISMA statement. *J Clin Epidemiol*. (2009) 62:1006–12. doi: 10.1371/journal.pmed.1000097
27. Hong QN, Pluye P, Fabregues S, Bartlett G, Boardman F, Cargo M. et al. *Microsoft Word—MMAT_2018_criteria-manual_2018-08-08.docx* (2018).
28. Hong QN, Pluye P, Bujold M, Wassef M. Convergent and sequential synthesis designs. Implications for conducting and reporting systematic reviews of qualitative and quantitative evidence. *Syst Rev*. (2017) 6:61. doi: 10.1186/s13643-017-0454-2
29. Kuckartz U, Rädiker S. Datenaufbereitung and Datenbereinigung in der qualitativen Sozialforschung. Nina Baur and Jörg Blasius (Hg) Handbuch Methoden der empirischen Sozialforschung Wiesbaden. *Springer Fachmedien Wiesbaden:*(2019) 441–56. doi: 10.1007/978-3-658-21308-4_31
30. van der Sanden RLM, Bos AER, Stutterheim SE, Pryor JB, Kok G. Stigma by association among family members of people with a mental illness. A qualitative analysis. *J Community Appl Soc Psychol*. (2015) 25:400–17. doi: 10.1002/casp.2221
31. Cogan N, Riddell S, Mayes G. Children living with an affectively ill parent. How do they cope? *Educ Child Psychol*. (2005) 22:16–28.
32. Cogan N, Riddell S, Mayes G. The anderstanding and experiences of children affected by parental mental health problems. A qualitative study. *Qual Res Psychol*. (2005) 2:47–66. doi: 10.1191/1478088705qp024oa
33. Blakeman M, Martin C, Gupta A. Making sense of growing up with a parent with psychosis. An interpretative phenomenological analysis study. *Psychosis*. (2019) 11:54–62. doi: 10.1080/17522439.2019.1573916
34. Bolas H, van Wersch A, Flynn D. The well-being of young people who care for a dependent relative. An interpretative phenomenological analysis. *Psychol Health*. (2007) 22:829–50. doi: 10.1080/14768320601020154
35. Carroll L, Tuason MTG. Perpetually self-reflective. *Couns Psychol*. (2015) 43:1059–83. doi: 10.1177/0011000015602316
36. Dam K, Joensen DG, Hall EOC. Experiences of adults who as children lived with a parent experiencing mental illness in a small-scale society. A qualitative study. *J Psychiatric Mental Health Nurs*. (2018) 25:78–87. doi: 10.1111/jpm.12446
37. Davison J, Scott J. Should we intervene at stage 0? A qualitative study of attitudes of asymptomatic youth at increased risk of developing bipolar disorders and parents with established disease. *Early Interven Psychiatry*. (2018) 12:1112–9. doi: 10.1111/eip.12421
38. Haug Fjone H, Ytterhus B, Almvik A. How children with parents suffering from mental health distress search for 'Normality' and avoid stigma. *J Glob child Res*. (2009) 16:17.
39. Fudge E, Mason P. Consulting with young people about service guidelines relating to parental mental illness. *Aus e-J Adv Mental Health*. (2004) 3:50–8. doi: 10.5172/jamh.3.2.50
40. Griffiths J, Norris E, Stallard P, Matthews S. Living with parents with obsessive-compulsive disorder. Children's lives and experiences. *Psychol Psychother*. (2012) 85:68–82. doi: 10.1111/j.2044-8341.2011.02016.x
41. Kadish Y. Five women's recollections and reflections on being raised by a mother with psychosis. *S Afr J Psychol*. (2015) 45:480–94. doi: 10.1177/0081246315581565
42. Karnieli-Miller O, Perlick DA, Nelson A, Mattias K, Corrigan P, Roe D. Family members' of persons living with a serious mental illness. Experiences and efforts to cope with stigma. *J Mental Health*. (2013) 22:254–62. doi: 10.3109/09638237.2013.779368
43. Krupchanka D, Kruk N, Murray J, Davey S, Bezborodovs N, Winkler P, et al. Experience of stigma in private life of relatives of people diagnosed with schizophrenia in the Republic of Belarus. *Soc Psychiatry Psychiatr Epidemiol*. (2016) 51:757–65. doi: 10.1007/s00127-016-1190-y
44. Leahy MA. Children of mentally ill parents. Anderstanding the effects of childhood trauma as it pertains to the school setting. *Int J Educ Res*. (2015) 71:100–107. doi: 10.1016/j.ijer.2015.04.005
45. Leinonen A, Seikkula J, Alasuutari M. Can I tell just by myself? Discussing a parental mental disorder with a child in a research interview. *Int J Child, Youth Family Stud*. (2016) 7. doi: 10.18357/ijcyfs.71201615675

46. McCormack L, White S, Cuenca J. A fractured journey of growth. Making meaning of a 'Broken' childhood and parental mental ill-health. *Community, Work Family*. (2016) 20:327–45. doi: 10.1080/13668803.2015.1117418
47. Moore T, McArthur M, Noble-Carr D. Different but the same? Exploring the experiences of young people caring for a parent with an alcohol or other drug issue. *J Youth Stud.* (2010) 14:161–77. doi: 10.1080/13676261.2010.522561
48. Mordoch E, Hall WA. Children's perceptions of living with a parent with a mental illness. Finding the rhythm and maintaining the frame. *Qual Health Res.* (2008) 18:1127–44. doi: 10.1177/1049732308320775
49. Murphy G, Peters K, Wilkes L, Jackson D. Adult children of parents with mental illness. *Navigating stigma. Child Family Social Work.* (2015) 22:330–8. doi: 10.1111/cfs.12246
50. Nieto-Rucian V, Furness PJ. The experience of growing up with a parent with schizophrenia—A qualitative study. In: *Qualitative Psychology* (2017).
51. Oskouie F, Zeighami R, Joolaee S. Outcomes of parental mental illness on children. A qualitative study from Iran. *J Psychosocial Nurs Mental Health Serv.* (2011) 49:32–40. doi: 10.3928/02793695-20110802-06
52. Ostman M. Interviews with children of persons with a severe mental illness. Investigating their everyday situation. *Nordic J Psychiatry*. (2008) 62:354–9. doi: 10.1080/08039480801960065
53. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of Children of Parents With Mental Illness. A Necessity to Provide Relevant Support. *Front Psychiatry*. (2019) 9:358. doi: 10.3389/fpsyt.2018.00728
54. Stengler-Wenzke K, Trosbach J, Dietrich S, Angermeyer MC. Experience of stigmatization by relatives of patients with obsessive compulsive disorder. *Arch Psychiatr Nurs.* (2004) 18:88–96. doi: 10.1016/j.apnu.2004.03.004
55. Tabak I, Zabłocka-Zytka L, Ryan P, Poma SZ, Joronen K, Viganò G, et al. Needs, expectations and consequences for children growing up in a family where the parent has a mental illness. *Int J Ment Health Nurs.* (2016) 25:319–29. doi: 10.1111/inm.12194
56. Trondsen MV. Living with a mentally ill parent: exploring adolescents' experiences and perspectives. *Qual Health Res*. (2012) 22::174–88. doi: 10.1177/1049732311420736
57. Tamutiene I, Jogaite B. Disclosure of alcohol-related harm: children's experiences. *Nordisk Alkohol Narkotikatidskrift.* (2019) 36:209–22. doi: 10.1177/1455072518807789
58. Trondsen MV, Tjora A. Communal normalization in an online self-help group for adolescents with a mentally ill parent. *Qual Health Res.* (2014) 24:1407–17. doi: 10.1177/1049732314547708
59. Wahl P, Otto C, Lenz A. dann würde ich traurig werden, weil alle über meinen Papa reden"—Die Rolle des Stigmas in Familien mit psychisch erkranktem Elternteil. *Gesandheitswesen (Bandesverband der Arzte des Offentlichen Gesandheitsdienstes (Germany)).* (2017) 79:987–92. doi: 10.1055/s-0043-122234
60. Widemalm M, Hjärthag F. The forum as a friend. Parental mental illness and communication on open Internet forums. *Social Psychiatry Psychiatric Epidemiol.* (2015) 50:1601–7. doi: 10.1007/s00127-015-1036-z
61. Haverfield MC, Theiss JA. Parent's alcoholism severity and family topic avoidance about alcohol as predictors of perceived stigma among adult children of alcoholics: Implications for emotional and psychological resilience. *Health Commun.* (2016) 31:606–16. doi: 10.1080/10410236.2014.981665
62. Gray B, Robinson C, Seddon D. Invisible children. Young Carers of Parents with Mental Health Problems—The Perspectives of Professionals. *Child Adolesc Mental Health.* (2008) 13:169–72. doi: 10.1111/j.1475-3588.2007.00477.x
63. Rezayat F, Mohammadi E, Fallahi-Khoshknab M, Sharifi V. Experience and the meaning of stigma in patients with schizophrenia spectrum disorders and their families. A qualitative study.*Jpn J Nurs Sci.* (2019) 16:62–70. doi: 10.1111/jjns.12212
64. Haug Fjone H, Ytterhus B, Almvik A. How children with parents suffering from mental health distress search for 'normality' and avoid stigma. *Childhood.* (2009) 16:461–77. doi: 10.1177/0907568209343743
65. Rezayat F, Mohammadi E, Fallahi-Khoshknab M. The process of responding to stigma in people with schizophrenia spectrum disorders and families. A groanded theory study. *Perspect Psychiatric Care.* (2019) 56:564–73. doi: 10.1111/ppc.12468
66. CORRIGAN PW, Markowitz FE, Watson AC. Structural levels of mental illness stigma and discrimination. *Schizophr Bull.* (2004) 30:481–91. doi: 10.1093/oxfordjournals.schbul.a007096
67. Schomerus G, Angermeyer MC. Stigma and its impact on help-seeking for mental disorders. What do we know? *Epidemiologia e psichiatria sociale.* (2008) 17:31–7. doi: 10.1017/S1121189X00002669
68. van der Sanden RLM, Bos AER, Stutterheim SE, Pryor JB, Kok G. Experiences of stigma by association among family members of people with mental illness. *Rehabil Psychol.* (2013) 58:73–80. doi: 10.1037/a0031752
69. Link BG, Phelan JC. Conceptualizing stigma. *Annu Rev Sociol.* (2001) 27:363–85. doi: 10.1146/annurev.soc.27.1.363
70. Havighurst RJ. *Developmental Tasks and Education.* 3rd ed. New York, NY: McKay and Company (1972).
71. Park S, Park KS. Family Stigma. A Concept Analysis. *Asian Nurs Res.* (2014) 8:165–71. doi: 10.1016/j.anr.2014.02.006
72. van der Sanden RLM, Pryor JB, Stutterheim SE, Kok G, Bos AER. Stigma by association and family burden among family members of people with mental illness. The mediating role of coping. *Soc Psychiatry Psychiatr Epidemiol.* (2016) 51:1233–45. doi: 10.1007/s00127-016-1256-x
73. Ostman M, Kjellin L. Stigma by association. Psychological factors in relatives of people with mental illness. *Br J Psychiatry*. (2002) 181:494–8. doi: 10.1192/bjp.181.6.494
74. Riebschleger J, Tableman B, Rudder D, Onaga E, Whalen P. Early outcomes of a pilot psychoeducation group intervention for children of a parent with a psychiatric illness. *Psychiatr Rehabil J.* (2009) 33:133–41. doi: 10.2975/33.2.2009.133.141
75. Reupert AE, Maybery J, Darryl K, Kowalenko NM. Children whose parents have a mental illness. Prevalence, need and treatment. *MJA Open*. (2012) 1:7–9. doi: 10.5694/mjao11.11200
76. Coughlin SS. Recall bias in epidemiologic studies. *J Clin Epidemiol.* (1990) 43:87–91. doi: 10.1016/0895-4356(90)90060-3
77. Bauer A, Best S, Malley J, Christiansen H, Goodyear M, Zechmeister-Koss I, et al. Towards a program theory for family-focused practice in adult mental health care settings. an international interview study with program leaders. *Front Psychiatry*. (2021) 12:741225. doi: 10.3389/fpsyt.2021.741225

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 22 February 2022
doi: 10.3389/fpsyt.2022.791294

The Triple P Positive Parenting Program for Parents With Psychosis: A Case Series With Qualitative Evaluation

Lauren Wolfenden[1], Rachel Calam[1], Richard J. Drake[1,2] and Lynsey Gregg[1]*

[1] Division of Psychology and Mental Health, School of Health Sciences, Faculty of Biology, Medicine and Health, Manchester Academic Health Science Centre, The University of Manchester, Manchester, United Kingdom, [2] Greater Manchester Mental Health NHS Foundation Trust, Manchester, United Kingdom

OPEN ACCESS

Edited by:
Joanne Nicholson,
Brandeis University, United States

Reviewed by:
Laelia Benoit,
Yale University, United States
Dirk Richter,
Universitäre Psychiatrische Dienste
Bern, Switzerland

***Correspondence:**
Lynsey Gregg
lynsey.gregg@manchester.ac.uk

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: 08 October 2021
Accepted: 11 January 2022
Published: 22 February 2022

Citation:
Wolfenden L, Calam R, Drake RJ and Gregg L (2022) The Triple P Positive Parenting Program for Parents With Psychosis: A Case Series With Qualitative Evaluation.
Front. Psychiatry 13:791294.
doi: 10.3389/fpsyt.2022.791294

Although many people with psychosis are parents, managing the dual demands of poor mental health and parenting can be stressful and may contribute to poorer outcomes for both parent and child. Parenting interventions have the potential to improve outcomes for the whole family but need evaluation of feasibility in this context. The Triple-P Self-Help Workbook was implemented with guidance and support with 10 parents experiencing psychosis in a multiple baseline case series study. Sessions were weekly and home-based. Outcome measures examined facets of parenting, child behavior, self-efficacy and parental mental health. Follow up interviews explored parents' perspectives of the perceived impact of the intervention and apparent mechanisms of change. The program resulted in clinically significant change (>25% improvement) in mental health, parenting and child behavior measures post-intervention for the 50% who completed all 10 sessions and improvements were maintained at 3 and 6 month follow up. Interviews with those who completed the program revealed it to have been transformative: parents reported positive changes in parenting style; they were empowered with regard to their parenting and had a greater sense of control over their mental health. This study provides preliminary evidence that self-directed Triple P might be able to reduce the symptoms of psychosis by improving family functioning. Findings could inform the future development or adaptation of evidence-based parenting interventions for parents with psychosis in order to improve their mental health, aid recovery, and intervene early in the lives of children at risk of poor long-term outcomes.

Keywords: parental mental health, parenting, schizophrenia, SMI, parent-child interaction

INTRODUCTION

Serious mental illness (SMI), such as psychosis, can be debilitating and interfere with social, emotional and psychological functioning. The chronicity and severity of symptoms experienced in psychosis can have multifaceted and debilitating implications for daily life (1). Without support, the impact on mood, relationships and quality of life can be profound (2). Such challenges can be further exacerbated when people with psychosis also have dependent children with elevated emotional reactivity to stress, making parenting a particularly stressful aspect of their lives. At the same time, the parental role can create meaning, belonging and increase self-worth and as such, is an important part of self-identity for both men and women with psychosis (3, 4) and may be an important focus for recovery (5, 6).

It has been estimated that up to 55% of men and 62% of women experiencing psychosis are parents (7) but current treatments may neglect the challenges experienced by these parents, particularly mothers (4).

The link between parental SMI and reduced quality parent-child interactions, poor attachments and limited sensitivity is well established (8–11) and the influence of parental factors in the development, maintenance and amelioration of disruptive behaviors and psychological health of children has been evidenced within practice and comprehensive reviews (12–16).

However, experiencing SMI does not need to impede the ability to parent effectively (17, 18); interventions that target parenting and child behaviors and parental mental health have potential to prevent long-term adverse consequences for families and should be prioritized (14, 19–21). The use of evidence-based parenting interventions is recommended by the National Institute for Health and Clinical Excellence to improve child behavioral problems and reduce intergenerational cycles of poor mental health (22) however, there remains a lack of appropriate and timely parenting interventions to support parents experiencing SMI, and in particular, psychosis.

One widely used parenting intervention is the Triple P-Positive Parenting Program (23, 24). Based on social learning theory and cognitive behavioral principles, it aims to improve confidence in the parenting role and modify maladaptive parenting behaviors (25). The self-directed variant also targets coercive family interactions and offers skill acquisition and problem solving (26, 27). Large scale trials of the self-help variant have demonstrated positive outcomes for both parents and children similar to that of standard parenting interventions (24, 28, 29) including with parents experiencing bipolar disorder (30, 31).

To date, no study has evaluated the use of self-help Triple P with parents experiencing psychosis. The aim of the current study was to ascertain whether the use of this intervention in parents' homes was feasible and acceptable and whether there were any clinical effects for parents or children during and upon completion of the intervention in terms of child behavior and parental mental health. A single case design with multiple participants was used in order to capture the impact of the intervention on each individual participant rather than obtain an aggregate group effect which was not concerned with individual experience.

METHODS

Design

A within-subject A-B-A single case design across participants with follow-up was implemented. In this design, which employs a multiple baseline phase, participants act as their own controls. Following the baseline period (A), the ten sessions of the intervention (B) were delivered weekly over 10–14 weeks. After the intervention, participants were followed up at 3 and 6 months (A). The intervention was initiated only if the repeated measurements at baseline were stable, or else the baseline phase was increased. The repeated assessment of dependent variables during all three phases allows for the dependent variables to be assessed prior the implementation of the intervention and confidence is increased that any observable changes are attributable to the intervention rather than alternative explanations.

Qualitative methodology was employed to generate knowledge surrounding implementation, usefulness and perceived change based on parents' personal accounts of engaging in the program. Study procedures were registered prior to recruitment (clincaltrials.gov: NCT02622048).

Participants

Participants were eligible to take part if they met the following criteria: (1) ability to provide informed consent; (2) English speaking; (3) diagnosed with Schizophrenia Spectrum Disorder; (4) parent of a child aged 3–10 years old with whom they had more than 10 h of face to face contact per week; (5) over 18 years old; (6) medication stable; and (7) no change in care plans and no other parenting support being received. Case note review corroborated diagnosis or symptoms and was additionally considered by a psychiatrist (author 3) using the International Classification of Diseases (ICD) version 10 (32). Participants were referred from Early Intervention Services (EISs), Community Mental Health Teams (CMHTs) and local authority services, including local council family and housing teams.

Data Collection and Evaluation

Data were collected for parents who took part in the intervention to monitor feasibility, change over time and acceptability. Delivery adaptations were also recorded. In line with process evaluation planning (33–35), 11 key areas were highlighted as priorities: recruitment, maintenance, context, resources, implementation, reach, barriers, exposure, initial use, continued use and contamination.

Primary outcomes included attrition rate monitoring, proportion of data points completed and acceptability. In line with the MRC Framework, mechanisms of impact were explored using the client satisfaction questionnaire to provide a quantified measure of perceived acceptability, usefulness and practicality. Qualitative interviews were also undertaken following completion of the intervention. Secondary outcome measures consisted of weekly (continuous) measurement of symptoms, mood, parental efficacy and child behaviors. Additional outcome measures assessed social functioning, parenting practices and family relationships at the start and end of baseline phases and at the end of the intervention. During such preliminary research, incorporating a range of outcome measures at different time points can help to ascertain how participants interpret each measure and understand perceived usefulness.

Measures

Well-established and validated semi-structured interview schedules were employed to assess symptoms and functioning: The Positive and Negative Syndrome Scale [PANSS; (36)], Psychotic Symptoms Rating Scale [PSYRATS; (37)] and the

Personal and Social Performance Scale [PSP; (38)] were used to assess psychopathology and functioning. The self-report Depression, Anxiety and Stress Short Form Scale [DASS-21; (39)] was used to determine parental mood and stress levels. Parenting and child behaviors were explored using a range of measures: the Parenting Task Checklist [PTC, (40)] assessed parental self-efficacy, the Eyberg Child behavior Inventory [ECBI, (41)] assessed intensity and frequency of problematic child behaviors and the Parenting Scale [PS, (42)] assessed a range of parenting behaviors. Additionally, the Family Background Questionnaire [FBQ, (43)] was used to collect demographic and psychosocial information. The Client Satisfaction Questionnaire [CSQ; (24)] assessed parents' thoughts and beliefs regarding the acceptability and effectiveness of the parenting intervention for themselves, their family and their child.

Procedure

Following referral from the healthcare professionals working with parents, parents who met the inclusion criteria were given a written participant information sheet (PIS) to read before participating in an initial visit with the researcher. At this visit study processes and the nature of the work book were explained and discussed. If literacy problems were noted, the researcher read the PIS aloud before giving parents the opportunity to ask questions. At a second visit, at least 48 h after the first, informed consent was obtained. Participants with more than one child were asked to identify a target child with whom they experienced the most difficulties. Data were recorded for this child only.

Participants were monitored and assessed using a multiple baseline approach (A), during weekly home-visits over the 10-week intervention (B), repeat of baseline (A) and at three- and 6-month follow-up. The first baseline phase acted as an engagement opportunity and built rapport with participants; this ensured safety and trust, and facilitated commitment to the program. Following the engagement phase, the intervention began and weekly symptom monitoring occurred. Changes to mental health, current parenting behaviors and child behavior were measured using the PANSS, DASS-21, ECBI and PTC. In addition to measures used during the pre-and post-intervention multiple baseline phases (A) the PSYRATS, PSP and the PS were used during weekly monitoring.

Typically, baseline visits lasted 45–60 min over a minimum of three sessions. The baseline phase controlled for potential confounds, ensuring any change could more likely be attributed to participation in the program (44, 45). The intervention phase consisted of a minimum of 10 weekly visits lasting 1.5 h.

Follow up interviews took place in participants' homes, where a flexible interview schedule consisting of open-ended questions was used to explore experiences of the program in two broad domains: (i) experiences of taking part in the program in relation to self, child and parent-child relationship; and (ii) perceived intervention appropriateness and effectiveness. Interviews were audio recorded and transcribed verbatim. Duration varied between 46–57 min (mean = 52 min).

The Triple P Positive Parenting Program

The self-directed variant of the Triple P Positive Parenting Program was used in its manualized "Every Parent"s Self-Help Workbook' format. The workbook aims to promote self-sufficiency and independence to achieve sustainable behavior change over 10 weeks. The work book focusses on 17 core strategies; 10 of which are designed to promote child development (i.e., attention through quality time and talking; affection; praise; engaging activities; incidental teaching; parent as role model; and daily/weekly behavior monitoring charts). The final seven encourage parents to actively manage misbehavior (i.e., appropriate rule setting and boundaries; directed discussion and instructions; logical consequences, quiet-time and time-out) (26). The skills acquired aim to help parents to form clear plans that can be used at home and in the community to respond well in situations that become difficult or "high risk".

Sessional role plays to practice learnt techniques were used to reinforce the development of self-evaluation and problem-solving capabilities. Parents were aided to explore and understand their child's needs and causes of behavior, emotional and social problems. Such tasks aimed to enable participants to acquire skills to aid their awareness of appropriate expectations and child development. These were deemed low level adaptations that did not alter the intervention (46). All sessions and assessments occurred in the client's home and were conducted by the first author.

Due to complex family circumstances and readiness levels, a flexible, parent-led approach was used throughout. Additional support was offered in the form of (a) help reading the workbook, ensuring parents understood the content and examples used and (b) conversations linking mental health to parenting. This facilitated engagement, problem solving, and implementation of strategies within the workbook. The conversations linking mental health to parenting helped participants who were not already doing so make links between their parenting and mental health. These conversations took place after the outcome measures were assessed each week. Participant's responses to the self-reported mental health, parenting and child behavior measures were discussed in relation to the strategies in the workbook the parent was attempting to develop. In this way, parents were encouraged to begin to recognize their triggers, their child's triggers and also their strengths rather than deficits.

Potential facilitators and barriers to implementation were identified within baseline sessions. In this phase parenting strengths, struggles and areas of desired change were identified and fed into the intervention to ensure goals were individual to participants' needs. Participants were sent text messages the day before and the day of each session to remind them of the upcoming session and ensure tasks had been completed beforehand.

Contextual Factors: Guided Self-Help Adaptations

There were high rates of challenging circumstances for each family, such as: literacy problems, family conflicts and disruptions, social isolation, fear, poverty and financial stressors. Therefore, during the initial baseline sessions time was spent collaboratively identifying parents' strengths and struggles. More than half experienced literacy problems (60%), some reported that they had never actively read a book before (30%), and most had never read for pleasure (80%). It was therefore essential to make minor adaptations to the delivery of the self-directed workbook and time spent completing practical exercises. Participants required assistance to understand tasks and required support and guidance when planning and implementing the strategies. This was deemed "low risk" because it did not change the core elements of the intervention or the measures used (47).

Data Analysis

SPSS 25.0 was used for all statistical analyses. Visual inspection of graphical data, percentage change calculations of outcome measures and descriptive statistics were used to ascertain the acceptability and feasibility of the intervention. Means were derived from measurements at each time point. Data were graphically represented for weekly measurements to explore change over time. There are numerous established ways to calculate clinical significance which produce similar outcomes (48). We used the Reliable Change Index approach to calculate clinically significant change (49). Clinically significant change was defined as > 25% reduction from pre to post intervention (50). A threshold of 25% change was classified as a "moderate outcome" and > 50% classified as a "good outcome" based on previous studies using the PANSS and PSYRATS (51, 52). For participants who elected to finish using the workbook before session 10, weekly scores to that point were used to monitor change.

End of program interview transcripts were analyzed using a framework analysis approach (53) in which the interview topic guide was used to provide an initial framework for the analysis. Coding was also inductive allowing for the expansion or collapsing of initial themes and categories before production of the final classification framework. To preserve anonymity participants' names were replaced with a pseudonym and names of children were replaced with "X".

RESULTS

Participant Characteristics

All participants were experiencing psychosis and were mothers of at least one child aged between 3–10 years old. **Table 1** provides a breakdown of participant characteristics and family circumstances.

Participants were white British (8, 80%), Black African (1, 10%) and Chinese (1, 10%). They were mainly single parents (9, 90%) who had a diagnosis of schizophrenia (6, 60%) or paranoid schizophrenia (4, 40%). One participant had completed higher education (10%), qualifications were otherwise at high school (2, 20%) or sixth-form level (7, 70%). The majority (90%) were unemployed and all reported that they were struggling financially with either "just enough to meet essential expenses only" (3, 30%) or "unable to meet essential expenses" (7, 70%).

Feasibility

Enrolment

Recruitment was successful: a total of 19 parents referred to the study were eligible to take part and all expressed an interest in participating, however only 11 progressed to the initial baseline phase. No participants declined participation. The CONSORT diagram is presented in **Figure 1**.

Attendance and Attrition Rates: Uptake of Intervention

The maximum number of visits from baseline to follow-up was 23 per participant, 230 in total across the sample. Of these 171 (74%) were attended. Four participants attended all possible sessions, one attended 91%, three attended 52% and two attended 48%.

Of the 11 parents who progressed to the initial baseline phase one did not complete baseline due to more than five missed appointments. Ten completed the baseline phase and progressed to the intervention. Of these, five completed all

TABLE 1 | Participant and family characteristics.

	Parent details							Target child details	Completed programme
	Sex	**Age**	**Diagnosis**	**Ethnicity**	**Marital status**	**Employment**	**Number of children**		
Participant 1	Female	33	Schizophrenia	White British	Single	Unemployed	3	Male, 7	No
Participant 2	Female	36	Schizophrenia	White British	Single	Unemployed	3	Female, 10	No
Participant 3	Female	26	Schizophrenia	White British	Single	Unemployed	4	Male, 10	No
Participant 4	Female	48	Schizophrenia	White British	Single	Unemployed	2	Male, 9	Yes
Participant 5	Female	25	Schizophrenia	White British	Single	Unemployed	2	Male, 4	Yes
Participant 6	Female	28	Schizophrenia	Black African	Single	Unemployed	2	Male, 9	Yes
Participant 7	Female	33	Schizophrenia	White British	Single	Working part time	2	Female, 8	No
Participant 8	Female	27	Schizophrenia	White British	Single	Unemployed	2	Male, 6	Yes
Participant 9	Female	40	Schizophrenia	White British	Cohabiting	Unemployed	5	Female, 8	Yes
Participant 10	Female	33	Schizophrenia	Chinese	Single	Unemployed	1	Male, 9	No

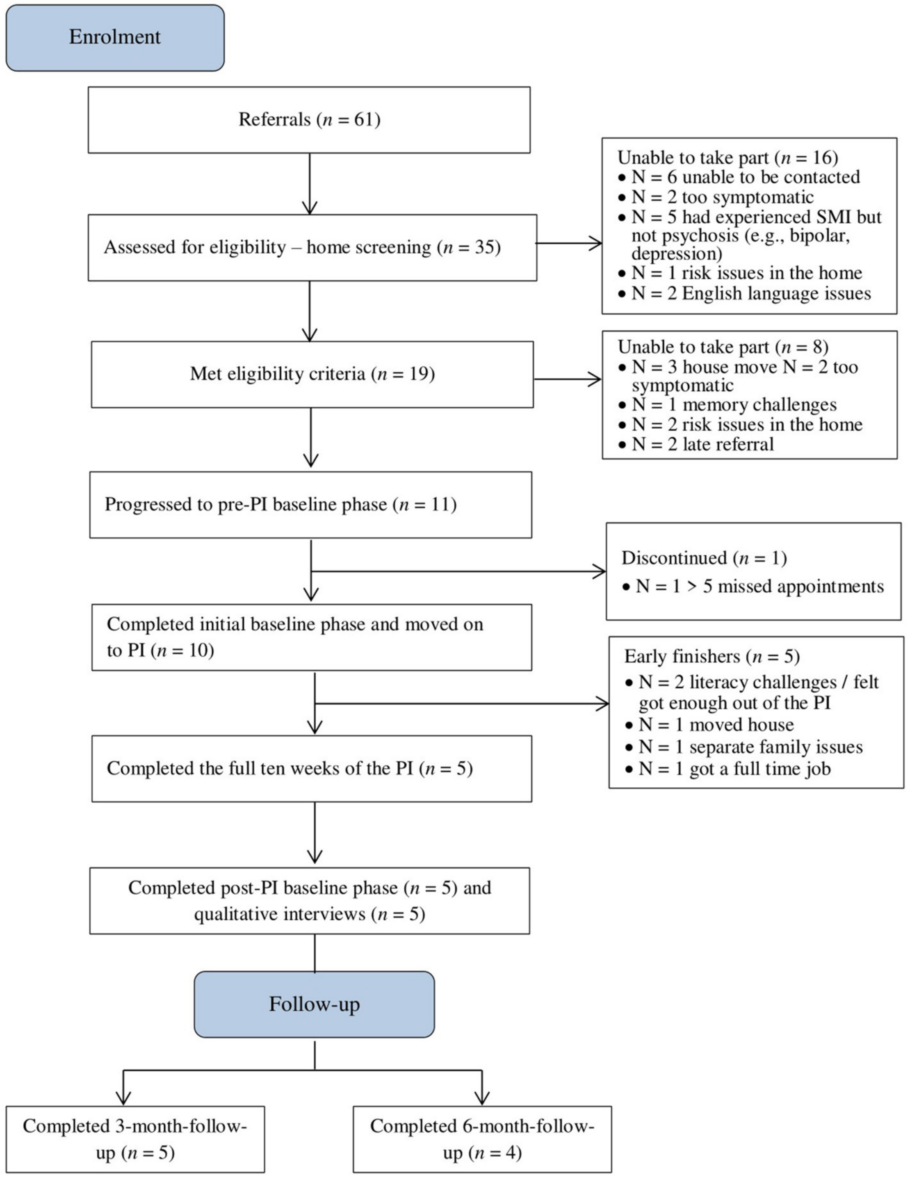


FIGURE 1 | CONSORT diagram showing participant flow through the study.

10 weeks of the workbook, final baseline phase and 3-month-follow-up. Four of these completed the 6-month-follow-up. The remaining five partially completed the workbook, completing to either week 4 or week 6. The five participants who did not complete it discontinued for practical reasons (a house move; new employment and challenges with another child in the family, $N = 3$) or because they found the workbook challenging due to poor literacy ($N = 2$).

All parents completed the first half of the workbook which contains the core strategies which promote positive parenting practices, self-monitoring strategies, stress reduction, and management of disruptive child behaviors. The five parents who finished early failed to cover troubleshooting, practice strategies, and identifying future challenges in high risk situations. No adverse events, hospital admissions or worsening of symptoms were reported post-intervention or at follow-up visits.

Acceptability Ratings

Overall, the intervention was rated as highly acceptable by eight participants with an average score of 95% (range: 76–100%) on the client satisfaction questionnaire. It was rated as 87% (range 71–100%) "Useful and informative", 89% (range 71–100%) "Interesting" and 79% (range 57–100%) "Practical".

Clinical Outcomes

Analyses of the effects of taking part in the intervention on mental health and parenting measures are summarized in **Table 2**. Tests of significance (t-tests) and effect sizes (Cohen's d)

TABLE 2 | Pre and post intervention measures.

Measures	Pre PI baseline mean (SD)	End of PI mean (SD)	Post PI baseline mean (SD)	Follow-up 3 month (SD)	Follow-up 6 month (SD)	Pre to post PI			Pre to follow-up 3 months			Pre to follow-up 6 months		
	(*n* = 10)	**(*n* = 5)**	**(*n* = 5)**	**(*n* = 5)**	**(*n* = 4)**	***t***	***p***	***d***	***t***	***p***	***d***	***t***	***p***	***d***
Parenting and child behavior measures														
PTC behavior	49.7 (18.78)	89.2 (14.33)	98.26 (2.21)	98.4 (1.88)	98.84 (2.1)	9.5	0.001	2.49	12.1	0.001	4.08	−8.9	0.003	4.24
PTC setting	52 (22.96)	89.6 (14.1)	98.62 (1.9)	97.7 (3.9)	97.6 (3.9)	8.2	0.001	2.09	12.8	0.001	3.1	9.6	0.002	3.19
PS total	4.06 (0.77)	2.33 (0.622)	1.75 (0.81)	2.15 (0.45)	2.01 (0.67)	5.88	0.004	2.76	10.12	0.001	3.39	8.9	0.003	3.28
ECBI intensity	69.4 (12.6)	46.6 (6.7)	46 (4.7)	45.8 (4.6)	43.2 (3.6)	8.5	0.001	2.38	6.04	0.003	2.78	5	0.01	3.26
ECBI problem	70.3 (10.6)	47.6 (6.55)	45.2 (3.4)	42.4 (1.67)	41.2 (0.5)	7.5	0.001	2.71	8.9	0.001	4.11	7.11	0.005	4.47
Mental health measures														
PSYRATS hallucination	16.1 (14.7)	6.3 (9.2)	3.81 (7.01)	5.4 (7.6)	5.3 (6.7)	2.79	0.021	0.82	3.23	0.032	0.96	5.78	0.010	1.0
PSYRATS delusions	14.5 (3.8)	7.8 (5.32)	5.03 (3.01)	4.0 (2.91)	7.65 (2.3)	5.30	0.000	1.20	6.45	0.003	3.13	9.02	0.003	2.24
DASS-21 total	81.7 (35.26)	38 (32.37)	30.5 (33.4)	32 (36.9)	38 (41.6)	4.13	0.003	1.36	2.3	0.083	1.54	1.5	0.230	1.26
WEMWBS	31 (2.89)	52 (6.16)	47 (16)	53 (9.08)	57 (8.04)	6.16	0.004	4.88	4.5	0.011	3.65	5.5	0.012	5.17
PSP	46.8 (6.68)	62.4 (2.4)	63.2 (4.86)	63.4 (4.7)	65.5 (4.1)	5.24	0.006	3.43	6.12	0.004	2.91	4.52	0.020	3.46
PANSS positive	18 (5.6)	14.6 (3.78)	12.2 (4.54)	13.4 (4.39)	14.5 (3.87)	1.14	0.319	0.72	1.65	0.174	0.92	1.13	0.34	0.74
PANSS negative	13.4 (2.04)	10.6 (0.89)	9.2 (2.86)	10.2 (1.78)	10.0 (2.7)	2.44	0.071	1.9	2.02	0.114	1.68	1.43	0.247	1.43
PANSS general	33.5 (9.28)	26.6 (2.96)	23.4 (6.9)	25.6 (2.9)	25.5 (3.3)	1.59	0.186	1.13	1.99	0.117	1.3	1.73	0.182	1.27
PANSS total	64.8 (15.7)	51.8 (6.6)	44.8 (13.4)	49.2 (8.3)	50 (7.4)	1.61	0.182	1.16	2.01	0.115	1.3	1.50	0.230	1.28

are presented. Graphical representations of weekly mental health outcomes can be found in **Figures 2A,B**, and weekly parenting and parent-child outcomes can be found in **Figures 3A–D**.

Parental Mental Health

All participants experienced some reductions in symptom severity according to the PANSS (positive, negative and general subscales and overall total). This change was not statistically significant on the PANSS subscales but four of the five participants who completed 100% of the 10 week program achieved clinically significant reductions in PANSS total scores. At post-intervention symptoms were reduced by 25–44% from baseline for all participants. At the three-month-follow-up, clinically significant reductions were reported for all participants from baseline ranged from 29 to 39% and from 36 to 45% at 6 month-follow-up.

There were statistically significant improvements in both the hallucinations and delusions dimensions according to the PSYRATS. These improvements were maintained at both follow-up time points. Clinically significant change of >25% was observed for five of the six participants experiencing hallucinations and 9 of 10 participants experiencing delusions. For hallucinations, reduction in symptoms ranged from 66 to 100% post-intervention compared to baseline, from 47 to 100% at 3-month-follow-up and from 53 to 100% at 6-month-follow-up. For delusions, change ranged from 25 to 100% post-intervention compared to baseline, from 49 to 100% at 3-month-follow-up and 37–64% at 6-month-follow-up. Participants 1, 2, 4, 5 and 6 showed significant reductions in hallucinations frequency, severity and levels of distress. Participant three was no longer experiencing hallucinations or delusions. Graphical representations can be seen in **Figure 2.**

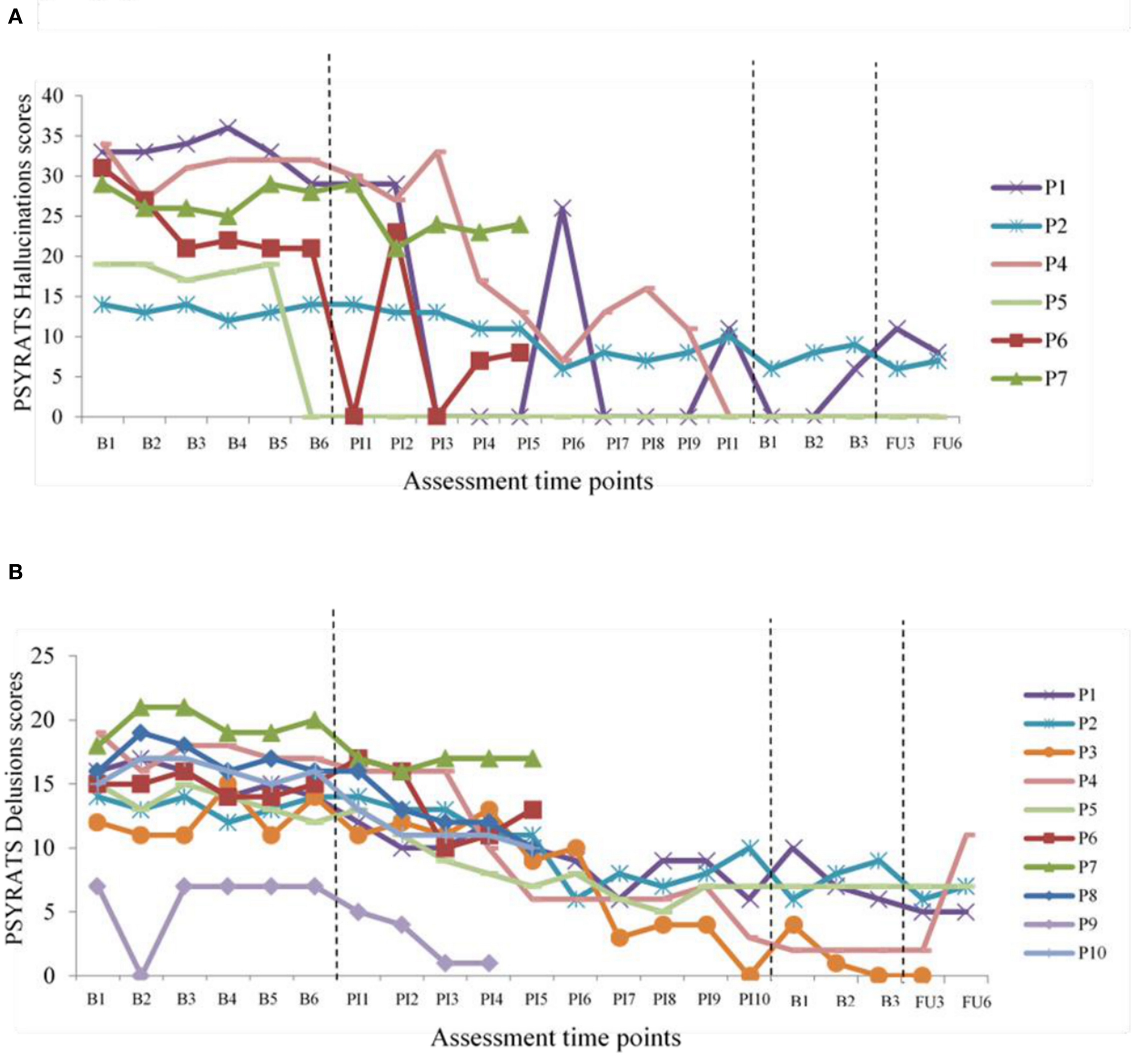


FIGURE 2 | Changes in PSYRATS Hallucinations and Delusions. **(A)** PSYRATS Hallucination scores over time. **(B)** PSYRATS Delusions scores over time.

There were significant improvements in depression, anxiety and stress (DASS-21) pre to post intervention. However, this change was not maintained at follow-up. At baseline most participants fell into "severe" or "extremely severe" categories for depression (60%), anxiety (80%) and stress (80%). All scores improved and moved into "normal", "mild" and "moderate" categories at completion, with 20–40% scoring zero across subscales. Percentage change improvements of >25% were observed for nine out of 10 participants on subscale and total scores at all time points. Change ranged from 27 to 100% post-intervention or final session (chosen end point) compared to baseline, and from 55 to 86% at follow-up. Graphical representations can be seen in **Figure 2**.

There were significant improvements in social functioning (PSP) post-intervention. These changes were maintained at both follow-up time points. At baseline, eight out of 10 participants were categorized as having "marked" or "severe" poor social functioning and two out of 10 into "marked" or "manifest" difficulties. Social functioning scores showed improvements at all-time points for all participants. Social functioning improved post-intervention and categorized participants as "mild" and "manifest". Percentage change improvements of >25% was observed for three out of five participants who completed the program at all-time points. Changes ranged from 27 to 63% post-intervention compared to baseline, from 25 to 63% at 3-month-follow-up, and 35–78% at 6-month-follow-up. Participant 3 and 5 showed small improvements across all time points. This suggests improvements across facets of relationships, activities, self-care and aggressive behaviors.

Parenting

There were significant improvements in parenting on all PS subscales and total scores. These improvements were maintained at both follow-up time points. At baseline, eight out of 10 participants were within the clinical range expressing difficulties in their parenting role. Following the intervention, all participants fell below the clinical cut-off (3.2) and were within the normal range for parenting difficulties. Percentage change improvements >25% were reported for all five participants on PS total post-intervention compared to pre-intervention and at follow-up, and subscale improvements ranged from 18to 68%. PS total score improvements ranged from 28 to 54% post-intervention compared to baseline, from 41 to 53% at 3-month-follow-up and 40 to 58% at 6-month-follow-up.

Parental confidence also improved according to PTC behavior and setting subscales. These improvements were maintained at both follow-up time points. For PTC behavior subscale, improvements ranged from 58 to 142% post-intervention compared to baseline, from 59 to 115% at 3-month-follow-up and 59–121% at 6-month-follow-up. For PTC setting subscale, improvements ranged from 43 to 173% post-intervention compared to baseline, 50–94% at 3-month-follow-up and 55–97% at 6-month-follow-up. These improvements demonstrated change in parental self-efficacy in most settings and when dealing with most child behaviors following the intervention. Graphical representations can be seen in **Figure 3**.

Child Behavior

There were significant improvements in child behavior according to ECBI intensity and problem subscales. These improvements were maintained at both follow-up time points. At baseline, eight out of 10 participants fell into the 'clinical' range for intensity scores (>131) and nine out of 10 on problem scores (>15). Following the intervention, participants who completed T1 and T2 were no longer scoring in the clinical category for child behavior problems or parental distress. Child behavior improvements of >25% was observed for nine out of 10 participants on the intensity subscale and all participants on the problem subscale post-intervention or chosen end point compared to baseline. For ECBI-intensity subscale, child behavior improvements ranged from 31%-59% post-intervention compared to baseline, from 38 to 61% at 3-month-follow-up and 42–55% at 6-month-follow-up. For ECBI-problem subscale, child behavior improvements ranged from 58 to 100% post-intervention compared to baseline, from 90 to 100% at 3-month-follow-up and 97–100% at 6-month-follow-up. Graphical representations can be seen in **Figure 3**.

End of Study Qualitative Evaluation

The qualitative interviews aimed to uncover parents' experiences of the program, identifying aspects they found valuable or difficult in order to inform future implementation. In an attempt to uncover mechanisms of change we asked parents to reflect on their experiences of parenting prior to starting the program, and describe any positive impacts during and since its completion. The analytical framework organized the data into two main themes: The first theme describes the family's journey (parenting prior to the program, their expectations of the program and finally, program outcomes) and the second theme details the aspects of the program they found most valuable and any suggestions parents made for adaptations that they felt might be beneficial for other parents in in the future.

Theme 1: The Family's Journey

Parenting Prior to the Program

Parents described themselves variously as "hopeless", "lost" and a "bad parent" before the program. One parent went as far as to say "I wasn't a Mum". All five felt they were failing in the parental role and reported poor relationships with their children. Parental accounts highlighted a lack of control. They described cycles of behavior in which children's behavior was exacerbated via conflict between the parent and child, with "screaming and shouting" the norm. Attempts at control were sometimes futile: "everything I tried to do backfired in my face" (participant 9) and parents sometimes chose the "easy option" of not responding to challenging behavior and giving in "for a quiet life" despite recognizing that this could serve to worsen behavior.

"that's probably resulted in his difficult behavior cause he knows that it's all right, I'll just kick off get emotional and my mum will just end up getting it me, she won't follow through with it" (participant 6)

It was clear that self-efficacy for all five parents was low and this was also reflected in the quality of parent-child interactions.

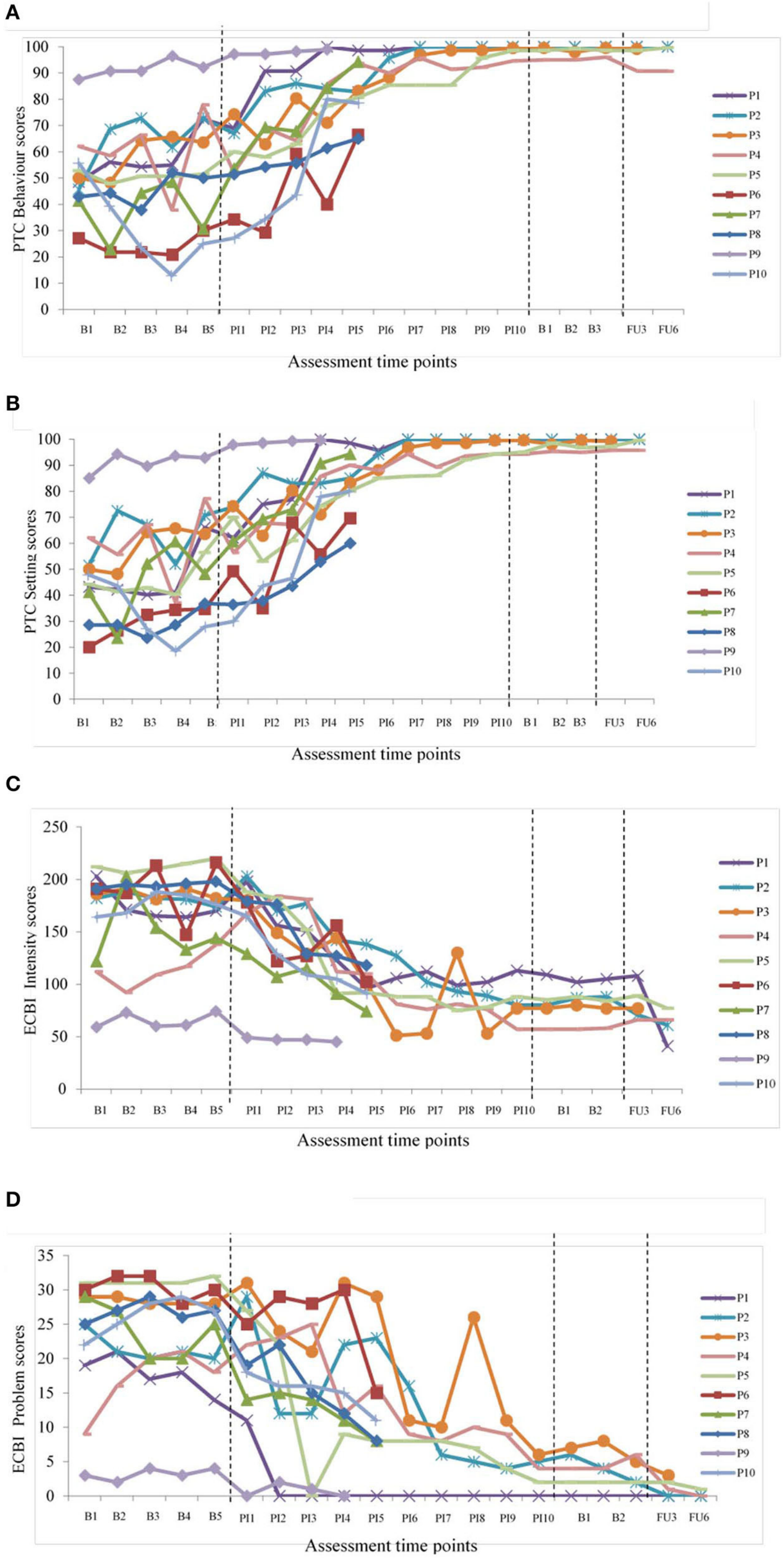


FIGURE 3 | Changes in parenting and child behavior. **(A)** PTC behavior scores over time. **(B)** PTC setting scores over time. **(C)** ECBI intensity scores over time and **(D)** ECBI problem scores over time as per the file in the original submission.

Parents described dreading spending time with their children and reported a desire to escape:

"I used to dread picking him up from school. I used to dread just being on my own with him… I'd say to my parents please let him come and stay at yours" (participant 4).

Significantly, although all were asked, no parents were able to identify anything positive about their parenting prior to the program and this question elicited an emotional response for some.

Parents reported their children's behavior to have been very problematic, describing disruptive and sometimes destructive behavior that drew comments from others, including wider family members and school teachers. This sometimes led to parents feeling judged and embarrassed and in some cases made them reluctant to be out in public with their child: "I was scared of being alone with him, I was worried if I had to take him out in how you say it, a public situation" (participant 4).

Parents noted the interplay between their children's behavior, their own mental health and parenting before the program, revealing very stressful family environments:

"I think probably my mental health and the fact I was always moody and miserable and shouting was obviously just fueling the fire… it just meant that he could play up even more and he could be as destructive as he wanted and stubborn as he wanted because I was not able because of my mental health to deal with it" (participant 4).

"…just all the stress, all the worries, all the screaming, all the fighting, all the shouting probably kept me up at night, so that's probably what caused the mental illness more than anything else really, just not being able to know how to deal with things" (participant 8)

"there was no rules, there was no instructions, there was no backup plans, there was no charts for doing good behaviors, there was no praise, there was no organization, it was chaos" (participant 8).

Expectations at the Start of the Program

Feelings of hopelessness prior to the program were echoed in low expectations about what the program could achieve. The two parents who had previously completed parenting courses reported not to have derived any benefit and expectations were again low: "when I was first told about this one I was like oh no not again" (participant 9). Parents who had not previously attended parenting courses were no more optimistic:

"I kind of felt like it wouldn't work at the start erm, just reading through it was kind of like, how did it feel like, how, can't believe I'm doing this… I'm going to bear to do this but in my heart of hearts I feel like this is totally not going to work" (Participant 6)

Another had not felt that she needed support despite struggling with her children "because I didn't think that there was a special parenting way" (Participant 8)

These misgivings were echoed in participant's preliminary perceptions of the workbook. Initial views were negative or neutral at best. Some parents found it daunting, describing it as "off-putting" and likening it to a school textbook in appearance (participants 4, 6 and 9):

"What put me off was the book itself because it looks like a textbook and the first thing you think of is oh my god I feel like a kid you know, I'm 40 years old. I really don't want to be doing this again" (participant 9)

Despite these reservations, "quick wins" in the first weeks boosted parents' confidence in the program and kept them motivated to take part. Parents reported particular success in this period with behavior charts, using descriptive praise and being consistent with ground rules.

"when I started I was at ground zero you know the, I was at rock bottom, I couldn't have got any lower so them few initial weeks are fantastic… you know it's just the tiny little things but just repeat, repeat, repeat I repeat the times out.. just little things and you get to a place where the child is responding" (participant 4).

Impact of the Program

Parents reported many positive impacts of the program, on their children, on themselves and more broadly on the wider family. Accounts indicated that by the end of the program, parents were more actively engaged with their children; more consistent in their parenting, and generally more structured in their approach and had a greater sense of control. As evidenced in the quantitative data, self-efficacy was very much improved and children's behavior was perceived to be less problematic overall. Parents took pride in the change that had been achieved and reported increases in their own wellbeing and mental-health. Reduced stress in relation to parenting was highlighted as a particular benefit and their improved mental health was attributed to this.

Impact on the Parent: Parents' accounts of the change within themselves were striking. The program had been transformative for all five who completed it. Not only were they all able to identify specific ways in which their parenting skills had improved, there were global statements that they were "better parents" and "changed people" as a result of the program. They were less stressed, calmer and happier in themselves and were looking forward with enjoyment to time and activities with their children.

"It has kept me calm, it was like being a new Mum again in a way… even though [X] is eight" (participant 5)

"I feel a lot more like a parent now than I was then" cause then I was just feeling embarrassed, low mood, really low, like I have no control over these children whatsoever what I do, now I feel I'm a better parent because of it (participant 8)

"The fact that I smile and laugh every day and I don't necessarily, I can't remember the last time I've asked my parents to have my kids overnight because I enjoy weekends" (participant 4)

"It's completely changed my whole world, 6 months ago is it, yeah summertime, I was in a terrible place with my mental health and it's just turned everything around it really has… having a strategy and a routine for the children and knowing how to handle their difficult behavior, it helps improve my wellbeing, knowing that I can cope, knowing that I can deal with it, erm if I get stuck I just refer to the book" (participant 6)

"It's made me feel a lot more confident in myself, knowing that these things can happen and that I can actually deal with them instead of trying to run away and hide which is what I used to do, bury my head in the sand like an ostrich" (participant 9).

Impact on the Child: Parents reported their children to be happier and more affectionate as a result of the changes parents has been able to make, describing a better child-parent relationship as well as noting significant improvements in behavior, which also transferred to other environments, including school. Two parents used the phrase "a new child".

"[X] is a completely different child. I get hugs, I get kisses, temper tantrums are virtually gone erm, you know, the need for shouting or having to really deal with [X] in that way is not required (participant 4)

"The kids are a lot happier, they're a lot happier now, 'cause I'm taking more time with them, sitting down and giving them this and some attention and I didn't know what I was doing before. I do now though, I'm glad I do it (participant 8)

Children were described as less oppositional overall and displayed a greater understanding of consequences. Parents recognized their own role in changing their own parenting behavior to achieve these outcomes and discussed improvements with pride. Communication between parents and children was also better and parents described themselves as feeling 'closer' to their children. Parents reported enjoying time with their children and looking forward to their time together in marked contrast to their reflection of before the program. "I enjoy spending time with my children now, erm quality of life is just so different" (participant 4).

Broader Impacts

Parents reported additional impacts from the program relating to better communication within the wider family and more openness between all family members. Parents were more hopeful about their own lives and the future life chances of their children.

"[X] was going to grow up and at 13, he was going to be out, he was going to end up smoking weed or drinking or hanging round with the wrong crowd or I dread to think, but I think I'm going to have a lovely well rounded teenager and I'm not scared of that thought" (participant 4)

Participants with younger children also recognized the opportunity for them to benefit from the program: "all the things I've got in this book I didn't have when [X] and [X] were little so obviously it's going to be easier to put them into place for him because he's down here and not up here you know, I can teach him without the mistakes (participant 9).

Theme 2: Valuable Aspects and Suggestions for Improvement

Parents unanimously felt that the support they had been given to complete the program was invaluable. When asked whether they felt it was a necessary component of success all five felt that some face to face contact would be required for anyone completing the program in the future. Views on the possibility of group delivery were mixed and although some thought that the normalizing aspects of a group session with peers might be beneficial, reservations about sharing essentially private information with strangers meant this was not a viable option. It was clear that the one-to-one support they had received had ensured understanding of the book, and provided opportunity to discuss strategies:

"I think I'd have given up, I think by week 2, week 3 if I'd not seen a response on my own without having someone to talk to. Yes I would have just given up and it's awful to admit that but it's the truth" (participant 4)

"... sometimes I found myself slipping like I did have to actually look at the book and think "what should I do now" ... it was good if like there was sometimes I didn't understand something or knowing how to approach a situation correctly ... sometimes it's better to have that bit more input you know, am I doing it right?" (Participant 5)

Participants discounted entirely the (hypothetical) option of online delivery although there was some agreement that it could potentially be feasible if accompanied by effective telephone support, particularly at crisis points:

"It's a robot, it's a computer, it's not one to one, if you had a question it can't answer you" (participant 6)

"Doing it on a computer or tablet you're not going to stick with it, I wouldn't have, you need that interaction... that [online delivery with telephone contact] might be good if you know, you just sort of came on week one and there was maybe, like there's in mental health, a crisis team that you can ring" (participant 4)

Others felt that that it would be difficult to discuss parenting challenges on the telephone indicating the need for a rapport to be built first:

"nooooo... like things that we talk about well I would be like 'is she pulling her face' or like 'is she' cause like you can't see her you just don't know" (participant 5)

Parents were primarily recruited to the study via referral from adult mental health services and the independence of the study from children's services seemed to facilitate openness about parenting difficulties. Parents valued the normalizing aspects of the intervention, particularly discussion of the interaction between parenting stress and mental health and did not seem to experience their involvement as stigmatizing:

"A lot of parents clam up as soon as that starts to happen [struggling with children] because they're scared of letting someone like social services in... they're going to come in, they're going to sweep in, take your kids, bye bye, there goes your family" (participant 9)

"well this is the thing 'cause I didn't know that this psychosis was very common and that you'd had people, clients like me, who were err suffering the same sort of thing but, it makes me happy that they will accept this is a mental illness and try and help you with it and still be a good Mum" (participant 8)

Parents made several practical suggestions for improvements to the workbook. They recommended a more aesthetically pleasing design to counter its textbook appearance and encourage better engagement. The need for a more inclusive book was highlighted, such as greater representation of more diverse family types, especially single parent families, and a larger font and simplified text.

Parents found some wording and phrases difficult to understand at first, for example 'incidental teaching' and felt that some simplification was required but on the whole found the workbook clear and concise. Opportunities to discuss any sections that were difficult to follow were valued and showed the importance of having someone to discuss progress with:

"There were a couple of sections on 1 week that I just couldn't get my head round, I couldn't understand the phrasing, I couldn't get behind the concept and I struggled to deal sort of that week… I struggled to reach the full benefits but then I think when I saw [facilitator] to review the week she said don't get hung up on the things that you can't, just concentrate on the things that you do" (participant 4).

Participation in the case series involved weekly assessment of mental health and parents valued these discussions, which enabled the linking of mood and well-being to family stress and challenges. One parent recommended the monitoring of parental health in the workbook:

"Maybe something about your mental health and how you're going through the book and, and you know a graph or something or a place to erm, I don't know week one make a few notes or you know you, at the end of each chapter… a summary section for parents to write in of how they've thought that the week went and maybe a graph as well just so the parent could see because all about mental health, it's peak and troughs you know it's ups and downs, ups and downs and I think maybe if I could have seen you know erm, oh I've had a good week or on a bad week but I put a little note at the side what that blip was that maybe you know that, that could have been beneficial" (participant 4).

DISCUSSION

This is the first study to systematically explore the use of a guided self-help parenting intervention with parents experiencing psychosis. Recruitment rates were good and considerable change in outcomes was reported over time for the 50% who completed follow up. The remaining five completed only 40–60% of the intervention but it is noteworthy that all participants saw significant improvements across measures during the program. Very high acceptability ratings were reported and the qualitative evaluation was extremely positive. The practicality aspect of the intervention was rated with the lowest satisfaction for those ending prior to week 10.

Previous research had highlighted the challenges of recruiting parents and service users into research and retaining participants in parenting interventions, therefore weekly guidance and assistance was offered. This was largely due to low levels of confidence, poor literacy skills, cognitive deficits and motivation. Initial barriers to progress and engagement were described as feeling overwhelmed and a mistrust of services. Two participants exited the study early due to struggling with literacy and chaotic environments including house moves. Support to understand and implement strategies and exercises in the workbook were essential. This improved as perceptions of self and parent-child relationship shifted throughout the course of the intervention.

Clinically significant reductions (>25%) across mental health and parent-child outcome measures were demonstrated on weekly measures. The magnitude of change was significant across mental health and parenting practices, parental confidence, and child behavior outcome measures, except for PANSS. The frequency, severity and levels of distress caused by hallucinations and delusions on PSYRATS was significantly reduced during the ten weeks of the intervention and continued to improve post-intervention and at follow-up. At completion, all participants were no longer meeting criteria for 'clinical' categories on outcome measures. Similarly, improvements across facets of child disruptive behaviors, positive parenting practices and parental self-efficacy was comparable to previous research (21, 25, 28, 31). Participants moved from the "clinical" category showing fewer child behavior problems and less parental distress on the ECBI and were within the "normal" range for parenting practices on the PS. Parental self-efficacy showed a substantial increase on the PTC. Improvements were maintained at follow-up, except for depression, anxiety and stress scores on the DASS-21. Where significant effects were found, effect sizes were very large (above 1.0).

The qualitative evaluation revealed that parents who completed the full 10 weeks of the intervention were extremely positive about its impacts, despite initial reservations and doubts about its ability to effect change. Their accounts also gave insights into the mechanisms of change by which parental mental health and wellbeing were improved. Parents described greater self-efficacy in their parenting as a result of the intervention. A more positive approach to parenting and improved behavior management strategies combined to improve child behavior. Parents spoke of enjoying time with their children rather than finding interactions stressful. Stress reactivity relating to psychosis has been demonstrated in numerous studies (54) and stress has been shown to worsen psychiatric symptoms in people with psychosis. It is also well established that high conflict family environments are linked to greater symptoms and increased likelihood of relapse in schizophrenia (55). In addition, there is evidence that more severe symptoms are associated with higher levels of parenting stress in people with serious mental illness and, conversely, that parenting improves when symptoms decline (56). Hence, it seems likely that parental wellbeing is enhanced by the stress of parenting being reduced. The accounts of the five participants who did not fully complete the intervention are missing of course, and it may be assumed that a different picture may well have emerged had they been included. Nonetheless, the finding that 50% of those who started the program experienced such significant change is important, and indicates that with the required adjustments to enable parents to engage with such an intervention, significant positive impacts are possible.

Strengths and Limitations

No participants had previously taken part in research and all ten presented with chronic symptomology. Therefore, the process of change could be conceptualized differently than those within other services presenting with acute as opposed to chronic mental health challenges. Despite aiming to recruit both mothers and fathers, no fathers took part. The majority of these mothers were also single, experiencing socioeconomic disadvantage and facing adversity. Although this reflects the typical household makeup of children living with a parent with psychosis (57), future research should seek to include fathers and parents from broader socioeconomic backgrounds where possible. The majority of participants were also White British. Spoken and written English was an inclusion criterion that will have precluded some groups of parents from taking part (for example immigrants). Suitability of the intervention for more diverse populations would need to be established in a larger trial.

With parents acting as their own baseline controls, the extent to which change can be attributed to the intervention cannot be fully established. To counteract this limitation, any significant lifestyle, family or medication changes were monitored. No significant changes were reported. Case series methodology is restrictive in its ability to demonstrate treatment efficacy; however, applying a multiple baseline design, session-by-session measures and reporting effect sizes, strengthens the findings. Autocorrelation of data was not assessed and controlled for which may have increased the likelihood of type I error.

The same researcher completed all assessments from baseline to follow-up which could cause biases in design and interpretation of outcomes. Although developing a relationship over time with the participants was a strength, to mitigate potential interpretation biases, self-report measures were used and a subset of mental health interviews were listened to by the wider research team. Follow-up at 3 and 6 months showed that gains acquired during the intervention were largely durable; however, this length of follow was insufficient to determine whether improvements can be maintained in the longer term. A longer follow up period, ideally a year or more, would offer insight into the strategies, knowledge and techniques that remain useful and implemented by families in the longer term.

Conclusion and Future Directions

Preliminary indications arising from this study are that a home-visiting parenting intervention for parents experiencing psychosis could be feasible, effective and valuable. Further studies involving larger and more diverse samples and a randomized controlled design are needed to substantiate these outcomes and more work surrounding successful implementation is needed.

Establishing the impact of parenting interventions for parents with different levels of need, varied illness length and the impact on quality of life will ensure support can be targeted and appropriate. It is also essential to identify the active ingredients within a parenting intervention that drive or prevent change (34).

The optimum modality and duration of parenting interventions needs to be adequately examined to ascertain the most beneficial method. Feedback from parents suggested that less repetition and using audio-guidance or visual supports such as video animations or infographics could assist those with poor literacy. This could also address some of the practical challenges reported. Utilizing technology (for example, smartphone applications and electronic behavior diaries) could also be of use to self-monitor behavior change. There are clear benefits from simply being listened to, having distress recognized and receiving warmth. Exploring the role of face-to-face support will disentangle intervention benefits from the benefits of modality type. Staff engagement and awareness of the dual demands of parenting when experiencing a serious mental health challenge needs to be of focus to ensure efficacious parenting interventions are no longer under-utilized in mainstream services. Examination of the most appropriate and useful ways of disseminating and integrating future work into services with a multi-agency approach in mind is essential to target hard-to-reach families.

This study has established that the use of the Triple-P Self-Help Workbook, using a guided and supportive framework, is feasible to deliver to parents experiencing psychosis. Positive outcomes were apparent across mental health measures with some participants no longer experiencing delusions or hallucinations and others moving out of clinical ranges. Child behavior, parenting practices and parent-child interactions all improved for each participant completing the intervention highlighting its potential promise as an intervention. Future development work with a focus on implementation should seek to increase the acceptability of the intervention to ensure completion and increase retention to follow up so that its effects may be determined more robustly.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Greater Manchester West National Research Ethics Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

LW: conceptualization, designing the study, data collection, quantitative data analysis, and writing the manuscript. RC: conceptualization, designing the study, supervision, and reviewing and editing the manuscript. RD: conceptualization, designing the study, and supervision LG: conceptualization, designing the study, supervision, qualitative data analysis, and writing, reviewing and editing the manuscript. All authors contributed to the article and approved the submitted version.

FUNDING

LW was supported by a President's Doctoral Scholar award from The University of Manchester.

ACKNOWLEDGMENTS

We gratefully acknowledge the commitment and support of each parent who took part in the study and thank the staff who referred them to us.

REFERENCES

1. Booij S, Snippe E, Jeronimus B, Wichers M, Wigman J. Affective reactivity to daily life stress: Relationship to positive psychotic and depressive symptoms in a general population sample. *J Affect Disorders*. (2018) 225:474–81. doi: 10.1016/j.jad.2017.08.051
2. Reininghaus U, Depp CA, Myin-Germeys I. Ecological interventionist causal models in psychosis: targeting psychological mechanisms in daily life. *Schizophr Bull.* (2016) 42:264–9. doi: 10.1093/schbul/sbv193
3. Perera DN, Short L, Fernbacher S. There is a lot to it: Being a mother and living with a mental illness. *Adv Ment Health.* (2014) 12:167–81. doi: 10.1080/18374905.2014.11081895
4. Firmin RL, Zalzala AB, Hamm JA, Luther L, Lysaker PH. How psychosis interrupts the lives of women and men differently: a qualitative comparison. *Psychol Psychother.* (2021) 94:704–20. doi: 10.1111/papt.12317
5. Waller S, Reupert A, Ward B, McCormcik RN, Kidd S. Family-focused recovery: Perspectives from individuals with a mental illness. *Int J Ment Health Nurs.* (2018) 27:247–55. doi: 10.1111/inm.12528
6. Reupert A, Price-Robertson R, Maybery, D. Parenting as a focus of recovery: A systematic review of current practice. *Psych Rehab J.* (2017) 40:361–70. doi: 10.1037/prj0000240
7. Nicholson J, Biebel K, Katz-Leavy J, Williams V. *The Prevalence of Parenthood in Adults With Mental Illness: Implications for State and Federal Policymakers, Programs, and Providers.* Rockville, MD: Psychiatry Publications (2004). Available online at: https://escholarship.umassmed.edu/cgi/viewcontent.cgi?article=1148&context=psych_pp
8. Goodman S, Brumley H. Schizophrenic and depressed mothers: Relational deficits in parenting. *Dev Psychol.* (1990) 26:31–9. doi: 10.1037/0012-1649.26.1.31
9. Gowers S. Family Matters: Interfaces between child and adult mental health. *Br J Psychiat.* (2001) 178:582–582. doi: 10.1192/bjp.178.6.582-a
10. Wan MW, Abel KM, Green J. The transmission of risk to children from mothers with schizophrenia: a developmental psychopathology model. *Clin Psychol Rev.* (2008) 28:613–37. doi: 10.1016/j.cpr.2007.09.001
11. Rosenblum K, Lawler J, Alfafara E, Miller N, Schuster M, Muzik M. Improving maternal representations in high-risk mothers: A randomized, controlled trial of the mom power parenting intervention. *Child Psychiatry Hum Dev.* (2018) 49:372–84. doi: 10.1007/s10578-017-0757-5
12. Serketich WJ, Dumas JE. The effectiveness of behavioural parent training to modify antisocial behaviour in children: A meta-analysis. *Behav Ther.* (1996) 27:171–86. doi: 10.1016/S0005-7894(96)80013-X
13. Dretzke J, Davenport C, Frew E, Barlow J, Stewart-Brown S, Bayliss S. The clinical effectiveness of different parenting programmes for children with conduct problems: a systematic review of randomised controlled trials. *Child Adolesc Psychiatry Ment Health.* (2009) 3:7. doi: 10.1186/1753-2000-3-7
14. Bee P, Bower P, Byford S, Churchill R, Calam R, Stallard P, et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. *Health Technol Assess.* (2014) 18:1–250. doi: 10.3310/hta18080
15. Clarke B, Younas F, Family Kids and Youth project team. *Helping Parents to Parent*. London: Social Mobility Commission (2017).
16. Ayano G, Betts K, Maravilla JC, Alati. The risk of anxiety disorders in children of parents with severe psychiatric disorders: a systematic review and meta-analysis. *J Affect Disord.* (2021) 282:472–87. doi: 10.1016/j.jad.2020.12.134
17. Seeman M. *Schizophrenia and motherood*. In Gopfert M, Webster J, Seeman MV, editors. Parental psychiatric disorder: Distressed parents and their families. Cambridge: Cambridge University Press. (2004) pp. 161–71. doi: 10.1017/CBO9780511543838.013
18. Nicholson J, Sweeney EM, Geller JL. Focus on women: mothers with mental illness: I. the competing demands of parenting and living with mental illness. *Psychiatric Services.* (2014) 49:643–9. doi: 10.1176/ps.49.5.643
19. Howard L, Underdown H. Hilfebedarfe von Eltern mit psychischen Erkrankungen - eine Literaturübersicht. *Psychiatr Prax.* (2011) 38:8–15. doi: 10.1055/s-0030-1265976
20. Reupert A, Maybery D. Programmes for parents with a mental illness. *J Psychiatr Ment Health Nurs.* (2011) 18:257–64. doi: 10.1111/j.1365-2850.2010.01660.x
21. Nicholson J, Deveney W. Why not support (ed) parenting? *Psychiatr Rehabil J.* (2009) 33:79–82. doi: 10.2975/33.2.2009.79.82
22. National Institute for Health and Clinical Excellence. *Antisocial behaviour and conduct disorders in children and young people: Recognition, intervention and management*. (2013). Available online at: http://www.nice.org.uk/guidance/cg158/resources/guidance-antisocial-behaviour-and-conduct-disorders-in-children-and-young-peoplerecognitionintervention-and-management-pdf
23. Sanders MR. Triple P-Positive Parenting Program: Towards an empirically validated multilevel parenting and family support strategy for the prevention of behavior and emotional problems in children. *Clin Child Fam Psychol Rev.* (1999) 2:71–90.
24. Sanders MR, Markie-Dadds C, Tully LA, Bor W. The Triple P - Positive Parenting Program: A comparison of enhanced, standard and self-directed behavioural family intervention for parents of children with early onset conduct problems. *J Consult Clin Psychol.* (2000) 68:624–40. doi: 10.1037/0022-006X.68.4.624
25. Sanders MR, Kirby JN, Tellegen CL, Day JJ. The triple P-Positive parenting program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clin Psychol Rev.* (2014) 34:337–57. doi: 10.1016/j.cpr.2014.04.003
26. Markie-Dadds C, Sanders M. Self-Directed Triple P (Positive Parenting Program) for mothers with children at-risk of developing conduct problems. *Behav Cogn Psychother.* (2006) 34:259–75. doi: 10.1017/S1352465806002797
27. Sanders MR. Development evaluation, and multinational dissemination of the triple P-positive parenting program. *Annu Rev Clin Psychol.* (2012) 8:345–79. doi: 10.1146/annurev-clinpsy-032511-143104
28. Morawska A, Sanders MR. Self-administered behavioural family intervention for parents of toddlers: Effectiveness and dissemination. *Behav Res Ther.* (2006) 44:1839–48. doi: 10.1016/j.brat.2005.11.015
29. Sanders MR, Markie-Dadds C, Rinaldis M, Firman D, Baig N. Using household survey data to inform policy decisions regarding the delivery of evidence-based parenting interventions. *Child Care Health Dev.* (2007) 33:768–83. doi: 10.1111/j.1365-2214.2006.00725.x
30. Jones S, Calam R, Sanders M, Diggle PJ, Dempsey R, Sadhnani V. A pilot web based positive parenting intervention to help bipolar parents to improve perceived parenting skills and child Outcomes. *Behav Cogn Psychother.* (2014) 42:283–96. doi: 10.1017/S135246581300009X
31. Jones SH, Jovanoska J, Calam R, Wainwright LD, Vincent H, Asar O, et al. Web-based integrated *bipolar* parenting intervention for parents with bipolar disorder: a randomised controlled pilot trial. *J Child Psychol Psychiatry.* (2017) 58:1033–41. doi: 10.1111/jcpp.12745
32. World Health Organization. *The ICD-10 Classification of Mental and Behavioural Disorders*. Geneva: World Health Organization (1993). Available online at: https://www.who.int/classifications/icd/en/GRNBOOK.pdf
33. Baranowski T, Stables G. 'Process evaluations of the 5-a-day projects'. *Health Educ Behav.* (2000) 27:157–66. doi: 10.1177/109019810002700202
34. Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M, Medical Research Council Guidance. Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ (Clinical Research Ed.).* (2008) 337:a1655. doi: 10.1136/bmj.39486.541725.80
35. Moore GF, Audrey S, Barker M, Bond L, Bonell C, Hardeman W, et al. Process evaluation of complex interventions: Medical Research Council guidance. *Br Med J.* (2015) 350:h1258. doi: 10.1136/bmj.h1258
36. Kay SR, Fiszbein A, Opler LA. The positive and negative syndrome scale (PANSS) for schizophrenia. *Schizophr Bull.* (1987) 13:261–76. doi: 10.1093/schbul/13.2.261
37. Haddock G, McCarron J, Tarrier N, Faragher EB. Scales to measure dimensions of hallucinations and delusions: the psychotic symptom rating scales (PSYRATS). *Psychol Med.* (1999) 29:879–89. doi: 10.1017/S0033291799008661
38. Morosini PL, Magliano L, Brambilla L, Ugolini S, Pioli R. Development, reliability and acceptability of a new version of the DSM-IV Social and Occupational Functioning Assessment Scale (SOFAS) to assess routine social functioning. *Acta Psychiatr Scand.* (2000) 101:323–9. doi: 10.1111/j.1600-0447.2000.tb10933.x

39. Lovibond PF, Lovibond SH. The structure of negative emotional states: Comparison of the Depression Anxiety Stress Scales (DASS) with the Beck Depression and Anxiety Inventories. *Behav Res Ther*. (2005) 33:335–43. doi: 10.1016/0005-7967(94)00075-U
40. Sanders M, Woolley ML. The relationship between maternal self-efficacy and parenting practices: Implications for parent training. *Child Care Health Dev*. (2005) 31:65–73. doi: 10.1111/j.1365-2214.2005.00487.x
41. Eyberg SM, Pincus D. *Eyberg Child Behavior Inventory and Sutter-Eyberg Student Behavior Inventory-Revised: Professional Manual. Odessa, FL: Psychological Assessment Resources.* (1999).
42. Arnold DS, O'Leary SG, Wolff LS, Acker MM. The Parenting Scale: A measure of dysfunctional parenting in discipline situations. *Psychol Assess.* (1993) 5:137–44. doi: 10.1037/1040-3590.5.2.137
43. Sanders MR, Markie-Dadds C, Turner KMT. *Practitioner's manual for Standard Triple P*. Brisbane Queensland, Australia: Triple P International Pty Ltd. (2001).
44. Wells A, Fisher P, Myers S, Wheatley J, Patel T, Brewin CR. Metacognitive Therapy in Recurrent and Persistent Depression: A multiple-baseline study of a new treatment. *Cognit Ther Res.* (2009) 33:291–300. doi: 10.1007/s10608-007-9178-2
45. Hutton P, Morrison AP, Wardle M, Wells A. Metacognitive therapy in treatment-resistant psychosis: A multiple-baseline study. *Behav Cogn Psychother*. (2014) 42:166–85. doi: 10.1017/S1352465812001026
46. Moore L, Moore GF. Public health evaluation: which designs work, for whom and under what circumstances? *J Epidemiol Community Health*. (2011) 65:596–7. doi: 10.1136/jech.2009.093211
47. Mazzucchelli TG, Sanders MR. Facilitating practitioner flexibility within an empirically supported intervention: Lessons from a system of parenting support. *Clin Psychol.* (2010) 17:238–52. doi: 10.1111/j.1468-2850.2010.01215.x
48. Lambert MJ, Hansen NB, Bauer S. Assessing the clinical significance of outcome results. In A. M. Nezu & C. M. Nezu (Eds.), *Evidence-based outcome research.* New York, NY: Oxford University Press. (2008) p. 359–378. doi: 10.1093/med:psych/9780195304633.003.0017
49. Jacobson NS, Truax P. Clinical significance: a statistical approach to defining meaningful change in psychotherapy research. *J Consult Clin Psychol.* (1991) 59:12–9. doi: 10.1037/0022-006X.59.1.12
50. Durham RC, Guthrie M, Morton RV, Reid DA, Treliving LR, Fowler D, et al. Tayside–Fife clinical trial of cognitive–behavioural therapy for medication-resistant psychotic symptoms. *British Journal of Psychiatry.* (2003) 182:303–11. doi: 10.1192/bjp.182.4.303
51. Leucht S, Kissling W, Davis JM. The PANSS should be rescaled. *Schizophr Bull.* (2010) 36:461–2. doi: 10.1093/schbul/sbq016
52. Morrison AP, Pyle M, Chapman N, French P, Parker SK, Wells A. Metacognitive therapy in people with a schizophrenia spectrum diagnosis and medication resistant symptoms: A feasibility study. *J Behav Ther Exp Psychiatry*. (2014) 45:280–4. doi: 10.1016/j.jbtep.2013.11.003
53. Ritchie J, Spencer L. Qualitative data analysis for applied policy research. In: Bryman A, Burgess RG, editors. *Analyzing Qualitative Data*. London: Routledge (1994). p. 173–94.
54. Myin-Germeys I, van Os J. Stress-reactivity in psychosis: evidence for an affective pathway to psychosis. *Clin Psychol Rev.* (2007) 27:409–24. doi: 10.1016/j.cpr.2006.09.005
55. Butzlaff RL, Hooley JM. Expressed emotion and psychiatric relapse, a meta-analysis. *Archives of General Psychiatry*. (1998) 55:547–52. doi: 10.1001/archpsyc.55.6.547
56. Kahng SK, Bybee D, Oyserman D, Mowbray. Mothers with serious mental illness: When symptoms decline does parenting improve? *Journal of Family Psychology*. (2008) 22:162–6. doi: 10.1037/0893-3200.22.1.162
57. Ranning A, Munk Laursen T, Thorup A, Hjorthøj C,Nordentoft M. Children of parents with serious mental illness: With whom do they grow up? A Prospective, Population-Based Study. *J Am Acad Child Adolesc Psychiatry*. (2016) 55:953–961. doi: 10.1016/j.jaac.2016.07.776

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 28 February 2022
doi: 10.3389/fpsyt.2022.781332

Concerns of Parental Substance Abuse and Mental Health Problems Reported to Child Welfare Services—Testing a Moderated Mediation Model for Paths From Reports to Substantiated Concern and Service Provision

Svein Arild Vis [1]*, *Camilla Lauritzen* [1], *Karen J. S. Havnen* [2], *Charlotte Reedtz* [1] and *Bjørn Helge Handegård* [1]

[1] *Regional Center for Child and Youth Mental Health and Child Welfare, UiT-The Arctic University of Norway, Tromsø, Norway,*
[2] *Regional Center for Child and Youth Mental Health and Child Welfare, NORCE Norwegian Research Centre, Bergen, Norway*

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
John Cosgrove,
University of Maryland, Baltimore,
United States
Kate Blake-Holmes,
University of East Anglia,
United Kingdom

***Correspondence:**
Svein Arild Vis
svein.arild.vis@uit.no

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: 22 September 2021
Accepted: 24 January 2022
Published: 28 February 2022

Citation:
Vis SA, Lauritzen C, Havnen KJS,
Reedtz C and Handegård BH (2022)
Concerns of Parental Substance
Abuse and Mental Health Problems
Reported to Child Welfare
Services—Testing a Moderated
Mediation Model for Paths From
Reports to Substantiated Concern
and Service Provision.
Front. Psychiatry 13:781332.
doi: 10.3389/fpsyt.2022.781332

Background: Parental mental health and substance abuse problems are found in reports of concern to child protection and welfare services. The aim of this study was first to investigate what characterized these reports and how they differed from reports with other types of concerns. Two hypotheses were tested. The first hypothesis was (i) if a report contains concerns about mental health and substance abuse problems, the likelihood of service provision was mediated by substantiation status. The second hypothesis was (ii) that the threshold for substantiation of such problems differed depending on child age, single parent status, and the presence of other child and parent related problems.

Method: The study was designed as a case file study which was carried out retrospectively ($N = 883$). A conceptual model was tested in two steps. First a mediation model with direct and indirect paths from reports of concerns through substantiation decision to service provision was tested. Then a second model was expanded to also include moderators for the indirect effects of reported concerns on substantiation decisions.

Results: A total of 33.1% of reports about substance abuse and 41.7% of reports about parental mental illness concerns were provided services. The first hypothesis was confirmed. There is a negative direct effect and a positive mediated effect of reported concern on service provision. The second hypothesis was not confirmed. We failed to identify any significant moderating effect of child age, single caregiver status, or number of child problems, upon the threshold for substantiation of mental health and drug abuse problems.

Conclusions: The total effect of reports about mental illness and substance abuse upon service provision was low. Service provision in cases with suspected substance abuse and/or mental illness is highly dependent upon substantiation of that specific problem. Substantiation threshold is not impacted by other case characteristics. This is surprising because there are good theoretical reasons to assume that parental drug abuse and or mental illness are potentially more detrimental to child health, development and safety if the child is younger, if the parent is a single caregiver, and there are many other parallel concerns.

Keywords: child protection, investigation, assessment, parental mental health, parental substance abuse, decision making, substantiation, service provision

INTRODUCTION

A public health approach to prevent adverse outcomes and improve quality of life for children when there is a concern about the parent's mental health or substance use is important. The Norwegian Child Welfare and Protection Services (CWPS) are obligated to ensure that children and youth who live in conditions that may be detrimental to their health and development receive necessary support and care. A substantial proportion of the reports of concern the CWPS receives is about parental mental health problems (12.0%) and parental substance abuse (16.9%) (1). There is little knowledge about whether these circumstances lead to reports of concern being substantiated. Substantiation implies that a case is considered by the CWPS to cause a serious concern for the child's health and development. When a case is substantiated, the family will normally be offered some form of voluntary support service by the CWPS. Usually this takes the form of parental guidance and counseling conducted by a social worker. This may be in addition to, but usually not as a substitute for other health care or social services.

It should be an aim for health care and social services to work together to identify and provide services for children and their parents in families struggling with parental mental health problems and/or substance abuse. Little is known about how factors such as child age, parental custody, and the presence of other risk factors influence the judgement and risk assessments of social workers within the CWPS. It is important to learn more about this because early identification and service provision may help prevention of trans-generational transmission of mental health problems. When the problems influence the child's everyday life and functioning the CWPS can offer supportive interventions for these families. However, this depends on the problems being identified and reported by other services or individuals.

One of the major challenges in mental health care and other health services has been to identify children of parents with a mental illness (COPMI) (2). According to the Norwegian Child Welfare Act of 1992, professionals in institutions bound by the professional duty of confidentiality (i.e., teachers, day care personnel, health care services) are required to report cases of concern to the CWPS. Adult psychiatric services have started to recognize the parenting challenges patients have, and to acknowledge the need for interventions to support parenting and the patients minor children (3). One approach to increase identification of COPMI and increase provision of services in Norway has been to appoint designated child responsible personnel in adult mental health services. The role of the child responsible personnel has been to record information about patients' children and establish collaboration with services that could provide patients and their families with support (2). We are therefore interested in looking more closely at what proportion of cases reported to the CWPS originate from health care services.

Parents struggling with mental health issues may potentially experience the development of a variety of problems in their offspring. These children run a higher risk of abuse and neglect, depression, anxiety disorders, substance abuse, eating disorders, conduct problems, and academic failure (4–6). The ongoing spiral, where mental disorders are being transmitted from one generation to the next, is one of the core mechanisms in the development of mental illness (3). Previous research found that one in five minors have a parent with mental illness (7), and that between 44 and 74% of these children develop psychosocial or mental health problems (8).

In the same way parental substance abuse (PSA) is a well-established risk factor for a variety of negative psychosocial outcomes for children (9–14), but some studies have also focused on "resilient" children of substance abusers, like the "classical" study by Werner (15). American studies have widely documented the association between PSA and child maltreatment, and both American and British studies have recognized PSA as a major concern for the CWPS (16–18).

Early parent-child interactions have shown to be important in child development (19). A focus on how parental substance abuse and mental health problems influence the interaction between parents and their children is crucial for the CWPS in order to determine risk for the child's health and development. Being a single parent or caregiver leaves the child more dependent on one caregiver because there is no one else to compensate for parent-child interaction problems. Hence, single caregiver status is considered a strong risk factor for child developmental problems in such cases (20). Additionally, the potential risk

---

Abbreviations: COPMI, Children of parents with a mental illness; CWPS, Child Welfare and Protection Services; PSA, Parental Substance Abuse.

is larger for very young children compared to older children. One of the reasons for this is that young children, i.e., under school age are more dependent upon their parents for provision of basic care and safety. Whereas, parental mental health problems also affect the emotional support and guidance for older children, deficiencies in parenting due to mental health problems also poses a potential safety risk when young children are involved.

Children and adolescents in contact with the CWPS have been found to have an elevated risk for mental health problems. In a large Norwegian study, Iversen et al. (21) found that 56% of children receiving in-home services from the CWPS had mental health problems, compared to 8% among those who did not. Additionally, several Norwegian studies (22, 23) and international studies (24–26) have documented that children in out-of-home care have mental health problems or disorders to a significantly larger extent than the general population of minors. In a recent study in Norway, researchers found that youth in foster care had lower scores on life quality compared to a general sample of adolescents, and that the life quality among those in foster care was comparable to COPMI (27).

In some contrast to the above mentioned studies Havnen et al. (28) found that Norwegian children placed in out-of-home care due to parental substance abuse had less mental health problems and better prosocial behavior than children placed due to other reasons. Research on mental health issues of children with PSA problems in the CWPS is however scarce, and the findings are somewhat divergent (26, 29). Findings about sociodemographic characteristics of the PSA children, are more consistent, as several studies have reported that children placed out-of-home because of PSA were younger, more often girls and more often had single caregivers than other children placed out-of-home (16, 18, 28, 29). In a study of COPMI conducted by Reedtz et al. (30), similar characteristics were discovered. The study assessed the circumstances and characteristics of COPMI when a parent was receiving treatment in the adult mental health services. Two thirds of the children were aged 0–11, and a large proportion had single caregivers (30).

A recent Norwegian study of the CWPS (31) compared problems identified in reported concerns to the services with concerns described by the CWPS in their concluding investigation reports. Reports about abuse/neglect were substantiated in about half the cases and rarely identified as new problems, while concerns about children's functioning, parenting competencies and parent-child interactions were often assessed as worrying, also in cases where this was not reported initially. Reports about parental substance abuse were substantiated in $<50\%$ of the cases and seldom detected as a new problem, whereas parental mental health problems were more likely to be detected as new problems irrespective of the content in the initial report of concern. The systematic change in problem profile from initial reports of concern to the concluding assessment reports from the CWPS, led the authors to suggest that the assessment process during the investigation is influenced by a tendency of CWPS workers to identify certain risk factors more than others (31). A seemingly predominant focus on parent-child interactions and a lack of suitable interventions that target many of the possible risk factors, might hinder CWPS workers from detecting problems related to parental mental health, as well as parental substance abuse. If this is the case, it may represent an erroneous basis for assessment conclusions made by the CWPS, as these problems represent serious risk factors in child development.

To understand the context of our study it is important to note that the Norwegian CWPS differs in some ways from the British and the US CPS (32). Whereas, the British and US services traditionally have been described as directed primarily toward protecting children against neglect and abuse, the Nordic CWPS are described as more directed toward supporting children and families in need, in order to prevent out-of-home placements (33–35). Although the development toward prevention and home-based support through differential response systems has been seen in the US and other countries (36) there is still a disproportionate number of children of PSA being placed into out-of-home care. In the US the proportion of PSA children in out-of-home care increased from 18.5% in the year 2000 to 38.9% in 2019. There were however substantial differences between states (37). Results from a Canadian longitudinal study on placement risk (38) show that for younger children below the age of nine, increased placement risk is explained by family difficulties whereas increased placement risk for older children is explained by behavioral problems. There are currently no epidemiological studies linking PSA or parental mental health problems to risks for out-of-home care in Norway. The main activity of the Norwegian CWPS, consists of voluntary consent-based interventions (71%), while relatively few of the families receiving services (29%) are placed out-of-home by court orders (1). In line with the public welfare tradition, the threshold to report cases of concern to the Norwegian CWPS is low. About 80% of all reports of concern to the CWPS in Norway are screened-in for further investigation. Of these, only 40% of the investigations are concluded with service provision (31, 39). The most common reason for service provision from the CWPS is parenting problems (23%), while parental mental health problems and substance abuse only account for 8 and 5%, respectively (1).

The current study is based upon the theory of the General Assessment and Decision-Making model in child protection (40). The theory states that the assessment dimension of risk, i.e., the level of concern in a CWPS investigation, is dependent upon the case factors influencing the assessment and the threshold for taking action. If the evaluation of case factors indicate that the weight and amount of evidence is large enough to raise an alarming concern for the child, then the case is substantiated, and services will be offered to the family. The threshold level for substantiation is however not defined by an objective proxy but is rather influenced by the views, experience, and knowledge of the decision-maker. The actions of social workers are in turn influenced by organizational factors, such as the routines and regulations within the CWPS agency, as well as factors external to CWPS, e.g., the capacity and service provision by other health care services. This study is focusing on the interaction of different case factors and how these influence the threshold levels determined by social workers when concluding an investigation of parental mental health problems or substance abuse.

AIMS AND HYPOTHESES

The purpose of the current study is to investigate (a) who reports concerns about parental substance abuse or mental health problems and what are the characteristics of these reports of concern, (b) what are the direct and mediated effects of reports about COPMI and PSA upon service provision, and (c) what are the thresholds for substantiation of parental mental health problems and parental substance abuse moderated by other case characteristics.

We hypothesize that service provision is dependent upon the CWPS's investigation and assessment of the case. Furthermore, we believe the threshold for substantiation of concerns as well as provision of services is lower the younger the child is, in cases where parents have substance abuse problems or mental health problems and are single caregivers. We also hypothesize that in cases where the reported substance abuse problems and mental health problems coinside with other child problems and parenting problems, there is a lower threshold for substantiation and service provision.

METHODS

The study was designed as a case file study which was carried out retrospectively. A total of 1,365 cases were randomly drawn from all referrals registered in the 16 participating agencies in the period of January 2015 to June 2017. The agencies represented (i) six districts from the three major cities in Norway with a population ranging from 190,000 to 680,000, (ii) six regional cities with a population ranging from 20,000 to 80,000, and (iii) four agencies from smaller towns and rural areas with a population below 15,000. The number of cases from each agency varied between 50 and 150 depending on the size of the agency. The reason why we sampled agencies by size is that we wanted the number of cases drawn from each agency to be approximately the same proportion of the total available sample from that agency.

Data was collected and coded from case records. A data entry form was developed and tested for interrater reliability by independent coding of 20 cases by two researchers. The results showed an average interrater agreement of 86.9%. A total of 13 variables had low reliability (<80% interrater agreement). Three of those were eliminated from the form due to the conclusion that reliable information could not be obtained. The remaining 10 variables were reformulated, and the coding manual was revised with better explanation of codes. After this revision the reliability of the instrument was re-tested by independent coding of 42 cases by two researchers. At this second step, average interrater agreement was 90.8%. In health research, an interrater agreement over 80% is generally considered acceptable (41). The variables and the codes from the form are available from the corresponding author upon request.

Participants

For the present analysis we included all the cases that were subject of a child protection investigation, and which were concluded in an investigation report ($n = 883$). We did not include cases that were screened out without any further investigation ($n = 242$) and cases that did not have a concluding report ($n = 240$). The reason for this is that we were not able to determine whether or not the reported concern had been substantiated in those cases.

There were 54.0% boys ($n = 477$) in the sample and the mean age was 9.0 years (SD = 5.0). In a total of 40.8% of the referrals, the family had immigrant background. Immigrant background was defined as the child or one of the parents being born in a country other than Norway.

The sample was representative for the population of cases involved with the CWPS in Norway with respect to child age, child gender, and the proportion of cases screened out or screened in for service provision.

Measures

The dependent variables investigated in this article are whether parental mental illness or substance abuse was substantiated and if services were provided. Predictors were (i) child age (ii) caregivers' civil status, (iii) number of substantiated concerns about child development and health, (iv) number of substantiated concerns about parenting problems.

Possible substantiated child development and health related concerns were age-adequate development, mental health problems, child crime/substance abuse, externalizing behavior problems, functioning in school/kindergarten, emotional problems, social problems with peers, social problems with adults or conflict with adults. Each problem was counted as substantiated or not.

Possible substantiated concerns about parenting problems were deficiencies in parental stimulation/guidance/boundary setting, basic care for the child, parents' emotional availability, or parents' protection of the child.

Summary of coding of included variables and their possible values are shown in **Table 1**.

Statistical Analyses

Analysis was carried out in Mplus. The conceptual model shown in **Figure 1** was tested in two steps. First the mediation model (a, b, and c paths) was tested for each of the paths from reported mental illness or substance abuse to service provision. Then a second model was expanded to also include the moderators (**Figure 1**). When programming the model in Mplus we followed the recommendations by Stride et al. (42) to test indirect and total effects of each possible combination of high, medium and low values for the moderators using the model constraint function. For the age variable, low was set to 4 years, medium was 9 years and high was 14 years. For the variables: number of parenting problems and number of child problems, low was set to zero, medium was one and high was three.

We used bootstrapping and the weighted least square mean and variance adjusted estimations.

Chi-square tests were used to test differences in distribution of reporters for cases with mental health problems/substance abuse problems and reporters in cases without mental health problems/substance abuse problems.

TABLE 1 | Included variables and their coding.

Data source	Variables	Values
Reports of concern	Reported parental substance abuse problem	0 = no 1 = yes
	Reported parental mental illness	0 = no 1 = yes
	Single caregiver	0 = no 1 = yes
	Child age	0–17
Investigation report	Number of child problems substantiated	0–9
	Number of parenting problems substantiated	0–4
	Substance abuse substantiated	0 = not substantiated 1 = substantiated
	Mental health problems substantiated	0 = not substantiated 1 = substantiated
CPS conclusion	Service provision	0 = no 1 = yes

RESULTS

Cases with concerns about parental mental illness and substance abuse problems are not surprisingly often reported by healthcare services. It is worth noting however that private parties, such as a relative, a friend or a neighbor seem to be much more involved in reporting those kinds of concerns compared to the CWPS reports in general. This is particularly true for reports concerning substance abuse problems where about one third of the reported concerns originated from a non-mandated reporter. For the overall reports of concerns to CWPS in Norway however, about four out of five reported concerns were submitted by a professional (**Table 2**).

There are also other differences between concerns about parental mental illness and substance abuse compared to the total amount of reported concerns (**Table 3**). First, these reports were related to children who were about 1 year younger compared to the other reports of concern, and their caretakers were more often a single parent. There were some differences between reports of concerns regarding parental mental illness and those with concerns about substance abuse, with respect to what other problems were substantiated because of the investigation. When the report was about substance abuse, problems related to child development and heath were less frequently identified. When the reported concern was about parental mental illness more parenting problems were identified. The reported concerns about mental illness and substance abuse problems were substantiated

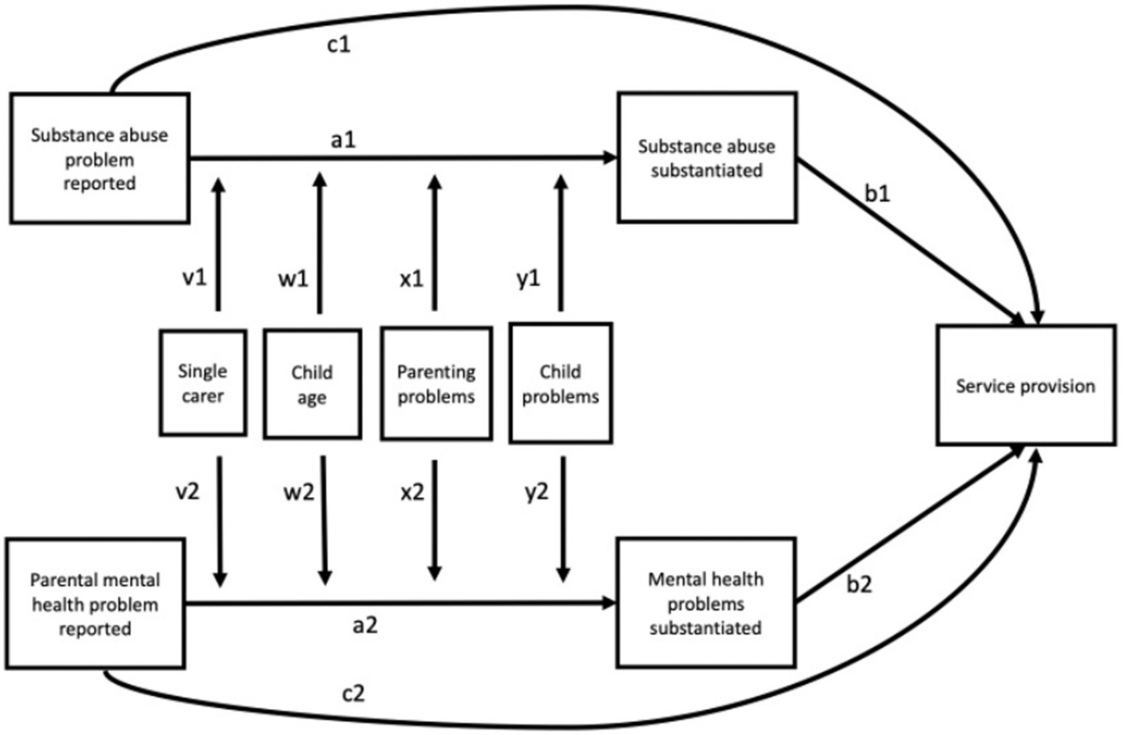


FIGURE 1 | The conceptual model. In the moderation analysis, the substance abuse paths (a1, b1, c1) and the parental mental health paths (a2, b2, c2) were tested separately.

TABLE 2 | Reporters of cases with concerns about parental mental health problems or substance abuse to CPS.

	Reports with parental mental health problems (*n* = 132) *N* (%)	**Reports with parental substance abuse problems (*n* = 160) *N* (%)**	**Total sample (*n* = 883) *N* (%)**
Health care services	50 (37.9)	40 (25.0)	175 (19.8)
School	11 (8.3)	9 (5.6)	188 (21.3)
Police	12 (9.1)	28 (17.5)	127 (14.4)
Social services	10 (7.6)	5 (3.1)	53 (6.0)
CPS agency	14 (10.6)	25 (15.6)	132 (14.9)
Private parties	32 (24.2)	48 (30.0)	187 (21.2)
Other	3 (2.3)	5 (3.1)	21 (2.4)

The overall test for differences in who referred cases with mental illness [$\chi^2_{(6)} = 44.1$] and substance abuse [$\chi^2_{(6)} = 36.8$] compared to other cases were both highly significant ($p < 0.001$). The tests are comparisons of the distribution of reporters for cases with mental health problems/substance abuse problems and reporters in cases without mental health problems/substance abuse problems.

TABLE 3 | Characteristics of reports with suspected parental mental illness or substance abuse compared to the total sample.

	Total Sample (*n* = 883) *M* (SD)	**Reports with alleged parental mental health problems (*n* = 132) *M* (SD)**	**Reports with alleged parental substance abuse (*n* = 160) *M* (SD)**
Child age	9.0 (5.0)	8.0 (5.4)*	7.7 (5.1)**
Number of substantiated child related problems	1.1 (1.7)	0.9 (1.6)	0.6 (1.4)***
Number of substantiated parenting problems	0.6 (0.9)	0.8 (1.1)*	0.5 (1.0)
	***N* (%)**		
Single care	299 (33.9)	56 (42.4)*	69 (43.1)**
Substantiated mental health problem	144 (16.3)	77 (58.3)***	43 (26.9)***
Substantiated drug abuse problem	110 (12.5)	24 (18.2)*	86 (53.8)***
Service provision	339 (38.4)	55 (41.7)	53 (33.1)

*Significance levels are *$p < 0.05$; **$p < 0.01$; and ***$p < 0.001$. Testes are t-tests and chi-square tests for differences between cases where the referral concern is present vs. all other cases.*

in about 50–60% of the cases. Substantiation of a problem does however not always lead to provision of services for the family. Services were provided in about 38% of the cases and was not more or less likely in cases about mental illness and substance abuse than in other types of cases.

The results from the mediation analysis showed, as should be expected, that the total effects of the reported concerns about parental mental illness (OR = 1.22, $p = 0.33$) or substance abuse (OR = 0.72, $p = 0.10$) upon service provision is low and non-significant. As shown in **Figure 2**, the direct effects are negative, and the indirect effects are positive. This indicates that service provision in these types of reports is highly dependent upon whether or not that specific problem is substantiated by CWPS. It is also worth noting that there were strong effects from reported concerns to substantiation decision. This means that substantiation of parental substance abuse or mental health problems is highly dependent upon being identified as a concern in the report to the CWPS, substance abuse problems more so than other mental health problems. The total odds ratio for reports of mental health problems to service provision via substantiation was OR = 1.86 (95% CI: 1.40–2.33) and the odds ratio from substance abuse reports to service provision through substantiation was OR = 1.81 (95% CI: 1.23–2.39). The path effects for the mediation model are shown in **Figure 2**. The log scale estimates for indirect effects are found in the table's two sub-notes.

The analysis of moderated paths from reports of concern to substantiation did not identify any statistically significant moderators, i.e., there were no statistically significant interaction effects between reported concerns and child age, single care status, or the number of substantiated child or parenting problems, upon substantiation decision for either mental health problems or substance abuse. The regression estimates for the moderated mediation models are shown in **Tables 4**, **5**.

Since there were no significant moderating effects of child age, single care status or the number of substantiated child or parenting problems on the association between reported concerns and substantiation decision, conditional indirect effects are not reported. Thus, we have not sufficient evidence to claim that the indirect effects vary depending on the age of the child, single care status, or the number of substantiated child or parenting problems. The highest indirect effect for reported concerns about substance abuse with moderators present, were in cases where the parent was not a single caregiver, the child age was young (4 years) and there were three other substantiated child and parenting concerns (1.235, S.E = 0.416, $p = 0.003$). The lowest indirect effect with moderators present were when there was a single caregiver, the child was older (14 years), there were no substantiated child related concerns and one substantiated parenting concern (1.042, S.E. = 0.238, $p < 0.001$).

The highest indirect effect for reported concerns about parental mental illness with moderators present, were in cases where the parent was not a single caregiver, the child age was older (14 years) and there were no other substantiated child concerns, but three substantiated parenting concerns (1.312, S.E. = 0.338, $p < 0.001$). The lowest indirect effect with moderators present were when there was a single caregiver, the child was older (14 years) there were no substantiated child related concern and one substantiated parenting concern (0.456, S.E. = 0.248, $p = 0.066$).

DISCUSSION

Our initial aim was to study who reports concerns about parental substance abuse or mental illness and what are the characteristics

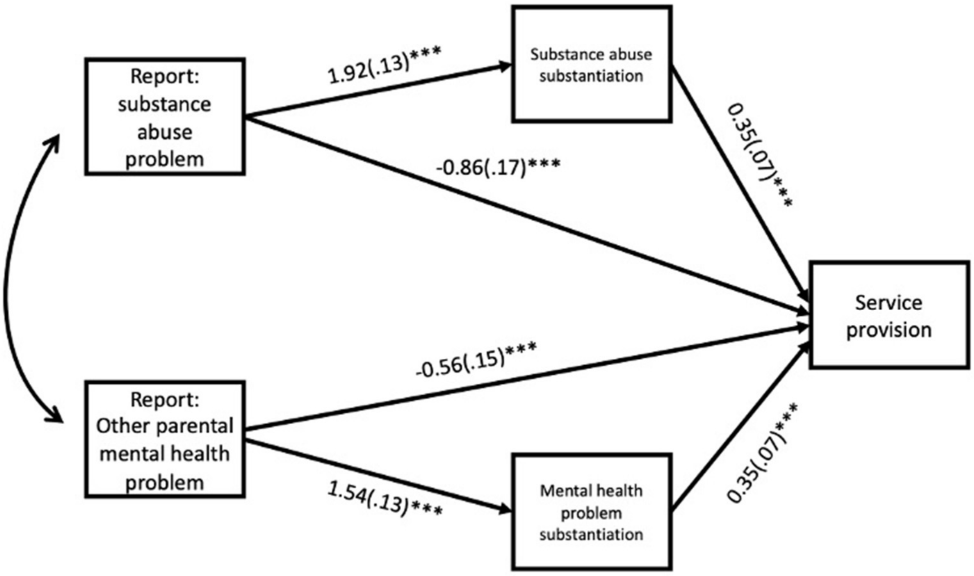


FIGURE 2 | Direct and indirect effects from referral concern to service provision. ***Indicate $p < 0.001$. The specific indirect effect from reported substance abuse problem to service provision was 0.67 (0.11)***. The specific indirect effect from reported parental mental health problem to service provision was 0.67 (0.14)***.

TABLE 4 | Regression estimates for the moderated mediation paths from referrals of parental mental health problems to service provision ($n = 883$).

Dependent variable	Predictor	WLSM estimate	Standard error	P
Service provision				
	Constant	0.629	0.118	<0.001
	Referral for mental illness (c2 path)	−0.537	0.351	0.126
	Substantiated mental illness (b2 path)	0.534	0.065	<0.001
Substantiated mental illness				
	Constant	1.476	0.165	<0.001
	Referral for mental illness	1.459	0.303	<0.001
	Child age	−0.031	0.014	0.022
	Singelcare = yes	0.142	0.127	0.265
	Substantiated child problems	0.164	0.036	<0.001
	Substantiated parenting problems	0.463	0.070	<0.001
	Referral by age (w2 path)	0.002	0.029	0.933
	Referral by single care (v2 path)	−0.410	0.280	0.143
	Referral by child problems (y2 path)	−0.068	0.095	0.475
	Referral by parenting problems (x2 path)	0.322	0.173	0.063

of these reports of concern. The results showed, not surprisingly, that a majority of these reports came from different healthcare services. Health care services refers to a large group of workers, such as public health nurses, school nurses, general practitioners, and hospital personnel. In their meetings with the patient, health personnel working with adult patients who struggle with mental health issues or substance abuse are in a key position to address the impact mental health problems may have on parenting quality. They are also an important agent to initiate collaboration with services such as the CWPS, which can provide supporting (and preventive) interventions for these families. However, we believe the reports of concerns from adult mental health services are still too low. Based on the large proportion of children and adolescents who have parents with mental illness or substance abuse in the general population, and on the large proportion of those who are in contact with the CWPS who have parents with such problems, it is reason to believe that the threshold to report concerns about COPMI is still too high. Even though COPMI have received a lot of attention in these services during the last decade, there still seems to be a long way to go before the collaboration between adult mental health services and the CWPS is adequate and leads to a functional service-provision (2).

A large proportion, about one third of the reports of concerns about PSA were reported from private parties, such as a relative, a friend or a neighbor. This is particularly disturbing if it means that non-mandated reporters of concern take on a larger

TABLE 5 | Regression estimates for the moderated mediation paths from referrals of parental substance abuse problems to service provision ($n = 883$).

Dependent variable	Predictor	WLSM estimate	Standard error	*P*
Service provision				
	Constant	0.576	0.122	<0.001
	Referral for substance abuse (c1 path)	−1.177	0.328	<0.001
	Substantiated substance abuse (b1 path)	0.553	0.082	<0.001
Substantiated substance abuse				
	Constant	2.267	0.282	<0.001
	Referral for substance abuse	2.101	0.362	<0.001
	Child age	−0.009	0.016	0.575
	Single care = yes	0.002	0.156	0.988
	Substantiated child problems	0.159	0.036	<0.001
	Substantiated parenting problems	0.672	0.076	<0.001
	Referral by age (w1 path)	−0.013	0.029	0.633
	Referral by single care (v1 path)	−0.042	0.286	0.885
	Referral by child problems (y1 path)	0.049	0.168	0.771
	Referral by parenting problems (x1 path)	0.014	0.184	0.939

Path name refers to ***Figure 1***.

responsibility for children at risk from neglect because of parental substance abuse, compared to professionals working with this group of patients/parents. These parents may not be identified as having such problems and may not receive help for their abuse problems from health care services. In such cases, reports of concern from private parties represent a safety net for children in families with substance abuse problems. The rest of the reported concerns related to such problems were reported by professionals that have a mandate to report. Because of the social stigma related to PSA, it is challenging for the CWPS to gather information about such problems through their contact with parents and children. In Norway, the police regularly report episodes of PSA to the CWPS, which can provide these services with important information and hence strengthen the potential for substantiation of concern and provision of services. From a prevention perspective, it is positive if parental mental illness and substance abuse is discovered while the children are relatively young, as interventions aimed at younger children have greater potential for strong effects on the child's development (43, 44).

Our second aim was to investigate direct and mediated effects of reports about COPMI and PSA upon service provision. We hypothesized that service provision is dependent upon the CWPS's investigation and subsequent substantiation of the reported problem. This hypothesis was confirmed. In terms of whether reports of concern were met with service provision by the CWPS, the results showed that total of 33.1% of reports with PSA and 41.7% of reports about parental mental illness were provided with services. Although the rate of service provision in cases where reports of concern is related to parental mental illness and/or substance abuse is similar to other types of reports, the analysis showed that there was a partial mediation effect that goes through the substantiation process. Primarily, this means that when parental mental health problems or substance abuse is reported, service provision is highly dependent upon the substantiation of such specific problems. Identification and substantiation of other types of problems seem less important. The CWPS's difficulties in substantiating parental mental illness and substance abuse emphasize the need for a broad assessment of child development, familial social resources and network, as well as general parenting problems in these cases (45).

It is an interesting finding that when the CWPS receive reports of concerns related to PSA, problems related to the development and health status of the child were less frequently identified. This may imply that PSA among parents is evaluated as important enough in itself, regardless of the child's developmental status. However, it may also imply that the CWPS become very focused on gathering "evidence" of substance abuse problems in the parents, as opposed to assessing the mental and social status and development of the child. If the latter is the case, the CWPS run the risk of being blind to the consequences of living with parental substance abuse, even though the services are finding it difficult to document such problems to a satisfactory degree. Havnen et al. (28) found that children placed out-of-home because of parental substance abuse had lower scores on mental health problems than children placed in out-of-home care because of other reasons. It is however possible that this is at least partly mediated by earlier intervention in cases with serious substance abuse, and that this protects the child from developing mental health problems later. Therefore, we would be very careful to dismiss the need for a broad assessment of the child's needs at the investigation phase of these cases.

When the reported concern was about parental mental illness, more parenting problems were identified. It is unclear why the CWPS are able to detect more parenting problems in these cases compared to cases where the concern is related to parental substance abuse. However, since it may be more intuitive why mental illness poses a threat to everyday parenting functions, it may seem more natural to assess such difficulties, as opposed to substance abuse problems which are difficult to substantiate by normal assessment procedures. It may be easier for parents to admit and seek help for mental health problems compared to substance abuse problems. Admitting to substance abuse problems in encounters with the CWPS may be considered more likely to invoke considerations of custody issues. Another explanation could be the stronger stigma connected to substance abuse problems than mental health issues, which in turn could make it easier for parents to admit and receive help for mental health problems.

Our final aim was to investigate if the thresholds for substantiation of parental mental health problems and parental substance abuse were moderated by other case characteristics. We hypothesized that the threshold for substantiation of cases was moderated by child age, single parent vs. two parent household and the number of other child and parenting problems that were identified. This hypothesis was rejected.

It is probable that the investigation process following reports about PSA in particular, more often aims to substantiate or unsubstantiate the substance use and that the investigation perhaps is not as broad and needs-oriented as in other cases. An implication of this may be that parents who struggle with substance abuse issues, may not be offered supporting interventions to prevent out-of-home placements. If this is the case one might question if this practice is in line with the broad family-oriented mandate of the Norwegian CWPS (33–35). However, it may also be the case that the CWPS have not implemented relevant interventions for these families. This interpretation is in line with the results of Christiansen et al. (31), who found that a lack of suitable interventions for these families hinders service-provision. As parental substance abuse represents a serious risk factor in child development, a lack of service provision in these cases needs to be met with implementation of adequate interventions and services by the CWPS.

It is also apparent that mental health problems are risk factors that the CWPS are not very likely to look for unless the concern has already been raised in the report. An implication of this may be that an important opportunity to discover COPMI is missed, because the CWPS does not systematically assess if the reported problems within a family may be due to mental illness or substance abuse. Subsequently, this has important implications for the partners of the CWPS that provide the reports of concerns, such as schools, health care services, social services, and the police. First, the reports should be specific and explicit about any suspicion that children are struggling due to parental mental health or substance abuse problems. If the report of concern is too vague or other vicarious concerns are provided as cause for the report to the CWPS, perhaps out of fear of offending parents or damaging a therapeutic relation, then the CPWS will most likely not look for such risk factors in their investigation. Although we do not know for certain how prevalent this problem may be, we do find it strange that private parties are so often the reporters of substance abuse and mental health problems. It is worrying if other public services are either not aware of or not sufficiently alarmed by such problems in the 25–30% of cases that are reported by family, friends, and neighbors. A solution to this could be to raise awareness among mandated reporters who work with children, e.g., schools, kindergartens, public health nurses, about the importance of identification and reporting of such problems.

Second, we do find it worrying that the CWPS assessment in cases with parental mental health problems or substance abuse seem to have a more limited scope than other reported concerns. This conclusion is supported by the failure in this study to identify any interaction effects between reported concern and child age, single care, number of child related problems or number of parenting problems on the substantiation decision. If the CWPS assessments had not primarily focused on the occurrence or non-occurrence of substance use and mental health problems but had also taken into consideration the effects this may have upon parents' ability to care for the child and on child functioning and health, such interaction effects should be expected. In a public health perspective, thresholds for substantiation of parental mental health problems and subsequent service provision should be much lower for young children of single caregivers due to the added effects of early intervention and prevention (3, 46). Additionally, with the current knowledge about the cumulative effects of family risk factors on child development and health (8), the threshold for substantiation and service provision should be lower when maladaption or child mental health problems are starting to manifest. Sadly, this does not seem to be the case since we found no evidence for the moderation effect hypothesis.

Limitations

We do not have information about the types of substance use parents were reported for, or the types of substance use that was substantiated. We do however assume that certain types of substance abuse such as use of heroin or methamphetamines that may have a more easily identifiable effect upon users are more likely to be substantiated. Additionally, does different types of drugs have differing effects upon the user's ability to care for the child. It is therefore likely that the types of substances that are being used affect both the chance of detection, and the likelihood that it will be considered serious enough to constitute a problem.

We only included variables at case level in this study. As indicated in the introduction we do acknowledge that not all variation in child welfare decision making is determined by case level factors. It may be important which social worker is processing the case. How serious a concern has to be in order to be substantiated as risk is not only influenced by the facts of the case but also by how these facts are understood and interpreted by the decision-maker. Unfortunately, we are unable to control for variability in professional judgement because we do not have information about which workers were involved in which cases. We do know how cases are nested within agencies. However, because the study was designed primarily to look at case level factors, we have relatively few agencies included. Therefore, we consider analysis of variation at the agency level to be outside the scope of this article.

CONCLUSIONS

The total effect of reports about mental illness and substance abuse upon service provision was low. This means that the overall chance that children and parents receive services from CWPS in these types of cases are about the same as when the reports of concern contain other types of problems. Service provision for reported concerns about substance abuse and/or mental illness is however much more dependent upon substantiation status. This indicates that action is likely taken in high-risk cases, but that preventive measures in low-risk cases are not as commonly provided.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because our license from the Data Protection Authority does not include permission to store raw data in open data repositories. Requests to access the datasets should be directed to svein.arild.vis@uit.no.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Norwegian Centre for Research Data, the Council for Duty of Confidentiality and the Norwegian Data Protection Authority. Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study following a decision by the directorate for children and family affairs.

AUTHOR CONTRIBUTIONS

SV designed the study. SV, KH, and CL collected the data. SV and BH conducted the analyses. All authors contributed to the writing of the article manuscript and agreed to be accountable for the content of the work.

FUNDING

The study was funded by the Norwegian Directorate for Children, Youth and Family Affairs (Bufdir) and UiT The Arctic University of Norway.

ACKNOWLEDGMENTS

This study is part of a larger project deigned to research the decision-making process in Norwegian Child Protection Services, and many have contributed. The authors of this article would like to acknowledge the contribution of the following researchers to the project: SV (PI), Øivin Christiansen, CL, KH, Anette Iversen, Toril Tjelflaat, Sturla Fossum, Marte Knag Fylkesnes, Veronika Paulsen, Gro Ulset, Eirinn Ljones, Kirsten Buck Rustad, Reidun H. Nygård, Frederikke Jarlby, Jannike Kaasbøl, Stein Erik Ulvund. The authors also want to thank the Norwegian Directorate for Children, Youth and Family affairs and UiT The Arctic University of Norway for funding the project. Additionally, the authors wish to thank the Research Support Team at RKBU/UiT The Arctic University of Norway.

REFERENCES

1. Statistics Norway (2020). Retrieved from: https://www.ssb.no/en/sosiale-forhold-og-kriminalitet/barne-og-familievern/statistikk/barnevern (accessed September 15, 2021).
2. Lauritzen C, Reedtz C. Child responsible personnel in adult mental health services. *Int J Mental Health Syst*. (2016) 10:64. doi: 10.1186/s13033-016-0098-y
3. Thanhäuser M, Lemmer G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? results of a systematic review and meta-analysis. *Curr Opin Psychiatry*. (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342
4. Beardslee WR, Versage EM, Van de Velde P, Swatling S, Hoke L. Preventing depression in children through resiliency promotion. In: Peters R, McMahon R, editors. *The Effects of Parental Dysfunction on Children*. Boston, MA: Springer (2002). p. 71–86. doi: 10.1007/978-1-4615-1739-9_4
5. Goodman SH, Rouse MH, Connell AM, Broth MR, Hall CM, Heyward D. Maternal depression and child psychopathology: a meta-analytic review. *Clin Child Family Psychol Rev*. (2011) 14:1–27. doi: 10.1007/s10567-010-0080-1
6. Goodman SH, Gotlib IH. (editors). Children of Depressed Parents: Mechanisms of Risk and Implications for Treatment. *American Psychological Association*. (2002). doi: 10.1037/10449-000
7. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry*. (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
8. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Austr e-J Adv Mental Health*. (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
9. Christoffersen MN, Soothill K. The long-term consequencies of parental alcohol abuse: a cohort study of childre in Denmark. *J Subst Abuse Treat.* (2003) 25:107–16. doi: 10.1016/S0740-5472(03)00116-8
10. Dore MM, Doris JM, Wright P. Identifying substance abuse in maltreating families: a child welfare challenge. *Child Abuse Negl.* (1995) 19:531–43. doi: 10.1016/0145-2134(95)00013-X
11. Haugland BSM. *Parental Alcohol Abuse. Family Functioning and Child Adjustment*. Bergen: University of Bergen (2003).
12. Johnson JL, Leff M. Children of substance abusers: overview of research findings. *Pediatrics.* (1999) 103:1085. doi: 10.1542/peds.103.S2.1085
13. Johnson JL, Sher KJ, Rolf JE. *Models of Vulnerability to Psychopathology in Children of Alcoholics, Alcohol Health and Research World, Vol. 15*. Rockville, MD: ProQuest Medical Library (1991). p. 32.
14. Steinhausen HS. Children of alcoholic parents. *Euro Child Adolesc Psychiatry.* (1995) 4:143. doi: 10.1007/BF01980453
15. Werner EE. Resilient offspring of alcoholics: a longitudinal study from birth to age 18. *J Stud Alcohol.* (1986) 47:34–40. doi: 10.15288/jsa.1986.47.34
16. Besinger BA, Garland AF, Litrownik AJ, Landsverk JA. Caregiver substance abuse among maltreated children placed in out of home care. *Child Welfare.* (1999) 78:221–39.
17. Cleaver H, Undell I, Aldgate J. *Children's Needs - Parenting Capacity: The Impact of Parental Mental Illness, Problem Alcohol and Drug Use, and Domestic Violence on Children's Development*. London: Jessica Kingsley (1999).
18. Forrester D, Harwin J. Parental substance misuse and child care social work: findings from the first stage of a study of 100 families. *Child Family Social Work.* (2006) 11:325–35. doi: 10.1111/j.1365-2206.2006.00415.x
19. Van Doesum KTM, Riksen-Walraven JM, Hosman CMH, Hoefnagels C. A randomized controlled trial of a home-visiting intervention aimed at preventing relationship problems in depressed mothers and their infants. *Child Dev.* (2008) 79:547–61. doi: 10.1111/j.1467-8624.2008.01142.x

20. McLanahan S, Sandefur G. *Growing Up With a Single Parent: What Hurts, What Helps*. Cambridge, MA: Harvard University Press (2009).
21. Iversen AC, Jakobsen R, Havik T, Hysing M, Stormark KM. Mental health problems among child welfare clients living at home. *Child Care Practice.* (2007) 13:387–99. doi: 10.1080/13575270701488790
22. Havnen KS, Jakobsen R, Stormark KM. Mental health problems in Norwegian school children placed outside home - the importance of family risk factors. *Child Care Practice.* (2009) 15:235–50. doi: 10.1080/13575270902891115
23. Lehmann S, Havik OE, Havik, T. Mental disorders in foster children: a study of prevalence, comorbidity and risk factors. *Child Adolesc Psychiatry Ment Health.* (2013) 7:39. doi: 10.1186/1753-2000-7-39
24. Ford T, Goodman R, Meltzer H. The British child and adolescent mental health survey 1999: the prevalence of DSM-IV disorders. *J Am Acad Child Adolesc Psychiatry.* (2003) 42:1203–11. doi: 10.1097/00004583-200310000-00011
25. Ford T, Vostanis P, Meltzer H, Goodman R. Psychiatric disorder among British children looked after by local authorities: comparison with children living in private households. *Br J Psychiatry.* (2007) 190:319–25. doi: 10.1192/bjp.bp.106.025023
26. Egelund T, Lausten M. Prevalence of mental health problems among children placed in out-of-home care in Denmark. *Child Family Social Work.* (2009) 14:156–65. doi: 10.1111/j.1365-2206.2009.00620.x
27. Larsen M, Goemans A, Baste V, Wilderjans TF, Lehmann S. Predictors of quality of life among youths in foster care-a 5-year prospective follow-up study. *Qual Life Res.* (2021) 30:543–54. doi: 10.1007/s11136-020-02641-z
28. Havnen KJS, Breivik K, Stormark KM, Jakobsen R. Why do children placed out-of-home because of parental substance abuse have less mental health problems than children placed for other reasons? *Child Youth Serv Rev.* (2011) 33:2010–7. doi: 10.1016/j.childyouth.2011.05.028
29. Vanderploeg JJ, Conell CM, CAron C, Saunders L, Katz KH, Tebes JK. The impact of parental alcohol or drug removals on foster care placement experiences: a matched comparison group study. *Child Maltreat.* (2007) 12:125–36. doi: 10.1177/1077559507299292
30. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness: a necessity to provide relevant support. *Front Psychiatry*. (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
31. Christiansen Ø, Havnen KJ, Vest RKBU. *Barnevernets undersøkelsesarbeid–fra bekymring til beslutning [Child protection investigations in Norway]*. Tromsø: Delrapport 4-Når barnevernet Undersøker (2019).
32. Kritz K, Skivenes M. Systemic differences in views on risk: A comparative case vignette study of risk assessment in England, Norway and the United States (California). *Child Youth Serv Rev.* (2013) 35:1862–70. doi: 10.1016/j.childyouth.2013.09.001
33. Samsonsen V. *Assessment in child protection: a comparative study Norway – England* (Ph.D. thesis), University of Stavanger, Faculty of Social Science, Stavanger (2016).
34. Pösö T, Skivenes M, Hestbæk AD. Child protection systems within the Danish, Finnish and Norwegian welfare states—time for a child centric approach? *Euro J Social Work.* (2014) 17:475–90. doi: 10.1080/13691457.2013.829802
35. Kojan BH. Norwegian child welfare services: a successful program for protecting and supporting vulnerable children and parents? *Austr Social Work.* (2011) 64:443–58. doi: 10.1080/0312407X.2010.538069
36. Fuller T. Beyond investigations: differential response in child protective services. In: *Handbook of Child Maltreatment* Dordrecht: Springer (2014). p. 443–61. doi: 10.1007/978-94-007-7208-3_24
37. National Center on Substance Abuse and Child Welfare (2019). Available online at: https://ncsacw.samhsa.gov/research/child-welfare-and-treatment-statistics.aspx (accessed February 14, 2022).
38. Esposito T, Trocmé N, Chabot M, Shlonsky A, Collin-Vézina D, Sinha V. Placement of children in out-of-home care in Québec, Canada: when and for whom initial out-of-home placement is most likely to occur. *Child Youth Serv Rev.* (2013) 35:2031–9. doi: 10.1016/j.childyouth.2013.10.010
39. Lauritzen C, Vis SA, Ulset G, Tjelflaat T, Rustad KB. *Barnevernets undersøkelsesarbeid. [Reports to child protection services]*. Tromsø:Delrapport 3. Meldinger til barnevernet (2019).
40. Baumann DJ, Dalgleish L, Fluke J, Kern H. *The Decision-Making Ecology*. Washington, DC: American Humane Association (2011).
41. McHugh ML. Interrater reliability: the kappa statistic. *Biochem Medica.* (2012) 22:276–82. doi: 10.11613/BM.2012.031
42. Stride CB, Gardner S, Catley N, Thomas F. *Mplus Code for the Mediation, Moderation, and Moderated Mediation Model Templates From Andrew Hayes' PROCESS Analysis Examples.* (2015). Available online at: http://www.offbeat.group.shef.ac.uk/FIO/mplusmedmod.htm (accessed February 14, 2022).
43. Reedtz C, Handegård BH, Mørch W-T. Promoting positive parenting practices in primary pare: outcomes and mechanisms of change in a randomized controlled risk reduction trial. *Scand J Psychol.* (2011) 52:131–7. doi: 10.1111/j.1467-9450.2010.00854.x
44. Reedtz C, Klest S. Improved parenting maintained four years following a brief parent training intervention in a non-clinical sample. *BMC Psychol.* (2016) 4:43. doi: 10.1186/s40359-016-0150-3
45. Havnen KJS, Fossum S, Lauritzen C, Vis SA. How does the Kvello assessment framework attend to important dimensions of the children's needs and welfare? A comparison with the BBIC and the ICS frameworks for child welfare investigations. *Nordic Social Work Res.* (2021). doi: 10.1080/2156857X.2021.1891959. [Epub ahead of print].
46. Van Doesum KTM, Hosman CMH, Riksen-Walraven JM. A model-based intervention for depressed mothers and their infants. *Infant Ment Health J.* (2005) 26:157–76. doi: 10.1002/imhj.20037

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 07 March 2022
doi: 10.3389/fpsyt.2022.784995

Rationale and Description of Implementation of Regional Collaborative Service Model for Enhancing Psychosocial Wellbeing of Children and Families—Oulu Collective Impact Study

Tuula Takalo[1]*, *Sami Räsänen*[1,2], *Helinä Hakko*[2], *Aapo Juutinen*[3] *and Mika Niemelä*[2,3]

[1] Research Unit of Clinical Neuroscience, Psychiatry, Faculty of Medicine, University of Oulu, Oulu, Finland, [2] Department of Psychiatry, Oulu University Hospital, Oulu, Finland, [3] Faculty of Medicine, Center for Life Course Health Research, University of Oulu, Oulu, Finland

OPEN ACCESS

Edited by:
Joanne Nicholson,
Brandeis University, United States

Reviewed by:
Yaara Zisman-Ilani,
Temple University, United States
Mary Brolin,
Brandeis University, United States

Correspondence:
Tuula Takalo
tuula.takalo@ouka.fi

Specialty section:
This article was submitted to Child and Adolescent Psychiatry, a section of the journal Frontiers in Psychiatry

Received: *28 September 2021*
Accepted: *31 January 2022*
Published: *07 March 2022*

Citation:
Takalo T, Räsänen S, Hakko H, Juutinen A and Niemelä M (2022) Rationale and Description of Implementation of Regional Collaborative Service Model for Enhancing Psychosocial Wellbeing of Children and Families—Oulu Collective Impact Study. Front. Psychiatry 13:784995. doi: 10.3389/fpsyt.2022.784995

Background: The psychosocial wellbeing of children and adolescents is an ongoing global concern. Despite positive outcomes of child- and family-focused programs, the fragmentation of services presents challenges. To enhance harmonization and diminish fragmentation of service policies, we implement a preventive collaborative service model for children and families. The rationale for our study is based on analyses of national and local data before implementing the service model in the pilot area.

Methods: The need for a preventive service model for children and families was demonstrated using national and local data sources. First, a national school health survey was utilized to screen adolescents' perceptions of their depressive symptoms and support. Second, time trends in child and adolescent psychiatric and child protection service use were investigated. For these aims, epidemiological data of the study area (city of Oulu) were compared with data from the whole country (Finland). Third, local usage data of the preventive stand-alone Let's Talk About Children (LT) intervention before the service model implementation were evaluated. After these analyses, Let's Talk About Children Service Model (LT-SM) implementation in the pilot area is described in detail.

Results: The background data showed that 17% of the adolescents in the study area had reported depressive symptoms, and almost half of them had not received professional help. Child and adolescent psychiatric outpatient visits had increased during the last decade, but the number of visits remains lower in the study area compared with the country as a whole. The number of adolescent psychiatric inpatient days had increased contrary to a decreasing national trend. The number of urgent child welfare placements was also higher compared with the whole country. The local LT intervention data revealed very low utilization rates. These results indicated a necessity to enhance preventive and collaborative support for children and their families. This article describes the implementation of the LT-SM.

Discussion: We demonstrated excessive use of curative services in social and health care and insufficient usage of the stand-alone preventive intervention. The LT-SM is now piloted in one regional service area of the city of Oulu. Its effectiveness will be evaluated when enough data have been accumulated for statistical analyses.

Keywords: children, adolescents, families, collective impact, collaboration, implementation, Let's Talk about Children Intervention, Let's Talk about Children Service Model

INTRODUCTION

Recently, there has been an increase in child protection needs and acts in Finland, some Nordic countries, and in Europe as a whole. Studies and reports have documented increased rates of psychological symptoms, diagnosed mental illness, and the use of psychiatric and social services in persons younger than 18 years (1–4). It is unclear whether the increased use of curative services indicates a higher incidence of illnesses; research suggests that they may also be indicators of weaknesses, such as inefficiency and fragmentation, in the current child and family social and healthcare services (3).

Separate policies and lack of collaboration between service providers are associated with inefficiency of the service system (5–7). This is understandable, as it may be difficult to take into account individually varying needs, diverse family backgrounds, and differences between communities when arranging the required services (8, 9). In response to these challenges, in Finland, government-funded programs for social and healthcare, daycare, and schools have been launched over the last decade, and national and international evidence-based programs and interventions have been implemented (10–18). However, these approaches have been narrowly oriented, restricted, and usually limited to one service area (19).

The most recent solution to the inefficiency of services has been to develop comprehensive service models covering all public providers as well as non-profit organizations. Various stand-alone interventions are used in these models, but they are integrated into a larger entity serving the common goal shared by the sectors. Utilizing these comprehensive models is expected to enhance psychosocial wellbeing and decrease the need for curative healthcare services or custodial arrangements in social services (20). An encouraging initiative is the Let's Talk About Children Service Model (LT-SM), which is a community-based infrastructure model developed for reinforcing child and family wellbeing and resilience. The main principle is to create a cross-sectoral infrastructure from prevention to treatment across different services and hierarchies (21). The LT intervention is an essential stand-alone method within the LT-SM, and there is encouraging research evidence indicating that it enhances psychosocial wellbeing and prevents the intergenerational impact of parental problems, such as parental mental health conditions (17, 18, 22–24) and somatic illnesses (25, 26).

The LT-SM shares principles with the collective impact (CI) framework in organizing services introduced by Kania and Kramer (27). The CI framework includes five conditions: a common agenda, shared measurement system, mutually reinforcing activities, continuous communication, and backbone support organization (21, 27). CI has been reported to be utilized with varying initiatives, such as food, health, and education programs (20, 28–30), whereas the LT-SM is focused on children's psychosocial wellbeing (21, 31). However, both models are intended to decrease the fragmentation of and improve the collaboration between services (21, 27, 32).

Reliable research-based evidence of the effectiveness of comprehensive models is lacking. In practice, examining or making conclusions about a model's efficacy at cross-sectoral service and service user level is not possible until after comprehensive implementation of the model (30, 32). Therefore, it is important to thoroughly describe the implementation process of the CI framework-based LT-SM.

The rationale of our study is based on analyzing epidemiological data on the perceived need and use of child and adolescent health and social services in the study area (city of Oulu). In addition, numerical data from the study area on the use of a single, preventive stand-alone LT intervention for children, adolescents, and families in different services before the implementation of the LT-SM in the pilot area of the city of Oulu were evaluated.

First, as indicators of need of services, we explored adolescents' perceptions of their depressive symptoms and experiences of receiving support for these problems in the study area (city of Oulu) utilizing the national School Health Promotion (SHP) survey data. Second, we analyzed 10-year time trends in the study area in the use of child and adolescent psychiatric and child protection services. In these two aims, the study area was compared with the whole country. Third, we examined the local usage data of the stand-alone LT intervention before the implementation of the LT-SM in the pilot area. After these analyses of national and local data, we describe in detail the implementation process of the LT-SM in the pilot area aimed to enhance the psychosocial wellbeing of children and families in Oulu. The effectiveness of that model will be evaluated in follow-up studies after implementation and systematic utilization of the LT-SM.

METHODS

The methods and results sections are structured as four sections. The first section consists of analyses of the national survey data of adolescents' perceptions of their depressive symptoms and experiences on receiving support for these problems in the study area (city of Oulu). The second section includes analyses of national epidemiological data, including the study area, to assess and compare time trends in the need and use of children and adolescent social and psychiatric services. The third section includes evaluation of the stand-alone LT intervention based on the numerical usage data from local registers of the city of Oulu at the time before the implementation of the LT-SM. The fourth section includes a detailed description of the implementation process of the LT-SM, which is piloted in one regional welfare service area (WSA) of Oulu.

Perceived Symptoms of Depression Indicating Need for Support (Section 1)

Data regarding perceived symptoms of depression and experience of receiving support for them among 14–16-year-old adolescents (eighth and ninth graders from comprehensive schools) were based on the nationwide SHP study conducted in the years 2017 and 2019 (33). The national- and regional-level data were available from the SOTKANET databank produced by the National Institute for Health and Welfare. The SHP is administered nationwide every second year, with data gathered with an anonymous and voluntary classroom-administered questionnaire. The topics in the questionnaire include living conditions, schoolwork, health, health-related behavior, and school health services.

Perceived Depressive Symptoms

The Patient Health Questionnaire 2 (PHQ-2) (34) utilized in the SHP is a self-report assessment for screening depression, its severity, and patient treatment response used to examine the data on depressive symptoms. These data allowed us to estimate the number of the adolescents with depressive symptoms. The PHQ-2 assesses loss of interest in activities and low spirits, depression, and feelings of hopelessness over the last 2 weeks, scored from 0 to 2 (0 = no symptoms, 2 = loss of interest and mood involvement).

Experience on Receiving Support

Experience of receiving support for symptoms of depression was demonstrated by four indicators, which were based on two questions in the school health survey. The first question is: "Have you been worried about your mood during the past 12 months?" [answers: (1) No; (2) Yes, and I have told someone about it; (3) Yes, but I have not told anyone about it]. Question 2 asks: "Have you received support and help concerning your mood during the past 12 months?" [answers: (1) Yes, a lot; (2) Yes, some; (3) No, but I would have needed it; (4) I have not needed any help], and it consists of four subsections: (1) from school adults (teacher, school health nurse, physician, psychologist, social worker); (2) from services outside school (health center, mental health services, youth services, etc.); (3) from your own parents; (4) from friends and relatives. Indicators in **Figure 2A** for school adults, services outside school, own parents, or friends and relatives were calculated based on response alternative 2 for question 1 (Yes, had been worried about mood during the past 12 months and had told someone about it) and question 2 response alternatives 1–2 (Yes, received a lot or some support and help from school adults, services outside school, own parents, or friends and relatives). Furthermore, experiences of not having received help for depressive symptoms from either school adults/services outside school or from own parents/friends or relatives in the last 12 months despite a perceived need for help were illustrated by two indicators (**Figure 2B**). The proportions were calculated based on respondents who answered question 2 with response alternative 3 (did not receive support or help, although would have needed it). The indicators do not include those respondents who responded to question 2 that they had not needed any help.

Epidemiological Data for Need and Use of Child and Adolescent Services (Section 2)

Data regarding child and adolescent psychiatric services and the use of child protection were gathered from the SOTKANET databank produced by the Finnish Institute for Health and Welfare (THL; Sotkanet.fi).

Use of Child and Adolescent Psychiatric Services

The psychiatric inpatient care indicator reflects the number of days young people spent in psychiatric hospital care (i.e., all psychiatric inpatient wards in the public sector) per 1,000 persons of the same age. The psychiatric outpatient care indicator provides the number of outpatient visits within child (aged 0–12 years) and adolescent psychiatry (aged 13–17 years) per 1,000 persons of the same age. No psychiatric inpatient care was provided in the private sector.

Use of Child Protection Services

The child protection indicator per 1,000 population reflects the percentage of children and adolescents aged 0–17 years who received child welfare placements or urgent (emergency) child welfare placements, both voluntarily and involuntarily. This indicator also includes those placed in care who turned 18 years during that year.

Use of the Stand-Alone LT Intervention (Section 3)

The stand-alone LT intervention (35) as a single intervention method has been implemented and registered in the city of Oulu since 2015 and 2017, respectively, Agreement for Wellbeing 2013–2017 (36). The LT intervention was offered universally to all parents of our target population. The numerical data for utilization of the LT intervention were extracted from the local statistical register of the city of Oulu including the Primus, Effica, and LifeCare register systems (37). These electronic register tools are intended for monitoring and collecting work-related information for professionals in daycare, schools, and services. The statistics on the use of the LT intervention were calculated by dividing the number of users by the total number of children in the target population, including public daycare, school, maturity and child health clinics, and school health services.

Implementation (Section 4)

Model Applied in Implementation

The LT-SM is a community-based action model aimed to enhance child and family wellbeing and resilience and prevent child and family problems. This model includes specific preventive interventions. The LT intervention has two steps: LT Discussion

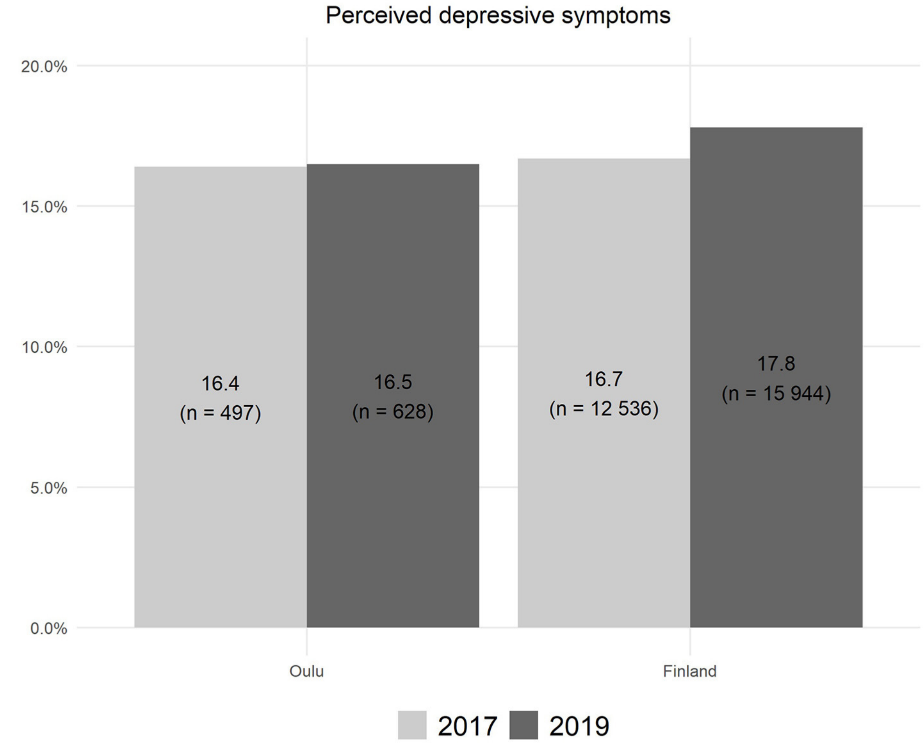


FIGURE 1 | Perceived symptoms of depression lasting at least 2 weeks among 14–16-year-old pupils in Oulu and Finland in 2017 and 2019.

with families mapping out and supporting the protective factors of the children and LT Network with cross-sectoral collaboration including the families and their social network (17, 18, 21, 31, 35). The LT-SM has two shared goals: to support children's everyday life in all developmental contexts and environments (i.e., at home, daycare, school, and leisure environments) and to build the corresponding service structure (21, 31).

A specific service structure was established with the following parts: (1) cross-sectoral Multiagent Management Group (MMG) with all core leaders from all relevant sectors, (2) a feedback system to collect information on implementation and collaboration quality, and (3) One Contact Service (OCS) to ensure that all families in the area would receive services and support within 1–2 weeks (21).

Pilot Region of Implementation

The implementation process was piloted in one (out of 18) regional WSAs in the city of Oulu, and it covered all social and healthcare services for children, adults, and families. It comprised three public schools (∼2,500 students), six daycare units (∼1,050 children), and a local healthcare center, including a well-child clinic. The participating daycare and school units included the school's student welfare services comprising nurses, social workers, and psychologists. The implementation process in the pilot area involved a total of 450 professionals: 150 from daycare, 260 from schools, and 40 social and healthcare professionals.

Statistical Methods

The Joinpoint Regression Program (version 4.8.0.1; Statistical Methodology and Applications Branch, Surveillance Research Program, National Cancer Institute, Bethesda, MD, USA) and the average annual percentage change (AAPC) was used to analyze time trends in the rates of psychiatric inpatient care days, outpatient visits, those placed in care, and those who received emergency placement, as well as to compare time trends between the city of Oulu and the whole country. Because of the relatively small number of observations causing high variability in the number of psychiatric inpatient care days and outpatient visits, a 3-year moving average (2-year moving average at the end of time periods) was used for the statistical modeling. The statistical significance of difference in AAPCs between the study area (city of Oulu) and the whole country (Finland) was calculated. In addition, the test of parallelism was used to determine whether the two regression mean functions between the study area and the whole country were parallel (same slope), allowing different intercepts. The remaining analyses and most of the **Figures 1–3** were performed using RStudio (version 1.2.1335; RStudio, Boston, MA, USA).

RESULTS

Perceived Symptoms of Depression and Received Support (Section 1)

In the study area (city of Oulu), a total of 3,028 (68% aged 14–16 years) adolescents in 2017 and 3,803 (76%) in 2019 participated in the nationwide school health promotion study. The corresponding figures for Finland as a whole were 75,068 (64%) in 2017 and 89,570 (75%) in 2019.

Perceived Depressive Symptoms

Approximately 16% ($n = 497$ and 628) of the adolescents reported symptoms of depression in the study area (city of Oulu) during 2017 and 2019, respectively (**Figure 1**). Accordingly, 16.7% ($n = 12{,}536$) and 17.8% ($n = 15{,}944$) of all Finnish adolescents reported depressive symptoms in 2017 and 2019, respectively. These proportions differed statistically significantly between Oulu and the whole country in the year 2019 ($\chi^2 = 4.14$, $p = 0.042$), but not in the year 2017 ($\chi^2 = 0.17$, $p = 0.679$).

Experience on Receiving Support

As illustrated in **Figure 2**, in the study area (city of Oulu), support was received from school adults (2017: 79.3%; 2019: 81.3%), services outside of school (2017: 68.3%; 2019: 67.8%), parents (2017: 81.2%; 2019: 81.5%), and friends and relatives (2017: 87.8%; 2019: 90.4%). The adolescents who reported a need for support for their depressive symptoms felt that they did not receive support from school adults or services outside school (2017: 48.8%; 2019: 49.4%) or from their parents, friends, and relatives (2017: 30.8%; 2019: 27.1%).

Nationally, adolescents received support from school adults (2017: 81.6%; 2019: 82.0%), services outside of school (2017: 71.6%; 2019: 72.9%), parents (2017: 81.5%; 2019: 81.7%), and friends and relatives (2017: 89.0%; 2019: 89.0%). There was also a substantial percentage of adolescents who perceived a lack of support from school adults or services outside school (2017: 43.8%; 2019: 42.5%) or from parents or friends and relatives (2017: 29.2%; 2019: 28.5%).

Epidemiological Data on Need and Use of Child and Adolescent Services (Section 2)

The time trends over the years 2007–2017 in the need and use of child and adolescent services as well as the use of child protection services were based on the population-adjusted data obtained from the nationwide indicator databank. The results are presented in **Figure 3**.

Use of Child and Adolescent Psychiatric Services and Child Protection Services

As illustrated in **Figure 3A**, the rate of outpatient care visits showed a significant increase in both the study area (city of Oulu) (AAPC = 9.9; 95% confidence interval = 8.9–11.0; $p < 0.001$) and the whole country (AAPC = 8.0; 95% confidence interval = 7.5–8.5; $p < 0.001$). The AAPC of the study area was significantly higher compared with Finland (difference = −1.9; 95% confidence interval = −2.9 to −0.9; $p < 0.001$). The overall test for parallelism also revealed a statistically significant difference in time trends between the study area (city of Oulu) and the whole country ($p = 0.028$).

From 2007 to 2017, the rate of inpatient care days (**Figure 3B**) showed a significant increase in the study area (city of Oulu) (AAPC = 0.9; 95% confidence interval = 0.6–1.2; $p < 0.001$) compared with a decreasing trend in Finland (AAPC = −4.6; 95% confidence interval = −5.0 to −4.3; $p < 0.001$). Furthermore, there was a statistically significant difference in AAPCs between the study area and Finland (difference = −5.5; 95% confidence interval = −5.9 to −5.1; $p < 0.001$). Following the reverse time trends observed in the study area and Finland, the overall test for parallelism showed a statistically significant difference between the study area and the whole country ($p = 0.002$).

Use of Child Protection Services

As seen in **Figure 3C**, over the entire time period, the percentage rates of adolescents placed in care showed a significant increase in both the study area (city of Oulu) (AAPC = 4.6; 95% confidence interval = 2.9–6.4; $p < 0.001$) and the whole country (AAPC = 0.4; 95% confidence interval = 0.2–0.7; $p = 0.005$). The increase in AAPC of the study area was significantly higher compared with Finland (difference = −4.2; 95% confidence interval = −5.7 to −2.7; $p < 0.001$). In addition, the overall test for parallelism revealed a statistically significant difference in time trends between the study area and the whole country ($p = 0.002$).

As shown in **Figure 3D**, a significant change occurred in time trends in the year 2011. During the first 4 years from 2008 to 2011, the percentage rates of urgent placements showed a significant increase in both the study area (city of Oulu) and the whole country (AAPC = 10.2; 95% confidence interval = 1.0–20.1; $p = = 0.031$). After 2011 up to 2018, the percentage rates of urgent placements remained stable in both the study area and Finland as a whole (AAPC = −0.04; 95% confidence interval = −2.3 to 2.3; $p = 0.969$). Throughout the whole 10-year time period, from 2008 to 2018, the percentage rates of urgent placements showed a significant increase in the study area and Finland (AAPC = 2.9; 95% confidence interval = 0.06–5.9; $p = 0.045$).

Use of Stand-Alone LT Intervention in Study Area (City of Oulu) (Section 3)

As demonstrated in **Figure 4**, the use of the LT intervention varied between 0% (school healthcare) and 5.8 and 9.1% (comprehensive schools) in the study area (city of Oulu) and in the implementation pilot area including one WSA in Oulu, respectively.

Description of CI Implementation Process (Section 4)

The CI implementation process was divided into three different phases. The starting point was the meeting where the common agenda was agreed. The phases from the starting point were the preparing phase (0–3 months), working phase I (4–12 months), and working phase II (13 months). The implementation research with the University of Oulu was prepared from the preparing phase of the process. It started 8 months after the starting point, during working phase I, once all necessary administrative permissions and funding for research had been obtained. The actions of the implementation are described in **Figure 5**.

Preparing Phase

The preparing phase began in October 2018. It included cross-sectoral meetings, nominating an MMG, creating a concrete

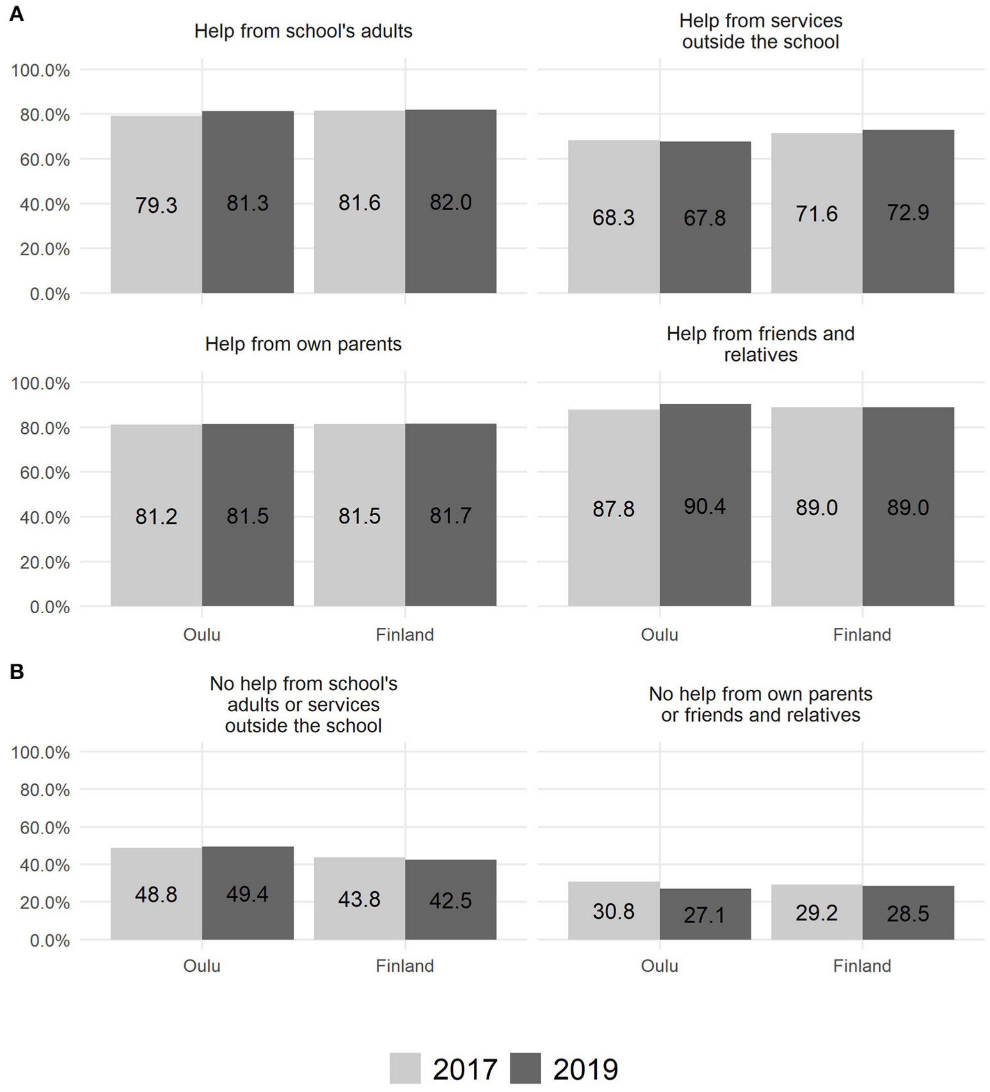


FIGURE 2 | Experience of receiving **(A)** and not receiving **(B)** help for perceived symptoms of depression among 14–16-year-old adolescents ($n = 628$).

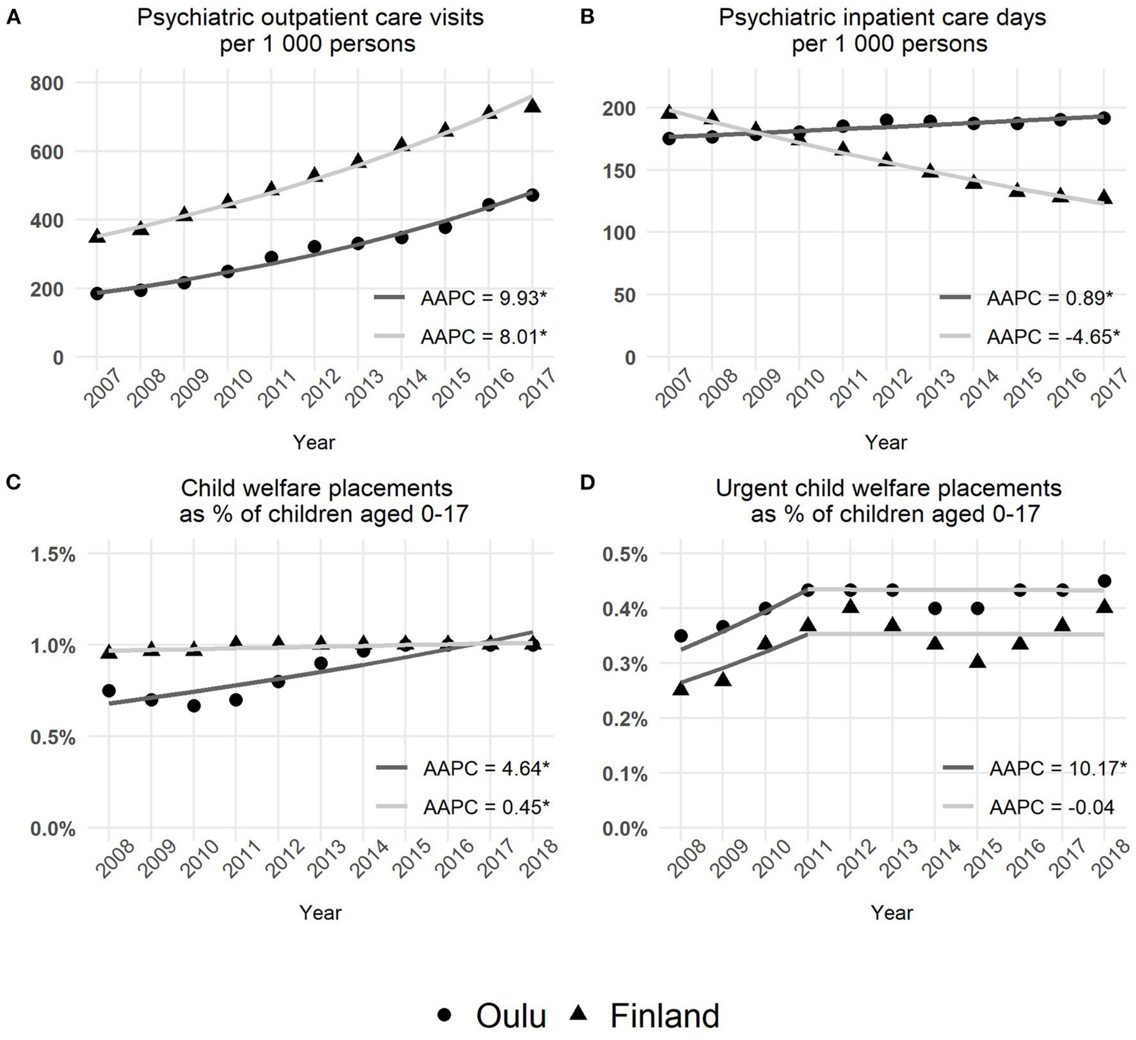
action plan for implementation, and determining which research to include.

Organizing Starting Point Meetings

In October 2018, three starting point meetings were arranged to prepare for the implementation. The participants were the core local leaders of the pilot area (WSA) from daycare, education, social and healthcare services, and as backbone support, an external consultant with prior extensive experience in the LT-SM implementation process. At the conclusion of the meetings, commonly accepted shared goals were set: these comprised supporting children's everyday lives in all service sectors and in the following developmental environments: daycare, school, and leisure time. This goal was accepted by the division leaders of the city of Oulu and all local leaders, who committed to organize the required meetings, seminars, and trainings. In addition, scientific research was integrated into the implementation process from the start (**Figure 5**).

Nominating the MMG

In October 2018, the MMG consisting of core leaders from different sectors in the pilot area (WSA) was nominated, aiming to ensure that the units were committed to fulfilling the implementation tasks and providing consultation and support to the professionals and unit leaders. The MMG drew up a concrete step-by-step action plan on how to take into consideration the views of the unit leaders and professionals in all sectors. In addition, the MMG informed and organized training for professionals aimed to enhance and encourage communication between participants. The MMG was instructed to collect and analyze feedback from the units on a regular basis and perform all necessary actions based on the feedback.



* Indicates that the Annual Percentage Change (AAPC) is significantly different from zero at the alpha = 0.05 level.

FIGURE 3 | Use of child and adolescent psychiatric services and child protection services. **(A)** The number of outpatient care visits per 1,000 persons. **(B)** The number of inpatient care days per 1,000 persons. **(C)** The percentage rates of child welfare care placements. **(D)** The percentage rates of urgent child welfare care placement.

Working Phase I

From January to June 2019, the MMG met six times (i.e., once per month). The MMG prepared and organized seminars to activate stakeholders from different sectors and explain which concrete tasks were intended to be carried out by particular professionals.

Activating Stakeholders

The first meeting for sharing information and mutual conversation was organized at the beginning of 2019. At least one leader or representative per unit and professionals from all services, daycare, schools, social and healthcare services, and student welfare services attended the meeting. During this meeting, the implementation plan prepared by MMG was introduced, and the proposed common agenda for the intended project was described.

Based on written feedback memos from the meetings, collaboration between daycare, education, and social and healthcare services was ranked according to importance by the participants. Eliminating delays in accessing child and family services was highlighted in particular.

During spring 2019, four information and open conversation meetings were arranged for the target group, which enabled a mutual understanding of what the implementation process required from the point of view of professionals, units, and organizations in different sectors. The background theories and core principles of the implementation were introduced. In

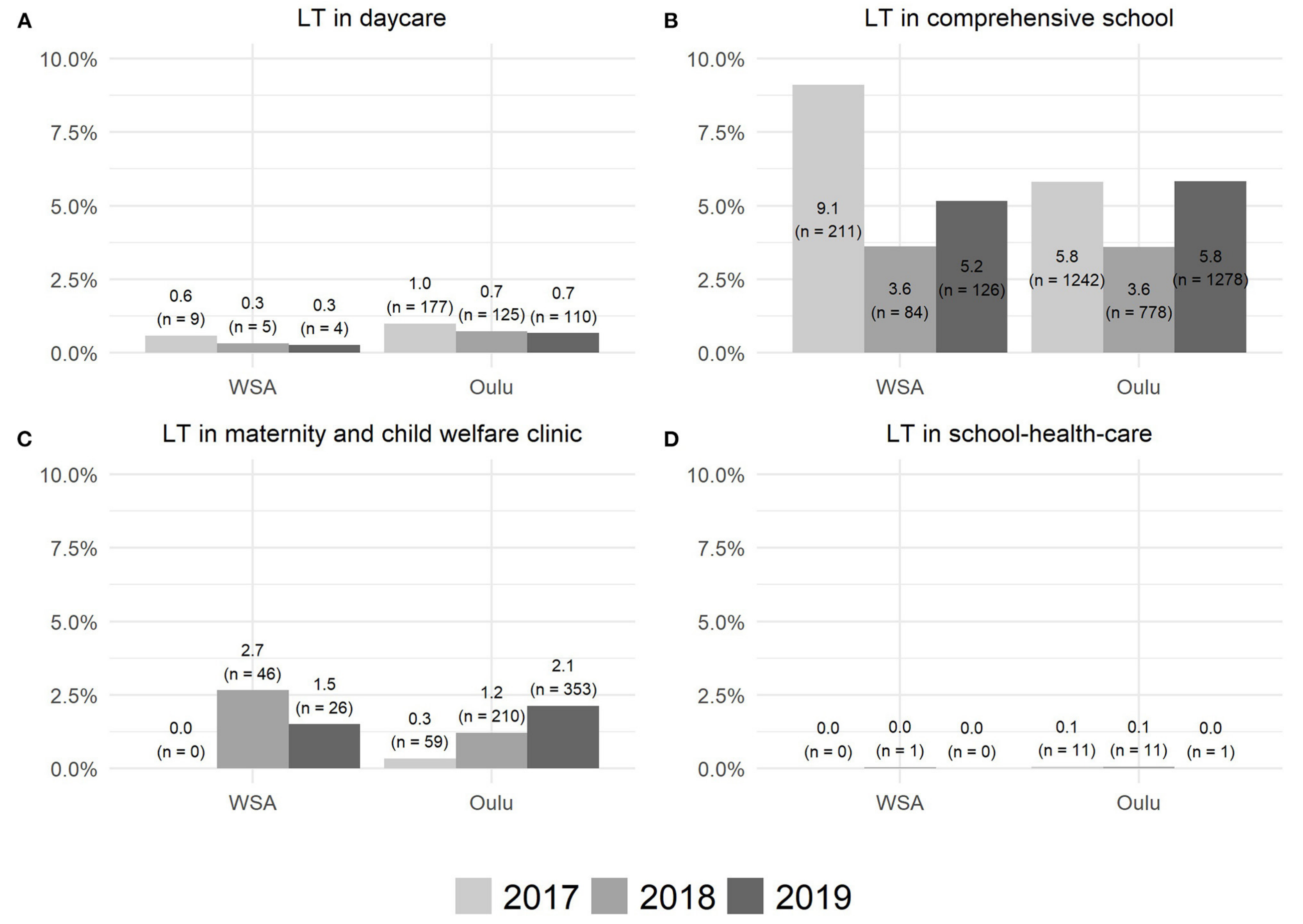


FIGURE 4 | The use of the LT intervention in daycare **(A)**, comprehensive school **(B)**, maternity and child welfare clinic **(C)**, and school-health-care **(D)**.

particular, the professionals' opinions on how to develop the services were of interest.

Conducting the Survey

In spring 2019, the core stakeholders were surveyed with open questions administered to the professionals and units to determine their opinions of the necessary actions on conducting the implementation process. Altogether 61 responses were received from 9 unit meetings, with ∼4–15 participants at each meeting. The participants were asked to describe how to enhance the availability of social and healthcare services and cross-sectoral collaboration between different service sectors in practice. The content was analyzed by bringing together the main themes that were most frequently brought up. The responses highlighted that (1) there is a need to build a shared unified service model because the current collaboration does not work appropriately and there is a need for coordination of the services; (2) cross-sectoral collaboration should be based on a common and united agenda to which all sectors will commit; (3) the commitments should be reliable and such that all participants can trust them; (4) there should be strict follow-up on how the guidelines of the model have been fulfilled; (5) the service model should enhance goal-oriented and responsible work; (6) the service model should allow families to be in key role in the collaboration; (7) there should be a simple method to invite professionals into the collaboration; and (8) the professionals emphasized the importance of knowing each other and of lively exchange of information between collaborative partners.

Establishing OCS

The MMG prepared the OCS model for the child and family services in the pilot area (WSA). The OCS was decided to be placed in the social and family services unit. Students' welfare services and social and healthcare services committed to participate in the LT Network meeting within a predetermined time. Urgent child protection actions (e.g., cases of family violence) continued as usual, according to the Child Protection Act.

Constructing Feedback System

The next task was to systematize the feedback system, which was initiated at the beginning of the implementation process. The feedback system served two purposes: (1) guiding and supporting professionals in different service sectors to fulfill

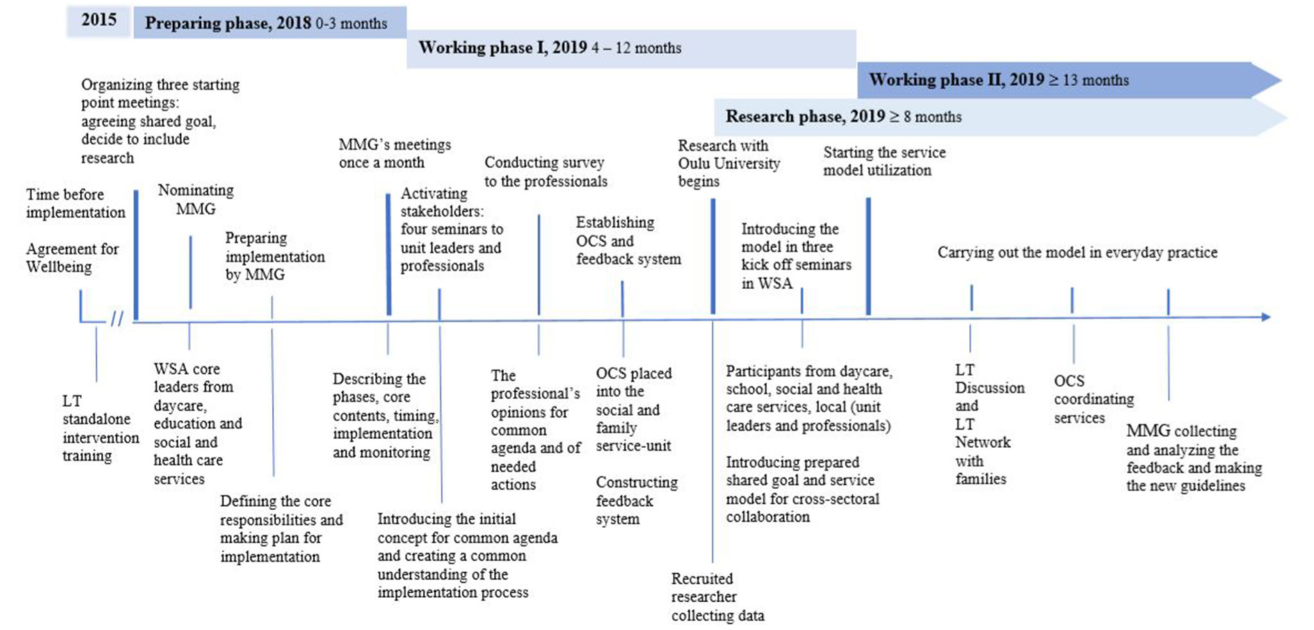


FIGURE 5 | CI implementation process in 2018–2020. LT, Let's Talk About Children intervention; LT-SM, Let's Talk About Children Service Model; CI, collective impact framework; MMG, Multiagent Management Group; OCS, One Contact Service; WSA, the pilot area of Oulu, one regional welfare service area.

their work according to the shared goal and (2) developing collaboration structures according to experiences accumulated from everyday work.

The feedback data (e.g., written feedback, experiences of LT Discussions, etc.) were collected by email from all units once per month before the MMG meeting. The MMG analyzed the feedback and the numerical usage data of the LT intervention and prepared the actions and new guidelines according to the feedback as necessary.

Introducing the LT-SM Model in Kickoff Seminars

After the preparation phase and working phase I, the LT-SM was introduced to all professionals in all service sectors: daycare, schools, social and healthcare services, and student welfare services. The concrete utilization of the model began on September 2, 2019.

The concrete service model was formed as follows:

When the professionals close to the child noticed stressful changes in a child's life (e.g., behavioral problems, parental illness, etc.), they were instructed to do the following:

1) Contact the child and parents: get a picture of the child's current situation.

2) Meet with children and parents, use LT intervention if possible, and include everyday adults in the child's life.

3) Use OCS, if necessary, for social and healthcare professionals: utilize required services.

4) Arrange an LT Network meeting: make concrete action plans for helping the child and family in collaboration with separate service sectors.

5) Arrange LT Network meetings with as many follow-ups as required: ensure reliable collaboration and services for the family.

In August 2019, the MMG organized three kick-off seminars to introduce the information to professionals in all sectors. The professionals from the OCS group participated in the seminars as well to familiarize them with future collaboration. The MMG introduced the prepared service model based on professional feedback and opinions. After the seminars, each unit was informed about the start of concrete CI service model utilization by sending out handouts with a detailed description of the main principles and agreed actions during the implementation process.

Research Phase

The research was included in the implementation process during working phase I. As a result of the negotiations with the University of Oulu, one researcher began to gather documentation data of the implementation process and baseline statistics before the implementation.

Working Phase II

Working phase II was initiated in September 2019. In working phase II, the aim was to conduct everyday practice and management as discussed. Thus, LT Discussions were meant to be conducted with families when there were changes in the family situation or problems with the child. In addition, OCS was to coordinate the required services to support the child and the close adults to manage these challenges. MMG collected and analyzed monthly feedback to develop the collaboration further.

DISCUSSION

Psychosocial wellbeing of children and adolescents is an ongoing societal concern (38, 39). Despite various local and national efforts and programs to promote the psychosocial wellbeing of young people, several child and adolescent health and social care indicators demonstrate concerning trends. Although child- and family-focused programs have generally reported positive outcomes (10–12), they have been unable to respond to the challenges caused by the fragmentation of services and separate service models. These concerns are acknowledged as a major contributor to the problems in the service sector.

In the present study, we describe in detail the implementation of the LT-SM in the pilot area, one regional WSA of the city of Oulu. This model is assumed to unify policies and solve major problems caused by the fragmented services. The follow-up data and forthcoming analyses of the model will produce research-based evidence on the efficacy of such a model. To demonstrate the rationale for the implementation of this model, we first evaluated various population-based child and adolescent health and social care indicators and the results of a nationwide school-health survey in the study area (city of Oulu) in comparison with those of the average of the whole country. We also explored the numerical usage data of the stand-alone LT intervention in the study area (city of Oulu) in the period before the implementation of the LT-SM.

We found that the number of child and adolescent psychiatric inpatient days has increased in the study area (city of Oulu), whereas in the whole country, the respective rates have decreased. Furthermore, psychiatric outpatient visits have increased in this area, but the level has remained lower when compared with that in Finland as a whole. These findings are consistent with the previous reports of increased mental health service use among children and adolescents (40–42). A high level of use in inpatient psychiatric services and a nationally low level in outpatient service use in the study area are a concerning finding and advocate for more effective preventive and outpatient-oriented service approaches. In addition to healthcare services, urgent child welfare placements by social services were at a notably higher level in the study area compared with the average of the whole country. This is alarming because the rates of child welfare placements in Finland are already high in international comparisons, and they are also regarded as too high by national professional and scientific communities (4, 38, 39). This alarming finding from the study area (city of Oulu) may be due to the nationally low rates of use of children and adolescent outpatient mental health services in the city of Oulu. Primarily, this may suggest not only insufficient levels of services in this area, but also unwillingness to seek help.

We found that nationally, approximately one-fifth of the adolescents who responded to the nationwide SHP survey reported that they suffered from depressive symptoms. A large majority of respondents reported that they received help from their close relatives and friends. With regard to professionals, adolescents reported most commonly that they received support from school personnel, such as teachers and school welfare professionals, but less commonly from social and healthcare services. On the other hand, almost half of the respondents who reported a need for help with their depressive symptoms had not received professional help. This lack of support was more common among adolescents from the study area (city of Oulu) compared with the national average. This indicates a lack of necessary services. The perceived need for support among adolescents is consistent with the increased trends observed in the use of adolescent social and healthcare services (40–42).

A worrisome finding was that before the implementation of the comprehensive LT-SM, the use of the stand-alone LT intervention in the study area (city of Oulu) as a single preventive method decreased to the minimum level soon after its initiation. This finding confirmed the previously known challenge of the implementation of stand-alone interventions, that is, that single interventions rarely remain part of everyday practice despite proper training and other implementation efforts (43).

Furthermore, our results on the epidemiological data are consistent with the previously internationally recognized need for preventive psychosocial and outpatient mental health services for children and adolescents (44, 45). The city of Oulu has attempted to respond to these challenges of preventive work with local actions (46) and by participating in national child and family development programs (10–12). The current study clearly demonstrated that despite these actions, the use of institutionalized services among adolescents has remained at high level. At the national level, the need for cross-sectoral collaboration between professionals has been a subject of ongoing debate. Even in Finnish legislation, there are regulations calling for collaboration between service sectors in areas such as education, student welfare, and social and healthcare (Basic Education Act 628/1998; Student Welfare Act 1287/2013; Social Welfare Act 1301/2014; Healthcare Act 1326/2010). However, there is no clear roadmap for professionals from different sectors on how to fulfill these requirements.

According to Kania and Kramer (27), CI framework initiatives have successfully established collaboration between services surrounding various initiatives such as HIV prevention and food and obesity programs (20). In Finland, promising results have been obtained from psychosocial children and family-focused preventive work when applying the LT-SM based on the CI framework (21).

In the implementation process of the LT-SM described in this article, the common agenda (i.e., supporting the everyday life of children and families) on collaboration with social and health care services and developmental environments was determined at the beginning of the preparation phase. The implementation process in the pilot area followed the principles of CI-related literature, which highlights the significance of a common agenda as a first condition for successful implementation. It is usually determined by core actors around the same topic who have connections with relevant stakeholders (47–49).

In the pilot area of Oulu, as part of the implementation process, the MMG collected monthly feedback from all collaborative units. Feedback-related communication in the MMG revealed the need for the required actions, including various common agenda-related discussions with professionals on how they can apply the common goal in their everyday work in different sectors. This was carried out in accordance with previous research literature where feedback-based measurement and communication were regarded as essential to lead the CI process and build collaboration (29, 50, 51).

In the current implementation process, the role of the OCS (20) was emphasized in terms of reinforcing activities (27). OCS invited the appropriate participants to the LT Network meetings based on contacts (e.g., from school and daycare). Consequently, the professionals in the OCS became aware of the needs and services required for the families. This information, together with general feedback collected from the units and service use statistics, helped the MMG focus on necessary activities. The OCS helped to integrate the actions of different services by inviting professionals to collaborate during network meetings. In summary, the OCS provided a possibility to increase integration and prevention within the services.

The role of backbone organization was essential in the implementation process in the pilot area of Oulu, which is consistent with previous recommendations in studies highlighting the need of backbone in collaborative organizations (20). Overall, backbone organizations are related to project funding (52), non-governmental and intermediary organizations to collaboration with various actors (53), and local administration to relevant stakeholders (54). In the pilot area, the MMG was the backbone of the whole organization because its members knew the whole implementation process, were able to facilitate the required actions, and monitor that all CI conditions were fulfilled.

The literature of the CI framework recognizes four steps in the change of collaboration into practice: (1) fulfilling the five CI conditions, (2) early changes and their connections with CI conditions, (3) systems changes in the core organizations and their connections with early changes and CI conditions, and (4) population changes and their connections with systems changes (55). The implementation process described in the current article is the first step toward the change of collaboration between the service sectors. Thus, profound population-level changes, such as a decrease in the rates of child welfare placements and adolescent psychiatric hospitalizations in the city of Oulu, will take time (30, 55).

Strengths and Limitations

To our knowledge, this is the first detailed description of the CI implementation process aimed at child and family service integration in Nordic WSAs. The nationwide register data on the use of child and adolescent services as well as survey data on depressive symptoms and support perceived by adolescents were obtained from the SOTKANET databank, which has been acknowledged to be reliable for research purposes (56). This epidemiological data were related to the period prior to the implementation of the LT-SM in one WSA of the city of Oulu. The numerical data on the use of the stand-alone LT intervention in the pilot area were very limited and do not allow more detailed analyses, such as how many parents started using the model but did not finish it. The number of cases for **Figure 2** was not available for our study. However, we believe that **Figure 2** serves well to illustrate that the majority of the adolescents had received support and help from someone in their everyday life, but there is also a notable number of adolescents reporting need for help with their mental health who have not received it.

The present study clearly demonstrated a high rate of use of curative services in social and health care and insufficient usage of the stand-alone preventive intervention. The process described in this article indicates that a comprehensive CI-based service model can be implemented in the municipal service system, including all existing sectors. In this way, separate services are likely able to act as an integrated service entity. The effectiveness of the service model will be evaluated in the future when enough follow-up data have been accumulated for justifying reliable statistical analyses.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

AUTHOR CONTRIBUTIONS

TT and MN were responsible for the implementation process. TT and MN wrote the first draft of the article. SR provided consultation during the implementation process, including research into it, and made significant contributions to the manuscript. HH and AJ were responsible for statistical analyses, including reporting and interpretation of the results based on these analyses. All authors participated in critical drafting of the manuscript and approved the final version of the manuscript and agreed to be accountable for all aspects of the work to ensure that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

FUNDING

The scholarship of TT (2020–2021) and professorship of MN (2020–2023) have been funded by ITLA Children's Foundation.

REFERENCES

1. Ketchen Lipson S, Lattie EG, Eisenberg D. Increased rates of mental health service utilization by U.S. College students: 10-year population-level trends. (2007–2017). *Psychiatr Serv*. (2018) 70:60–3. doi: 10.1176/appi.ps.201800332
2. McManus S, Gunnell D, Cooper C, Bebbington PE, Howard LM, Brugha T, et al. Prevalence of non-suicidal self-harm and service contact in England, 2000–14: repeated cross-sectional surveys of the general population. *Lancet Psychiatry.* (2019) 6:573–81. doi: 10.1016/S2215-0366(19)30188-9
3. Kaltiala-Heino R, Marttunen M, Fröjd S. Lisääntyvätkö nuorten mielenterveyden ongelmat? *Suom Laakaril.* (2015) 70:1908–12.
4. Kääriälä A, Hiilamo H. Children in out-of-home care as young adults: a systematic review of outcomes in the Nordic countries. *Child Youth Serv Rev.* (2017) 79:107–14. doi: 10.1016/j.childyouth.2017.05.030
5. Tamhne RC. Who is responsible for child mental health? The increasing fragmentation of health services for children is the main problem. *BMJ.* (1997) 315:310–1. doi: 10.1136/bmj.315.7103.310a

6. Horwath J, Morrison T. Collaboration integration and charge in children's services: critical issues and key ingredients. *Child Abuse Negl.* (2007) 31:55–69. doi: 10.1016/j.chiabu.2006.01.007
7. Lelliott P, Gallagher U. *Review of Children and Young People's Mental Health Services. Phase One Report.* (2017) Care Quality Commission. Review of children and young People's mental health services. 10/2017. Available online at: https://www.cqc.org.uk/sites/default/files/20171103_cypmhphase1_report.pdf (accessed October 10, 2020).
8. Heinonen O-P, Ikonen A-K, Kaivosoja M, Reina T. *Yhdyspinnat yhteiseksi mahdollisuudeksi - Selvitys lapsi- ja nuoriso- ja perhepalveluiden toteuttamiseen liittyvistä yhdyspinnoista muuttuvassa toimintaympäristössä 8/2018.* (2018). Available online at: http://urn.fi/URN:ISBN:978-952-00-3904-2 (accessed October 10, 2020).
9. Frith E. *Progress and Challenges in the Transformation of Children and Young People's Mental Health Care: 17 August 2016.* Education Policy Institute 2016 (2016). Available online at: http://dera.ioe.ac.uk/id/eprint/27929.
10. KASTE (2008–2011). Lähteenmäki-Smith K, Terävä E. *Sosiaali- ja terveydenhuollon kansallisen kehittämisohjelman (KASTE) 2008–2011 arviointi. Loppuraportti. National Development Program for Social and Health Care. Ministry of Social Affairs and Health. Sosiaali- ja terveysministeriön julkaisuja 2012:12. Sosiaali- ja terveysministeriö.*. Helsinki (2012). Available online at: https://julkaisut.valtioneuvosto.fi/bitstream/handle/10024/73326/URN%3ANBN%3Afife201504224195.pdf?sequence=1 (accessed October 10, 2020).
11. KASTE (2012–2015). Kallinen S. *(toim.) Sosiaali- ja terveydenhuollon kansallinen kehittämisohjelma KASTE 2012–2015. Loppuraportti. Sosiaali- ja terveysministeriön raportteja ja muistioita 2016:8. Sosiaali- ja terveysministeriö*. Helsinki 2016. National Development Program for Social and Health Care. Ministry of Social Affairs and Health (2016). Retrieved from: http://julkaisut.valtioneuvosto.fi/bitstream/handle/10024/74927/Rap%20ja%20mui_2016_8_PDF__.pdf?sequence$=$1&isAllowed=y (accessed October 10, 2020).
12. LAPE. *The program to address Child and Family Services*. Available online at: https://stm.fi/documents/1271139/4067344/LAPE_k%C3%A4rkihanke_loppuraportti_2019.pdf/87d61c67-cf44-75a7-7d4f-cc5d0b258b8c/LAPE_k%C3%A4rkihanke_loppuraportti_2019.pdf.SummaryofLAPE. Available online at: https://stm.fi/documents/1271139/3208911/LAPE-esite-verkko_ENG_web.pdf/b13d5d01-6ee6-464b-9dbe-399cb018e9da/LAPE-esite-verkko_ENG_web.pdf. Ministry of Social Affairs and Health (2016–2019) (accessed October 10, 2020).
13. Webster-Stratton C. *Incredible Years®. Parents, Teachers and Children's Training Series. Program Content, Methods, Research and Dissemination 1980–2011.* (2011). Available online at: http://www.incredibleyears.com/ (accessed October 10, 2020).
14. McGrath PJ, Lingley-Pottie P, Thurston C, MacLean C, Cunningham C, Waschbusch DA, et al. Telephone-based mental health interventions for child disruptive behavior or anxiety disorders: randomized trials and overall analysis. *J Am Acad Child Adolesc Psychiatry.* (2011) 50:1162–72. doi: 10.1016/j.jaac.2011.07.013
15. McGrath PJ, Sourander A, Lingley-Pottie P, Ristkari T, Cunningham C, Huttunen J, et al. Remote population-based intervention for disruptive behavior at age four: study protocol for a randomized trial of Internet-assisted parent training (Strongest Families Finland–Canada). *BMC Public Health.* (2013) 13:985. doi: 10.1186/1471-2458-13-985
16. Glick B, Goldstein AB. Aggression replacement training. *J Counsel Dev.* (1987) 65:356–62. doi: 10.1002/j.1556-6676.1987.tb00730.x
17. Solantaus T, Toikka S, Alasuutari M, Beardslee WR, Paavonen EJ. Safety, feasibility and family experiences of preventive interventions for children and families with parental depression. *Int J Ment Health Promot.* (2009) 11:15–24. doi: 10.1080/14623730.2009.9721796
18. Solantaus T, Paavonen EJ, Toikka S, Punamäki RL. Preventive interventions in families with parental depression: children's psychosocial symptoms and prosocial behavior. *Eur Child Adolesc Psychiatry.* (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
19. Stange KC. The problem of fragmentation and the need for integrative solutions. *Ann Fam Med.* (2009) 7:100–3. doi: 10.1370/afm.971
20. Virtanen P, Ristikari T, Niemelä M. Partnership and backbone organizations as enablers of children's wellbeing. In: Leal Filho W, editor. *Encyclopedia of the UN Sustainable Development Goals.* Cham: Springer. (2020). p. 1–14. doi: 10.1007/978-3-319-71067-9_111-1
21. Niemelä M, Kallunki H, Jokinen J, Räsänen S, Ala-Aho B, Hakko H, et al. Collective impact on prevention: let's talk about children service model and decrease in referrals to child protection services. *Front Psychiatry.* (2019) 10:64. doi: 10.3389/fpsyt.2019.00064
22. Allchin B, Goodyear M, O'Hanlon B, Weimand BM. Leadership perspectives on key elements influencing implementing a family-focused intervention in mental health services. *J Psychiatr Ment Health Nurs.* (2020) 27:616–27. doi: 10.1111/jpm.12615
23. Maybery D, Goodyear M, Reupert A, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry.* (2017) 17:198. doi: 10.1186/s12888-017-1357-4
24. Ueno R, Osada H, Solantaus T, Murakoshi A, Inoue T. Safety, feasibility, fidelity, and perceived benefits of an intervention for parents with mood disorders and their children - "Let's Talk about Children" in Japan. *J Fam Psychother.* (2019) 30:272–91. doi: 10.1080/08975353.2019.1678092
25. Niemelä M, Repo J, Wahlberg KE, Hakko H, Räsänen S. Pilot evaluation of the impact of structured child-centered interventions on psychiatric symptom profile of parents with serious somatic illness: struggle for life trial. *J Psychosoc Oncol.* (2012) 30:316–30. doi: 10.1080/07347332.2012.664258
26. Niemelä M, Marshall CA, Kroll T, Curran M, Koerner SS, Räsänen S, et al. Family- focused preventive interventions with cancer co survivors: a call to action. *Am J Public Health.* (2016) 106:1381–7. doi: 10.2105/AJPH.2016.303178
27. Kania J, Kramer M. Collective impact. *Stanford Soc Innov Rev.* (2011) 9:36–41. doi: 10.48558/5900-KN19
28. Kania J, Kramer M. Embracing emergency: how collective impact addresses complexity. *Stanford Soc Rev.* (2013). doi: 10.48558/ZJY9-4D87
29. Hanleybrown F, Juster JS, Kania J. (2014). Essential mindset shifts for collective impact. *Stanford Soc Innov Rev.* (2014). 12, A2–A5. doi: 10.48558/VV1R-C414
30. Stachowiak S, Gase L. Does collective impact really make an impact? *Stanford Soc Innov Rev.* (2018) 9. doi: 10.48558/6GD9-MB47
31. Solantaus T, Niemelä M. Arki kantaa - kun se pannaan kantamaan. *Perheterapia.* (2016) 32:21–33.
32. Hanleybrown F, Kania J, Kramer M. Channeling change: making collective impact work. *Stanford Soc Innov Rev.* (2012) 26. doi: 10.48558/2T4M-ZR69
33. SHP. *The School Health Promotion.* (2020). Available online at: https://thl.fi/en/web/thlfi-en/research-and-expertwork/population-studies/school-healthpromotionstudy. The Finnish Institute for Health and Welfare (accessed August 8, 2020)
34. Kroenke K, Spitzer RL, Williams JB. The Patient Health Questionnaire-2: validity of a two item depression screener. *Med Care.* (2003) 41:1284–92. doi: 10.1097/01.MLR.0000093487.78664.3C
35. Mieli. *Mental Health Finland. Let's Talk Child (LT).* (2020). Available online at: https://mieli.fi/en/let%E2%80%99s-talk-about-children-lt (accessed October 10, 2020).
36. Pohjois-Pohjanmaa. Available online at: https://www.hyvinvointikertomus.fi/ci/user_uploads/1639/MUUTOS_NYT_HYVINVOINTISOPIMUS.pdf.Agreementforwellbeing2013--2017. *Muutos nyt Hyvinvointisopimus* (2013–2017) (accessed October 10, 2020).
37. Oulu Open Data (2020). Available online at: https://data.ouka.fi/en_gb/ (accessed August 8, 2020).
38. Kestilä A, Paananen R, Väisänen A, Muuri A, Merikukka M, Heino T, et al. Kodin ulkopuolelle sijoittamisen – rekisteripohjainen seurantatutkimus Suomessa vuonna 1987 syntyneistä. *Yhteiskuntapolitiikka*. 77:34–52. Available online at: https://urn.fi/URN:NBN:fi-fe201209117910
39. Kuoppala T, Forsell M, Säkkinen S. *Lastensuojelu 2018. Tilastoraportti 23/2019. Terveyden ja hyvinvoinnin laitos.* (2019). Available online at: https://www.julkari.fi/bitstream/handle/10024/138211/Tr23_19_LASU.pdf?sequence=5 (accessed November 30, 2020].

40. Olfson M, Druss BG, Marcus SC. Trends in mental health care among children and adolescents. *N Engl J Med.* (2015) 372:2029–38. doi: 10.1056/NEJMsa1413512
41. Ringeisen H, Miller S, Munoz B, Rohloff H, Hedden SL, Colpe LJ. Mental health service use across adolescence: findings from the national survey on drug use and health. *Psychiatr Serv*. (2016) 67:787–9. doi: 10.1176/appi.ps.201400196
42. Filatova S, Upadhyaya S, Kronström K, Suominen A, Chudal R, Luntamo T, et al. Time trends in the incidence of diagnosed depression among people aged 5–25 years living in Finland 1995–2012. *Nord J Psychiatry*. (2019) 73:475–81. doi: 10.1080/08039488.2019.1652342
43. Beardslee WR, Versage EM, Gladstone TR. Children of affectively ill parents: a review of the past 10 years. *J Am Acad Child Adolesc Psychiatry*. (1998) 37:1134–41. doi: 10.1097/00004583-199811000-00012
44. Lawrence D, Johnson S, Hafekost J, Boterhoven de Haan K, Sawyer M, Ainley J, et al. *The Mental Health of Children and Adolescents. Report on the Second Australian Child and Adolescent Survey of Mental Health and Wellbeing.* (2015). Available at: https://www1.health.gov.au/internet/main/publishing.nsf/content/9DA8CA21306FE6EDCA257E2700016945/\protect\T1\textdollarFile/child2.pdf. Australian Government (accessed November 30, 2020).
45. Lipari RN, Hedden S, Blau G, Rubenstein L. *Adolescent Mental Health Service Use and Reasons for Using Services in Specialty, Educational, and General Medical Settings*. CBHSQ Rep: May 5, 2016. Rockville, MD: Center for Behavioral Health Statistics and Quality, Substance Abuse and Mental Health Services Administration (2016). Available online at: https://www.ncbi.nlm.nih.gov/books/NBK362074/ (accessed October 5, 2020).
46. Oulu. *Lasten ja nuorten hyvinvointisuunnitelma.* Oulun Kaupunki (2018). Available online at: https://www.ouka.fi/documents/64277/104164/Hyvinvointisuunnitelma_2018_2021.pdf/dc30f99b-4228--4685-987c-75f18132018a (accessed August 29, 2020)
47. Buchbinder SP, Havlir DV. Getting to zero San Francisco: a collective impact approach. *J Acquir Immune Defic Syndr.* (2019) 82(Suppl. 3):S176–82. doi: 10.1097/QAI.0000000000002200
48. Terrile VC. Public library support of families experiencing homelessness. *J Child Pover.* (2016) 22:133–46. doi: 10.1080/10796126.2016.1209166
49. Wills R, Gabriel B, Matthews KM. The Ngatahi project: competency development for the vulnerable children's workforce. *Policy Q.* (2019) 15:73–9.
50. Gillam RJ, Counts JM, Garstka TA. Collective impact facilitators: how contextual and procedural factors influence collaboration. *Commun Dev.* (2016) 47:209–24. doi: 10.1080/15575330.2015.1133684
51. Raderstrong J, Boyea-Robinson T. The why and how of working with communities through collective impact. *Commun Dev.* (2016) 47:181–93. doi: 10.1080/15575330.2015.1130072
52. Calderon B. Achieving "first in the world" hispanic serving institutions and closing attainment gap. *Harvard J Hispanic Policy*. (2014) 27:60–4.
53. Weaver NL, Thompson J, Shoff CR, Copanas K, McMillin SE. A conceptual model for pathways of effect for intermediary organizations: a case study from maternal and child health. *Eval Program Plann.* (2017) 63:69–73. doi: 10.1016/j.evalprogplan.2017.03.006
54. Cox M. Logan together: how working across sectors for our kids is the answer to everything. *Int J Integr Care.* (2018) 18:1. doi: 10.5334/ijic.s1062
55. Lynn J, Gase L, Roos J, Oppenheimer SD, Stachowiak S, Akey T, et al. *When Collective Impact Has an Impact. A Cross-Site Study of 25.* Seattle, WA: Collective Impacts Initiatives. Spark Policy Institute and ORS Impact of Seattle.
56. Miettunen J, Suvisaari J, Haukka J, Isohanni M. Use of registers data for psychiatric epidemiology in the Nordic countries. In: Tsuang MT, Tohen M, Jones PB editors. *Textbook in Psychiatric Epidemiology.*, 3rd Edn. West Sussex: John Wiley & Sons, Ltd (2011) 117–31. doi: 10.1002/9780470976739.ch8

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

FURTHER READING

Nordic Social Statistical Committee. Social Protection in the Nordic Countries 2013/2014. Scope, Expenditure and Financing. Nordic Social Statistical Committee (2015) (NOSOSCO) 60:2015. Copenhagen, Denmark. [Accessed 5.10.2020]. Available at: https://norden.diva-portal.org/smash/get/diva2:1059435/FULLTEXT02.pdf.

Social welfare act 1301/2014. [Accessed 30.11.2020]. Available at: https://finlex.fi/en/laki/kaannokset/1982/en19820710. Finland: The Ministry of Social Affairs and Health.

Basic education Act 628/1998. [Accessed 30.11.2020]. Available at: https://finlex.fi/en/laki/kaannokset/1998/en19980628?search%5Btype%5D=pika&search%5Bkieli%5D%5B0%5D=en&search%5Bpika%5D=628%2F1998. Finland: The Ministry of Education and Culture.

Health care act 1326. [Accessed 30.11.2020]. Available at: https://www.finlex.fi/en/laki/kaannokset/2010/en20101326?search%5Btype%5D=pika&search%5Bkieli%5D%5B0%5D=en&search%5Bpika%5D=1326%20%2F%202010. Finland: The Ministry ofSocial Affairs and Health (2010).

Student welfare act 1287. [Accessed 30.11.2020]. Available at: https://www.finlex.fi/en/laki/kaannokset/2013/en20131287?search%5Btype%5D=pika&search%5Bkieli%5D%5B0%5D\protect$\relax=$en&search%5Bpika%5D\protect$\relax=$student%20Welfare%20. Finland: The Ministry of Education and Culture (2013).

Social and healthcare services in Finland: Expert Evaluation. Autumn 2018. (ed.) Rissanen, P. 5/2019. [Accessed 30.10.2020]. Available online at: https://urn.fi/URN:ISBN:978-952-343-360-1

sotkanet.fi. Statistical information on welfare and health in Finland. [Accessed 5.10.2020]. Available at: https://sotkanet.fi/sotkanet/en/haku?g=354. National Institute for Health and Welfare(2020).

The City of Oulu. The Department of Educational and Cultural Services and the Department of Well-Being Services. The City of Oulu. Non-Published Material, 2017–2019 (2020). The statistics of the Let's Talk about Children Intervention years.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 09 March 2022
doi: 10.3389/fpsyt.2022.778236

Support for Children of Parents With Mental Illness: An Analysis of Patients' Health Records

Kjersti Bergum Kristensen, Camilla Lauritzen and Charlotte Reedtz*

Regional Center for Child and Adolescent Mental Health and Child Welfare, Faculty of Health Sciences, UiT The Arctic University of Norway, Tromsø, Norway

Introduction: Children of parents with a mental illness (COPMI) are at risk of behavioral, emotional, and cognitive difficulties and diagnoses. Support and information about parents' mental illness may contribute to improve their lives, which is the purpose of the intervention Child Talks (CT). This study aimed to investigate the participation rate of CT, characteristics of participating patients and children, and themes in sessions with children.

Materials and Methods: Data were collected from 424 electronic patient journals written by healthcare professionals (H) for patients admitted to a clinic for mental health and substance use disorders in the years 2010–2015. Both quantitative statistical analysis and qualitative thematic analysis were carried out.

Results: Eighteen percent of assessed parents with minor children received the CT intervention and children participated in half of them. Participating children more often knew about their parent's treatment and condition when initially assessed, and more often lived with the hospitalized parent. Three main themes were identified in sessions with children; communication about parental mental illness within the family, childrens' struggles, and healthcare professionals' (HCPs) evaluation of the child's situation and need for further support.

Discussion: Sessions with patients' children appeared to be relatively rare, and participating children did not necessarily receive appropriate information, support, or follow-up. To ensure that HCPs provide quality support and follow-up to COPMI, the routines and the training of HCPs need to be improved.

Keywords: children of parents with mental illness, parental mental illness, mental healthcare services for adults, healthcare professionals, preventive intervention, support of patients' children

OPEN ACCESS

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Mary V. Seeman,
University of Toronto, Canada
Jessica Radley,
University of Oxford, United Kingdom

Correspondence:
Kjersti Bergum Kristensen
Kjersti.b.kristensen@uit.no

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *16 September 2021*
Accepted: *14 February 2022*
Published: *09 March 2022*

Citation:
Kristensen KB, Lauritzen C and Reedtz C (2022) Support for Children of Parents With Mental Illness: An Analysis of Patients' Health Records. Front. Psychiatry 13:778236. doi: 10.3389/fpsyt.2022.778236

Abbreviations: CAMHS, The Child and Adolescent Mental Health Services; COPMI, Children of parents with a mental illness; CT, Child Talks; CWPS, The Child Welfare and Protection Services; DMHSD, The Division for Mental Health and Substance Use Disorders; HCPs, healthcare professionals; PMI, Parental mental illness.

INTRODUCTION

In Norway, 12.2% of children have parents who are receiving treatment for mental illness and/or alcohol use disorder each year (1). According to Norwegian (2), and international estimates (3) approximately one third of patients in adult psychiatric services are parents of minor children. Children of parents with a mental illness (COPMI) have an elevated risk of developing emotional, behavioral, and cognitive difficulties that can result in poorer life outcomes regarding educational level, ability to work, socioeconomic status, and ability to establish meaningful relationships with others (4–8). COPMI are at risk of developing the same illness as their parents, as well as other mental illnesses (9). Half of the children of parents with a severe mental illness (SMI) are at risk of developing a mental illness themselves by the age of 20, and one third are at risk of developing a SMI (10).

The transmission of mental illness from one generation to the next is a complex process. Such transmission is influenced by the interaction of factors related to the mentally ill parent, the child, the family, and the social environment (11). Protective factors can reduce the prevalence and/or severity of problems for COPMI (12). Supportive relationships, coping skills, positive relationships between parents, well-functioning communication within the family, and high socio-economic status are examples of such protective factors (13, 14). Several studies and meta-analysis have found significant effects of preventive interventions for COPMI (15–18). The results imply that preventive interventions with a psychoeducational focus reduce the risk for psychopathology and psychiatric symptoms and increase prosocial behavior for COPMI (15–18).

Knowledge about mental health provides resilience against mental illness (14, 19). Children who receive accurate, non-stigmatizing information about parental mental illness (PMI), treatment, and recovery may be able to understand their parent's behavior, talk to others about their situation, and feel less alone (20). Knowledge and openness about PMI may reduce the stigma and burden of worrying for their parent and make it easier for children to seek professional help (14, 21). A lack of information about PMI can cause misunderstandings and misattributions of the causes of parent's behaviors and treatment, and may increase feelings of concern, confusion, and stress for these children (14). In studies of which information COPMI value, children reported that they preferred to learn about PMI from healthcare professionals (HCPs) and regarded support and information as helpful (22, 23). They valued opportunities to ask questions and wanted to learn about the organization of health services. Several children wanted to be assured that it was not their fault that their parent was ill (22). Children expressed that they needed information about what a mental illness is, different types of illnesses, etiology and prognosis, how to cope with parents' symptoms, where to seek help and support, and how to communicate with others about PMI (14).

Child Talks (CT) is a brief preventive intervention, developed in the Netherlands for COPMI aged 0–25 (24, 25). To this date there are no effect studies of the CT intervention. However, the CT has a clear and well-described theoretical foundation, focusing on psychoeducation. The intervention is delivered to patients with a mental illness and their children through three sessions. The patient's child should be included in at least one session, to get information, ask questions and share any concerns. The intervention aims to strengthen parents' knowledge of possible consequences for COPMI and increase parents' focus on the child's situation. By providing children with emotional and social support, and information about their parent's disorder, treatment, and recovery, the intervention aims to reinforce children's ability to cope with their situation. Another objective is to detect early signs of psychopathology and/or problem behaviors in children and initiate further support and referrals if needed. The intervention is manual-based and the sessions are described in detail in the CT manual (24). There is also a Logbook associated with the manual that HCPs should complete during or after CT sessions. The Logbook is described further in section "Child Talks Logbooks."

The content of the CT intervention accords to §10 a) of the Health Personnel Act (26). The CT intervention was implemented in the participating clinic at the University Hospital of Northern Norway (UNN) when the amendments to the Health Personnel Act were made in 2010. The law states that HCPs are obligated to contribute to meet COPMIs need for information and support regarding their parents' diagnoses and treatment. If necessary, HCPs should invite children to participate in a conversation to offer information and support. Despite the legal obligations, studies show that COPMI are not provided with the information they are entitled to (27, 28). Fewer than one third of HCPs had conversations with COPMI (28). Moreover, about 40% of parents in treatment reported that their children were unaware they were receiving treatment or being hospitalized, and over 40% reported that their children were not informed about their condition (27).

Most HCPs have positive attitudes toward a family-focused practice in adult mental health services (29). Still, studies have found numerous barriers for a family-focused practice (28–31). Important predictors for a family-focused practice are worker skills, knowledge, resources, and confidence, whereas families' lack of time and fear of involving children are hindering factors (28, 29, 31, 32). Insecurities among HCPs about their role when meeting patients' children and the lack of knowledge of how to have age-appropriate conversations about PMI with COPMI affected HCPs' tendency to invite children negatively (33).

There is a lack of knowledge about how factors related to the parent and the child influence whether children are given information and support by the parent's HCPs. Little is known about the extent to which COPMI participate in psychoeducational interventions and whether the children who do participate are provided with support, information, and follow-up actions. In this study we aimed to address this knowledge gap by analyzing patients' health records.

The main aim of the present study was to evaluate the performance of CT sessions, with a particular focus on sessions with participating children. More specifically, we aimed to investigate:

(1) parents' participation rate in CT,

(2) children's participation rate in CT, and reasons for their exclusion,
(3) age, gender, and psychosocial differences between participating and non-participating children, and
(4) HCPs' support and information to children.

MATERIALS AND METHODS

Design

This is a retrospective study based on electronic patient journal data for the period 2010–2015. The approval from the data protection officer at UNN allowed us to extract the data from the electronic patient journals in 2015. The study has a mixed-methods approach since both quantitative data from electronic patient journal entries and written reports entered by HCPs to analyses qualitatively were used.

Participants

The total HCPs workforce at the Division for Mental Health and Substance Use Disorders (DMHSD) at UNN was 436 in 2010 (29), whereas 35 HCPs held CT sessions with participating children. Family Assessment Forms were filled out for 424 patients.

Data Material

The data material in this study is information extracted from the Family Assessment Forms and Logbooks from CT sessions, as recorded by HCPs in electronic patient journals. Over the course of the project, two different forms were implemented in the electronic patient journals at the DMHSD at UNN. These two forms were a Family Assessment Form and a Logbook from CT sessions. HCPs were instructed to fill out the Family Assessment Form for patients admitted to the DMHSD who had minor children. Secondly, the patients were to be invited to participate in CT, and HCPs were instructed to write a short report of the sessions in the electronic patient journal, labeled CT Logbooks. Information from these two forms was extracted from the electronic patient journals in 2015.

Family Assessment Forms

The Family Assessment Form consists of five categories of questions: (1) general information about the child, (2) the child's network, (3) concerns for the child, and how the child is coping, (4) the child's knowledge and information about PMI, and (5) the family's need for support and follow-up.

Child Talks Logbooks

Logbooks from cases with participating children were used in the thematic analysis ($n = 39$). In the CT Logbooks, background information such as date, duration of session, place, participants, and parents' diagnoses are requested. HCPs are also asked whether they have any concerns or issues regarding the family. In the following sections, HCPs are asked to respond to openly formulated questions about each session. Five sections are to be filled out for session one, two, and three about which topics and concerns were discussed, support options for the children and families, any questions regarding the child posed by the parents, any additional details, and agreements for the next session. For session three, there are additional sections for follow-up agreements and advice given to the child and parents, as well as for referrals and necessary follow-up actions that HCPs are to take. In our analyses, we used all the sections from sessions in which children participated. The amount of information and degree of details in the CT logbooks varied. In most logbooks HCPs had written a response in all sections. Some logbooks were several pages long, while others only contained a few paragraphs.

Data Analyses

In the Family Assessment Forms each patient stated how many minor children they had. This information enabled us to calculate the total number of children for the assessed patients, and the number of children for the patients participating in CT. We detected how many children participated in CT from the Logbooks. Based on this information we calculated (1) children's participation rate and (2) number of non-participating children whose parent participated. For non-participating children, we used information from the Logbooks to detect and quantify reasons for their absence.

Descriptive information of participating and non-participating children and parents was compared by analyzing information reported from CT sessions and Family Assessment Forms. For our analyses, we used information from the Family Assessment Form on parent gender, parent diagnosis, child age and gender, and where the children lived. We also used two questions about whether the children had received information about the parent's treatment/hospitalization and condition: "*Does your child know that you receive treatment/are hospitalized?*" and "*Has your child received information about your condition?*" The response categories for these two items are "*no*", "*partially*," and "*yes*." Descriptive statistics, *t*-tests, and chi-square testing were computed in IBM SPSS Statistics 25.

Statistical Analysis

We were not able to test differences between participating and non-participating children regarding parents' diagnoses in reliable ways because of the small sample size, resulting in few parents in each diagnosis category. Partial receival of information was treated as having received information in the analysis. Chi-square tests was conducted to analyze differences between participating and non-participating children in terms of parent gender, child gender, information received and where the children normally lived. For all chi-square tests, we reported phi (φ) for effect size measurement. To test for differences in the mean age for participating and non-participating children, we initially performed a Levene's test to determine if the variance of the groups was unequal or equal. The results from the Levene's test showed that the variance of the groups was unequal. Therefore, we performed a two-tailed t-test with unequal variance for the groups to test for age differences. We calculated the effect size of the mean differences using Cohen's *d*. The magnitude for all effect sizes was interpreted in accordance with Cohen (34).

Thematic Analysis

Child Talks Logbooks were analyzed to identify the thematic content of the sessions in which children participated, and all CT sessions with participating children were imported into the qualitative analysis program NVivo 12 Pro. The logbooks are a secondary source of information of the CT sessions, written and processed by HCPs.

For the thematic analysis of the CT sessions with participating children, the authors and researchers of the present study used the six-step phase guide by Braun and Clarke (35). This is a flexible approach in which the aim is to identify, analyze, and report the patterns found in the material (35). Our aim was to explore characteristics and patterns in the sessions with participating children; therefore, an inductive approach to the material was chosen.

The first step in the guide by Braun and Clarke (35) is getting to know the dataset by reading it multiple times. We transcribed the forms from paper to electronic format in order to familiarize ourselves with the material. Secondly, we started the initial coding of the material by identifying aspects of the data that reoccurred and which were an important focus in the sessions. The principle of data saturation was used, and hence we ended the initial coding when further coding no longer added new information. In the third step, we formed themes and sub-themes from the codes. In the fourth step, we reviewed all the themes and adjusted them as necessary so that the themes were more meaningful and comprehensive in respect of the codes included. In the fifth step, we defined the themes by writing a few sentences on their content that were suitable for all the codes included. The final, sixth step consisted of describing the themes in the present paper.

RESULTS

Parents' Participation Rate in Child Talks

Around 5,500 patients were receiving treatment at the DMHSD each year during the project period (2). A total of 424 patients were assessed as having minor children by using the Family Assessment Form, and 78 of these patients (18%) received the CT intervention.

Children's Participation Rate and Reasons for Exclusion

In 39 (50%) of the performed CT interventions all or some of patients' children participated. The 78 patients who received the CT intervention had 157 children in total. Of these children, 62 (39%) took part in the intervention, leaving 95 children (61%) not participating despite their parent receiving the intervention.

Based on the Family Assessment Forms, a total of 864 children were identified. Of these children, 62 participated in CT, resulting

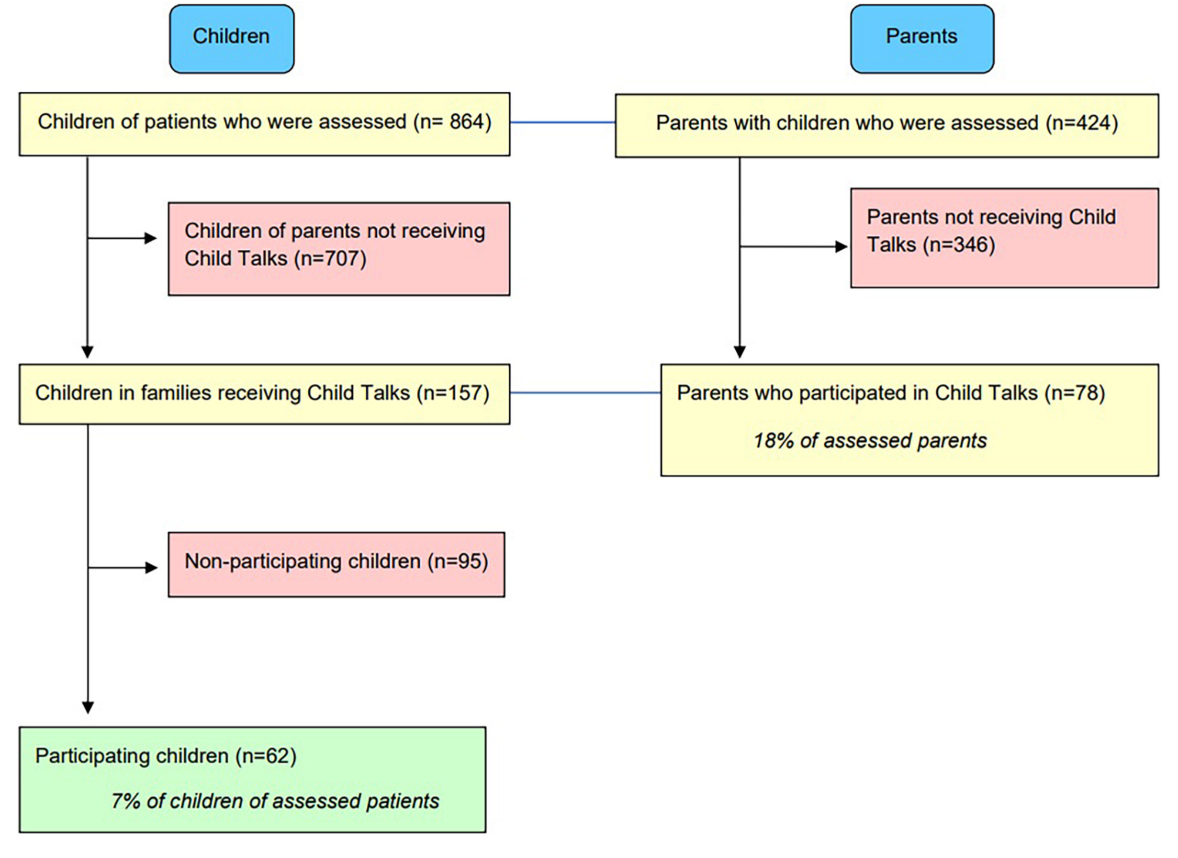


FIGURE 1 | Childrens' participation in Child Talks.

in a total participation rate of 7% for the identified children. See **Figure 1** for a flowchart of childrens' participation in CT.

Healthcare professionals provided information about the reasons for children's absence in some of the records (n = 23). The reasons stated in records were: (1) the patient was soon to be discharged from hospital and therefore the task of talking to the children was postponed to a later occasion or transferred to personnel in other services(n = 7); (2) the patient had little contact with the child/children (n = 7); (3) the patient rejected the offer of CT with participating children (n = 5); and (4) the other parent of the child did not consent to the child participating, or personnel had not been given a response from the family (n = 4).

Comparison of Participating and Non-participating Children

Diagnosis and Gender of Children's Parents

The diagnosis and gender of participating and non-participating children's parents are given in **Table 1**. Twelve parents had multiple diagnoses.

The difference between participating and non-participating children in terms of parent's gender was not significant at $p < 0.05$. The result from the chi-square test was X^2 (1, N = 113) = 3.805, p = 0.051 and had a small to medium effect size (φ = 0.18). Information about parents' gender was missing for 44 of parents' children.

Childrens' Age and Gender

Children participating in the intervention were between 3 and 22 years of age. We observed that the proportion of participating children increased with age (see **Figure 2**). Two children of preschool age (<6 years) participated (see **Table 2**). Of the participating children, 80% were more than nine years old. **Figure 2** illustrates children's age distribution for participating and non-participating children.

TABLE 1 | Characteristics of parents with participating and non-participating children.

Characteristics of parents	With participating children (n = 39)	Without participating children (n = 39)
Diagnosis		
Alcohol and substance dependence	2	8
Paranoid schizophrenia and psychosis	7	1
Manic episodes	0	1
Bipolar disorder	5	4
Major depression disorder	15	17
Anxiety disorder	7	6
Post-Traumatic Stress Disorder	11	5
Eating disorder	1	1
Personality disorder	1	6
	49	49
Patient's kinship to children		
Mother	33	26
Father	6	12
Sibling		1
	39	39

Information missing on diagnoses of two parents with participating children. Since some parents had several diagnoses, the sum of diagnoses exceeds the number of patients participating in CT.

To test for difference in the mean of age of participating children [M (51) = 11.69, SD = 3.78] and non-participating children [M (62) = 10.13, SD = 5.27], we performed a t-test. The results from Levene's test for difference of variance between the groups were significant at $p < 0.05$: F (1,111) = 5.764, p = 0.018. We therefore preformed a two-tailed t-test for which unequal variance for the groups was assumed. Cohen's effect size value (d = 0.34) implied a small to medium magnitude of difference between the two groups, but the difference was not statistically significant at $p < 0.05$: t (111) = 1.83, p = 0.07.

Of the participating children, 33 were girls and 23 were boys. Of the non-participating children, 36 were girls and 31 were boys (see **Table 2**). Information about gender was missing for 6 participating and 28 non-participating children. A higher proportion of girls than boys participated. However, this difference was not statistically significant at $p < 0.05$, X^2 (1, N = 123) = 0.335, p = 0.563 and had a very small effect size (φ = 0.05).

Information Provided to Children

Participating children knew that their parent was receiving treatment or was being hospitalized more often than children who did not participate in the sessions, measured at assessment point. Of the participating children, 97.7% (42 out of 43) were aware of their parent's treatment or hospitalization, whereas 72.5% (29 out of 40) of the non-participating children were aware of this. However, answers to this question were missing for 19 participating children and 55 non-participating children. We compared participating and non-participating children by performing a chi-square test and found a significant difference of $p < 0.05$ between the groups, X^2 (1, N = 83) = 10.62, p = 0.001, with a medium magnitude of difference (φ = 0.36).

Participating children had also received information about their parent's condition more often than non-participating children, at assessment point. Of the participating children, 85.7% (36 out of 42) were aware of their parent's condition, compared to 60.5% (23 out of 38) of the non-participating children. Answers to this question were missing for 20 participating and 57 non-participating children. The results from the chi-square test comparing the two groups showed a significant difference at $p < 0.05$ level, X^2 (1, 80) = 6.54, p = 0.011, with a small-to-medium effect size (φ = 0.28).

Where the Children Lived

For participating children, 94% (47 out of 50) lived with the hospitalized parent. In the case of the non-participating children, 65.9% (29 out of 44) were registered as living with the patient. Answers to the question of where the children lived were missing for 12 out of 62 participating children and 51 out of 95 non-participating children. A chi-square test conducted to assess the difference between participating and non-participating children in terms of living with the hospitalized parent showed a

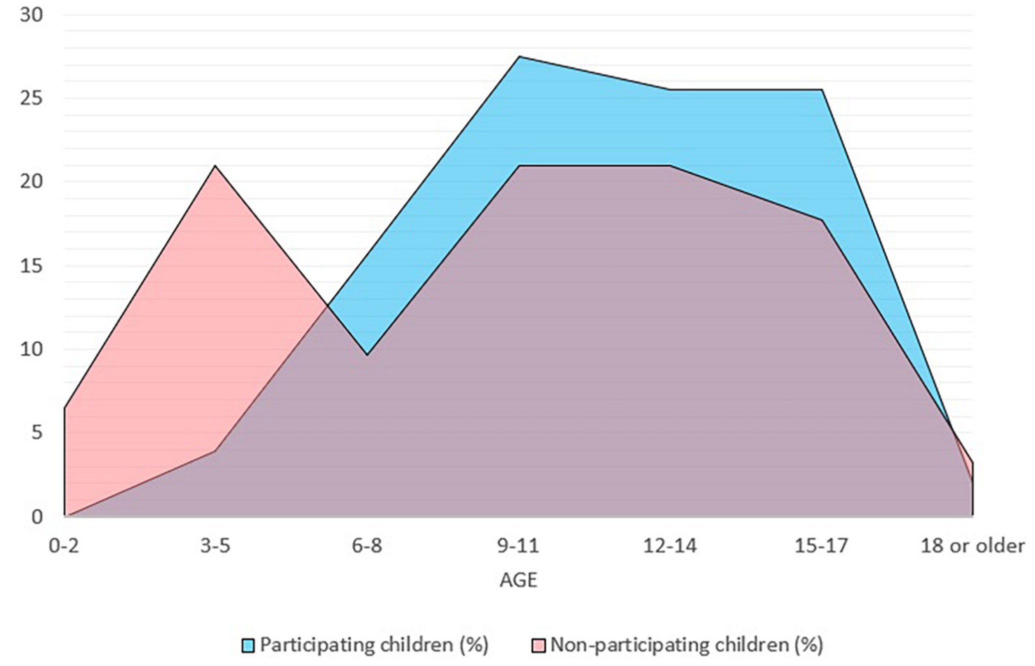


FIGURE 2 | Age distribution for participating and non-participating children. *n* = 113, age missing for 11 participating children and 33 non-participating children.

TABLE 2 | Characteristics of participating and non-participating children.

Characteristics of children	Participating children (*n* = 62)	Non-participating children (*n* = 95)
Gender		
Girls	33	36
Boys	23	31
	56	67
Age		
0–2	0	4
3–5	2	13
6–8	8	6
9–11	14	13
12–14	13	13
16–18	13	11
18 or older	1	2
	51	62

Information missing about gender for participating 6 participating and 28 non-participating children, and information missing about age for 11 participating children and 33 non-participating children.

significant difference at $p < 0.05$, X^2 (1, $N = 94$) = 11.93, $p = 0.001$, with a medium effect size ($\varphi = 0.36$).

Themes in Sessions With Participating Children

The thematic analysis of the written reports from CT sessions involving children resulted in three main themes and ten sub-themes. The main themes were communication about PMI within the family, children's struggles, as well as HCPs' evaluation of the child's situation and identification of further support. See **Figure 3** for an overview of main themes and sub-themes.

Informing Children About Parental Mental Illness

Healthcare professionals frequently explained to parents why talking openly about mental health within the family is important. Some of the parents were open about their illness, and many said they wanted to be even more open. In many families, however, there was little to no communication about the parent's illness. Hence, for some children the CT session was the first time they received information about their parent's illness. The children usually expressed that they were glad to receive information from HCPs. However, some children did not want information and did not want to visit or have contact with the mental health services.

In some cases, the parents were unwilling to talk about mental illness and prevented their children from receiving information or being given the opportunity to talk about their situation. Reasons were that they did not want their children to be unnecessarily worried or to focus on the negative characteristics and psychiatric diagnosis of the sick parent. A mother who was diagnosed with depression, anxiety, and an eating disorder protected her 10-year-old daughter (ID 35), from words such as "mental illness" and "psychiatric hospital."

The information that children received was not always correct or clarifying. Proper explanations about parents' illness, symptoms and behavior were often missing. Some children knew that something was wrong or that their parents were struggling, but not what or how. Often parents and HCPs used word such as "exhausted" or "having headache" when explaining parents' mental illness.

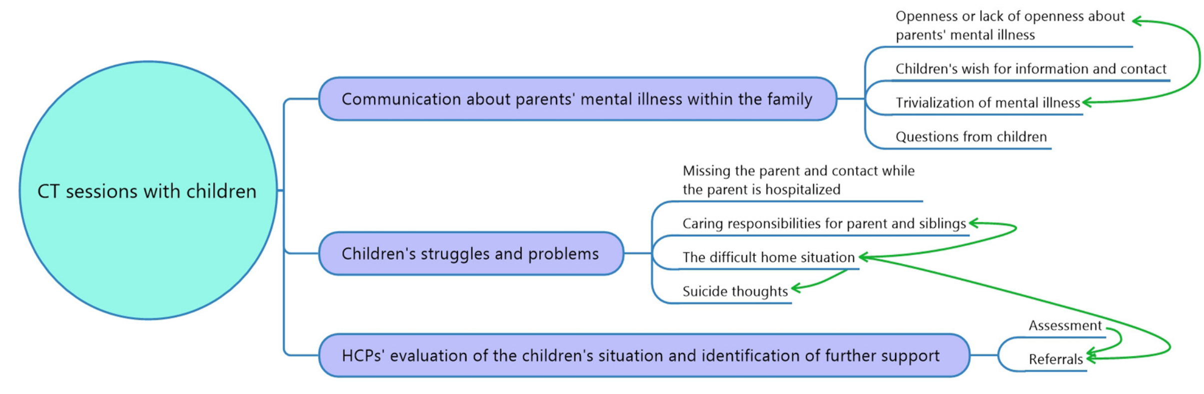


FIGURE 3 | Map over main themes and sub themes.

Questions from children appeared in a few records. The questions reported concerned the care of younger siblings, heredity, and the home situation. Children did not usually ask directly, but rather expressed their curiosity about a theme in the session.

Children's Struggles and Problems

Children reported missing their hospitalized parents and being worried about them. Two boys aged 10 and 11 years (ID 78) missed their father while he was hospitalized and reported being worried about him. This was also given as the reason for the youngest boy having trouble concentrating at school. Some children kept in contact with the hospitalized parent by using video calls and some had also visited their hospitalized parent on several occasions. Some children missed the way things used to be before their parent became ill, like a 15-year-old girl (ID 17) who said that she was missing her "healthy" mother. She missed spending time with her mother in the evenings, lighting candles, and watching movies. She also missed tasty and healthy meals.

Several children in the records were worried about, and had great responsibilities for, the care of their sick parent and/or younger siblings. A 12-year-old girl (ID 42) had to physically stop her mother from dying by suicide. The girl was worried about what might happen once her mother was discharged from the hospital. HCPs emphasized the importance of making the daughter aware that it was not her responsibility to take care of and look after her mother, yet they advised her to contact the police if her mother did anything like that again. The girl also expressed her concerns for the care of her younger siblings while her mother was hospitalized, especially the one-year-old. She was not sure her father would be able to take care them. She had trouble sleeping at night. The girl was advised to contact help services if things became difficult or if she needed someone to talk to.

Some children were overinvolved in the illness of their parent, such as a 12-year-old boy (ID 18) whose mother had been diagnosed with anxiety:

> *The boy said he was going to look after his mother until he became an adult. He was worried about his mother when she got her anxiety attacks. When she got the anxiety attacks, he massaged her.*

Another child, a 16-year-old boy (ID 60), frequently had to participate in his mother's doctor appointments and translate letters from the Ministry of Foreign Affairs. He was also involved in the conflict between his parents and described how he had to stop his father from being violent with his mother:

> *In the beginning, after they came to Norway, his father was physically violent toward his mother. One night he got up and told his father that if he ever beat her again, he would call the police. The father went out and did not return until much later. Since the incident the father has never beaten her. The boy cried while telling this. He said this was the very first time he had cried in front of his sister.*

Many children had a difficult home situation, living in families with severe, long-lasting problems. Many of the children had experienced frightening episodes at home. The children's situations at home were often described as unpredictable, stressful, and characterized by high conflicts levels and violence between family members. In one case a 16-year-old girl (ID 39) who was living with her mother, who had been diagnosed with a psychotic illness, was physically abused:

> *There was general concern for the family because of the mother's mental state. There was also concern for the daughter's situation and whether she was given help for her own mental health problems, which she had had for several years. Her mother was unstable and had on several occasions pushed and thrown things after the girl. The mother had also called her names. It was difficult for the daughter because her mother was suspicious and seemed to be in a paranoid state of mind.*

The patient in the example above was discharged from the hospital and sent home to her daughter. A report of concern was sent to The Child Welfare and Protection Services (CWPS). The daughter was advised to contact the school nurse when she returned to school at the end of the summer.

Several children had more or less concrete thoughts about dying by suicide. For one of these children, HCPs stated that the child's mental health problem was taken care of by their general practitioner. In another case a 16-year-old boy (ID 60) was invited to call HCPs if he wanted to talk, after he had told them that he had thought about cutting his main artery if he was sent back to Afghanistan. He had even looked in the kitchen drawer for a knife. For a child who had attempted to die by suicide earlier, HCPs were concerned for the child's mental health problems and whether appropriate help was provided.

Evaluating and Supporting the Children

In many of the records, HCPs observed and evaluated the situation of the children and the parent's caring abilities. In some cases, HCPs explicitly wrote down the agenda for evaluating the children, for example to "look at the interaction between the mother and the daughter" or to "observe the relation and the interaction between the children and parents." In one case (ID 26), the HCPs evaluated the attachment and how the child acted around his parents. HCPs even talked with the four-year-old boy while his parents were waiting outside.

Healthcare professionals explored the children's network and support options as part of the intervention. Many sessions led to referrals to CWPS and The Child and Adolescent Mental Health Services (CAMHS), but in some cases where children described severe problems, no referrals were made. HCPs frequently encouraged parents and children to contact their general practitioner, their teacher, or the school nurse. Responsibility for establishing contact with help services or professionals was often left with the child. Some families were already in contact with CWPS and/or CAMHS. School nurses were frequently recommended as a support option. In the case of the 10-year-old girl (ID 35) who was being shielded from words such as "mental illness" and "psychiatric hospital," the school nurse was recommended as a support option because the parent and the child knew of her. Likewise, in a case with a 17-year-old boy (ID 09), the school nurse was emphasized as a support option as a neutral person the boy could talk to about everyday life and other relevant topics of conversation for adolescents. In addition, HCPs often gave children and parents the opportunity to have several sessions and to get in touch with them outside of the sessions if they needed to talk. The HCPs also gave children the opportunity to call them if they had any questions. In some cases, there was a mutual agreement that the family would benefit from staying in touch with the ward.

DISCUSSION

Is the Participation Rate of Patients at an Acceptable Level?

Of the thousands of patients at the DMHSD at UNN during the period 2010–2015 (2), only 424 were assessed with the Family Assessment Form. This means that for most of the patients, there were no records of minor children they might be parenting. Furthermore, only 78 patients received the CT intervention, meaning that only a small fraction of COPMI were attended to in the manner mandated by legislation in Norway. These results are in line with previous research, suggesting that it is challenging to implement new routines related to COPMI in Norway (36, 37) and illustrating the need for a better implementation strategy.

Is the Participation Rate of Children at an Acceptable Level?

Seven percent of children identified in the Family Assessment Forms participated in CT. However, the number of children identified in the Family Assessment Form does not represent all minor children of patients at the DMHSD. In fact, around 5,500 patients were receiving treatment at the DMHSD each year during the project period (2), and likely one third of them had a mean of 1.75 minor children each (3, 27, 38), which equals more than three thousand minor children of patients each year. The participation rate based on the actual number of minor children of patients will therefore be considerably lower than 7%.

According to the CT manual, children should participate in the second session and optionally in the third session (24). However, children were only present in half of the cases in which the CT intervention was utilized, and since not all siblings participated in cases including children, children's participation rate in received CT was only 39%. Children's participation rate reveals that the intervention manual is not being adhered to, and consequently, the obligation of HCPs to provide COPMI necessary information about PMI, support and follow up does not seem fulfilled. It is especially important that HCPs provide information to COPMI since previous research has shown that parents themselves often did not inform their child about their treatment/hospitalization or condition (27).

The main reasons for children not participating reported in this study were reluctance of one or both parents, little contact with the children or ending of the parent's treatment. In cases where the parent does not have custody or contact with the children, inviting the children to participate in a session is not appropriate, thus these patients would not have been invited to participate in the CT intervention. Large demographical distances between the clinic and the childrens home might have made childrens participation difficult in some cases, especially for the youngest children. In cases in which parents were reluctant to bring their children to a CT session, HCPs are in a good position to argue in favor of child participation. The reasoning behind including children is available in the CT manual and motivating parents and planning how to inform the children is the core activity in session one (24). Aligned with previous research, HCPs seem to need better awareness of the importance of giving children information and support, greater skill at motivating parents to invite their children, and greater skill, or perhaps greater confidence, in performing conversations with children present (28, 31).

Which Factors Influence Child Participation?

In terms of factors relating to the parent, differences between participating and non-participating children regarding parents' diagnoses could not be tested in reliable ways because of the sample size. However, the parent's gender might be a factor influencing child participation, with a difference between participating and non-participating children close to our chosen significant level, with a small to medium effect size. COPMI more often lived with their mothers as a sole caregiver and therefore were in the care of relatives while their mother was hospitalized (27). A more dramatic change in these children's life situations calls for more information and support, which might explain why children of mothers participated more often. This is also in correspondence with our results showing that participating children more often lived with the parent in treatment. Furthermore, deciding and planning for child participation is easier when the child is fully under the custody of the patient.

Mostly older children participated in CT. Child participation increased with child age, and the difference between participating and non-participating children in mean age was close to our chosen significance level, with a small to medium effect size. In earlier studies HCPs have reported feeling insecure about who has the responsibility of children visiting patients and how to have age-appropriate conversations (33). Furthermore, the study found that HCPs' confidence level influenced their initiative to motivate patients to invite their children. For the youngest children, it is possible that HCPs' insecurities were amplified, since younger children are less independent and require more adjustments by HCP. HCPs might need more knowledge and training in child development and age-appropriate conversations about PMI. Interventions for COPMI can also be more adaptable and user-friendly for HCPs, by making recommendations for different age groups. For example, for children under two years a visit to the hospital to assure them their parent is safe can be recommended. For children from three years and up, in addition to recommending a visit, guidelines for age-appropriate information and communication principles can be provided in the intervention manual. Increasing HCPs' information and support to the youngest children is of great importance since younger children are the most dependent on their parents, and not mature and autonomous to seek information and support elsewhere. We found no differences in child gender for participation.

In terms of knowledge about PMI, there was a significant difference, of a small to medium magnitude, between participating and non-participating children. Participating children more often already knew about parents' treatment/hospitalization and condition. Families that are more open about PMI might be more willing to have children participating in a conversation with HCPs. This is in coherence with earlier studies in which families' fear of involving children was perceived as an important hindering factor for a family-focused practice by HCPs (32).

Are Children Supported and Informed?

The main themes in the CT sessions with children reflected the objectives of the CT intervention (24): communication about PMI within the family, children's struggles and HCPs' evaluation of the child's situation and identification of further support. However, the content of the CT sessions uncovered a large variation in the quality of the support and information children were provided.

Children were glad to receive information, which is in line with earlier studies which show that children appreciate support and information from HCPs (22, 23). Parents, however, were sometimes reluctant and unsure about informing their children because they did not want to make their child additionally worried, a barrier for family-focused practice found in another study (32). Our result confirmed, what is described by other researchers (14), that children often know that something was wrong and that not having information could lead to frustration. It was also found that the children were missing their parent and were worried about them. These results underpin the importance of information and contact with the parent in treatment for COPMI (14, 19–22).

Healthcare professionals evaluated and explored children's situations but were reluctant to refer to other services or provide further support. High conflict levels within the family, domestic violence, physical and mental abuse, mental health problems and suicide thoughts among children were described. However, few appropriate actions were taken by HCPs. Despite HCPs' obligation by law to refer children to the CWPS when concerned with their living situation, HCPs did not take appropriate actions in all cases. In previous studies HCPs have reported hesitation against referrals because of insecurities of whether there were grounds for referral, whether a referral would benefit the child and whether a referral would harm the family and their relationship with the patient (39). The lack of action by HCPs does not only take away children's chance for help but does also trivialize the problems and struggles the children are experiencing. HCPs need to know which support they can offer, and which actions to take. Educating HCPs about follow-up options and help services for children might contribute to providing COPMI better support.

Strengths and Limitations

One limitation of this study is missing data for several variables, particularly for the non-participating group of children. The results must therefore be interpreted carefully. The small sample size may be a factor contributing to the lower sensitivity of the t-test, resulting in less reliable results.

The journal data were a secondary source of information of the CT sessions, written and processed by HCPs, based on their perception of what is important and of interest. What was written in the logbooks was partially decided and influenced by the CT logbook and the questions HCPs were to answer. However, the questions were openly formulated and did invite HCPs to give detailed descriptions of the conversations and share a range of information. Despite this, it varied how much and how specific the written information about the conversations were. By focusing on the themes that were discussed in sessions, rather than looking for meanings behind the text, the data material was suitable to answer the associated research aim in the present study. The benefit of the research design is that it enabled a reduction of the disturbance and influence of an observing researcher. A researcher present in sessions might have made participants more hesitant to speak openly about sensitive and personal subjects. In addition, HCPs should be able to perform the sessions at a time they found appropriate in respect of the patient's course of treatment and time management. Having to plan for a third person's participation would have made the feasibility of the project weaker.

Only CT Logbooks in the electronic patient journal were assessed; hence information written elsewhere was not available and not included in the analysis. What is logged from the sessions is partially prearranged from the Logbook forms. Since the Logbook forms are directly based on the manual's description of the intervention, the data may incorrectly confirm the HCPs' adherence to the manual. The HCPs were aware that the reports were going to be used in a quality-assurance project and they might therefore have reported the session more in line with the guidelines of the manual. There was, however, sections in the Logbook form with open formulated questions, which gave HCPs the opportunity to share a wide range of information.

Future Research

To facilitate and strengthen the degree to which children are given information and support they are entitled, more research is needed to gain detailed knowledge about factors influencing children's participation. Future research should identify reasons why HCPs are not including children and investigate whether it is due to lack of consent from parents, institutional constraints, or unfulfilled professional needs. It would be useful to know whether certain characteristics of the parent's illness, such as a sudden onset or a significant change in the parent's functional level and behavior, influence the need to give and receive information among HCPs, parents, and children. Whether child participation is influenced by parent gender also needs to be explored in future studies after adjusting for where the child usually lives. In addition, the difference between participating and non-participating children in terms of received information about PMI should be investigated when adjusting for confounding factors, such as the child age.

CONCLUSION

Child Talks is an intervention that seeks to reinforce COPMI's ability to cope with their family situation by the provision of age-appropriate information about their parent's illness and treatment. The intervention also aims to provide additional support and follow-up for the children who require it. Of patients who were registered as having minor children, less than one fifth received the intervention, and only half of the patients who participated also had their children participating. Of the registered minor children, less than one in ten received CT. Ideally, children who participate in the intervention emerge better informed, supported and are, when necessary, provided with follow-up. However, this study shows that even participating children were not always followed-up or judged to have been adequately informed. Routines and training of HCPs to support parents with mental illness and their children need improvement. Initial identification of children of patients is important, and subsequent support and provision of adequate services to the identified children always needs to follow.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Data Protection Officer at the University Hospital of Northern Norway (UNN). The regional ethics committee (REK) categorized the project as a quality assurance project. Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

KK made the analysis and the draft of the manuscript. CR was the primary investigator in the main project. CR and CL collected the data in collaboration with the project coordinator, Lisbeth Mørch, at UNN. All authors contributed to the writing of the manuscript.

FUNDING

This study has been funded by The Regional Center for Child and Youth Mental Health and Child Welfare (RKBU North) at UiT The Arctic University of Norway.

ACKNOWLEDGMENTS

This article is part of a larger study at RKBU North in cooperation with the University Hospital of Northern Norway (UNN) and the non-governmental organization Adult for Children (Voksne for Barn). We would like to thank UNN for carrying out the study and making the data available to us. We also thank Bjørn Helge Handegård, statistician at RKBU North, for valuable advice on the statistical analysis.

REFERENCES

1. Torvik FA, Rognmo K. *Barn av Foreldre med Psykiske Lidelser eller Alkoholmisbruk: Omfang og Konsekvenser*. Oslo: Nasjonalt folkehelseinstitutt (2011).
2. Reedtz C, Mørch L, Lauritzen C. Registreres psykiatriske pasienters barn i elektronisk pasientjournal? *Nordisk Sygeplejeforskning*. (2015) 5:36–45.
3. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry*. (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427

4. Reupert AE, Maybery D, Kowalenko N. Children whose parents have a mental illness: prevalence, need and treatment. *Med J Aust.* (2013) 199:S7–9. doi: 10.5694/mja11.11200
5. Weissman M, Wickramaratne P, Gameroff M, Warner V, Pilowsky D, Kohad R, et al. Offspring of depressed parents: 30 years later. *Am J Psychiatry.* (2016) 173:1024–32. doi: 10.1176/appi.ajp.2016.15101327
6. Sundfær A. *God Dag, Jeg er et Barn – Om Barn som Lever Med rus Eller Psykisk Sykdom i Familien [Good Day, I am a Child – About Children Living With Substance use or Mental Illness Within Their Family]*. Oslo: Fagbokforlaget (2012).
7. Brennan PA, Hammen C, Andersen MJ, Bor W, Najman JM, Williams GM. Chronicity, severity, and timing of maternal depressive symptoms: relationships with child outcomes at age 5. *Dev Psychol.* (2000) 36:759–66. doi: 10.1037//0012-1649.36.6.759
8. Farahati F, Marcotte DE, Wilcox-Gök V. The effects of parents' psychiatric disorders on children's high school dropout. *Econ Educ Rev.* (2003) 22:167–78.
9. van Santvoort F, Hosman CMH, Janssens JMAM, van Doesum KTM, Reupert A, van Loon LMA. The impact of various parental mental disorders on children's diagnoses: a systematic review. *Clin Child Fam Psychol Rev.* (2015) 18:281–99. doi: 10.1007/s10567-015-0191-9
10. Rasic D, Hajek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder, and major depressive disorder: a meta-analysis of family high-risk studies. *Schizophr Bull.* (2014) 40:28–38. doi: 10.1093/schbul/sbt114
11. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Adv Ment Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
12. Foster K, O'Brien L, McAllister M. Addressing the needs of children of parents with a mental illness: current approaches. *Contemp Nurse.* (2004) 18:67–80. doi: 10.5172/conu.18.1-2.67
13. Foster K, O'Brien L, Korhonen T. Developing resilient children and families when parents have mental illness: a family-focused approach. *Int J Ment Health Nurs.* (2011) 21:3–11. doi: 10.1111/j.1447-0349.2011.00754.x
14. Riebschleger J, Grove C, Cavanaugh D, Costello S. Mental health literacy content for children of parents with a mental illness: thematic analysis of a literature review. *Brain Sci.* (2017) 7:141. doi: 10.3390/brainsci7110141
15. Siegenthaler E, Munder T, Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17.e8. doi: 10.1016/j.jaac.2011.10.018
16. Solantaus T, Paavonen EJ, Toikka S, Punamäki R-L. Preventive interventions in families with parental depression: children's psychosocial symptoms and prosocial behaviour. *Eur Child Adolesc Psychiatry.* (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
17. Thanhäuser M, Lemmer G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. *Curr Opin Psychiatry.* (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342
18. Lannes A, Bui E, Arnaud C, Raynaud JP, Revet A. Preventive interventions in offspring of parents with mental illness: a systematic review and meta-analysis of randomized controlled trials. *Psychol Med.* (2021) 51:2321–36. doi: 10.1017/S0033291721003366
19. Reupert AE, Maybery D. "Knowledge is power": educating children about their parent's mental illness. *Soc Work Health Care.* (2010) 49:630–46. doi: 10.1080/00981380903364791
20. Grové C, Melrose H, Reupert A, Maybery D, Morgan B. When your parent has a mental illness: children's experiences of a psycho-educational intervention. *Adv Ment Health.* (2015) 13:127–38.
21. Pihkala H, Sandlund M, Cederstrom A. Children in Beardslee's family intervention: relieved by understanding of parental mental illness. *Int J Soc Psychiatry.* (2012) 58:623–8. doi: 10.1177/0020764011419055
22. Grové C, Reupert A, Maybery D. The perspectives of young people of parents with a mental illness regarding preferred interventions and supports. *J Child Fam Stud.* (2016) 25:3056–65.
23. Drost LM, van der Krieke L, Sytema S, Schippers GM. Self-expressed strengths and resources of children of parents with a mental illness: a systematic review. *Int J Ment Health Nurs.* (2016) 25:102–15. doi: 10.1111/inm.12176
24. Reedtz C, Lauritzen C. *Håndbok – Prosjekt Barn som Pårørende. Regionalt Kunnskapssenter for Barn og Unge*. Oslo: RKBU Nord (2011).
25. Doesum KTV, Koster C. *KOPP Praten met Ouders Kinderen: Handleiding Preventieve Huisbezoeken voor Ouders met Psychiatrishe Problemen en Hun Kinderen Manual Child Talks*. (2008). Deventer, The Netherlands: Dimence, Community Mental Health Center.
26. The Health Personnel Act, §10 a (2010). *Act Relating to Health Personnel (LOV 1999-07-02-64)*. Lovdata. Available online at: https://lovdata.no/dokument/NL/lov/1999-07-02-64/KAPITTEL_2#%C2%A710a
27. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness: a necessity to provide relevant support. *Front Psychiatry.* (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
28. Skogøy BE, Ogden T, Weimand B, Ruud T, Sørgaard K, Maybery D. Predictors of family focused practice: organisation, profession, or the role as child responsible personnel? *BMC Health Serv Res.* (2019) 19:793. doi: 10.1186/s12913-019-4553-8
29. Lauritzen C, Reedtz C, Van Doesum K, Martinussen M. Factors that may facilitate or hinder a family-focus in the treatment of parents with a mental illness. *J Child Fam Stud.* (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
30. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
31. Maybery D, Goodyear M, Reupert AE, Grant A. Worker, workplace or families: what influences family focused practices in adult mental health? *J Psychiatr Ment Health Nurs.* (2016) 23:163–71. doi: 10.1111/jpm.12294
32. Korhonen T, Vehviläinen-Julkunen K, Pietilä A-M. Implementing child-focused family nursing into routine adult psychiatric practice: hindering factors evaluated by nurses. *J Clin Nurs.* (2008) 17:499–508. doi: 10.1111/j.1365-2702.2007.02008.x
33. O'Brien L, Brady P, Anand M, Gillies D. Children of parents with a mental illness visiting psychiatric facilities: perceptions of staff. *Int J Ment Health Nurs.* (2011) 20:358–63. doi: 10.1111/j.1447-0349.2011.00740.x
34. Cohen J. *Statistical Power Analysis for the Behavioral Sciences*. Mahwah, NJ: Lawrence Erlbaum Associates (1988).
35. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* (2006) 3:77–101.
36. Reedtz C, Jensaas E, Storjord T, Kristensen KB, Lauritzen C. Identification of children of mentally Ill patients and provision of support according to the Norwegian health legislation: a 11-year review. *Front Psychiatry.* (2022) 12:815526. doi: 10.3389/fpsyt.2021.815526
37. Lauritzen C, Reedtz C, Van Doesum KTM, Martinussen M. Implementing new routines in adult mental health care to identify and support children of mentally ill parents. *BMC Health Serv Res.* (2014) 14:58. doi: 10.1186/1472-6963-14-58
38. Statistics Norway. *Slik er Norske Barnefamilier [How Norwegian Families with Children Are]*. (2018). Available online at: https://www.ssb.no/befolkning/artikler-og-publikasjoner/slik-er-norske-barnefamilier (accessed May 15, 2021).
39. Lauritzen C, Reedtz C. Adult mental health services and the collaboration with child protection services. *J Hosp Admin.* (2016) 5. doi: 10.5430/jha.v5n5p72

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Public Health

PERSPECTIVE
published: 11 March 2022
doi: 10.3389/fpubh.2022.756066

It Takes a Village to Raise a Child: Understanding and Expanding the Concept of the "Village"

***Andrea Reupert*[1]*, *Shulamith Lala Straussner*[2], *Bente Weimand*[3,4] and *Darryl Maybery*[5]**

[1] Faculty of Education, Monash University, Clayton, VIC, Australia, [2] Silver School of Social Work, New York University, New York City, NY, United States, [3] Faculty of Health and Social Sciences, University of South-Eastern Norway, Drammen, Norway, [4] Division of Mental Health Services, Akershus University Hospital, Lørenskog, Norway, [5] School of Rural Health, Monash University, Warragul, VIC, Australia

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Miriam Heyman,
Brandeis University, United States
Emma Maynard,
University of Portsmouth,
United Kingdom
Lucy Thompson,
University of Glasgow,
United Kingdom
Kate Blake-Holmes,
University of East Anglia,
United Kingdom
Rachel Calam,
The University of Manchester,
United Kingdom

Correspondence:
Andrea Reupert
andrea.reupert@monash.edu

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Public Health

Received: *10 August 2021*
Accepted: *11 February 2022*
Published: *11 March 2022*

Citation:
Reupert A, Straussner SL, Weimand B and Maybery D (2022) It Takes a Village to Raise a Child: Understanding and Expanding the Concept of the "Village". Front. Public Health 10:756066. doi: 10.3389/fpubh.2022.756066

This perspective article defines and discusses the concept of the "village" when working with families who are experiencing multiple adversities. The article starts with a discussion on what is meant generally by a village approach, followed by a historical overview of how families living in adversity have been defined and positioned. The need to move past a siloed, professional centric approach when working with families is then presented. Using a model of social connections, based on Bronfenbrenner's ecological theory, we then identify who the "villagers" might be. Some potential principles for how the village might work with families living with adversity are presented, along with two case studies, to demonstrate how these principles might be enacted. This perspective article provides an overview and discussion of "the village" concept, rather than present a definitive set of guidelines or recommendations.

Keywords: perspective, community, children, parent, adversity, parents, caregivers

INTRODUCTION

Globally, many families face multiple adversities. These advertises may include mental illness, substance use and addiction problems, physical illness, domestic and community violence, poverty, insecure housing and war. Moreover, many of these problems are accumulative, with one problem, for example, parental mental illness, cascading into other problems, such as relationship breakdowns, unemployment and poverty (1). Preventing and mitigating the impact of these problems on parents and children is critical for improving population health for families now and in the future. However, no one sector or organization is in a position to address all the issues that these families may face. Hence, it is proposed that a "village approach" is needed when bringing up children.

The genesis for this perspective article comes from the *It takes a village,* an international conference held in Oslo, 2018. The conference brought together those with lived experience, researchers, practitioners and policy makers to discuss the needs of such families but arguably more importantly, optimal service responses. Given its audience, efforts were made, when putting together symposiums and accepting articles, to highlight ways the village might work together. Others also employ the term "village", for example, the Austrian *How to raise the village to raise the child,* an initiative funded by the Ludwig Boltzmann Society and the Medical University of Innsbruck. The initiative aims to strengthen formal and informal support for children living with parental mental illness. Drawing on these initiatives, this article documents what is meant by the concept of a village approach. This article constitutes an attempt to "move toward" clarifying and discussing the concept of "the village" rather than provide a definitive set of guidelines or recommendations.

In this perspective, we first define what we mean by the "village" and then provide some discussion about what we mean by the term "families". The need to move past traditional practice silos and how the village might work with families is then discussed using two, brief case studies.

DEFINING THE "VILLAGE"

The phrase "it takes a village to raise a child" originates from an African proverb and conveys the message that it takes many people ("the village") to provide a safe, healthy environment for children, where children are given the security they need to develop and flourish, and to be able to realize their hopes and dreams. This requires an environment where children's voices are taken seriously (2) and where multiple people (the "villagers") including parents, siblings, extended family members, neighbors, teachers, professionals, community members and policy makers, care for a child. All these 'villagers' may provide direct care to the children and/or support the parent in looking after their children. However, the village, in many countries today, is dissipated and fragmented and individuals are increasingly isolated and are not eager to ask for, or provide help to, others. Family breakdown, economic pressures, long working hours and increased mobility have all contributed to families feeling less connected to extended family members and others around them (3).

In this perspective article, we propose a village that has the capacity to provide support and guidance to families living with adversity. Inherent in the concept of the village is the notion that caring for children is a shared responsibility amongst many. In this article we explore the notion of the village further, provide case studies of when it is occurring and provide principles of a village approach.

DEFINING FAMILY

Families mean different things for different people. Osher and Osher (4) suggested that family is "defined by its members, and each family defines itself" (p. 48). Likewise, Eassom et al. (5) provided a broad approach to the definition of family, which may not necessarily include one's biological family, but instead consists of those who share a common purpose, set of conventions and customs. Hence, there are different types of families, which may include the traditional nuclear (two parent) families, single parent families, adoptive families, same-sex parents, foster families, stepfamilies, and those in which children are raised by grandparents or other relatives.

One important role of families is to provide love, guidance, care, and support for its members. How they do this will differ, according to culture, family values, and the availability of educational, economic, and welfare resources. Through an interpretative framework, parents convey to their children the values, standards and rules about relationships and social structures. In turn, parents' beliefs and practices reflect the norms and expectations of their time and the culture in which they live. All of these factors impact the family environment and inform how family members show emotions, make decisions, resolve conflicts, interact with, and care for each other. When one family member is ill, facing addiction problems, or is otherwise under stress, other family members, including children, are inevitably impacted (1). In these instances, other family members may support the family member who is ill or under stress; alternatively (or in some instances, additionally), the family may itself be the source of trauma and ongoing stress and anxiety (6).

Multiple studies have shown that compared to other children, children growing up in such families may experience negative impacts on their own mental health and well-being, physical health and education (7). However, not all children whose families experience adversity will be negatively impacted, nor will all children be affected in the same way (1). Moreover, Gladstone et al. (8) argue that rather than being passive victims, many young people living in these families have their own agency, and in the face of great adversity, can be highly resilient and active contributors to family life.

Throughout recent history there have been different ways of describing families experiencing multiple adversities. In an address at a 1946 conference, Wofinden, a public health researcher, defined families who experience problems as "families with social defectiveness of such a degree that they require care, supervision and control for their own well-being and for the well-being of others" [(9), p. 127]. He continued by suggesting that "help from outside [the family] can hardly be of permanent value, except in proportion as it tends to develop the self helping faculties" (p. 130). In more recent times, public policy has mirrored similar sentiments. The 2011 Troubled Families Programme launched in England aimed to "turn around" the lives of the 120,000 most troubled families in England by 2015. In that policy, these "troubled families" were seen to "have" problems and "cause" problems to those around them (10). Such simplistic arguments condoned and extenuated the complex and interrelated relations between socioeconomic and psychosocial problems that many families experience, often over multiple generations. Such positioning also negates the responsibility of the "village" to support families. Helming et al. (11) consolidated such arguments when they write:

> The concept of "multi-problem families" includes only the level of the family system ("families that have many problems") and hides social deprivation [and] the deprivation of these families.... The term also neglects the... the obligation of the state to intervene to regulate equal opportunities (translated from the original, p. 74)

Tausendfreund et al. (12) advocated for the term "families in multi-problem situations" rather than "multi-problem families" so that the location of the "problem" is ascribed (semantically at least) to the family's environment rather than the family itself. Similarly, Goerge and Wiegand (13) employed the term "multisystem families", though acknowledge that this only captures those problems that families seek assistance for, and that services are able to address. When responding to these families, Hayden and Jenkins (10) advocated for a two-prong government approach that involves: (i) providing immediate responses for supporting the whole family, and not only the individual adult and child "problems", and (ii), targeting underlying driving forces behind family problems, especially pertaining to unemployment and insecure housing. Defining problems by the systems families engage with and the need to look at underlying forces, underscores the need for a village approach.

SILOED PRACTICE

Typically, organizational responses and policies for families living with adversity have been siloed, for example, supporting a parent presenting for cancer treatment without consideration of the needs of his or her children (14), or working with a client's mental illness without acknowledging his or her substance use problem (15, 16). Changing siloed practices is difficult, because they are grounded in professional development and education, laws and regulations, health policy and funding models (17).

Roberts (18) described silos as the "inability to share information and integrate system activity" (p. 677). Goerge and Wiegand (13) investigated families experiencing multiple adversities in the state of Illinois (USA) and found that 23% of families surveyed received services from two or more public services, including child welfare, mental health, substance abuse services and adult and juvenile incarcerations, mirroring findings from an audit of adult and child mental health services in Northern Ireland (19). Even though these families accounted for 86% of the funding for these services, there was little coordination or collaboration of care and little or no sharing of information between services. This siloed approach results in either an overlap of services or alternatively misses critical problems that a family may want and need to address. Siloed practice models are a problem that appears to be pervasive across countries, agencies and funding models (17).

Problems that may arise in families can correlate, for example, when parents who have a history of substance use also have a mental illness (20), or when one family member who has a mental illness has other family members who experience their own mental health issues (21). The reciprocal impact between children's and parents' health should not be underestimated (22) and will also reverberate in families. Exposure to one problem often leads to other problems, such as unemployment, inadequate housing, and in some cases violence and child neglect (1). Social complexity theory may help understand the problems families face; what might seem like chaotic behaviors are instead highly organized with rehearsed patterns. Complexity theory shifts attention from a "decontextualised and universalized essence to a concern with contextualized and contingent, complex wholes" [(23), p. 119]. This necessitates looking past presenting behaviors (e.g., the reaction of children to a parent's symptomology) and instead, appreciating the ways in which interactions with others, material resources and services contribute to family experiences. Rather than see families as dysfunctional or beyond hope, we need to recognize that they may be striving for meaning and balance and doing the best that they can, in their given circumstances (24).

The complexities of these adversities further underscore the need for coordinated responses across health, housing, employment services, education, policing and other agencies and community groups, from the perinatal period through to adulthood. Different services will be needed at different times, especially for key developmental milestones, such as the birth of a new child or the death of a grandparent (25). Moreover, the impact of these adversities can be intergenerational, as the impact of the adversity is passed on through parenting practices, violence, substance misuse and mental health issues (26).

SOCIAL CONNECTEDNESS

Even though parents may be a child's primary caregivers, a family does not exist in a vacuum. Social connectedness has been defined as those subjective psychological bonds that people experience in relation to others including, for example, a sense of belonging and feeling cared for (27). It also includes objective measures such as the frequency of social participation and marital status (27). There is much evidence that strong, positive connections are linked to positive mental health and well-being, especially in times of stress or trauma (27). Social connectedness is one way of describing the members of the village and the need for families to have multiple supports. Given that responding to, and overcoming adversity, occurs in a social context that extends beyond individual and family levels, social connections for families living in adversity includes but extends past members of the immediate family.

There are, however, many families who are not included within their communities. Likewise, families with complex health and social needs may be excluded from services, for many reasons, leading to poor health outcomes and multiple morbidities and in some cases early mortality (28). Families may be excluded because of the stigma associated with adversity (such as mental illness or poverty) and an inappropriate representation in the media (29) or because they are not recognized by a government's criteria of "troubled", and are missing from public policy (10). Rather than being "hard to reach", some families may not have the ability to access services (because of transport or time), may have had negative experiences with similar services previously and/or find them intimidating or unhelpful. Some may not be aware of services that could assist them and may need professionals to serve as conduits to other services (30). The village concept implies a need to identify the magnitude of exclusion (that is, who is being excluded and from what), specify why they are excluded and, on that basis, promote access to essential services for individuals and their families and challenge societal attitudes and media misrepresentations. Families need different forms of connections, formal and informal, from the individual level to the policy and government level, to address the upstream causes of exclusion and disadvantage, including adverse childhood experiences and poverty.

WHO IS IN THE VILLAGE?

Bronfenbrenner's ecological theory (31) highlights the various factors that impact on children's learning and development. We

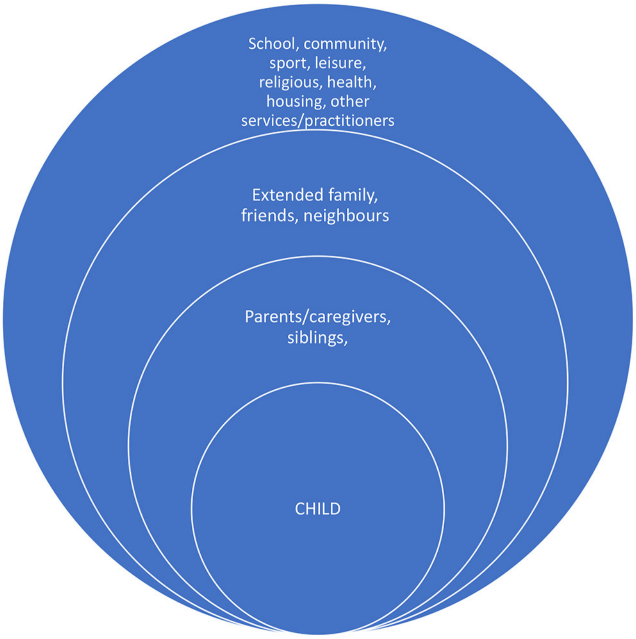


FIGURE 1 | Possible connections for families living in adversity.

have extrapolated from that model to highlight the connections that families might have, in each sphere, as one possible indication of who might be the "villagers" (see **Figure 1**). This figure demonstrates how different social connections impact children's outcomes, across varying proximity levels (though this may also vary for different families). Culture, socioeconomic status and language provide further context to this figure. It is the richness (quality and quantity) of these connections that can have a significant influence on the quality of the child rearing that a parent provides and the types of connections that children might make (32). Synergy is an important aspect of this model, which implies that families, schools, community groups and agencies working together can achieve more than either could alone (33).

Supportive connections with village members are valuable for both children and parents. In her seminal longitudinal study of high-risk children, Werner (34) found that children from high risk backgrounds, who formed bonds with caring and trusting other adults, turned out to be more resilient, than those who did not form such connections. Connections also help parents; Garbarino and Sherman (35) found that parents who have access to social networks and supports when looking after children report less parenting stress, than other parents. Likewise, communities with strong formal and informal networks are associated with lower rates of child maltreatment, compared to communities characterized by social disorganization and low levels of social cohesion (36).

Though the importance of social connections might be self-evident, Kesselring (32) argued that in western societies there is a trend toward parenting as a private concern, and when any presenting problems (experienced by the child or family) are referred to professionals rather than shared amongst the family's social networks. In this approach, the village shrinks considerably, especially when professional services are limited or are not accessed by the family (for whatever reason). However, in many societies, nonparental caretaking is either the norm or occurs frequently. Donner (37) found that in Polynesian society both parents and nonparents were involved in the upbringing of other people's children. Polynesian adults viewed the western ideal of sole parental responsibility as a "lack [of] compassion" for other people's children (p. 703). Likewise, Otto (38) found that Cameroonian Nso mothers discouraged maternal exclusivity, believing that multiple caregivers are optimal, with one mother stating, "Just one person cannot take care of a child throughout" (p. 95).

There are, however, times when parents in western cultures draw on different members of the village. In the UK, Edwards and Gillies (39) found that although many parents receive less informal support than in the past (because of divorce, or because extended families are geographically dispersed), parents still identified relatives and friends as the main source of emotional support and advice about their children's behavior. In the USA, Burchinal et al. (40) found that in communities where neighbors trust each other, parents are more likely to utilize informal childcare from their neighbors, rather than relying exclusively on their relatives to look after their children, when working or ill. Both neighbors and parents can be involved in caring for children when they have "shared expectations and mutual engagement by adults in the active support and social control of children" [(41), p. 635]. Professionals, such as teachers and youth workers, play a role in these neighborhoods by organizing neighborhood activities and events and by "caring" for children (42). Governments in many western countries focus on parenting in public provision and policy, and provide some families with government financial support and information and hands-on support through different initiatives and parenting programs (43). There are also different parenting blogs and other online sites that parents might access, to meet other parents and/or obtain emotional support and advice. In sum, the different connections that a family might make (**Figure 1**) help us understand the different forms of support that may be provided, and those that may be missing.

POTENTIAL PRINCIPLES OF THE VILLAGE APPROACH

The connections within the village approach are important, but how these connections might confer protection or buffer the impact of adversity is not always clear. Articulating principles for a village approach serves as the first step in operationalising the village approach. Based on our collective experiences as researchers and clinicians, these principles have been outlined in **Table 1**, along with practice and/or policy implications. These principles might be used to develop new initiatives and evaluate existing ones, an important future direction in the field.

Applying these principles in practice is the next step to which might challenge the social factures that inhibit the notion of the village that may intentionally or unintentionally exclude

TABLE 1 | Potential principles of the village approach.

Village principle	Practice and policy implication
Interdisciplinary	Practitioners from various professional disciplines, including but not limited to physical health, psychology, social work, and education, are provided with the training and time to work collaboratively
Interagency	Coordinated interagency support is provided to families depending on need, including but not limited to housing, employment, child care and education
Strength based	Family, parenting and children's strengths and resources are identified, recorded and celebrated.
Prevention focused	Support aims to prevent immediate and long-term problems.
Developmental, lifespan approach	Different support is provided to parents/caregivers and children at different times, depending on key developmental milestones.
Promoting parents' agency and empowerment	The views and perspectives of parents is actively sought when defining problems and solutions. Parents are partners in the planning and delivery of services.
Giving children a voice	Children of all ages are encouraged to present their perspectives on the issues and potential solutions to existing and future family issues
Culturally sensitive	Individual, familial and communal cultures are acknowledged and considered when addressing problems and solutions.
Feedback and evaluation	Feedback and evaluation processes are built into Village-focused policies and practices

families. In this final section we provide two case studies which demonstrate the ways in which "the village" might be enacted.

Harlem Children's Zone

Aiming to improve the educational and developmental outcomes for children in one of America's most impoverished communities, the Harlem Children's Zone (HCZ) is a non-profit organization for children and families that includes community building, the promotion of parent networks and neighborhood safety, and child-oriented education and health programs (44). By promoting a sense of community, HCZ addresses a constellation of factors that might negatively impact families. Individual programs may be delivered, for example, that focus on housing, but these incorporate a mandate to foster community connections and support healthy physical and social environments. HCZ services are structured into a "pipeline" of continuous support from a child's birth through to college graduation. Services include parenting supports, which provide a safe space for parents to connect with others and provide information on parenting best practices and pathways to coordinate and navigate services. Evaluations indicate that HCZ significantly increased academic achievement for children living in adversity (45), and has positive impacts on children's weight and physical health (46).

The HCZ incorporates many of the principles covered by the village approach (**Table 1**) by providing an interdisciplinary, interagency approach and before that is prevention and youth focused. The "pipeline" of supports is clearly developmentally orientated and its focus on parenting support promotes parents' agency and autonomy. Nonetheless, there have been calls for further evaluation to demonstrate the efficacy of this approach, especially in regards to impacts on children's well-being, in addition to their academic outcomes (44).

Strategies With Kids–Information for Parents

Developed in New Zealand, SKIP is a government funding program that aims to increase the opportunities for communities to promote positive parenting, for families living in adversity (47). SKIP employs an open tender process in which the government invites local organizations and groups to submit proposals that aim to support families in a holistic, culturally sound manner. For example, one initiative brings parents together to share successful strategies for positively managing challenging behavior in their preschool children, while another identifies community and agency partnerships for addressing community violence. Its approach affirms the role of parent and provides the pathways for normalizing help seeking in communities, in culturally appropriate ways. The initiative's locally driven, strength-based approach aligns with the village principles (**Table 1**) as does its focus on promoting parents' agency and empowerment. However, children's voices appear to be lacking as is any form of rigorous evaluation and monitoring.

The two case studies illustrate community led approaches that aim to promote the development of a village approach that benefits children and their families. Both demonstrate the large number and range of initiatives offered, which draw on existing capacity and address the specific needs of the local community. The most common activities appear to be the active involvement of parents in the planning and development of programs, active community engagement, and promoting safe, family friendly environments. The potential to use community settings, such as schools, to upscale interventions is also evident, allowing local communities to drive programs adapted to their context within existing resources.

The Austrian project mentioned at the start of this perspective, titled *How to raise a village to raise a child*, has a program theory model that the authors argued promotes the capacity of the village to care for children and families; this model outlines resource inputs, systems and individual context considerations and triggers for behavior changes (48), with a particular focus on translation and implementation (49). A subsequent article further emphasized the importance of regional context specific solutions and engagement with local and experienced stakeholders to ensure service models are implementation ready (50). As the authors themselves conceded, their work to date has not yet demonstrated the effectiveness of this approach, especially in terms of child outcomes (50). A major issue in the field appears to be that many of these broad community projects have not been rigorously evaluated, especially in regard to how children may benefit. Some of the reasons for this, at least partially, may be that the principles of a village approach are rarely articulated, hence the need for this article. Additionally, as Nicholson (51) argued, the complexity involved in an ecological model of family functioning makes gold standard evaluations (typically employing a randomized controlled trial) difficult to conduct; we would suggest that a village approach makes conducting an evaluation even more challenging but one that researchers are currently addressing [see for example, (48)].

Many of those who organized and participated in the 2018 *It takes a village* conference, were involved in the writing of an editorial which outlined various recommendations for systems and workforce change, (52) and which generated much traditional and social media interest. The recommendations article, the two case studies shown, and the recent Austrian project, indicate that there is interest in the concept of the village. However, further research is required to demonstrate how a village approach might be enacted in different settings and with different families, and in particular, evaluating its long term impact on families.

CONCLUSION

This article provided one perspective of a "village" approach when supporting families who experience various challenges. We describe a village approach, which ranged from immediate child and family level responses through to government lead initiatives that services and governments might need to consider when developing practice guidelines and public health policy. The connections and principles identified in this perspective might serve as the framework from which new initiatives could be developed and existing programs evaluated. These connections and principles are even more pertinent given the struggles experienced by families and communities throughout the COVID-19 pandemic (53).

AUTHOR CONTRIBUTIONS

All authors were responsible for the conceptualization of the article and contributed to its writing.

REFERENCES

1. Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman M. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. Cambridge, UK: Cambridge University Press (2015)
2. Gladstone B, Exenberger S, Weimand B, Lui V, Haid-Stecher N, Geretsegger M. The capability approach in research about children and childhood: a scoping review. *Child Indic Res.* (2021) 14:453–75. doi: 10.1007/s12187-020-09766-0
3. Phillips NK, Straussner SLA, editors. *Children in the Urban Environment: Linking Social Policy and Clinical Practice (3rd Ed).* Springfield, IL: Charles C. Thomas (2017)
4. Osher TW, Osher DM. The paradigm shift to true collaboration with families. *J Child Fam Stud.* (2002) 11:47–60. doi: 10.1023/A:1014715527823
5. Eassom E, Giacco D, Dirik A, Priebe S. Implementing family involvement in the treatment of patients with psychosis: a systematic review of facilitating and hindering factors. *BMJ Open.* (2014) 4:e006108. doi: 10.1136/bmjopen-2014-006108
6. Bouverie Centre. *From Individuals to Families: A Client-Centred Framework For Involving Families*. (2016). Available online at: https://www.bouverie.org.au/images/uploads/Bouverie_Centre_Framework.pdf
7. Straussner SLA, Fewell CF. A review of recent literature on the impact of parental substance use disorders on children and the provision of effective services. *Curr Opin Psychiatry*. (2018) 31:363–7. doi: 10.1097/YCO.0000000000000421
8. Gladstone BM, Boydell KM, McKeever P. Recasting research into children's experiences of parental mental illness: beyond risk and resilience. *Soc Sci Med.* (2006) 62:2540–50. doi: 10.1016/j.socscimed.2005.10.038
9. Wofinden RC. Problem families. *Eugen Rev.* (1946) 38:127–32
10. Hayden C, Jenkins C. 'Troubled families' programme in England: 'wicked problems' and policy-based evidence. *Policy Stud.* (2014) 35:631–49. doi: 10.1080/01442872.2014.971732
11. Helming E, Schattner H, Blüml H, editors. Handbuch sozialpädagogische Familienhilfe [Handbook social pedagogical family support]. Baden-Baden, Germany: Nomos (2004).
12. Tausendfreund T, Knot-Dickscheit J, Schulze GC, Knorth EJ, Grietens H. Families in multi-problem situations: Backgrounds, characteristics, care services. *Child Youth Serv.* (2016) 37:4–22. doi: 10.1080/0145935X.2015.1052133
13. Goerge RM, Wiegand ER. Understanding vulnerable families in multiple service systems. *J Soc Sci.* (2019) 5:86–104. doi: 10.7758/rsf.2019.5.2.05
14. Ellis SJ, Wakefield CE, Antill G, Burns M, Patterson P. Supporting children facing a parent's cancer diagnosis: a systematic review of children's psychosocial needs and existing interventions. *Eur J Cancer Care.* (2016) 26:e12432. doi: 10.1111/ecc.12432
15. Reupert A. Maybery D. Practitioners' experiences of working with families with complex needs. *J Psychiatr Ment Health Nurs.* (2014) 21:642–51. doi: 10.1111/jpm.12149
16. Senreich E, Straussner SLA. Screening and brief interventions. In Stanhope V, Straussner SLA, editors. *Social Work and Integrated Health Care: From Policy to Practice and Back*. New York: Oxford University Press (2017).
17. Nicholson J, Reupert AE, Grant A, Lees R, Maybery DJ, Mordoch E, et al. The policy context and change for families living with parental mental illness. In Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman M, Editors. *Parental psychiatric disorder: Distressed parents and their families* (pp. 354-364). Cambridge UK: Cambridge University Press (2015).
18. Roberts NC. Beyond smokestacks and silos: open-source, web-enabled coordination in organizations and networks. *Public Adm Rev.* (2011) 71:677–93. doi: 10.1111/j.1540-6210.2011.02406.x
19. McCartan C, Bunting L, Davidson G, Devaney J, Donaghy M, Dufy J, et al. *Think Family NI Audit of Adult Mental Health and Children's Services*. Queens's University, Belfast (2020). Available online at: https://pure.qub.ac.uk/en/publications/think-family-ni-audit-in-adult-mental-health-and-childrens-servic
20. Ali MM, Dean D, Hedden SL. The relationship between parental mental illness and/or substance use disorder on adolescent substance use disorder: results from a nationally representative survey. *Addict Behav.* (2016) 59:35–41. doi: 10.1016/j.addbeh.2016.03.019
21. Naughton M, Maybery D, Sutton K, Goodyear M. Impact of parental mental illness on children's HoNOSCA results in a regional child and adolescent mental health service. *Int J Ment Health Nurs.* (2019) 28:1053–64. doi: 10.1111/inm.12609
22. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family focused practice in mental health care: an integrative review. *Child Youth Serv.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
23. Mason M. Making educational development and change sustainable: insights from complexity theory. *Int J Educ Dev.* (2009) 29:117–24. doi: 10.1016/j.ijedudev.2008.09.005

24. Sanders J, Munford R, Liebenberg L. Young people, their families and social supports: understanding resilience with complexity theory. In: Ungar M, Editor. *The Social Ecology of Resilience*. New York, NY: Springer (2012). p. 233–43.
25. McGoldrick M, Carter B, Garcia-Preto N. *The expanded family life cycle: Individual, family, and social perspectives* (4th ed). New York: Guilford Press (2011).
26. Spratt T. Families with multiple problems: some challenges in identifying and providing services to those experiencing adversities across the life course. *J Soc Work.* (2011) 11:343–57. doi: 10.1177/1468017310379256
27. Haslam C, Cruwys T, Haslam A, Jetten J. Social connectedness and health. In: Pachana N, Editor. *Encyclopedia of geropsychology*. Singapore: Springer (2015).
28. Luchenski S, Maguire N, Aldridge RW, Hayward A, Story A, Perri P, et al. What works in inclusion health: overview of effective interventions for marginalised and excluded populations. *Lancet.* (2018) 391:266–80. doi: 10.1016/S0140-6736(17)31959-1
29. Beddoe L. Feral families, troubled families: The spectre of the underclass in New Zealand. *N Z Sociol.* (2014) 29:51–68.
30. Reupert A, Deppeler J, Sharma U. Perspectives of parents of children with autism spectrum disorder. *Australas J Spec Educ.* (2015) 39:85–96. doi: 10.1017/jse.2014.17
31. Bronfenbrenner U. Ecological systems theory. In: Bronfenbrenner U, editor. *Making human beings human: Bioecological perspectives on human development*. Thousand Oaks, CA: Sage Publications Ltd (2005). p. 106–73.
32. Kesselring M, De Winter M, Van Yperen T, Lecluijze S. Partners in parenting: An overview of the literature on parents' and nonparental adults' perspectives on shared responsibilities in childrearing. *Soc Probl.* (2016) 4:69–97. doi: 10.5296/iss.v4i1.8764
33. Christenson SL. The family-school partnership: an opportunity to promote the learning competence of all students. *School Psych Rev.* (2004) 33:454–82. doi: 10.1080/02796015.2004.12086233
34. Werner EE. Risk, resilience, and recovery: perspectives from the Kauai longitudinal study. *Dev Psychopathol.* (1993) 5:503–15. doi: 10.1017/S095457940000612X
35. Garbarino J, Sherman D. High-risk neighborhoods and high-risk families: the human ecology of child maltreatment. *Child Dev.* (1980) 51:188–98. doi: 10.2307/1129606
36. Garbarino J, Kostelny K. Child maltreatment as a community problem. *Child Abuse and Neglect.* (1992) 16:455–64. doi: 10.1016/0145-2134(92)90062-V
37. Donner WW. Sharing and compassion: fosterage in a polynesian society. *J Comp Fam Stud.* (1999) 30:703–18. doi: 10.3138/jcfs.30.4.703
38. Otto H. *Culture specific attachment strategies in the Cameroonian Nso. Cultural solutions to a universal developmental task*. Dissertation, Osnabrück (2008). Available online at: https://osnadocs.ub.uni-osnabrueck.de/bitstream/urn:nbn:de:gbv:700-2009050119/2/E-Diss881_thesis.pdf.
39. Edwards R, Gillies V. Support in parenting: values and consensus concerning who to turn to. *J Soc Policy.* (2004) 33:627–47. doi: 10.1017/S0047279404008037
40. Burchinal M, Nelson L, Carlson M, Brooks-Gunn J. Neighborhood characteristics and child care type and quality. *Early Educ Dev.* (2008) 19:702–25. doi: 10.1080/10409280802375273
41. Sampson RJ, Morenoff JD, Earls F. Beyond social capital: spatial dynamics of collective efficacy for children. *Am Sociol Rev.* (1999) 64:633–60. doi: 10.2307/2657367
42. Laletas S, Reupert A. Exploring pre-service teachers' understanding of care. *Teach Teach.* (2015) 22:485–503. doi: 10.1080/13540602.2015.1082730
43. Daly M, Bruckhauf Z, Byrne J, Pecnik N, Samms-Vaughan M, Bray R, et al. *Family and Parenting Support: Policy and Provision in a Global context*. Florence: Innocenti Insight, UNICEF Office of Research (2015). Available online at: https://www.unicef-irc.org/publications/770-family-and-parenting-support-policy-and-provision-in-a-global-context.html
44. Jean-Louis B, Benitex L. Creating a village that nurtures children at the Harlem Children's Zone. In: Gordon EW, et al., editors. *Strengthening Families, Communities and Schools to Support Children's Development*. Taylor and Francis (2017). p. 135–57. doi: 10.4324/9781315161754-11
45. Dobbie W, Fryer RG. Are high quality schools enough to increase achievement among the poor? Evidence from the harlem children's zone. *Am Econ J.* (2011) 3:158–87. doi: 10.1257/app.3.3.158
46. Mabli J, Bleeker M, Fox MK, Jean-Louis B, Fox M. Randomized controlled trial of Healthy Harlem's Get Fit Program: an after-school intervention for childhood overweight and obesity in Harlem's Children's Zone. *Child Obes.* (2020) 16:479–87. doi: 10.1089/chi.2020.0012
47. Ministry of Social Development. Strategies with Kids – Information for parents, research report. New Zealand Government (2005). Available online at: https://www.msd.govt.nz/about-msd-and-our-work/publications-resources/research/skip-research/index.html
48. Bauer A, Best S, Malley J, Christiansen H, Goodyear M, Zechmeister-Koss I, et al. Towards a program theory for family-focused practice in adult mental health care settings: an international interview study with program leaders. *Front Psychiatry*. (2021) 12:741225. doi: 10.3389/fpsyt.2021.741225
49. Zechmeister I, Goodyear M, Tüchler H, Paul JL. Supporting children who have a parent with a mental illness in Tyrol: a situational analysis for informing co-development and implementation of practice changes. *BMC Health Serv Res.* (2020) 20:326. doi: 10.1186/s12913-020-05184-8
50. Goodyear M, Zechmeister-Koss I, Bauer A, Christiansen H, Glatz-Grugger M, Paul JL. Development of an evidence-informed and co-designed model of support for children of parents with a mental illness – "It takes a village" approach. *Front Psychiatry*. (2022) 12:806884. doi: 10.3389/fpsyt.2021.806884
51. Nicholson J. Building the evidence base for families living with parental mental illness. *AeJAMH*. (2009) 8:222–6. doi: 10.5172/jamh.8.3.222
52. Reupert A, Bee P, Hosman C, van Doesum K, Drost LM, Falkov A, et al. Prato research collaborative for change in parent and child mental health: principles and recommendations for working with children and parents living with parental mental illness. *J Child Psychol Psychiatry*. (2021) 63:350–3. doi: 10.1111/jcpp.13521
53. Berger E, Reupert A, Miko A, Negar J, Jobson L. A systematic review of the mental health implications for children impacted by infectious outbreaks. *Child Adolesc Ment Health.* (2021) 26:157–66. doi: 10.1111/camh.12453

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 14 March 2022
doi: 10.3389/fpsyt.2022.814185

A Challenging Yet Motivating Journey: The Experiences of Young Adult Parents With Serious Mental Health Conditions in the USA

***Kathryn Sabella*[1]*, *Amanda Baczko*[1], *Ian A. Lane*[1], *Laura Golden*[2], *Emma Pici-D'Ottavio*[1] and *Murron O'Neill*[3]**

[1] Transitions to Adulthood Center for Research, Implementation Science and Practice Advances Research Center (iSPARC), Department of Psychiatry, University of Massachusetts Chan Medical School, Worcester, MA, United States, [2] Sociology Department, University of Massachusetts Boston, Boston, MA, United States, [3] School of Social Work, Boston University, Boston, MA, United States

In recent decades the average age of becoming a parent has increased, the rate of teen pregnancies has decreased, and a new developmental period of emerging adulthood is marked by diverse pathways into adulthood. Today, those who become parents in young adulthood (18–24 years old) and their children may be vulnerable to poor outcomes observed in teen parents (13–19 years old) of previous generations. Young adults with serious mental health conditions (SMHC) who encounter additional challenges navigating young adulthood and tend to parent earlier than their peers may be at particularly increased risk of poor outcomes. To date, little research has been done to understand the experiences of young adult parents, especially those with SMHC. This study describes themes from qualitative interviews with 18 young adults with SMHC in the United States who became parents before the age of 25. Life story narrative interviews, conducted mostly by young adults with lived experience, asked participants to describe their parenting and mental health experiences and their school, training, and work experiences. Participants described the challenges of simultaneously parenting young children and managing a mental health condition, experiences of discrimination, and fear of future discrimination related to their mental health condition. However, parents also expressed that their children motivated them to maintain recovery and build a good life for their family. This is the first study to qualitatively explore the experiences of young adult parents with SMHC. While many of these findings align with prior qualitative research on mothers with mental illness, by exclusively focusing on individuals who become parents earlier than their peers and including father experiences, this research adds to our understanding of how individuals simultaneously navigate parenting and managing a serious mental health condition. These findings should inform larger-scale research studies on the experiences and outcomes of young adults with SMHC who become parents in their late teens or early twenties. A better understanding of their experiences should inform public mental health services that incorporate parenting as an important element of an individual's personal recovery model.

Keywords: emerging adult, mental health conditions (MHCs), young adulthood (18 years older), parenting with mental illness, qualitative

Edited by:
Joanne Nicholson,
Brandeis University, United States

Reviewed by:
Miriam Heyman,
Brandeis University, United States
Tracy Gladstone,
Wellesley College, United States

Correspondence:
Kathryn Sabella
kathryn.sabella@umassmed.edu

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *17 December 2021*
Accepted: *07 February 2022*
Published: *14 March 2022*

Citation:
Sabella K, Baczko A, Lane IA, Golden L, Pici-D'Ottavio E and O'Neill M (2022) A Challenging Yet Motivating Journey: The Experiences of Young Adult Parents With Serious Mental Health Conditions in the USA. Front. Psychiatry 13:814185. doi: 10.3389/fpsyt.2022.814185

INTRODUCTION

As the average age of becoming a parent has increased in recent decades, those who become parents in young adulthood (18–25 years old) and their children may be vulnerable to poor outcomes like those of teen parents (13–19 years old) and their children from previous generations. Particularly at risk are young adults with serious mental health conditions (SMHC)[1] who often have poorer education, employment, and housing outcomes and tend to become parents earlier than their peers. By virtue of their age and their mental health condition, young adults with SMHC, and their children, may be at higher risk for negative outcomes (1, 2). Thus, targeted services to support young adult parents with SMHC are imperative. However, such services will only be effective if they are informed by the lived experiences of young adult parents with SMHC. To date, research on this population is scant. This paper will describe results from qualitative interviews with young adults with SMHC who became parents in their teens or early twenties. This information should be used to inform public health and mental health programming.

Teen Parents and Their Children: Historical Health Outcomes and Public Response

In the 1990s, the birth rate for females ages 15–19 peaked at 61.6 births per 1,000 women (3). During this time, research increasingly showed that teen parents and their children were at increased risk of several poor outcomes. Teen parenting was linked to increased rates of poverty and decreased social well-being in parents (4, 5). Additionally, the educational pursuits of teen parents were often negatively impacted. In 2008, over 90% of the general population received their high school diploma by age 22 compared to only 50% of teen mothers (4). Completing continued education was even less likely; only 10% of teen mothers pursued and completed an associate or bachelor degree program (4). Children born to teen parents experienced disproportionate rates of health and development issues including low birth weight and infant mortality (6, 7). Studies also showed that children born to teen parents were more likely to be victims of neglect with rates of maltreatment estimated at over 50% (8, 9). As children of teen parents grew up, they also experienced higher rates of teen pregnancy, poorer educational outcomes, and were at increased risk of substance use disorders and SMHC (10, 11).

In response, several public health initiatives emerged to reduce the rate of unplanned teen pregnancies (12). For example, the National Campaign to Prevent Teen and Unplanned Pregnancy, launched in 1996, developed and distributed public health knowledge to inform the conversation on teen pregnancy and parenting (13). The Teen Pregnancy Prevention Program (TPPP), an evidence-based program enacted in 2010, distributes grant funding to programs across the US aimed at preventing teen pregnancy (14). TPPP programs and many other successful public health initiatives have helped to develop and disseminate comprehensive sexual education and provide funding for family planning and young adult development (14). Many of these initiatives boast success in reducing teen pregnancies; teen pregnancy rates have steadily declined and currently hover around 7–16% (15).

Young Adulthood and Young Parenthood in the 21st Century

Over the last two decades, the period of young adulthood (broadly ages 18–30) has evolved into a newly understood life-phase known as "emerging adulthood" (16). Normative activities that previously marked one's transition into adulthood (e.g., completing education, securing stable employment, marrying, and becoming a parent) now typically occur several years later than in previous generations (16). In the United States, a 2015 report by the National Academy of Medicine (formerly the Institute of Medicine) and the National Research Council concluded that young adulthood (17–25) is a critical developmental period marked by increasingly diverse pathways (26).

In the United States, the lengthening transition from adolescence to adulthood is evident in patterns of employment, education, and living status. Between 1960 and 2018 the percentage of recent high school graduates who went on to higher education increased from 45.1 to 69.1 percent (17). In recent years, even before the COVID pandemic, the rate of young adults living at home with their parents was the highest rate since the Great Depression (18). Emerging adulthood has also been marked by a steady increase in the average age of first birth. Between 1970 and 2000, the mean age for women having their first live birth in the United States increased by 3.6 years (19). That rate has accelerated over the last two decades; census data show that between 1994 and 2018, the average age for women having their first live birth increased another three years from age 23 to 26 (20). This trend is evident across other datasets (21). The average age men are becoming fathers has also increased by several years (22, 23).

At the end of the 20th century, poor outcomes among teen parents and their children, such as those described earlier, reflected concerns that teen parents did not have sufficient physical and emotional maturity to provide effective care for a child and that they may not have adequate economic resources (24). In the U.S., public health officials framed these data to portray teen pregnancy as a social problem that needed to be addressed. However, as the average age of first birth has increased over this time, and a unique developmental period known as emerging adulthood has evolved, individuals who become parents significantly earlier than their peers, even if not technically "teen parents," may face challenges similar to those of teen parents in earlier generations.

Historically, research on teenage parenting utilized an absolute age cut-off (i.e., the age of 20) to define a group of parents most at risk for poor outcomes. However, the timing of what constitutes "early" parenting differs by society and over time. A

---

[1] SMHC refers to having been diagnosed either with a serious emotional disturbance (often used in children) or a serious mental illness (often used with adults) as defined in the Federal Register [5/20/1993, FR, 58(96). P. 29422]. SMHC includes schizophrenia-spectrum disorders, bipolar disorder, and severe forms of depression, anxiety, and posttraumatic stress.

recent cross-national comparative study looking at educational differences in early childbearing defined "early childbearing" as the age by which 20% of first births have occurred to women in a given birth cohort and country (24). In this definition, "early childbearing" is defined in relative terms, i.e., in relation to the normative timing of childbearing within a given society or cohort. The authors argue that since teenage childbearing is relatively rare (and has been declining) in many countries, its utility as an absolute measure of "early childbearing" is waning. Taken together, elongated and diverse pathways from adolescence to adulthood and the increasing average age of first birth necessitate an updated definition of what constitutes a "young parent" or "early childbearing" in the United States. Following this logic, the high-risk category that was previously defined as "teen parents" should now be redefined in more relative terms. For instance, in the United States, only 30% of pregnancies happen prior to the age of 24 (25). Accordingly, the outcomes and needs of young parents in their late teens *or* early twenties might be poorer relative to people who become parents later in life.

Young Adult Parents With SMHC

Pathways through the transition to adulthood are more diverse and increasingly difficult to predict (26). Most mental health conditions are diagnosed by age 24 (27) and it is estimated that between 5 and 7 million young adults (ages 16–25) in the United States have serious mental health conditions (SMHC) (28, 29). The term "serious mental health conditions" (SMHC) is used to be inclusive of mental health diagnoses of "serious emotional disturbance" in childhood and diagnoses of "serious mental illness" in adulthood. SMHC includes schizophrenia-spectrum disorders, bipolar disorder, and severe forms of depression, anxiety, and post-traumatic stress disorders. In the U.S., mental health conditions are the cause of 45% of the burden of disability in older youth and young adults (26). Compared to their peers, young adults with SMHC experience lower rates of high school graduation (30), higher rates of college drop-out (31, 32), and are less likely to be employed (31, 33–35). Youth and young adults with SMHC are over-represented in homeless populations (36, 37) and have high rates of co-occurring substance use (28).

On average, young adults with SMHC become first-time parents at an earlier age than their peers without SMHCs (2, 38, 39). The average age of becoming a parent is currently 26 years old for females (40) and 31 years old for males (22). Meanwhile, depending on the diagnosis, the average age of becoming a parent for women living with SMHC ranges from 19 to 22 years and from 24 to 25 years of age for men with SMHC (2). Prior research shows that among young adults ages 18 to 26 years, 29% of those with SMHC are parents of at least one child, compared to just 19% of their peers without SMHC (39). Based on estimated mental illness prevalence rates cited earlier, we estimate there are 1–2 million young adults with SMHC who are parents of young children in the United States.

Based on the limited research that is available, parents with serious mental health conditions (SMHC) of any age face a myriad of challenges as they navigate adulthood. Parents with SMHC experience higher rates of poverty, lower employment rates, and higher rates of enrollment in Supplemental Security Income (SSI) or Social Security Disability Insurance (SSDI) than parents without SMHC (1, 2). They are more likely to be living without partners and managing parenting and financial responsibilities without spousal support (2). Parents with SMHC face the additional barrier of stigma and discrimination when disclosing their mental health diagnosis (41). As a result, parents with SMHC may not be forthcoming about their challenges or needs (42).

Research has found that parents with SMHC often experience less positive relationships and poor attachment with their children compared to parents without mental illness (43–46). Although experiences can vary by many contextual circumstances (e.g., race/ethnicity, social class, severity of mental health needs), parental mental illness can also be associated with low family cohesion (47) and higher family conflict, which can potentially lead to overall more problematic behaviors in the children (48). For instance, children of parents with SMHC can experience increased levels of distress, deviance, and heavy drinking in adolescence (49) and demonstrate less academic competence (50). Children of parents with mental illness may experience negative emotions such as anger, fear, and sadness more frequently, which may lead to additional internalizing problems (51, 52) or externalizing problems (53). One study found that about one-third of children of parents with SMHC are at risk of developing a serious mental illness themselves (54).

The Need to Better Support Young Adult Parents With SMHC

Taken together, young adult parents with SMHC may be particularly at risk for poor outcomes by virtue of both their age (relative to average age of becoming a parent) and their SMHC. Young adult parents are often unable to engage in age-normative activities (e.g., employment and post-secondary education), which are primary pillars of their mental health recovery and long-term career trajectories (55). Parenting in young adulthood creates additional psychological strain for those with preexisting SMHC. The existing research, mostly with mothers with mental illness, illustrate how the normal stresses of parenting are exacerbated when also struggling to manage one's own mental illness (56, 57) and managing competing identities of "mother" and "person with mental illness" (58). Furthermore, mothers with SMHC struggle to juggle the practical demands of parenting and stigma while taking care of themselves and their condition; maintaining consistent engagement with mental health services appears to be particularly challenging (56–58). Regardless of the challenges, many mothers with mental illness have reported that parenting and caretaking have positive effects on their overall well-being and that they feel a lot of pride for being a parent (42, 59, 60). However, most of this exploratory research was limited to only the experiences of mothers, and also mothers with mental illness who were in their late 20s or 30s.

Outcomes of children of parents with SMHC are the result of a complex web of risk and protective factors in the family and social environment (61, 62) that can create a particularly complex mental and public health issue with no easy solution

(63). Existing interventions have been found to significantly reduce the risk of children exhibiting internalizing symptoms or developing the same mental illness as their parent (64). However, many promising interventions are still being tested, are not widely disseminated, and have not been implemented in diverse settings (65).

To be effective, interventions to support parents with mental illness need to be informed by their needs and experiences. While there is some qualitative literature on parents with mental illness, especially mothers, descriptive and exploratory research on the experiences of young adult parents with SMHC is almost non-existent. The current study aims to describe the experiences of young adults with SMHC who became parents at age 25 or younger (i.e., young parents) and how they navigate young adulthood while managing a SMHC and parenting young children. Findings from this study will invaluably inform efforts in the public health sector to adequately support and tailor services to address the unique needs of this population.

MATERIALS AND METHODS

Community Based Participatory Research

This study applied principles of Community Based Participatory Research (CBPR) by collaboratively and equitably involving community members and researchers in all aspects of the research process (66, 67). The center that housed this research employs young adults with lived experience of a mental health condition as research coordinators. This research was motivated by the lived experience of a young adult staff member who was parenting a young child. She and other young adult staff contributed to the iterative design of the study, led recruitment activities, conducted most of the interviews, and participated in the analyses, interpretation, and dissemination of study findings, including the writing of this article. We also acquired input from the Youth and Young Adult Advisory Board (YAB) affiliated with the research center that housed this study. The board is comprised of 8–10 young adults with lived experience of a mental health condition from across the U.S. They met monthly to provide input and feedback to all center projects. The principal investigator and research coordinators met with the board several times to obtain input on the goals of the study, get feedback on recruitment activities and materials, and review, pilot, and edit the interview script.

Participants

The young adult parents in this paper represent a subgroup of participants from a larger qualitative interview study of 61 young adults with SMHC in the greater Boston and Central Massachusetts area. The purpose of the larger qualitative study was to explore and describe the experiences of navigating employment, education, and training in young adulthood while managing a mental health condition. In order to be eligible to participate in the larger study, participants had to be between the ages of 25–30, have some school or work experience, and report having been diagnosed with a serious mental health condition (e.g., major depression, anxiety disorder, bipolar disorder, schizophrenia) that was diagnosed by a medical provider at or before age twenty-two. Research and policy relative to individuals with mental health conditions often distinguish the level of impairment or seriousness associated with that condition (e.g., no impairment, moderate impairment, or serious impairment). Unfortunately, definitions of what entails "functional impairment" are not universal. The research team collectively identified several indicators that the young adult's ability to function was impaired at some point due to their mental health condition. As a result, participants had to endorse one of the following experiences: receiving intensive outpatient or inpatient mental health treatment, receiving services by the Massachusetts Department of Mental Health, receiving formal special education services because of the mental health condition (includes have an IEP or a 504 plan), or having to take a formal leave of absence from school or work due to their mental health condition. To explore our research aims regarding young parents with SMHC we (a) extended the age eligibility range down to age 22, and (b) oversampled young adults who had at least one child 12 months or older living with them for at least 50% of the time for a minimum of six consecutive months. Exclusion criteria for the study included being unable or unwilling to provide informed consent or being unable to read and understand written and spoken English.

Recruitment and Procedures

Participants were recruited via flyers and electronic announcements shared with community mental health agencies, clubhouses, and Department of Mental Health providers. The center shared the research opportunity on social media and through electronic newsletters. It is important to note that these more general recruitment strategies resulted in a sizable sample of young adults but did not reach a sufficient number of young adult parents with SMHC. After consulting with our advisory boards and providers in the area, we broadened our recruitment to more general social service agencies (e.g., those served by agencies other than Department of Mental Health) and homeless shelters. These additional recruitment strategies led to the recruitment of more young parents with SMHC. Study flyers offered contact information to provide more information about one's involvement and voluntary participation in the study. Interested young adults were contacted by research staff and completed brief phone screenings to assess eligibility. If deemed eligible, research staff scheduled their interview. All procedures were approved by the Institutional Review Board at the University of Massachusetts Chan Medical School.

Data Collection

The research team conducted semi-structured qualitative interviews in 2016 with study participants who identified as young adult parents ($n = 18$). Most interviews ($n = 17$) were facilitated by young adult research staff members with lived experience of a mental health condition and took about 90 min to complete. The remainder were conducted by the Principal Investigator. Interviews took place in-person in community settings, at a convenient location identified by the study participant. Interviewers reviewed informed consent procedures, asked participants to complete a brief demographic survey, and conducted the semi-structured interview. Two participants declined being recorded and instead detailed qualitative notes were taken by the interviewer. Participants were each compensated with a $30 gift card upon completion of the interviews.

The interview script was modeled as a life story narrative interview (68). The interviewer asked the participants to describe, (a) their educational, vocational training, and employment experiences, (b) how these activities occurred over time, and (c) how contextual life circumstances and experiences (e.g., family history, experiences with SMHC, major life events) influenced those activities. Participants who were also parents were also asked questions about their children, their parenting experiences, how their lives changed after becoming a parent, custody and living arrangements, and challenges or supports they experience related to parenting. Interviewers were instructed to ask about a certain set of mental health related experiences if details were not offered (e.g., suicidality, inpatient hospitalizations).

Data Analysis

Audio-files or interview notes were transcribed and entered into Dedoose Qualitative Coding Software (69). A team of three coders inductively created a qualitative codebook and coded all interviews using exploratory and grounded theory approaches (70). In the initial wave of coding, the coders read through a third of the transcripts to identify and discuss major topics and themes that would form the basis of the codebook. Each member of the research team then descriptively coded some of the same transcripts independently and met for regularly scheduled sessions to compare codes and achieve consensus on the further specification of codes. The codebook was continually refined by repeating this process until inter-rater reliability was consistently attained ($k = 0.80$). The remaining transcripts were coded by the first and second author. In the final wave of coding, the first and second authors completed thematic coding and extracted and chronologically described information about each young adult parent's parenting experiences (e.g., custody status, living arrangement) and accompanying employment and education activities (e.g., start date, end date, description of the experience, why ended).

RESULTS

Participants

Of the 18 young adult participants, 83% identified as female and the majority were white, non-Hispanic (55.6%) (**Table 1**). The mean participant age at the time of the interview was 26 years old. The majority of participants were never married (55.6%) and half the sample (50%) were living independently without the live-in support of their own parents or other extended family members. Two participants were living in homeless shelters with their children and one participant was living in a group home after having been released from prison the month before. For almost half of the participants (44.4%), a high school diploma or GED was the highest level of education they had ever obtained. About a third of participants (38.9%) had attempted college but not been able to complete a degree (38.9%). Only three participants completed a post-secondary degree. Most participants (55.6%) reported making <$10,000 per year (not including benefits).

TABLE 1 | Demographics.

Variable	N	%
Gender		
Female	15	83.3
Male	3	16.7
Race/ethnicity		
Non-Hispanic white	10	55.6
Non-Hispanic black	2	11.1
Hispanic (Puerto Rican, Columbian)	3	16.7
Mixed or other race	3	16.7
Age at time of interview		
22–23	4	22.2
24–25	3	16.7
26–27	6	33.3
28–30	5	27.8
Marital status		
Never married	10	55.6
Currently married or cohabitating	5	27.8
Divorced or separated	3	16.7
Living arrangement		
Living independently	9	50.0
Living with their own parents or extended family	6	33.3
Living in homeless shelter or group home	3	16.7
Educational attainment (at time of interview)		
High school graduate or GED	8	44.4
Some college, degree not obtained	7	38.9
Associates degree	2	11.1
Master's degree	1	5.6
Annual personal income in last year (not including benefits)		
$0–$10,000	10	55.6
$10,001–$20,000	5	27.8
$20,001–$30,000	2	11.1
$30,001–$40,000	0	0.0
$40,001–$50,000	1	5.6

All participants except one reported receiving multiple mental health diagnoses. The most frequently self-reported mental health conditions were anxiety (83.3%) and major depression (72.2%), followed by PTSD (50.0%) and bipolar disorder (50.0%). Approximately 30% of participants had engaged in a suicide attempt or self-harm. Twelve participants had been hospitalized overnight due to their SMHC and of those 12, half had been hospitalized five or more times (**Table 2**).

Most participants identified as mothers but three fathers were included in the sample (**Table 3**). Some participants gave birth to their first child prior to the age of 20, thereby (40%) fitting the traditional definition of "teen parents" who had their first child before age 20. Two parents had more than two children (11.2%) and overall, children's ages (not shown) ranged from several weeks to 12 years old. The majority of participants had their children living with them the majority of the time. None of

TABLE 2 | Mental health characteristics.

Variable	N	%
Serious mental health conditions		
Anxiety	15	83.3%
Major depression	13	72.2%
Post-traumatic stress syndrome (PTSD)	9	50.0%
Bipolar disorder	9	50.0%
Schizophrenia	1	5.6%
Schizoaffective	1	5.6%
Eating disorder	5	27.8%
Borderline personality disorder	1	5.6%
Other co-occurring conditions		
Attention deficit hyperactivity disorder	3	16.7
Substance use disorder	1	5.6
Ever engaged in suicide attempt or self-harm		
Yes	6	33.3
No	12	66.7
Number of overnight psychiatric hospitalizations		
None	6	33.3
1–2	3	16.7
3–4	3	16.7
5–10	4	22.2
10+	2	11.1

TABLE 3 | Parenting characteristics.

Variable	N	%
Parental role		
Mother	15	83.3%
Father	3	16.7%
Age at which participant first became a parent		
Under 20 years old	7	38.9
20–24 years old	11	61.1
Number of Children		
1	9	50.0
2	7	38.9
3	1	5.6
4	1	5.6
Proportion of time spent living with their children		
Most of the time (>75% of the time)	15	83.3
None of the time, visitation only	1	5.6
None of the time, no visitation	2	11.1

the fathers lived with their children the majority of the time, and only one of those had visitation rights.

Qualitative Findings

Several themes emerged from the qualitative interviews specific to parenting including the strategies that young adult parents employ to manage their mental health, how children act as a motivator for recovery, and experiences of stigma and discrimination.

Managing Symptomatology of MHC While Parenting

Participants often reported that mental health symptoms impeded their ability to fully concentrate on their children and be present. One participant described how their symptoms of depression would often get in the way of their ability to engage in play with their child: "I start slacking on my kids. And like I know like he'll get restless with me, and he'll be like "mommy, you're not playing with me anymore. You're not doing anything with me." And that kind of like puts it into perspective." In this way, children also helped increase their parent's cognizance of their mental health symptoms and serve as a reminder of when and how mental health symptoms got in the way of engagement.

Some participants described how the stress of parenting can trigger an increase in their overall mental health symptoms. For one participant, parenting a child at the age in which they themselves had experienced a traumatic event triggered an increase in PTSD symptoms: "My PTSD would kick in because I would think about when I was his age, like what my parents were doing to me at that time." Another mentioned, "I feel like when he is sad or when he is sick, my anxiety is like triggered by that. It's like I try to do as much as I can to be able to help him or the situation because I feel like… I didn't have that." To cope with increased symptoms and feelings of stress, some participants would be tempted to look to negative coping skills, like drug use, to manage:

> But with him, like I get aggravated also a lot……being with him in maybe such a close range, right there in the same room, like it gets overwhelming……before when he would aggravate me, I would feel like my triggers, and I would want to use. I'd be like I just want to get high. He's just so annoying. I don't want to deal with this.

Mental health symptoms would be triggered by the stressors of parenting, and in those moments, negative coping skills were harder to avoid.

When participants tried to take care of their mental health and practice self-care, feelings of guilt often arose. Taking time out to receive treatment or to work on symptom management was difficult:

> Yeah, it's hard… being a parent. And having kids and having to take care of them. And then having to stop everything because I needed to be hospitalized. It's very… I've always kind of had a little bit of guilt. I feel like in the back of my mind, every time I get hospitalized because it's like my—wherever what I'm supposed to be doing is just going to have to stop until I come back. And that can be stressful.

Others reported that their mental health got in the way of putting their child first, and touched on the associated feelings of guilt that came with it:

> For me, it's sad, but it makes it harder for me to put my child first. And makes it harder for me to focus more on his well-being than what I want to do for myself. Which sounds really bad. It sounds really sad. But it's like if you want the honest to God truth, that's the honest to God truth. And I wish that it would be better. And I

> wish that I could put more focus on to him in making sure that he's good before anything else. But sometimes it's just like my brain won't let me do it. It's just like I come first. Which I don't want to. He's my baby.

This participant seemed to battle the overwhelming feeling that putting themselves first was selfish.

Children as Sources of Motivation and Recovery

While participants reported various obstacles to managing their mental health while parenting, the act of parenting itself was described by many as a primary motivator to initiate and maintain their mental health recovery. Participants reported that prior to becoming a parent, they were the ones most often influenced by their decisions in life. However, after having children, they needed to recognize that their behaviors and decisions, good or bad, would directly influence their children. This realization often acted as a motivator to power through the difficult times. Says one mother,

> After I had my son, it's been like a situation where I refuse to like not [to] be able to provide for my son. And then being without me and it's not just me anymore, my family, you know? Even though I have my depression and my anxiety that was like a weight on my legs. I still go forward.

In some cases, participants described their children as a positive source of distraction, providing them the ability or need to focus on something other than the negative impact of their mental health:

> And I have experienced dealing with the schizophrenia with them, and it's—most of the time they kind of are able to distract me. So, I can kind of focus on them and not worry so much about what's going on in my head.

Participants recognized the desire for a healthy distraction from their ongoing mental health symptoms, and parenting was sometimes cited as that source of refuge.

Other participants described their children as the motivator needed to embrace their recovery and persevere. Three participants shared experiences of suicidal ideation and how the need to be present for their children was strongly related to a sense of purpose, which helped provide a reprieve from acting on these thoughts. One participant in particular spoke about a shift in mindset taking place after almost being hospitalized for suicidal ideation:

> You know I kind of like came home, put my foot down, and you know kind of did what I had to do. Because honestly, I don't want to leave my kids. You know because nobody will take care of them the way that I do.

Another participant described the need to look outside of themselves to not act on their ideation, and instead look to the impact on their family:

> But when I get those thoughts, like I need to think about what is there to look forward to? And that's easier to think about when you're thinking outside of yourself, at least for me. My daughter made me, especially with her dad not being stable… I was always like… it would be selfish for me to leave because then where would that leave her.

One parent summed up their belief about the journey of parenting with a mental health condition, and offered advice and words of inspiration to other parents who should find themselves in a similar situation:

> That having a mental illness and being a parent is possible. You know, and you don't have to be like ashamed. And I've met a lot of people who have told me—who have had like bad things happen to them. They're afraid that they won't be good parents. And I mean everything depends on the person, but I mean a person who is nervous about it, most likely they notice it. And I think that shows that they don't want that to happen. And I think just because you have like PTSD or …… something. It doesn't mean that your child is going to go through that with you. Like it doesn't mean you're bad. Like it doesn't—you know, like you can still be a great parent regardless of what your mental illness is.

Despite experiencing increased challenges and negative impact on mental health, participants still described the value of parenting on their recovery and were able to balance feelings of inadequacy with reminders that experiencing a mental illness did not equate to being a lesser parent.

Experiences of Discrimination and Feelings of Stigma

Some participants described discrimination and the resulting stigma they experienced as parents with mental health conditions. Often, these experiences were related to the ways in which others perceived their ability to be a parent. One participant described an experience of facing discrimination from a judge during an ongoing custody battle with their ex-partner:

> I've definitely been discriminated against by the judge. It was horrible. They'd talk to me like I was five. They looked at me like I was a disease of a father. I wanted to be there for my kid but was never given the opportunity by the courts to do it.

This participant went on to describe how their mental health was used against them and threatened their parental rights:

> His mother and I were in a relationship for not that long a time. We were both very, very, very young. She was 16 when she gave birth, turning 17. I was 19. And everything was great up until 8 months. We were brand new parents. Like everything's very hard, very stressful. And then the anxiety and the depression kicked in with wondering can I do this? Can we do this together? And we were very unhappy together. And then stuff that I confided in with her during our relationship, when we decided to end our relationship, she used my mental health against me in court. And it ended up not turning out very well.

Another participant described the added stressor of trying to manage going to school while parenting, and the stigma they faced as a student with a so-called "behavioral problem":

> And, definitely with teachers and school, they would look at you like you were a problem. Like a behavioral problem. If you were—like if I was like crying in class, they singled you out. They would single me out. And they'd go *Oh [Name] is crying again. What's going on with you? Is there something wrong? Did I offend you*?

Discrimination against a person due to their mental health condition is common. But for many parents in this sample, the fear or experience of discrimination was even more pronounced because of their role as a parent.

DISCUSSION

This paper contributes to the current understanding of the experiences of young adult parents with SMHC and the potential impact of mental health symptom management, stigma, and discrimination on navigating a successful path to adulthood. In this sample of 18 young adults with SMHC who became parents prior to the age of 25, many described struggling to manage the symptoms associated with their SMHC and its perceived effect on their ability to engage as parents. Furthermore, mental health symptoms were often exacerbated or triggered by the stressors of parenting. Symptoms may also impede the ability to fully engage in parenting, straining the relationship between parent and child. At the same time, an overwhelming majority of young adult participants described their children as their primary sources of motivation to stay on path of mental health recovery and to "better themselves." Children were described by their parents as the primary factor for pushing through when things felt overwhelming, and elicited feelings of pride and determination.

Despite the many disclosed benefits of parenting on mental health, young adult parents also shared the negative impact of disclosing mental health to others. It was clear that many young parents with SMHC had experienced or were afraid of experiencing stigma and discrimination as a result of disclosing their mental health condition. Participants described instances of discrimination in public settings, including court rooms and educational institutions. Discrimination prevents treatment, and it impedes recovery (55, 71). Within various social services there is a prevailing blame mentality and risk discourse in which parents with mental illness are constantly monitored and often easily suspected of abuse when their symptoms are acute (72). Parents that are forced into silence will not engage in needed services if accessing that care could lead to discrimination and potential loss of custody.

Taken together, most of our findings echo previous qualitative research with parents, mostly mothers, with serious mental illness (42, 56–58, 60). However, given the research in this area is limited and the changing cultural norms of emerging or young adulthood, these findings can still uniquely inform public health and mental health services and approaches for parents with mental illness. Most people with mental illness will not only become parents but become parents at an earlier age compared to their peers (2). The earlier services are able to intervene to support parents with mental illness, the more likely they will succeed in supporting them and their families over time. Furthermore, given young adult parents in this study endorsed parenthood as meaningful and beneficial to their recovery, it is important for services to approach parent needs from a place of encouragement and motivation rather than too heavily focusing on risks or needs. The United Nations recently identified youth, defined as the period from 15 to 24 years of age, as a period of vulnerability worldwide (73). Young parents (i.e., those who have children prior to their mid-twenties) are still susceptible to experiencing the economic, social and health disadvantages that were often reserved for teen parents in previous generations. Young adults who are managing serious mental health conditions may be further susceptible to poor outcomes.

Implications

Findings from this study can help to inform public and mental health initiatives that more adequately meet the needs of young adult parents who are simultaneously managing their own mental health condition. First, participants in this study described feelings of guilt when having to put their parenting "on hold" to seek out needed mental health care. This is of no surprise, as a majority of young adult parents are single parents, without a co-parent to balance the weight of added responsibilities that parenting carries. Allocating transportation and daycare funds for young adult parents with SMHC could provide a platform for increasing access to care, as well as alleviating the feeling of burden placed on young adult parents to "do it all" without adequate support.

Young adult Access Centers or Drop-In Centers are an increasingly popular model in the United States aimed at supporting the mental health and overall career development of young adults with SMHC. Access Centers provide a non-judgmental safe space where young people can meet with peers and community-based mental health professionals to access mental health care, get basic needs met (e.g., laundry, showering, food) and focus on career development (e.g., resume building, GED practice) (74). Building upon the successful Young-adult Access Centers model, additional programming within Access Centers should be tailored to young adult parents with SMHC. Expanding programming to meet the needs of young parents would offer a safe environment where parents could bring their children and engage in peer support and parenting education and develop concrete tools to continue to foster a wellness recovery plan that emphasizes the parent-child relationship. Access Centers are designed to have flexible/evening hours and are structured in a way that lends well to providing childcare services. This is ideal for young parents who are often balancing parenting, employment, and education simultaneously and do not have the luxury of accessing needed care during traditional office hours. The Access Center approach has already begun to draw interest from young adult parents with SMHC; a recent analysis showed that within a Massachusetts Young Adult Access Center, young parents represented 10–15% of all young adults served (74).

Services aimed at supporting young adult parents with SMHC should attempt to strengthen the relationship between parent and child and be cognizant of the positive role that parenting can have in the recovery process. Many public health initiatives aimed at teen parenting in earlier generations focused primarily on the negative outcomes and risks associated with teen parenting. Similarly, parenting with a mental health condition is often seen as an impediment or liability to both the parent and the child. However, it is clear from this data that parenting may foster resilience in young adults with SMHC and can be capitalized on as a positive source of motivation and a tool to maintain recovery. Peer support services, tailored for young adult parents with SMHC, may be part of the solution. Peers offer special knowledge, drawn from personal experience, as a unique resource to help navigate the very practical day-to-day challenges that parents encounter when raising children and navigating treatment and recovery (75). Providers, policy makers, and practitioners need to recognize how young adult parents with SMHC are motivated by their children and capitalize on that motivation.

While many mental health services prioritize the medical model of recovery (i.e., the reduction or management of symptoms), a broader more personalized recovery model would be more supportive of parents with SMHC, especially those younger and earlier in their mental health recovery journey. Personal recovery has been defined as recovery that aims to emphasize the ability to lead a meaningful, purposeful life, with or without ongoing episodes of mental illness (76). One brief family support intervention in Australia, Let's Talk About Children, aims to enhance the recovery journey of parents with mental illness by acknowledging and addressing the parenting life domain (77). Research on the effectiveness of this intervention is underway, but the model's approach to parenting as a "value-add" in one's personal recovery journey is a promising practice in light of our current findings. Nicholson et al. (78) have adapted Let's Talk into the ParentingWell Practice Profile to support mental health practitioners in implementing family-focused practice approach with adults with mental illness. An initial pilot of the ParentingWell Learning Collaborative in Massachusetts provided preliminary support for the feasibility and impact of the model (79). Future research and evaluation efforts should also explore necessary modifications of this recovery intervention to fit the unique needs of young adult parents with SMHC.

Additionally, stigma and fear of discrimination are real and can impede young adult parents' willingness and ability to seek adequate care for themselves or their children. An ongoing yet silent threat of removal of their children prevents parents from being forthcoming about their needs or stressors, which in turn negatively impacts their mental health, their parenting, and their children (42). The general public and professionals within social services are in need of training and education to dispel myths about living with a mental health condition and its impact on one's ability to parent. Research tells us that when people share their stories of recovery with the public, it encourages people to challenge their negative beliefs and assumptions about mental health (55). Providing increased education on mental health cultural competency to professionals in direct family support positions (e.g., court officials, departments of children and families) could alleviate discrimination in care settings where young adult parents may fear disclosure.

Finally, providing increased access to comprehensive and confidential parental mental health screenings in community-based parent service settings (e.g., parenting classes and support groups) and general healthcare settings could help to close the gap in reaching parents who are fearful of being stigmatized when approaching mental health services directly. Young adults with SMHC have some of the lowest mental health help-seeking rates compared to other age groups and often cite stigma and embarrassment as primary barriers to help-seeking (80). For young adult parents with SMHC, the additional fear of custody loss may exacerbate the desire to steer clear from formal services, and in turn, enhance the desire to seek support from more informal peer services. However, despite grappling with these systemic and environmental barriers to psychological care, many young adults access primary care on an annual basis (81). Increasing training and funding to general health care spaces already accessed by this population would contribute to increased public health services coverage, while supporting and honoring the comfort and safety of young adults with SMHC.

Limitations

Limitations of this study include a lack of racial diversity in the sample. However, the sample's racial composition is not that different from that of Worcester County, MA where the majority of the sample resided (82). A lack of diversity limits generalizability as does a small sample size. The sample for this study was pulled from a larger study sample ($n = 61$). While we successfully oversampled to ensure about 1/3 of the full sample were young parents, given the diversity of pathways through young adulthood, these findings may not fully represent the full range of experiences of young adults with SMHC who are parents. The experiences of young adult parents and their children are highly contextualized and can vary on multiple characteristics including race, social class, and severity of mental health condition. However, these findings should inform a larger prospective study that will better understand the experiences of this population. Finally, only three of the participants were fathers. The experiences of fathers with mental illness have been largely missing from the literature and while this represents a small contribution, more research is needed to understand fathers with mental illness of all ages, especially those who are young and may not have full custody of their children.

CONCLUSION

Given increased recognition of a period of "emerging adulthood," young people who become parents in young adulthood (18–24 years old) and their children may be vulnerable to poor outcomes like those of teen parents (13–19 years old) from previous generations. Particularly at risk are young adults with serious mental health conditions (SMHC) who often have poorer education, employment, and housing outcomes and tend to parent earlier than their peers. Taken together, by virtue of their age and their mental health condition, young adults with SMHC, and their children, may be at higher risk for poor outcomes.

This was the first qualitative study to explore the experiences of young adults with SMHC who live in the United States and became parents prior to the age of 25. These young adult parents described the challenges of simultaneously parenting young children and managing a mental health condition, experiences of discrimination, and fear of future discrimination related to their mental health condition. Like prior qualitative research with mothers with mental illness, these parents also regarded their children as motivators for their recovery and important elements of their overall personal recovery. However, by exclusively focusing on individuals who become parents earlier than their peers, and including father experiences, this research adds to our understanding of how individuals simultaneously navigate parenting and managing their own mental health. These findings are relevant to public mental health services. Mental health services should be offered in low-barrier settings with convenient hours for young parents. Programming should foster resilience in young parents and incorporate their role of parenting as an asset in their personal recovery journey. The continued threat of discrimination due to mental health stigma is particularly poignant for parents and will continue to negatively influence their help-seeking habits if left unaddressed.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of Massachusetts Medical School Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

KS is the principal investigator and lead author. AB contributed substantially in the writing the manuscript. LG and EP-D'O participated in data collection, analysis and manuscript preparation. IL and MO'N assisted in the writing and editing of the manuscript. All authors contributed to the article and approved the submitted version.

FUNDING

This study was funded through a center grant to the University of Massachusetts Chan Medical School from the National Institute on Disability, Independent Living, and Rehabilitation Research (NIDILRR). NIDILRR is a Center within the Administration for Community Living (ACL), United States Department of Health and Human Services (HHS) (ACL grant# 90RT5031, The Learning and Working Transitions RRTC: PI, Maryann Davis, PhD).

REFERENCES

1. Luciano A, Nicholson J, Meara E. The economic status of parents with serious mental illness in the United States. *Psychiatr Rehabil J.* (2014) 37:242. doi: 10.1037/prj0000087
2. Nicholson J. For parents with mental health conditions: the chance to "have it all" at work and at home. *Psychiatr Rehabil J.* (2014) 37:153. doi: 10.1037/prj0000057
3. Centers for Disease Control and Prevention (CDC). Births: final data for 2017. *Natl Vital Stat Rep*. (2018) 67:1–50. Available online at: https://www.cdc.gov/nchs/data/nvsr/nvsr67/nvsr67_08-508.pdf
4. Hoffman SD, Maynard RA. (eds.). (2008). *Kids Having Kids: Economic Costs & Social Consequences of Teen Pregnancy, 2nd Edn*. Washington, DC: The Urban Institute Press.
5. Shuger L. *Teen Pregnancy and High School Dropout: What Communities Can Do to Address These Issues*. Washington, DC: The National Campaign to Prevent Teen and Unplanned Pregnancy and America's Promise Alliance (2012). Available online at: https://powertodecide.org/what-we-do/information/resource-library/teen-pregnancy-and-high-school-dropout (accessed February 18, 2022).
6. Martin JA, Hamilton BE, Sutton PD, Ventura SJ, Mathews TJ, Osterman MJK. Births: final data for 2008. *Natl Vital Stat Rep.* (2010) 59:1–102. Available online at: https://stacks.cdc.gov/view/cdc/23430
7. Mathews TJ, MacDorman MF. Infant mortality statistics from the 2006 period linked birth/infant death data set. *Natl Vital Stat Rep.* (2010) 58:1–31. Available online at: https://www.cdc.gov/nchs/data/nvsr/nvsr58/nvsr58_17.pdf
8. Bolton FG. The risk of child maltreatment in adolescent parenting. *Adv Adolesc Ment Health.* (1990) 4:223–37.
9. Easterbrooks MA, Jacobs FH, Bartlett JD, Goldberg J, Contreras MM, Kotake C, et al. *Initial Findings From a Randomized, Controlled Trial of Healthy Families Massachusetts: Early Program Impacts on Young Mothers' Parenting*. Washington, DC: Pew Charitable Trusts (2012).
10. Lipman EL, Georgiades K, Boyle MH. Young adult outcomes of children born to teen mothers: effects of being born during their teen or later years. *J Am Acad Child Adolesc Psychiatry.* (2011) 50:232–41. doi: 10.1016/j.jaac.2010.12.007
11. Stiles AS. Case study of an intervention to enhance maternal sensitivity in adolescent mothers. *J Obstetr Gynecol Neonatal Nurs.* (2010) 39:723–33. doi: 10.1111/j.1552-6909.2010.01183.x
12. Kost K, Henshaw S. *US Teenage Pregnancies, Births, and Abortions, 2010: National and State Trends by Age, Race, and Ethnicity*. New York, NY: Guttmacher Institute (2014). Available online at: http://www.guttmacher.org/pubs/USTPtrends10.pdf (accessed February 17, 2022).
13. The Urban Child Institute. (2011). *The National Campaign to Prevent Teen and Unplanned Pregnancy*. Available online at: http://www.urbanchildinstitute.org/articles/updates/the-national-campaign-to-prevent-teen-and-unplanned-pregnancy
14. Feldman Farb A, Margolis AL. The teen pregnancy prevention program (2010-2015): synthesis of impact findings. *Am J Public Health.* (2016). 106:S9–15. doi: 10.2105/AJPH.2016.303367
15. U.S. Department of Health and Human Services. *Trends in Teen Pregnancy and Childbearing*. (2019). Available online at: https://opa.hhs.gov/adolescent-health/reproductive-health-and-teenpregnancy/trends-teen-pregnancy-and-childbearing (February 17, 2022).
16. Arnett JJ, Tanner JL. (eds.). *Emerging Adults in America: Coming of Age in the 21st Century*. Washington, DC: American Psychological Association. (2006). p. 3. doi: 10.1093/acprof:oso/9780195309379.001.0001
17. De Brey C, Snyder TD, Zhang A, Dillow SA. *Digest of Education Statistics 2019 (NCES 2021-009)*. Washington, DC: National Center for Education Statistics, Institute of Education Sciences, U.S. Department of Education (2021).

18. Fry R, Passel JS, Cohn D. *A Majority of Young Adults in the U.S. Live With Their Parents for the First Time Since the Great Depression*. Washington, DC: Pew Research Center (2020). Available online at: https://www.pewresearch.org/fact-tank/2020/09/04/a-majority-of-young-adults-in-the-u-s-live-with-their-parents-for-the-first-time-since-the-great-depression/ (accessed February 17, 2022).
19. Centers for Disease Control and Prevention (CDC). Mean age of mother, 1970-2000. *Natl Vital Stat Rep*. (2002) 51:1—13. Available online at: https://www.cdc.gov/nchs/data/nvsr/nvsr51/nvsr51_01.pdf
20. Livingston, G. (2018). *They're Waiting Longer, But U.S. Women Today More Likely to Have Children Than a Decade Ago*. Washington, DC: Pew Research Center. Available online at: https://www.pewresearch.org/social-trends/2018/01/18/theyre-waiting-longer-but-u-s-women-today-more-likely-to-have-children-than-a-decade-ago/ (accessed February 17, 2022).
21. Martin JA, Hamilton BE, Osterman M, Driscoll AK. Births: final data for 2019. *Natl Vital Stat Rep*. (2021) 70:1–51.
22. Khandwala YS, Zhang CA, Lu Y, Eisenberg ML. The age of fathers in the USA is rising: an analysis of 168 867 480 births from 1972 to 2015. *Hum Reprod (Oxford, England)*. (2017) 32:2110–6. doi: 10.1093/humrep/dex267
23. Schweizer VJ. *30 Years of Change in Men's Entry into Fatherhood, 1987-2017. Family Profiles, FP-19-28*. Bowling Green, OH: National Center for Family & Marriage Research (2019).
24. Raymo JM, Carlson MJ, VanOrman A, Lim SJ, Perelli-Harris B, Iwasawa M. Educational differences in early childbearing: a cross-national comparative study. *Demogr Res*. (2015) 33:65–92. doi: 10.4054/DemRes.2015.33.3
25. Martin JA, Hamilton BE, Osterman M, Driscoll AK. Births: final data for 2018. *Natl Vital Stat Rep*. (2019) 68:1–47. Available online at: https://www.cdc.gov/nchs/data/nvsr/nvsr68/nvsr68_13-508.pdf
26. National Academy of Medicine (formerly Institute of Medicine) and National Research Council. *Investing in the Health and Well-Being of Young Adults*. Washington, DC: The National Academies Press (2015).
27. Kessler RC, Berglund P, Demler O, Jin R, Merikangas KR, Walters EE. Lifetime prevalence and age-of-onset distributions of DSM-IV disorders in the National Comorbidity Survey Replication. *Arch Gen Psychiatry*. (2005) 62:593–602. doi: 10.1001/archpsyc.62.6.593
28. Substance Abuse and Mental Health Services Administration (SAMHSA). *Key Substance Use and Mental Health Indicators in the United States: Results From the 2020 National Survey on Drug Use and Health (HHS Publication No. PEP21-07-01-003, NSDUH Series H-56)*. Rockville, MD: Center for Behavioral Health Statistics and Quality, Substance Abuse and Mental Health Services Administration (2021). Available online at: https://www.samhsa.gov/data/
29. U.S. Census Bureau. *Data Table: American Community Survey, 1-yr Estimates, Table S-101 Age and Sex Estimates*. (2022). Available online at: https://data.census.gov/cedsci/ (accessed on January 20, 2022).
30. U.S. Department of Education, Office of Special Education and Rehabilitative Science, and Office of Special Education Programs. *39th Annual Report to Congress on the Implementation of the Individuals With Disabilities Education Act, 2017*. (2017). Available online at: https://sites.ed.gov/idea/2017-annual-report-to-congress-on-the-individuals-with-disabilities-education-act/ (accessed February 17, 2022).
31. Newman L, Wagner M, Knokey AM, Marder C, Nagle K, Shaver D, et al. *The Post-High School Outcomes of Young Adults with Disabilities up to 8 Years After High School. A Report From the National Longitudinal Transition Study-2 (NLTS2) (NCSER 2011-3005)*. Menlo Park, CA: SRI International (2011).
32. Salzer MS, Wick LC, Rogers JA. Familiarity with and use of accommodations and supports among postsecondary students with mental illnesses. *Psychiatr Serv*. (2008) 59:370–5. doi: 10.1176/ps.2008.59.4.370
33. Frank AR. Transition of adolescents with behavioral disorders–is it successful? *Behav Disord*. (1991) 16:180–91. doi: 10.1177/019874299101600303
34. Neel RS, Meadows N, Levine P, Edgar EB. What happens after special education: a statewide follow-up study of secondary students who have behavioral disorders. *Behav Disord*. (1988) 13:209–16. doi: 10.1177/019874298801300306
35. Vander Stoep A, Beresford SAA, Weiss NS, McKnight B, Cauce AM, Cohen P. Community-based study of the transition to adulthood for adolescents with psychiatric disorder. *Am J Epidemiol*. (2000) 152:352–62. doi: 10.1093/aje/152.4.352
36. Substance Abuse and Mental Health Services Administration (SAMHSA). *Results From the 2013 National Survey on Drug Use and Health: Summary of National Findings, NSDUH Series H-48, HHS Publication No. (SMA) 14-4863*. Rockville, MD: Substance Abuse and Mental Health Services Administration (2014).
37. Castro AL, Gustafson EL, Ford AE, Edidin JP, Smith DL, Hunter SJ, et al. Psychiatric disorders, high-risk behaviors, and chronicity of episodes among predominantly African American homeless Chicago youth. *J Health Care Poor Underserv*. (2014) 25:1201. doi: 10.1353/hpu.2014.0124
38. Stykes J. *Fatherhood in the US: Men's Age at First Birth, 1987-2010 (FP-11-04)*. Bowling Green, OH: National Center for Family & Marriage Research (2011). Available online at: http://www.bgsu.edu/content/dam/BGSU/college-of-arts-and-sciences/NCFMR/documents/FP/FP-11-04.pdf (accessed February 17, 2022).
39. U.S. Government Accountability Office. *Young Adults With Serious Mental Illness: Some States and Federal Agencies Are Taking Steps to Address Their Transition Challenges (GAO-08-678)*. Washington, DC: United States Government Accountability Office (2008).
40. Guzzo KB, Payne KK. A*verage Age at First Birth, 1970 - 2017. Family Profiles, FP-18-25*. Bowling Green, OH: National Center for Family & Marriage Research (2018). doi: 10.25035/ncfmr/fp-18-25
41. Henderson C, Evans-Lacko S, Thornicroft G. Mental illness stigma, help seeking, and public health programs. *Am J Public Health*. (2013) 103:777–80. doi: 10.2105/AJPH.2012.301056
42. Boursnell M. The silent parent: developing knowledge about the experiences of parents with mental illness. *Child Care Pract*. (2007) 13:251–60. doi: 10.1080/13575270701353630
43. Oyserman D, Mowbray CT, Meares PA, Firminger BA. Parenting among mothers with a serious mental illness. *Am J Orthopsychiatry*. (2000) 70:297–315. doi: 10.1037/h0087733
44. Duncan S, Reder P. Children's experience of major psychiatric disorder in their parent: an overview. In: Reder P, McClure M, and Jolley A, editors, *Family Matters: Interfaces Between Child and Adult Mental Health*. London: Routledge (2000). p. 83–95.
45. Hammen C. Generalization of stress in the course of unipolar depression. *J Abnorm Psychol*. (1991) 100:555–61. doi: 10.1037/0021-843X.100.4.555
46. O'Donnell M, Maclean MJ, Sims S, Morgan VA, Leonard H, Stanley FJ. Maternal mental health and risk of child protection involvement: Mental health diagnoses associated with increased risk. *J Epidemiol Commun Health*. (2015) 69:1175–83. doi: 10.1136/jech-2014-205240
47. Nomura Y, Wickramaratne PJ, Warner V, Mufson L, Weissman MM. Family discord, parental depression and psychopathology in offspring: 10-year follow-up. *J Am Acad Child Adolesc Psychiatry*. (2002) 41:402–9. doi: 10.1097/00004583-200204000-00012
48. Burt KB, Van Dulmen MHM, Carlivati J, Egeland B, Sroufe LA, Forman DR, et al. Mediating links between maternal depression and offspring psychopathology: The importance of independent data. *J Child Psychol Psychiatry*. (2005) 46:490–9. doi: 10.1111/j.1469-7610.2004.00367.x
49. Farrell MP, Barnes GM, Banerjee S. Family cohesion as a buffer against the effects of problem-drinking fathers on psychological distress, deviant behavior, and heavy drinking in adolescents. *J Health Soc Behav*. (1995) 36:377–85. doi: 10.2307/2137326
50. Lannes A, Bui E, Arnaud C, Raynaud J-P, Revet A. Preventive interventions in offspring of parents with mental illness: A systematic review and meta-analysis of randomized controlled trials. *Psychol Med*. (2021) 51:2321–36. doi: 10.1017/S0033291721003366
51. Beidel DC, Turner SM. At risk for anxiety: I. Psychopathology in the offspring of anxious parents. *J Am Acad Child Adolesc Psychiatry*. (1997) 36:918–24. doi: 10.1097/00004583-199707000-00013
52. Weissman MM, Wickramaratne P, Nomura Y, Warner V, Pilowsky D, Verdeli H. Offspring of depressed parents: 20 years later. *Am J Psychiatry*. (2006) 163:1001–8. doi: 10.1176/ajp.2006.163.6.1001
53. Merikangas KR, Dierker LC, Szatmarie P. Psychopathology among offspring of parents with substance abuse and/or anxiety disorder: a high-risk study. *J Child Psychol Psychiatry*. (1998) 39:711–20. doi: 10.1017/S0021963098002522
54. Rasic D, Haljek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder, and major depressive disorder:

a meta-analysis of family high-risk studies. *Schizophrenia Bull.* (2014) 40:28–38. doi: 10.1093/schbul/sbt114
55. Corrigan P. Beat the stigma: come out of the closet. *Psychiatr Serv.* (2003) 54:1313. doi: 10.1176/appi.ps.54.10.1313
56. Nicholson J, Sweeney EM, Geller JL. Focus on women: Mothers with mental illness: I. The competing demands of parenting and living with mental illness. *Psychiatr Serv.* (1998) 49:635–42. doi: 10.1176/ps.49.5.635
57. Diaz-Caneja A, Johnson S. The views and experiences of severely mentally ill mothers A qualitative study. *Soc Psychiatry Psychiatr Epidemiol.* (2004) 39:472–82. doi: 10.1007/s00127-004-0772-2
58. Dolman C, Jones I, Howard LM. Pre-conception to parenting: A systematic review and meta-synthesis of the qualitative literature on motherhood for women with severe mental illness. *Arch Women's Ment Health.* (2013) 16:173–96. doi: 10.1007/s00737-013-0336-0
59. Ackerson BJ. Coping with the dual demands of severe mental illness and parenting: the parents' perspective. *Fam Soc.* (2003) 84:109–18. doi: 10.1606/1044-3894.69
60. Reupert A, Maybery D. Families affected by parental mental illness: a multiperspective account of issues and interventions. *Am J Orthopsychiatry.* (2007) 77:362–9. doi: 10.1037/0002-9432.77.3.362
61. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I the scientific basis to a comprehensive approach. *Aust J Adv Ment Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
62. Nicholson J, Biebel K, Hinden B, HenryA, Stier L. *Critical Issues for Parents With Mental Illness and Their Families*. Rockville, MD: Center for Mental Health Services/Substance Abuse and Mental Health Services Administration (2001). https://www.umassmed.edu/globalassets/center-for-mental-health-services-research/documents/products-publications/reports/families/critical_isues.pdf doi: 10.1037/e303792004-001
63. Foster K. Advancing the prevention of intergenerational mental illness: Where are we now, where do we go from here? *Adv Ment Health.* (2015) 13:97–9.
64. Siegenthaler E, Munder T, Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17. doi: 10.1016/j.jaac.2011.10.018
65. Nicholson J, de Girolamo G, Schrank B. Editorial: parents with mental and/or substance use disorders and their children. *Front Psychiatr.* (2019) 10:915. doi: 10.3389/fpsyt.2019.00915
66. Israel BA, Schulz AJ, Parker EA, Becker AB. Review of community-based research: assessing partnership approaches to improve public health. *Annu Rev Public Health.* (1998) 19:173–202. doi: 10.1146/annurev.publhealth.19.1.173
67. Wallerstein N, Duran B. The Conceptual, Historical, and Practice Roots of Community Based Participatory Research and Related Participatory Traditions. In: Minkler M, Wallerstein N, editors. *Community-based Participatory Research for Health*. San Francisco, CA: Jossey-Bass (2002). p. 27–52.
68. Atkinson R. *The Life Story Interview as a Bridge in Narrative Inquiry. Handbook of Narrative Inquiry: Mapping a Methodology.* Thousand Oaks, CA: Sage Publications, Inc (2007). p. 224–245. doi: 10.4135/9781452226552.n9
69. Dedoose. *Version 8.0.35, web application for managing, analyzing, and presenting qualitative and mixed method research data (2017)*. Los Angeles, CA: Socio Cultural Research Consultants, LLC (2017). Available online at: www.dedoose.com (accessed February 17, 2022).
70. Strauss A, Corbin J. *Basics of Qualitative Research Techniques*. Thousand Oaks, CA: Sage Publications (1998).
71. Hyman I. *Self-Disclosure and Its Impact on Individuals Who Receive Mental Health Services*. Rockville, MD: US Department of Health and Human Services. Substance Abuse and Mental Health Services Administration, Center for Mental Health Services (2008).
72. Krumm S, Becker T. Subjective views of motherhood in women with mental illness–a sociological perspective. *J Ment Health*. (2006) 15:449–60.
73. United Nations Educational, Scientific and Cultural Organization. *Youth Matters: Equipping Vulnerable Young People With Literacy and Life Skills. UNESCO Institute for Lifelong Learning Policy Brief 2.* (2013). Available online at: http://unesdoc.unesco.org/images/0022/002230/223022e.pdf (accessed February 17, 2022).
74. Henry AD, Fishman J, Pratt C. *Service Enhanced Access Centers for Young Adults in Massachusetts*. Worcester, MA: University of Massachusetts Medical School, Commonwealth Medicine (2020).
75. Nicholson J, Valentine A. Key informants specify core elements of peer supports for parents with serious mental illness. *Front Psychiatry.* (2019) 10:106. doi: 10.3389/fpsyt.2019.00106
76. Burgess P, Pirkis J, Coombs T, Rosen A. Assessing the value of existing recovery measures for routine use in Australian mental health services. *Aust N Z J Psychiatry.* (2011) 45:267–80. doi: 10.3109/00048674.2010.549996
77. Maybery D, Goodyear M, Reupert A, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry.* (2017) 17:198. doi: 10.1186/s12888-017-1357-4
78. Nicholson J, English K. *ParentingWell Practice Profile*. Waltham, MA: National Research Center for Parents with Disabilities, Brandeis University and the Massachusetts Department of Mental Health. (2019). Available online at: https://heller.brandeis.edu/parents-with-disabilities/parenting-well/index.html (accessed February 17, 2022).
79. Nicholson J, English K, Heyman M. The ParentingWell Learning collaborative feasibility study: training adult mental health service practitioners in a family-focused practice approach. *Commun Ment Health J.* (2021) 58:261—76. doi: 10.1007/s10597-021-00818-5
80. Gulliver A, Griffiths KM, Christensen H. Perceived barriers and facilitators to mental health help-seeking in young people: a systematic review. *BMC Psychiatry.* (2010) 10:1–9. doi: 10.1186/1471-244X-10-113
81. Mauerhofer A, Berchtold A, Michaud PA, Suris JC. GPs' role in the detection of psychological problems of young people: a population-based study. *Br J Gen Pract.* (2009) 59:e308–14. doi: 10.3399/bjgp09X454115
82. U.S Census Bureau. *"United States Census Bureau QuickFacts Worcester County, Massachusetts"*. (2019). Available online at: https://www.census.gov/quickfacts/fact/table/worcestercountymassachusetts (accessed February 17, 2022).

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

The handling editor JN declared a shared affiliation, though no other collaboration, with one of the authors KS, EP-D'O, and IL at the time of the review.

frontiers in Psychiatry

ORIGINAL RESEARCH
published: 18 March 2022
doi: 10.3389/fpsyt.2022.807251

Mandatory Check for COPMI in Adult Mental Healthcare Services in the Netherlands—A Quantitative and Qualitative Evaluation

Saskia Everts[1]*, *Therese van Amelsvoort*[1,2] *and Sophie Leijdesdorff*[2]

[1] *Division Child and Adolescent Mental Health, Mondriaan Mental Health Center, Maastricht, Netherlands,* [2] *Department of Psychiatry and Neuropsychology, Maastricht University, Maastricht, Netherlands*

Background: Children of parents with a mental disorder and/or addiction (COPMI) are at increased risk of developing a mental disorder. In spite of preventive interventions that can decrease the risk of problem development, COPMI are not automatically offered help. In 2013, a mandatory COPMI check was implemented in the Netherlands, requiring every mental health care professional to check whether their adult patients have children and to assess these children's safety and needs. Earlier research has shown that a gap between these regulations and the actual integration in clinical practice is not uncommon.

Method: In the current study, we evaluated the implementation of the mandatory COPMI check in the Netherlands, using quantitative as well as qualitative data from a large mental healthcare organization in the Netherlands that offers both Child and Adolescent Mental Health and Adult Mental Healthcare.

Results: Files from 14,469 patients were analyzed quantitatively and a sample of 150 files was further analyzed in depth. Findings were refined through 4 focus groups with adult mental healthcare professionals. It was found that while there are examples of the tool leading to interventions for COPMI, the tool is often not used, and when used tends to direct the focus away from COPMI needs and organizing help toward the more narrow and problematic focus on safety and reporting to child abuse authorities.

Conclusion: The potential of the COPMI check is currently not fully realized. Strategies to improve its effectiveness in clinical practice are needed to improve access to interventions for COPMI.

Keywords: COPMI, children, parental mental illness, mental disorders, intervention, mandatory check, prevention

Edited by:
Joanne Louise Riebschleger,
Michigan State University,
United States

Reviewed by:
Darpan Kaur,
Mahatma Gandhi Missions Medical College and Hospital, India
Andrea Reupert,
Monash University, Australia
Rebecca Kate Allchin,
Eastern Health, Australia

Correspondence:
Saskia Everts
s.everts@mondriaan.eu

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *01 November 2021*
Accepted: *07 February 2022*
Published: *18 March 2022*

Citation:
Everts S, van Amelsvoort T and Leijdesdorff S (2022) Mandatory Check for COPMI in Adult Mental Healthcare Services in the Netherlands—A Quantitative and Qualitative Evaluation. Front. Psychiatry 13:807251. doi: 10.3389/fpsyt.2022.807251

INTRODUCTION

It has been well documented that children of parents with a mental disorder and/or addiction (COPMI) are at considerable risk to develop mental disorders themselves (1–3). Accounting for this risk are both hereditary factors, as well as a potential inadequate developmental context (including child-abuse) that can arise when parents suffer from mental disorders or addiction. Moreover, congenital factors within the child and contextual factors in the family, influence each other as well (4).

Of course, not all children whose parents struggle with mental illness will develop mental problems themselves, nor will their development necessarily be problematic. There is quite some important literature on the resilience of children and on protective factors that can counter the risk of having a vulnerable parent (5, 6). Also, several Child and Adolescent Mental Health interventions exist to increase resilience of children and mitigate potentially negative developmental effects among COPMI (7, 8). These include programs directly targeting both children and parent(s) regarding their parenting tasks. Many of these interventions are found to be evidence-based (9–11) and/or are experienced by professionals and children or their parents as helpful (12, 13). Moreover, it has been established that early and preventive interventions can decrease the risk of problem development in COPMI with 40% (14, 15). One of the key questions is how these available interventions can be brought timely to those needing it. As COPMI are at greater risk, approaching them through their mentally ill caregivers (usually the parents) could provide an entry point for (early) detection and intervention. Therefore, organizations providing mental healthcare to parents can play an important role in the identification of those children at risk, enabling prevention and (early) intervention through specific programs for COPMI (16). This role has however not typically been taken up spontaneously by mental healthcare organizations or individual professionals and appears not easy to fulfill (17–20), partially due to different ways of working between child and adolescent mental health services (CAHMS) and adult mental health services (AMHS). Therefore professionals working with adults with mental disorders should be encouraged to play a role in enabling COPMI to access help. In a few countries, notably Norway and the Netherlands, as well as in the state of Victoria in Australia (21), a top-down approach has been chosen aimed at a routine identification of COPMI in adult mental health services: attention to COPMI has been required by law and tools are introduced that should be used routinely. The Norwegian COPMI project has been evaluated at different stages, showing that in the first 3 years after implementation, significantly more children were identified as being at risk, yet follow-up in terms of support for these children did not significantly increase (18). A follow up study found that after 5 years there was little increase in experience, attitude and knowledge or experience with family conversations among adult health care workers about COPMI (22). Similar limited results were found in Australia (21).

In the Netherlands, both the obligation to ask about the children of adult patients receiving mental health care (MHC) as well as an instrument to facilitate this mandatory check (*Kindcheck* or COPMI check), were introduced in 2013 (23). The present study (1) evaluates the implementation of this COPMI tool, (2) explores whether this has resulted in increased support for COPMI and (3) identifies potential strengths and barriers.

COPMI Check

The Dutch COPMI check was originally developed, and is still presented as part of a nationally implemented protocol aimed at reducing the incidence of child abuse and domestic violence (24, 25). This so called Reporting Protocol offers a five-step decision tree, detailing the best course of action in case of suspected child abuse or domestic violence. The COPMI check is presented as part of the first step, which is to document the warning signals that support or contradict such a suspicion. The COPMI check focuses on the "parental warning signals" that may indicate risk for child abuse, which include the (mental) health issues of the parent. The tool is meant to be used by professionals working in adult health care.

It is of note that the COPMI check was introduced with a focus on child *abuse*, while the present study is concerned with the broader issue of *mental health needs* and well-being of children and adolescents at risk. Child abuse or safety can be seen as one extreme of a continuum, with general mental and developmental needs of children at the other end. Of course the distinction between safety and mental health needs is gradual and the two foci overlap. For example, the broader issue of emotional neglect is often included in the official definitions of child abuse violence[1] (26). But although there is overlap, there is still a clear difference between the two ends of the continuum, with the COPMI check focused on the safety end. Despite this differing focus, it would seem reasonable to expect and hope that the COPMI check, as the only mandatory and widely implemented tool addressing COPMI, would contribute to an increased support for COPMI both regarding abuse and regarding broader mental health needs. An evaluation of the Dutch COPMI check has not been done from this perspective before, although reference to problems with its use are made in some Dutch studies (27–29). Thus, the present study evaluated the implementation, use and outcome of the COPMI check at Mondriaan Mental Health Center, a large mental healthcare organization in the South of the Netherlands, that incorporated the COPMI check in 2016 as mandatory tool in their standard intake assessment procedure.

MATERIALS AND METHODS

Patient Files

Subjects

Data were collected prospectively for a period of 4 years; between December 2016 (start of the implementation) and January 2021. To include a group of patients with a reasonable chance of having the responsibility over underage children, patients outside the age range of 20–65 years were excluded. As a result, completed COPMI checks of 14.469 patients were retrieved from electronic patient files. This data set was anonymized. The study protocol and procedure were assessed as non-invasive and approved by the medical ethical committee of Maastricht University (protocol number: 2021-2784).

Measures

The COPMI check was operationalized at Mondriaan Mental Health Center as a brief tool, existing of one question with three answering possibilities, integrated in the standard intake formats. The quantitative data of this study are the answers to this COPMI check question.

---

[1] https://www.rijksoverheid.nl/onderwerpen/kindermishandeling/wat-kindermishandeling-is

COPMI check question: Has the COPMI check been carried out?

1. Yes, no risk present
2. Yes, risk present (if so: put relevant information in COPMI-check form and record interventions in treatment plan)
3. No, not carried out (with follow-up question: Why not?)

If none of the boxes was ticked, we consider this category 0, described as "No answer given".

Procedure and Analysis

For each of the first two categories (responses 1 and 2), a random sample of 75 patient files was taken, using Excel, version 16.50 (Microsoft, Redmond, WA, USA). These patient files were further investigated to (1) extract in-depth information about how professionals came to their answers to the COPMI check question, and (2) whether they initiated further steps such as organizing or providing some form of help. For each file, a summary of the available information on the children, the home-situation, the COPMI check and related considerations was written. Following thematic analysis, these summaries were categorized into different themes relevant to understanding the decisions made by the professionals.

Focus Groups

Setting

To help interpret and enrich the results from the analysis of patient files, four focus groups were conducted. We made use of the regular team meetings of AMHS professionals, which we joined for 30–90 min in order to conduct a focus group discussion regarding our research questions. Participants were contacted in advance with comprehensive study information. Withdrawal from participation of the study was possible at any stage in the process which was repeatedly stated by the researchers. Oral informed consent was obtained. Focus groups were recorded with an audio-recorder and later transcribed to text in their original language. Any relevant notes made during the focus group were included in the transcript as well. At the end of each focus group, the researchers did a "member check" (30) by summarizing the discussion and asking participants to either adjust, add to or approve this summary.

Participants

To gain insight into the full scope of barriers and facilitators, we joined the meetings of different AMHS teams thus using a form of purposeful sampling (31). The following meetings were joined and used for a focus group session:

1. A routine multidisciplinary meeting from the team working on Anxiety, Compulsion, and Trauma.
2. A routine multidisciplinary meeting from the team working with Attention Deficit Hyperactivity Disorder and Autism Spectrum Disorders.
3. A routine meeting to discuss possible crises, in which AMHS professionals working with Anxiety disorders, Psycho trauma, and Personality Disorders participated.
4. A quarterly meeting of AMHS professionals that function as "internal ambassadors" for the implementation of the Reporting Protocol Domestic Violence and Child Abuse. These internal ambassadors are assigned to stimulate the use of the COPMI check and broader protocol of the Reporting Protocol Domestic Violence and Child Abuse.

By joining routine meetings, we were able to reach more AMHS professionals, as well as a more random selection of them, than if we would have scheduled separate focus group meetings. The teams were heterogeneous regarding expertise, treatment options, and department within the organization. Most AMHS professionals worked directly as therapists with adult out-patients in the domains mentioned above. Some others worked on an internship basis and had few independent contacts with patients. In one of the team meetings a psychiatrist was present. The majority were women, reflecting the gendered division of labor in this sector. Inclusion and data analysis ran in parallel, providing a constant feedback-loop between both processes. To provide adequate depth on this topic, inclusion was continued until no new insights with respect to the main research questions emerged (32, 33).

Procedure and Analysis

Because of the COVID-19 pandemic, all focus groups were held remotely. Focus groups were conducted in Dutch. Researchers SE and SL led all focus groups, alternating a leading or observational role. As participants were direct colleagues and thereby familiar with each other, focus groups started with researchers introducing themselves and the study. Next, two questions were asked, both oral and written in the chat function of the online environment. We used a variant of the 1–2–4-All technique (34), that is participants were asked to take 2 min to formulate their individual answer to these questions, followed by a brief discussion of another 2 min of their answers with one other participant in break-out rooms. Remaining time was used for a group discussion followed by a short summary by the researchers and a possibility for remaining questions and remarks. Researchers used probing questions to reach more in-depth answers. Inductive thematic analysis was conducted (32, 33). Analysis started with an explorative phase of open coding, in which basic themes were defined, followed by merging these themes into more conceptual categories, and after the major topics were identified, codes were analyzed further identifying the most important themes.

RESULTS

Use of the COPMI Check

Figure 1A shows the way professionals dealt with the COPMI question for the total of 14,469 patients aged 20–65 that were treated by Mondriaan between December 2016 and January 2021.

For 58% of the 14,469 patients aged 20–65, the COPMI check instrument was not used at all, that is: none of the three answering categories to the COPMI check question were selected. We call this category 0.

Figure 1B quantifies how professionals responded to the COPMI check question when they did not ignore it. 78% of these patients was judged to be *not at risk* regarding their children

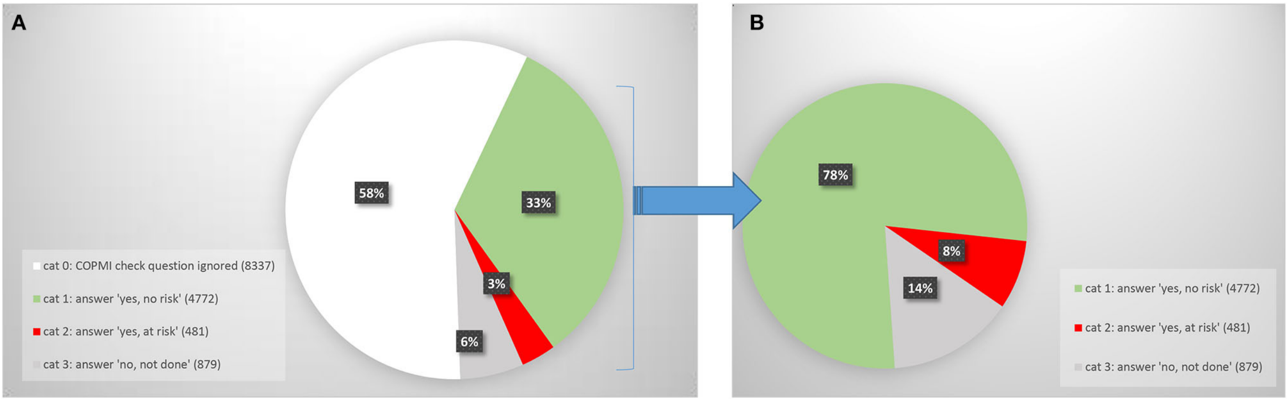


FIGURE 1 | (A) Answer to COPMI check question: all patients (*N* = 14,469). **(B)** Answer to COPMI check question: patients for whom the question was answered (*N* = 6,132).

(category 1), while 8% was judged to be *at risk* regarding their children (category 2). Category 3 (14%) exists of patients for whom the professional answered the COPMI question by saying they did *not do* the COPMI check.

It is important to note however that category 1 (judged not to be at risk regarding their children: 78%) includes many patients who simply do not have children. Thus, to interpret the figures, it is necessary to distinguish between patients who are and who are not caregivers for underage children.

Caregivers vs. Non-caregivers

Inspection of the data revealed that when patients did *not* have children, the question "have you carried out the COPMI check" created confusion for the professionals. When their patient was not a caregiver, some professionals responded that they had done the COPMI check but "no risk" exists (category 1) because no children are present. Yet others responded that they have "not carried out" the COPMI check (category 3) because no children were present. This is a result of an apparent multi-interpretability of the COPMI check question. This inconsistency in interpretation makes it necessary to quantify the number of caregivers within each category.

We checked parenting status in the representative sample of *N* = 75 taken from all patients in categories 1 and 2, the results of which were extrapolated to all patients in those two categories. In addition, we did a visual check of all patients in category 3. Parenting status is shown in **Figure 2** for each category respectively.

Of the patients in category 1 ("no risk regarding COPMI"), only 33% is caregiver. In other words, 2/3 of the patients who get the judgement "no risk", are given this judgement *because they have no children.* Of the patients judged to be at risk because of their children (category 2), predictably almost all are caregivers for children (96%). Of the patients in category 3 ("COPMI check not carried out"), only an estimated 2% is caregiver. Thus, where professionals responded that they had not carried out the COPMI check, this was almost always *because their patients had no children.*

Figure 3 shows how the COPMI check question was answered *for the caregivers only*. Because the patients without children have been filtered out, this presents more relevant numbers than **Figure 1B**.

Content Analysis: Professionals' Assessment and Actions When Patients Are Caregivers

Two random samples (*N* = 75 each) were taken for patients in category 1 and 2, respectively. Below, we report on the files of the 97 patients in the samples that were caregivers (**Table 1**).

For those patients who *were* caregivers of underage children, how did professionals estimate the risks to these children? Moreover, to what extent did they initiate further steps, including filing an official report to child abuse authorities and/or organizing help?

It was possible to categorize the situations that professionals encountered into five types of situations (**Table 2**). These situations were found both when there was judged to be risk or no risk.

Within each type of situation, we found a variety in courses of action taken by professionals. Among these were the following:

- Regular discussion of the situation of the children with the patient
- Monitoring whether the situation seems to get worse
- Psycho-education about COPMI
- Referring the children to a COPMI training (not necessarily taken up)
- Coordinating with support organizations already involved with the family
- Organizing new help
- Addressing parental interaction as part of treatment
- Inviting the children to therapy sessions
- Inviting co-caregivers to therapy sessions
- Reporting to GP (either separately or included in the regular reports to the GP)

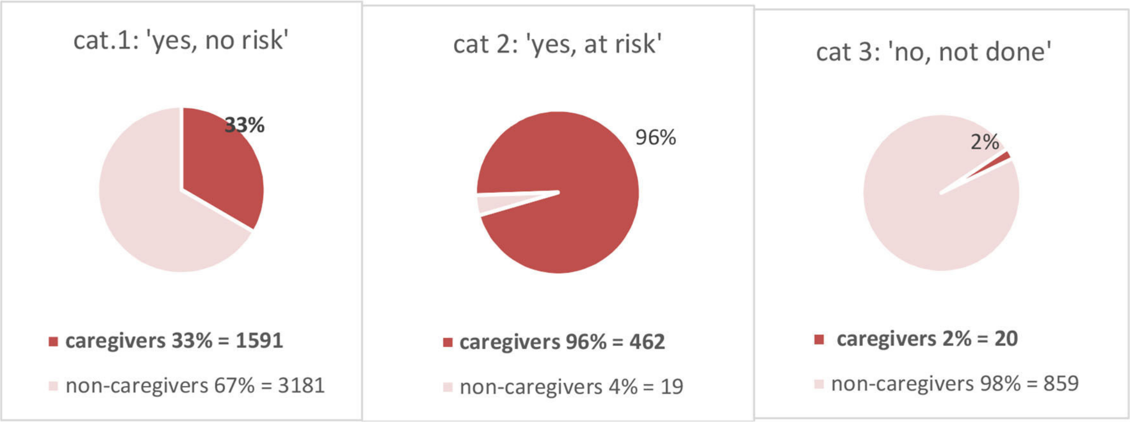


FIGURE 2 | Parenting status for each category.

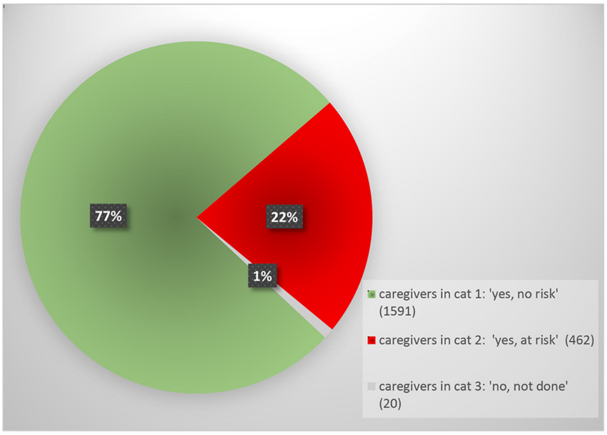


FIGURE 3 | Answer to COPMI check question: caregivers only (*N* = 2,073).

- Consulting with child abuse authority
- Reporting to child abuse authorities (Safe at Home).

Yet, among the 72 patients with children who were judged to be "at risk", in 24 cases (33%) no actions were reported in the patient's file, even though we qualified even limited interventions as actions, as can be seen in the list above. For example in situation 1 (the situation "in which there are concerns, but some treatment or help is already in place"), there were no actions in 15 cases. Yet, in other cases in the same type of situation, professionals did undertake action. They would for example ask for permission to contact the other help involved, if this was not given they conferred with colleagues whether to break the confidentiality. If the permission was given, in some cases there was intensive coordination with the other support organizations, such as drawing up a safety plan together. Likewise, in the seven cases which we classified as situation 5 ("before concerns can be addressed, the patient discontinues treatment"), in three cases nothing was done after the patient stopped the treatment, while in the other cases the general practitioner was informed that there were concerns about the children. Overall, in each of the situations categorized above, supportive actions were initiated for about 2/3rd of the patients, yet in one out of three nothing was done.

TABLE 1 | Caregivers in the subsamples.

Samples	Non-caregiver	Caregiver
Subsample from category 1 (answer "no risk") *N* = 75	50	25
Subsample from category 2 (answer "at risk") *N* = 75	3	72
Total sample (*N* = 150)	53	97

Focus Groups

The focus groups were used to help interpret and enrich the results from the file analysis. To find out more about the way professionals see their role and possibilities in relation to the COPMI and the COPMI check, focus groups started with a relatively open question:

"Given that you have an adult patient who is caregiver for (an) underage child(ren).

- What would you–as a mental healthcare professional–wish to be able to do in an ideal situation? And how could you be facilitated to do that?
- How do you experience the role of the Child check in this?"

1. The general line of answer was that professionals wished to have more information on what was really happening at their patients' homes. Several professionals mentioned that they wished they could talk to other people involved, including the partner, school, or neighbors, or doing home visits. In addition, the wish for some report from the GP was mentioned, so that

TABLE 2 | Types of situations encountered by mental healthcare professionals.

Types of situations	Description	Number of patients with this type of situation
1 Shared concern, some COPMI-related support already present	Professionals have concerns, patients share this concern to a degree. At the time of the intake, the children were already receiving treatment, or some parenting assistance organization was involved with the family. This includes situations where the patient is a divorced parent and his or her child lives with the other parent, while visitation takes place under the guidance of the Child protection agency.	43
2 Shared concern, no COPMI-related support present	Professionals have concerns, patients share this concern to a degree. At the time of the intake, there is no formal support given to family, children or parent regarding COPMI issues.	19
3 Shared concern, informal help seems present	There are concerns, but the patients rely on their social network to compensate.	4
4 Concern addressed by the professional, not shared by patients	Mental healthcare professional were concerned, however, patients emphasized that their vulnerabilities did not influence the well-being of their children. In some cases, children were mentioned as protective factor. In other cases contact with the children was very limited. Professionals seemed to agree with this.	9
5 Concern, without possibility to discuss with patients	Mental healthcare professionals were unable to discuss their concerns because patients avoid discussing the subject or discontinue treatment before any actions concerning the children could be taken.	7
6 Not enough information in files		15 (16%)

they would know beforehand whether there were risks at home. A form of "truth finding" seemed to be the dominant focus for professionals when confronted with the COPMI check and the question of COPMI. Without extensive information on the situation of their patients and their children, most professionals refrained from taking any action, making this wish for "truth finding" an important barrier in arranging any help for COPMI.

2. Another theme which was often voiced was the concern of professionals to harm the therapeutic relationship, as they could feel intrusive and stigmatizing when asking about the children. This concern seems also related to the truth finding focus, as the fear is related to a continued probing into the situation of the patients' children. Some respondents felt that an obligatory set of questions might help them ask about their home situation without having to seem suspicious and thereby hurting the relationship. Some others felt the COPMI check already provided this role of legitimizing the probing questions.

3. The focus among some of the professionals on truth finding – as opposed to introducing support - became especially clear when the professionals were asked what they would do if they had enough information on their patients' children. One professional admitted not yet to have thought about that and another one answered "to follow the steps of the Reporting protocol". Steering more toward the possibility of organizing help for the children of their patients, one professional said: "yes if you look at it that way, we should actually try to get every patient to have their children participate in a COPMI-prevention group".

4. Some focus group participants addressed the possibility of initiating support more directly. They also mentioned some barriers. For example the fear that there would be long waiting lists in CAMHS was mentioned as barrier to even introducing the subject to their patients.

5. Reflecting on the COPMI check tool, some focus group members addressed it as positive, because it was part of step by step guide toward reporting in case of child abuse. None said it helped them choose a form of help to offer or initiate.

6. There were also remarks that shed light on the large group of patients for which the COPMI check was not done at all. Professionals mentioned that they sometimes forget it, or skip it for lack of time, Also, there is some irritation about the proliferation of obligatory instruments that professionals have to use, more seem to added all the time. The COPMI check (although maybe the most relevant of all, one person said) sometimes becomes submerged in the total of such obligations.

7. All in all, the issue of what to do when your patient has underage children seemed to provoke anxiety and a feeling of falling short with quite a few professionals.

8. All recognized the importance though, and none said that this should not be part of their work.

DISCUSSION

While many studies have studied the effectiveness of interventions for COPMI, this study is one of few focusing on improving access to such interventions, and focusing on the potentially very effective access through the parents (35). The Dutch "COPMI check" has enabled us to study in a focused way whether such a mandatory check contributes to the increase of support, prevention and (early) intervention for COPMI.

Our results showed that for a majority (58%, $n = 8{,}337$) of all Mondriaan patients aged 20–65, the COPMI check tool was not used at all by the professionals, a high percentage given that it is mandatory. Among these patients, we expect that there were many patients with underage children. We did not take a sample of this group to quantify this, but recent research showed that in Norway the number of outpatients with children was 36% (36, 37). A conservative estimate would therefore be that at least 25–30% of this group of patients. Thus, we conclude that for *at least 500–600 patients each year who do have children, the COPMI check question was not answered*. In other words, the COPMI check as operationalized at Mondriaan Mental Health Center, is still often either overseen or for some other reason not given attention. Previous research showed that mandatory instruments in mental healthcare can indeed be experienced as "a paper-filling exercise for managers" if insufficient argumentation and feedback is given concerning the added value of the instrument (38). Focus group discussions suggest that with the COPMI check as well, even though the importance of general issue of COPMI is recognized, using the tool is not always perceived as helpful, especially given the number of other obligatory instruments and rules that professionals nowadays are confronted with. In the total of such requirements, it can become unclear where the priorities of organizations lie.

Of course it is possible that for the (estimated) 2,000–2,500 patients with children for whom the tool was not used, the COPMI issue was still taken on, even though the tool was skipped. We have not been able to check this in the present study. Given the awareness and motivation regarding COPMI encountered in the focus groups, in combination with being overburdened by mandatory tools, it may well be that at times professionals did address the COPMI question even while neglecting the tool. Thus we *cannot* conclude that for all these patient the issues of their children was neglected. Yet, the numbers are high enough to give a worrying indication that too many COPMI problems may remain unseen.

For an additional 6% of all patients ($n = 879$), the mental health professionals explicitly responded that they did not do the COPMI check. This group of patients almost always did not have children, which was also the reason given for not carrying out the check. Thus by responding that they are "not doing the check", in fact these professionals showed that they did check whether there were COPMI.

For the other 36% of patients aged 20–65, the COPMI check was carried out and patients were classified as either "risk" or "no risk" with regard to COPMI. In the no-risk group, we found that 66% the patients were not caregiver for underage children. Applying the COPMI check in those cases came down to stating that there was no risk, because there were no children involved. So in two-thirds of the cases where "no risk" was reported, the reason for this was the absence of any children. As we saw that other professionals whose patients had no children said that they had *not* done the COPMI check for that reason, we conclude that the wording and answering categories of the COPMI check question were multi-interpretable. This makes the quantitative data difficult to interpret without content analysis of the files to reveal parenting status. The multi-interpretability is also confusing to the user, as discussed and recognized in the focus group of the internal ambassadors. Recommendations will be necessary for a possible redesign of the tool, such as to reword the COPMI check, starting with a more basic question such as: "is the patient a caregiver for underage children or are there underage children in the household". Among the *caregivers* for whom the COPMI check was carried out, the qualification "at risk" with regard to COPMI was given in only 22% of the cases. If "at risk" means that these children may develop lasting and serious emotional problems, then we know from research that the percentage is likely to be higher than 22%. A meta-analysis from 2012 (14) showed that one out of two (50%) COPMI develop a mental illness, with 32% developing a severe one. Possibly, professionals interpret "at risk" as meaning: immediate safety risk. We have already discussed how the COPMI check is introduced with a focus on safety and child abuse. The focus groups show that many professionals have taken on this narrow focus, which may lead to an underestimation of the *needs* of COPMI.

Analysis of the patient files of a random sample of patients was carried out. Focusing on caregiving patients for whom the COPMI check was carried out ($N = 97$), five types of situations encountered by professionals were identified, the most prevalent being "shared concern, some COPMI-related support already present". We also identified a spectrum of actions that were taken by professionals to help support COPMI. However, for those COPMI judged to be at risk, in 33% of the cases no actions were taken at all. This is reason for concern, given the professionals' own judgement that there is risk (while, as mentioned above, that judgement itself already seems an underestimation). For the remaining 67% of patients on the other hand, there were examples of (sometimes relatively simple) sensitive and well informed actions to help COPMI. We conclude from this that in all the situations encountered by professionals, courses of actions are indeed available to them, as indeed the literature has shown as well (8, 39–41). Yet these actions toward support are still not readily or standardly carried out.

The focus group discussions shed light on the above. They showed that many professionals are more focused on truth finding (being clear whether there are severe problems at home) rather than on initiating support. We suggest that this focus comes from the fact that the COPMI check is presented as the first step in a protocol leading to the reporting of child abuse. As shown earlier, this protocol is concerned with "safety" rather than "needs" of COPMI. This study makes clear that this leads to a second bias, namely a focus on "deciding to report or not" (a focus that requires truth finding), vs. a focus on "organizing help".

Contrasting a *safety/reporting focus* with a *needs/support focus* helps put into perspective the COPMI check and its limitations. Other studies have also warned for the consequences of limited focus on questions of safety and reporting (42, 43). From a historical and political perspective, the positioning of a COPMI check in terms of *safety/reporting* is understandable: it reflects the fact that public opinion, media and political urgency are often safety/reporting focused (43). But a COPMI check with such a focus at an AMHS institution limits the potential for COPMI. Rather than helping professionals to undertake basic supporting actions for COPMI in general, it sets professionals on a course to find out which children are at immediate safety risk and should be reported for child abuse. This requires truth finding, which is understandably a burden since an adult mental health professional is not in a likely position to undertake truth finding. With it comes a fear to harm the therapeutic relationship and appearing suspicious, as questions concerning children are more threatening when posed from the perspective of safety/reporting.

In contrast, CAMHS organizations, such as the Child and Youth division or the Prevention division at Mondriaan, are, by the nature of their daily work, more *needs/support focused*. That is, they are geared to helping a larger group of children that need support (among them COPMI), also where there is no direct threat to safety in a narrow sense. It has been noted by professionals in Mondriaan's CAMHS that very few referrals of children are prompted by AMHS professionals from Mondriaan–an observation that merits to be researched. Likewise, a training offered to COPMI at Mondriaan Mental Health Center still receives fewer participants than it can provide for. Of course one must at the same time be realistic about the fact that in practice, support is not always available to children even when they are adequately identified. Long waiting lists and limits to the funding are unfortunately still a limiting factor in CAMHS. Nonetheless, identification, basic help and referral are the first steps. Our study shows that these can be improved with a differently focused COPMI check. We would like to conclude with a few specific practical recommendations.

Practical Recommendations

If the COPMI instrument could be redesigned toward a *needs/support focus,* a broader group of COPMI might be reached.

*Such a redesign would include basic guidance for professionals on how to initiate supportive actions, other than how to decide whether to report or not. We recommend that such practical guidance becomes part of the COPMI check.

*The guidance could make use of "best practice" examples, some of which we encountered in this study. Likewise the supportive actions that were taken up in 67% of the cases (as identified in this study), also provide a good starting point.

*CAMHS services should be given a role in the supportive actions, and in Mondriaan, where CAMHS and AMHS both take place within one organization, such collaboration could be institutionalized.

*Professionals should also be shown that they can take some supportive actions (for example: inviting the children to therapy sessions–possibly with help of colleagues from CAMHS) without having to know the exact extent of the problems at home. This would exempt them from some of the burden of truth finding. Possibly, such practical guidance on what to do next, once the professional has concluded that there are young children, would also contribute to a more widespread use of the COPMI check tool.

*The COPMI question(s) should be rephrased to resolve the multi-interpretability regarding patients that are not caregivers.

*AMHS organizations should carefully weigh the number of obligatory instruments that they introduce, in order for such instruments to retain their effectiveness.

Strengths and Limitations

A strength of this research project was its setting in a large Mental Health Center that provides both CAHMS and AMHS and features a mandatory tool to check for the needs of COPMI. It has enabled us to include a large data set concerning 14,469 patients, evaluate the impact of a mandatory tool and see whether the presence of CAHMS in the same organization plays a role in helping COPMI. Furthermore, combining quantitative and qualitative data allowed us to avoid an unrealistic interpretation of the quantitative data, and brought to light biases in the instrument and the effects they had. The results lead to direct recommendations for the local situation.

A limitation of the study is that we evaluated a tool which is presented as part of a protocol aimed at reporting child abuse; this limits its effectiveness in stimulating help for COPMI. Therefore, other barriers to stimulating this help came less clearly into sight. Yet we feel that it is very important that we brought to light how confounding helping COPMI with reporting child abuse hampers the way COPMI can be helped through professionals working in AMHS.

We studied only one organization and the results are not representative for all Dutch organizations working with the COPMI check, nor of course for other countries working with other tools. Yet the organization studied is certainly relevant as it has implemented the COPMI check for a relatively long time and in a structured way, compared to other AMHS organizations.

Regarding the focus groups, it should be noted that only a limited number of professionals participated, moreover, the time spent on the issue was relatively short. These limitations did however enable us to talk to a relatively randomly selected and much larger group than if we had asked people to free a lot of time to talk about the COPMI check. The information we received was rich, varied, and useful. Another limitation of the focus group method is the risk of group-think evolving. We avoided this by our variant of the 1–2–4-All technique. Another limitation is that we cannot be sure to which degree the context (for example the presence of factual or perceived hierarchies within group participants) has influenced results: it may for example have led to participants paying more lip service to the importance of taking responsibility for COPMI than they are actually experiencing.

Future Steps

It will be necessary to elaborate our study by focusing on those professionals who have focused more on helping than on reporting and have undertaken some action. One could then explore which barriers and enabling factors they encountered and by what means they could be assisted.

Our analysis of whether patients are caregivers for children touches upon the important question of parenting status among

AMHS patients in general. Unfortunately we could not draw definite conclusions on this issue, as we did not have data on the parenting status of the large group of patients for whom the COPMI check question was ignored. A content analysis of a representative sample of this group would be a worthwhile future research endeavor.

Studying the files of patients where the COPMI check tool was *not* used would also give information about to which degree in these cases (serious) COPMI issues were missed.

Finally, it would be quite interesting to implement some of the recommendations that emerge from this research regarding the redesign of the COPMI check. This could then be evaluated as to whether more COPMI are receiving help, as well as with regard to whether professionals feel more supported in their task to take on COPMI issues.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Maastricht University Medical Ethics Committee. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

SE, TA, and SL were involved in setting up the research and played an active role in the writing process. Data collection and analysis were done by SL and SE. All authors contributed to the article and approved the submitted version.

ACKNOWLEDGMENTS

We would like to express our gratitude to all participants in the focus groups. We thank Mondriaan's board and scientific committee for allowing us full access to all relevant information within their organization and Jasper van der Biezen for his voluntary assistance during the sampling.

REFERENCES

1. Rasic D, Hajek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder, and major depressive disorder: a meta-analysis of family high-risk studies. *Schizophr Bull.* (2014) 40:28–38. doi: 10.1093/schbul/sbt114
2. Leijdesdorff S, van Doesum K, Popma A, Klaassen R, van Amelsvoort T. Prevalence of psychopathology in children of parents with mental illness and/or addiction: an up to date narrative review. *Curr Opin Psychiatry.* (2017) 30:312–7. doi: 10.1097/YCO.0000000000000341
3. Campbell TC, Reupert A, Sutton K, Basu S, Davidson G, Middeldorp CMet al. Prevalence of mental illness among parents of children receiving treatment within child and adolescent mental health services (CAMHS): A scoping review. *Eur Child Adolesc Psychiatry*. (2021) 30:997–1012. doi: 10.1007/s00787-020-01502-x
4. Hosman CM. van Doesum, KT, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I The scientific basis to a comprehensive approach. *Adv Ment Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
5. Williams VE. *Living with a Parent with Mental Illness: Descriptions of Caring, Concealing and Coping*. Diss. Grand Canyon University. (2021).
6. Radicke A, Barkmann C, Adema B, Daubmann A, Wegscheider K, and Wiegand-Grefe S. Children of parents with a mental illness: predictors of health-related quality of life and determinants of child–parent agreement. *Int J Environ*. (2021) 18:379. doi: 10.3390/ijerph18020379
7. Tapias E, Coromina M, Grases N, Ochoa S. Psychological treatments with children of parents with mental illness: a systematic review. *Child Youth Care Forum.* (2021) 50:1107–30. doi: 10.1007/s10566-021-09608-2
8. van Doesum KT, Hosman CM. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: II. interventions. *Aus J Adv Ment Health.* (2009) 8:264–76. doi: 10.5172/jamh.8.3.264
9. Reupert AE, Cuff R, Drost L, Foster K, Van Doesum KT, Van Santvoort F. Intervention programs for children whose parents have a mental illness: a review. *Med J Aust.* (2013) 199:S18–22. doi: 10.5694/mja11.11145
10. Breslend NL, Parent J, Forehand R, Peisch V, Compas BE. Children of parents with a history of depression: the impact of a preventive intervention on youth social problems through reductions in internalizing problems. *Dev Psychopathol.* (2019) 31:219–31. doi: 10.1017/S0954579417001821
11. Rosenblum KL, Muzik M, Morelen DM, Alfafara EA, Miller NM, Waddell RM, et al. A community-based randomized controlled trial of Mom Power parenting intervention for mothers with interpersonal trauma histories and their young children. *Arch Womens Ment Health.* (2017) 20:673–86. doi: 10.1007/s00737-017-0734-9
12. Pitman E, Matthey S. The SMILES program: a group program for children with mentally ill parents or siblings. *Am J Orthopsychia.* (2004) 74:383–8. doi: 10.1037/0002-9432.74.3.383
13. Wolpert M, Hoffman J, Martin A, Fagin L, Cooklin A. An exploration of the experience of attending the Kidstime programme for children with parents with enduring mental health issues: parents' and young people's views. *Clin Child Psychol Psychiatry.* (2015) 20:406–18. doi: 10.1177/1359104514520759
14. Siegenthaler E, Munder T, Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17 e8. doi: 10.1016/j.jaac.2011.10.018
15. Thanhäuser, M, Lemmer, G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. *Curr Opin Psychiatry*. (2017) 30:283–99, Wolters Kluwer. doi: 10.1097/YCO.0000000000000342
16. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness: A necessity to provide relevant support. *Front Psychiatry.* (2019) 9:728. doi: 10.3389/fpsyt.2018.00728
17. Karibi H. Arblaster K. Clinician experiences of "Let's Talk about Children" training and implementation to support families affected by parental mental illness. *J Ment Health Train Educ Pract.* (2019) 14:201–11. doi: 10.1108/JMHTEP-08-2018-0044
18. Lauritzen C, Reedtz C, Van Doesum K, Martinussen M. Factors that may facilitate or hinder a family focus in the treatment of parents with a mental illness. *J Child Fam Stud.* (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
19. Pfeiffenberger AS, D'Souza AJ, Huthwaite MA, Romans SE. The well-being of children of parents with a mental illness: the responsiveness of crisis mental health services in Wellington, New Zealand. *Child Fam Soc Work.* (2016) 4:600–7. doi: 10.1111/cfs.12186

20. Maybery D, Goodyear M, O'Hanlon B, Cuff R, Reupert A. Profession differences in family focused practice in the adult mental health system. *Fam Process.* (2014) 53:608–17. doi: 10.1111/famp.12082
21. Tchernegovski P, Maybery D, Reupert A. Legislative policy to support children of parents with a mental illness: revolution or evolution? *Int J Ment Health Promot.* (2017) 19:1–13. doi: 10.1080/14623730.2016.1270847
22. Lauritzen C, Reedtz C, Rognmo K, Nilsen MA, Walstad A. Identification of and support for children of mentally ill parents: a 5 year follow-up study of adult mental health services. *Front Psychiat.* (2018) 9:507. doi: 10.3389/fpsyt.2018.00507
23. Besluit verplichte meldcode huiselijk geweld en kindermishandeling. *Staatsblad.* (2013) 1–18. Available online at: https://zoek.officielebekendmakingen.nl/stb-2013-324.html (accessed February 17, 2022).
24. The Child Check. Signalling Child Abuse based on Parental Signals. (2020). Available online at: https://www.huiselijkgeweld.nl/publicaties/factsheets/2018/11/15-september-2020/the-child-check (accessed February 17, 2022).
25. Model Reporting Code Domestic Violence and Child Abuse, action plan for responding to signs of domestic violence and child abuse. (2013). Available online at: https://www.government.nl/documents/reports/2013/03/14/model-reporting-code-domestic-violence-and-child-abuse (accessed February 17, 2022).
26. Definitions of Child Abuse and Neglect. *Child Welfare Information Gateway and U.S. Department of Health and Human Services.* (2019). Available online at: https://www.childwelfare.gov/pubpdfs/define.pdf (accessed February 17, 2022).
27. Ridderbos-Hovingh M, Frederiks BJM, Veen C, Tingen A, Beukers M, Geertsema JB, et al. Winter *Evaluatie Wet verplichte meldcode huiselijk geweld en kindermishandeling. ZonMw Den Haag.* Den Haag: ZonMW (2020)
28. Implementatie impuls kindcheck bij de GGZ. In: *Huiselijk Geweld en Kindermishandeling Projectenbundel.* ZonMw DenHaag. (2020)
29. De werking van de meldcode huiselijk geweld en kindermishandeling. Available online at: https://www.rijksoverheid.nl/documenten/rapporten/2015/06/15/rapportage-onderzoek-onder-artsen-naar-de-werking-van-de-meldcode-huiselijk-geweld-en-kindermishandeling (accessed February 17, 2022).
30. Lincoln YS, Guba, EG. Naturalistic Inquiry. Newbury Park, CA: Sage Publications. (1985) doi: 10.1016/0147-1767(85)90062-8
31. Palinkas LA, Horwitz SM, Green CA, Wisdom JP, Duan N, Hoagwood K. Purposeful sampling for qualitative data collection and analysis in mixed method implementation research. *Adm Policy Ment Health.* (2015) 42:533–44. doi: 10.1007/s10488-013-0528-y
32. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* (2006) 3:77–101. doi: 10.1191/1478088706qp063oa
33. Boeije, H. Analyseren in kwalitatief onderzoek. *Denken en doen. Boom Uitgevers Amsterdam.* (2005). doi: 10.5117/2006.011.001.015
34. Kimball L. Liberating structures: A new pattern language for engagement. *Systems Thinker.* (2012) 23:2–6. Available online at: https://thesystemsthinker.com/wp-content/uploads/pdfs/230101pk.pdf
35. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child Youth Serv Rev.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
36. Ruud T, Maybery D, Reupert A, Weimand B, Foster K, Grant A, et al. Adult mental health outpatients who have minor children: Prevalence of parents, referrals of their children, and patient characteristics. *Front Psychiat.* (2019) 10:163. doi: 10.3389/fpsyt.2019.00163
37. Maybery D. Reupert A. The number of parents who are patients attending adult psychiatric services. *Current Opinion Psychiat.* (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
38. Jacobs R, Moran V. *Uptake of mandatory outcome measures in mental health services.* Cambridge University Press. (2018)
39. Marston N, Stavnes K, Van Loon LM, Drost LM, Maybery D. Mosek A. Reupert A content analysis of Intervention Key Elements and Assessments (IKEA): What's in the black box in the interventions directed to families where a parent has a mental illness? *Child and Youth Services.* (2016) 37:112–28. doi: 10.1080/0145935X.2016.1104041
40. Grové C, Riebschleger J, Bosch A, Cavanaugh D, van der Ende PC. Expert views of children's knowledge needs regarding parental mental illness. *Child Youth Serv Rev.* (2017) 1:249–55. doi: 10.1016/j.childyouth.2017.06.026
41. van Doesum KTM, Reedtz C, Lauritzen C. Child Talks+: A new intervention to support families affected by parental mental illness. *Journal of Parent and Family Mental Health.* (2021) 6:2639–9997. doi: 10.7191/parentandfamily.1016
42. Brown R, Strozier M. Resisting Abuse at What Cost? The Impact of mandated reporting laws on the process and content of therapy. *Contemporary Family Therapy.* (2004) 26:45–60. doi: 10.1023/B:COFT.0000016911.48414.7d
43. O'Donnell M, Scott D. Stanley F. Child abuse and neglect — is it time for a public health approach? *Aust N Z J Public Health.* (2008) 32:325–30. doi: 10.1111/j.1753-6405.2008.00249.x

Conflict of Interest: SE and TA are involved as employees at the mental healthcare facility where the research was conducted.

The remaining author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Psychiatry

SYSTEMATIC REVIEW
published: 24 March 2022
doi: 10.3389/fpsyt.2022.765118

Maternal Suicide Ideation and Behaviour During Pregnancy and the First Postpartum Year: A Systematic Review of Psychological and Psychosocial Risk Factors

Holly E. Reid[1,2], **Daniel Pratt**[1,2,3], **Dawn Edge**[1,2] **and Anja Wittkowski**[1,2,3]*

[1] Division of Psychology and Mental Health, School of Health Sciences, Faculty of Biology, Medicine and Health, University of Manchester, Manchester, United Kingdom, [2] Manchester Academic Health Science Centre, Manchester, United Kingdom, [3] Greater Manchester Mental Health NHS Foundation Trust, Manchester, United Kingdom

Suicide is a leading cause of maternal death during pregnancy and up to a year after birth (perinatal period). Many psychological and psychosocial risk factors for maternal suicidal ideation and behaviour have been investigated. Despite this, there have been no attempts to systematically search the literature on these risk factors. Additionally, few studies have described how the risk factors for suicidal ideation, attempted suicides and suicide deaths differ, which is essential for the development of tools to detect and target suicidal ideation and behaviour. Seven databases were searched up to June 2021 for studies that investigated the association between suicidal ideation and/or suicidal behaviour and psychological/psychosocial risk factors in pregnant and postpartum women. The search identified 17,338 records, of which 59 were included. These 59 studies sampled a total of 49,929 participants and investigated 32 different risk factors. Associations between abuse, experienced recently or during childhood, and maternal suicide ideation, attempted suicide and death were consistently reported. Social support was found to be less associated with suicide ideation but more so with suicide attempts. Identifying women who have experienced domestic violence or childhood abuse and ensuring all women have adequate emotional and practical support during the perinatal period may help to reduce the likelihood of suicidal behaviour.

Keywords: women, mothers, perinatal, suicide deaths, suicide attempts, suicidal ideation

Edited by:
Laura Orsolini,
Marche Polytechnic University, Italy

Reviewed by:
Suwa Goden Goar,
University of Jos, Nigeria
Giuseppe Tavormina,
Independent Researcher, Provaglio d'Iseo, Italy

***Correspondence:**
Anja Wittkowski
anja.wittkowski@manchester.ac.uk

Specialty section:
This article was submitted to Psychopathology, a section of the journal Frontiers in Psychiatry

Received: *26 August 2021*
Accepted: *14 February 2022*
Published: *24 March 2022*

Citation:
Reid HE, Pratt D, Edge D and Wittkowski A (2022) Maternal Suicide Ideation and Behaviour During Pregnancy and the First Postpartum Year: A Systematic Review of Psychological and Psychosocial Risk Factors. Front. Psychiatry 13:765118. doi: 10.3389/fpsyt.2022.765118

INTRODUCTION

In the United Kingdom and Ireland, maternal suicide is the leading direct cause of death between 6 weeks and a year after the end of pregnancy and the second commonest direct cause of death occurring during or within 6 weeks of the end of pregnancy (1). This phenomenon is not exceptional to the United Kingdom; maternal suicide has been shown to be among the leading causes of death in other high-income countries, such as Australia (2), France (3), Italy (4), and the five Nordic countries (5). The United Kingdom and Ireland Confidential Enquiries into Maternal Deaths and Morbidity (1) reported that the rate of suicide during the perinatal period (i.e., pregnancy and the first postpartum year) has slightly increased over the last decade, with 0.60 deaths by suicide per 100,000 maternities reported in 2012–2014 and 0.63 deaths by suicide per 100,000 maternities reported in 2016–2018. Many more women who die by suicide will attempt suicide and a recent meta-analysis of 14 studies found the worldwide prevalence of suicide attempts during pregnancy was 680 per 100,000 and 210 per 100,000 during the postpartum period (6). Even greater numbers of women experience thoughts of suicide and/or self-harm, which is not only distressing, but may lead to a suicide attempt. The prevalence of self-harm ideation during the perinatal period was found to range from 5 to 14% (7).

Suicide prevention and risk reduction should be key intervention targets since maternal death by suicide has a profound effect on the woman's child(ren), including loss of a primary care giver, feelings of separation and abandonment (8), increased rate of depressive symptoms (9), increased risk of bipolar disorder (10), and an increased risk of death by suicide themselves (11). Furthermore, maternal suicidal ideation alone has been associated with poorer child cognitive outcomes, including motor skills and language development (12).

Maternal suicides during the perinatal period appear to be distinguished from non-perinatal suicides in several ways. Firstly, these suicides tend to occur through more violent methods (e.g., hanging or jumping from a height) when compared with suicides of non-perinatal women (13–15). Secondly, during pregnancy and the first postpartum year most women have significantly more contact with healthcare professionals than at other times in their lives, but despite this, suicide has remained a leading cause of death in mothers. Thus, regular access to healthcare services alone may not be enough to encourage mothers to seek help for suicidal thoughts and/or behaviour and ultimately to avoid maternal death by suicide. An additional complexity is the supposed protective factor of being a parent; parenthood is associated with a lower risk of suicide in both men and women (16), but parenthood has also been found to be an impetus for suicide attempt among those who report high parenting stress (17). Given these unique characteristics of maternal suicidal ideation and behaviour, it is important to establish the risk factors associated with suicide during the perinatal period rather than extrapolate from research with non-perinatal samples.

Perinatal mental disorders are the most common complication of childbearing (18–20) and the early postpartum period is a particularly risky time for first and recurrent episodes of severe mental illness (21, 22), which may go some way towards explaining the high incidence of suicidal ideation and behaviour in the perinatal population. The risk of maternal suicide is significantly increased in mothers with first-onset severe psychiatric disorders compared to mothers with no psychiatric history (23). However, most people with a mental health problem never become suicidal and fewer than 5% of people admitted to hospital for the treatment of an affective disorder die by suicide (24). Therefore, the presence of a mental health problem has little predictive power and more specific risk factors for suicidal ideation, attempts and maternal death by suicide need to be identified.

In their epidemiological review of suicidal ideation during pregnancy only, Gelaye, Kajeepeta, and Williams (25) selected 57 studies for inclusion and identified intimate partner violence (IPV), less than 12 years of education and major depressive disorder as risk factors for antepartum suicidal ideation. In their review of 129 studies that investigated risk factors and clinical correlates of suicide during both pregnancy and postpartum, Orsolini et al. (26) identified that suicides were more likely to occur among younger women, during unwanted and unintended pregnancies and in those with psychiatric diagnoses. Despite the comprehensiveness of their review, Orsolini et al. (26) did not clarify whether the factors increased the risk of suicidal ideation or non-fatal suicidal behaviour or deaths, and only studies published in English were included. A more recent meta-analysis of 39 studies reported on the prevalence and correlates of self-harm regardless of suicidal intent, during the perinatal period (27). The review authors identified mental disorder, substance misuse, younger age, being unmarried, and obstetric and neonatal complications as key correlates of maternal self-harm. Taken together, these reviews highlight a very broad range of variables that have been investigated and they provide an indication of demographic groups of women more at risk of suicide during the perinatal period. However, most of these factors are "non-modifiable," and therefore offer limited help when developing new interventions to reduce suicidal ideation and behaviour in perinatal women.

Previous studies have started to investigate a range of modifiable, psychological, and psychosocial factors that may increase a woman's risk of suicide during the perinatal period, such as hopelessness and impact of childhood trauma (14, 28). A review of these psychological and psychosocial risk factors has yet to be conducted, but it would offer an important first step towards the development of new interventions targeting the reduction of suicidal ideation and behaviour. Therefore, this review aimed to (1) summarise the psychological and psychosocial risk factors associated with maternal suicide outcomes (i.e., suicidal ideation, suicide attempts, and suicide deaths) during the perinatal period; (2) describe how these risk factors differ between women experiencing suicidal ideation alone, women who attempt suicide, and women who die by suicide, during the perinatal period.

MATERIALS AND METHODS

The systematic review was reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (29). The review protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO) on May 20, 2019 (registration number CRD42019107795).

Search Strategy and Eligibility Criteria

A systematic search of the literature was conducted in seven databases: EMBASE, Medline, PsycINFO, CINAHL Plus, Maternity and Infant Care, Applied Social Sciences Index and Abstracts and Web of Science. A search was also conducted using Google Scholar. To uncover any relevant unpublished studies and grey literature, the Centre for Reviews and Dissemination databases, ProQuest Dissertations and Theses (United Kingdom and Ireland, Health and Medicine) and EThOS were also searched. The first author and university librarian developed a combined search strategy of free text terms and exploded Medical Subject Heading (MeSH) terms for the topics of suicide and the perinatal period, and MeSH terms were adapted for each database. The search strategy for MEDLINE is presented in **Appendix A** as an example. The reference lists of all papers identified for inclusion within this review and of existing reviews and position papers were also examined for any additional papers. The search was most recently conducted in June 2021.

The eligibility criteria were developed using the PICOS (Population, Intervention, Comparison, Outcome, Study design) framework (30) and are outlined in **Table 1**. Studies were included if the sample comprised pregnant and/or postpartum women aged 18 years old or over. Samples comprising adolescents only or majority adolescents were excluded because teenage motherhood brings its own unique challenges for a young woman, such as mothering while dealing with her own adolescent development, negative public attitudes and a lack of preparation for motherhood (31), which may confound the risk factors for suicide. Studies that only included women following a miscarriage, stillbirth, termination of pregnancy, or ectopic pregnancy were excluded because the factors associated with these losses are likely to be different from those experienced by suicidal women who have not experienced a loss or termination. Studies were required to measure at least one psychological or psychosocial risk factor. For the purposes of this review, definitions of psychological and psychosocial factors were adapted from O'Connor and Nock (32): Variables were deemed psychological factors if they represented cognitive factors (e.g., rumination, defeat, entrapment, agitation, belongingness, and burdensomeness) or personality and individual differences (e.g., hopelessness, impulsivity, perfectionism). Variables were deemed psychosocial factors if they represented social factors (e.g., exposure to death by suicide, social isolation) or negative life events (e.g., childhood adversities, traumatic life events during adulthood). Studies that measured psychological or psychosocial risk factors, which were found not to be correlated with a suicide variable (i.e., positively or negatively), were included to ensure the review was not biased towards statistically significant findings.

Given the wide-ranging definitions and measures of suicide outcomes as well as the difficulties of establishing suicidal intent, defining suicidal ideation and behaviour presents a challenge. For this review, initially, we included only studies that used any measure of suicide ideation, any measure of suicide attempts and suicide deaths. However, once we scrutinised the search results, we discovered that many studies that claimed to measure suicide ideation, used item 10 of the *Edinburgh Postnatal Depression Scale* (*EPDS)* (33) to assess ideation. The item asks the respondent whether "*the thought of harming myself has occurred to me*" and the respondent may answer "*yes quite often*," "*sometimes*," "*hardly ever*," or "*never.*" There is uncertainty as to whether thoughts of harming oneself equate to suicidal ideation and the item does not explicitly ask the respondent whether the thoughts of self-harm are driven by suicidal intentions. However, endorsing "*yes quite often*" on the *EPDS* item 10 was found to be concordant with suicidality measured by the Clinical Interview Schedule–Revised measure of suicidality (34). In order to take an all-inclusive approach to defining suicide outcomes, we decided to include any studies that used the *EPDS* item 10, but we refer to the outcome measured by this item as "self-harm ideation" rather than suicide ideation. Studies that specifically focused on non-suicidal self-injury (i.e., self-harm with no suicidal intent) were excluded. To ensure all papers that met the eligibility criteria were identified, there were no restrictions on date of publication or language in which the study was reported.

Study Selection, Data Extraction, and Analysis

All database search results were imported into EndNote Online, a reference management service. The first author removed duplicates and screened all the identified studies to assess eligibility, according to the pre-specified inclusion and exclusion criteria. A peer outside of the research team also independently screened 50% of the identified abstracts, the percentage agreement between the independent raters was 99% (Cohen's $\kappa = 0.89$). The first author and peer discussed and resolved any discrepancies regarding eligibility of studies. When a study was published in a language other than English, translation of the abstract was sought to identify whether the study met the inclusion criteria. If the study met the inclusion criteria, translation of the entire paper was sought. The number of studies identified, screened and selected are presented in the PRISMA flow diagram (see **Figure 1**). Data were extracted from the included studies into a table in Microsoft Word by the first author. To verify the accuracy of the data extraction, data from 50% of the included studies were also extracted independently by a peer outside of the research team, and any discrepancies or uncertainties were discussed and resolved.

Using the extracted data, a narrative synthesis was conducted following Popay et al.'s (35) guidance. Studies were organised into the four categories adapted from O'Connor and Nock (32), depending on the risk factors measured: negative life events, social factors, cognitive factors and personality and individual differences. This then allowed for clustering of studies depending on the suicide outcome investigated and the interrogation of similarities and differences between the clusters and the studies in each cluster.

Risk of Bias and Quality Assessment

The studies included in the review were subject to a methodological quality assessment using the Quality Assessment Tool for Studies with Diverse Designs (QATSDD) (36). The review eligibility criteria are inclusive of a range of study designs and therefore the QATSDD was selected because it allows for the comprehensive examination of studies that employ different designs but address similar research questions. The QATSDD tool consists of a total of 16 criteria, two of which are solely applicable to quantitative studies and another two of the criteria are solely applicable to qualitative studies. Therefore, only mixed methods studies are assessed *via* all 16 criteria, and quantitative or qualitative studies are assessed by 14 criteria. Each criterion is weighted equally and rated 0

TABLE 1 | Eligibility criteria for inclusion of studies in the review.

	Inclusion criteria	Exclusion criteria
Population	• Pregnant women and/or women during the first 12 months following birth (perinatal period). • From any location. • With or without a psychiatric diagnosis. • Women aged 18 years and over. For studies that included a mixed sample of adolescents and adults, the mean or median age of the sample needed to be 18 years or older. • Both inpatient and community samples.	• Women who had suffered a miscarriage or stillbirth, following a termination of pregnancy for any reason or ectopic pregnancies. • Adolescents (less than 18 years).
Intervention (exposure)	• Psychological/psychosocial risk factors. • Assessed using any objective or subjective measure.	• Investigate the presence of mental health problems (e.g., psychosis, major depressive disorder) but with no measurement of psychological and/or psychosocial factors.
Comparison	• Exposure versus non-exposure to the psychological/psychosocial risk factor(s) of interest.	• Purely descriptive studies (e.g., case report studies).
Outcome	• Suicide, which can include: – Suicide deaths – Suicide attempts – Suicidal ideation – Suicide planning – Self-harm ideation – Self-harm with suicidal intent or suicidal intent is unclear • Assessed using any objective or subjective measure.	• A focus on non-suicidal self-injury.
Study design	• Report original quantitative findings.	• Report qualitative findings only. • Reviews, practice recommendations or guidelines, comments, replies, letters, and opinion/position papers.

(not at all), 1 (very slightly), 2 (moderately), or 3 (complete) using the scoring guidance notes. As this review only included quantitative studies (no mixed methods studies were identified), a total score out of a possible 42 points was calculated for each study (i.e., a maximum score of 3 for 14 criteria). The total percentage was also calculated to provide an overall indication of methodological quality and similarly to the guide outlined by Gillham and Wittkowski (37), a percentage score over 75% was considered "high" quality, 50–75% "good," 25–49% "moderate," and below 25% "poor." Furthermore, the percentage of studies that scored 1, 2, or 3 for each item was calculated to give an indication of how many of the studies addressed each of the 14 criteria. The first author rated all studies and made notes to elaborate on the QATSDD scoring guidelines to provide more detailed, tailored guidance to ensure consistency when scoring. A peer also rated 51% of the studies independently, using the additional scoring guidance notes provided by the first author. The percentage agreement between the independent raters was 86% before differences were discussed and resolved. None of the studies were excluded due to a "low" quality rating in order to ensure a broad range of risk factors were reviewed, although studies with particularly low ratings are highlighted and their methodologies discussed in the narrative synthesis.

RESULTS

Study Characteristics

The systematic search strategy identified 17,338 titles (see **Figure 1**). After removal of duplicates and screening of abstracts, 333 full text publications were evaluated. Once publications that did not meet inclusion criteria and/or lacked relevant data were removed, 59 studies reporting quantitative associations between suicide outcomes and psychological and/or psychosocial factors during the perinatal period were included (see **Supplementary Table 1** for study characteristics).

The 59 studies sampled a total of 49,929 participants and sample sizes ranged from 28 to 5,960. Twenty-three of the studies included pregnant women alone, 17 included postpartum women alone and 19 studies included both pregnant and postpartum women. Studies were conducted in a wide range of locations: 13 were conducted in the United States, 14 in Asia, 14 in Africa, nine in South America, seven in Europe, one in Australia, and one did not specify the location (38). Of the 59 studies included, 36 were cross-sectional in design (61%), 18 used a cohort design (31%), four were case-control studies (7%), and one used a mixed cohort and cross-sectional design whereby 121 of the 748 participants were seen at two time points (39).

Studies could be categorised depending on the suicide outcome measured: 43 studies measured suicidal or self-harm ideation alone (73%), 13 studies measured suicidal or self-harm ideation and suicide attempts (22%) either using a combined measure that assessed both the ideation and the attempts or *via* multiple measures that assessed the ideation and attempts separately, one study measured suicide attempts alone (2%) and two studies measured suicide deaths alone (3%).

A varied range of 32 psychological and psychosocial factors were investigated across the 59 studies. Those 32 studies were then grouped into the four categories adapted from O'Connor and Nock (32): negative life events, social factors, cognitive factors, and personality and individual differences. **Figure 2** illustrates the risk factors and suicide outcomes measured.

Risk of Bias and Quality Assessment of the Included Studies

Overall quality ratings for the 59 studies included in this review ranged between 33 and 86%, 44 of the studies were considered "good" quality (50–75%), 9 studies were "high" quality (>75%), and 6 were of "moderate" quality (25–49%). Ratings for each study on each of the QATSDD criteria are outlined in **Supplementary Table 2**. As no qualitative studies were included, criteria 11 and 14 were not utilised and removed. As evident in **Supplementary Table 2**, user involvement in the design of the studies was relatively poor (criterion 15), only seven studies used a pilot study, of which three used any pilot study feedback to inform changes to the study design and only one of the studies (40) consulted stakeholders about the study procedures. However, all 59 studies included some description of the research setting (criterion 3), recruited samples representative of perinatal women (criterion 5), used suitable methods of analysis (criterion 12), and included some explanation of choice of analytic method (criterion 13).

Negative Life Events

Negative life events were commonly investigated as risk factors for perinatal suicide, with 47 studies investigating psychological/psychosocial risk factors that could be categorised as negative life events. The methodological quality ratings of these 47 studies ranged from 36 to 86% with 35 considered "good" quality, 7 were "high," and 5 "low" quality. These negative life events could be clustered into four groups of very similar risk factors: experiences of abuse during childhood only or adverse childhood experiences, experiences of abuse at any time of life (childhood and/or adulthood), experiences of abuse during adulthood only or intimate partner violence (IPV), and stress (e.g., stress experienced during pregnancy or the postpartum, exposure to traumatic life events, post-traumatic stress symptoms, and 1st degree relative suicide).

Experiences of Abuse During Childhood Only or Adverse Childhood Experiences

Experiences of childhood abuse had significant associations with maternal suicide and self-harm ideation, and reports of physical childhood abuse appeared to be one of the strongest predictors across study designs. Sit et al.'s (41) cross-sectional findings from a sample of 628 postpartum women demonstrated that childhood physical abuse increased the odds of self-harm ideation by 68%, but childhood sexual abuse was not significantly associated with self-harm ideation. In their matched case-control study of 255 pregnant women, Leeners et al. (42) also showed childhood physical abuse increased the odds of suicide ideation by 20%; however, childhood sexual abuse was not significantly associated with suicide ideation, although there is limited information regarding whether the suicide ideation was experienced during the perinatal period or at another time during the participants' lives. Moreover, in Giallo et al.'s (43) longitudinal study of 1,507 women, the univariate analysis revealed that experiences of childhood physical abuse increased the odds threefold and childhood sexual abuse increased the odds almost twofold of reporting self-harm ideation persistently at each time point (3 months, 6 months, 12 months, 18 months, and 4 years postpartum). However, in Giallo et al.'s (43) multivariable model, childhood sexual abuse was no longer a significant predictor of self-harm ideation.

It is not only abusive experiences during childhood that have been investigated. Doi and Fujiwara (44) assessed adverse childhood experiences, which included asking participants about parental death, parental divorce, parental mental illness, neglect, economic hardship, and IPV against their mother as well as about physical and psychological abuse. The authors found that postpartum women with three or more adverse childhood experiences were almost five times more likely to experience recent self-harm ideation.

Experiences of childhood trauma were also strongly associated with suicidal behaviour. In one of only three papers in this review to provide separate data for those reporting suicidal ideation only and suicide attempt, Levey et al. (45) discovered that a history of childhood abuse increased the odds of suicidal ideation 2.57-fold, increased the odds of suicide planning almost threefold and increased the odds of suicide attempt 2.43-fold in a sample of 2,062 pregnant women. In their longitudinal study that followed 306 women from pregnancy to 16 months postpartum, Martini et al. (46) identified that childhood abuse or rape was significantly associated with suicidality (defined as thoughts of death or self-harm, suicide planning or suicide attempt).

Gressier et al. (28) examined suicide attempts and the association with childhood abuse and adverse childhood experiences, using a database created by the French Network of Mother and Baby Units (MBUs) which contains information about perinatal women admitted to 16 MBUs (13 in France, 3 in Belgium) between 2001 and 2010. The authors retrospectively assessed 1,439 women for suicide attempts and of these, 105 had attempted suicide during the postpartum period, and 49 had made attempts during pregnancy. The study compared three groups: women who attempted suicide during pregnancy, women who attempted suicide during the postpartum period and women who did not attempt suicide. There was no difference across the groups on measures of foster care in childhood, maltreatment in childhood or childhood sexual abuse.

In summary, consistent evidence suggests that physical abuse experienced during childhood, rather than sexual abuse, increases the odds of perinatal suicide and self-harm ideation. The findings also suggest that childhood abuse increases the odds of suicide planning but there are mixed results regarding its association with suicide attempts during the perinatal period.

Experiences of Abuse at Any Time of Life

One study could not be included in Sections "Experiences of Abuse During Childhood Only or Adverse Childhood Experiences" or "Experiences of Abuse During Adulthood Only or Intimate Partner Violence" because it measured abusive experiences during the lifetime (both during childhood and adulthood). Therefore, we cannot determine when the abuse was experienced. In their cohort study of 200 women living with HIV, Knettel et al. (40) reported no association between experiences of abuse at any point in life and suicidal/self-harm ideation during pregnancy or at 6 months postpartum. However, it is difficult to evaluate the measure of abusive experiences because information about how this was assessed was omitted from the report.

Experiences of Abuse During Adulthood Only or Intimate Partner Violence

Similar to experiences of childhood abuse, recent experiences of IPV, particularly physical IPV, were shown to increase the likelihood of suicidal or self-harm ideation. Cross-sectional studies include Doi and Fujiwara (44) who found IPV during pregnancy increased odds of self-harm ideation over fourfold (verbal IPV) and almost fivefold (physical IPV) in a large sample of 5,960 postpartum women. The association was demonstrated in reverse by Iyengar et al. (47) who found those who reported suicidal ideation to be 10 times more likely to experience IPV in a smaller sample of 120 pregnant; however, details about the assessment of suicidal ideation are very limited in this study. In their "high" quality study of 426 women, Islam et al. (48) measured IPV during the first 6 months postpartum and found the odds of postpartum self-harm ideation were 2.65 times higher when a woman had experienced physical IPV following birth; however, neither sexual IPV nor psychological IPV experienced after birth affected the odds of self-harm ideation. Longitudinal findings have demonstrated a continuous effect of IPV during the perinatal period. According to Rodriguez et al. (49), having experienced physical IPV within the previous 4 weeks increased the odds of experiencing self-harm ideation during pregnancy and again at 12 months postpartum by 17% in a sample of 681 women. In their longitudinal study, Fisher et al. (50) used a modified version of the *EPDS* item 10 which asked "*I have had thoughts that I do not want to live any more*" to measure suicide ideation and noted that any form of lifetime IPV (emotional, sexual, or physical) significantly increased the odds of having thoughts of not wanting to live any more. Moreover, experiencing two or three forms of lifetime IPV was associated with almost eight times increased odds of suicide ideation. Aside from IPV, sexual trauma experienced during military service and self-harm ideation in women veterans in the United States were investigated by Gross et al. (51). In a sample of 620 women veterans, both military sexual harassment and trauma were significantly associated with self-harm ideation during pregnancy but not postpartum.

Previous studies also consistently found significant associations between experiences of abuse during adulthood and suicidal ideation and attempts, although the majority of studies was homogenous in design (i.e., cross-sectional) and conducted with pregnant women only. For example, in Asad et al.'s (52) sample of 1,369 pregnant women, 48% of participants had experienced verbal abuse and 20% had experienced physical or sexual abuse during the pregnancy or within the 6 months prior to the pregnancy. Asad et al. (52) conducted one of only three studies in this review (45, 52, 53) that provided separate data for suicide ideation and suicide attempts (rather than a combined measure of "suicidal behaviour"). Not only were there significant associations between experiences of any form of abuse and suicidal ideation and suicide attempts, but the frequency of the abuse was also associated with an increased likelihood of suicidal ideation and attempts. However, the authors did not find experiences of abuse to be more or less associated with suicidal ideation, nor more or less associated with suicide attempt. Current IPV was also associated with a sixfold increase in likelihood of suicidal ideation and attempts (a combined measure) in 214 pregnant women (54), and IPV experienced at any point during a woman's lifetime was also found to be associated with increased odds of suicidal ideation, planning and attempts in a larger sample of 2062 pregnant women (45). According to Supraja et al.'s (55) bivariate analyses, any form of IPV (psychological, sexual, or physical) was strongly correlated with ideation during pregnancy, but IPV did not emerge as a significant predictor of ideation in their multivariate analysis. The authors asked participants who had attempted suicide during pregnancy to give a reason for their attempt and all eight of the women cited abuse and/or conflict with spouse as a reason for their attempt. Findings also demonstrated a link between history of rape and suicidal behaviour (56). In a sample of 988 women, Belete and Misgan (56) identified that an experience of rape increased the odds of suicidal behaviour by twofold at 6 weeks postpartum, although it is not clear whether "behaviour" refers to suicidal ideation, attempts, or both.

Two studies examined IPV and suicide deaths (57, 58). Both were case-control studies and used data extracted from the United States National Violent Death Reporting System whereby death by suicide was defined as a record of "death resulting from the intentional use of force against oneself." Gold et al. (58) compared pregnant and postpartum women with women of reproductive age who were not pregnant or postpartum at the time of death. The authors identified a total of 2,083 female suicide victims of reproductive age from 17 US states between 2003 and 2007. Pregnant women who died by suicide were three times more likely to have experienced intimate partner conflict and postpartum women were over two times more likely to have experienced intimate partner conflict. Similarly, Adu et al. (57) used data gathered from 18 US states from 2003 to 2012 to compare suicides of pregnant and postpartum women with non-pregnant females (15–54 years) and then compared urban and rural differences. The authors identified a total of 4,306 female suicide victims and found recent intimate partner problems were associated with increased odds of the victim being pregnant or postpartum compared to non-perinatal. This observation supports Gold's (58) findings and demonstrates that intimate partner problems as a key correlate for suicide during the perinatal period from 2007 to 2012. Two issues should be highlighted: a large proportion of Adu et al.'s (57) data were likely to have been the same data used in Gold et al.'s (58) study and neither study clarified what constituted an "intimate partner problem" or "conflict" and when and how frequently the "conflict" occurred.

To summarise, these studies consistently demonstrate that abuse experienced during adulthood increases the odds of suicide and self-harm ideation. There is also evidence that abuse experienced before and during the perinatal period was associated with ideation, and physical abuse appears to have the strongest association. IPV significantly increases odds of suicide planning, attempts and deaths, suggesting that receipt of abuse during adulthood cannot only trigger thoughts of suicide, but also enable women to act on those thoughts, and in some cases result in a fatal outcome.

Stress

Eleven studies measured risk factors that can be broadly grouped as "stress." These risk factors include stressful life events experienced during pregnancy (59, 60) or experienced at any point in life (39, 53, 61), psychosocial stress during pregnancy (62), general perceived stress (63), pregnancy stress (61), parenting stress (64), stress about debt (65), post-traumatic stress symptoms (39, 66), and first-degree relative suicide (67). When grouped, these studies reported mixed findings with regards to stress and its association with ideation and behaviour. In Tavares et al.'s (60) unadjusted multivariate analysis, they found the prevalence of suicide ideation to be over four times greater in postpartum women who experienced two or more stressful life events during pregnancy, but this was no longer significant after adjustment for all significant correlates. The authors also failed to provide any information regarding which life events were considered stressful. It should also be noted that Tavares et al.'s (60) methodological quality was relatively poor (QATSDD rating of 45%). Gelabert et al. (59) used the *St Paul Ramsey Life Experience Scale* (68) to assess the impact of stressful life events, experienced during pregnancy, in six categories: primary support, social environment, housing, work, health, and economy. The authors treated stressful life events as a dichotomous variable whereby a score of at least two, in one or more of the categories, was classed as presence of stressful life events. Women who reported the presence of stressful life events were 88% more likely to experience self-harm ideation at some point during the first 32 weeks following childbirth (59). Gavin et al. (62) used the *Prenatal Psychosocial Profile* (69) which assesses the extent to which 11 events are causing stress, such as financial problems, feeling generally "overloaded" and current abuse. The authors found high levels of psychosocial stress experienced during pregnancy increased the odds of experiencing suicide ideation during pregnancy threefold. Focussing their investigations on parenting stress, Paris et al. (64) categorised a sample of 32 women with postpartum depression into "low" and "high" suicidality groups and noted that the high suicidality group perceived overall parenting as significantly more stressful.

With regards to suicidal ideation and attempts, Mezey et al. (53) reported data that show a significant association between lifetime exposure to traumatic events (e.g., life threatening illness, physical assault, imprisonment) and suicide ideation only and suicide attempts, in a sample of 200 pregnant and postpartum women. Palfreyman (65) assessed stress about debt specifically and found it was correlated with suicide ideation and behaviour in 1000 Sri Lankan pregnant women. With regards to posttraumatic stress symptoms, Maré et al. (39) reported those classed as "high risk" for suicidal ideation and behaviour were more likely to report post-traumatic stress than those who reported no suicidal ideation and behaviour in 748 pregnant and postpartum women.

Summary of Negative Life Events

Taken together, the studies (41–52, 54, 56–58) demonstrate that experiences of abuse, particularly physical abuse, are significantly associated with ideation and attempts when measured separately and collectively, and therefore may be involved in enabling a woman to transition from having suicidal thoughts alone to then act on those thoughts. Furthermore, the findings suggest a persistent psychological effect of abuse (e.g., lifetime IPV and childhood abuse) which can manifest during the perinatal period. There is some evidence to suggest experiencing stress during the perinatal period is associated with perinatal self-harm and suicide ideation and suicidal behaviour. However, the studies presented used different measures of perinatal stress and there was no consistency with regards to what constituted a stressful or traumatic life event, which limits the overall findings. Additionally, the life events measured may incorporate other psychosocial risk factors, such as IPV.

Social Factors

Twenty-eight studies investigated social factors which were clustered into three groups of risk factors related to mother-infant bond/mother-infant interactions, social support, and loneliness. The methodological quality ratings of these 28 studies ranged from 36 to 86%, with 19 of the studies considered "good" quality, 7 "high," and 2 "moderate."

Mother-Infant Bond and Interactions

Takegata et al. (70) conducted a longitudinal study of three time points (during third trimester of pregnancy, 5 days postpartum, one month postpartum) with 243 women. They found those with self-harm ideation at any time point scored significantly higher on the "rejection and fear" and "anger and restrictedness" subscales of the *Japanese Postpartum Bonding Questionnaire* (*PBQ*) (71) at 5 days postpartum, and significantly higher on the "lack of affection," "rejection and fear," and "anger and restrictedness" *PBQ* subscales at one month postpartum. In another longitudinal study of 545 women, Gordon et al.'s (72) high quality study discovered that women who reported self-harm ideation at baseline (within 3 weeks of first antenatal midwifery appointment) had *PBQ* scores 6.28 points higher (i.e., poorer perceived bond) than those who did not report self-harm ideation. However, this difference was no longer significant after adjustment for depression at baseline. Furthermore, women who reported baseline self-harm ideation were significantly more controlling and infants significantly more compulsive in mother-infant interactions at 3 months postpartum. Two cross-sectional studies that assessed suicidal ideation report evidence of impaired mother-infant bonding and interactions. Faisal-Cury et al. (73) found bonding impairment was associated with almost five times increased odds of suicide ideation 6–9 months after birth, even when postpartum depressive symptoms were controlled for. Paris et al. (64) rated mother-infant interactions in 32 women with postpartum depression. Participants were categorised as "low suicidality" if they scored below 12 on the *Postpartum Depression Screening Scale* (*PDSS*) (74), or "high suicidality" if they scored 12 or higher on the *PDSS*. Compared to mothers identified as "low suicidality," during unstructured interactions, the "high

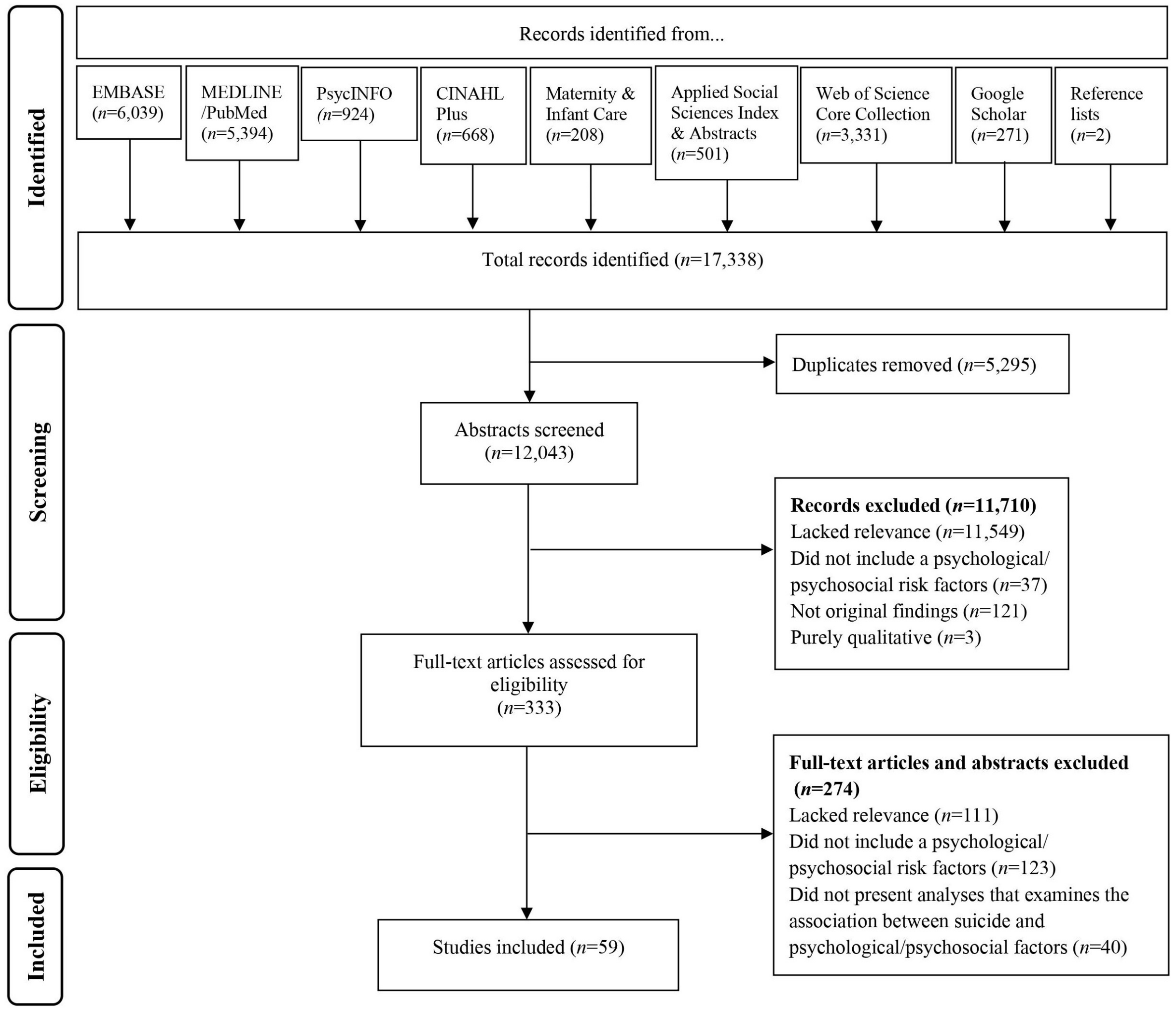


FIGURE 1 | Flowchart of paper selection based on PRISMA guidance.

suicidality" mothers were significantly less aware of, and less able to consistently respond to, their infants' social signals. Infants of the "high suicidality" mothers also exhibited less positive affect and more negative affect. However, the two groups behaved similarly in structured interactions (e.g., asking the parent to guide the infant to follow a rattle). In contrast, in a cohort of 430 women, Kubota et al. (75) did not observe any group difference in the mother-infant bond between those who did and did not report self-harm ideation. However, it should be noted that Kubota et al. (75) measured mother-infant bonding during early pregnancy (mean 5.3 months gestation) using the *Mother Infant Bonding Questionnaire* (76) which was designed to be used postnatally.

Martini et al. (46) investigated suicidal ideation and attempts and the mother-infant bond in a cohort of 306 mothers: mothers who reported perinatal suicidal behaviour indicated higher mean scores of bonding impairment with their infants compared to mothers with no perinatal suicidal behaviour, although these differences were not statistically significant.

Social Support

Twenty-four studies (28, 40, 43, 44, 46, 48, 54–56, 59–61, 65, 67, 75, 77–85) assessed social support, yet the types of social support (e.g., material, emotional, informational, affective, and social interaction) and sources of support (e.g., partner, family, friends, and neighbours) varied across these studies. With regards to the 16 studies that measured social support and suicide/self-harm ideation alone, seven studies found no association (48, 67, 80–82, 84, 85), six (40, 43, 44, 59–61) reported a significant association (i.e., poor social support increased the odds), and three (75, 78, 79) found a significant protective effect of social support (i.e., good-quality support reduced the odds). Both Doi and Fujiwara (44) and Tavares et al. (60) reported that a lack of social support was significantly associated with self-harm and suicide ideation, respectively, in their initial analyses but this association was no longer significant in their adjusted analyses. It should be noted that neither Doi and Fujiwara (44) nor Tavares et al. (60) provide information about the items used to assess support. In their longitudinal study, Gelabert et al. (59) measured

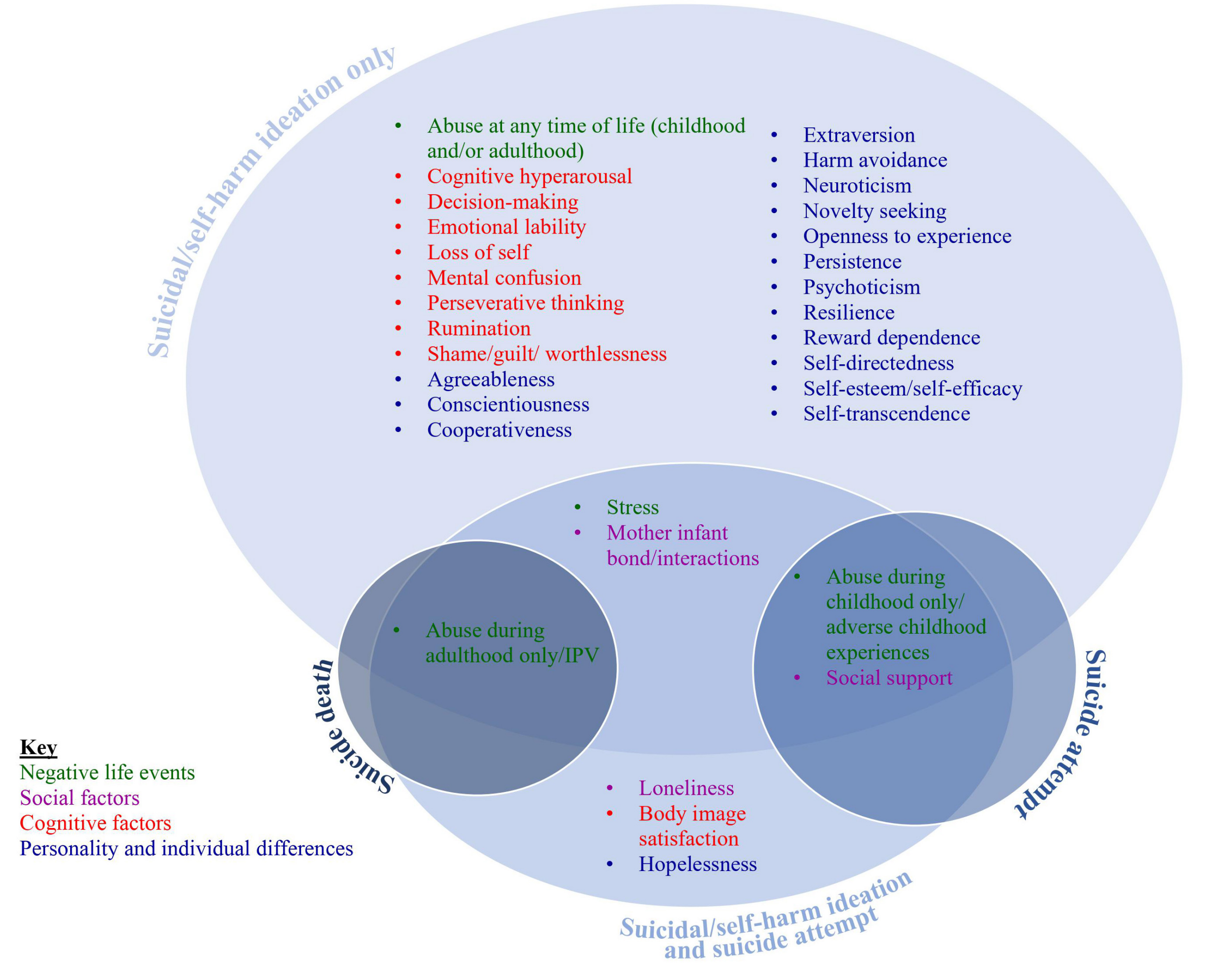


FIGURE 2 | Diagrammatic illustration of the included risk factors and suicide outcomes.

functional social support (e.g., "*I have someone who goes to the doctor with me*") and found that the women who reported self-harm ideation had significantly lower mean scores of functional social support at 2–3 days postpartum and at 8 weeks postpartum but no difference in scores at 32 weeks postpartum. Peltzer (79) used three items from the *Social Supportood Questionnaire* (86): "*If I were sick and needed someone to take me to a doctor, I would have trouble finding someone,*" "*I feel that there is no one I can share my most private concerns and fears,*" and "*I feel a strong emotional bond with at least one other person.*" In their sample of 580 postpartum women, the authors reported that the odds of self-harm ideation were reduced by 19% in women who perceived having social support, whilst being accompanied by the baby's father to antenatal care was not found to affect the odds of self-harm ideation postpartum. In Giallo et al.'s (43) longitudinal study, a sample of 1,507 women at 12 months postpartum were asked if they would have liked more emotional support (e.g., someone to talk to about how they were feeling) over the past month. The sample were categorised into a "persistent self-harm ideation" class (38–43% probability of endorsing self-harm ideation across the study time point) and a "minimal self-harm ideation" class (1–3% probability of endorsing self-harm ideation across the study time points). Women who reported a need for emotional support at 12 months postpartum were almost twice as likely to be a member of the "persistent self-harm ideation" class.

Seven studies investigated social support and suicidal ideation and behaviour (46, 54–56, 65, 77, 83) with mixed findings, although it should be noted that most of these studies used a different self-report measure of social support. Only Belete and Misgan (56) and Belete et al. (77) used the same measure of social support, namely, the *Oslo-3 Social Support Scale* (87) which asks: "*how many people are you so close to that you can count on them if you have great personal problems?,*" "*how much interest and concern do people show in what you do?,*" and "*how easy is it to get practical help from neighbours if you should need it?.*" Belete and Misgan (56) assessed a sample of 988 postpartum women and did not find a significant association between social support and suicidal behaviour. However, in their sample of 738 pregnant women, Belete et al. (77) found that participants who reported poor social support were over three times more likely to indicate having seriously considered attempting suicide while pregnant. It is therefore unsurprising that Belete et al. (77) also reported that poor social support was correlated with suicide attempts during pregnancy. Onah et al. (83) used the *Multidimensional Scale of Perceived Social Support* (88), which consists of 12 items that measure closeness and emotional support from friends, family and a significant other (e.g., "*I can count on my friends when things go wrong,*" "*my family is willing to help me make decisions,*" "*there is a special person with whom I can share my joys and sorrows*"). In their sample of 376 pregnant women, the authors found that perceived support from a significant other did not have an effect on suicide ideation only, but perceived support was found to be associated with a reduced likelihood of suicide behaviour (defined by the authors as those who had suicidal thoughts and had proceeded to plan or prepare or attempt suicide). Martini et al.'s (46) longitudinal study used the *F-SozU* (89) which assesses general perceived or anticipated social support, excludes any support from healthcare professionals and includes items such as "*when I am sick, I can ask friends/relatives to handle important things for me without hesitation*" and "*there are people who accept me the way I am without reservations.*" In their cohort of 306 women, Martini et al. (46) reported that higher levels of perceived social support during pregnancy decreased the odds of perinatal suicidal behaviour, although ideation, planning and attempts were combined and therefore we cannot discern the specific effect of social support upon these unique outcomes.

As described previously, Gressier et al. (28) used the French Network of MBUs to retrospectively assess 1,439 women for suicide attempts and of these, 105 attempted suicide during the postpartum period and 49 had made attempts during pregnancy. Women who attempted suicide during pregnancy reported poor family/social support significantly more than those who attempted suicide when postpartum and those who did not attempt suicide during the perinatal period. However, no independent effect of family/social support on suicide attempt was observed in the logistic regression. The logistic regression did, however, reveal that tobacco and alcohol use significantly increased the odds of suicide attempts during pregnancy. The regression also identified history of miscarriage as having a significant independent association on suicide attempts during pregnancy. Gressier et al. (28) suggested that pregnancy ignited the previous trauma of losing a baby. Overall, Gressier et al.'s (28) results indicated that a mother's distress had a greater effect on a woman's inclination to act on suicidal thoughts than poor social support.

Loneliness

One study assessed loneliness: Kugbey et al. (54) failed to find a bivariate or multivariate association between loneliness and current suicidal behaviour (i.e., a composite score of suicidal ideation, suicide plans, and suicide attempts) in 214 pregnant women. However, they used just one item to measure loneliness without providing any more information on this item.

Summary of Social Factors

In the six studies that measured the mother-infant bond and/or interactions, the results start to advocate a link between the mother-infant relationship and ideation but not with suicidal behaviour. Several studies provided evidence that social support was important during the early postpartum period, when a woman appeared to be particularly vulnerable, and that emotional support demonstrated an association with self-harm ideation. Whilst the association between social support and ideation appears less strong, the studies that measured support and suicidal behaviour provided stronger evidence for an association. When viewed in combination, these 24 studies start to suggest that support may be more pertinent in the decision to act on thoughts rather than a trigger for the initial ideation.

Cognitive Factors

A range of cognitive factors was investigated in seven studies (54, 63, 64, 82, 90–92). These studies' methodological quality ratings ranged from 33 to 71%, with six of the seven studies considered "good" quality and one study was of "moderate" quality.

In a sample of 100 pregnant women, the association between decision-making function, assessed using the *Iowa Gambling Task* (93) and self-harm ideation was investigated (90). The participants were allocated to one of three groups: those who reported self-harm ideation, those who scored highly for depressive symptoms on the *EPDS* (33), but reported no self-harm ideation, and those who reported no self-harm ideation and did not score highly on the *EPDS*. Women in the self-harm ideation group demonstrated impaired decision-making compared to the other two groups in the fifth block of the task. Paris et al. (64) clustered a sample of 32 women with postpartum depression into "low" and "high" suicidality groups. When these two groups were compared, the mothers in the high suicidality group reported feeling significantly more emotionally labile, mentally confused and experienced a greater loss of self. Paris et al. (64) was the only study to investigate these cognitive factors and although the study employed a very small sample, it was rated high for methodological quality. Two studies explored rumination as a risk factor for perinatal self-harm ideation (63, 92) and both found negative perinatal-specific nocturnal rumination (i.e., having stressful thoughts about the pregnancy/infant while attempting to fall asleep) was associated with increased odds of self-harm ideation. In their more recent study, Kalmbach et al. (63) also found those who reported nocturnal cognitive hyperarousal (e.g., not being able to shut off thoughts when trying to fall asleep) at baseline were over 11 times more likely to report new onset self-harm ideation. However, this same study found perseverative thinking during the daytime was not associated with self-harm ideation, suggesting that cognitive-emotional dysregulation specifically at night plays a unique role in self-harm ideation.

Guilt, shame and worthlessness were assessed by three studies [guilt/shame (64); worthlessness/guilt (91); maltreatment-related shame (82)] and all three reported significant associations between their assessment of worthlessness/guilt/shame and suicidal ideation. Muzik et al. (82) identified maltreatment-related shame as the only risk factor in their logistic regression to be independently associated with the presence of suicidal ideation at 4 and 12 months postpartum; other factors included in the analysis, which did not demonstrate independent associations, were resilience, household income and marital status. As previously described, Paris et al. (64) categorised their sample as either "high suicidality" or "low suicidality." On the guilt/shame subscale of the *PDSS* the "high suicidality" mothers scored significantly higher than the "low suicidality" mothers. Similarly, Benute et al. (91) divided their sample into those who reported suicidal ideation and those who did not and discovered that significantly more women in the suicidal ideation group endorsed feeling worthless/guilty; however, the authors only used one item to assess worthlessness/guilt.

Personality and Individual Differences

Ten of the included studies assessed 16 different risk factors that could be categorised as personality and individual differences. Eight of the studies were rated as "good" quality and two were "high" quality with QATSDD scores ranging from 57 to 81%.

The association of self-esteem/self-efficacy was investigated with mixed results. Using the *Rosenberg Self-esteem Scale* (94), Islam et al. (48) reported high self-esteem to be protective against self-harm ideation postpartum, whereas Shi et al. (61) found those with self-harm ideation had relatively higher self-esteem, but self-esteem did not predict presence/absence of self-harm ideation before birth or postpartum. Paris et al. (64) used the *Maternal Self-Report Inventory-Short Form* (95) which assesses self-esteem and self-perceptions of parenting and motherhood over five domains: the mother's perceived caretaking abilities, general ability and preparedness for motherhood, acceptance of her baby, expectations of a positive relationship with baby and feelings towards the birth. Mothers in the "high suicidality" group had lower total self-esteem scores, perceived they were less prepared for motherhood and expected a poorer relationship with their infants compared to those in the "low suicidality" group. Similarly, Bodnar-Deren et al. (80) investigated self-efficacy using five items that assessed perceived ability to care for the baby, other family members and the household. Self-efficacy showed a protective effect with almost 50% lower odds of suicide ideation in a sample of 1073 postpartum women.

Several personality traits were assessed by Takegata et al. (70) using the Japanese version of the *Temperament and Character Inventory* (96). In their cohort of 243 women, those who reported self-harm ideation at any time point (i.e., during the third trimester of pregnancy, 5 days postpartum or one month postpartum) had significantly lower mean scores for cooperativeness and self-directedness when compared to women who never reported self-harm ideation. Neither novelty-seeking, harm avoidance, persistence, reward-dependence nor self-transcendence demonstrated a significant association with self-harm ideation.

Neuroticism was assessed by three studies with mixed findings. Both Duan et al. (97) and Gelabert et al. (59) chose the *Eysenck Personality Questionnaire* (98) to measure neuroticism: Duan et al. (97) found neuroticism did not predict postpartum suicidal ideation, whereas Gelabert et al. (59) reported that neuroticism was associated with a marginal increase in the odds of self-harm ideation of 3% throughout the first 32 weeks following birth. Enătescu et al. (99) used the *NEO-Five Factor Inventory* (100) with a cohort of 202 women and reported higher levels of neuroticism in postnatal women with self-harm ideation than in those without, although neuroticism did not predict self-harm ideation.

Like neuroticism, there were mixed findings for an association between extraversion and self-harm ideation. Extraversion was negatively associated with self-harm ideation 2–3 days postpartum in a longitudinal study of 1795 women (59), whereas no differences in extraversion was found between those who reported self-harm ideation and those who did not, neither during pregnancy nor postpartum (99).

Only one study investigated the association between a personality/individual difference and suicide ideation and behaviour. In a sample of veterans, Szpunar et al. (66) did not find a significant correlation between hopelessness and suicidal behaviour. However, none of the participants reported a severe level of hopelessness and the lack of associations found may be due to the low variability of hopelessness reported and the very small sample of 28 women.

DISCUSSION

This review aimed to summarise the psychological and psychosocial risk factors associated with maternal suicide outcomes during the perinatal period and describe how these risk factors vary across self-harm and suicidal ideation, attempts and deaths. This was a comprehensive review of 59 studies making a novel contribution to the literature by synthesising findings of studies with a range of designs, conducted in low-, middle- and high-income countries with samples of both pregnant and postpartum women.

Most of the included studies used cross-sectional designs and therefore causal relationships could not be inferred. Regression models were commonly used to determine the shared relationships between the risk factors and suicide outcomes, and this paves the way for the use of alternative study designs and analyses in the future to investigate how apparently pertinent factors affect the trajectory of suicidal ideation and behaviour during the perinatal period. Only three studies measured suicide ideation and attempts separately and provided sufficient information to allow for a comparison of risk factors between these suicide outcomes (45, 52, 53), the other ten studies that investigated ideation and attempts either used a combined measure, arriving at one score for "suicidal behaviour" or did not provide sufficient data to make a comparison between the suicide outcomes. Comparing those women who experience suicidal ideation alone with those who have attempted suicide provides an opportunity to tease out which risk factors are unique and specific to suicide ideation or suicide attempts. It can be assumed that people who attempt suicide are also likely to experience suicidal ideation, and so only comparing those who experience suicidal ideation or attempt suicide with those who do not is no longer sufficient. Klonsky and May's (101) ideation to action framework highlighted the need to better understand the distinct pathways and processes, i.e., what factors specifically contribute to the development of maternal suicide ideation, and then separately, what factors specifically promote the progression from ideation to maternal suicide attempts. That being said, all three studies that include specific measures of ideation and behaviour demonstrated the same risk factors for those with suicide ideation and those who had attempted suicide. Much of the identified research investigated psychosocial risk factors, such as negative life events and social factors, whereas there were far fewer studies that assessed cognitive factors and individual differences. This could be due to ease of measurement; for example, negative life events and social factors may be assessed with very few yes/no questions, whereas cognitive factors and personality traits require more complex validated measures. Furthermore, the psychological effects of negative life events and social factors have received greater research attention compared to those of the myriad cognitive factors and individual differences. It is important to bear in mind that many of the identified factors are not mutually exclusive. For example, IPV can cause long-term damage to the victim's self-esteem (102), likewise feelings of guilt often persist into adulthood following abuse experienced during childhood (103). Therefore, determining pathways between psychological and psychosocial risk factors and suicidal behaviour in women is incredibly complex, but disentangling this web of risk factors is crucial for the development of assessments to identify and interventions to target maternal suicide ideation and behaviour.

To measure suicidal or self-harm ideation, 24 studies used the *EPDS* (33) item 10 which reads "*the thought of harming myself has occurred to me*" in the past 7 days, which may be interpreted in one of three ways: self-harm with suicide intent (i.e., suicide ideation), intentional self-harm with no suicide intent, or unintentional self-harm (e.g., accidentally falling down the stairs). Authors using this item often failed to provide a measure of severity of suicide/self-harm ideation. The widespread use is understandable because the *EPDS* is commonly used in primary care and maternity services to identify depressive disorders during pregnancy and the postpartum period (104). However, we cannot be sure of how respondents interpreted this item and whether self-harm with or without suicide intent was measured in the 24 studies. Current suicidal ideation screening instruments validated for use during the perinatal period, including the use of *EPDS* item 10, were primarily developed to assess maternal depression (25). It is therefore essential that future research efforts focus on designing a suicidal ideation measure for use during the perinatal period to assess suicidal thoughts, thoughts of self-harm with suicidal intentions and severity of suicidal thoughts.

Negative Life Events

In multivariate models, experiences of IPV, particularly physical abuse, are associated with ideation, attempts and death by suicide, and the more frequently abuse is experienced, the greater the odds of ideation and attempt. This is interesting because the findings start to suggest that intimate partner abuse could trigger ideation and is then involved in a woman's decision to act on those thoughts of self-harm and suicide. Similarly, childhood abuse was a strong predictor of both ideation and attempts. This dose-response relationship between adult and childhood trauma and suicide risk has been found by large-scale studies in the general population (e.g., 105, 106). There is also evidence to suggest that the relationship between depressive symptoms and IPV is bidirectional, with women who experience IPV at increased risk of depression, and women who report depressive symptoms being more likely to experience IPV, but curiously this finding has not been found in men (107). This might indicate the profundity of the effect of abuse on women. Experiencing violence as a perinatal woman may pose a particular risk because the stable and safe environment for her infant and herself, while she is very vulnerable, is threatened.

It is not necessarily surprising that abuse results in the victim feeling suicidal. Nevertheless, the findings included in this review solidify that abuse experienced in childhood and adulthood is prevalent around the world and that the effects can persist and manifest during this important period of a woman and infant's life. It is essential for suicide prevention during the perinatal period to identify and help women who have experienced or are currently experiencing abuse. The volume of studies and the strength of the findings also reinforce the need to identify which psychological factors are most affected when abuse is experienced, which could then help with the development of an intervention to target these mediating psychological mechanisms in order to reduce suicidal thoughts and behaviour.

Social Factors

The review findings suggest that support may play a lesser part in the development of suicidal ideation but provides some evidence that lack of support is associated with suicidal behaviour. Lack of support appeared to be particularly risky during pregnancy, which could demonstrate that women are especially aware of the support around them during pregnancy, fearful if there is no support network in place for the baby's arrival, and of being unsupported when coping with the major changes a baby will bring. Previous research has demonstrated that lack of social support is significantly associated with postnatal depression and health-related quality of life (108). Reid and Taylor (109) found that support from an intimate partner and from friends and family was protective against postpartum depression but was insufficient to reduce the effect of exposure to stress (e.g., sexual assault, IPV, parenting-related stress), found to be significantly associated with postpartum depression. Qualitative research conducted with women living with HIV and perinatal depression reported that women described social support as being composed of interaction, encouragement, and "offloading"/sharing worries (110). Interaction served as a distraction from their worries, encouragement was helpful to alleviate depressive feelings because it gave women strength, and "offloading" was viewed as an opportunity for women to alert others of their suicidal thoughts (110). Not only do these findings highlight the importance of social support for perinatal women because it alleviated perinatal depression using different functions, but it also identified offloading as particularly critical for preventing maternal suicide.

The review identified some evidence to suggest an association between self-harm and suicidal ideation and the mother-infant bond and interactions. An association between poor mother-infant bonding and postpartum depressive symptoms has been well-documented (e.g., 111, 112). However, how bonding contributes to maternal mood and how depressive symptoms contribute to poor bonding is not well understood. Understandably, the mother-infant bond does not feature in models of suicidal behaviour based on research conducted with the general population, further highlighting the need to develop a model of suicidal behaviour specific to the perinatal period.

Cognitive Factors

A variety of cognitive factors were investigated. Decision-making was found to be impaired in those who reported self-harm ideation after five blocks of the decision-making task, suggesting an impairment only occurs over time and/or after making many decisions. Interestingly, one study suggests impaired decision-making may influence the occurrence of interpersonal problems and increase the risk of problematic affective relationships. Worthlessness, guilt, emotional lability, mental confusion, loss of self, emotional lability, and shame were found to be associated with suicide and self-harm ideation. Feeling worthless is a common symptom of depression (113, 114) and has been shown to predict lifetime suicide attempts in those who have experienced serious trauma (e.g., a physical or sexual assault) but not in those who have not experienced trauma (115). This finding suggests that worthlessness can mediate the relationship between negative life events (e.g., IPV and childhood abuse) and suicidal behaviour. With regards to loss of self, there is very limited empirical research that investigates this factor and suicide. However, qualitative research has linked "loss of former identity" to postnatal depression (116).

Personality and Individual Differences

The one study that investigated hopelessness, failed to find an association with suicidal ideation and behaviour, although the sample reported low variability of hopelessness (66). Interestingly, findings in the general population suggest that hopelessness is involved in the development of suicidal ideation but it is not useful for predicting the transition from ideation to suicide attempts and death (32).

Self-esteem, defined as one's judgement of self-worth and others' judgement of oneself (117), and self-efficacy, defined as one's perceived ability and motivation (117), were both investigated as risk factors for suicide. Findings start to suggest that high levels of self-esteem and self-efficacy could be protective and low levels could be a risk for self-harm and suicide ideation. In psychiatric outpatients, other-based self-esteem (i.e., beliefs about others' judgement of oneself) was found to be the strongest predictor of suicidal ideation after controlling for depression and hopelessness (118). The postpartum period is a time of learning new skills while developing the new identity of "mother" and it is therefore unsurprising that a woman's perceived ability to be a mother and care for her infant, or self-efficacy, could impact on a mother's mental wellbeing. Previous research has identified that support from family and friends (119) and marital support (120) are positively associated with maternal self-efficacy postpartum and negatively associated with maternal depression. Although risk factors were treated as separate entities for the purposes of this review, these associations between support and postpartum self-efficacy highlight the interconnectedness of the risk factors for maternal suicide, and in turn, the complex nature of teasing out the causal and mediational relationships.

Strengths and Limitations of the Review

This novel review is the first to focus on psychological and psychosocial factors rather than include any or all correlates of maternal suicide, many of which cannot be targeted by psychological interventions. Strengths of this review include the use of a prospective protocol and adherence to the PRISMA statement to ensure the review was conducted and the findings reported in a transparent manner, as well as no limitations on language and date of publication, which resulted in a comprehensive and thorough review of studies conducted around the world. Furthermore, an independent reviewer also conducted reliability checks to ensure rigour of study screening, selection, data extraction and quality appraisal of the identified studies.

The review is somewhat limited by the narrative, rather than statistical, synthesis of results. Most of the risk factors were investigated by few studies that employed heterogenous designs and therefore a meta-analysis would not have been suitable for the majority of risk factors. The intentionally inclusive sampling strategy resulted in the inclusion of studies with wide ranging samples, such as veterans (51, 66) and HIV-positive women (40, 49, 79, 121, 122). Although it is important that suicidal ideation and behaviours are investigated in all groups of women, this may have resulted in the inclusion of some risk factors that will not apply to all women, such as military sexual trauma. Finally, only two papers investigated suicide deaths (57, 58) and these two studies only investigated IPV. It is therefore difficult to draw conclusion about the impact of different risk factors on this most severe suicide outcome.

Implications and Future Research

Four implications for identification of women at increased risk of suicide and potential methods for intervening to reduce suicidal ideation and behaviour in clinical practice are evident. The review findings suggest it is very important that perinatal women who have experienced domestic violence whether it be from an intimate partner or another member of the household, or childhood abuse, are identified. Therefore, ensuring midwives routinely ask women about childhood abuse and domestic violence during the booking appointment is the first clinical implication of this research. Secondly, those who indicate they have had these negative experiences should be helped to leave the abuse if ongoing and offered psychological support. The occurrence of negative life events cannot be modified, but the psychological sequelae could be, therefore future research should aim to determine which psychological factors are affected by abuse and how they trigger or sustain suicidal ideation and behaviour. The review also suggests ensuring women have emotional and practical support, whether it be from an intimate partner or friends and family, especially during pregnancy, may help to reduce the likelihood of suicidal behaviour. Therefore, ensuring midwives help women identify their supportive network during pregnancy offers a third clinical implication. Fourthly, midwives should also facilitate a mother to strengthen her access to support if required.

The theoretical basis for which psychological and psychosocial factors are involved in the development of suicidal ideation and behaviour is derived from research with the general population. We know that the perinatal period is unique in that women are facing many new changes and challenges and that perinatal suicides differ to non-perinatal suicides [e.g., perinatal suicides often occur through more violent means (13–15)]. Thus, to establish psychological and psychosocial factors involved in perinatal suicide, research needs to focus specifically on perinatal samples.

Many of these correlational findings are mixed and do not consider that women's situations vary dramatically, therefore missing the more important nuances that could impact on a woman's risk of suicide. Qualitative approaches offer more flexibility and scope to investigate what could lead a woman to feeling suicidal during the perinatal period and help untangle the web of interconnected risk factors. There are many other psychological factors known to be associated with suicide risk in other populations, such as impulsivity and optimism (32), which have not been assessed by any of the included studies. Future qualitative research could also investigate potential risk factors which have not been investigated previously in the perinatal population.

CONCLUSION

This novel review was the first to focus on psychological and psychosocial factors rather than include any or all correlates of maternal suicide, many of which cannot be targeted by psychological interventions. There was strong evidence to indicate that abuse, either experienced recently or during childhood, is associated with suicide ideation, attempted suicide and death. There was also convincing evidence that a lack of social support is particularly important during the perinatal period and was significantly associated with suicidal behaviour. Clinically, investigating the role of psychological and psychosocial factors in the development of suicidal ideation and behaviour is essential to the generation of assessments and interventions to identify and reduce maternal suicidal ideation and behaviour.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

HR, AW, and DP developed the initial idea for this review. HR undertook the systematic review, under the supervision of AW, DP, and DE. HR wrote the first draft of the manuscript. All authors contributed to the article and have approved the final manuscript.

FUNDING

This work was conducted as part of the HR's Ph.D. research funded by the Medical Research Council Doctoral Training Partnership (MR/N013751/11). The funders had no role in the study design, collection, analysis or interpretation of the data, writing the manuscript, or the decision to submit the manuscript for publication.

ACKNOWLEDGMENTS

We would like to thank Claire Hodkinson at the University of Manchester library for providing guidance and advice on developing the search strategy, Richard Green for independently screening, data extracting, and quality appraising studies.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.765118/full#supplementary-material

REFERENCES

1. Knight M, Bunch K, Tuffnell D, Shakespeare J, Kotnis R, Kenyon S, et al. *Saving Lives, Improving Mothers' Care – Lessons Learned to Inform Maternity Care from the UK and Ireland Confidential Enquiries into Maternal Deaths and Morbidity 2016-18*. Oxford: National Perinatal Epidemiology Unit, University of Oxford (2020).
2. Humphrey MD. Maternal mortality trends in Australia. *Med J Aust.* (2016) 205:344–6. doi: 10.5694/mja16.00906
3. Guillard V, Gressier F. Suicidalité en périod périnatale. *Presse Méd.* (2017) 46:565–71. doi: 10.1016/j.lpm.2017.05.018
4. Donati S, Maraschini A, Lega I, D'Aloja P, Buoncristiano M, Manno V, et al. Maternal mortality in Italy: results and perspectives of record-linkage analysis. *Acta Obstet Gynecol Scand.* (2018) 97:1317–24. doi: 10.1111/aogs.13415
5. Vangen S, Bødker B, Ellingsen L, Saltvedt S, Gissler M, Geirsson RT, et al. Maternal deaths in the Nordic countries. *Acta Obstet Gynecol Scand.* (2017) 96:1112–9. doi: 10.1111/aogs.13172
6. Rao WW, Yang Y, Ma TJ, Zhang Q, Ungvari GS, Hall BJ, et al. Worldwide prevalence of suicide attempt in pregnant and postpartum women: a meta-analysis of observational studies. *Soc Psychiatry Psychiatr Epidemiol.* (2021) 56:711–20. doi: 10.1007/s00127-020-01975-w

7. Lindahl V, Pearson J, Colpe L. Prevalence of suicidality during pregnancy and the postpartum. *Arch Womens Ment Health.* (2005) 8:77–87. doi: 10.1007/s00737-005-0080-1
8. Busch T, Kimble CS. Grieving children: are we meeting the challenge? *Pediatr Nurs.* (2001) 27:414–24.
9. Pfeffer CR, Karus D, Siegel K, Jiang H. Child survivors of parental death from cancer or suicide: depressive and behavioral outcomes. *Psychooncology.* (2000) 9:1–10. doi: 10.1002/(SICI)1099-1611(200001/02)9:13.0.CO;2-5
10. Tsuchiya KJ, Agerbo E, Mortensen PB. Parental death and bipolar disorder: a robust association was found in early maternal suicide. *J Affect Disord.* (2005) 86:151–9. doi: 10.1016/j.jad.2005.01.006
11. Lieb R, Bronisch T, Höfler M, Schreier A, Wittchen HU. Maternal suicidality and risk of suicidality in offspring: findings from a community study. *Am J Psychiatry.* (2005) 162:1665–71. doi: 10.1176/appi.ajp.162.9.1665
12. Mebrahtu H, Sherr L, Simms V, Weiss HA, Rehman AM, Ndlovu P, et al. Effects of maternal suicidal ideation on child cognitive development: a longitudinal analysis. *AIDS Behav.* (2020) 24:2421–9. doi: 10.1007/s10461-020-02802-8
13. Grigoriadis S, Wilton AS, Kurdyak PA, Rhodes AE, VonderPorten EH, Levitt A, et al. Perinatal suicide in Ontario, Canada: a 15-year population-based study. *CMAJ.* (2017) 189:E1085–92. doi: 10.1503/cmaj.170088
14. Khalifeh H, Hunt IM, Appleby L, Howard LM. Suicide in perinatal and non-perinatal women in contact with psychiatric services: 15 year findings from a UK national inquiry. *Lancet Psychiatry.* (2016) 3:233–42. doi: 10.1016/S2215-0366(16)00003-1
15. Lega I, Maraschini A, D'Aloja P, Andreozzi S, Spettoli D, Giangreco M, et al. Maternal suicide in Italy. *Arch Womens Mental Health.* (2020) 23:199–206. doi: 10.1007/s00737-019-00977-1
16. Dehara M, Wells MB, Sjöqvist H, Kosidou K, Dalman C, Wallin AS. Parenthood is associated with lower suicide risk: a register-based cohort study of 1.5 million Swedes. *Acta Psychiatr Scand.* (2021) 143:206–15. doi: 10.1111/acps.13240
17. Cerel J, Frey LM, Maple M, Kinner DG. Parents with suicidal behaviour: parenting is not always protective. *J Child Fam Stud.* (2016) 25:2327–36. doi: 10.1007/s10826-016-0377-x
18. Howard LM, Molyneaux E, Dennis C, Rochat T, Stein A, Milgrom J. Non-psychotic mental disorders in the perinatal period. *Lancet.* (2014) 384:15–21. doi: 10.1016/S0140-6736(14)61276-9
19. Howard LM, Khalifeh H. Perinatal mental health: a review of progress and challenges. *World Psychiatry.* (2020) 19:313–27. doi: 10.1002/wps.20769
20. Jones I, Chandra PS, Dazzan P, Howard LM. Bipolar disorder, affective psychosis, and schizophrenia in pregnancy and the post-partum period. *Lancet.* (2014) 384:1789–99. doi: 10.1016/S0140-6736(14)61278-2
21. Munk-Olsen T, Maegbaek ML, Johannsen BM, Liu X, Howard LM, di Florio A, et al. Perinatal psychiatric episodes: a population-based study on treatment incidence and prevalence. *Transl Psychiatry.* (2016) 6:e919. doi: 10.1038/tp.2016.190
22. Woody CA, Ferrari AJ, Siskind DJ, Whiteford HA, Harris MG. A systematic review and meta-regression of the prevalence and incidence of perinatal depression. *J Affect Disord.* (2017) 219:86–92. doi: 10.1016/j.jad.2017.05.003
23. Johannsen BMW, Larsen JT, Laursen TM, Bergink V, Meltzer-Brody S, Munk-Olsen T. All-cause mortality in women with severe postpartum psychiatric disorders. *Am J Psychiatry.* (2016) 173:635–42. doi: 10.1176/appi.ajp.2015.14121510
24. Bostwick JM, Pankratz VS. Affective disorders and suicide risk: a reexamination. *Am J Psychiatry.* (2000) 157:1925–32. doi: 10.1176/appi.ajp.157.12.1925
25. Gelaye B, Kajeepeta S, Williams MA. Suicidal ideation in pregnancy: an epidemiologic review. *Arch Womens Ment Health.* (2016) 19:741–51. doi: 10.1007/s00737-016-0646-0
26. Orsolini L, Valchera A, Vecchiotti R, Tomasetti C, Iasevoli F, Fornaro M, et al. Suicide during perinatal period: epidemiology, risk factors, and clinical correlates. *Front Psychiatry.* (2016) 7:138. doi: 10.3389/fpsyt.2016.00138
27. Ayre K, Gordon HG, Dutta R, Hodsoll J, Howard LM. The prevalence and correlates of self-harm in the perinatal period: a systematic review. *J Clin Psychiatry.* (2020) 81:19r12773. doi: 10.4088/JCP.19r12773
28. Gressier F, Guillard V, Cazas O, Falissard B, Glangeaud-Freudenthal NM, Sutter-Dallay A. Risk factors for suicide attempt in pregnancy and the post-partum period in women with serious mental illnesses. *J Psychiatr Res.* (2017) 84:284–91. doi: 10.1016/j.jpsychires.2016.10.009
29. Moher D, Liberati A, Tetzlaff J, Altman DG, Group TP. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med.* (2009) 6:e1000097. doi: 10.1371/journal.pmed.1000097
30. Methley AM, Campbell S, Chew-Graham C, McNally R, Cheraghi-Sohi SPICO. PICOS and SPIDER: a comparison study of specificity and sensitivity in three search tool for qualitative systematic review. *BMC Health Serv Res.* (2014) 14:579. doi: 10.1186/s12913-014-0579-0
31. Hanna B. Negotiating motherhood: the struggles of teenage mothers. *J Adv Nurs.* (2001) 34:456–64. doi: 10.1046/j.1365-2648.2001.01774.x
32. O'Connor RC, Nock MK. The psychology of suicidal behaviour. *Lancet Psychiatry.* (2014) 1:73–85. doi: 10.1016/S2215-0366(14)70222-6
33. Cox JL, Holden JM, Sagovsky R. Detection of postnatal depression: development of the 10-item Edinburgh postnatal depression scale. *Br J Psychiatry.* (1987) 150:782–6. doi: 10.1192/bjp.150.6.782
34. Howard LM, Flach C, Mehay A, Sharp D, Tylee A. The prevalence of suicidal ideation identified by the Edinburgh postnatal depression scale in postpartum women in primary care: findings from the RESPOND trial. *BMC Preg Childbirth.* (2011) 11:57. doi: 10.1186/1471-2393-11-57
35. Popay J, Roberts H, Sowden A, Petticrew M, Arai L, Rodgers M, et al. *Guidance on the Conduct of Narrative Synthesis in Systematic Reviews: A Product from the ESRC Methods Programme*. Lancaster: Lancaster University (2006).
36. Sirriyeh R, Lawton R, Gardner P, Armitage G. Reviewing studies with diverse designs: the development and evaluation of a new tool. *J Eval Clin Pract.* (2011) 18:746–52. doi: 10.1111/j.1365-2753.2011.01662.x
37. Gillham R, Wittkowski A. Outcomes for women admitted to a mother and baby unit: a systematic review. *Int J Womens Health.* (2015) 7:459–76. doi: 10.2147/IJWH.S69472
38. Farber EW, Herbert SE, Reviere SL. Childhood abuse and suicidality in obstetrics patients in a hospital- based urban prenatal clinic. *Gen Hosp Psychiatry.* (1996) 18:56–60. doi: 10.1016/0163-8343(95)00098-4
39. Maré KT, Pellowski JA, Koopowitz S-M, Hoffman N, van der Westhuizen C, Workman L, et al. Perinatal suicidality: prevalence and correlates in a south African birth cohort. *Arch Womens Ment Health.* (2021) 24:737–48. doi: 10.1007/s00737-021-01121-8
40. Knettel BA, Mwamba RN, Minja L, Goldston DB, Boshe J, Watt MH. Exploring patterns and predictors of suicidal ideation among pregnant and postpartum women living with HIV in Kilimanjaro, Tanzania. *AIDS.* (2020) 34:1657–64. doi: 10.1097/qad.0000000000002594
41. Sit D, Luther J, Buysse D, Dills JL, Eng H, Okun M, et al. Suicidal ideation in depressed postpartum women: associations with childhood trauma, sleep disturbance and anxiety. *J Psychiatr Res.* (2015) 66–67:95–104. doi: 10.1016/j.jpsychires.2015.04.021
42. Leeners B, Rath W, Block E, Gorres G, Tschudin S. Risk factors for unfavorable pregnancy outcome in women with adverse childhood experiences. *J Perinat Med.* (2014) 42:171–8. doi: 10.1515/jpm-2013-0003
43. Giallo R, Pilkington P, Borschmann R, Seymour M, Dunning M, Brown S. The prevalence and correlates of self-harm ideation trajectories in Australian women from pregnancy to 4-years postpartum. *J Affect Disord.* (2018) 229:152–8. doi: 10.1016/j.jad.2017.12.064
44. Doi S, Fujiwara T. Combined effect of adverse childhood experiences and young age on self-harm ideation among postpartum women in Japan. *J Affect Disord.* (2019) 253:410–8. doi: 10.1016/j.jad.2019.04.079
45. Levey EJ, Rondon MB, Sanchez S, Zhong QY, Williams MA, Gelaye B. Suicide risk assessment: examining transitions in suicidal behaviors among pregnant women in Peru. *Arch Womens Ment Health.* (2019) 22:65–73. doi: 10.1007/s00737-018-0884-4

46. Martini J, Bauer M, Lewitzka U, Voss C, Pfennig A, Ritter D, et al. Predictors and outcomes of suicidal ideation during peripartum period. *J Affect Disord.* (2019) 257:518–26. doi: 10.1016/j.jad.2019.07.040
47. Iyengar R, Bondade S, Raj R. A cross sectional hospital-based study of intimate partner violence and psychiatric comorbidity in pregnancy. *Arch Psychiatry Psychother*. (2020) 4:12–21. doi: 10.12740/APP/120441
48. Islam M, Broidy L, Mazerolle P, Baird K, Mazumder N, Zobair KM. Do maternal depression and self-esteem moderate and mediate the association between intimate partner violence after childbirth and postpartum suicidal ideation? *Arch Suicide Res.* (2019) 24:609–632. doi: 10.1080/13811118.2019.1655507
49. Rodriguez VJ, Mandell LN, Babayigit S, Manohar RR, Weiss SM, Jones DL. Correlates of suicidal ideation during pregnancy and postpartum among women living with HIV in rural South Africa. *AIDS Behav.* (2018) 22:3188–97. doi: 10.1007/s10461-018-2153-y
50. Fisher J, Tran TD, Biggs B, Dang TH, Nguyen TT, Tran T. Intimate partner violence and perinatal common mental disorders among women in rural Vietnam. *Int Health.* (2013) 5:29–37. doi: 10.1093/inthealth/ihs012
51. Gross GM, Kroll-Desrosiers A, Mattocks K. A longitudinal investigation of military sexual trauma and perinatal depression. *J Womens Health.* (2020) 29:38–45. doi: 10.1089/jwh.2018.7628
52. Asad N, Karmaliani R, Sullaiman N, Bann CM, McClure EM, Pasha O, et al. Prevalence of suicidal thoughts and attempts among pregnant Pakistani women. *Acta Obstet Gynecol Scand.* (2010) 89:1545–51. doi: 10.3109/00016349.2010.526185
53. Mezey G, Bacchus L, Bewley S, White S. Domestic violence, lifetime trauma and psychological health of childbearing women. *BJOG.* (2005) 112:197–204. doi: 10.1111/j.1471-0528.2004.00307.x
54. Kugbey N, Ayanore M, Doegah P, Chirwa M, Bartels SA, Davison CM, et al. Prevalence and correlates of prenatal depression, anxiety and suicidal behaviours in the Volta region of Ghana. *Int J Environ Res Public Health.* (2021) 18:5857. doi: 10.3390/ijerph18115857
55. Supraja TA, Thennarasu K, Satyanarayana VA, Seena TK, Desai G, Jangam KV, et al. Suicidality in early pregnancy among antepartum mothers in urban India. *Arch Womens Ment Health.* (2016) 19:1101–8. doi: 10.1007/s00737-016-0660-2
56. Belete H, Misgan E. Suicidal behaviour in postnatal mothers in northwestern Ethiopia: a cross-sectional study. *BMJ Open.* (2019) 9:e027449. doi: 10.1136/bmjopen-2018-027449
57. Adu A, Brown SV, Asaolu I, Sanderson W, Brown SW. Understanding suicide in pregnant and postpartum women, using the national violent death reporting system data: are there differences in rural and urban status? *Open J Obstet Gynecol.* (2019) 9:547–65. doi: 10.4236/ojog.2019.95054
58. Gold KJ, Singh V, Marcus SM, Palladino CL. Mental health, substance use and intimate partner problems among pregnant and postpartum suicide victims in the national violent death reporting system. *Gen Hosp Psychiatry*. (2012) 34:139–45. doi: 10.1016/j.genhosppsych.2011.09.017
59. Gelabert E, Gutierrez-Zotes A, Navines R, Labad J, Puyane M, Donadon M, et al. The role of personality dimensions, depressive symptoms and other psychosocial variables in predicting postpartum suicidal ideation: a cohort study. *Arch Womens Ment Health.* (2020) 23:585–93. doi: 10.1007/s00737-019-01007-w
60. Tavares D, Quevedo L, Jansen K, Souza L, Pinheiro R, Silva R. Prevalence of suicide risk and comorbidities in postpartum women in Pelotas. *Rev Bras Psiquiatr.* (2012) 34:270–6. doi: 10.1016/j.rbp.2011.12.001
61. Shi P, Ren H, Li H, Dai Q. Maternal depression and suicide at immediate prenatal and early postpartum periods and psychosocial risk factors. *Psychiatry Res.* (2018) 261:298–306. doi: 10.1016/j.psychres.2017.12.085
62. Gavin A, Tabb K, Melville J, Guo Y, Katon W. Prevalence and correlates of suicidal ideation during pregnancy. *Arch Womens Ment Health.* (2011) 14:239–46. doi: 10.1007/s00737-011-0207-5
63. Kalmbach DA, Ahmedani BK, Gelaye B, Cheng P, Drake CL. Nocturnal cognitive hyperarousal, perinatal-focused rumination, and insomnia are associated with suicidal ideation in perinatal women with mild to moderate depression. *Sleep Med.* (2021) 81:439–42. doi: 10.1016/j.sleep.2021.03.004
64. Paris R, Bolton RE, Weinberg M. Postpartum depression, suicidality, and mother-infant interactions. *Arch Womens Ment Health.* (2009) 12:309–21. doi: 10.1007/s00737-009-0105-2
65. Palfreyman A. Addressing psychosocial vulnerabilities through antenatal care – depression, suicidal ideation, and behavior: a study among urban Sri Lankan women. *Front Psychiatry.* (2021) 12:554808. doi: 10.3389/fpsyt.2021.554808
66. Szpunar MJ, Crawford JN, Baca SA, Lang AJ. Suicidal ideation in pregnant and postpartum women veterans: an initial clinical needs assessment. *Mil Med.* (2020) 185:e105–11. doi: 10.1093/milmed/usz171
67. Farias DR, de Jesus Pereira Pinto T, Teofilo MMA, Vilela AAF, Vaz JdS, Nardi AE, et al. Prevalence of psychiatric disorders in the first trimester of pregnancy and factors associated with current suicide risk. *Psychiatry Res.* (2013) 210:962–8. doi: 10.1016/j.psychres.2013.08.053
68. Paykel ES. Methodological aspects of life events research. *J Psychosom Res.* (1983) 27:341–52. doi: 10.1016/0022-3999(83)90065-X
69. Curry MA, Campbell RA, Christian M. Validity and reliability testing of the prenatal psychosocial profile. *Res Nurs Health.* (1994) 17:127–35. doi: 10.1002/nur.4770170208
70. Takegata M, Takeda S, Sakanashi K, Tanaka T, Kitamura T. Perinatal self-report of thoughts of self-harm, depressive symptoms, and personality traits: prospective study of Japanese community women. *Psychiatry Clin Neurosci.* (2019) 73:707–12. doi: 10.1111/pcn.12917
71. Ohashi Y, Kitamura T, Sakanashi K, Tanaka T. Postpartum bonding disorder: factor structure, validity, reliability and a model comparison of the postnatal bonding questionnaire in Japanese mothers of infants. *Healthcare.* (2016) 4:50–61. doi: 10.3390/healthcare4030050
72. Gordon H, Nath S, Trevillion K, Moran P, Pawlby S, Newman L, et al. Self-harm, self-harm ideation, and mother-infant interactions: a prospective cohort study. *J Clin Psychiatry.* (2019) 80:18m12708. doi: 10.4088/JCP.18m12708
73. Faisal-Cury A, Levy RB, Matijasevich A. The relationship between mother-child bonding impairment and suicidal ideation in São Paulo, Brazil. *Matern Child Health J.* (2021) 25:706–14. doi: 10.1007/s10995-020-03024-z
74. Beck CT, Gable RK. Postpartum depression screening scale: development and psychometric testing. *Nurs Res.* (2000) 49:272–82. doi: 10.1097/00006199-200009000-00006
75. Kubota C, Inada T, Shiino T, Ando M, Sato M, Nakamura Y, et al. The risk factors predicting suicidal ideation among perinatal women in Japan. *Front Psychiatry.* (2020) 11:441. doi: 10.3389/fpsyt.2020.00441
76. Kumar R. "Anybody's child": severe disorders of mother-infant bonding. *Br J Psychiatry.* (1997) 171:175–81. doi: 10.1192/bjp.171.2.175
77. Belete K, Kassew T, Demilew D, Zeleke TA. Prevalence and correlates of suicide ideation and attempt among pregnant women attending antenatal care services at public hospitals in southern Ethiopia. *Neuropsychiatr Dis Treat.* (2021) 17:1517–29. doi: 10.2147/ndt.s309702
78. Akram B, Ahmed MA, Maqsood F, Bibi B. Postpartum depression and suicidal ideation in new mothers with hearing loss: perceived social support as a moderator, a multicentre study. *J Pak Med Assoc.* (2020) 70:213–8. doi: 10.5455/jpma.11537
79. Peltzer K. Prevalence of suicidal ideation and associated factors among postpartum HIV-positive women in health facilities, South Africa. *J Psychol Afr.* (2015) 25:547–50. doi: 10.1080/14330237.2015.1124615
80. Bodnar-Deren S, Klipstein K, Fersh M, Shemesh E, Howell EA. Suicidal ideation during the postpartum period. *J Womens Health.* (2016) 25:1219–24. doi: 10.1089/jwh.2015.5346
81. Castro e Couto T, Brancaglion MY, Cardoso MN, Faria GC, Garcia FD, Nicolato R, et al. Suicidality among pregnant women in Brazil: prevalence and risk factors. *Arch Womens Mental Health.* (2016) 19:343–8. doi: 10.1007/s00737-015-0552-x
82. Muzik M, Brier Z, Menke RA, Davis MT, Sexton MB. Longitudinal suicidal ideation across 18-months postpartum in mothers with childhood maltreatment histories. *J Affect Disord.* (2016) 204:138–45. doi: 10.1016/j.jad.2016.06.037
83. Onah MN, Field S, Bantjes J, Honikman S. Perinatal suicidal ideation and behaviour: psychiatry and adversity. *Arch Womens Ment Health.* (2017) 20:321–31. doi: 10.1007/s00737-016-0706-5

84. Tabb KM, Huang H, Valdovinos M, Toor R, Ostler T, Vanderwater E, et al. Intimate partner violence is associated with suicidality among low-income postpartum women. *J Womens Health.* (2018) 27:171–8. doi: 10.1089/jwh.2016.6077
85. Zewdu LB, Reta MM, Yigzaw N, Tamirat KS. Prevalence of suicidal ideation and associated factors among HIV positive perinatal women on follow-up at Gondar town health institutions, Northwest Ethiopia: a cross-sectional study. *BMC Preg Childbirth.* (2021) 21:42. doi: 10.1186/s12884-020-03529-z
86. Brock D, Sarason I, Sarason B, Pierce G. Simultaneous assessment of perceived global and relationship-specific support. *J Soc Pers Relationsh.* (1996) 13:143–52.
87. Abiola T, Udofia O, Zakari M. Psychometric properties of the 3-item Oslo social support scale among clinical students of Bayero University Kano, Nigeria. *Malays J Psychiatry.* (2013) 22:32–41.
88. Zimet GD, Dahlem NW, Zimet SG, Farley GK. The multidimensional scale of perceived social support. *J Pers Assess.* (1988) 52:30–41.
89. Fydrich T, Sommer G, Tydecks S, Brähler E. Social support questionnaire (F-SozU): standardization of short form (K-14). *Z Med Psychol.* (2009) 18:43–8. doi: 10.1016/j.jclinepi.2014.11.003
90. Bao CQ, Xu L, Tang WN, Sun SY, Zhang WM, He JC, et al. Poor sleep and decision-making disturbance are associated with suicidal ideation in pre-natal depression. *Front Psychiatry.* (2021) 12:680890. doi: 10.3389/fpsyt.2021.680890
91. Benute GRG, Nomura RMY, Jorge VMF, Nonnenmacher D, Fraguas R, de Lucia MCS, et al. Risk of suicide in high risk pregnancy: an exploratory study. *Rev Associacao Med Bras.* (2011) 57:583–7. doi: 10.1590/s0104-42302011000500019
92. Kalmbach DA, Cheng P, Ong JC, Ciesla JA, Kingsberg SA, Sangha R, et al. Depression and suicidal ideation in pregnancy: exploring relationships with insomnia, short sleep, and nocturnal rumination. *Sleep Med.* (2020) 65:62–73. doi: 10.1016/j.sleep.2019.07.010
93. Bechara A, Tranel D, Damasio H. Characterization of the decision-making deficit of patients with ventromedial prefrontal cortex lesions. *Brain.* (2000) 123:2189–202. doi: 10.1093/brain/123.11.2189
94. Rosenberg M. *Society and the Adolescent Self-Image*. Princeton, NJ: Princeton University Press (1965).
95. Shea E, Tronick EZ. The maternal self-report inventory: a research and clinical instrument for assessing maternal self-esteem. *Theory Res Behav Pediatr.* (1988) 4:101–39.
96. Kijima N, Saito R, Takeuchi M. Cloninger's seven-factor model of temperament and character and the Japanese version of the temperament and character inventory (TCI). *Arch Psychiatr Diagn Clin Eval.* (1996) 7:379–99. doi: 10.1002/(sici)1097-4679(199812)54:8<1043::aid-jclp4>3.0.co;2-a
97. Duan Z, Wang Y, Tao Y, Bower JL, Yu R, Wang S, et al. Relationship between trait neuroticism and suicidal ideation among postpartum women in China: testing a mediation model. *J Affect Disord.* (2019) 256:532–5. doi: 10.1016/j.jad.2019.06.030
98. Eysenck S. *Eysenck Personality Questionnaire-Revised (EPQ-R) and Short Scale (EPQ-RS)*. London: Hooder & Stoughton (1985).
99. Enătescu I, Craina M, Gluhovschi A, Giurgi-Oncu C, Hogea L, Nussbaum LA, et al. The role of personality dimensions and trait anxiety in increasing the likelihood of suicide ideation in women during the perinatal period. *J Psychosom Obstet Gynecol.* (2020) 42:242–52. doi: 10.1080/0167482X.2020.1734790
100. Costa PT, McCrae RR. *NEO Personality Inventory (NEO-PI-R) and NEO Five-Factor Inventory (NEO-FFI) (Revised)*. Odessa, FL: Psychological Assessment Resources (1992).
101. Klonsky ED, May AM. Differentiating suicide attempters from suicide ideators: a critical frontier for suicidology research. *Suicide Life Threat Behav.* (2014) 44:1–5. doi: 10.1111/sltb.12068
102. Matheson FI, Daoud N, Hamilton-Wright S, Borenstein H, Pedersen C, O'Campo P. Where did she go? The transformation of self-esteem, self-identity, and mental well-being among women who have experienced intimate partner violence. *Womens Health Issues.* (2015) 25:561–9. doi: 10.1016/j.whi.2015.04.006
103. Street AE, Gibson LE, Holohan DR. Impact of childhood traumatic events, trauma-related guilt, and avoidant coping strategies on PTSD symptoms in female survivors of domestic violence. *J Traum Stress.* (2005) 18:245–52. doi: 10.1002/jts.20026
104. Gibson J, McKenzie-McHarg K, Shakespeare J, Price J, Gray R. A systematic review of studies validating the Edinburgh postnatal depression scale in antepartum and postpartum women. *Acta Psychiatr Scand.* (2009) 119:350–64. doi: 10.1111/j.1600-0447.2009.01363.x
105. Dube SR, Anda RF, Felitti VJ, Chapman DP, Williamson DF, Giles WH. Childhood abuse, household dysfunction, and the risk of attempted suicide throughout the life span: findings from the adverse childhood experiences study. *JAMA.* (2001) 286:3089–96. doi: 10.1001/jama.286.24.3089
106. Stein DJ, Chiu WT, Hwang I, Kessler RC, Sampson N, Alonso J, et al. Cross-national analysis of the associations between traumatic events and suicidal behavior: findings from the WHO world mental health surveys. *PLoS One.* (2010) 5:e10574. doi: 10.1371/journal.pone.0010574
107. Devries KM, Mak JY, Bacchus LJ, Child JC, Falder G, Petzold M, et al. Intimate partner violence and incident depressive symptoms and suicide sttempts: a systematic review of longitudinal studies. *PLoS Med.* (2013) 10:e1001439. doi: 10.1371/journal.pmed.1001439
108. Webster J, Nicholas C, Velacott C, Cridland N, Fawcett L. Quality of life and depression following childbirth: impact of social support. *Midwifery.* (2011) 27:745–9. doi: 10.1016/j.midw.2010.05.014
109. Reid KM, Taylor MG. Social support, stress, and maternal postpartum depression: a comparison and supportive relationships. *Soc Sci Res.* (2015) 54:246–62. doi: 10.1016/j.ssresearch.2015.08.009
110. LeMasters K, Dussault J, Barrington C, Bengtson A, Gaynes B, Go V, et al. "Pain in my heart": understanding perinatal depression among women living with HIV in Malawi. *PLoS One.* (2020) 15:e0227935. doi: 10.1371/journal.pone.0227935
111. Taylor A, Atkins R, Kumar R, Adams D, Glover V. A new mother-to-infant bonding scale: links with early maternal mood. *Arch Womens Ment Health.* (2005) 8:45–51. doi: 10.1007/s00737-005-0074-z
112. Moehler E, Brunner R, Wiebel A, Reck C, Resch F. Maternal depressive symptoms in the postnatal period are associated with long-term impairment of mother–child bonding. *Arch Womens Ment Health.* (2006) 9:273–8. doi: 10.1007/s00737-006-0149-5
113. Beck AT. Cognitive therapy of depression: new perspectives. In: Clayton PJ, Barett JE editors. *Treatment for Depression: Old Controversies and New Approaches*. New York, NY: Raven Press (1983). p. 265–90. doi: 10.1016/j.brat.2006.09.011
114. Zahn R, Lythe KE, Gethin JA, Green S, William Deakin JF, Young AH, et al. The role of self-blame and worthlessness in the psychopathology of major depressive disorder. *J Affect Disord.* (2015) 186:337–41. doi: 10.1016/j.jad.2015.08.001
115. Jeon HJ, Park JI, Fava M, Mischoulon D, Sohn JH, Seong S, et al. Feelings of worthlessness, traumatic experience, and their comorbidity in relation to lifetime suicide attempt in community adults with major depressive disorder. *J Affect Disord.* (2014) 166:206–12. doi: 10.1016/j.jad.2014.05.010
116. Nicolson P. Understanding postnatal depression: a mother-centred approach. *J Adv Nurs.* (1990) 15:689–95. doi: 10.1111/j.1365-2648.1990.tb01892.x
117. Bandura A. *Self-Efficacy: The Exercise of Control*. New York, NY: Worth Publishers (1997).
118. Bhar S, Ghahramanlou-Holloway M, Brown G, Beck AT. Self-esteem and suicide ideation in psychiatric outpatients. *Suicide Life Threat Behav.* (2008) 38:511–6. doi: 10.1521/suli.2008.38.5.511
119. Leahy-Warren P, McCarthy G, Corcoran P. First-time mothers: social support, maternal parental self-efficacy, and postnatal depression. *J Clin Nurs.* (2012) 21:388–97. doi: 10.1111/j.1365-2702.2011.03701.x
120. Teti D, Gefland D. Behavioural competence among mothers of infants in the first year: the mediational role of self-efficacy. *Child Dev.* (1991) 65:918–29.
121. Jones DL, Rodriguez VJ, Alcaide ML, Weiss SM, Peltzer K. The use of efavirenz during pregnancy is associated with suicidal ideation in postpartum women in rural South Africa. *AIDS Behav.* (2019) 23:126–31. doi: 10.1007/s10461-018-2213-3
122. Rodriguez VJ, Cook RR, Peltzer K, Jones DL. Prevalence and psychosocial correlates of suicidal ideation among pregnant women living with HIV in Mpumalanga Province, South Africa. *AIDS Care.* (2017) 29:593–7. doi: 10.1080/09540121.2016.1238442

123. Crandall M, Sridharan L, Schermer C. Depression and thoughts of death among disadvantaged mothers: risk factors and impact on maternal and child health. *Arch Suicide Res.* (2010) 14:248–60. doi: 10.1080/13811118.2010.494143
124. Abdelghani M, Saad A, Khalil Y, Ibrahem MA, Badr MS, Saraya Y, et al. Can lifetime exposure to intimate partner violence predict suicidality in a sample of Egyptian pregnant women: a cross-sectional study? *Eur J Psychiatry*. (2021) 35:83–91. doi: 10.1016/j.ejpsy.2020.11.005
125. Zhang X, Sun J, Wang J, Chen Q, Cao D, Wang J, et al. Suicide ideation among pregnant women: the role of different experiences of childhood abuse. *J Affect Disord.* (2020) 266:182–6. doi: 10.1016/j.jad.2020.01.119
126. Rurangirwa AA, Mogren I, Ntaganira J, Govender K, Krantz G. Intimate partner violence during pregnancy in relation to non-psychotic mental health disorders in Rwanda: a cross-sectional population-based study. *BMJ Open.* (2018) 8:e021807. doi: 10.1136/bmjopen-2018-021807
127. Shamu S, Zarowsky C, Roelens K, Temmerman M, Abrahams N. High-frequency intimate partner violence during pregnancy, postnatal depression and suicidal tendencies in Harare, Zimbabwe. *Gen Hosp Psychiatry*. (2016) 38:109–14. doi: 10.1016/j.genhosppsych.2015.10.005
128. Zhong QY, Wells A, Rondon MB. Childhood abuse and suicidal ideation in a cohort of pregnant Peruvian women. *Am J Obstet Gynecol.* (2016) 215:501.e1-8. doi: 10.1016/j.ajog.2016.04.05
129. Alhusen JL, Frohman N, Purcell G. Intimate partner violence and suicidal ideation in pregnant women. *Arch Womens Ment Health.* (2015) 18:573–8. doi: 10.1007/s00737-015-0515-2
130. Fonseca-Machado MD, Alves LC, Haas VJ, Monteiro JCD, Gomes-Sponholz F. Under the shadow of maternity: pregnancy, suicidal ideation, and intimate partner violence. *Rev Panam Salud Publica.* (2015) 37:258–64.

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

APPENDIX A. MEDLINE SEARCH

The search strategy for MEDLINE.

1	exp SUICIDE
2	exp SUICIDAL IDEATION
3	exp SUICIDE, ATTEMPTED
4	exp SELF-INJURIOUS BEHAVIOR
5	(suicid* OR parasuicid* OR self*harm* OR self-injur* OR self-inflict* OR poisoning OR drug overdose OR self-poison* OR overdose OR self mutilat*).ti,ab.
6	1 OR 2 OR 3 OR 4 OR 5
7	exp MOTHERS
8	exp WOMEN
9	exp PREGNANCY
10	exp POSTPARTUM PERIOD
11	exp PERIPARTUM PERIOD
12	exp PERINATAL DEATH
13	(mother* OR pregnan* OR antenatal OR postpartum OR peripartum OR perinatal OR postnatal OR puerper*).ti,ab.
14	7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13
15	6 AND 1

frontiers | Frontiers in Psychiatry

STUDY PROTOCOL
published: 04 April 2022
doi: 10.3389/fpsyt.2022.809807

The Danish High-Risk and Resilience Study—VIA 15 – A Study Protocol for the Third Clinical Assessment of a Cohort of 522 Children Born to Parents Diagnosed With Schizophrenia or Bipolar Disorder and Population-Based Controls

Anne Amalie Elgaard Thorup[1,2,3]*, *Nicoline Hemager*[3,4], *Vibeke Fuglsang Bliksted*[3,5,6], *Aja Neergaard Greve*[3,5,6], *Jessica Ohland*[3,4], *Martin Wilms*[3,4], *Sinnika Birkehøj Rohd*[3,4], *Merete Birk*[3,6], *Anette Faurskov Bundgaard*[3,6], *Andreas Færgemand Laursen*[3,6], *Oskar Hougaard Jefsen*[3,6], *Nanna Lawaetz Steffensen*[3,6], *Anna Krogh Andreassen*[3,5,6], *Lotte Veddum*[3,5,6], *Christina Bruun Knudsen*[3,5,6], *Mette Enevoldsen*[3,4], *Marie Nymand*[3,4], *Julie Marie Brandt*[2,3,4], *Anne Søndergaard*[2,3,4], *Line Carmichael*[3,4], *Maja Gregersen*[2,3,4], *Mette Falkenberg Krantz*[1,3], *Birgitte Klee Burton*[1,2,3,7], *Martin Dietz*[8], *Ron Nudel*[3,4], *Line Korsgaard Johnsen*[1,2,9], *Kit Melissa Larsen*[1,9], *David Meder*[9], *Oliver James Hulme*[9], *William Frans Christiaan Baaré*[9], *Kathrine Skak Madsen*[9,10], *Torben Ellegaard Lund*[8], *Leif Østergaard*[9], *Anders Juul*[2,11], *Troels Wesenberg Kjær*[12], *Carsten Hjorthøj*[2,4,13], *Hartwig Roman Siebner*[2,9,14], *Ole Mors*[3,5,6] and *Merete Nordentoft*[2,3,4]

[1] Child and Adolescent Mental Health Centre, Copenhagen University Hospital, Mental Health Services, Capital Region Psychiatry, Copenhagen, Denmark, [2] Faculty of Health and Medical Sciences, Institute for Clinical Medicine, University of Copenhagen, Copenhagen, Denmark, [3] The Lundbeck Foundation Initiative for Integrative Psychiatric Research (iPSYCH), Aarhus, Denmark, [4] Research Unit CORE, Mental Health Centre, Copenhagen University Hospital, Mental Health Services, Capital Region Psychiatry, Copenhagen, Denmark, [5] Department of Clinical Medicine, Faculty of Health and Medical Services, Aarhus University, Aarhus, Denmark, [6] The Psychosis Research Unit, Aarhus University Hospital, Aarhus, Denmark, [7] Child and Adolescent Mental Health Center, Roskilde, Denmark, [8] Center of Functionally Integrative Neuroscience, Aarhus University Hospital, Aarhus, Denmark, [9] Danish Research Centre for Magnetic Resonance, Centre for Functional and Diagnostic Imaging and Research, Copenhagen University Hospital Amager and Hvidovre, Copenhagen, Denmark, [10] Department of Radiography, Department of Technology, University College Copenhagen, Copenhagen, Denmark, [11] Department of Growth and Reproduction, Rigshospitalet, Section 5064, University of Copenhagen, Copenhagen, Denmark, [12] Department of Neurology, Zealand University Hospital, Roskilde, Denmark, [13] Department of Public Health, Section of Epidemiology University of Copenhagen, Copenhagen, Denmark, [14] Department of Neurology, Hospital Bispebjerg, Copenhagen University, Copenhagen, Denmark

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Roser Granero,
Universitat Autònoma de Barcelona, Spain
Miriam Heyman,
Brandeis University, United States
Louise Claire Johns,
Oxford Health NHS Foundation Trust,
United Kingdom

***Correspondence:**
Anne Amalie Elgaard Thorup
Anne.Amalie.Elgaard.Thorup@regionh.dk

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: 05 November 2021
Accepted: 28 January 2022
Published: 04 April 2022

Citation:
Thorup AAE, Hemager N, Bliksted VF, Greve AN, Ohland J, Wilms M, Rohd SB, Birk M, Bundgaard AF, Laursen AF, Jefsen OH, Steffensen NL, Andreassen AK, Veddum L, Knudsen CB, Enevoldsen M, Nymand M, Brandt JM, Søndergaard A, Carmichael L, Gregersen M, Krantz MF, Burton BK, Dietz M, Nudel R, Johnsen LK, Larsen KM, Meder D, Hulme OJ, Baaré WFC, Madsen KS, Lund TE, Østergaard L, Juul A, Kjær TW, Hjorthøj C, Siebner HR, Mors O and Nordentoft M (2022) The Danish High-Risk and Resilience Study—VIA 15 – A Study Protocol for the Third Clinical Assessment of a Cohort of 522 Children Born to Parents Diagnosed With Schizophrenia or Bipolar Disorder and Population-Based Controls. Front. Psychiatry 13:809807. doi: 10.3389/fpsyt.2022.809807

Background: Children born to parents with severe mental illness have gained more attention during the last decades because of increasing evidence documenting that these children constitute a population with an increased risk of developing mental illness and other negative life outcomes. Because of high-quality research with cohorts of offspring with familial risk and increased knowledge about gene–environment interactions, early interventions and preventive strategies are now being developed all over the world. Adolescence is a period characterized by massive changes, both in terms of physical, neurologic, psychological, social, and behavioral aspects. It is also the period of life with the highest risk of experiencing onset of a mental disorder. Therefore, investigating the impact of various risk and resilience factors in adolescence is important.

Methods: The Danish High-Risk and Resilience Study started data collection in 2012, where 522 7-year-old children were enrolled in the first wave of the study, the VIA 7 study. The cohort was identified through Danish registers based on diagnoses of the parents. A total of 202 children had a parent diagnosed with schizophrenia, 120 children had a parent diagnosed with bipolar disorder, and 200 children had parents without these diagnoses. At age 11 years, all children were assessed for the second time in the VIA 11 study, with a follow-up retention rate of 89%. A comprehensive assessment battery covering domains of psychopathology, neurocognition, social cognition and behavior, motor development and physical health, genetic analyses, attachment, stress, parental functioning, and home environment was carried out at each wave. Magnetic resonance imaging scans of the brain and electroencephalograms were included from age 11 years. This study protocol describes the third wave of assessment, the VIA 15 study, participants being 15 years of age and the full, 3-day-long assessment battery this time including also risk behavior, magnetoencephalography, sleep, and a white noise paradigm. Data collection started on May 1, 2021.

Discussion: We will discuss the importance of longitudinal studies and cross-sectional data collection and how studies like this may inform us about unmet needs and windows of opportunity for future preventive interventions, early illness identification, and treatment in the future.

Keywords: familial high risk, schizophrenia, bipolar disorder, adolescent mental health, developmental trajectories

INTRODUCTION

Parental mental illness is known to affect children in many ways, including risk of negative influence on upbringing circumstances, home environment, neurodevelopment, and increased risk of developing mental problems and disorders. This fact has been documented in several studies (1, 2), but less is known about how mental illness affects offspring during the adolescent years. As adolescence is a period characterized by significant changes in brain structure, connectivity, and functioning, as well as changes in physical appearance, hormonal status, and psychological and social constitution (3, 4), it is a period of life with dramatic development and changes. Adolescence is the time where the young person is searching for individuation and autonomy, while having a strong focus on peer relationships and at the same time start to separate from home and especially from the parents. It is concurrently the period with the highest incidence rates for mental disorders (3) and risk behavior (5). From a developmental perspective, it is a period in life that is highly not only formative but also challenging to study because of the complex interplay of biological (e.g., genetics, hormonal status, neuroplasticity) and social, environmental, and psychological (e.g., education, peers, sexual debut) risk factors.

Brain Development

While early childhood includes the first and very sensitive periods for development of the sensory and motor systems (6), adolescence constitutes a second, but also very sensitive period for further development of the social, emotional, and higher cognitive domains (7). The networks that serve and constitute these brain functions are undergoing neuroplastic changes based on the experiences of the individual. The adolescent brain development can be characterized as a continuous maturation of cognitive functions mediated by higher associative cortices such as the prefrontal cortex including working memory, planning, concept formation, inhibitory control, and emotion regulation (8). A thinning of the cortex within the prefrontal cortex (and many other brain regions) and an increase in white matter density and volume are taking place (9, 10). In parallel, the brain is undergoing regional heterogeneous maturational changes with primary sensory and motor areas maturing before high associative cortical regions. Apparent cortical thickness is continuously decreasing from ∼4 years of age, and surface area is increasing until early adolescence, whereas cortical gray matter volume steadily decreases after a peak in early adolescence (11). Such changes are thought to reflect both synaptic pruning and cortical myelination (12). White matter volume continues to increase into adulthood, with specific white matter fiber tracts displaying heterogeneous maturation with frontal–temporal association tracts such as the cingulum and uncinate fasciculus maturing well into adulthood (13).

With this maturation of complex structures and underlying brain networks, reflecting a high level of plasticity and learning potential, comes a heightened vulnerability to disease, disorder, and risk exposures that can compromise functional and structural maturation. External influence may lead to an increased possibility that functional and structural maturation can become abnormal and psychopathology may emerge (14). Puberty plays a role in brain maturation. Its onset in each individual varies widely, and so does its contribution (5, 15).

Adolescent brain development is not linear as the brain gets more connected and specialized in some areas, whereas others are reduced through a pruning process (16). The changing dynamic between frontal/executive and limbic/arousal/reward regions strongly influences the behavior of the individual. The malleability of the developing brain represents a high level of plasticity and learning potential but at the same time also represents vulnerability to disease, disorder, and risk exposures (14). Human brain development and functioning are also highly dependent on precise epigenetic regulation, and aberrant changes are increasingly reported to be associated with mental disorders (17, 18). Thus, DNA methylation plays a pivotal role in regulation of neuronal development and functioning, and its levels can be modified by environmental factors. Moreover, the genetic background of an individual is also associated with epigenetic variability, and risk single-nucleotide polymorphisms for mental disorders are reported to alter DNA methylation.

Adolescence is also the time for social transition from childhood to adulthood (7). In this transition period, research shows that young people are much more orientated toward and interested in their peers and how they look and behave than in adults (19). The social context is larger and more unpredictable, which implies a risk for social isolation, bullying, or peer rejection; it can be hard to cope with for vulnerable individuals (20). These processes and changes involve the networks of social cognition including mentalization and emotion regulation, which are some of the latest developed areas in humans. Good emotion regulation and well-developed mentalization (i.e., ability to think about others' thoughts, intentions and preferences) are protective against misunderstanding or interpreting others' behavior as directed negatively toward one self and to help to adapt to a stressful social situation [e.g., a peer rejection (20)]. On the other hand, these processes may also be involved in risk-taking behavior like experiments with drugs and alcohol or deliberate self-harm behavior (21).

In summary, adolescence can be understood as a window of vulnerability due to the significant neural changes, the changes in social roles, the onset of puberty, the increased risk of substance abuse, and other kinds of risk behavior, which can explain why the adolescent is at an increased risk of developing depression, psychosis, and many other mental health problems (22). A thorough review of the current knowledge and evidence on adolescence, brain development, and psychopathology can be found in *Biological Psychiatry* (23), where this was the special theme for the full issue (https://doi.org/10.1016/j.biopsych.2020.06.012).

Familial High-Risk Studies

Schizophrenia and bipolar disorder are among the most costly and debilitating disorders both in terms of personal suffering for those affected, for the children and other relatives, and for society (24). Identifying disease mechanisms and possibilities for prevention before onset of illness will therefore be extremely valuable. As schizophrenia and bipolar disorder are rare conditions in the general population, studies of enriched populations (like children with familial high risk) can be fruitful and provide insight into the early disease processes. Approximately 55% of the children born to parents with schizophrenia, bipolar disorder, and severe depression will develop some kind of mental illness themselves during early adult life (25). Thus, the offspring have both a higher risk of developing the same disorder as their parents, or another severe mental disorder.

Familial high-risk studies have been conducted for decades (1, 26, 27). Previous familial high-risk studies have reported neurointegrative problems, social impairments, poorer neurocognitive and neuromotor functions, and early symptomatology among offspring of parents with severe mental illness (1, 2, 26, 28–30). However, because of limitations in previous studies such as small sample sizes, poor representativeness and wide age ranges, high attrition rates, lack of specific measures that inform about the underlying neurobiological processes, and lack of longitudinal follow-up, it is not clear whether these abnormalities abate, prevail, or worsen (30) over time.

Most of the previous studies were mainly based on convenience samples and were thus not representative. They included only a single assessment during childhood, and participating children were in different age groups (1). Developmental trajectories require at least three assessments, and longitudinal clinical cohort studies are therefore very valuable, although time consuming and costly. Attrition/dropout rates can be high, too.

Former waves of the study presented in this article have documented that children born to parents with schizophrenia and bipolar disorder show signs of vulnerability in a range of domains. In the first wave, The Danish High-Risk and Resilience Study—VIA 7, we found that as a group children with familial risk for schizophrenia and to some extent also bipolar disorder at age 7 years were impaired in, for example, neurocognitive functioning (31–33), social functioning (34), motor functioning (35), and mental health (36–38), while also living in environments with poorer levels of stimulation and support (39). The Danish High-Risk and Resilience Study (40)—of which the third wave, the VIA 15 study, is presented here—has overcome the obstacles described above by recruiting a large sample through national registers, all in a narrow age range that has been maintained in all three waves. The longitudinal method allows inference about development in the repeated waves of cross-sectional examinations, ultimately following developmental pathways in the longitudinal design. Therefore, conducting regular follow-ups on the defined outcomes is crucial for the end results of the study.

Structural and functional brain changes are present in drug-naive adult patients with schizophrenia, and some of the strongest risk factors exert their influence already in the prenatal or perinatal period (41). Notably, structural and functional neuroimaging of a large group of familial high-risk children before and during puberty, using a longitudinal design, has never been carried out before (42). In a recent study of offspring with familial risk for schizophrenia and bipolar disorder, the analysis of structural and functional brain networks revealed prominent group differences in brain organization, comparing vulnerable groups within a broad age range, and a relatively small sample

(43). Brain imaging before, during, and after puberty is lacking in order to study brain development during this crucial period in human life. No previous studies have performed follow-up magnetic resonance imaging (MRI) of the brain of a large group of adolescents with a familial predisposition for schizophrenia and bipolar disorder.

AIMS AND HYPOTHESES

The overall aim of this third wave of The Danish High-Risk and Resilience Study—VIA 15 is to follow up on the already defined domains of development and function in order to describe developmental trajectories, which are of great importance for mental health. The domains are psychopathology, neurocognition, motor function, and somatic health including sleep, physical activity, social cognition and social functioning, structural brain development, functional brain development, and environmental risk assessment including family situation, childhood trauma, and risk behavior.

We aim to

(1) improve insight into early disease processes of schizophrenia and bipolar disorder including early symptom formation and psychopathology, impairments or delays of maturation in different domains of cognitive functioning including social cognition, and changes in brain structure and task-related brain activation;
(2) identify the influence of genetic, epigenetic, and environmental exposures by analyzing associations between outcomes, such as psychopathology, risk behavior, and social and cognitive functioning, and structural and functional brain readouts and exposures, such as polygenic risk scores for schizophrenia, major depressive disorder, and educational attainment, and direct and indirect measures of the emotional climate in the family;
(3) identify early modifiable risk and resilience factors, such as low levels of stimulation and support in the home, traumatic life events during childhood, conflicting relation parents, neurocognitive and social cognitive deficits, risk behavior, and early signs of psychopathology, leading to development of good prediction models; and
(4) communicate the very important knowledge gained in this project about a vulnerable and overlooked group of children and adolescents to professionals who work with this population.

METHODS

Design

The Danish High-Risk and Resilience Study is a representative nationwide longitudinal multi-informant cohort study consisting of 522 children born to parents with schizophrenia, bipolar disorder, or population-based controls. The participating families were recruited from Danish registers and investigated thoroughly during 2013–2015 when the children were 7 years old. This first assessment is referred to as the VIA 7 study (40). The second wave of assessments, the VIA 11 study (44), was carried out when the children were 11 years of age from 2017 to 2020 with an 89% retention rate. See **Figure 1** for the flowchart and **Figure 2** for image of recruitment folder sent to each family by mail.

The cohort consists of (a) 202 children with at least one parent diagnosed with schizophrenia spectrum psychosis (familial high risk of schizophrenia [FHR-SZ]); (b) 120 children with at least one parent diagnosed with bipolar disorder (familial high risk of bipolar disorder [FHR-BP]); (c) 200 children with neither of the parents treated in mental health services for the above diagnoses (population-based controls).

The control children were matched to FHR-SZ children on municipality, sex, and age. We included FHR-BP children as a nonmatched group; however, the group was comparable to the two other groups with respect to age and sex. The representative cohort is based on data from The Danish Civil Registration System (45) linked to the Danish Psychiatric Central Research Register (46). Analyses have shown that our cohort in many aspects is representative of the Danish population and have been described elsewhere (Falkenberg Krantz, submitted). Because of limitation of resources, we were able to include only 120 FHR-BP children.

Earlier Assessments

In the VIA 7 study, saliva from the children and blood samples from the parents were used for genome-wide association analyses (GWASs, PsychChip). When the children were 7 and 11 years of age, the children and their parents were thoroughly examined with interviews, neurocognitive and social cognitive tests, questionnaires, home visits, and observations. In the VIA 11 study, MR scans and electroencephalographic (EEG) recordings were also performed. Assessments were supplemented with data from questionnaires sent to schoolteachers. Outcomes for the children were assessed thoroughly in the domains of neuromotor functioning, neurocognitive and social cognitive functioning, social functioning, and psychopathology at both ages. In addition, parents were interviewed about their mental health, and data on their neurocognitive functioning were collected. All assessors were kept blinded to whether the children were at familial high-risk or were population-based controls. Registration of unblinding in the former waves showed that assessors were unblinded in ∼10% of all families.

The full assessment batteries in the VIA 7 and the VIA 11 study lasted ∼3 days, and the vast majority of the families completed the whole battery. Parents were always offered feedback on their child's performance, and all participants received a gift card for their time taken, and practical obstacles such as transportation and catering were taken care of by the researchers. For families traveling longer distances, a hotel stay will be arranged for each family, like in the former waves. All families were informed at their previous visit that a follow-up at age 15 years was being planned.

Assessment in the VIA 15 Study

The test battery in the VIA 15 study primarily focuses on the adolescent offspring, whereas only the primary caregiver's current level of daily functioning will be assessed with the Personal and Social Performance Scale [PSP Interview (47)]. Like

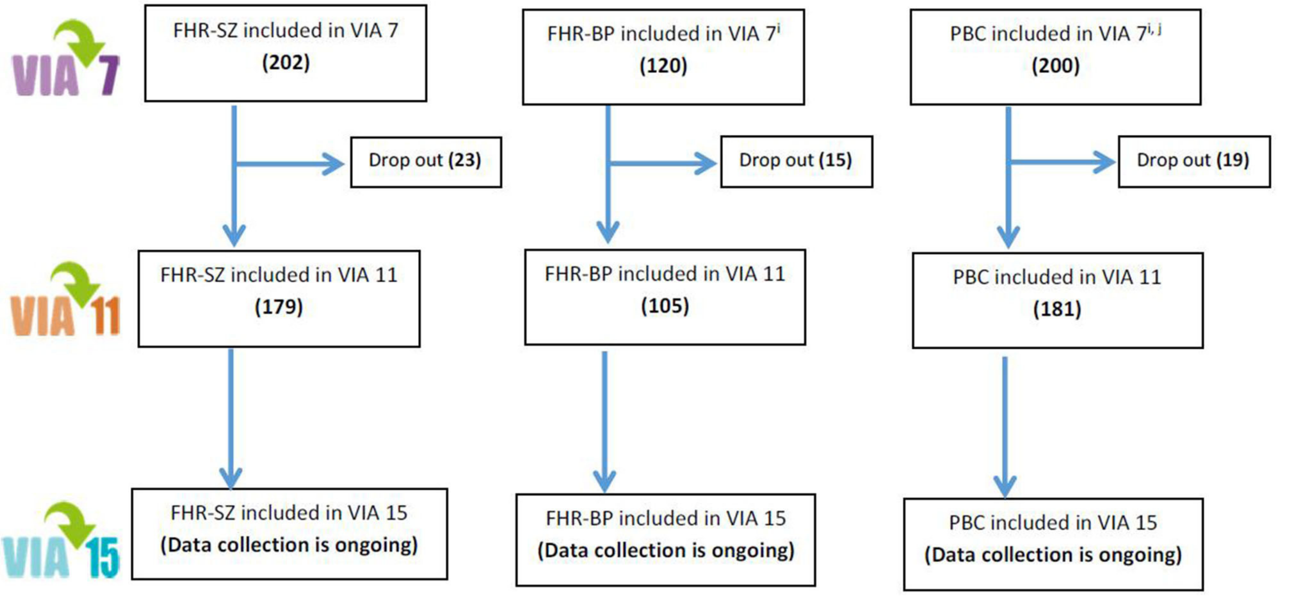


FIGURE 1 | Flowchart of The Danish High-Risk and Resilience Study.

in the former waves, a "primary caregiver" will be identified in each family (this may be a biological parent but could also be another adult), that is, a person, who is responsible for the adolescent's well-being on a daily/regular basis and preferably lives at the same address as the adolescent. This person may thus be different from the "primary caregiver," who participated in the VIA 7 or the VIA 11 study. The primary caregiver will be asked to give information about the adolescent's mental health status and daily functioning both in interviews and from questionnaires.

The test battery for the adolescent will last ∼3 days (5–6 h duration per day including breaks) and one night's sleep. Most tests and interviews will take place at the research clinic, unless the family for some reason needs the assessment to take place somewhere else, for example, in their home. However, measurement of sleep and assessment of the home environment will always take place in the home of the adolescent. All outcome measures are being examined with validated instruments, specifically developed and selected for this age group, sensitive to small changes, and suitable for later follow-up. Many variables will be measured for the third time, making analyses of trajectories possible. The battery consists of interviews, tests, observation, and questionnaires (**Table 1**). The battery is comprehensive, and some may find it exhausting, but individual needs (e.g., for breaks or shorter test days) are always taken into account to ensure a positive experience for the participants. As in the former waves, all adolescent assessors will be blinded to the familial risk status of each family.

The assessors are highly skilled and educated psychologists, doctors, and research nurses who have been part of the preparation phase in the VIA 15 study and are trained and accredited in all tests and interviews. Weekly clinical conferences will be held in order to ensure uniformity between sites and testers, and a specialist in child and adolescent psychiatry (A.A.E.T.) will be present when psychiatric diagnoses are determined. For some instruments, interrater reliability will be measured [Vineland (48), Movement Assessment Battery for Children [ABC] (49)], whereas for others [Children's Global Assessment Scale [C-GAS] (50), PSP (47), psychotic experiences [PEs]], ratings will be made in consensus.

Overview of Domains and Instruments in the via 15 Study

Adolescent Assessment

Neuromotor Function

Manual dexterity, ball skills, and balance are assessed with Movement ABC-2 (49), a clinical, gold-standard test for motor function that has also been used in the two former waves. To investigate manual dexterity, the participants will also be performing a circle-drawing task with their right and left hands on a pressure-sensitive digitizing tablet (WACOM Intuos4 large PTK-840; Wacom Technology Corporation, Vancouver, WA, USA) recording their writing trace from which the kinematics of the movements can be derived (e.g., movement velocity, frequency, and variability) (51).

Neurocognitive Function

Neurocognitive functions will be assessed with Rey's Complex Figure Test (52), Rapid Visual Information Processing [from Cambridge Neuropsychological Test Automated Battery [CANTAB] (53)], Verbal Fluency 1–3, and Trail Making Test conditions 2–4 A/B from the Delis-Kaplan Executive Function System (54), Symbol Search and Coding from the Wechsler Intelligence Test for Children—Fourth Edition [WISC-IV (55)], Stockings of Cambridge, Intra–Extra Dimensional Shift and

FIGURE 2 | Image of the folder sent to all participating families in the VIA 15 study by mail.

Spatial Working Memory [from CANTAB (28)], Letter–Number Sequencing and Arithmetic [WISC-IV (55)] Word Selective Reminding from the Test of Memory and Learning—Second Edition (56), and Reynolds Intellectual Screening Test (57). Smell identification is measured with the Brief Smell Identification Test (58).

Social Cognition

Social cognition is measured by Animated Triangles (59, 60), consisting of short movie clips with two animated triangles moving around either in an intentional or arbitrary manner (note that the Animated Triangles Task measures theory of mind, a social cognitive domain), Emotion Recognition Task [from CANTAB (28), The Awareness of Social Inference Test—Part A2, and the Social Cognition paradigm from the Human Connectome Project (61) (performed during MRI).

Psychopathology

General psychopathology and PEs will be examined with the gold-standard diagnostic interview Kiddie Schedule for Affective Disorders and Schizophrenia [K-SADS-PL (62)]. This interview also includes a score based on a general assessment of the adolescent's daily functioning in the current month, the C-GAS (50). As before, we will include a specialized assessment of subthreshold psychotic-like experiences (PEs) inspired from the Scale of Prodromal Symptom Scale (63). Possible diagnoses and all PEs will be discussed at clinical conferences with a child and adolescent psychiatrist present. We used a modified version of the Attention Deficit/Hyperactivity Disorder Rating Scale [mADHD-RS (64)] to assess symptoms of attention-deficit/hyperactivity disorder and oppositional defiant disorder, rated both by the primary caregiver and the teacher. Affective liability will be measured using Children's Affective Liability Scale [CALS (65)]. We will also include Youth Experience Tracker Instrument [YETI (66)], a new brief self-report measure designed to facilitate early identification of risk for severe forms of mental illness, including major depressive disorder, bipolar disorder, and schizophrenia. By using the white noise paradigm (67), we will be able to investigate if a subgroup of children is more likely than the others to appraise an ambiguous situation as, for example, threatening. We will apply a Danish version of the white noise paradigm, which is a series of 75 very short sound clips with white noise. In two of three sound clips, short and neutral sentences are included in the sound of the white noise, 25 clearly audible and 25 barely audible, whereas the remaining 25 sound clips included only white noise. The respondents can select the following responses: 1 = "hearing positive voice," 2 = "hearing negative voice," 3 = "hearing neutral voice," 4 = "no speech heard," and 5 = "uncertain."

Data from school will also be included via questionnaires sent to the schoolteachers if parents give permission (i.e., sign a consent form). Executive functioning including affective regulation and flexibility will be assessed with the questionnaire Behavior Rating Inventory of Executive Function [BRIEF (68)] from both the primary caregiver and the teacher. Autism spectrum traits are evaluated with Social Responsiveness Scale [SRS (69)] also completed by the caregiver and the teacher. Dimensional measures of psychopathology will be covered with Youth Self-report version of the Child Behavior Checklist [CBCL (70)] and also from the primary caregiver and the teacher. The adolescent will also be asked to complete the Strengths and Difficulties Questionnaire (71). The ratings of the clinical impression of the adolescent during the testing are reported with Tester's Observation Form (72). All the mentioned questionnaires have been used in the VIA 7 study and in the VIA 11 study as well.

Social Functioning, Self-Esteem, Deliberate Self-Harm, Risk-Taking Behavior, and Resilience

Adaptive social functioning of the adolescent is captured by parental interview using the Vineland-2 (48). Self-esteem is covered by the questionnaire "Sådan er jeg" ("This Is Me"), a questionnaire about self-esteem in school, in the family, and in a peer context (73).

TABLE 1 | Assessment battery for the adolescents in The Danish High Risk and Resilience Study VIA 15, all domains.

Domains	Outcomes	Instrument	Type of test	Approximate duration	In VIA 7	In VIA 11
Neuromotor and physical measures	Motor function: manual dexterity, aiming & catching and balance	Movement Assessment Battery for Children-2 (Movement ABC-2)	Test	45 min	Yes	Yes
	Anthropometry	Measure of height, weight, waist	Observations in clinic	5 min	Yes	Yes
	Physical activity and sleep	SENS chip	Chip on thigh for 1 week	5 min + 1 week	No	Yes
	Polysomnography	Polysomnogram (PSG)	PSG monitor	Overnight	No	No
		Pittsburgh Sleep Quality Index (PSQI)	Questionnaire	5–10 min	No	No
	Motor screening	Motor Screening Test (MOT) from CANTAB	Computer test	5 min	Yes	Yes
	Smell identification	Brief Smell Identification Test (B-SIT)	Test	10 min	Yes	No
Neurocognition	Verbal Memory and visual memory	Word Selective Reminding from the Test of Memory and Learning–Second Edition TOMAL-2	Test	10 min	Yes	Yes
		Rey's Complex Figure Test (RCFT)	Test	8–10 min	Yes	Yes
	Attention	Rapid Visual Information Processing (RVP) from CANTAB	Computer test	10–15 min	Yes	Yes
	Flexibility and processing speed	Trail Making Test 2-4 from Delis-Kaplan Executive Function System (D-KEFS)	Test	8 min	Yes	Yes
		Symbol Search and Coding test from Wechsler Intelligence Test for Children – Fourth Edition (WISC-IV)	Test	5 min	Yes	Yes
		Verbal Fluency 1-2 from Delis-Kaplan Executive Function System (D-KEFS)	Test	4 min	Yes	Yes
	Executive functions (planning and flexibility)	Stockings of Cambridge (SOC) and Intra-Extra Dimensional Set Shift (IED) from CANTAB	Computer test	20–30 min	Yes	Yes
		Verbal Fluency 3 from Delis-Kaplan Executive Function System (D-KEFS)	Test	2 min	Yes	Yes
	Executive functions (error monitoring)	Flanker Task	Test before and during fMRI		Yes	Yes
	Executive functions (visual and verbal working memory)	Spatial Working Memory (SWM) from CANTAB	Computer test	10-15 min	Yes	Yes
		Letter-number Sequencing and Arithmetic from Wechsler Intelligence Test for Children – Fourth Edition (WISC-IV)	Test	10–15 min	Yes	Yes
	Social cognition	Human Connectome Project Social Cognition Paradigm	Test during MRI	10 min	No	Yes
		The Animated Triangles Task	Test	12 min	Yes	Yes
		The Awareness of Social Inference Test – Part A2 (TASIT A2)	Test	10 min	No	No

(Continued)

TABLE 1 | Continued

Domains	Outcomes	Instrument	Type of test	Approximate duration	In VIA 7	In VIA 11
	Intelligence	Reynolds Intellectual Screening (RIST)	Test	15 min	Yes	Yes
Psychopathology	Psychiatric symptoms	Kiddie Schedule for Affective Disorders and Schizophrenia (K-SADS-PL)	Interview	45–100 min	Yes	Yes
	Psychotic experiences	PE Hallucinations and PE Delusions	Interview inspired from the SOPS scale	1–10 min	Yes	Yes
	General functioning	Children's Global Assessment Scale (C-GAS)	Interview	20 min	Yes	Yes
	Speech illusions	White Noise Test	Test	20 min	No	No
	Risk factors for mental illness	Youth Experience Tracker Instrument (YETI)	Questionnaire	3–5 min	No	No
	Self-harm	Semi structured interview adapted to use in VIA 15 with items from Deliberate Self-Harm Inventory – Youth Version (DLSH-I) and iClinician-Administered Non-Suicidal Self-Injury Disorder Index (CANDI)	Interview	5–10 min	No	No
	Dimensional psychopathology	Youth Self Report for age 11-16 (YSR)	Questionnaire			
Social function and behavior	Behavior, affect, and test-taking-style	Tester's Observation Form (TOF)	Clinician rating	5–10 min	Yes	Yes
	Self-esteem	I think I am (Sådan er jeg")	Questionnaire	5 min	Yes	Yes
	Bullying	Semi structured interview based on Olweus Bully/Victim Questionnaire	Interview	1–10 min	No	Yes
	Resilience	Child and Youth Resilience Measure (CYRM-12)	Questionnaire	1–3 min	No	Yes
	Social functioning	Strength and Difficulties Questionnaire (SDQ)	Questionnaire	3 min	No	Yes
	Risk behavior	Adapted questionnaire from 2019 National Risk Behavior Survey (USA)	Questionnaire	5–10 min	No	No
Environment and emotional climate	Perceived social support	Multidimensional Scale of Perceived Social Support (MSPSS)	Questionnaire	2 min	No	No
	Expressed emotions/ emotional family climate/familiar relations	Five Minute Speech Sample (FMSS)	Interview	7 min	No	No
		Family Assessment Device (FAD)	Questionnaire	2 min	No	No
	Social network and contact	Social contact questionnaires (from Lasgaard et al.)	Questionnaire	2 min	No	No
	Childhood trauma	Childhood Trauma Questionnaire, short form (CTQ-SF)	Questionnaire	3–5 min	No	No
Biological measures and physical health	Pubertal status	Tanner stages incl. menarche	Illustrations, test	3 min	No	Yes
		Hormone level	Blood sample		No	Yes
	Physical health	HbA1c. leucocytes, CRP	Blood sample		No	Yes
	Stress, biological measure	Hair test for long term level osf cortisol	Hair sample	5 min	Yes	Yes

(Continued)

TABLE 1 | Continued

Domains	Outcomes	Instrument	Type of test	Approximate duration	In VIA 7	In VIA 11
	Bodily distress	Body Distress Symptoms checklist (BDS)	Questionnaire	2 min	No	No
	Health anxiety and somatization	Whiteley Index 6-R (Wi-6)	Questionnaire	1 min	No	No
Genetic and epigenetic analyses	Polygenic risk scores	Blood sample, saliva sample, dry blood spot and dry blood spots from Danish Neonatal Screening Biobank	Blood samples	5–10 min	Yes	Yes
	Inflammatory and infectious markers	Blood samples and dry blood spots	Blood sample	5–10 min	No	Yes
Brain scan	Brain structure and brain activity	Functional and Structural MRI and EEG	Brain scan at hospital	90 min	No	Yes
	Electrophysiology (only in Copenhagen)	The Copenhagen psychophysiological Test Battery: 40 Hz auditory steady state response Mismatch negativity Modified Eriksen Flanker task	Brain scan at hospital	120 min	No	Yes
	Magnetoencephalography (MEG, only Aarhus)	Paradigms: Roving auditory oddball + 40 Hz auditory steady state response	Brain scan at hospital	90 min	no	no

TABLE 2 | Assessment battery for the primary caregiver in The Danish High Risk and Resilience Study—VIA 15.

Domains	Instrument	Type of test	Duration	In VIA 7	In VIA 11
Family relations, education, stressors, health and social life	Anamnesis	Interview	30–40 min	Yes	Yes
Mental health status in adolescent	Kiddie Schedule for Affective Disorders and Schizophrenia (K-SADS-PL)	Interview	45–90 min	Yes	Yes
Attention/hyperactivity	ADHD-Rating Scale	Questionnaire	5–10 min	Yes	Yes
Executive functions	Behavior Rating Inventory of Executive Function (BRIEF)	Questionnaire	10 min	Yes	Yes
Autism spectrum traits	Social Responsiveness Scale (SRS-2)	Questionnaire	10 min	No	Yes
Affect regulation	The Children's Affective liability Scale (CALS)	Questionnaire	2–5 min	No	No
Social development	Vineland Adaptive Behavior Scales – II	Interview	20 min	Yes	Yes
Daily functioning	Personal and Social Performance Scale (PSP)	Interview	10 min	Yes	Yes
Family Functioning	Family Assessment Device (FAD)	Questionnaire	3 min	No	Yes
Environment and emotional climate	Five minutes Speech Sample (FMSS)	Interview	7 min	Yes	Yes
Behavior	Child Behavior Checklist (CBCL)	Questionnaire	10 min	Yes	Yes
Genetic and epigenetic analyses	Saliva sample	Saliva sample	5 min	Yes	Yes

Deliberate self-harm is a questionnaire made by our own research group in collaboration with specialists in the area. We collapsed items from two longer questionnaires, the Deliberate Self-harm Inventory—Youth Version (74) and Clinician-Administered Non-Suicidal Self-injury Disorder Index [CANDI (75)] and will be administered as a semistructured interview in the VIA 15 study.

Risk-taking behavior will be assessed with a modified and adapted questionnaire based on Youth Risk Behavior Surveillance System (76), whereas school performance, leisure activities, social relations, and use of social media are included

TABLE 3 | Assessment battery for the adolescents' teachers in The Danish High Risk and Resilience Study VIA 15.

Domains	Instrument	Type of test	Duration	In VIA 7	In VIA 11
Psychosocial functioning and behavior	Teachers Rating Form (TRF)	Questionnaire	10 min	Yes	Yes
Attention/Hyperactivity	ADHD-Rating Scale	Questionnaire	5 min	Yes	Yes
Affect regulation/flexibility	Behavior Rating Inventory of Executive Function (BRIEF)	Questionnaire	5 min	Yes	Yes
Autism spectrum traits	Social Responsiveness Scale (SRS-2)	Questionnaire	5–10 min	Yes	Yes
Communication and social interaction	The Children's Communication Checklist CCC-2	Questionnaire	10 min	Yes	Yes

in the anamnestic interview (i.e., interview about what has happened in the adolescent's life within the previous 4 years, since the VIA 11 study) made primarily with the primary caregiver as informant. Alcohol and drug use is also covered by interview, partly as part of the K-SADS-PL (diagnostic level of misuse) and in a specific, short interview suited for this specific age group. Level of stress will be captured from hair cortisol. Perceived social support will be assessed with a questionnaire, Multidimensional Scale of Perceived Social Support [MSPSS (77)].

Resilience is measured by a short version of the questionnaire Child Youth Resilience Measurement—Youth Version (78). Affective regulation is captured by the questionnaire CALS (65).

Environmental Factors

The family environment in terms of family functioning will be assessed by both the parent and the adolescent by using the questionnaire Family Assessment Device [FAD (79)], which was also in the VIA 11 study. The 5 Min Speech Sample [FMSS (80)] was used in the VIA 7 and the VIA 11 studies for the primary caregiver to talk about the child, but this time it will be administered with both the primary caregiver and the adolescent. Adverse life events including unwanted sexual experiences will also be assessed by a questionnaire, Childhood Trauma Questionnaire—Short Form (81, 82), and is also included in the anamnestic interview. Further, childhood trauma is measured directly from the adolescent and the primary caregiver in the K-SADS-PL (62) interview section about traumatic events and PTSD. Social network is captured by MSPSS (77).

Biological Measures and Physical Health

We will make a clinical evaluation of anthropometry of the adolescent (height, weight, and waist circumference) at the time when the adolescent visits the clinic. Further, three different biological samples will be acquired, including a small hair sample to measure the levels of the stress hormone cortisol, a blood sample that will provide data on the immune system, diabetes, and so on, and a saliva sample used for genetic and epigenetic analyses. Physical activity will be measured by a sensor in an easily wearable adhesive patch [SENS motion® (83)], which directly measures the amount and level of physical activity during a 1-week observation period. Retrospective report on menarche and growth will be obtained, and puberty status will be assessed from the four Tanner stages by asking the adolescents to estimate their current developmental state from a figure (84, 85). Sex hormones (i.e., testosterone and estradiol) will be measured from the blood sample. Bodily distress symptoms are covered by the questionnaire Body Distress Symptoms checklist (86), and screening for somatization and hypochondriasis is covered by Whiteley Index 6-R (87).

Neuroimaging

Structural and Functional MRI and Magnetoencephalography/EEG. We will repeat the anatomical and functional MRI (fMRI) of the whole brain at 3.0 T, which was carried out at age 11 years. MRI with harmonized scan parameters will be performed at Aarhus University, Center for Integrative Neuroscience (CFIN) and Hvidovre Hospital, Danish Research Center for Magnetic Resonance (DRCMR). We will acquire three-dimensional high-resolution MP2RAGE structural scans and diffusion-weighted MRI to derive, respectively, measures of brain structure, including global and regional cortical thickness, area, volume, and gyrification; subcortical brain structure (and microstructure); and myelin sensitive brain tissue maps and microstructural properties of gray and white matter brain tissue (e.g., fractional anisotropy, mean diffusivity), as well as measures of structural connectivity by means of, for example, tractography and structural covariance. Task-related functional brain activity and connectivity will be assessed while participants perform well-established paradigms as in the VIA 11 study, that is, Eriksen Flanker Task (88) and the Social Cognition Task from the Human Connectome Project, that is, Animated Triangles Test (59, 60), which, respectively, probe executive cognitive control (89) (i.e., distractor resistance during fast response choices cued by directional cues) and social cognition (i.e., inferring the intentionality of moving objects[1]). In addition, and new to the VIA 15 study, we have included a reward paradigm. In the reward paradigm, participants start out with 100 DKK and are then repeatedly presented with two different stimuli in random order. Each stimulus presentation is accompanied with varying outcomes adding or subtracting to their current wealth. Participants thus can learn about the

---

[1] Note that The Animated Triangles Task and the social cognition paradigm from The Human Connectome Project are not exactly the same. The latter both contain triangles, squares, and circles. However, the premise is the same (the figures are moving around either in an intentionally or random manner).

reward probability distributions of the two stimuli. The aim is to investigate whether the dopaminergic reward system represents the entire reward probability distribution, as recently suggested by an experiment in mice (90), and whether this neural distribution is changed in the high-risk groups. We have chosen these tasks because task-related networks are hypothesized to be implicated in the pathophysiology of neurodevelopmental disorders. Functional profiling of these brain systems will enable us to infer specific network properties and dynamics that contribute to disease formation or resilience.

EEG (DRCMR only). We will repeat the EEG assessments performed in the VIA 11 study. Specifically, an auditory oddball paradigm to measure Mismatch negativity (91) and an auditory paradigm (using 40-Hz click trains) to measure steady-state oscillations (92) will be used. In addition, we will repeat the Eriksen Flanker task that is both performed during the fMRI and EEG.

By combining fMRI and EEG data (although not acquired concurrently), we will be able to get a deeper understanding of lower-order processing as well as the interaction of specific brain regions during the emerging of psychopathology, on the one hand, and cognitive control, on the other hand, during this age period.

Magnetoencephalography (CFIN only). We will perform magnetoencephalographic (MEG) recordings of all participants assessed at the Aarhus study site, expecting a total sample size of 175–200. We will collect MEG data using the ELEKTA Neuromag TRIUX MEG system with 204 planar gradiometers and 102 magnetometers. Like EEG, MEG measures brain activity with high temporal resolution; however, MEG can achieve slightly higher spatial resolution compared with EEG. As for the EEG recordings, we will apply two auditory paradigms: the roving auditory oddball paradigm (to elicit mismatch negativity) and the 40-Hz auditory steady-state response, to investigate evoked and induced responses, respectively. Both paradigms are well-replicated in patients with both schizophrenia and bipolar disorder, with medium–large effect sizes, compared with healthy controls. Our MEG data can subsequently be combined with T1-weighted structural images from MRI scans for source localization. We will investigate effective connectivity within and between brain regions using dynamic causal modeling, which will allow us not only to investigate the clinical usefulness of two putative biomarkers for schizophrenia and bipolar disorder, but also to investigate the pathophysiological trajectory leading from a familial high-risk state to manifest illness.

Polysomnography

Polysomnography (PSG) is a noninvasive EEG-based method, considered the gold standard of sleep analysis, and widely applied both in clinical practice and for research purposes (93). We will examine the sleep pattern and sleep stage architecture of participants with PSG. For PSG recordings, we will apply a portable recording device, the Somnomedics Somno HD with the 32-channel Somnomedics EEG+ headbox attached to capture EEG signals from the scalp, electrocardiographic signal from the chest, electromyographic signals from the chin and thigh and electro-oculographic signals from the outer lateral canthus left and right sides. Electrodes will be placed according to the American Academy of Sleep Medicine guidelines for extended EEG montage. Trained personnel will fit the PSG equipment on location in each participant's home. Participants will wear the PSG equipment for one night at home, sleeping as normal. Next morning, after the recording, participants remove the equipment and store it for collection by our staff. Following the PSG recording, participants must complete the Pittsburgh Sleep Quality Index (94). Except for potential mild discomfort from sleeping with the equipment, there are no known adverse effects or complications to the method.

Data will be analyzed in order to score the expression of the various sleep stages based on the complete recording period, to produce a hypnogram for each participant. The occurrence of individual sleep spindles (95) and K-Complex' (96) in the EEG recording will be marked for each participant as well.

Genetic and Epigenetic Analyses

DNA samples were obtained from a subset of the VIA study sample, which included both parents and children. These were genotyped on the Illumina PsychChip v1-1_15073391_C. The genetic data were subject to quality control measures adapted for a family-based sample, as outlined in our previous papers (97, 98). Genetic analyses include family-based GWASs and analyses for the detection of parent-of-origin effects as well as generation of polygenic risk scores for use in downstream studies either directly or to account for genetic predisposition to an array of traits, including psychiatric disorders (e.g., schizophrenia) and physiological traits (e.g., body mass index [BMI]).

The VIA 7-11-15 studies have the unique opportunity to study neonatal epigenetic signatures from birth through childhood and adolescence toward development of mental disorders diagnosed later in life and integrate them with genetic and environmental data. We will additionally assay DNA methylation for all 522 children in peripheral samples collected at birth from dried and saved bloodspots (phenylketonuria test made at birth and stored for all children in Denmark) and at all three follow-up visits (the VIA 7 study, the VIA 11 study, and the VIA 15 study) to provide longitudinal assessment of epigenetic changes from birth and during child–adolescent development. Genome-wide DNA methylation will be assayed with the use of Infinium Methylation EPIC BeadChip (tagging 850,000 sites across the genome).

This epigenetic data will be subjected to stringent quality control and data processing using well-established Bioconductor packages (99–101). In order to account for cellular heterogeneity and reduce the confounding in the sample, we will predict blood cell proportions from the epigenetic data and further adjust for these measures in our association models (102). We will perform cross-sectional epigenome-wide association analyses to identify epigenetic markers of brain structure and activation, as well as social cognition, language, olfactory function, measures of hormones, and immune function. We will also investigate interaction scenarios between DNA methylation, genetics, and environmental exposures with measures of brain structure and functioning as outcome.

Primary Caregiver Assessment

The primary caregiver is the actual caregiver and defined as an adult who knows the adolescent very well, lives with the adolescent, or has daily contact with and is caring for the young person and who can thus provide reliable information. The primary caregiver will be asked to participate in an anamnestic semistructured interview concerning the previous 4 years (since the assessment at age 11 years, the VIA 11 study) about development, school performance, and daily behavior of the adolescent. The primary caregiver will also be asked to provide information about the adolescent's mental health status through the K-SADS-PL interview (62) and from a series of questionnaires (**Table 2**). Further, the primary caregiver will be asked to give a short speech sample about the adolescent, the FMSS (80). The primary caregiver will be asked about his/her daily functioning during the previous month by the interview Personal and Social Provision Scale [PSP (47)], and the adult will be asked to fill in a questionnaire about the family functioning by the FAD (79) (**Table 2**).

Teacher as Informant

If the parents give permission and the adolescent accepts it, a series of five questionnaires will be sent to the school teacher to ensure information from school: the SRS (69) the mADHD-RS (64), Teacher's Reports Form [similar to CBCL (70)], and BRIEF (68). See also **Table 3**.

Practical Issues

The dropout rate between the first and the second wave, the VIA 7 and the VIA 11 study was only 11%, and we believe that this has to do with our aim and great effort to meet each family with a friendly and flexible approach when arranging their participation. Therefore, as before, testing can be conducted over several days and take place at time points and places that suit the adolescents' needs and the families' specific preferences. If there are any special tests, interviews, or questionnaires that the informants for some reason do not want to take part in, this is always respected and will not lead to exclusion from the study. Transportation and catering are arranged in collaboration with the family. All participants will receive gift cards for their time taken, and travel reimbursement is offered.

Both the adolescent and the primary caregiver will be offered a verbal feedback with the conclusions from the assessments completed. Participation in the study does not include any interventions or treatment. In case of obvious needs for psychiatric treatment, or medical or psychological assistance, the adolescent (and the parents if the adolescent allows it) will be guided in how to find relevant assistance or help. In cases where referral to secondary mental health service system (i.e., hospital treatment) is urgent, we will make the referral immediately. Health professionals including researchers are obliged to make referrals to the Child Protection Services in the municipalities, when needed (in some cases without consent if the problems revealed are very serious). When milder problems arise during the assessment, the researchers will give the adolescent a list of public and nongovernmental organizations (NGOs) and institutions, which can be contacted without referral, including telephone counseling, chat forums, and open-door services.

Funding

The VIA 15 study has received financial support from The Lundbeck Foundation: 20 million DKK (∼2.6 million euros), The Novo Foundation: 10 million DKK, Mental Health Services, and Capital Region of Denmark: 10 million DKK (∼1.3 million euros). Further financial support is currently being sought to cover extra costs including delays caused by COVID-19 and other unforeseen events.

Ethics and Data Protection

A minor proportion of the families, who participated in the VIA 7-study, declined to participate in the VIA 11 study for various reasons (11% of all). This information is carefully registered in our cohort files. All data from the VIA 7 study are stored at Statistics Denmark and linked to register-based information about use of mental and somatic health services for parents and children [National Patient Register (46, 103)], parental education, and source and level of income (104). Data from the VIA 11 study and the VIA 15 study will also be stored at Statistics Denmark. The study protocol was approved by the ethical committee in March 2021 (Journal-nr.: H-20067908), and all guidelines and regulations for data security and data protection are being followed carefully. Data collection started on May 1, 2021, and all data are collected and stored in REDCap[2] (105).

Statistics

The analyses from the VIA 7 study have shown that the sample is large enough to show group differences of 0.25 Z scores and larger in tests of neurocognition and social cognition. The size of the sample allows for analyses of mediation via home environment or other environmental exposures from the VIA 7 and the VIA 11 studies and for latent class analyses of trajectories.

Differences between the three groups will be analyzed with multivariate and univariate analyses of variance or χ^2 test as appropriate. Between-group differences of diagnoses will be evaluated using logistic regression adjusting for the adolescent's sex. Multiple imputations will be applied with 20 imputations using a multivariate normal distribution. Multiple imputations will be followed by a standardizing of continuous data into z scores, using the control mean as reference. Mixed models, Cox regression, and latent class growth analysis will be applied in the longitudinal data analyses.

---

[2] Study data were collected and managed using REDCap electronic data capture tools hosted at Mental Health Services, Capital Region of Denmark.1 REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies, providing (1) an intuitive interface for validated data entry, (2) audit trails for tracking data manipulation and export procedures, (3) automated export procedures for seamless data downloads to common statistical packages, and (4) procedures for importing data from external sources.

RESULTS

Results will be presented within the context of both cross-sectional and longitudinal analyses, that is, comparing the results from the first assessments at ages 7 and 11 years, this time giving us the opportunity to present developmental trajectories with three time points. Results will be reported in all domains that have been included from the outset (neurocognition, psychopathology, social behavior, and social cognition and daily functioning, motor function, and family/home environment). Follow-up on the domains introduced in the VIA 11 study (i.e., MR scans, EEG recordings, SENS motion data, and blood sample data) will be of special interest. In addition, for many of the domains covered partly by questionnaires, we can at this time create trajectories and compare the three groups with the exact same instrument. Results concerning the actual indicators of subthreshold psychopathology and symptoms that meet the diagnostic criteria will be analyzed into mental health status at ages 7 and 11 years. This time, we will be able to include data on deliberate self-harm, risk-taking behavior including alcohol and drug use, reports of current or previous experiences of social exclusion, or bullying and physical health (immune system status, BMI, sleep, etc.). Finally, we will be able to investigate how differences and/or changes in structural and functional brain readouts are related to differences and/or changes in clinical and behavioral measures and how these are modulated and/or mediated by biological and environmental factors.

For all domains, we have strived to use instruments that can be used for a wide age span. We therefore have a huge amount of data with similar methods, and analyses will take into account to what extent these children's deficits or advantages measured at ages 7 and 11 years remain stable, deteriorate, or diminish over time.

DISCUSSION

In this article, we have described the outline for the third wave of The Danish High-Risk and Resilience Study—VIA 15, a follow-up study on 522 children born in Denmark, most of them with a familial predisposition for schizophrenia or bipolar disorder. We aim to follow up on all the central domains that have already been thoroughly investigated at ages 7 and 11 years, and thus, we will be able to demonstrate trajectories for both good and poor outcomes and at-risk states in adolescence. The overall purpose is to contribute to the existing knowledge about etiology and development of mental illness and to propose optimal time points and domains or specific profiles relevant and especially targeted preventive strategies and early interventions for offspring with familial risk for severe mental illness.

There are other research groups around the world, who are also assessing children with familial risk for both schizophrenia and bipolar disorder, and some also include children born to parents with moderate to severe depression (106). Those who are closest to our study are the Bipolar and Schizophrenia Young Offspring Study in Spain (107) and the Families Overcoming Risks and Building Opportunities for Well-being Study in Canada (108). The latter is also testing different models of interventions, while investigating antecedents, symptom development, and behavior while in the same study testing different models of interventions. These and other familial high-risk studies have confirmed what earlier high-risk studies revealed, namely, that the increased risk for the offspring to be ill is not specific for the illness of the parent, but rather is seen as a generally increased risk for developing any mental disorder (25). Further, research has shown that a developmental perspective is needed when trying to disentangle, understand, and interpret the importance of unspecific and early mental health problems and subthreshold symptoms in terms of seeing these early signs as markers of emerging psychiatric disorders (109, 110).

The Danish High-Risk and Resilience Study—VIA 15 also implies some challenges that we are completely aware of. Of utmost importance is the willingness of the families to participate again. From the former waves, we already learned that practical issues and logistics such as arranging transportation and remembering exact time and meeting point can be troublesome especially for the families, who struggle with ongoing or acute episode of mental illness. In the VIA 11 study, we saw that some of the children already at that age had developed various mental problems that could make further participation difficult for them (111). A 15-year-old teenager will have more influence on the decision, and if he/she is reluctant, the primary caregivers may not want to force/put pressure on him/her. On the other hand, at age 15 years, the adolescents are familiar with the study from the former waves, and many of them expressed that they liked being part of it. Most of them will still be in elementary school, and not in high school, which may also make participation a bit easier to find time for.

For domains such as psychopathology, risk behavior, and social relations, we know that being 15 years of age implies some very specific behavioral patterns and social processes that we need to be aware about and well-educated to capture and document. For example, we expect that adolescents at age 15 years will present with mental health issues that include symptoms from many different diagnostic entities and when an exact diagnosis can be hard to determine (112). This age group often present with many mild to moderate transdiagnostic symptoms pointing in different directions (113). For example, mood swings, deliberate self-harm, isolation, and some irregular alcohol use can be both normal teenager problems and signs of underlying mental disorder. Therefore, this must be scrutinized in order to avoid overdiagnosing or underdiagnosing. Some of the young people may also describe some more subjective changes in sensory functions such as heightened perceptions of light or sound and self-disturbances that may be early warnings of later psychotic illness, which to some degree is covered by the questionnaire YETI (66) but not in the K-SADS-PL (62).

If the current situation is evaluated to be acutely unsafe and the adolescent's health situation is at risk, we will offer a statutory referral to the municipality's Child Protection Services or to the Center for Child and Adolescent Mental Health Services, depending on the type of problem presented. For less severe or acute cases (e.g., few occasions of deliberate self-harm that happened some months ago), we will hand out a list of low threshold, that is, easy to access and open-door services and organizations, which can be contacted for all kinds of unspecific problems with school, parents, friends, and peers, to get advice and support (e.g., headspace and other NGOs, general practitioners, municipality's open office, telephone counseling, and chat forums). We will also provide feedback on the test results to both the adolescent and the parents if they wish.

For some of the adolescents with familial high risk for mental disorder, the transition to adulthood can be troublesome for other reasons than those shared by everyone. Being a relative to a person with a severe mental illness can seriously impact daily life functioning and behavior, both for the other parent, often identified as the "well parent," and for the children. The process of finding one's own identity, being more independent, expressing oppositional viewpoints, and separating from the home and the family structure is a natural process in this phase of life. But in families with parental mental illness, these processes can be much more difficult, if the adolescent at the same time has a huge responsibility for practical tasks in the family or for the emotional and psychological well-being of the parent. Many adolescents worry about what will happen to their ill parent, if they stay out long or even leave home 1 day, and some have a very close relationship to the parent, which makes it difficult to be an individual without thinking about the parent's needs (114).

Potential Clinical Implications

Prevention and early intervention are important and possible and are being developed and tested in many areas of psychiatry (115). Children born to parents with severe mental illness have been overlooked and "fallen between chairs," but longitudinal studies such as this can help change that. Early intervention programs can be developed and tested in accordance with knowledge about the children's developmental trajectories and early signs of mental illness with specific focus on various outcomes. A recent review of intervention studies targeting children with familial risk for mental disorder documented that it is possible to influence the risk profiles of the individuals by rather simple, general, or short interventions (116). Risk of mental illness was reduced as were both internalizing and externalizing symptoms. Interventions varied a lot but were primarily cognitive behavioral therapy, parental training, and psychoeducation. Some of the factors that children born to parents with mental illnesses live with are shared with children in families, where a parent has a serious somatic disorder, such as worrying about the parent, being a "young carer." Other similarities include having a hospitalized parent, whereas other factors are more specific (change of the personality, emotions, and behavior and high levels of stigma). When children are relatives, the parent's illness poses a risk on their environment, which could be further included in treatment and prevention strategies by providing information and knowledge to the children about the parent's situation (117).

One of our long-term goals of the comprehensive study is to be able to—on the basis of the results from the three measurements—detect the most vulnerable individuals by assessing their profiles at an early time point and use this knowledge to inform intervention studies and develop specialized interventions that are directed against the specific problems or symptoms that they display. However, most of the knowledge about preventive interventions for children and adolescents emphasize the importance of also including parents and other important adults around the child/adolescent at risk to have a more holistic approach. Also, school, social environment, access to leisure time activities, and local communities have a potentially important role in providing options for resilience and self-esteem (115).

CONCLUSION

Longitudinal studies are time- and resource-consuming but have a major potential for highlighting developmental processes for individuals with familial risk of severe mental illness such as schizophrenia and bipolar disorder. The large and unique cohort of 522 individuals in The Danish High-Risk and Resilience Study has already provided striking results in terms of higher rates of early markers of vulnerability, developmental delays, and clinical problems compared with population-based controls. The cohort is now being followed up for the third time to inform preventive strategies and early interventions in the future.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of the Capital Region of Denmark. Written informed consent was obtained from the individuals and minors' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

AT and MNo wrote the manuscript. All authors contributed to development of study design and preparation of the manuscript, and have commented and approved the submitted version.

FUNDING

This work was funded by Lundbeckfonden—R-277-2018-594, MEG part: Lundbeckfonden—R322-2019-2711, Novo Nordisk Fonden—NNF20OC0060468, and Region Hovedstadens Psykiatris Forskningspulje.

ACKNOWLEDGMENTS

We would like to thank Physiotherapist Camilla Tjott, Child and Adolescent Mental Health Center, Capital Region of Denmark, Denmark. Psychologist, Ph.D. Jens Richard Møller Jepsen, Clinical Intervention and Neuropsychiatric Schizophrenia Research, Mental Health Services, Glostrup, Capital Region of Denmark, Denmark. Professor Carsten B Pedersen, Department of Economics and Business Economics—CIRRAU—Center for Integrated Register-based Research, Aarhus, Denmark. MSc Marianne Gjørz Pedersen, Department of Economics and Business Economics—National Center for Register-based Research, Aarhus, Denmark. Secretary Hanne Junge Larsen, CORE, Copenhagen Research Unit, Mental Health Center, Capital Region of Denmark.

REFERENCES

1. Hameed MA, Lewis AJ. Offspring of parents with schizophrenia: a systematic review of developmental features across childhood. *Harv Rev Psychiatry.* (2016) 24:104–17. doi: 10.1097/HRP.0000000000000076
2. Birmaher B, Axelson D, Monk K, Kalas C, Goldstein B, Hickey MB, et al. Lifetime psychiatric disorders in school-aged offspring of parents with bipolar disorder: the Pittsburgh Bipolar Offspring study. *Arch Gen Psychiatry.* (2009) 66:287–96. doi: 10.1001/archgenpsychiatry.2008.546
3. Berenbaum SA, Beltz AM, Corley R. The importance of puberty for adolescent development: conceptualization and measurement. *Adv Child Dev Behav.* (2015) 48:53–92. doi: 10.1016/bs.acdb.2014.11.002
4. Blakemore SJ. Development of the social brain during adolescence. *Q J Exp Psychol (Hove).* (2008) 61:40–9. doi: 10.1080/17470210701508715
5. Vijayakumar N, Op de Macks Z, Shirtcliff EA, Pfeifer JH. Puberty and the human brain: Insights into adolescent development. *Neurosci Biobehav Rev.* (2018) 92:417–36. doi: 10.1016/j.neubiorev.2018.06.004
6. Bick J, Nelson CA. Early experience and brain development. *Wiley Interdiscip Rev Cogn Sci.* (2017) 8:1–2. doi: 10.1002/wcs.1387
7. Kilford EJ, Garrett E, Blakemore SJ. The development of social cognition in adolescence: an integrated perspective. *Neurosci Biobehav Rev.* (2016) 70:106–20. doi: 10.1016/j.neubiorev.2016.08.016
8. Sydnor VJ, Larsen B, Bassett DS, Alexander-Bloch A, Fair DA, Liston C, et al. Neurodevelopment of the association cortices: Patterns, mechanisms, and implications for psychopathology. *Neuron.* (2021) 109:2820–46. doi: 10.1016/j.neuron.2021.06.016
9. Shaw P, Kabani NJ, Lerch JP, Eckstrand K, Lenroot R, Gogtay N, et al. Neurodevelopmental trajectories of the human cerebral cortex. *J Neurosci.* (2008) 28:3586–94. doi: 10.1523/JNEUROSCI.5309-07.2008
10. Casey B, Jones RM, Somerville LH. Braking and accelerating of the adolescent brain. *J Res Adolesc.* (2011) 21:21–33. doi: 10.1111/j.1532-7795.2010.00712.x
11. Amlien IK, Fjell AM, Tamnes CK, Grydeland H, Krogsrud SK, Chaplin TA, et al. Organizing Principles of Human Cortical Development–Thickness and Area from 4 to 30 Years: Insights from Comparative Primate Neuroanatomy. *Cereb Cortex.* (2016) 26:257–67. doi: 10.1093/cercor/bhu214
12. Walhovd KB, Fjell AM, Giedd J, Dale AM, Brown TT. Through Thick and Thin: a Need to Reconcile Contradictory Results on Trajectories in Human Cortical Development. *Cereb Cortex.* (2017) 27:1472–81. doi: 10.1093/cercor/bhv301
13. Lebel C, Beaulieu C. Longitudinal development of human brain wiring continues from childhood into adulthood. *J Neurosci.* (2011) 31:10937–47. doi: 10.1523/JNEUROSCI.5302-10.2011
14. Dow-Edwards D, MacMaster FP, Peterson BS, Niesink R, Andersen S, Braams BR. Experience during adolescence shapes brain development: From synapses and networks to normal and pathological behavior. *Neurotoxicol Teratol.* (2019) 76:106834. doi: 10.1016/j.ntt.2019.106834
15. Paus T, Keshavan M, Giedd JN. Why do many psychiatric disorders emerge during adolescence? *Nat Rev Neurosci.* (2008) 9:947–57. doi: 10.1038/nrn2513
16. Pfeifer JH, Allen NB. Puberty initiates cascading relationships between neurodevelopmental, social, and internalizing processes across adolescence. *Biol Psychiatry.* (2021) 89:99–108. doi: 10.1016/j.biopsych.2020.09.002
17. Kundakovic M, Champagne FA. Early-life experience, epigenetics, and the developing brain. *Neuropsychopharmacology.* (2015) 40:141–53. doi: 10.1038/npp.2014.140
18. Rutten BP, Mill J. Epigenetic mediation of environmental influences in major psychotic disorders. *Schizophr Bull.* (2009) 35:1045–56. doi: 10.1093/schbul/sbp104
19. Foulkes L, Blakemore SJ. Is there heightened sensitivity to social reward in adolescence? *Curr Opin Neurobiol.* (2016) 40:81–5. doi: 10.1016/j.conb.2016.06.016
20. Andrews JL, Ahmed SP, Blakemore SJ. Navigating the social environment in adolescence: the role of social brain development. *Biol Psychiatry.* (2021) 89:109–18. doi: 10.1016/j.biopsych.2020.09.012
21. Crone EA, van Duijvenvoorde AC, Peper JS. Annual research review: neural contributions to risk-taking in adolescence–developmental changes and individual differences. *J Child Psychol Psychiatry.* (2016) 57:353–68. doi: 10.1111/jcpp.12502
22. Patel PK, Leathem LD, Currin DL, Karlsgodt KH. Adolescent Neurodevelopment and Vulnerability to Psychosis. *Biol Psychiatry.* (2021) 89:184–93. doi: 10.1016/j.biopsych.2020.06.028
23. Leibenluft E, Barch DM. Adolescent brain development and psychopathology: introduction to the special issue. *Biol Psychiatry.* (2021) 89:93–5. doi: 10.1016/j.biopsych.2020.11.002
24. Patel V, Saxena S, Lund C, Thornicroft G, Baingana F, Bolton P, et al. The Lancet Commission on global mental health and sustainable development. *Lancet.* (2018) 392:1553–98. doi: 10.1016/S0140-6736(18)31612-X
25. Rasic D, Hajek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder, and major depressive disorder: a meta-analysis of family high-risk studies. *Schizophr Bull.* (2013) 40:28–38. doi: 10.1093/schbul/sbt114
26. Niemi LT, Suvisaari JM, Tuulio-Henriksson A, Lonnqvist JK. Childhood developmental abnormalities in schizophrenia: evidence from high-risk studies. *Schizophr Res.* (2003) 60:239–58. doi: 10.1016/S0920-9964(02)00234-7
27. Jablensky A, McNeil TF, Morgan VA. Barbara Fish and a Short History of the Neurodevelopmental Hypothesis of Schizophrenia. *Schizophr Bull.* (2017) 43:1158–63. doi: 10.1093/schbul/sbx094
28. Duffy A, Horrocks J, Doucette S, Keown-Stoneman C, McCloskey S, Grof P. Childhood anxiety: an early predictor of mood disorders in offspring of bipolar parents. *J Affect Disord.* (2013) 150:363–9. doi: 10.1016/j.jad.2013.04.021
29. Bora E, Ozerdem A. A meta-analysis of neurocognition in youth with familial high risk for bipolar disorder. *Eur Psychiatry.* (2017) 44:17–23. doi: 10.1016/j.eurpsy.2017.02.483
30. Agnew-Blais J, Seidman LJ. Neurocognition in youth and young adults under age 30 at familial risk for schizophrenia: a quantitative and qualitative review. *Cogn Neuropsychiatry.* (2013) 18:44–82. doi: 10.1080/13546805.2012.676309
31. Hemager N, Plessen KJ, Thorup A, Christiani C, Ellersgaard D, Spang KS, et al. Assessment of Neurocognitive Functions in 7-Year-Old Children at Familial High Risk for Schizophrenia or Bipolar Disorder: the danish high risk and resilience study VIA 7. *JAMA Psychiatry.* (2018) 75:844–52. doi: 10.1001/jamapsychiatry.2018.1415
32. Spang KS, Ellersgaard D, Hemager N, Christiani CJ, Burton BK, Greve AN, et al. Executive functions in 7-year-old children of parents with schizophrenia or bipolar disorder compared with controls: the danish high risk and resilience study-VIA 7, a population-based cohort study. *Eur Child Adolesc Psychiatry.* (2021) 30:1871–84. doi: 10.1007/s00787-020-01650-0
33. Burton BK, Vangkilde S, Petersen A, Skovgaard LT, Jepsen JR, Hemager N, et al. Sustained Attention and Interference Control Among 7-Year-Old Children With a Familial High Risk of Schizophrenia or Bipolar Disorder-A Nationwide Observational Cohort Study. *Biol Psychiatry Cogn Neurosci Neuroimaging.* (2018) 3:704–12. doi: 10.1016/j.bpsc.2018.04.012
34. Christiani CJ, Jepsen JRM, Thorup A, Hemager N, Ellersgaard D, Spang KS, et al. Social Cognition, Language, and Social Behavior in 7-Year-Old Children at Familial High-Risk of Developing Schizophrenia or Bipolar Disorder: The Danish High Risk and Resilience Study VIA 7-A Population-Based Cohort Study. *Schizophr Bull.* (2019) 45:1218–30. doi: 10.1093/schbul/sbz001
35. Burton BK, Thorup AAE, Jepsen JR, Poulsen G, Ellersgaard D, Spang KS, et al. Impairments of motor function among children with a familial risk of schizophrenia or bipolar disorder at 7 years old in Denmark: an observational cohort study. *Lancet Psychiatry.* (2017) 4:400–8. doi: 10.1016/S2215-0366(17)30103-7
36. Ellersgaard D, Gregersen M, Ranning A, Haspang TM, Christiani C, Hemager N, et al. Quality of life and self-esteem in 7-year-old children with familial high risk of schizophrenia or bipolar disorder: the Danish High Risk and Resilience Study-VIA 7-a population-based cohort study. *Eur Child Adolesc Psychiatry.* (2020) 29:849–60. doi: 10.1007/s00787-019-01397-3
37. Ellersgaard D, Gregersen M, Spang KS, Christiani C, Burton BK, Hemager N, et al. Psychotic experiences in seven-year-old children with familial high risk of schizophrenia or bipolar disorder in: The Danish High Risk and Resilience Study - VIA 7; A population-based cohort study. *Schizophr Res.* (2021) 228:510–18. doi: 10.1016/j.schres.2020.11.045

38. Ellersgaard D, Jessica Plessen K, Richardt Jepsen J, Soeborg Spang K, Hemager N, Klee Burton B, et al. Psychopathology in 7-year-old children with familial high risk of developing schizophrenia spectrum psychosis or bipolar disorder - The Danish High Risk and Resilience Study - VIA 7, a population-based cohort study. *World Psychiatry.* (2018) 17:210–9. doi: 10.1002/wps.20527
39. Gantriis DL, Thorup AAE, Harder S, Greve AN, Henriksen MT, Zahle KK, et al. Home visits in the Danish High Risk and Resilience Study - VIA 7: assessment of the home environment of 508 7-year-old children born to parents diagnosed with schizophrenia or bipolar disorder. *Acta Psychiatr Scand.* (2019) 140:126–34. doi: 10.1111/acps.13057
40. Thorup AA, Jepsen JR, Ellersgaard DV, Burton BK, Christiani CJ, Hemager N, et al. The Danish High Risk and Resilience Study - VIA 7 - a cohort study of 520 7-year-old children born of parents diagnosed with either schizophrenia, bipolar disorder or neither of these two mental disorders. *BMC Psychiatry.* (2015) 15:233. doi: 10.1186/s12888-015-0616-5
41. Insel TR. Rethinking schizophrenia. *Nature.* (2010) 468:187–93. doi: 10.1038/nature09552
42. Johnsen LK, Ver Loren van Themaat AH, Larsen KM, Burton BK, Baaré WFC, Madsen KS, et al. Alterations in Task-Related Brain Activation in Children, Adolescents and Young Adults at Familial High-Risk for Schizophrenia or Bipolar Disorder - a systematic review. *Front Psychiatry.* (2020) 11:632. doi: 10.3389/fpsyt.2020.00632
43. Collin G, Scholtens LH, Kahn RS, Hillegers MHJ, van den Heuvel MP. Affected Anatomical Rich Club and Structural-Functional Coupling in Young Offspring of Schizophrenia and Bipolar Disorder Patients. *Biol Psychiatry.* (2017) 82:746–55. doi: 10.1016/j.biopsych.2017.06.013
44. Thorup AAE, Hemager N, Sondergaard A, Gregersen M, Prosch AK, Krantz MF, et al. The Danish High Risk and Resilience Study-VIA 11: Study Protocol for the First Follow-Up of the VIA 7 Cohort—522 Children Born to Parents With Schizophrenia Spectrum Disorders or Bipolar Disorder and Controls Being Re-examined for the First Time at Age 11. *Front Psychiatry.* (2018) 9:661. doi: 10.3389/fpsyt.2018.00661
45. Pedersen CB, Gøtzsche H, Møller JØ, Mortensen PB. The Danish Civil Registration System - A cohort of 8 million people. *Dan Med Bull.* (2006) 53:441–9.
46. Mors O, Perto GP, Mortensen PB. The Danish Psychiatric Central Research Register. *Scand J Public Health.* (2011) 39(7 Suppl):54–7. doi: 10.1177/1403494810395825
47. Morosini P, Magliano L., Brambilla L, Ugolini S, Pioli R. PSP - Personal and Social Performance Scale. Acta Psychiatrica Scandinavica Development,reliability and acceptability of a new version of the DSM-IV social and occupational scales (SOFAS) to assess routine social functioning. (2000) 101:323–9.
48. Sparrow SS, Balla DA, Cicchetti DV. *Vineland*. Circle Pines, MN: American Guidance Service (1984).
49. Henderson SE, Sugden DA, Barnett AL. *Movement Assessment Battery for Children*. 2nd ed. London, UK: The Psychological Corporation (2007).
50. Shaffer D, Gould MS, Brasic J, Ambrosini P, Fisher P, Bird H, et al. A children's global assessment scale (CGAS). *Arch Gen Psychiatry.* (1983) 40:1228–31. doi: 10.1001/archpsyc.1983.01790100074010
51. Marquardt C, Mai N. A computational procedure for movement analysis in handwriting. *J Neurosci Methods.* (1994) 52:39–45. doi: 10.1016/0165-0270(94)90053-1
52. Meyers JE, Meyers KE. *Rey Complex Figure Test and Recognition Trial: Professional Manual.* Odessa, FL: Psychological Assessment Resources (1995).
53. CANTAB. *Cambridge Neuropsychological Automated Test Battery*. Cambridge Cognition Ldt. Available online at: website: http://www.cantab.com CANTAB®[Cognitive assessment software]. Cambridge Cognition (2019). All rights reserved.
54. Delis DC, Kaplan E, Kramer, J.H. *D-KEFS*. San Antonio, TX: The Psychological Corporation (2001).
55. Wechsler D. *WISC-IV The Wechsler Intelligence Scale for Children—4th ed.* San Antonio: The psychological Corporation (2003).
56. Reynolds CR, Voress JK. *Test of Memory and Learning-Second Edition (TOMAL-2)*. Austin, TX: PRO-ED (2007).
57. Reynolds CR, Kamphaus RW. *Reynolds Intellectual Assessment Scales Second Edition*. (2003). Available online at: https://www.parinc.com/Products/Pkey/365
58. Doty RL. B-SIT. *wwwsensonicscom.* (2007).
59. Abell F, Happe F, Frith U. Do triangles play tricks? Attribution of mental states to animated shapes in normal and abnormal development. *Cogn Dev.* (2000) 15:1–16. doi: 10.1016/S0885-2014(00)00014-9
60. Castelli F, Happe F, Frith U, Frith C. Movement and mind: a functional imaging study of perception and interpretation of complex intentional movement patterns. *Neuroimage.* (2000) 12:314–25. doi: 10.1006/nimg.2000.0612
61. Barch DM, Burgess GC, Harms MP, Petersen SE, Schlaggar BL, Corbetta M, et al. Function in the human connectome: task-fMRI and individual differences in behavior. *Neuroimage.* (2013) 80:169–89. doi: 10.1016/j.neuroimage.2013.05.033
62. Kaufman J, Birmaher B, Brent D, Rao U, Flynn C, Moreci P, et al. Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL): initial reliability and validity data. *J Am Acad Child Adolesc Psychiatry.* (1997) 36:980–8. doi: 10.1097/00004583-199707000-00021
63. Miller TJ, McGlashan TH, Rosen JL, Cadenhead K, Cannon T, Ventura J, et al. Prodromal assessment with the structured interview for prodromal syndromes and the scale of prodromal symptoms: predictive validity, interrater reliability, and training to reliability. *Schizophr Bull.* (2003) 29:703–15. doi: 10.1093/oxfordjournals.schbul.a007040
64. Makransky G, Bilenberg N. Psychometric properties of the parent and teacher ADHD Rating Scale (ADHD-RS): measurement invariance across gender, age, and informant. *Assessment.* (2014) 21:694–705. doi: 10.1177/1073191114535242
65. Gerson AC, Gerring JP, Freund L, Joshi PT, Capozzoli J, Brady K, et al. The Children's Affective Lability Scale: a psychometric evaluation of reliability. *Psychiatry Res.* (1996) 65:189–98. doi: 10.1016/S0165-1781(96)02851-X
66. Patterson VC, Pencer A, Pavlova B, Awadia A, MacKenzie LE, Zwicker A, et al. Youth Experience Tracker Instrument: A self-report measure of developmental antecedents to severe mental illness. *Early Interv Psychiatry.* (2020). doi: 10.1111/eip.13007
67. Rimvall MK, Clemmensen L, Munkholm A, Rask CU, Larsen JT, Skovgaard AM, et al. Introducing the White Noise task in childhood: associations between speech illusions and psychosis vulnerability. *Psychol Med.* (2016) 46:2731–40. doi: 10.1017/S0033291716001112
68. Gioia GA, Isquith PK, Guy SC, Kenworthy L. Behavior rating inventory of executive function. *Child Neuropsychol.* (2000) 6:235–8. doi: 10.1076/chin.6.3.235.3152
69. Constantino JN, Gruber CP. *Social Responsiveness Scale-Second Edition (SRS-2)*. Los Angeles, CA: Western Psychological Services (2012).
70. Achenbach TM, Edelbrock, C. *CBCL, Manual for child behavior checklist and revised child behavior profile*. University of Vermont, Burlington (1983).
71. Obel C, Dalsgaard S, Stax HP, Bilenberg N. Strengths and Difficulties Questionnaire (SDQ-Dan). A new instrument for psychopathologic screening of children aged 4-16 years. *Ugeskr Laeger.* (2003) 165:462–5.
72. McConaughy SH, Achenbach TM. *Manual for the Test Observation Form for Ages 2-18*. Burlington, VT: University of Vermont (2004).
73. Ouvinen-Birgerstam P. *Sådan er jeg/This is me*. Manual, Hogrefe (2000).
74. Gratz KL, Latzman RD, Young J, Heiden LJ, Damon J, Hight T, et al. Deliberate self-harm among underserved adolescents: the moderating roles of gender, race, and school-level and association with borderline personality features. *Personal Disord.* (2012) 3:39–54. doi: 10.1037/a0022107
75. Gratz KL, Dixon-Gordon KL, Chapman AL, Tull MT. Diagnosis and Characterization of DSM-5 Nonsuicidal Self-Injury Disorder Using the Clinician-Administered Nonsuicidal Self-Injury Disorder Index. *Assessment.* (2015) 22:527–39. doi: 10.1177/1073191114565878
76. Centers for Disease C, Prevention, Brener ND, Kann L, Shanklin S, Kinchen S, et al. Methodology of the Youth Risk Behavior Surveillance System—2013. *MMWR Recomm Rep*. (2013) 62:1–20.

77. Zimet GD, Powell SS, Farley GK, Werkman S, Berkoff KA. Psychometric characteristics of the multidimensional scale of perceived social support. *J Pers Assess.* (1990) 55:610–7. doi: 10.1207/s15327752jpa5503&4_17
78. Liebenberg L, Ungar M, LeBlanc JC. The CYRM-12: a brief measure of resilience. *Can J Public Health.* (2013) 104:e131–5. doi: 10.1007/BF03405676
79. Epstein N, Baldwin L, Bishop D. *The McMaster family assessment device* (1983) 2:171–180.
80. Daley D, Sonuga-Barke EJ, Thompson M. Assessing expressed emotion in mothers of preschool AD/HD children: psychometric properties of a modified speech sample. *Br J Clin Psychol.* (2003) 42(Pt 1):53–67. doi: 10.1348/014466503762842011
81. Bernstein DP, Stein JA, Newcomb MD, Walker E, Pogge D, Ahluvalia T, et al. Development and validation of a brief screening version of the Childhood Trauma Questionnaire. *Child Abuse Negl.* (2003) 27:169–90. doi: 10.1016/S0145-2134(02)00541-0
82. Kongerslev MT, Bach B, Rossi G, Trauelsen AM, Ladegaard N, Lokkegaard SS, et al. Psychometric validation of the Childhood Trauma Questionnaire-Short Form (CTQ-SF) in a Danish clinical sample. *Child Abuse Negl.* (2019) 94:104026. doi: 10.1016/j.chiabu.2019.104026
83. Bartholdy C, Gudbergsen H, Bliddal H, Kjaergaard M, Lykkegaard KL, Henriksen M. Reliability and Construct Validity of the SENS Motion(R) Activity Measurement System as a Tool to Detect Sedentary Behaviour in Patients with Knee Osteoarthritis. *Arthritis.* (2018) 2018:6596278. doi: 10.1155/2018/6596278
84. Marshall WA, Tanner JM. Variations in pattern of pubertal changes in girls. *Arch Dis Child.* (1969) 44:291–303. doi: 10.1136/adc.44.235.291
85. Marshall WA, Tanner JM. Variations in the pattern of pubertal changes in boys. *Arch Dis Child.* (1970) 45:13–23. doi: 10.1136/adc.45.239.13
86. Budtz-Lilly A, Fink P, Ornbol E, Vestergaard M, Moth G, Christensen KS, et al. A new questionnaire to identify bodily distress in primary care: The 'BDS checklist'. *J Psychosom Res.* (2015) 78:536–45. doi: 10.1016/j.jpsychores.2015.03.006
87. Carstensen TBW, Ornbol E, Fink P, Pedersen MM, Jorgensen T, Dantoft TM, et al. Detection of illness worry in the general population: A specific item on illness rumination improves the Whiteley Index. *J Psychosom Res.* (2020) 138:110245. doi: 10.1016/j.jpsychores.2020.110245
88. Eriksen BA, Eriksen CW. Effects of noise letters upon the identifaction of a target letter in a nonsearch task. *Percept Psychophys.* (1974) 16:143–9. doi: 10.3758/BF03203267
89. Hillebrandt H, Friston KJ, Blakemore SJ. Effective connectivity during animacy perception–dynamic causal modelling of Human Connectome Project data. *Sci Rep.* (2014) 4:6240. doi: 10.1038/srep06240
90. Dabney W, Kurth-Nelson Z, Uchida N, Starkweather CK, Hassabis D, Munos R, et al. A distributional code for value in dopamine-based reinforcement learning. *Nature.* (2020) 577:671–5. doi: 10.1038/s41586-019-1924-6
91. Rydkjaer J, Mollegaard Jepsen JR, Pagsberg AK, Fagerlund B, Glenthoj BY, Oranje B. Mismatch negativity and P3a amplitude in young adolescents with first-episode psychosis: a comparison with ADHD. *Psychol Med.* (2017) 47:377–88. doi: 10.1017/S0033291716002518
92. Thune H, Recasens M, Uhlhaas PJ. The 40-Hz Auditory Steady-State Response in Patients With Schizophrenia: A Meta-analysis. *JAMA Psychiatry.* (2016) 73:1145–53. doi: 10.1001/jamapsychiatry.2016.2619
93. Conley S, Knies A, Batten J, Ash G, Miner B, Hwang Y, et al. Agreement between actigraphic and polysomnographic measures of sleep in adults with and without chronic conditions: a systematic review and meta-analysis. *Sleep Med Rev.* (2019) 46:151–60. doi: 10.1016/j.smrv.2019.05.001
94. Buysse DJ, Reynolds CF 3rd, Monk TH, Berman SR, Kupfer DJ. The Pittsburgh Sleep Quality Index: a new instrument for psychiatric practice and research. *Psychiatry Res.* (1989) 28:193–213. doi: 10.1016/0165-1781(89)90047-4
95. Chan MS, Chung KF, Yung KP, Yeung WF. Sleep in schizophrenia: a systematic review and meta-analysis of polysomnographic findings in case-control studies. *Sleep Med Rev.* (2017) 32:69–84. doi: 10.1016/j.smrv.2016.03.001
96. Zhang Y, Quiñones GM, Ferrarelli F. Sleep spindle and slow wave abnormalities in schizophrenia and other psychotic disorders: Recent findings and future directions. *Schizophr Res.* (2020) 221:29–36. doi: 10.1016/j.schres.2019.11.002
97. Nudel R, Christiani CAJ, Ohland J, Uddin MJ, Hemager N, Ellersgaard DV, et al. Language deficits in specific language impairment, attention deficit/hyperactivity disorder, and autism spectrum disorder: An analysis of polygenic risk. *Autism Res.* (2020) 13:369–381. doi: 10.1002/aur.2211
98. Nudel R, Christiani CAJ, Ohland J, Uddin MJ, Hemager N, Ellersgaard D, et al. Quantitative genome-wide association analyses of receptive language in the Danish High Risk and Resilience Study. *BMC Neurosci.* (2020) 21:30. doi: 10.1186/s12868-020-00581-5
99. Gentleman RC, Carey VJ, Bates DM, Bolstad B, Dettling M, Dudoit S, et al. Bioconductor: open software development for computational biology and bioinformatics. *Genome Biol.* (2004) 5:R80. doi: 10.1186/gb-2004-5-10-r80
100. Aryee MJ, Jaffe AE, Corrada-Bravo H, Ladd-Acosta C, Feinberg AP, Hansen KD, et al. Minfi: a flexible and comprehensive Bioconductor package for the analysis of Infinium DNA methylation microarrays. *Bioinformatics.* (2014) 30:1363–9. doi: 10.1093/bioinformatics/btu049
101. Fortin JP, Labbe A, Lemire M, Zanke BW, Hudson TJ, Fertig EJ, et al. Functional normalization of 450k methylation array data improves replication in large cancer studies. *Genome Biol.* (2014) 15:503. doi: 10.1186/s13059-014-0503-2
102. Houseman EA, Accomando WP, Koestler DC, Christensen BC, Marsit CJ, Nelson HH, et al. DNA methylation arrays as surrogate measures of cell mixture distribution. *BMC Bioinformatics.* (2012) 13:86. doi: 10.1186/1471-2105-13-86
103. Andersen TF, Madsen M, Jorgensen J, Mellemkjoer L, Olsen JH. The Danish National Hospital Register. A valuable source of data for modern health sciences. *Dan Med Bull.* (1999) 46:263–8.
104. *Danmarks Statistik (The Danish National Bureau of Statistics).* Copenhagen (1991). Available online at: https://www.dst.dk
105. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)–a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform.* (2009) 42:377–81. doi: 10.1016/j.jbi.2008.08.010
106. Sandstrom A, Sahiti Q, Pavlova B, Uher R. Offspring of parents with schizophrenia, bipolar disorder, and depression: a review of familial high-risk and molecular genetics studies. *Psychiatr Genet.* (2019) 29:160–9. doi: 10.1097/YPG.0000000000000240
107. Sanchez-Gistau V, Romero S, Moreno D, de la Serna E, Baeza I, Sugranyes G, et al. Psychiatric disorders in child and adolescent offspring of patients with schizophrenia and bipolar disorder: a controlled study. *Schizophr Res.* (2015) 168:197–203. doi: 10.1016/j.schres.2015.08.034
108. Uher R, Cumby J, MacKenzie LE, Morash-Conway J, Glover JM, Aylott A, et al. A familial risk enriched cohort as a platform for testing early interventions to prevent severe mental illness. *BMC Psychiatry.* (2014) 14:344. doi: 10.1186/s12888-014-0344-2
109. Thapar A, Riglin L. The importance of a developmental perspective in Psychiatry: what do recent genetic-epidemiological findings show? *Mol Psychiatry.* (2020) 25:1631–9. doi: 10.1038/s41380-020-0648-1
110. Beauchaine TP, Constantino JN, Hayden EP. Psychiatry and developmental psychopathology: Unifying themes and future directions. *Compr Psychiatry.* (2018) 87:143–52. doi: 10.1016/j.comppsych.2018.10.014
111. Gregersen M, Søndergaard A, Brandt JM, Ellersgaard D, Rohd SB, Hjorthøj C, et al. Mental disorders in preadolescent children at familial high-risk of schizophrenia or bipolar disorder - a four-year follow-up study: The Danish High Risk and Resilience Study, VIA 11: The Danish High Risk and Resilience Study, VIA 11. *J Child Psychol Psychiatry.* (2021). doi: 10.1111/jcpp.13548 [Epub ahead of print].
112. Drabick DAG, Kendall PC. Developmental psychopathology and the diagnosis of mental health problems among youth. *Clin Psychol Sci Pract.* (2010) 17:272–80. doi: 10.1111/j.1468-2850.2010.01219.x
113. Meyer HC, Lee FS. Translating Developmental Neuroscience to Understand Risk for Psychiatric Disorders. *Am J Psychiatry.* (2019) 176:179–85. doi: 10.1176/appi.ajp.2019.19010091
114. Yamamoto R, Keogh B. Children's experiences of living with a parent with mental illness: A systematic review of qualitative studies

using thematic analysis. *J Psychiatr Ment Health Nurs.* (2018) 25:131–41. doi: 10.1111/jpm.12415

115. Arango C, Diaz-Caneja CM, McGorry PD, Rapoport J, Sommer IE, Vorstman JA, et al. Preventive strategies for mental health. *Lancet Psychiatry.* (2018) 5:591–604. doi: 10.1016/S2215-0366(18)30057-9
116. Lannes A, Bui E, Arnaud C, Raynaud JP, Revet A. Preventive interventions in offspring of parents with mental illness: a systematic review and meta-analysis of randomized controlled trials. *Psychol Med*. (2021) 51:2321–336. doi: 10.1017/S0033291721003366
117. Oja C, Edbom T, Nager A, Mansson J, Ekblad S. Informing children of their parent's illness: A systematic review of intervention programs with child outcomes in all health care settings globally from inception to 2019. *PLoS ONE.* (2020) 15:e0233696. doi: 10.1371/journal.pone.0233696

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 07 April 2022
doi: 10.3389/fpsyt.2022.801065

The ParentingWell Practice Approach: Adaptation of Let's Talk About Children for Parents With Mental Illness in Adult Mental Health Services in the United States

Joanne Nicholson[1], Miriam Heyman[2]*, Kelly English[3] and Kathleen Biebel[4]

[1] The Heller School for Social Policy and Management, Institute for Behavioral Health, Brandeis University, Waltham, MA, United States, [2] The Heller School for Social Policy and Management, Lurie Institute for Disability Policy, Brandeis University, Waltham, MA, United States, [3] Children, Youth and Family Services, Massachusetts Department of Mental Health, Boston, MA, United States, [4] Massachusetts Rehabilitation Commission, Boston, MA, United States

Background: Despite the importance of family and parent-focused practice, there has been a dearth of research on interventions for parents with mental illness. This paper describes the process and outcome of adapting an evidence-based intervention, Let's Talk about Children (LTC), in the context of adult mental health services in Massachusetts, United States.

Methods: Specific objectives included: (1) to specify the core components, functions, and principles of LTC essential to adapting the intervention (i.e., program theory), (2) to consider contextual factors related to the new setting; (3) to pre-test the adapted materials with diverse practitioners; and (4) to compile the program model and materials (i.e., the practice profile) for use by adult mental health service providers in Massachusetts. The Adaptation Team included individuals with expertise in psychiatric rehabilitation and clinical care, policymaking, program development and research, and parents. Activities occurred between 2015–2019 and included: (1) consulting with experts to specify the core elements and theory behind the selected intervention (i.e., with the LTC purveyor and international experts); (2) consulting with key stakeholders for input regarding the Massachusetts target population and context to inform adaptations (i.e., individual and group key informant interview sessions); (3) pretesting the initial adapted materials (i.e., training and coaching sessions with adult mental health practitioners); and (4) using feedback to refine and compile the final intervention manual (i.e., the ParentingWell Practice Profile). Participants reflected diverse, oftentimes multiple roles and perspectives, including those of parents with mental illness, adult children, and family members.

Results: ParentingWell is practitioner- and setting-agnostic, addresses parenting across the lifespan, fits into the routine workflow, and builds on practitioners' existing skills. Eight themes emerged, which were translated into four core elements (engage, explore, plan, access and advocate) consistent with Self-Determination Theory and four underlying principles (trauma-informed, strengths-based, family-focused,

Edited by:
Yuka Kotozaki,
Iwate Medical University, Japan

Reviewed by:
Michelle Lisa Benstead,
Deakin University, Australia
Frederike Jörg,
University Medical Center Groningen, Netherlands
Tracy Gladstone,
Wellesley College, United States

****Correspondence:***
Miriam Heyman
miriamheyman@brandeis.edu

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *24 October 2021*
Accepted: *28 February 2022*
Published: *07 April 2022*

Citation:
Nicholson J, Heyman M, English K and Biebel K (2022) The ParentingWell Practice Approach: Adaptation of Let's Talk About Children for Parents With Mental Illness in Adult Mental Health Services in the United States. Front. Psychiatry 13:801065. doi: 10.3389/fpsyt.2022.801065

culturally sensitive) in keeping with the LTC model. The ParentingWell Practice Profile operationalizes each core element and addresses the underlying principles.

Conclusion: ParentingWell makes talking about parenting and family experiences a routine part of the therapeutic conversation with adults with mental illness. Future research will test the adaptation, implementation, and impact of ParentingWell.

Keywords: parents with mental illness, adult mental health services, intervention adaptation, family-focused practice, recovery

INTRODUCTION

Family-focused practice has received increasing attention over the past decade, particularly in relation to parents with mental illness receiving care in adult mental health services (1–8). The relative lack of research into interventions for families with parents with serious mental illness has been highlighted (9). Practitioners have reported or been found to have significant deficits in relevant skills, knowledge and confidence in working with adults who are parents and their families (4, 5, 7–12). Challenges in integrating family-focused interventions into everyday routine in adult mental health are context- as well as practitioner-related (13). Contextual issues that may impede adoption or result in the adaptation of specific interventions include perceptions of workplace support (12); the need for training, mentoring, supervision and co-worker support (5); the fact that the implementation of new routines is a time and resource consuming effort (14); and the challenge of taking an open approach to the definition of family—a "whole of family" approach—in a context that is focused on the assessment and treatment of the individual (1, 6). We undertook the task of addressing these issues as we navigated the process of identifying and adapting an evidence-based intervention, Let's Talk about Children, to meet the needs of adults with serious mental illness who are parents or planning to become parents, in the context of adult mental health services in Massachusetts, United States.

Extensive groundwork for the selection and adaptation of an appropriate evidence-based practice was laid in previous research on the prevalence, experiences and needs of parents living with mental illness (15, 16), prior assessment of community capacity and needs (10, 17), and the prior identification and evaluation of existing models (18–22). Prior work was conducted in partnership with parents, practitioners, and policymakers, in the spirit of participatory action research and the mantra "nothing about us without us." We reviewed existing evidence-based interventions, selecting Let's Talk about Children (LTC), a three session, well-articulated, prescribed model developed in Finland (15, 23–26) and replicated and tested in Australia (13, 27–29), Greece (30), and Japan (31). The research background and replication of LTC in different countries were described in detail in a recent paper published in the current Frontiers special topic collection (32).

The goal of LTC is to promote parenting and child development and prevent children's mental health problems by providing their parents information and opportunity to talk about their children. The provider is trained to use a semi-structured interview tool in three or four prescribed sessions to guide the discussion about parenting to address the child's life, the parent's mental illness and its meaning for the family, development of a plan to promote the child's wellbeing and family life, and the engagement of supports and services. It is important to note that in randomized controlled trials in countries other than Finland, adaptations have been made to enable engagement with the parent (e.g., changing language to fit a parent's needs or culture), to fit the service system or model of care (e.g., delivering LTC in shorter sessions over a longer period of time), and to tailor LTC materials (e.g., to incorporate changes in questions asked) (13, 32).

In addition to specific guidelines for implementing the LTC model, authors have recommended the importance of six core and inter-related principles of family-focused practice for families living with parental mental illness that informed our work including: (1) family care planning and goal setting; (2) liaison between families and services including family advocacy; (3) instrumental, emotional and social support; (4) assessment of family members and family functioning; (5) psychoeducation; and (6) a coordinated system of care (1). Others stress the benefit of assessing strengths within families, a non-judgmental and supportive approach, transparency to build trust, and the normalization of parenting difficulties (8, 33). International efforts to extract and replicate key elements of family-focused practice and develop program theory have recently been described, with a focus on consideration of the relationships among contextual factors, action mechanisms, and impact (34).

The tension between intervention fidelity and fit as evidence-based practices are implemented in real world settings has given rise to the science of adaptation (35). In the context of innovation diffusion theory, Rogers defined adaptation as "the degree to which an innovation is changed or modified by a user in the process of its adoption and implementation" (36). Adaptation has come to represent a more active, intentional process "to make an intervention fit a specific or new use or situation often by modification" (37). Adaptation of interventions, developed and tested in more controlled conditions, has been suggested as a requirement for achieving sustainable real-world outcomes, attending to intervention fidelity while adjusting to local needs and contingencies operating in the environment (38). Aarons and colleagues further specify types of "scaling out," when evidence-based practices are adapted to new populations or new delivery systems or both (37, 39). Strategic, well-considered implementation of an evidence-based practice with a different population or in a different setting may contribute to more expeditious testing in a shorter timeframe. Strength may be "borrowed" from evidence obtained in prior effectiveness trials to the extent that core elements or core functions and forms are retained (39, 40).

Authors consistently highlight the importance of a systematic approach, both to navigating the adaptation process as well as documenting the adaptations made (35, 37, 39, 41, 42). Prior studies of intervention adaptation have focused on public health interventions including HIV prevention, teen pregnancy, and sexually transmitted infection (41). The recommendation has been made that more studies of the adaptation process to improve fit between interventions and contexts would inform adaptation strategies in the context of implementation (35).

Several models of the steps or phases in the adaptation process have been outlined (36, 37, 40–44). Common among them are steps involving exploration, preparation, implementation, and sustainment, with similar accompanying activities. Authors acknowledge that the adaptation process is not necessarily linear, but is best described as an iterative, dynamic process in which steps may overlap, with feedback loops informing next steps and refinements (36). At best, key stakeholders are engaged throughout the process, to ensure that all interests are represented, that the adapted intervention is culturally sensitive and relevant, and to promote stakeholder buy-in, thereby increasing the likelihood of successful implementation and positive outcomes (41). The ultimate goal of the adaptation process is to maintain as much fidelity to the essential ingredients of the original model as possible, while facilitating fit and feasibility with the new target population or context (42).

A multidisciplinary Adaptation Team, including researchers, practitioners, implementers, and service recipients or consumers, is recommended to guide and navigate this process (35). The first phase, exploration, generally involves assessing needs, selecting an intervention, and gathering and reviewing relevant intervention or program model materials. If possible, the developer or purveyor, and other experts are involved to ensure the Adaptation Team fully understands the selected intervention and the context in which it was originally implemented. Core elements or components (i.e., key ingredients necessary to make the intervention effective), core functions and forms (i.e., intervention activities that produce change) or best practice characteristics (i.e., characteristics common to effective programs) of the original model are identified, along with the internal logic or theory of change (36, 39–41, 44).

The second phase of the adaptation process focuses on the preparation of the adapted intervention or program model and materials (44). Common activities include the identification of mismatches between the original intervention or program model and the new context (e.g., culture, health care system, social and economic disparities), to enhance the potential fit and feasibility of the adapted model. This task may be informed by interviews with key community stakeholders to promote understanding of the contextual differences. Description of the adapted intervention model and materials (e.g., manual, training resources) may be reviewed by community partners and representatives of the target population, and feedback solicited (44).

The activities of the implementation phase generally involve the pre- or pilot testing of the adapted model, with training of staff, taking model adaptations into account. Intervention components and procedures are evaluated and refined (44). Finally, in the sustainment step, the adapted intervention is implemented and evaluated further, training and supervision provided on an ongoing basis, and a dissemination plan implemented. Attention to issues of training and ongoing technical assistance promote better intervention results (38).

The purpose of this paper is to describe the process of adapting an evidence-based intervention, Let's Talk about Children (LTC), targeting parents with mental illness receiving services in the adult mental health system in Massachusetts. The specific objectives included: (1) to explore and specify the core components, functions, and principles of LTC essential to adapting the intervention (i.e., program theory), (2) to consider contextual factors related to the new setting (i.e., practice, organizational and systemic factors); (3) to pre-test the adapted materials with diverse practitioners working with parents (i.e., in training and coaching sessions); and (4) to compile the program model and materials (i.e., the practice profile) for use by adult mental health service providers in Massachusetts. The overall project goal was to adapt LTC and clearly specify a program model for parents with mental illness that could be implemented, tested and sustained in the context of adult mental health services. We partnered with diverse stakeholders including parents with mental illness, their children, and family members to specify and adapt an appropriate model, pre-test, and refine the model for scale-up and future, larger-scale implementation, rigorous testing, and sustainment.

MATERIALS AND METHODS

A developmental evaluation design and qualitative methods provided the framework for the iterative process of exploration and innovation in adapting the LTC model (45). Cycles of data collection, reflecting, feedback and refinement were not linear, as adaptation activities informed each other in a reflective manner and changes were made based on emergent conditions and information. Consequently, findings from multiple perspectives were integrated systematically over time to inform the final program model and practice profile.

Procedures

Adaptation activities occurred between 2015–2019 and included: (1) consulting with experts with professional and lived experience to explore and specify the core elements and theory behind the selected intervention (i.e., with the LTC purveyor and international experts, in individual and group in-person and videoconference sessions); (2) consulting with key stakeholders with professional and lived experience for input regarding the Massachusetts target population and context to inform program model adaptations (i.e., individual and group stakeholder interview sessions); (3) pretesting the initial adapted materials (i.e., training and coaching sessions with adult mental health practitioners working with parents); and (4) using feedback to make further modifications and compile the final intervention manual (i.e., the ParentingWell Practice Profile). These steps were not completely linear, in that an iterative process of considering adaptations and checking back with stakeholders occurred over time, consistent with a developmental evaluation design. A core Adaptation Team met over time to facilitate the adaptation process. It is important to note that, while stakeholders may be referred to as "practitioners," for example, practitioners may reflect diverse perspectives and multiple roles and responsibilities (e.g., peer specialists who themselves are parents with mental illness).

Procedures for each phase of the project were reviewed by the relevant university and state agency institutional review boards. When activities met criteria for human subjects research *per se*, the appropriate written or verbal consents were obtained, as recommended by the institutional review boards. Stakeholders (i.e., agency staff, policymakers, parents, adult children, family members, and advocates) were volunteers who did not receive stipends for participation, as all activities took place during routine working hours as part of ongoing professional and agency activities and commitments.

The Adaptation Team

A core group served as the Adaptation Team ($n = 4$), including parents and individuals with backgrounds and expertise in psychiatric rehabilitation and clinical care, policymaking, program development and research. The Adaptation Team met in person, bi-weekly, and communicated more frequently via email and text message throughout the 4 years of the project. Detailed minutes were typed directly into electronic documents for qualitative analysis and stored in secure digital files by independent research staff who observed the meetings.

The Let's Talk About Children Purveyor and International Expert Group

The Let's Talk about Children (LTC) purveyor (Solantaus) and colleagues met quarterly in 2-h sessions over the course of 2 years (2015–2017) via video conferencing as the LTC Worldwide Group ($n = 20$). The group included purveyors, researchers and practitioners from Finland, Japan, Australia, Sweden, Italy and the United States, with professional and lived experience, who discussed implementation issues in diverse practice settings internationally. Adaptation Team members presented draft materials and Massachusetts-specific implementation considerations for input and feedback from LTC Worldwide members. In addition, individual LTC Worldwide participants provided in-depth review of draft project materials and detailed feedback. Meeting presentations, detailed minutes and reviewers' comments were transcribed into electronic documents by research staff and stored in secure digital files.

Key Massachusetts Stakeholders

Twelve individual or group interview sessions of approximately 1 to 2 h each were completed early in the project (2016) by telephone or in-person, involving a convenience sample of 70 participants with professional and lived experience recruited by telephone and email to represent the Massachusetts Department of Mental Health leadership and Planning Council, advocates from the National Alliance of Mental Illness, and two community-based agencies providing outpatient and residential mental health services to adults, and parents themselves. The agencies provide diverse mental healthcare services to 40,000–50,000 individuals and families annually with sites located in diverse geographic areas in Massachusetts. Agency representatives included practitioners (i.e., peer specialists, clinicians, case managers), supervisors, program managers and agency administrators, who participated in invited staff gatherings. Members of the Adaptation Team provided informational and draft materials for participants to review and facilitated discussion regarding topics including: (1) the experiences of parents and practitioners; (2) services currently provided; (3) challenges and unmet needs; and (4) implementation issues, current or anticipated, related to the agency and community contexts. In-person interviews took place in comfortable agency settings (e.g., a large office or conference room). Detailed verbatim notes were compiled electronically and stored in secure digital files by an independent research staff member attending meetings and using a laptop computer. A draft manual was compiled, based on the input and feedback of stakeholders to this point.

Adult Mental Health Service Practitioners

Two, 2-h in-person training sessions, held 2 weeks apart in May 2017, were conducted by members of the Adaptation Team in each of the two community-based agencies providing mental health services to adults. Practitioners were provided with the draft manual, and training content focused on information and materials in the draft manual to pretest materials. Twelve practitioners (e.g., social workers, peer specialists, case managers, supervisors) working with parents with mental illness participated. Participants then attended 41-to-2-h in-person coaching sessions, held in each agency at 1-month intervals following the training, in a comfortable conference room, facilitated by the Adaptation Team members. Participants were encouraged to describe contacts with parents served and supported in sharing suggestions for strategies to deal with challenges in service provision. Again, detailed notes were entered into document files by independent research staff members using laptop computers; documents were then stored in secure digital files.

Analysis

The goal of the project was to describe the process of intervention adaptation and compile the refined intervention model, rather than assess the impact of an intervention on practitioner or parent outcomes. Consequently, detailed background and demographic data on individuals participating were not solicited. Participants represented multiple roles and responsibilities, professional and lived experience, and their "in the moment" contributions reflected any of these. Detailed notes, systematically obtained and recorded in many diverse settings from multiple perspectives over time, were captured via laptop or transcribed where necessary into electronic documents by independent research staff, uploaded into Dedoose software to facilitate data management and coding, and reviewed systematically by members of the research team (46). The Adaptation Team and research staff debriefed after each consultation, interview, training or coaching session to review key points, add to notes as necessary to insure the thoroughness of documentation, and plan for the next data collection opportunity. Themes were identified and elaborated in content analysis of transcribed documents, across participants' perspectives, by members of the research team, experienced qualitative researchers, who met regularly to develop a shared understanding of themes and related codes. Transcripts were coded independently by members of the research staff who met to discuss and reconcile any differences, and to inform and refine subsequent coding. Themes identified through review and coding of prior sessions were explored further in subsequent sessions with diverse participants, to corroborate and elaborate data and thematic codes, and to obtain input and feedback from multiple perspectives over time (i.e., triangulation and member checking) (47). Memos were generated by research staff, describing and elaborating themes across data sources, to facilitate the identification of patterns and relationships among themes. These memos were reviewed by research staff and Adaptation Team members with professional and lived experience for further elaboration and feedback until consensus was achieved on key findings. Findings were compiled and translated into the final ParentingWell Practice Profile, to operationalize program theory and key intervention components, and provide guidelines for practitioners' interactions with parents served.

RESULTS

Findings relate to the study goal of adapting the LTC intervention to the new context and service setting. Named "ParentingWell," the core elements of LTC are retained, while shaping the practice approach to fit the service context and practitioners' recommendations. Concrete strategies for intervening are elaborated, based on the core elements and practice principles derived from LTC and translated for application in the U.S. setting.

Specifying the ParentingWell Program Theory

The ParentingWell program theory or logic model, adapted from the key elements of the Let's Talk about Children intervention, is based on Self-Determination Theory (48) with core elements of Engage and Explore (*autonomy*: identifying personal circumstances and motivation), Plan (*competence*: setting goals, assessing progress, and building self-efficacy), and Access and Advocate (*relatedness*: linking to social and professional supports and resources). Recent work by Australian colleagues corroborates the consistency of LTC underpinnings with Self-Determination Theory, that is, of the need to feel autonomous, effective and connected as drivers of the mental health recovery of parents (49). The original LTC model was found to enable practitioners to support parents with mental illness in building agency and self-regulation (49). We hypothesize practitioner outcomes to include enhanced skill and comfort, increased use of ParentingWell resources, and more frequent interactions with adults regarding parenting and family life, strengths, goals and needs, which will contribute to the model mediators of practitioner-parent alliance, hope and optimism, and supports and resources. We hypothesize proximal parent outcomes to include enhanced parent self-efficacy and reduced parenting stress. Distal outcomes include improved adult/parent wellbeing and functioning which will, in turn, contribute to and benefit from improvements in the parent-child relationship and enhanced child outcomes (see **Figure 1**).

Considering the New Context

We found numerous challenges to the implementation and spread of the adapted ParentingWell model when closely replicating the three-session LTC model. We learned that what practitioners needed and wanted, rather than a tightly prescribed intervention protocol, was a more loosely described practice approach, with well-specified principles and core elements, that fit into their routine work flow, drew from the skills and competencies they already had, was perceived as enhancing their

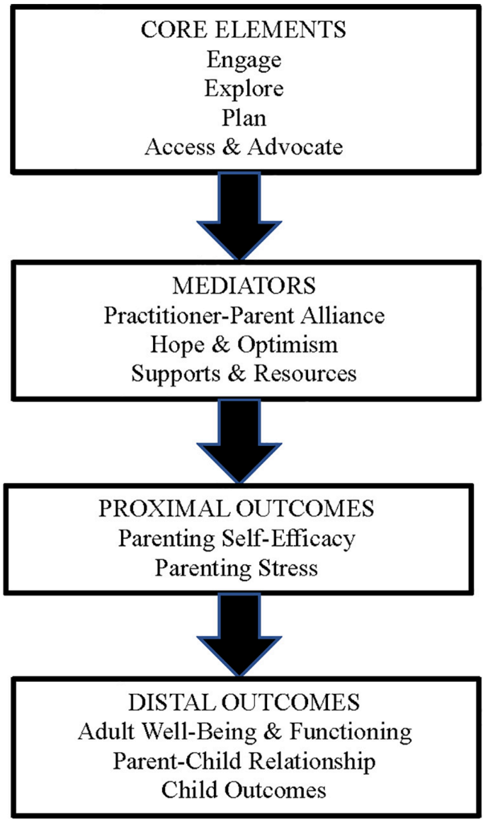


FIGURE 1 | The ParentingWell program theory is based on Self-Determination Theory (46) with core elements of Engage and Explore (autonomy: identifying personal circumstances and motivation, to feel and do better. Plan (competence: setting goals, assessing progress, and building self-efficacy), and Access and Advocate (relatedness: linking to natural and professional supports and resources).

work and promoting the recovery of the people with whom they worked, and could be of value in working with the parent of children at any age, at any point in the therapeutic relationship. In addition, work with parents and the spread of ParentingWell practice was perceived to benefit from training, coaching and ongoing support, professionally and organizationally.

Specific points of feedback included the following: (1) practitioners should routinely ask about parenting and family life; (2) parenting and family life must be integrated into program staff and agencies' routine practice, and not isolated as a specialty service; (3) working together with parents is complex and practitioners would benefit from targeted, ongoing support; (4) shifting to a focus on parenting and family life with adult providers requires organizational champions and support; and (5) organizations must be prepared to adapt policies, practices, and the agency context to be parent- and family-friendly. The first and second points both broadly relate to consistent integration of parenting considerations, as opposed to the implementation of an isolated intervention. In illustration of this need for consistent integration of parenting conversations into routine practice, one practitioner (coaching session, July 2017) noted the need to "link the parenting goals to other goals including symptom management, community education, community inclusion." Another practitioner said (coaching session, November 2017), "Making things general, like symptom management, or adult daily living skills or housing or self-care—all these things can work back around to parenting (e.g., you need to do self-care so you can be able to care for your son)," clearly conveying synergies between parenting and other topics. A coaching participant expressed that she was hesitant to tack on an "additional component" to her routine, and thus supported the idea of more holistic integration into practice (coaching session, June 2017).

The third, fourth, and fifth points of feedback each depict the need to address organizational considerations to adequately support parents. These organizational considerations include support for practitioners who do this work, having organizational infrastructure and champions to ensure the consistency of the work, and having family-friendly policies within the agency. For example, one stakeholder mentioned "coaching and supervision" as key considerations for peer specialists (Adaptation Team session, December 2016). Another practitioner commented that "In our [comprehensive assessment] we have a risk assessment and different addendums—this could be an addendum—other ones are substance abuse history, medications, legal issues. We use these to develop treatment plans (key informant session, August 2016)." This practitioner thus conveyed the need to infuse parenting questions into other agency routines—a task related to organizational infrastructure. Suggested family-friendly policies included allowing toys in the waiting room and permitting parents to bring their children in vehicles when transportation was provided by the agency.

Our notion of success thus loosened to focus less on the specific details of ParentingWell as a prescribed intervention, and more on translating the underlying goals and principles into concrete practice recommendations and skills, and building the capacity of practitioner/adopters to adapt the ParentingWell practice approach in their own settings (50). As such, the adapted practice approach ultimately focuses on the following three questions, which should be woven into routine interactions between behavioral health providers and their clients: (1) What are your parenting and family circumstances? (2) How are things going? and (3) How would you like them to be?

Pre-testing the ParentingWell Practice Approach

Eight themes (each described below) emerged from conversations with stakeholders and practitioners. These themes were ultimately translated into the core elements and underlying principles of the ParentingWell model, as described in a subsequent section.

Stakeholders consistently noted the need for a **family-focused** approach to behavioral health across the lifespan. As an LTC Purveyor explained, "Being able to respond to the family and child feeds into the sense of agency, which is a key ingredient in resilience" (LTC-Worldwide session, September 2015). Another stakeholder alluded to the role of the family in recovery, and the implications for behavioral health practice, as she said, "A lot of the clients that are parents do not have custody of their children and do not have visitation. We want these clients involved, because we feel issues of family life and children are incredibly important to recovery" (key informant session, October 2016). This stakeholder thus conveyed that a family-focused approach is relevant for the unique experiences of each client, including issues of custody loss and/or visitation, where relevant, and for those with children of any age.

Stakeholders acknowledged the fact that culture is largely influential in parenting and mental health (i.e., there is "diversity and cultural competency and different attitudes about parenting"; key informant session, June 2016), and thus recognized that the approach should be **culturally sensitive.** In discussing how to adapt existing models, one stakeholder stated that it will be important to "develop, when necessary, informed cultural adaptations in the Let's Talk model without sacrificing its principles" (LTC-Worldwide session, September 2015). These adaptations may occur in the context of individual practitioner-client relationships, so that practitioners can explore the implications of clients' cultures for their parenting experiences, family life and recovery.

Conversations with stakeholders frequently reflected their inclination to focus on the strengths of parents served, to inspire hope and to capitalize strengths to facilitate goal achievement. One practitioner reflected, "She (the mother) finds the conversations helpful in realizing the abilities she has within herself in helping her child and improving her parenting" (coaching session, September 2017). Thus, the **strengths-based** approach of these conversations was integral for this client in enabling her to focus on and grow positive aspects of her parenting experience. Another practitioner offered the following question that would be helpful to use within the ParentingWell approach: "Would you be interested in talking about your strengths and goals around parenting?" (key informant session, September 2016).

Stakeholders were also aware of the fact that many of their clients had experienced trauma, in some cases related to parenting, and thus the approach should be **trauma-informed**. A trauma informed approach would recognize the reality that "sometimes people are concerned that these types of questions [about parenting] will upset or retraumatize their clients" (key informant session, August 2016), especially for clients who do not have contact with their children. An Adaptation Team member (practitioner) noted that the approach should be trauma-informed, meaning that it needed "dependable, reliable, follow-through, non-judgmental" (Adaptation Team session, April 2018). The non-judgmental element is especially relevant given the wide array of parenting experiences, including the experience of separation from children.

Many stakeholders raised considerations regarding ways to **engage** with parents about family life respectfully and non-judgmentally and doing so in a way that aligns with the parents' needs and preferences. Stakeholders raised concerns associated with engagement, as reflected in the following quote: "Sometimes people are concerned that these types of questions will upset or retraumatize their clients—that is an issue" (key informant session, August 2016). Despite these hesitations, stakeholders acknowledged the importance of engaging their clients in these potentially difficult conversations. As one stakeholder said, "Part of the wellness role is to validate experiences, including parenting ones" (key informant session, June 2017) Given the recognized need to include conversations about parenting in their interactions with clients, stakeholders shared strategies for doing so, including meeting the parents where they are at, and bringing up parenting when the parent seems ready and willing to do so, pacing the conversations. Stakeholders emphasized that listening to the parent is key: "Listen. Listen to what's going on. Lots of people think they know what's going on, but you really need to listen. Don't be directive. Be collaborative in the way you work with someone. When people are directive, it pushes people away and people can get angry" (coaching session, August 2017).

Stakeholders also reflected on how to **explore** the wide range of their clients' parenting experiences, some of which may be emotional experiences that are charged with shame and self-blame. One practitioner explained, "Some clients are afraid to even talk with their children about their diagnoses—a lot of the time children don't even know what their parents are dealing with" (key informant session, August 2016). Stakeholders conveyed the importance of discussing family-related transitions and associated stressors: "Relating to how scary the leap of faith is as a parent when having their child moving in with them" (coaching session, September 2017) Another stakeholder conveyed the wide range of parenting experiences, and their associated implications for conversations about parenting, as she said, "We need to also realize that there are some parents with adult children. . . some parents want to make a connection with their older child" (key informant session, February 2016). Another stakeholder simply noted, "There are many ways to be a parent" (training session, May 2017).

Additionally, stakeholders explained that behavioral health practitioners should help parents in making plans to improve their experiences related to parenting. Thus, helping parents **plan** should be a key element of an approach to parenting-focused behavioral health approach. Parents will ultimately be encouraged to weave parenting goals into their overall wellness and recovery plan. As one participant said, "Feels like you could have a ParentingWell conversation about what was positive re: parenting and use that to start thinking about a plan. Focusing on how things are going, and how you'd like things to be" (coaching session, August 2017).

Stakeholders clearly conveyed that part of their role was to help their clients **advocate and access** peer supports, opportunities for self-care, supports related to basic living needs, and culturally relevant resources. Demonstrative quotes include: "Peer specialists are the ones who relate to the family, who the client will listen to. From the perspective of the peer specialist—'I understand your situation. What would work for you? Who will be in your life? Who will be there to support you?" (key informant session, March 2016); and regarding what training or preparation workers would need: ". . .to help the client find home or shelter, things the baby would need, parenting classes, the social welfare benefits process, information about what is changing in the system, employment and benefits applications, Mass Health (health care payer) applications" (key informant session, March 2016).

Compiling the ParentingWell Practice Profile

In light of the feedback we received, we shifted our focus to compiling the agency- and practitioner-agnostic ParentingWell Practice Profile (PWPP), relevant to parents across the lifespan (51). A practice profile describes the program or practice approach, including essential functions, operational definitions, and practical performance strategies (i.e., the theory of action). The PWPP provides concrete discussion points and topics (core activities) that practitioners can use to address the four core elements and four underlying principles. The core activities also embody action mechanisms (i.e., information sharing, reflecting and reframing, goal-setting, and skills-building; examples provided in the next paragraph). Thus, designed to reflect the core elements and underlying principles, and inclusive of concrete action mechanisms, the PWPP is the culmination of the adaptation process. The PWPP is also the operationalization of the core elements and practice principles into a specific theory of action (52) (see **Table 1**).

For example, a core activity suggests that during the first meeting, the practitioner welcomes the parents and asks initial questions about parenting and family status (core element **engage**, key principle **family focused**, action mechanism **information sharing**). A second core activity suggests that the practitioner support the parent in identifying strengths and resources, particularly as they relate to parenting/relationships with children and family life, social support, and self-care (**explore**, **strengths-based**, **reflecting and reframing**). A core activity pertaining to goal setting is to help the parents identify what they want to change and picture the outcomes; an activity pertaining to skills-building is to assist with a problem-solving approach if parents cannot "put the pieces in place" to take steps forward. Thus, the core activities provide concrete action steps that put into motion the core elements and underlying principles.

Core activities are not necessarily meant to occur in a particular order, activities from different core elements may occur

TABLE 1 | The ParentingWell Practice Profile action mechanisms.

Information sharing	Respectful, non-judgmental curiosity
	Positive messaging (e.g., encouragement, empathy)
Reflecting and reframing	Exploring experiences, thoughts and feelings
	Understanding relationship between attitudes, thoughts and behavior Unraveling and challenging faulty thinking
	Recognizing patterns
	Taking the other's point of view
	Shifting perspective to see a situation differently
Goal-setting	Forming intentions
	Identifying necessary resources (e.g., motivation, time and energy, natural, and professional supports)
	Pinpointing barriers and strategies for overcoming
	Setting SMART goals (Specific, Measurable, Achievable, Relevant, Time- Bound)
	Celebrating successes
Skills-building	Observing and recording (e.g., journaling) Instructing
	Modeling or demonstrating the behavior Rehearsing and experimenting
	Providing relevant, appropriate feedback

The ParentingWell theory of action, related activities and practitioner skills are informed by Social Cognitive and Cognitive Behavioral Theory and the Information-Motivation Behavioral Model.

simultaneously, and practitioners and clients may work back and forth among activities over time. For each client, a more complete picture of the person as a parent and their priorities for family life will emerge. Practitioners will be able to work with parents to help them weave their goals for parenting and family life into their vision for change and plans for the future.

In addition to compiling the ParentingWell Practice Profile as a guide for practitioners, the ParentingWell Workbook of activities for practitioners and parents is available, along with the ParentingWell Self-Assessment and Supervisory Tools for use by practitioners and their supervisors. These resources are available in **Supplementary Material** linked to this article.

DISCUSSION

This study describes the process and outcome of adapting an intervention for parents with mental illness, for implementation and sustainment in Massachusetts adult mental health service agencies. Specific objectives were as follows: (1) identify the core components and principles of the original LTC intervention; (2) consider service delivery contextual factors, which would be sustained during the adaptation process; (3) pre-test the adapted materials, resulting in the specification of new core components and principles; and (4) compile the practice profile, translating core components and principles into a theory of action and core activities.

Regarding the first study objective, conversations with stakeholders yielded the program theory or logic model for Let's Talk About Children. The elements of the logic model comprise the core components and principles of the original intervention that were retained during the compilation of the adapted ParentingWell model. Adaptations that fail to retain the key elements of an intervention may reduce the effectiveness of that intervention (53). While the goal of adaptation is to improve the efficacy of an intervention for a new specified context, the assumption is that the original intervention remains intact enough for evidence of its effectiveness to remain relevant even in its adapted form (53). To ensure that the adaptation is fidelity consistent (and thus that evidence for its effectiveness "translates"), the identification and maintenance of key components of the original intervention are critical to the adaptation process (53). In examining the extent and impact of adaptation, other considerations relate to both process and outcome, such as whether modifications were planned/proactive or unplanned/reactive; who made the decisions; what is modified (e.g., components, delivery method, etc.); and factors that influenced decisions (e.g., improve fit, align with cultural values, norms or priorities) (53). The adaptation process described in this paper included a consistent focus on the key elements of Let's Talk About Children, contributing to a strong likelihood that evidence for the effectiveness of Let's Talk About Children will also apply to the ParentingWell Practice Approach. However, as future research explores implementation of the ParentingWell Practice Approach, it will be important to investigate how the original intervention and its adapted elements each contribute to its impact.

Conversations with stakeholders, including intervention purveyors, ParentingWell Training and Coaching Participants, reflected themes that addressed the second and third study objectives. Regarding contextual considerations (the second objective), the first theme that emerged from our data relates to workflow; specifically, stakeholders emphasized that the adaptation should ultimately result in a framework that can be consistently integrated into practice, rather than a stand-alone intervention. As such, the resultant ParentingWell is an approach to routine practice that makes talking about parenting, children, and family experiences a natural part of the conversation and of an adult's recovery process. The ParentingWell approach thus addresses contextual considerations, namely by avoiding challenges that would accompany "tacking on" an additional intervention, which may require extensive time and training (3). Stakeholders who are familiar with adult mental health service agencies in Massachusetts emphasized the benefits of this routine integration. Future research should also investigate the extent to which the approach is relevant for agencies in other states and perhaps countries.

Also related to future implementation beyond Massachusetts, the use of the ParentingWell approach does not require extensive clinical, counseling or practice skill specific to addressing parenting. This may facilitate implementation in a wide variety of settings. Research in several contexts has established that practitioners often lack knowledge and skills related to addressing their clients' parenting roles (4, 5, 7–12). The ParentingWell approach enables and encourages practitioners to draw from the skills they already possess, while keeping parenting in mind. As such, it does not require a vast set of skills that are specific to addressing parenting. Future research will need to explicitly address this characteristic as it relates to scale-up.

Regarding the third objective, stakeholders specified the following themes, which comprise the underlying principles and the core elements of the ParentingWell Practice Profile:

the need for an approach that is family-focused, trauma-informed, culturally sensitive, and strengths-based; and for conversations in which the provider and the client engage, explore, plan, and access and advocate around issues related to parenting and family life. The identification of themes fulfills the third objective, which is the specification and the compilation of the ParentingWell Practice Profile. The Profile includes operationalized core activities for each element (i.e., for *engage*, the practitioner might ascertain where children are living and who is caring for them; for *explore*, the practitioner might discus daily routines, household chores, and taking care of the children). It also identifies how the underlying principles map onto each core element. For instance, *explore* is strengths-based as the activities reflect the understanding that parents, especially those who are quite depressed or see themselves as "failures," may require assistance in identifying strengths and resources in themselves and in their children. Future research should assess the feasibility of implementing the approach, the impact on practitioner behavior, and ultimately, the impact on parents. Also, because the adaptation involved modifying a stand-alone intervention into a continuous and holistic approach, future research that assesses impact should seek to understand how this change impacts parents. Considerations might include whether the timing and/or duration of impact differs as a result of the transition from a stand-alone intervention to an ongoing approach.

The adapted model (the fourth objective) fills a critical gap as it addresses the lack of evidence-based interventions for parents with serious mental illness and it reflects the need for a flexible practice approach. The ParentingWell Practice Approach includes well-specified principles and core elements, aligned with core activities that constitute a theory of action. It fits into routine workflow at any point in the therapeutic relationship, and draws from practitioners' existing skills and competencies, ultimately with the potential to enhance clients' recovery.

Limitations

Despite this promise, this study has its limitations. The stakeholders are reflective of the Massachusetts mental health workforce and, consequently, are mostly White. Meanwhile, both parenting and mental health are culturally bound. ParentingWell addresses this consideration, as a key principle is to be culturally sensitive, but it is still critical to engage more diverse stakeholders. This should be the focus of future testing and refinement of the ParentingWell Practice Profile. Additionally, the context of mental health service provision has changed with COVID-19, as has the context of parenting. The approach is designed to be flexible, delivered, however, and wherever mental health services are delivered. However, as future research explores the feasibility and impact of the approach, changing contextual factors should be kept in mind.

CONCLUSION

Ultimately, this study and the adapted ParentingWell resources address the critical lack of evidence-based interventions for parents with serious mental illness. Future research will provide needed insight pertaining to its implementation and impact.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because given the qualitative nature of the data and the concern for the privacy of participants, many of whom are agency leaders, data are held by the authors. Requests to access the datasets should be directed to JN, jnicholson@brandeis.edu.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Brandeis University Institutional Review Board, Dartmouth College Institutional Review Board, and Massachusetts Department of Mental Health Central Office Research Review Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JN was responsible for the overall design and implementation of study activities and the preparation of the manuscript. KE and KB contributed to study design and implementation. JN and KE were responsible for drafting the ParentingWell Practice Profile. MH contributed to data analysis and writing and editing of the manuscript. All authors contributed to the manuscript, read and approved the submitted version.

FUNDING

This study was funded by the National Institute on Disability, Independent Living, and Rehabilitation Research (grant #90DPGE0001-01-01) and the Massachusetts Department of Mental Health.

ACKNOWLEDGMENTS

We would like to acknowledge the contribution of the many agency stakeholders and parents who participated in all stages of this study. Joan Mikula, Commissioner, Massachusetts Department of Mental Health (ret.), and Toni Wolf, Commissioner, Massachusetts Rehabilitation Commission, have long been champions of this work.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.801065/full#supplementary-material

REFERENCES

1. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child Youth Serv.* (2016) 37:129–55. doi: 10.1080/0145935x.2016.1104048
2. Goodyear M, Maybery D, Reupert A, Allchin R, Fraser C, Fernbacher S, et al. Thinking families: a study of the characteristics of the workforce that delivers family-focussed practice. *Int J Ment Health Nurs.* (2017) 26:238–48. doi: 10.1111/inm.12293
3. Lauritzen C, Reedtz C, Doesum K, Martinussen M. Factors that may facilitate or hinder a family-focus in the treatment of parents with a mental illness. *J Child Fam Stud.* (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
4. Maybery D, Goodyear M, O'Hanlon CR, Reupert A. Profession differences in family focused practice in the adult mental health system. *Fam Process.* (2014) 53:608–17. doi: 10.1111/famp.12082
5. Maybery D, Goodyear M, Reupert AE, Grant A. Worker, workplace or families: What influences family focused practices in adult mental health? *J Psychiatr Ment Health Nurs.* (2016) 23:163–71. doi: 10.1111/jpm.12294
6. Mulligan C, Furlong M, McGilloway S. Promoting and implementing family-focused interventions for families with parental mental illness: scoping and installation. *Adv Ment Health.* (2020) 18:202–16. doi: 10.1080/18387357.2019.1614466
7. Skogoy BE, Ogden T, Weimand BM, Ruud T, Sorgaard K, Maybery D. Predictors of family focused practice; organisation, profession, or the role as child responsible personnel? *BMC Health Serv Res.* (2019) 19:793. doi: 10.1186/s12913-019-4553-8
8. Tchernegovski P, Reupert AE, Maybery D. How do Australian adult mental health clinicians manage the challenges of working with parental mental illness? A phenomenological study. *Child Fam Soc Work.* (2018) 23:381–9.
9. Adderley H, Wittkowski A, Calam R, Gregg L. Adult mental health practitioner beliefs about psychosis, parenting, and the role of the practitioner: a Q methodological investigation. *Psychol Psychother Theory Res Pract.* (2020) 93:657–73. doi: 10.1111/papt.12249
10. Biebel K, Nicholson J, Woolsey K. Implementing an intervention for parents with mental Illness: Building workforce capacity. *Psychiatr Rehabil J.* (2014) 37:209–15. doi: 10.1037/prj0000046
11. Maybery D, Reupert A. Workforce capacity to respond to children whose parents have a mental illness. *Aust N Z J Psychiatry.* (2006) 40:657–64. doi: 10.1080/j.1440-1614.2006.01865.x
12. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
13. Allchin B, O'Hanlon B, Weimand BM, Goodyear M. Practitioners' application of Let's Talk about Children intervention in adult mental health services. *Int J Ment Health Nurs.* (2020) 29:899–907. doi: 10.1111/inm.12724
14. Lauritzen C, Reedtz C, van Doesum KTM, Martinussen M. Factors that may facilitate or hinder a family-focus in the treatment of parents with a mental illness. *J Child Fam Stud.* (2015) 24:864–71. doi: 10.1007/s10826-013-9895-y
15. Nicholson J, de Girolamo G, Schrank B (editors). *Parents with mental and/or substance use disorders and their children [Internet]. Frontiers Media SA*. (2020). Available online at: https://www.frontiersin.org/research-topics/7178/parents-with-mental-andor-substance-use-disorders-and-their-children (accessed March 18, 2020).
16. Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman MV. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. 3rd ed. Cambridge: Cambridge University Press (2015).
17. Biebel K, Nicholson J, Wolf T. Shifting the intervention paradigm from individuals to families living with parental mental illness. 3rd ed. In: Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman MV editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. Cambridge: Cambridge University Press (2015). p. 343–53. doi: 10.1017/cbo9781107707559.033
18. Hinden BR, Biebel K, Nicholson J, Henry AD. A survey of programs for parents with mental illness and their families: Identifying common elements to build an evidence base. *J Behav Health Serv Res.* (2006) 33:21–38. doi: 10.1007/s11414-005-9007-x
19. Hinden BR, Wolf T, Biebel K, Nicholson J. Supporting clubhouse members in their role as parents: Necessary conditions for policy and practice initiatives. *Psychiatr Rehabil J.* (2009) 33:98–105. doi: 10.2975/33.2.2009.98.105
20. Nicholson J, Wolf T, Biebel K. Creating positive parenting experiences: family options. 3rd ed. In: Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman MV editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. Cambridge: Cambridge Unviersity Press (2015). p. 266–76. doi: 10.1017/cbo9781107707559.026
21. Nicholson J, Deveney W. Why not supported parenting? *Psychiatr Rehabil J.* (2009) 33:79–82.
22. Nicholson J, Friesen BJ. Developing the evidence base for families living with parental psychiatric disabilities: crossing the bridge while we're building It. *Psychiatr Rehabil J.* (2014) 37:157–61. doi: 10.1037/prj0000092
23. Niemala M, Kallunki H, Jokiinen J, Rasanen S, Ala-Aho B, Hakko H, et al. Collective impact on prevention: let's talk about children service model and decrease in referrals to child protection services. *Front Psychiatry.* (2019) 10:1–5. doi: 10.3389/fpsyt.2019.00064
24. Solantaus T, Toikka S, Alasuutari M, Beardslee WR, Paavonen EJ. Safety, feasibility and family experiences of preventive interventions for children and families with parental depression. *Int J Ment Health Promot.* (2009) 11:15–24. doi: 10.1111/j.1545-5300.2009.01281.x
25. Solantaus T, Paavonen EJ, Toikka S, Punamäki R-L. Preventive interventions in families with parental depression: children's psychosocial symptoms and prosocial behaviour. *Eur Child Adolesc Psychiatry.* (2010) 19:883–92. doi: 10.1007/s00787-010-0135-3
26. Solantaus T, Reupert A, Maybery D. Working with parents who have a psychiatric disorder. 3rd ed. In: Reupert A, Maybery D, Nicholson J, Gopfert M, Seeman MV editors. *Parental Psychiatric Disorder: Distressed Parents and their Families*. Cambridge: Cambridge University Press (2015). p. 238–47. doi: 10.1017/cbo9781107707559.023
27. Maybery D, Goodyear M, Reupert A, Sheen J, Cann W, Dalziel K, et al. Developing an Australian-first recovery model for parents in Victorian mental health and family services: a study protocol for a randomised controlled trial. *BMC Psychiatry [Internet].* (2017) 17:198. doi: 10.1186/s12888-017-1357-4
28. Maybery D, Goodyear M, Reupert A, Sheen J, Cann W, O'Hanlon B, et al. A mixed method evaluation of an intervention for parents with mental illness. *Clin Child Psychol Psychiatry.* (2019) 30:11. doi: 10.1177/1359104518822676
29. Tchernegovski P, Reupert A, Maybery D. "Let's Talk about Children": a pilot evaluation of an e-learning resource for mental health clinicians. *Clin Psychol.* (2015) 19:49–58. doi: 10.1111/cp.12050
30. Giannakopoulos G, Solantaus T, Tzavara C, Kolaitis G. Mental health promotion and prevention interventions in families with parental depression: a randomized controlled trial. *J Affect Disord.* (2021) 278:114–21. doi: 10.1016/j.jad.2020.09.070
31. Ueno R, Osada H, Solantaus T, Murakoshi A, Inoue T. Safety, feasibility, fidelity, and perceived benefits of an intervention for parents with mood disorders and their children - "Let's Talk About Children" in Japan. *J Fam Psychother.* (2019) 30:272–91. doi: 10.1080/08975353.2019.1678092
32. Allchin B, Solantaus T. An evidence-based practice developed in-situ: let's Talk About Children and a consolidation of its evidence base. *Front Psychiatry.* (2022) 13:1–9. doi: 10.3389/fpsyt.2022.824241
33. Goodyear M, Hill T-L, Allchin B, McCormick F, Hine R, Cuff R, et al. Standards of practice for the adult mental health workforce: Meeting the needs of families where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120
34. Bauer A, Best S, Malley J, Christiansen H, Goodyear M, Zechmeister-Koss I, et al. Towards a program theory for family-focused practice in adult mental health care settings: an international interview study with program leaders. *Front Psychiatry.* (2021) 12:1–12. doi: 10.3389/fpsyt.2021.741225
35. Chambers DA, Norton WE. The adaptome: advancing the science of intervention adaptation. *Am J Prev Med.* (2016) 51(4 Suppl. 2):S124–31. doi: 10.1016/j.amepre.2016.05.011
36. McKleroy VS, Galbraith JS, Cummings B, Jones P, Harshbarger C, Collins C, et al. Adapting evidence–based behavioral interventions for new settings and target populations. *AIDS Educ Prev.* (2006) 18(Suppl.):59–73. doi: 10.1521/aeap.2006.18.supp.59
37. Aarons GA, Green AE, Palinkas LA, Self-Brown S, Whitaker DJ, Lutzker JR, et al. Dynamic adaptation process to implement an evidence-based child

maltreatment intervention. *Implement Sci.* (2012) 7:32. doi: 10.1186/1748-5908-7-32

38. Ghate D. From programs to systems: deploying implementation science and practice for sustained real world effectiveness in services for children and families. *J Clin Child Adolesc Psychol.* (2016) 45:812–26. doi: 10.1080/15374416.2015.1077449
39. Aarons GA, Sklar M, Mustanski B, Benbow N, Brown CH. "Scaling-out" evidence-based interventions to new populations or new health care delivery systems. *Implement Sci.* (2017) 12:111. doi: 10.1186/s13012-017-0640-6
40. Kirk MA, Moore JE, Wiltsey Stirman S, Birken SA. Towards a comprehensive model for understanding adaptations' impact: the model for adaptation design and impact (MADI). *Implement Sci.* (2020) 15:56. doi: 10.1186/s13012-020-01021-y
41. Card JJ, Solomon J, Cunningham SD. How to adapt effective programs for use in new contexts. *Health Promot Pract.* (2011) 12:25–35. doi: 10.1177/1524839909348592
42. Escoffery C, Lebow-Skelley E, Udelson H, Böing EA, Wood R, Fernandez ME, et al. A scoping study of frameworks for adapting public health evidence-based interventions. *Transl Behav Med.* (2019) 9:1–10. doi: 10.1093/tbm/ibx067
43. Fernandez ME, Ruiter RAC, Markham CM, Kok G. Intervention mapping: theory- and evidence-based health promotion program planning: perspective and examples. *Front Public Health.* (2019) 7:209. doi: 10.3389/fpubh.2019.00209
44. Movsisyan A, Arnold L, Evans R, Hallingberg B, Moore G, O'Cathain A, et al. Adapting evidence-informed complex population health interventions for new contexts: a systematic review of guidance. *Implement Sci.* (2019) 14:105. doi: 10.1186/s13012-019-0956-5
45. Patton MQ. *Developmental Evaluation: Applying Complexity Concepts to Enhance Innovation and Use*. New York, NY: The Guilford Press (2011).
46. Dedoose. *Dedoose Version 8.0.35, Web Application for Managing, Analyzing, and Presenting Qualitative and Mixed Method Research Data [Internet]*. Los Angeles, CA: SocioCultural Research Consultants, LLC (2018).
47. Levitt HM, Bamberg Mi, Creswell JW, Frost DM, Josselson R, Suarez-Orozco C. Journal article standards for qualitative primary, qualitative meta-analytic, and mixed methods research in psychology. *Am Psychol.* (2018) 73:26–46. doi: 10.1037/amp0000151
48. Deci EL, Ryan RM. Self-determination theory in health care and its relations to motivational interviewing: a few comments. *Int J Behav Nutr Phys Act.* (2012) 9:24. doi: 10.1186/1479-5868-9-24
49. Goodyear M, Allchin B, Burn M, von Doussa H, Reupert A, Tchernagovski P, et al. Promoting self-determination in parents with mental illness in adult mental health settings. *J Fam Nurs.* (2022) 2:1–13. doi: 10.1177/10748407211067308
50. Horton T, Illingworth J, Warburton W. *The Spread Challenge: How to Support the Successful Uptake of Innovations and Improvements in Health Care*. London: The Health Foundation (2018).
51. Nicholson J, English K. *ParentingWell Practice Profile [Internet]*. Waltham, MA: National Research Center for Parents with Disabilities (2019).
52. Abraham C, Michie S. A taxonomy of behavior change techniques used in interventions. *Health Psychol.* (2008) 27:379–87. doi: 10.1037/0278-6133.27.3.379
53. Wiltsey Stirman S, Baumann AA, Miller CJ. The FRAME: an expanded framework for reporting adaptations and modifications to evidence-based interventions. *Implement Sci.* (2019) 14:58. doi: 10.1186/s13012-019-0898-y

Author Disclaimer: The statements presented in this publication are solely the responsibility of the authors and do not necessarily represent the views of the funders.

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 08 April 2022
doi: 10.3389/fpsyt.2022.800037

Children of Parents With a Mental Illness—Stigma Questionnaire: Development and Piloting

Lisa-Marie Dobener*, Markus Stracke, Kathrin Viehl and Hanna Christiansen

Department of Clinical Child and Adolescent Psychology, Philipps University Marburg, Marburg, Germany

Children of parents with a mental illness are a particularly vulnerable group as they have a high risk to develop a mental disorder themselves and those are associated with high stigma. Moreover, just like primary recipients of stigma, they are affected by the social taboo surrounding mental illness: they do not receive enough information, are often left alone with their problems, and are thus considered "invisible children". In previous research, family stigma has only been assessed through general questionnaires for all family members. What has not yet been adequately investigated is how stigma difficulties affect the children of parents with mental illness in particular. To address these limitations, we developed the Children of Parents with Mental Illness-Stigma-Questionnaire (COPMI-SQ), a self-report instrument for young people aged 12–19 years, designed to assess young people's stigma experiences in daily life. Based on a systematic review preceding the questionnaire, we identified relevant stigma dimensions for children of parents with a mental illness that resulted in 93 items that according to theory were assumed to load on four different scales: experienced stigma, anticipated stigma, self-stigma, and structural discrimination. An expert discussion, and a comprehensibility analysis with the target group followed. In this paper, we report on the development process and initial pilot data ($N = 32$) on the psychometric properties of the COPMI-SQ. Item analyses via an item difficulty index, discriminatory power, as well as internal consistency analysis resulted in a revised instrument reduced to 67 items. We observed very high internal consistencies (between $\alpha = 0.868$ and $\alpha = 0.975$) for the subscales. The approach taken to develop the COPMI-SQ followed scientifically accepted principles by ensuring different construction phases and is considered a solid basis for further reliability and validity studies. The study is ongoing and undergoing a further validation investigation; dimensionality and factor structure will also be examined.

Keywords: children of parents with a mental illness, stigma by association, family stigma, COPMI-SQ, questionnaire, instrument development, piloting

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Emma Maynard,
University of Portsmouth,
United Kingdom
Kate Blake-Holmes,
University of East Anglia,
United Kingdom
Andrea Reupert,
Monash University, Australia

***Correspondence:**
Lisa-Marie Dobener
lisa-marie.dobener@uni-marburg.de

Specialty section:
This article was submitted to
Child and Adolescent Psychiatry,
a section of the journal
Frontiers in Psychiatry

Received: *22 October 2021*
Accepted: *04 February 2022*
Published: *08 April 2022*

Citation:
Dobener L-M, Stracke M, Viehl K and Christiansen H (2022) Children of Parents With a Mental Illness—Stigma Questionnaire: Development and Piloting. Front. Psychiatry 13:800037. doi: 10.3389/fpsyt.2022.800037

INTRODUCTION

Approximately 17–25% of all children worldwide live with at least one parent who has a mental illness (1–4). Evidence shows that children of parents with a mental illness (COPMI) are exposed to serious and diverse risks compared to their peers of similar age (3). The high prevalence and known risks contrast with the very low visibility of these children (5). Mental illnesses of parents can influence the living environment of children and adolescents in many ways, and they carry various risks associated with reduced mental health, poorer academic achievement, and impaired social well-being (3, 6). Moreover, COPMI carry a significantly increased risk of developing mental health problems themselves (1). In addition to genetic factors and familial influences resulting from or accompanying the parental illness, there are environmental factors that contribute to whether children develop a mental illness themselves during their lives (1, 7). One social environment mechanism that influences the entire family system and is known to be relevant in terms of children's personal development, wellbeing, and help-seeking behavior is the stigma associated with a parent's mental illness (8, 9). Despite high prevalence rates of children of parents with a mental illness, they are remaining "invisible" due to the lack of recognition and formal identification within (adult) health systems (10, 11). Through the fear of stigma and negative repercussions of those children and their families, many of these children remain hidden (8, 9).

Although the appearance of stigma is liable to historical, cultural, and temporal changes, there is hardly a country, society, or culture in the world where mental illness is not stigmatized (12, 13). Stigma is an attribute leading to widespread social disapproval, and encompasses the negative effects of a label placed on any group (14, 15). It occurs in social situations, meaning that stigma does not reside in the person itself, but is the result of the social context and the perception of the public (16). People with a mental illness are frequently viewed as dangerous, unpredictable, incompetent, abnormal, and of weak character (17). While this is widely true for all types of mental disorders, the perception of different disorders varies and therefore the stigma attached to it: for instance, schizophrenia is associated with much more "dangerousness" than depression, resulting in greater social distancing from the public to the people it is associated with (18).

With regard to the stigma of mental illness, as with other stigmatized conditions, there is evidence that those affected themselves by mental illness are not the only individuals who suffer social stigma (19). Goffman, who raised the concept of stigma, has already outlined the phenomenon of the so-called *courtesy stigma* in 1963 (15). Nowadays, it is widely known as *stigma by association (SBA)* or *family stigma*, describing family members as also being exposed to stigma resulting from a family member's mental illness (20). An abnormality attributed to individual family members or the family as a whole is considered key to the development of family stigma (21). Another crucial factor in determining whether a SBA occurs appears to be the entitavity, i.e. the degree to which two or more people form a significant social unit (22, 23). The higher the entitavity, the greater the likelihood of being stigmatized on the basis of association (23). Intimate groups such as families are attributed the highest degree of entitavity (23)—the association with a stigmatized family member and one's own experience of stigmatization is therefore very likely.

Children and adolescents who grow up with parents with a mental illness are thus—due to their dependent and close relationship to the affected person—a particularly vulnerable group for stigma. They may face the stigma due to the parental mental illness itself, and due to associated peculiarities and "otherness" within their families as described above (21, 24, 25). Negative effects in the affected children and young people arise both from the actual stigma experienced as well as the fear of being stigmatized and the internalization of stigmatizing attitudes toward them and their families (24, 26, 27).

There are many theories clarifying the various facets of stigma for people with a mental illness. Yet there is no comprehensive theoretical model for SBA. Research suggests that family stigma is no monolithic phenomenon, because it varies depending on the relationship the family member has with the person with the mental illness: studies have shown that parents of children with a mental illness are more likely to experience the stigma of neglect and blame for their children's disease onset (12, 28), and the siblings of children with a mental illness and spouses of someone with a mental illness are more likely to experience the stigma of blame for the persistence of their relative's disease (12); COPMI are more likely to face the stigma of "contamination"—meaning that the general population believes that parental illness, and especially regarding COPMI, is passed on to children (29). However, studies in this regard are very sparse. By describing a "contamination" stigma, authors are often referring to the public perception of stigma toward COPMI [e.g., (30, 31)], and are thereby missing out to understand children's lived experiences. Thus, these studies possess little informative value if we hope to understand how children perceive and experience family stigma. A recent integrative review on the evidence of stigma concepts for children of parents with a mental illness has shed light on those limitations by identifying stigma-related experiences and outcomes as reported by parents and children (29). Nevertheless, this review shows that a concept which includes the various dimensions of stigma experiences of children whose parent has a mental illness is missing. The results of the review show that children report feelings of embarrassment, shame, and the need to hide their parental mental illness, but the authors do not integrate those findings into an overall framework of different stigma dimensions.

To fill this gap, we have conducted a systematic review to analyze the COPMI's experiences of stigma and to identify specific stigma dimensions and their characteristics for this specific target group (28). Our review resulted in four stigma dimensions: (1) *Experienced SBA* describes personally experienced prejudice and discrimination (32, 33); (2) *anticipated SBA* incorporates expectations that others will devalue and discriminate against them in the future (32); (3) *affiliate stigma* describes the self-stigma of people associated with a mental illness, i.e. the internalization of public stigma (34); and (4) *structural discrimination*, entailing social institutions and ideological systems that reproduce and maintain the stigmatized status (14).

In a literature search, we identified nine scales measuring mental illness SBA or family stigma (34–42) as well as one family-experiences interview with a stigma subscale (43). Most of the scales only measure the stigma component self-stigma/internalized stigma (34–36, 40). One of the scales was a slightly modified version of a scale constructed for primary stigma recipients, i.e., people with a mental illness (39). Another scale was developed to measure the SBA of relatives of patients with schizophrenia (42); the others were not restricted to a specific relative's mental illness. Six scales as well as the interview schedule stigma scale were designed for all family members, while two of the scales measure the stigma parents of children with a mental illness suffer from (40, 41). None of the scales was constructed or validated for minor children. The items' wording is often awkward [e.g., *"Most people believe their friends would not visit them as often if a member of their family were hospitalized for serious mental illness"* (37)]. In addition, the way the items are phrased often presuppose a great deal of basic knowledge of certain emotional and cognitive processes, such as *"I feel emotionally disturbed because I have a family member with mental illness/intellectual disability"* (34), or do not seem stigma-specific: *"Having a family member with mental illness/intellectual disability imposes a negative impact on me"* (34).

Furthermore, we know that stigma varies depending on the individual's role within the family, and none of the established instruments combines all the stigma dimensions highlighted in the review that are relevant to children with a mentally ill parent. Our systematic review has revealed aspects of stigma that are quite specific to their role as children, for example, being bullied and teased in school, being isolated because of feeling ashamed and therefore not inviting friends home, being afraid of passing on the illness to another generation, being responsible as a child for the parent's wellbeing when psychiatrists turn them away, having nobody to talk to, and not getting enough information about their parent's mental illness.

Overall, for children of parents with a mental illness, no suitable instrument exists yet to measure their stigma experiences, expectations, self-stigma and the stigma's structural dimensions—especially none considering the special role of children. The current study aimed to describe the development and initial pilot data on our newly developed Children of Parents with a Mental Illness—Stigma Questionnaire (COPMI-SQ). This scale can provide important information to better understand the complex phenomenon of stigma by association among young people and shed light on experiences and needs of this group in intervention studies or anti-stigma campaigns.

METHODS

Figure 1 shows the different stages of development for the COPMI-SQ. We have oriented ourselves to two guidelines for the development of new scales (44, 45). We are currently reporting on phase 3 and present our pilot data. Phase 4 is still under way.

Ethics

Research with children and adolescents is always in a field of tension between the respect of the various rights of children and their special need for protection. In recent decades, a paradigm shift has occurred within research, in which children are now rather seen as *"moral agents in their own right"* (46). As a result, more research is now being conducted with children to increase

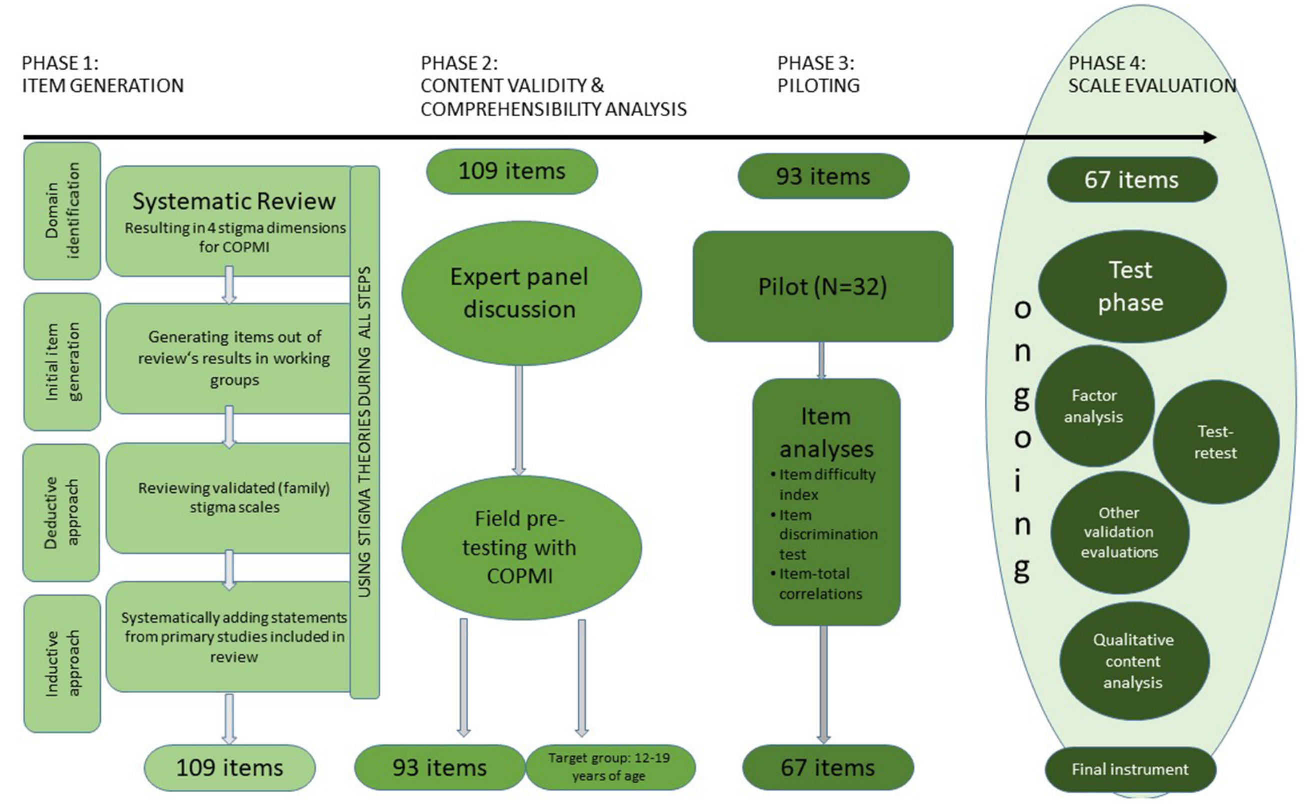


FIGURE 1 | Phases of development of the instrument measuring stigma in COPMI (COPMI-SQ).

their participation. Researchers and adults in general cannot assume that their view of the world is congruent with that of children (46). In order to comply with the UN Convention on the Rights of the Child, we consider it of fundamental importance to interview children and adolescents themselves when it comes to their life experiences and the intention to improve their living conditions. Since it can be assumed that stigma may affect a lot of children of parents with mental illness in particular, it is necessary to obtain their perspectives before interventions are directed past their needs.

In order to ensure the special protection of children under guardianship, we followed the guidelines of the Central Ethics Committee of the German Medical Association (47), which stipulates a need for an informed consent both from their parents with a mental disorder and the children themselves in age-appropriate language prior to study participation. We considered the costs and benefits of a survey, which could be potentially burdensome and concluded that the results of the survey offered great insights into the experiences of this population which could be beneficially used for further interventions for this population. However, since it can be assumed that children of parents with mental illness are particularly exposed to stresses that may also be related to stigmatizing experiences, they were offered to contact the persons responsible for the study, who are child and adolescent psychotherapists, at any time. In addition, they were advised to contact any trusted adult person in case of need. Furthermore, prior to study participation, they were informed that they could withdraw from their participation in the online study at any time and without giving reasons, up to when the data were analyzed.

The study was approved by the Ethics Committee of the Psychology Department of Philipps University of Marburg (study approval number: 2020-20).

Inclusion Criteria

Young people were eligible to participate in the study as long as they were between 12 and 19 years old and one or both parents had a mental illness. Information regarding the illness was obtained through a self-report by the parents. Otherwise, there were no exclusion criteria.

Phase 1: Item Generation

The first step toward designing this new instrument, the systematic review, resulted in four different dimensions of family stigma in COPMI, i.e. experienced SBA, anticipated SBA, affiliate stigma (self-stigma), and structural discrimination. Those findings were presented by a PhD student to 50 scientific collaborators in Clinical Psychology and Clinical Child and Adolescent Psychology working groups of Marburg University, Department of Psychology, including pre-, post-docs, and professors. Post-docs and professors further possessed licenses as psychotherapists and PhD students were psychotherapists in training. Quotes from primary studies based on a systematic review with the instruction to collect ideas for items for a survey were presented to all participants. Initial items that may be used in a survey for this population and for the different dimensions were generated through group discussions.

The inductively generated items were then compared with already validated instruments measuring (family) stigma (see above) to apply formulations already established whenever possible. Using the established stigma scales, the generated items were developed further and supplemented in their formulation.

To ensure that every aspect of stigma was covered, the first pool of items was then supplemented by adding further inductively formed items for every (sub-)sub-category identified in the review [for further information see Dobener et al. (28)]. Statements from the included primary studies were included also to reflect as accurately as possible the comments of the target group members themselves in the items. For each category identified in the review, 5–15 items were formed. A total of 109 items was generated in phase 1.

Phase 2: Content Validity and Comprehensibility Analysis

Expert Discussion

The entire item pool was then reviewed and discussed by a panel of experts from our and other known working groups, with knowledge in developing questionnaires as well as in the field of stigma. The experts were asked to rate all items according to the extent to which they seemed to be an appropriate measure of the intended construct. They were also asked to provide open-ended feedback. Items agreed upon by at least two of the three experts were retained or reworded according to their comments. This ensured that only items that were understandable and meaningful in terms of both content and language were used. Through this process, the item pool was reduced to 96 items since the expert panel revealed problems like overly complex wording, repetition, or other violations of "best-practices" regarding item structure (48).

Comprehensibility Analysis With Children

In a further step, five children and adolescents of different ages whose parents have a mental illness filled in the questionnaire to check comprehensibility. They completed the COPMI-SQ in the presence of the author L-MD, to enable immediate feedback on any difficulties in understanding. In addition to the comprehensibility check, this allowed us to see whether filling in the COPMI-SQ was associated with any mental stress when confronted with the effects of their parents' illness. According to the children's feedback, some items were reworded, and three were removed entirely.

At the end of this second phase, the item pool was reduced to 93 items, of which 91 were closed questions and two were open-ended. Due to the complexity of wording and the general need for any mental illness concept as well as children's feedback, we found our survey to be most suitable for children and adolescents aged 12–19 years. For younger children, the language and delivery as an online survey does not seem the right format.

Phase 3: Piloting

Our questionnaire was piloted in an online survey with a sample of children and adolescents having at least one parent with a mental illness.

The study was conducted using the online platform SosciSurvey (49) between March and May 2021. All participants were part of a convenience sample—either contacted as part of an ongoing study [COMPARE-family, see (4, 50), via university mailing lists or clinical institutions; e.g., psychiatric clinics, psychotherapy outpatient clinics]. Participants could participate in a raffle of several gifts as study reimbursement. Inclusion criteria for the COPMI-SQ-validation were having a parent with a mental illness and being 12–19 years of age. Participants were not excluded if they themselves had a mental illness.

Measures

Participants responded to questions pertaining to socio-demographic characteristics (age, school grade, living situation, etc.) and questions about their parent's mental illness and their own health status.

COPMI-SQ

Stigma experiences in COPMI were measured via the 93 items in the newly developed COPMI-SQ. Participants were first asked to complete two open-ended sentences: *"I think about people with a mental illness ..."* and *"Others think about people with a mental illness ..."*. Afterwards, they were asked to rate each of the 91 closed items on a visual analog scale (VAS). The VAS consisted of a line with verbal anchors ("strongly disagree" on the far left and "strongly agree" on the far right). All of the items were scored from 1 to 101, with 1 meaning no agreement at all, 101 meaning full agreement. Most of the items were preceded by an item root like *"Because my mother/my father has a mental illness,..."*, others were presented separately. The questionnaire consists of 26 reversed coded items, whose agreement represented a low stigma experience to control for and identify acquiescence response bias (51), see **Supplementary Table S1**. For all other items, the higher the agreement, the greater the experience of stigma.

The first theoretically assumed dimension, *experienced stigma by association (SBA)*, consisted of 18 items representing varied aspects of directly experienced stigmatization as a result of the parental mental illness; for example, experiencing rejection by others, inappropriate and hurtful language or warnings, and obviously discriminatory behavior such as bullying.

The second theoretically assumed dimension, *anticipated SBA*, was captured across 20 items and included fears of stigmatizing behavior or the attitudes of others if they found out about the parental mental illness.

For the third theoretically assumed dimension, *self-stigma,* 23 items were constructed to capture different facets of self-stigma: the feeling of being "contaminated" by the parental illness, the shame associated with the parental illness, feelings of guilt, and feeling generally inferior compared to other people.

The fourth and last theoretically assumed dimension represents the experienced *structural discrimination* and was captured by 30 items. This included discrimination in the education system, the health system, the media, and discrimination in society as a whole. Our instrument's entire structure and all its items are found in **Supplementary Table S1**.

Statistics

Sample Size

We aimed to recruit about 200 participants to validate the scale's psychometric properties. The sample size of 200 is considered adequate for validating an instrument, i.e., for item analyses, and factor analyses to examine the questionnaire's multidimensionality (52, 53).

Missing Values and Outliers

The online survey was designed so that all items had to be answered. Thus, none of the included cases had any missing values. However, there was a filter question addressing parental hospitalization. If this was answered positively, all the subsequent questions were posed only to COPMI whose parent(s) had been hospitalized; this group formed a smaller subsample of only 18 children. All items were analyzed for univariate outliers using boxplots. Conspicuous cases were assessed as realistic after examination and were in a generally inconspicuous range across the mean values achieved; thus they were not excluded from further analyses.

Item Analyses

General Procedure

The item analyses were carried out in three phases. In the first phase, items were checked at the subscale level for appropriate item difficulty and discriminatory power, and subscales were adjusted by removing inappropriate items. Due to content considerations, some psychometrically inappropriate items were retained. In the second phase, the adjusted subscales were combined into an overall scale, the items were rechecked for their characteristic values, and unsuitable items were removed accordingly to improve global consistency and increase the suitability of the overall instrument. In the third phase, the subscales of the COPMI-SQ were adapted to the structure of the final overall instrument, and the final instrument's internal consistency was calculated again. Statistical analyses were carried out using SPSS, version 20.

Item Difficulty Index

Given that the COPMI-SQ is intended to allow differentiation across a broad spectrum of trait values, we aimed for a distribution of item difficulties of $20 < P_i < 80$ [see (54)]. Since extremely difficult, or very easy items hardly allow differentiation between test persons, items with values below 20 and above 80 were considered more closely. Items of very low or extreme difficulty were checked for their discriminatory power and their retention or removal from the item pool was supplemented with content considerations. The exact reasons for retaining or deleting a given conspicuous item are provided in **Supplementary Table S1**.

Discriminatory Power

The item discrimination index was calculated using the corrected item-total-correlation. Values of r_{it} between 0.3 and 0.5 were considered acceptable; $0.5 < r_{it} < 0.7$ is considered good discriminatory power (54). All items whose discriminatory power was below 0.3 were scrutinized, and usually removed to ensure high internal consistency of the subscales and the overall instrument. However, those items with cut-off values below 0.3 were checked for their relevance to the content and either deleted or reformulated when evaluating strongly relevant content. They were then removed one after the other and new item-total correlations were calculated in each case to see how the items interacted. The order of removal of the items and content-related considerations is illustrated in **Supplementary Table S1**.

Internal Reliability

Cronbach's alpha was calculated to measure the COPMI-SQ's internal reliability (55). This should attain a value of at least 0.7 for the subscales—the widely accepted criterion for Cronbach's alpha [e.g., (56)].

RESULTS

Sample

All participants ($N = 32$) were between 13 and 19 years of age (M = 15.75, SD = 2.05). Sixty-five percent of the participants were female. The COPMI usually lived together with their parent with a mental illness (93.8%). Three COPMIs reported having both parents with a mental illness. Mothers were most often affected (60%). The most common parental diagnosis was a mood disorder such as bipolar disorder and depression (maternal: 54.2%; paternal: 82.3%), followed by anxiety disorders (maternal: 16.7%; 11.8% paternal). Almost half of the children stated to have already suffered from a mental illness themselves (46.9%). These were, similar to the parental diagnoses, usually mood disorders (32%). All the sample characteristics are provided in **Table 1**.

As COPMI are an exceedingly difficult target group to reach (57, 58), we were unable to recruit enough COPMI to assess decisively the questionnaire's psychometric goodness of fit, despite intensive recruitment efforts. Nevertheless, according to Johanson and Brooks (59), 30 representative participants from the population of interest suffice for a pilot study to test a new instrument in terms of preliminary item analyses, estimates of internal consistency and proportions of people responding to specific options. For factor and further analyses, more participants are needed, so we focused on the item analyses, especially to reduce the number of items requiring further validation and evaluation of the COPMI-SQ, in order to make the questionnaire more practical and reduce participation thresholds as well as raise the quality of the instrument.

TABLE 1 | Familial and COPMI-specific sample characteristics T1, $N = 32$.

Characteristics	Categories	*n* (%)
Gender[a]	Female	21 (65.6)
	Male	10 (31.3)
	Diverse	1 (3.1)
Age[a]	M (years)	15.75
	SD	2.05
	Min	13
	Max	19
Living situation[b]	Together with ill parent	30 (93.8)
	Without ill parent	2 (6.2)
Parental mental illness[b,c]	Mother	21 (58.3)
	Father	14 (38.9)
Diagnoses mother[b,c]	Mood disorders	13 (54.2)
	Phobia/anxiety disorder	4 (16.7)
	Personality disorders	3 (12.5)
	Posttraumatic stress disorder	1 (4.1)
	Burn-out	1 (4.1)
	Obsessive compulsive disorder	1 (4.1)
	Schizophrenia	1 (4.1)
Diagnoses father[b,c]	Mood disorders	14 (82.3)
	Phobia/anxiety disorder	2 (11.8)
	Pain disorder	1 (5.9)
Children's mental illness[a]	Yes	15 (46.9)
	No	17 (53.1)
Diagnoses child[a,c]	Mood disorder	8 (32.0)
	Phobia/anxiety disorder	4 (16.0)
	AD(H)D	3 (12.0)
	Eating disorder	2 (8.0)
	PTSD	1 (4.0)
	Autism spectrum disorder	1 (4.0)
	OCD	1 (4.0)
	Sleeping disorder	1 (4.0)
	Attachment disorder	1 (4.0)
	Not to be classified	3 (12.0)

n = stated characteristics. [a]Information is based solely on information provided by COPMI themselves; [b]Information is based solely on information provided by parents; [c]Multiple answers possible.

Item Analysis on Subscale Level

Experienced SBA Subscale

Initial Cronbach's alpha equalled 0.940 (see **Table 4**). In the experienced SBA scale, two items revealed low item difficulty, and one item a low item-total correlation. The item with the low item-total correlation was thus removed. However, the two items of low item difficulty were retained after content considerations. Both had a discriminatory power above 0.7. The item *"Because my mother/my father has a mental illness, my friends no longer want to be friends with me"* was retained because this aspect addressing the loss of friendship stood out in the review as a relevant experience for COPMI, and that aspect was not covered by any other item. Similar considerations applied to the item *"Because my mother/my father has a mental illness, I'm bullied at school/university/work"*: the aspect of bullying has been described in the literature, and as it is not covered by any other item in the questionnaire, we retained this item.

Anticipated SBA Subscale

In the anticipated SBA subscale (initial Cronbach's alpha of 0.927), four items revealed a low item-total-correlation; one of them also revealed low difficulty. The item with low discriminatory power and low difficulty was removed first. After considering the content, the other items with too little discriminatory power were also removed one after the other, because they were either covered by other items or classified as non-essential. The step-by-step removal of the items led to an increased item-total correlation within the subscale.

Affiliate Stigma Subscale

In the Affiliate Stigma subscale with an initial alpha of 0.904, two items were conspicuous due to low difficulty and two items due to low discriminatory power. The item with negative discriminatory power was removed first, as it seems to be misleading. Furthermore, its content was already covered by other items. The other item with low discriminatory power was also removed. The item *"Because my mother/father has a mental illness, I don't think I should have children of my own later"* was retained despite its low difficulty because it measures the belief in contamination, which has consequences for future plans, and is not covered by any other items. Furthermore, the other item of low difficulty *"I feel like I'm carrying around a sign: He/she has a mother/father with a mental illness"* was also retained because, it was adapted from an evaluated stigma scale and, additionally, the aspect of "contamination" is otherwise not sufficiently embodied in comparison to "inferiority."

Structural Discrimination Subscale

Out of the Structural Discrimination subscale with an initial alpha of 0.817, 16 items revealed low discriminatory power. Of these, one had very high and one very low item difficulty. The first ones deleted stepwise were those three items with negative item-total correlation, indicating that they measured something completely different. Further items of very low discriminatory power were then successively removed, starting with those intended to measure discrimination within the mental health system. This was because it was clearly overrepresented compared to the other dimensions of the subscale.

The item *"At school, I'd like to learn more about mental illness"* was retained despite its low discriminatory power, firstly because it was on the edge of acceptance ($r_{it} = 0.295$), secondly, in order to maintain the proportions to the other structural stigma dimensions to some extent, and thirdly, because the content of the item was attributed a special significance concerning possible interventions. For the media as a source of structural discrimination, the item "*In the media, mental illness is portrayed negatively, which contributes to many people developing prejudices*" was retained despite its low item-total-correlation; for the reason of having at least two items representing this source of discrimination. In addition, we reworded the item in order to avoid multidimensionality as follows *"Mental illness is portrayed negatively in the media."*

Item Analysis on Total Scale

In the second phase of the item analyses, i.e., when assessing the internal reliability of the overall instrument, one item in the Affiliate Stigma scale and an item in the Structural Discrimination scale showed low item-total correlations and were therefore deleted. **Supplementary Table S1** shows which items were affected.

Revised COPMI-SQ

Our instrument was reduced to a total of 67 items after the three phases of item analyses. The revised instrument is illustrated in **Table 2**. The final version of the *Experienced SBA* subscale now consists of 17 items, with Cronbach's alpha = 0.948 and item-total-correlations from good (lowest 0.395 for the item *"There are people I can talk to about my fears and worries."*) to very high (highest 0.905 for item *"Because my mother/father has a mental illness, my classmates/colleagues/work colleagues tease me."*). The final subscale *Anticipated SBA* (Cronbach's alpha = 0.949) shows item-total correlations between 0.426 (item *"If others found out about my mother's/father's illness, it wouldn't change their behavior toward me")* and 0.877 (item "they'd bad-mouth me."), see **Supplementary Table S1**. The final subscale of *Affiliate Stigma* consists of 19 items, with item-total correlations ranging between 0.449 (item: "I'm just a normal kid like any other.") to 0.820 (item: "Because my mother/my father has a mental illness, I think there's something wrong with me"), and with Cronbach's alpha = 0.933. The item-total-correlations for the final subscale of *Structural Discrimination (STD)* are the lowest compared to the other subscales, and range from 0.282 (item *"In media, mental illness is appropriately portrayed"*), which is actually below acceptable discriminatory power, but will be kept to ensure the media source of discrimination is represented, to 0.705 (item: *"In school, I don't feel disadvantaged because of my mother's/father's illness"*). The revised STD subscale results in a Cronbach's alpha of 0.868.

Table 3 shows our sample's scores on the final subscales and the overall COPMI-SQ instrument. Overall, the sample shows very similar average levels of family stigma on the subscales *Experienced SBA, Anticipated SBA, Affiliate Stigma* and the overall instrument. Here COPMI, with a mean score of 30, tend to be in the lower third of the possible values. On the *STD* subscale, COPMI are in the middle range of values. **Supplementary Table 2** illustrates the intercorrelations of the final instrument scales. Overall, these tend to demonstrate small to very large correlations, all of which are significant.

Internal Reliability

Table 4 shows the internal consistencies for both the initial subscales and subscales adjusted by the item analyses as described above. The internal consistencies for the overall instrument are also visible in **Table 4**. We observed an improvement in all the scales' consistencies. We detected internal consistencies between $\alpha = 0.868$ and $\alpha = 0.975$ across the scales.

DISCUSSION

This study reports on the development and piloting of a questionnaire to assess stigma in children of parents with a mental illness. The COPMI-SQ is the first instrument incorporating different stigma by association dimensions tailored to the stigma experiences of children of parents with a mental illness. After reducing the item number, the final version of the COPMI-SQ consists of 67 items representing four subscales: experienced stigma (17 items), anticipated stigma (16 items), self-stigma (19 items) and structural discrimination (15 items). The

TABLE 2 | Overview of the revised COPMI-SQ after item analyses.

New itemname	Original item	Translation	Intended theoretical dimension	Scoring
Experienced SBA				
	Preceded by: **Weil meine Mutter/mein Vater eine psychische Erkrankung hat, . . .**	Preceded by: **Because my mother/my father has a mental illness, . . .**		
ESBA_01	machen sich andere über meine Mutter/meinen Vater lustig.	others make fun of my mother/father.	hostile behaviors of others	1–101
ESBA_02	reden andere hinter meinem Rücken über die Erkrankung meiner Mutter/meines Vaters.	others talk about my mother's/father's illness behind my back.	hostile behaviors of others	1–101
ESBA_03	lästern andere über mich.	others say awful things about me.	hostile behaviors of others	1–101
ESBA_04	lachen andere mich aus.	others laugh at me.	hostile behaviors of others	1–101
ESBA_05	tratschen andere das weiter.	others gossip about it.	hostile behaviors of others	1–101
ESBA_06	gehen andere mir aus dem Weg.	others avoid me.	withdrawal and rejection	1–101
ESBA_07	haben andere Angst vor meiner Mutter/meinem Vater oder mir.	others are afraid of my mother/father or me.	inappropriate language and contents	1–101
ESBA_08	möchten meine Freund*innen nicht mehr mit mir befreundet sein.	my friends no longer want to be friends with me.	withdrawal and rejection	1–101
ESBA_09	wollen sich meine Mitschüler*innen / Kommiliton*innen/ Arbeitskolleg*innen nicht mehr mit mir treffen.	my classmates/ colleagues/ work colleagues no longer want to get together with me.	withdrawal and rejection	1–101
ESBA_10	ärgern mich meine Mitschäler*innen/ Kommiliton*innen/ Arbeitskolleg*innen.	my classmates/colleagues/work colleagues aggravate me.	hostile behaviors of others	1–101
ESBA_11	werde ich in der Schule / Uni / auf der Arbeit gemobbt.	I'm bullied at school/university/work.	hostile behaviors of others	1–101
ESBA_12	wussten andere nicht, wie sie passend darauf reagieren / damit umgehen sollten.	others did not know how to react to or deal with it appropriately.	inappropriate language and contents	1–101
ESBA_13	haben andere mir geraten, selbst keine Kinder zu bekommen.	others have advised me not to have children myself.	inappropriate language and contents	1–101
ESBA_14	haben andere verletzende Sachen über mich oder meine Mutter/meinen Vater gesagt.	others have said hurtful things about me or my mother/father.	hostile behaviors of others	1–101
ESBA_15	Es gibt Leute, mit denen ich über meine Ängste und Sorgen reden kann.	There are people I can talk to about my fears and worries.	withdrawal and rejection	101–1
	Separate items without an item root			
ESBA_16	Andere Leute möchten nicht mit mir über die Erkrankung meiner Mutter/meines Vaters sprechen.	Other people don't want to talk to me about my mother's/father's illness.	withdrawal and rejection	1–101
ESBA_17	Wenn ich wegen der Erkrankung meiner Mutter/meines Vaters Hilfe brauche, gibt es Personen. mit denen ich sprechen kann.	If I need help because of my mother's/father's illness, there are people I can turn to.	withdrawal and rejection	101–1

(Continued)

TABLE 2 | Continued

New itemname	Original item	Translation	Intended theoretical dimension	Scoring
Anticipated SBA				
	Preceded by: **Wenn andere von der Erkrankung meiner Mutter/meines Vaters erfahren würden,. . .**	Preceded by: **If others found out about my mother's/ father's illness,. . .**		
ASBA_01	würden sie sich über meine Mutter/meinen Vater lustig machen.	they'd make fun of my mother/father.	fearing hostile behaviors	1–101
ASBA_02	würden sie hinter meinem Rücken schlecht über die Erkrankung meiner Mutter/meines Vaters reden.	they'd talk badly about my mother's/father's illness behind my back	fearing hostile behaviors	1–101
ASBA_03	würden sie über mich lästern.	they would badmouth me.	fearing hostile behaviors	1–101
ASBA_04	würde das an ihrem Verhalten mir gegenüber nichts ändern	it wouldn't change their behavior toward me.	fearing hostile behaviors	1–101
ASBA_05	würden sie mich auslachen.	they'd laugh at me.	fearing hostile behaviors	1–101
ASBA_06	würden sie das für sich behalten.	they'd keep it to themselves.	fearing hostile behaviors	1–101
ASBA_07	würden sie mir aus dem Weg gehen.	they'd avoid me.	fearing lack of understanding rejection	1–101
ASBA_08	würden sie Angst vor meiner Mutter/meinem Vater oder mir bekommen	They'd become afraid of my mother/father or me.	fearing of negative attitudes and ascriptions	1–101
ASBA_09	würden sich meine Mitschüler*innen/ Kommiliton*innen/ Arbeitskolleg*innen nicht mit mir treffen wollen.	my classmates/ fellow students/ colleagues would no longer want to get together with me.	fearing lack of understanding rejection	1–101
ASBA_10	würden mich meine Mitschüler*innen/ Kommiliton*innen/ Arbeitskolleg*innen ärgern.	my classmates/ fellow students/ colleagues at work would get angry with me.	fearing hostile behaviors	1–101
ASBA_11	würde ich in der Schule / Uni / auf der Arbeit gemobbt werden.	I'd be bullied at school/university/work.	fearing hostile behaviors	1–101
ASBA_12	würden andere mir raten, selbst keine Kinder zu bekommen.	others would advise me not to have children myself.	fearing of negative attitudes and ascriptions	1–101
ASBA_13	würden sie verletzende Sachen über mich oder meine Mutter/ meinen Vater sagen.	they'd say hurtful things about me or my mother/father.	fearing hostile behaviors	1–101
	Preceded by: **Wenn Fachleute (Jugendamt/Psycholog*innen/Sozialarbeiter*innen. etc.) von der Erkrankung meiner Mutter/meines Vaters erfahren würden. . . .**	Preceded by: **If professionals (youth welfare office/psychologists/social workers. etc.) found out about my mother's/father's illness. . . .**		
ASBA_14	könnte ich weiterhin zu Hause wohnen bleiben.	I could still keep living at home.	fearing of negative attitudes and ascriptions	101–0
ASBA_15	bringe ich selten neue Freund*innen mit nach Hause, aus Angst. dass sie dann nicht mehr mit mir befreundet sein wollen.	I rarely bring new friends home for fear that they won't want to be friends with me anymore.	fearing lack of understanding and rejection	1–101

(Continued)

TABLE 2 | Continued

New itemname	Original item	Translation	Intended theoretical dimension	Scoring
	Seperate item without an item root			
ASBA_16	Ich habe kein Problem damit, meinen Freund*innen meine (erkrankte) Mutter/meinen (erkrankten) Vater vorzustellen.	I have no problem introducing my (ill) mother/father to my friends.	fearing any harmful reaction	101–1
Affiliate Stigma				
	Preceded by: **Weil meine Mutter/mein Vater eine psychische Erkrankung hat, . . .**	Preceded by: **Because my mother / my father has a mental illness, . . .**		
AS_01	denke ich, mit mir stimmt etwas nicht.	I think there's something wrong with me.	beliefs of being inferior	1–101
AS_02	versuche ich, mich besonders normal und unauffällig zu verhalten, damit andere mir nicht anmerken, dass ich nicht normal bin.	I try to act particularly normal and inconspicuous so that others do not notice that I am not normal.	beliefs of being contaminated	1–101
AS_03	nehmen andere wahr, dass ich anders / komisch bin.	others notice that I am different/weird.	beliefs of being contaminated	1–101
AS_04	habe ich Angst, mich anstecken zu können.	I am afraid of catching the illness.	beliefs of being contaminated	1–101
AS_05	denke ich bei kleinsten Anzeichen, dass ich dieselbe Erkrankung habe wie meine. . .	I think at the slightest sign that I have the same illness as my mother/father.	beliefs of being contaminated	1–101
AS_06	denke ich, dass ich später keine eigenen Kinder bekommen sollte.	I don't think I should have children of my own later on.	beliefs of being contaminated	1–101
AS_07	fühle ich mich weniger wert.	I feel less worthy.	beliefs of being inferior	1–101
AS_08	ist meine Familie nicht richtig.	my family is not right.	beliefs of being inferior	1–101
AS_09	fühle ich mich schuldig.	I feel guilty.	beliefs of being inferior	1–101
AS_10	schäme ich mich.	I feel ashamed.	beliefs of being inferior	1–101
	Seperate items without an item root			
AS_11	Ich fühle mich, als würde ich ein Schild mit mir herumtragen: "Er/Sie hat eine Mutter/einen Vater mit einer psychischen Erkrankung"	I feel like I'm carrying around a sign: "He/she has a mother/father with a mental illness".	beliefs of being contaminated	1–101
AS_12	Ich bin ein ganz normales Kind, wie jedes andere auch.	I'm just a normal kid like any other.	beliefs of being inferior	101–1
AS_13	Weil ich so ein schwieriges Kind bin, ist meine Mutter/ ist mein Vater erkrankt.	Because I am such a difficult child, my mother/ father has become ill.	beliefs of being inferior	1–101
AS_14	Ich bin (mit-)verantwortlich dafür, dass sich der Zustand meiner Mutter/meines Vaters nicht verbessert.	I am (co-)responsible for the fact that the condition of my mother/father isn't improving.	beliefs of being inferior	1–101

(Continued)

TABLE 2 | Continued

New itemname	Original item	Translation	Intended theoretical dimension	Scoring
AS_15	Ich muss die Erkrankung meiner Mutter/meines Vaters geheim halten.	I have to keep my mother's/father's illness a secret.	beliefs of being inferior	1–101
AS_16	Wenn ich die Erkrankung meiner Mutter/meines Vaters beschreibe, spiele ich die Schwere der Erkrankung herunter.	When I describe my mother's/father's illness, I downplay the severity of it.	beliefs of being inferior	1–101
AS_17	Mir ist es peinlich, dass meine Mutter/mein Vater eine psychische Erkrankung hat.	I'm embarrassed that my mother/father has a mental illness.	beliefs of being inferior	1–101
AS_18	Ich schäme mich dafür, dass meine Mutter/mein Vater nicht wie andere Mütter/Väter ist.	I'm ashamed that my mother/father isn't like other mothers/fathers.	beliefs of being inferior	1–101
AS_19	Wenn meine Mutter/mein Vater wegen ihrer/seiner Erkrankung verurteilt wird, fühle ich mich auch verurteilt.	If my mother/father is judged because of her/his illness, I feel judged too.	beliefs of being contaminated	1–101
Structural Discrimination				
	Preceded by (after a filter question whether the parent ever has been hospitalized before): **Wenn meine Mutter/ mein Vater aufgrund der psychischen Erkrankung im Krankenhaus war, . . .**	Preceded by: **When my mother/father was in hospital because of the mental illness, . . .**		
STD_01	konnte ich das Personal immer ansprechen, wenn ich Fragen zur Erkrankung meiner Mutter/meines Vaters hatte.	I could always approach the staff if I had any questions about my mother's/father's illness.	health care system	101–1
STD_02	hätte ich gerne mehr Informationen vom Krankenhauspersonal bekommen.	I'd have liked to get more information from the hospital staff.	health care system	1–101
STD_03	fühlte ich mich vom Krankenhauspersonal gut einbezogen und informiert.	I felt well integrated and informed by the hospital staff.	health care system	101–1
STD_04	fühlte ich mich, als wäre ich dort unerwünscht.	I felt like I was unwanted there.	health care system	1–101
STD_05	war die Beziehung zwischen mir und dem Krankenhauspersonal gut.	the relationship between me and the hospital staff was good.	health care system	101–1
	Seperate Item without an item root:			
STD_06	Meiner Mutter/meinem Vater wurde durch das Gesundheitssystem nicht genug geholfen.	My mother/father wasn't helped enough by the health system.	health care system	1–101

(Continued)

TABLE 2 | Continued

New itemname	Original item	Translation	Intended theoretical dimension	Scoring
	Preceded by: **In der Schule...**	Preceded by: **At school....**		
STD_07	würde ich gerne mehr über psychische Erkrankungen erfahren.	I'd like to learn more about mental illness.	educational system	1–101
STD_08	kann ich mit meinen Lehrer*innen über die Erkrankung meiner Mutter/meines Vaters sprechen.	I can talk to my teachers about my mother's or father's illness.	educational system	1–101
STD_09	gehen die Lehrer*innen auf mich und meine Schwierigkeiten zu Hause ein.	the teachers respond to me and my difficulties at home.	educational system	101–1
STD_10	fühle ich mich wegen der Erkrankung meiner Mutter/meines Vaters nicht benachteiligt.	I don't feel disadvantaged because of my mother's/father's illness.	educational system	101–1
	Preceded by: **In den Medien...**	Preceded by: **In the media,**		
STD_11	werden psychische Erkrankungen angemessen dargestellt.	Mental illness is portrayed appropriately.	media	101–1
STD_12	werden psychische Erkrankungen negativ dargestellt.	Mental illness is portrayed negatively.	media	1–101
	Seperate items without an item root			
STD_13	Ich erhalte von niemandem ausreichend Informationen über die Erkrankung meiner Mutter/meines Vaters.	I don't get enough information from anyone about my mother's/father's illness.	general	1–101
STD_14	Ich weiß genau, an welche (professionellen) Stellen in mich wenden kann, wenn ich Hilfe wegen der Erkrankung meiner Mutter/meines Vaters benötige.	I know exactly which (professional) places I can turn to if I need help because of my mother's/father's illness....	general	101–1
STD_15	Es gibt ausreichend Hilfsangebote für meine Eltern und mich.	There's enough help available for my parents and me.	general	101–1

Items with scoring 1–101 are the normal coded items, with 1 meaning no agreement at all, 101 meaning full agreement. The higher the agreement the higher the stigma experiences. Items with scoring 101–1 are the reversed coded items, also referring to 101 as full agreement. The higher the agreement, the lower the stigma for these items.

TABLE 3 | Sample characteristics revised instrument version COPMI-SQ.

Scale	n	Min	Max	M	SD
Experienced SBA	32	1.19	73.18	28.36	22.40
Anticipated SBA	32	1.00	73.94	34.59	25.28
Affiliate stigma	32	1.05	62.16	30.00	20.25
Structural discrimination	32	12.73	82.33	54.27	16.78
COPMI SQ total	32	7.64	66.24	35.50	18.36

Scale characteristics of the sample on the revised subscales and the overall instrument COPMI-SQ after adjustments according to the item analyses.

TABLE 4 | Changes in the instrument structure through internal consistency analysis.

Scale	α initial	α final	Initial number of items	Revised number of items
Experienced SBA	0.940	0.948	18	17
Anticipated SBA	0.927	0.949	20	16
Affiliate stigma	0.904	0.933	23	19
Structural discrimination	0.817	0.868	31	15
COPMI-SQ—total	0.970	0.975	92	67

Results of the consistency analysis. The initial internal consistency refers to all original items of a scale.

COPMI-SQ, once validated, can be used to better understand the complex phenomenon of stigma by association among young people and also to target the experiences and needs of this group in intervention studies or anti-stigma campaigns. As the questionnaire also captures which parental condition in which parent is present, it can be used to create insights to the extent to which stigma experiences vary for different parental disorders. In addition, it could potentially support shedding light on the gendered theoretical landscape of parenting.

Developing the Instrument

The approach we took to develop the instrument followed scientifically accepted principles (20) by ensuring various construction phases. The construction process relied on (family) stigma theories (14, 20, 39, 60, 61), incorporated qualitative results from a comprehensive systematic review to best reflect the expressed stigma experiences of affected children (28), and included expert opinions on the initially constructed items. The COPMI-SQ is thus considered a solid basis for further reliability and validity studies.

Item Reduction

We ran into difficulty with item analyses due to the fact that it was not a test construction for which clear rules could be applied while considering the required item difficulties or discriminatory power. Although recommendations could be used as a guide, each change or reduction of the item pool had to be made by carefully considering the content. For example, the subscale "Structural Discrimination" represents a very heterogeneous set of items, as they were meant to represent different institutional sources of discrimination, for example the educational system, as well as the media and the health care system. In this respect, we had to assume that discrimination perceived in one societal structure does not necessarily go hand in hand with a high level of discrimination in another, as there are unintended public and private sector policies that restrict opportunities for some groups (62) and we do not yet know precisely how they interact (62, 63). This means that the item-total correlations it did for not have to meet the same requirements as did the other subscales. We also had to make sure we did not over- or under-represent some sub-aspects.

Nevertheless, a detailed explanation of the reasons for deleting or retaining the items ensures transparency, and very high item-total correlations were achieved overall. Only in the "Structural Discrimination" subscale one item with a low item-total-correlation, namely "In the media, mental illness is appropriately portrayed" remained in the questionnaire. This can be justified due to the item's relevance to the content and that its discriminatory power of 0.282 was only minimally below the acceptable limit of 0.3. Nevertheless, items of low discriminatory power or showing low item-total correlations must be examined more closely in the further validation course in a factor analysis.

The very high values regarding internal reliability and the increase in Cronbach's alpha in each subscale, as well as in the final instrument compared to its first version suggest that we made the right decisions here.

We detected overall very strong correlations between the various subscales in the questionnaire's revised form. This may be an initial indication that also a one factor model may be considered. In a further validation study, factor structure and dimensionality have to be examined.

Evaluating Reliability and Validity

The final COPMI-SQ's internal reliability can be considered as very good. It demonstrates good to excellent internal consistency in the subscales, as well as excellent internal reliability of the instrument as a whole. We ensured content validity through an expert panel, and evaluated the difficulty of understanding items with children themselves. However, due to the impossibility of conducting a factor analysis, whether these subscales are confirmed by the instrument's factor structure and multidimensionality cannot be assessed yet.

Test-retest reliability could not be assessed due to an insufficient sample size. However, we can assume that stigma experiences are also not constant variables. New experiences can always emerge, so that if test-retest reliability is to be measured, there should be a shorter time span between them. Furthermore, the mere questioning of these experiences could also lead to some young people, especially when confronted with the topic for the first time through the questionnaire, potentially being sensitized to subsequent experiences and accordingly revealing higher values at a later point in time.

Strengths and Limitations

The greatest strength of the present study is that the COPMI-SQ was theory-based and developed supported by an extensive systematic review. In addition, the various steps in its development are presented transparently and in detail.

There are several areas for improvement and further investigations. About half of the participants reported having a mental illness of their own—a good representation of the population of children of parents with mental illness (64), but this means that their responses may be cofounded by experiencing stigma due to their own illness. Due to recruitment difficulties, we were unable to identify enough participants to permit a factor analysis and investigate multidimensionality and factor structure. The assumed four factors are not supported by the very high inter-correlations of the assumed subscales, but they cannot be ruled out either. However, the study is still ongoing. With the reduced number of items due to the item analyses, we can reach our sample size more easily. Other recruitment strategies, such as recruiting via schools, must be integrated in the future to reach this vulnerable target group. Since this paper is concerned with the methodology of developing a new instrument, content-related considerations of the different dimensions of stigma have been marginalized. A more detailed discussion of the meaning and interconnectedness of the different dimensions can be found in the paper reporting the results of the systematic review (28).

Implications for Further Research and Use of the COPMI-SQ

In a further validation study, we will aim to recruit approximately 200 COPMI to examine our questionnaire's multidimensionality. In the first step, this should be examined via an exploratory factor analysis (EFA). An EFA is preferable to a confirmatory factor analysis (CFA), since it is possible that factor structures other than those expected might emerge (45). Especially as high inter-correlations between the subscales might indicate a one-factor structure, uni-dimensionality should also be checked. A CFA should then be conducted on a separate sample to confirm the structure of the scales resulting from the EFA.

A further validated COPMI-SQ could be used to help develop anti-stigma and general interventions for this population. Our first pilot data promises good reliability of the a priori assumed subscales. As our scale is constructed for young people aged 12–19, it can be used as a starting point to develop a scale addressing the same problem for younger children. A more creative and interactive way of getting those information from younger children, should be developed, to be able to incorporate younger children's views as well. Research in children's lived experiences is limited, and this is especially true for younger children.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Committee of the Psychology Department of Philipps University of Marburg. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

L-MD designed and completed the study, involved as the main person in charge in all development and analyses steps, and wrote the manuscript. MS co-organized the data collection process and supported the writing of the manuscript by reviewing and editing. KV assisted with data collection and data analysis. HC supervised the study and preparation of the manuscript and supported the writing of the manuscript by reviewing and editing.

FUNDING

Funding was provided by the Working Group on Clinical Child and Adolescent Psychology, Department of Psychology, Philipps University of Marburg. The funding for the raffle was provided by COMPARE-family. COMPARE-family is funded by the Federal Ministry of Education and Research (BMBF) (01GL1748B). The funders did not influence the collection, analysis, and interpretation of data and played no role in writing the manuscript.

ACKNOWLEDGMENTS

The authors would like to thank Simone Pfeiffer from the University of Landau and Cornelia Weise from Philipps University of Marburg for their crucial contributions regarding the formulation of the items, thanks to their expertise in the field of stigma and questionnaire development. The authors would also like to thank all the participating COPMI and their parents. We know it can be difficult and painful to provide information about such experiences. We hope to contribute to improving care and, above all, to breaking the silence.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.800037/full#supplementary-material

REFERENCES

1. van Santvoort F, Hosman CMH, van Doesum KTM, Janssens JMAM, et al. Children of mentally ill parents participating in preventive support groups. *Parental Diagnoses and Child Risk J Child Fam Stud.* (2014) 23:67–75. doi: 10.1007/s10826-012-9686-x
2. Maybery DJ, Reupert AE, Patrick K, Goodyear M, Crase L. Prevalence of parental mental illness in Australian families. *Psychiatr Bull.* (2009) 33:22–6. doi: 10.1192/pb.bp.107.018861
3. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands. I The scientific basis to a comprehensive

approach. *Aust e-J Adv Mental Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250

4. Christiansen H, Reck C, Zietlow A-L, Otto K, Steinmayr R, Wirthwein L, et al. Children of mentally III parents at risk evaluation (COMPARE). Design and methods of a randomized controlled multicenter study-Part I. *Front Psychiatry.* (2019) 10:128. doi: 10.3389/fpsyt.2019.00128
5. Gladstone BM, Boydell KM, McKeever P. Recasting research into children's experiences of parental mental illness. Beyond risk and resilience. *Soc Sci Med.* (2006) 62:2540–50. doi: 10.1016/j.socscimed.2005.10.038
6. Radicke A, Barkmann C, Adema B, Daubmann A, Wegscheider K, Wiegand-Grefe S, et al. Children of parents with a mental illness. Predictors of health-related quality of life and determinants of child-parent agreement. *Int J Environ Res Public Health.* (2021) 18:379. doi: 10.3390/ijerph18020379
7. Reupert AE, Maybery J, Kowalenko D. Children whose parents have a mental illness. Prevalence, need and treatment. *MJA Open.* (2012) 1:7–9. doi: 10.5694/mjao11.11200
8. Cogan N, Riddell S, Mayes G. Children living with an affectively ill parent. How do they cope? *Educ Child Psychol.* (2005) 22:16–28.
9. Gladstone BM, Boydell KM, Seeman MV, McKeever PD. Children's experiences of parental mental illness. A literature review. *Early Interv Psychiatry.* (2011) 5:271–89. doi: 10.1111/j.1751-7893.2011.00287.x
10. Reedtz C, Lauritzen C, Stover YV, Freili JL, Rognmo K. Identification of children of parents with mental illness. A necessity to provide relevant support. *Front Psychiatry.* (2019) 9:358. doi: 10.3389/fpsyt.2018.00728
11. Maybery D, Reupert AE. The number of parents who are patients attending adult psychiatric services. *Curr Opin Psychiatry.* (2018) 31:358–62. doi: 10.1097/YCO.0000000000000427
12. Koschorke M, Evans-Lacko S, Sartorius N, Thornicroft G. Stigma in different cultures. In: Gaebel W, Roessler, Sartorius N, editors. *The Stigma of Mental Illness - End of the Story?*. Berlin: Springer International Publishing (2017). p. 67–82.
13. Henderson C, Evans-Lacko S, Thornicroft G. Mental illness stigma, help seeking, and public health programs. *Am J Public Health.* (2013) 103:777–80. doi: 10.2105/AJPH.2012.301056
14. Bos AER, Pryor JB, Reeder GD, Stutterheim SE. Stigma. *Adv Theory Res Basic Appl Soc Psychol.* (2013) 35:1–9. doi: 10.1080/01973533.2012.746147
15. Goffman E. *Stigma*. Frankfurt/M: Über Techniken der Bewältigung beschädigter Identität (1967).
16. Hebl MR, Dovidio JF. Promoting the "social" in the examination of social stigmas. *Pers Soc Psychol Rev.* (2005) 9:156–82. doi: 10.1207/s15327957pspr0902_4
17. Corrigan P, Watson AC. Understanding the impact of stigma on people with mental illness. *World Psychiatry.* (2002) 1:16–20.
18. Rössler W. The stigma of mental disorders: A millennia-long history of social exclusion and prejudices. *EMBO Rep.* (2016) 17:1250–3. doi: 10.15252/embr.201643041
19. van der Sanden RLM, Bos AER, Stutterheim SE, Pryor JB, Kok G. Experiences of stigma by association among family members of people with mental illness. *Rehabil Psychol.* (2013) 58:73–80. doi: 10.1037/a0031752
20. Miller PW, Corrigan FE. Shame, blame, and contamination. A review of the impact of mental illness stigma on family members. *J Mental Health.* (2009) 13:537–48. doi: 10.1080/09638230400017004
21. Park S, Park KS. Family stigma. A concept analysis. *Asian Nurs Res.* (2014) 8:165–71. doi: 10.1016/j.anr.2014.02.006
22. Campbell DT. Common fate, similarity, and other indices of the status of aggregates of persons as social entities. *Syst Res.* (1958) 3:14–25. doi: 10.1002/bs.3830030103
23. Pryor JB, Reeder GD, Monroe AE. The infection of bad company. Stigma by association. *J Pers Soc Psychol.* (2012) 102:224–41. doi: 10.1037/a0026270
24. Dam K, Hall EOC. Navigating in an unpredictable daily life. A metasynthesis on children's experiences living with a parent with severe mental illness. *Scand J Caring Sci.* (2016) 30:442–57. doi: 10.1111/scs.12285
25. Griffiths J, Norris E, Stallard P, Matthews S. Living with parents with obsessive-compulsive disorder. Children's lives and experiences. *Psychol Psychother.* (2012) 85:68–82. doi: 10.1111/j.2044-8341.2011.02016.x
26. Blakeman M, Martin C, Gupta A. Making sense of growing up with a parent with psychosis. An interpretative phenomenological analysis study. *Psychosis.* (2019) 11:54–62. doi: 10.1080/17522439.2019.1573916
27. Wahl P, Otto C, Lenz A. "…dann würde ich traurig werden, weil alle über meinen Papa reden – Die Rolle des Stigmas in Familien mit psychisch erkranktem Elternteil („…I would be sad then, because they are all talking about my dad"—The Role of Stigma in Families Affected by Parental Mental Illness). *Gesundheitswesen.* (2017) 79:987–92. doi: 10.1055/s-0043-122234
28. Dobener L-M, Fahrer J, Purtscheller D, Paul J, Christiansen H. How do children of parents with mental illness experience stigma? A systematic mixed studies review. *Front Psychiatry.* (2022) 13:813519. doi: 10.3389/fpsyt.2022.813519
29. Reupert A, Gladstone B, Helena Hine R, Yates S, McGaw V, Charles G, et al. Stigma in relation to families living with parental mental illness. An integrative review. *Int J Ment Health Nurs.* (2021) 30:6–26. doi: 10.1111/inm.12820
30. Corrigan P, Watson AC, Miller FE. Blame, shame, and contamination. The impact of mental illness and drug dependence stigma on family members. *J Fam Psychol.* (2006) 20:239–46. doi: 10.1037/0893-3200.20.2.239
31. Koschade JE, Lynd-Stevenson RM. The stigma of having a parent with mental illness. Genetic attributions and associative stigma. *Aust J Psychol.* (2011) 63:93–9. doi: 10.1111/j.1742-9536.2011.00009.x
32. Fox AB, Earnshaw VA, Taverna EC, Vogt D. Conceptualizing and measuring mental illness stigma. The mental illness stigma framework and critical review of measures. *Stig Health.* (2018) 3:348–76. doi: 10.1037/sah0000104
33. Pescosolido BA, Martin JK. The stigma complex. *Annu Rev Sociol.* (2015) 41:87–116. doi: 10.1146/annurev-soc-071312-145702
34. Mak WWS, Cheung RYM. Affiliate stigma among caregivers of people with intellectual disability or mental illness. *J Appl Res Intellect Disabil.* (2008) 21:532–45. doi: 10.1111/j.1468-3148.2008.00426.x
35. Girma E, Möller-Leimkühler AM, Dehning S, Mueller N, Tesfaye M, Froeschl G, et al. Self-stigma among caregivers of people with mental illness. Toward caregivers' empowerment. *J Multidiscip Healthc.* (2014) 7:37–43. doi: 10.2147/JMDH.S57259
36. Morris E, Hippman C, Murray G, Michalak EE, Boyd JE, Livingston J, et al. Self-stigma in relatives of people with mental illness scale. Development and validation. *Br J Psychiatry.* (2018) 212:169–74. doi: 10.1192/bjp.2017.23
37. Struening EL, Perlick DA, Link BG, Hellman F, Herman D, Sirey JA, et al. Stigma as a barrier to recovery. The extent to which caregivers believe most people devalue consumers and their families. *Psychiatr Serv.* (2001) 52:1633–8. doi: 10.1176/appi.ps.52.12.1633
38. Werner P, Goldstein D, Heinik J. Development and validity of the Family Stigma in Alzheimer's disease Scale (FS-ADS). *Alzheimer Dis Assoc Disord.* (2011) 25:42–8. doi: 10.1097/WAD.0b013e3181f32594
39. Yin Y, Zhang W, Hu Z, Jia F, Li Y, Xu H, et al. Experiences of stigma and discrimination among caregivers of persons with schizophrenia in China. *A field survey PLoS ONE.* (2014) 9:e108527. doi: 10.1371/journal.pone.0108527
40. Zisman-Ilani Y, Levy-Frank I, Hasson-Ohayon I, Kravetz S, Mashiach-Eizenberg M, Roe D. Measuring the internalized stigma of parents of persons with a serious mental illness. The factor structure of the parents' internalized stigma of mental illness scale. *J Nerv Ment Dis.* (2013) 201:183–7. doi: 10.1097/NMD.0b013e3182845d00
41. Zuckerman KE, Lindly OJ, Reyes NM, Chavez AE, Cobian M, Macias K, et al. Parent perceptions of community autism spectrum disorder stigma. Measure validation and associations in a multi-site sample. *J Autism Dev Disord.* (2018) 48:3199–209. doi: 10.1007/s10803-018-3586-x
42. Yıldız M, İncedere A, Kiras F, Abut FB, Kircalı A, İpçi K. Development of self-stigma inventory for families of the patients with schizophrenia (SSI-F). Validity and reliability study. *Psychiatry Clin Psychopharmacol.* (2019) 29:463–71. doi: 10.1080/24750573.2018.1480858
43. Tessler RC. *Family Experiences With Mental Illness.* Westport, Conn: Richard Tessler and Gail Gamache (2000).
44. Boateng GO, Neilands TB, Frongillo EA, Melgar-Quiñonez HR, Young SL. Best practices for developing and validating scales for health, social, and behavioral research: a primer. *Front Public Health.* (2018) 6:149. doi: 10.3389/fpubh.2018.00149
45. Carpenter S. Ten steps in scale development and reporting. A guide for researchers. *Commun Methods Measures.* (2017) 12:25–44. doi: 10.1080/19312458.2017.1396583
46. Bell N. Ethics in child research: rights, reason and responsibilities. *Children's Geogr.* (2008) 6:7–20. doi: 10.1080/14733280701791827

47. Zentrale Ethikkommission. Stellungnahme der Zentralen Kommission zur Wahrung ethischer Grundsätze in der Medizin und ihren Grenzgebieten (Zentrale Ethikkommission) bei der Bundesärztekammer zur Forschung mit Minderjährigen. *Deutsches Ärzteblatt.* (2004) 101:1613–1617.
48. Hinkin TR. A brief tutorial on the development of measures for use in survey questionnaires. *Organ Res Methods.* (1998) 1:104–21. doi: 10.1177/109442819800100106
49. Leiner DJ. *SoSci Survey (Version 3, 1.06).* (2019). Available online at: https://www.soscisurvey.de (accessed September 22, 2021).
50. Stracke M, Gilbert K, Kieser M, Klose C, Krisam J, Ebert DD, et al. COMPARE Family (Children of Mentally Ill Parents at Risk Evaluation). Study protocol for a preventive intervention for children of mentally ill parents (Triple P, evidence-based program that enhances parentings skills, in addition to gold-standard cbt with the mentally ill parent) in a multicenter RCT-Part II. *Front Psychiatry.* (2019) 10:54. doi: 10.3389/fpsyt.2019.00054
51. Herche J, Engelland B. Reversed-polarity items and scale unidimensionality. *J Acad Market Sci.* (1996) 24:366–74. doi: 10.1177/0092070396244007
52. Guadagnoli E. *The Relationship of Sample Size to the Stability of Component Patterns: A Simulation Study*. University of Rhode Island, Open Access Master's Theses. Paper 1575 (1984). Available online at: https://digitalcommons.uri.edu/theses/157
53. Comrey AL. Factor-analytic methods of scale development in personality and clinical psychology. *J Consult Clin Psychol.* (1988) 56:754–61. doi: 10.1037/0022-006X.56.5.754
54. Moosbrugger H, Kelava A. *Testtheorie und Fragebogenkonstruktion.* Berlin: Springer (2012).
55. Cronbach LJ. Coefficient alpha and the internal structure of tests. *Psychometrika.* (1951) 16:297–334. doi: 10.1007/BF02310555
56. Hossiep R. Cronbachs alpha. In: Wirtz MA, editor. *Lexikon der Psychologie*. Bern: Hogrefe (2021).
57. Grové C. Using social networking sites in research. An emerging approach to engaging with young people who have a parent with a mental illness and/or substance abuse disorder. *Front Psychiatry.* (2019) 10:76. doi: 10.3389/fpsyt.2019.00281
58. van Doesum KTM, Riebschleger J, Carroll J, Grové C, Lauritzen C, Mordoch E, et al. Successful recruitment strategies for prevention programs targeting children of parents with mental health challenges. An international study. *Child Youth Serv.* (2016) 37:156–74. doi: 10.1080/0145935X.2016.1104075
59. Johanson GA, Brooks GP. Initial scale development. Sample size for pilot studies. *Educ Psychol Meas.* (2010) 70:394–400. doi: 10.1177/0013164409355692
60. Fife B, Wright ER. The dimensionality of stigma: a comparison of its impact on the self of persons with HIV/AIDS and Cancer. *J Health Soc Behav.* (2000) 41:50–67.
61. Link BG, Phelan JC. Conceptualizing stigma. *Annu Rev Sociol.* (2001) 27:363–85. doi: 10.1146/annurev.soc.27.1.363
62. Corrigan PW, Markowitz FE, Watson AC. Structural levels of mental illness stigma and discrimination. *Schizophr Bull.* (2004) 30:481–91. doi: 10.1093/oxfordjournals.schbul.a007096
63. Hatzenbuehler ML. Structural stigma. Research evidence and implications for psychological science. *Am Psychol.* (2016) 71:742–51. doi: 10.1037/amp0000068
64. Leijdesdorff S, van Doesum K, Popma A, Klaassen R, van Amelsvoort T. Prevalence of psychopathology in children of parents with mental illness and/or addiction. An up to date narrative review. *Curr Opin Psychiatry.* (2017) 30:312–7. doi: 10.1097/YCO.0000000000000341

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

PERSPECTIVE
published: 11 April 2022
doi: 10.3389/fpsyt.2022.806550

Perspective: Implications of the COVID-19 Pandemic for Family-Focused Practice With Parents With Mental Illness and Their Families

Angela Obradovic[1] *and Joanne Nicholson*[2]*

[1] Northern Area Mental Health Services, Melbourne, VIC, Australia, [2] The Heller School for Social Policy and Management, Institute for Behavioral Health, Brandeis University, Waltham, MA, United States

The goal of this perspective piece is to suggest challenges to family-focused practice with parents with mental illness and their children that have emerged during the COVID-19 pandemic. We discuss implications for practice, policy, and research that will benefit from rigorous study in the future, as we sift through lessons learned. The impact of the pandemic on the mental health and well-being of people around the world has been documented. Common adaptations in service delivery have included a shift to telehealth and digital tools. The pandemic has posed challenges to practice across the EASE Framework components for family-focused practice (i.e., Engage, Assess, Support, and Educate) for both parents/families and practitioners. Governmental policy and funding responses will be critical to addressing the impact of stresses, disruptions and losses endured during the past months. Pandemic experiences and consequences have implications for research measures, methods, and outcomes, given the dramatic changes in people's lives and the contexts in which they live. The shift to research implementation in virtual environments has resulted in challenges in maintaining confidentiality, and the privacy and security of data. As we move forward, it will be important to acknowledge the remaining uncertainty about the future and accommodate the profound changes in family life, professional practice, and research implementation related to the pandemic in our efforts to demonstrate the effectiveness of positive lessons learned while developing new approaches for dealing with the negative consequences of the pandemic.

Keywords: family-focused practice, parents with mental illness, children of parents with mental illness, pandemic, adaptations, COVID-19

OPEN ACCESS

Edited by:
Lawrence T. Lam,
University of Technology
Sydney, Australia

Reviewed by:
Joanna Fox,
Anglia Ruskin University,
United Kingdom

Correspondence:
Joanne Nicholson
jnicholson@brandeis.edu

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *31 October 2021*
Accepted: *21 March 2022*
Published: *11 April 2022*

Citation:
Obradovic A and Nicholson J (2022)
Perspective: Implications of the
COVID-19 Pandemic for
Family-Focused Practice With Parents
With Mental Illness and Their Families.
Front. Psychiatry 13:806550.
doi: 10.3389/fpsyt.2022.806550

INTRODUCTION

Over the past few decades, the practice, lived experience, and research communities have engaged in ongoing discussion to specify core principles of family-focused practice aimed at improving outcomes for parents experiencing mental illness and/or substance use disorders and their children. Consensus has emerged across diverse cultural and systemic settings where stakeholders collaborated to facilitate local adaptation of evidence-based interventions (1–11).

Practice principles underpinning these initiatives include the importance of asking about an adult's parenting and family status, or a parent's mental health and well-being when children are being seen. Treatment and recovery are viewed as relational and multidirectional processes, with families providing the context for change. Relationships with parents and family members are built on trust and a non-judgmental, trauma-informed approach. Family roles, responsibilities, needs, and resources are assessed and considered in care planning and goal setting, leveraging the strengths of parents and children to address vulnerabilities. The coordination and integration of supports across diverse service sectors and natural networks are advised, in the context of a collaborative, therapeutic partnership approach. Psychoeducation about mental health benefits both parents and children (4, 5, 11–15).

Any changes, temporary or otherwise, in the biopsychosocial challenges families face and the factors that mediate risk, enhance resilience, and support personal and relational recovery require re-examination in light of the COVID-19 global pandemic (16). A contemporaneous understanding of the pivotal elements of socio-cultural and service system contexts is paramount to ensuring that future practice, policy, and research in this field remain evidence-based, and that lessons learned during and due to the pandemic are incorporated into our efforts.

In this perspective piece, we lay out a range of possible impacts of the COVID-19 pandemic on the core elements of family-focused practice. We borrow the EASE Framework (4), a theory- and evidence-informed practice approach to relational recovery for parents with mental illness, to organize our comments for this perspective. Our discussions with mothers with mental and substance use disorders and practitioners over the past 2 years further inform our consideration of the ways in which the pandemic has challenged family-focused practice.

As experienced clinicians and researchers in two different countries, cultures, and service contexts, we recognize that our perspectives are shaped by our own lived experience of the pandemic and the lives, experiences, and stories of those around us. Our goal is not to provide recommendations based on research findings but, rather, to suggest challenges and discuss implications for family-focused practice, policy, and research that will benefit from rigorous study in the future, as we sift through lessons learned to endorse positive consequences of the pandemic and move past negative impacts.

THE PANDEMIC HAS CHALLENGED FAMILY-FOCUSED PRACTICE

The impact of the pandemic on the mental health and well-being of people around the world has been documented (17–20). Common adaptations in service delivery have included a shift to telehealth and digital tools (21). The pandemic has posed challenges to practice across the EASE Framework components: Engage, Assess, Support and Educate (4). Challenges have differed somewhat, depending on family circumstances and whether parents and families are newly referred or have existing relationships with practitioners (16).

Engage

Engagement is the process of building a relationship with a parent and relevant family members, establishing rapport and trust to promote further collaboration (4). The pandemic has disrupted traditional, in-person modes of contact for practitioners with parents and families in many ways. Emerging, increasingly routine solutions (e.g., video conferencing) have necessitated attention to building and strengthening relationships within new arrangements. Cognizant of the limitations of virtual contact, practitioners have had to adapt or seek new ways to build trust, develop rapport, and maintain the safe, collaborative therapeutic partnerships essential to family-focused practice (22).

Safety imperatives have curtailed face-to-face contact by suspending home visiting or changed how this might occur due to the need for PPE protection, physical distance and, where possible, outdoor exchange. The focus of all parties (i.e., parents, families, practitioners, and programs) on minimizing the threat of infection have fundamentally altered the ways and locations in which parents, families and practitioners connect. Staying mindful of family needs and promoting connection likely have been challenging within some settings (e.g., hospital emergency departments or adult psychiatry inpatient units). Fundamental physical safety priorities have curtailed family visiting and possibly contributed to the breakdown of inter-team care coordination. During a crisis of this proportion, staff may have been redeployed away from their usual duties, potentially disrupting established therapeutic relationships.

While digital technology enables virtual connection as an alternative to face-to-face contact, these modes are not without disparity in their availability, cost, quality, capacity, and user proficiency. They also largely create a de facto incursion into the home, with potential privacy issues, complicating engagement in a way that office-based visits, if they were the norm, do not. The presence of children and other household members who may not be the focus of the session may affect levels of comfort, openness to sharing, concentration, and focus. Where sensitive matters such as abusive relationships, substance use or issues undisclosed to others in the household exist, virtual home-based sessions can be problematic. Practitioners may be exposing their own households to the parents they work with in virtual sessions, in ways that may inhibit or, alternatively, promote discussions of children and family life.

Assess

Assessment involves asking key questions in the context of the parent-practitioner relationship (4). The pandemic may have mixed impact on assessment of parents and family members, particularly in new relationships. If assessments are done virtually, practitioners may be concerned about missing the cues or signals that inform clinical judgement. Peer recovery coaches, in our community engagement discussions, shared their concerns about possibly missing signs of substance use relapse that typically informed their "hunches" about how parents were doing.

Virtual sessions may provide glimpses into the parent's home and family life, previously unavailable. Parents may face the disclosure of information—as seen by the practitioner or overheard by the child or partner—they might not have provided in an initial assessment session. Parents may say more than they might have typically offered in the past, as being stressed and feeling frustrated, unhappy, or anxious have become commonplace in the COVID-19 context. Articles in the popular press highlight "parenting burnout" and underscore the stress accompanying remote learning for children due to lockdown. The pandemic may have, in some ways, given parents permission to openly discuss their challenges and ask for help, in a less judgmental context and with fewer negative consequences.

In many ways, family life has changed significantly. Routine assessment probes such as the request to "describe a typical day" have new meaning when days are no longer "typical" and a "new normal" is emerging over time. Life may have slowed down or become more intense, as obligations and commitments outside the home have been limited by travel restrictions and family members are spending more time together. When coping (e.g., with the threat of illness exposure or pandemic-induced isolation) is perceived as challenging for everyone, practitioners' assessment of risk or the identification of family members' strengths may be altered.

Parents' resilience and coping strategies may be enhanced by opportunities for practice or overwhelmed by stresses conveyed by the pandemic. The "mom-wine" culture touted in social media may well have influenced alcohol consumption over the past months, as parents cope with unprecedented pressures through increased substance use (23). Clearly, a non-judgmental, accepting stance on the part of the practitioner is warranted, with appropriate questions posed to clarify any assumptions made during an assessment.

Support

Support includes helping parents take steps toward realistic goals, to achieve their vision for the family (4). The provision of instrumental or emotional support directly, or via the sharing of information and connections or referrals to resources have likely been significantly impacted by the pandemic. Exploring immediate unmet needs has been complicated both by the awareness that fewer solutions are available, and that methods of access to them have been hampered. On the other hand, a parent may be more likely to identify needs that have become more readily apparent due to the pandemic, as people's awareness and acceptance of challenges in day-to-day living have become the norm.

Developing and bolstering natural and professional support networks for parents, their children, partners, and other family members—a key activity in family-focused practice—may well be a major challenge. The ability to establish crisis and family care plans may be undermined by the lack of access to the established safety net of the extended family, respite resources, and school or childcare programs. In some cases, the pandemic may have led to a permanent loss of kinship care options, particularly affecting proactive parent-led prevention of relapse, or planning for hospitalization when alternative care options are vital. A fear that hospitalization may expose an individual and, therefore, family to infection may have led to avoidance of help seeking or planned admission, undermining a parent's recovery. In addition, demands on hospitals to focus on the acute needs of COVID-19 patients, has made in-patient care harder to access for those with non-COVID-19 related concerns.

The safety valve and empowerment provided by peer support and group programs for parents and children may have also been diminished through cancellation, postponement, or transfer to online formats. Online platforms have allowed some continuity of support, but the effectiveness of online approaches may vary depending on the resources and technical capabilities of users and services. The capacity to refer to additional professional help for individuals or whole families (e.g., counseling, infant and maternal mental health, family therapy) may have been limited by demands on the workforce to serve a much larger proportion of the population who have been affected by the pandemic.

Educate

The provision of evidence-based psychoeducation to parents and children is a key tenant of family-focused practice (4). The pandemic has resulted in an "info-demic" as parents and families are bombarded by health information, constantly changing as new research findings emerge. The pandemic has placed mental health center stage, with more information available in the popular press than ever before (e.g., suggestions for recognizing depression, coping with anxiety, etc.). Health literacy skills have become increasingly important, as parents and families make decisions about whether and when to seek help for emotional and behavioral problems, along with pandemic-specific decisions about social distancing, mask wearing and vaccination. Parents may require or request assistance in sifting through available information.

Psychoeducation about coping with loss and grief has become increasingly important. Families may have lost family members to illness and death; lost regular contact with extended family and friends; and lost the normal routines that comprised pre-pandemic family life. Continuous proximity of family members without sufficient relief during lockdowns has for many, intensified relationships within the home and resulted in emotional dysregulation, relational strain, and interpersonal hostility (16). The practitioner may be faced with the challenge of teasing apart the impact of the pandemic and trauma conveyed from normal development issues, though the parent and family's needs and the professional's response may be similar in either case. The frame placed on the issues is critical in supporting parents and families in drawing upon their strengths, rather than succumbing to their vulnerabilities.

DISCUSSION: FUTURE IMPLICATIONS

The challenges to family-focused practice during this period of pandemic crisis require us to consider the future implications for families, communities, and the workforce. This Discussion highlights opportunities to strengthen core principles, to identify potential undermining of advances made to date, and to refresh the practice and research agendas for the future.

Families and Communities

As time passes, more detailed evidence of the impact of the COVID-19 pandemic is emerging, confirming expectations that the incidence of mental ill health has indeed increased, with females and younger age groups particularly affected by depressive and anxiety disorders (24). The social determinants of mental ill health are clearly reflected in the social and economic consequences of the pandemic and, in turn, to an exacerbation of pre-COVID mental illnesses. This is of particular concern in families where parents had prior mental or substance use disorders and find their ability to manage their illnesses and cope with the demands of childrearing overwhelmed by pandemic-related stresses.

However, while generating significant demand for mental health services, this increase in higher prevalence psychiatric disorders (e.g., depression, anxiety) and greater awareness of the multidimensional, interdependent nature of mental distress and adversity across the lifespan, also create opportunities to:

- lessen the stigma, shame and secrecy that often accompany mental ill health or substance use;
- normalize open discussion about mental health or substance use concerns and, therefore, encourage help-seeking behavior;
- mobilize previously untapped social support networks (e.g., community and neighborhood opportunities), particularly for children and adolescents (25);
- increase recognition of the need for co-design in planning sustainable local responses (26, 27);
- gain public and governmental support for addressing mental health and substance use challenges, overcoming policy-related barriers to services and increasing funding for prevention, early intervention, and treatment.

What the post-pandemic context holds for those affected by lower prevalence mental illnesses typically considered more serious (e.g., schizophrenia, bipolar disorder), either pre-existing or newly onset, is less clear. Given the historical failure to implement and scale up adequate responses to those living with mental ill health despite compelling evidence of effective interventions, it may be the case that this heightened public awareness will steer discourse, service orientation, and publicly funded resources and research away from serious and enduring mental illnesses to the more commonly experienced situational, adjustment, and higher prevalence psychiatric disorders.

Workforce

One of the implications of this much broader experience of adversity and crisis is the effect on practitioners (28, 29). Many family-focused practitioners have undoubtedly encountered significant stress in their personal and professional lives, which may have generated reflection and reappraisal. Career reassessment is occurring in health care fields and in the workforce in general (30). Future inquiries should examine the extent of pandemic-related secondary impacts on family-focused practitioners of trauma, helplessness, compassion fatigue, and work-related moral injury (i.e., the profound psychological distress that comes from actions or the lack of, which violate one's moral or ethical code) (31).

If the emotional reserves of professionals have been tested, how has that impacted therapeutic relationships? Are practitioners more attuned to their parent-clients and the needs of the children in these families (32)? In training and supervision, practitioners may be able to draw from their personal exposure to adversity to consider their perception of risk and their capacity to maintain a fundamentally non-judgmental, strengths-based approach in their work. With the significant community effort and experience in collective caring and interpersonal sharing during this crisis, we also have an opportunity to elevate the utility of both peer support and recovery approaches across health and social care sectors, given their natural alignment to pandemic responses and healing.

If we accept that reliance on digital platforms has been embraced as an acceptable practice adaptation, this could lead to framing Internet access as a basic right, requiring future mental health service delivery to maintain a flexible menu of virtual and face-to-face options tailored to individual need, preference, and suitability. This could include:

- peer group programs that consist of a hybrid combination of face-to-face and online sessions;
- home visiting prioritized for individuals and families without reliable digital access;
- face-to-face interaction prioritized where virtual contact interferes with micro-communication; and
- increase in the available workforce by including those who cannot undertake a full time, site based or home visiting role due to other commitments or restrictions (e.g., digital service delivery by those home-bound).

It would be wise for the practice community to prepare actively to advocate against the repetition of past experience. Competing priorities in any service reconfiguration because of the pandemic may mean the needs of parents with mental illness and their families go unrecognized or untreated, as has occurred historically when resources are scarce.

Research

Our pandemic experiences have implications for research measures, methods, and outcomes. Treatment targets and timeframes may have shifted, given changes in modes of engagement, contextual stresses, and coping strategies. The validity and reliability of research measures routinely employed to assess variables and outcomes may need to be re-evaluated, given the dramatic changes in people's lives and the contexts in which they live. For example, traditional measures of social networks, social support, social functioning, self-efficacy, family functioning and family relationships may require careful examination, given the likely impact of the pandemic at a population level on each of these. Intervention fidelity measures, possibly based on frequency or type of visits, may need to be adjusted to take virtual encounters into consideration.

The pandemic has likely had positive and negative impacts on research practices. The shift to virtual encounters has, in some ways, made research interviewing more streamlined and expeditious by eliminating the need for travel. On the other hand, issues of privacy and confidentiality have been raised, as both the participant and the researcher are more exposed on the video screen. The role and responsibilities of institutional review boards have become more complex, as issues of data collection, management, and security have been challenged by requirements for limited in-person contact and social distancing. Research with parents and families may have been limited during this time to those with access to the Internet and necessary equipment. Alternatively, the shift to virtual data collection may have resulted in the ability to access target populations previously untapped or more diverse than in-person data collection permitted.

CONCLUSION

In conclusion, we acknowledge that our perspective is based on our experiences as clinicians and researchers in two well-resourced, first world countries. Our goal is to bring attention to both the challenges and the opportunities the pandemic brings to family-focused practice, and the parents, children, and families with whom we partner. As we move forward, it will be important to acknowledge the remaining uncertainty about the future and accommodate to the profound changes in family life, professional practice, and research related to the pandemic, as we demonstrate the effectiveness of positive lessons learned while developing new approaches for dealing with negative consequences.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary materials; further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

AO and JN were jointly responsible for the development and preparation of the manuscript. All authors contributed to the manuscript, read, and approved the submitted version.

FUNDING

Community engagement activities serving as a source for this article were funded through two Patient-Centered Outcomes Research Institute® (PCORI®) Awards: #EAIN-00147 and #8285-BU.

ACKNOWLEDGMENTS

We would like to thank our clinical and research collaborators and the families with whom we have partnered, who have helped us develop our perspective, particularly during the global pandemic.

REFERENCES

1. Blanch AK, Nicholson J, Purcell J. Parents with severe mental illness and their children: the need for human services integration. *J Ment Health Adm.* (1994) 21:388–96.
2. Dearden C, Becker S. *Young Carers in the U.K.: The 2004 Report*. London: Carers UK. Young Carers Research Group (2004).
3. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child Youth Serv.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
4. Foster K, Goodyear M, Grant A, Weimand B, Nicholson J. Family-focused practice with EASE: a practice framework for strengthening recovery when mental health consumers are parents. *Int J Ment Health Nurs.* (2019) 28:351–60. doi: 10.1111/inm.12535
5. Goodyear M, Hill T-L, Allchin B, McCormick F, Hine R, Cuff R, et al. Standards of practice for the adult mental health workforce: meeting the needs of families where a parent has a mental illness. *Int J Ment Health Nurs.* (2015) 24:169–80. doi: 10.1111/inm.12120
6. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Adv Ment Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
7. Human Rights and Equal Opportunity Commission. Children of parents with mental illness. Chapter 16. In: *Report of the National Inquiry Into the Human Rights of People With Mental Illness* (1993).
8. Marston N, Reupert A, Mosek A, Stavnes KA, Drost LM, Solantaus T, et al. A content analysis of intervention key elements and assessments (IKEA): what's in the black box in the interventions directed to families where a parent has a mental illness? *Child Youth Serv.* (2016) 37:112–28. doi: 10.1080/0145935X.2016.1104041
9. Mulligan C, Furlong M, McGilloway S. Promoting and implementing family-focused interventions for families with parental mental illness: scoping and installation. *Adv Ment Health.* (2020) 18:202–16. doi: 10.1080/18387357.2019.1614466
10. Reupert A, Price-Robertson R, Maybery D. Parenting as a focus of recovery: a systematic review of current practice. *Psychiatr Rehabil J.* (2017) 40:361–70. doi: 10.1037/prj0000240
11. Reupert A, Drost LM, Falkov A, Gatsou L, Gladstone B, Goodyear M, et al. Editorial perspective: Prato Research Collaborative for change in parent and child mental health - principles and recommendations for working wiht children and parents living with parental mental illness. *J Child Psychol Psychiatry*. (2021) 63:350–53. doi: 10.1111/jcpp.13521
12. Nicholson J, Wolf T, Wilder C, Biebel K. *Creating Options for Family Recovery: A Provider's Guide to Promoting Parental Mental Health*. Marlborough, MA: Employment Options, Inc. (2014).
13. Nicholson J, English K. *ParentingWell Practice Profile*. Waltham, MA: National Research Center for Parents with Disabilities, Brandeis University and the Massachusetts Department of Mental Health (2019). Available online at: https://heller.brandeis.edu/parents-with-disabilities/parenting-well/index.html
14. Price-Robertson R, Olsen G, Francis H, Obradovic A, Morgan B. *Supporting Recovery in Families Affected by Parental Mental Illness (CFCA Practitioner Resource)*. Melbourne, VIC: Australian Institute of Family Studies, Child Family Community Australia Information Exchcange (2016).
15. Price-Robertson R, Obradovic A, Morgan B. Relational recovery: beyond individualism in the recovery approach. *Adv Ment Health.* (2017) 15:108–20. doi: 10.1080/18387357.2016.1243014
16. Furlong M, McGilloway S, Mulligan C, Killion MG, McGarr S, Grant A, et al. Covid-19 and families with parental mental illness: crisis and opportunity. *Front Psychiatry*. (2021) 12:567447. doi: 10.3389/fpsyt.2021.567447
17. Boden M, Zimmerman L, Azevedo KJ, Ruzek JI, Gala S, Abdel Magid HS, et al. Addressing the mental health impact of COVID-19 through population health. *Clin Psychol Rev.* (2021) 85:102006. doi: 10.1016/j.cpr.2021.102006
18. Gavin B, Lyne J, McNicholas F. Mental health and the COVID-19 pandemic. *Ir J Psychol Med.* (2020) 37:156–8. doi: 10.1017/ipm.2020.72
19. Khan KS, Mamun MA, Griffiths MD, Ullah I. The mental health impact of the COVID-19 pandemic across different cohorts. *Int J Ment Health Addiction*. (2022) 20:380–6. doi: 10.1007/s11469-020-00367-0

20. Kumar A, Rajasekharan N. COVID-19 and its mental health consequences. *J Ment Health*. (2021) 30:1–2. doi: 10.1080/09638237.2020.1757052
21. Inkster B, O'Brien R, Selby E, Smriti J, Subramanian V, Kadaba M, et al. Digital health management during and beyond the COVID-19 pandemic: opportunities, barriers, and recommendations. *JMIR Mental Health*. (2020) 7:e19246. doi: 10.2196/19246
22. Leggatt M. Families and mental health workers: the need for partnership. *World Psychiatry*. (2002) 1:52–4.
23. Adams RS, Ledingham E, Keyes KM. Have we overlooked the influence of "Wine-Mom" culture on alcohol consumption among mothers? *Addict Behav*. (2021) 124:107119. doi: 10.1016/j.addbeh.2021.107119
24. COVID-19 Mental Disorders Collaborators. Global prevalence and burden of depressive and anxiety disorders in 204 countries and territories in 2020 due to the COVID-19 pandemic. *Lancet*. (2021) 398:1700–12. doi: 10.1016/S0140-6736(21)02143-7
25. Bauer A, Stevens M, Purtscheller D, Knapp M, Fonagy P, Evans-Lacko S, et al. Mobilising social support to improve mental health for children and adolescents: a systematic review using principles of realist synthesis. *PLoS ONE*. (2021) 16:e0251750. doi: 10.1371/journal.pone.0251750
26. Nicholson J. Risks linked with parental mental illness and substance use are undeniable, but efforst to scale-out and sustain evidence-based practices are challenging [Internet]. *Frontiers in Psychiatry Featured News*. (2021). Available online at: https://blog.frontiersin.org/2021/10/13/joanne-nicholson-parental-mental-illness-substance-use/ (accessed March 28, 2022).
27. Zechmeister-Koss I, Goodyear M, Tüchler H, Paul JL. Supporting children who have a parent with a mental illness in Tyrol: a situational analysis for informing co-development and implementation of practice changes. *BMC Health Serv Res*. (2020) 20:326. doi: 10.21203/rs.2.15578/v1
28. Muller AE, Hafstad EV, Himmels JPW, Smedslund G, Flottorp S, Stensland SO, et al. The mental health impact of the covid-19 pandemic on healthcare workers, and interventions to help them: a rapid systematic review. *Psychiatry Res*. (2020) 293:113441. doi: 10.1016/j.psychres.2020.113441
29. Rauch SAM, Simon NM, Rothbaum BO. Rising tide: responding to the mental health impact of the COVID-19 pandemic. *Depress Anxiety*. (2020) 37:505–9. doi: 10.1002/da.23058
30. Cook I. Who is driving the great resignation? *Harvard Business Review*. (2021). Available online at: https://hbr.org/2021/09/who-is-driving-the-great-resignation (accessed March 28, 2022).
31. Williamson V, Murphy D, Greenberg N. COVID-19 and experiences of moral injury in front-line key workers. *Occup Med*. (2020) 70:317–9. doi: 10.1093/occmed/kqaa052
32. Goodyear M, Allchin B, Burn M, von Doussa H, Reupert A, Tchernagovski P, et al. Promoting self-determination in parents with mental illness in adult mental health settings. *J Fam Nurs*. (2022) 1–13. doi: 10.1177/10748407211067308

Author Disclaimer: The views and statements presented in this article are solely the responsibility of the authors and do not necessarily represent the views of the Patient-Centered Outcomes Research Institute® (PCORI®), its Board of Governors or Methodology Committee.

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 14 April 2022
doi: 10.3389/fpsyt.2022.815873

An Evaluation of the Acceptability, Appropriateness, and Utility of a Bibliotherapy for Children of Parents With a Mental Illness

Kelly Vetri [1,2,3,4]*, *Geneviève Piché* [1,2,3,4] *and Aude Villatte* [1,2,3,4]

[1] Laboratory LaPProche, Department of Psychoeducation and Psychology, Université du Québec en Outaouais, Saint-Jérôme, QC, Canada, [2] Centre de recherche universitaire sur les jeunes et les familles (CRUJEF), Québec, QC, Canada, [3] Réseau de recherche en santé des populations du Québec (RRSPQ), Montréal, QC, Canada, [4] Réseau Intersectoriel de Recherche en Santé de l'Université du Québec (RISUQ), Québec, QC, Canada

Background: Children of parents with a mental illness are at higher risk for various psychiatric problems and adaptive difficulties compared to those of parents without mental health problems. Certain preventive psychoeducational interventions target these children to promote their well-being and resilience and prevent the emergence of adaptive difficulties. However, few such interventions have been developed and evaluated specifically for elementary school-aged children of parents with a mental illness.

Objectives: This study aims to evaluate an interpersonal psychotherapy-based book targeting children living with a parent with a mental illness.

Methods: The study examines children, parents and psychosocial workers' perception of the acceptability, appropriateness and utility of the book. In total, 22 participants answered online open-ended questions after reading the book.

Results: The book was highly appreciated and positively perceived by the families and psychosocial workers. Results suggest that children, parents and psychosocial workers viewed it as an appropriate and useful tool for supporting children with a parent with a mental illness. The present study reveals that the bibliotherapy appears well adapted to the developmental level of school-age children.

Discussion: This study presents a book that shows promise for supporting the resilience of elementary school-aged children having a parent with a mental illness. Results highlight the importance of tailoring the content and modalities of interventions to the developmental level, needs and preferences of elementary school-aged children. The relevance of a collaborative method is also discussed, thus providing knowledge regarding this type of approach for the development of interventions targeting children.

Keywords: children, parental mental illness, bibliotherapy, psychoeducation, interpersonal psychotherapy, intervention

Edited by:
Jean Lillian Paul,
Medizinische Universität
Innsbruck, Austria

Reviewed by:
Kjersti Bergum Kristensen,
UiT the Arctic University of
Norway, Norway
Melinda Jane Goodyear,
Monash University, Australia

***Correspondence:**
Kelly Vetri
vetk01@uqo.ca

Specialty section:
This article was submitted to
Frontiers in Child and Adolescent
Psychiatry,
a section of the journal
Frontiers in Psychiatry

***Received:** 15 November 2021*
***Accepted:** 14 March 2022*
***Published:** 14 April 2022*

Citation:
Vetri K, Piché G and Villatte A (2022)
An Evaluation of the Acceptability,
Appropriateness, and Utility of a
Bibliotherapy for Children of Parents
With a Mental Illness.
Front. Psychiatry 13:815873.
doi: 10.3389/fpsyt.2022.815873

INTRODUCTION

Overall, about one in five children have a parent with a mental illness (1). The mental illness symptoms can have a significant impact on the parent and other family members, including children (2). Living with a parent with a mental illness may involve adverse family conditions (3, 4) characterized by high levels of conflict, poor family communication and low cohesion between family members (5, 6). Children of parents with a mental illness (COPMI) frequently report a poor parent-child relationship, few moments of shared family time, a tendency to be parentified (7) and, above all, developmental needs that often go unmet (1). Exposure to parental mental illness can also lead to several negative outcomes for a child. COPMI are at higher risk of a wide range of negative affects (e.g., fear, anxiety, sadness, guilt, shame, confusion), notably owing to the parent's symptoms, lack of information about the mental illness, and fear of developing a similar illness or being responsible for the parent's illness (8, 9). These children may also experience more isolation and loneliness than others (10). The stigma of their parent's mental illness may make them reluctant to invite friends over, talk about the mental illness, or ask for help from those around them, thus reinforcing their feelings of distress (10–12). Moreover, COPMI are at higher risk of academic difficulties (8, 13, 14) and emotional and behavioral problems (15, 16). The scientific literature reveals, furthermore, that parental mental illness significantly increases a child's risk of developing psychopathology during childhood, adolescence and well into adulthood (17–22). This risk of intergenerational transmission seems present for two future generations (22). Furthermore, the likelihood of these children developing a mental illness at a younger age is higher than that observed in children in the population at large (23, 24).

Despite these important risks, it should be noted that children exposed to parental mental illness do not all develop in the same manner, as is underlined by the multifinality concept. While some face significant psychosocial difficulties, others demonstrate resilience in the face of family adversity and avoid developing psychosocial problems (1, 25–28). Interestingly, according to a literature review by Drost et al. (25), several COPMI even report feeling "*more mature, independent, and empathic than their peers who did not have a parent with a mental illness*". These resilient children show that some aspects of their environment and family, including positive and strong social support or the presence of a parent with positive mental health, helped them maintain good mental health. At the same time, the literature indicates that interventions to promote the mental health, well-being and resilience of youth with a mentally ill parent should include several components underscoring protective psychosocial factors. Protective factors are: seeking and requesting social support; acquiring knowledge about the parent's mental illness; communication and problem-solving skills; self-esteem and social competence skills (e.g., recognizing and sharing feelings and needs); and capacity to replace negative cognitions (12, 29, 30). Coping skills are particularly important in a family context where the manifestations of parental mental illness can lead parents to be unable to manage daily problems and conflicts.

Generally speaking, results of several studies indicate that the preventive interventions designed for this at-risk clientele, including the targets mentioned above, are beneficial and have positive effects on children's mental health and overall well-being (9, 31–33). They are recommended as early as elementary school, in a prevention perspective (1, 5, 30, 34). As examples, the *Play and Talk* group program (21), targets 8-to-12-year-old children with a mentally ill parent and offers eight weekly 90 min group meetings and a family meeting. The intervention program shows beneficial effects in participating families. Indeed, in a study conducted with 254 families, a decrease in negative thoughts and emotional and behavioral problems was observed in young participants to this program compared to the control group (35). Another example of an intervention with beneficial effects is the *Family Focus DVD*, designed for families with a parent having a depressive or anxiety disorder and elementary school-aged children. An increase in mental health literacy and knowledge of strategies for talking about mental health problems was reported by the 29 young participants (36). More generally, a meta-analysis highlighted that COPMI have a 40% lower risk of developing psychopathology when they have access to effective preventive services (16). Therefore, it's essential to support COPMI and their families with effective and easily accessible interventions to help them cope with the various challenges they face (37). The challenges they face are exacerbated by the current epidemiological situation and the difficulty of the mental health services to respond quickly and in an adapted way to their important needs (38–40).

However, even if the effects of interventions are positive and seem to persist and even increase over time, they tend to be small and are therefore unlikely to be sufficient to effectively reduce these children's risk factors or lighten their burden (35, 41–43). Some researchers raise the hypothesis that the content and modalities of certain preventive programs and tools for elementary school children may not be adequately tailored to their needs, characteristics or developmental level (35, 41, 42). In fact, very few published studies present information on the satisfaction or perceived usefulness of their content or intervention modalities by participants or on how the program content is integrated into the children's and family's life (44, 45). Besides, few studies have consulted elementary school-aged children to determine their needs and preferences when designing or evaluating interventions for them (44). Hence, almost no information is available on how COPMI perceive and accept the tools and programs proposed to them or, for example, if they understand the terms, strategies or activities included (42, 44–46). In general, the effects of preventive interventions are usually evaluated without considering their implementation (47). The adequacy between the characteristics of the clientele, the intervention protocols and the service delivery can ensure successful implementation and utilization of available services among families (48–51). When a psychoeducational program or intervention tool is offered for school-aged children, the proposed content must be relevant and adapted to their developmental level, notably their ability to understand complex concepts, solve interpersonal problems, focus during long periods of time, and recognize and share their emotions. Garber et al. (42) suggest these factors may modulate the implementation and effect sizes of psychoeducational programs or interventions targeting children.

Some strategies could improve children's acceptability and understanding of the content of an intervention. The recommendation is to use simple language and clear and short sentences when giving instructions, and to provide tangible and visual materials, pictograms, playful games, concrete examples and practical experiences (42, 52). Bibliotherapy appears to be among the most promising ways to promote the understanding of psychoeducational content and its adaptation to children's developmental level (52–54). The term *bibliotherapy* refers to the use of written materials (e.g., storybooks) as therapeutic tools for preventing or treating a variety of specific issues related to a child's needs (54, 55). Empirical evidence underlines the positive effects of bibliotherapy on individuals with mental health problems and on mental health and well-being in general (55, 56). Also, although the evidence is scarce, some evidence tends to show that bibliotherapy could be beneficial for COPMI (57–59). In fact, some interventions for COPMI incorporate psychoeducation and informative materials in written form (e.g., pamphlets, books, symbolic stories), which supports the relevance of this type of medium for these children (30, 33, 60).

In Quebec, Canada, a book titled *Le Trésor de l'Île Rouge* (see https://lapproche.uqo.ca/le-tresor-de-lile-rouge/) was developed initially as part of a preventive intervention program for families with a depressed parent adapted from *Family Talk* (61), *FAMILLE+* (62, 63). The aim of the book is to help elementary-school COPMI learn coping strategies to deal with the challenges and difficult situations of everyday life. The book can be read by the child alone or facilitated by a professional, parent or other adult. The story describes an amusing pirate adventure where children are introduced to the main characters, their adventures and the strategies they use to overcome the obstacles and challenges they encounter. It uses different learning modalities and a series of activities offering young readers various opportunities to learn while having fun. Some of these are free to download and use on our website (e.g., Emotional-Pirate tools). The book's content is based on intervention targets acknowledged to be most relevant to COPMI as presented above (e.g., (19, 29, 64). It is also based on the Interpersonal Psychotherapy (IPT) approach, a commonly used and evidence-based preventive intervention treatment for depression in adolescents (65, 66) and pre-adolescents (8–12 years old; (67)) and a number of other disorders including anxiety (68). Several studies show its effectiveness for improving well-being and social functioning in young people (66). IPT focuses on improving interpersonal relationships, reducing symptoms of mental illness and improving well-being. This goal is achieved by improving communication and relational problem-solving skills and promoting positive interpersonal behaviors with significant others (e.g., parents, peers). Similarly, our objective via the IPT-based book is to help elementary school-aged COPMI learn concrete socio-emotional regulation skills, effective communication and interpersonal problem-solving skills, strategies for reducing conflict and ways to promote their social support network. In the long term, this may enable children dealing with parental mental illness to better cope with their parents and family. It may also help promote a positive parent-child relationship, children's well-being and, ultimately, their mental health (see **Table 1**).

It should be noted that the book addresses the main problems encountered by COPMI without addressing the specific issue of parental mental illness. Communication difficulties, impaired family functioning and a tendency to parentification are examples of themes highlighted in the characters' stories. For example, the main character, the ship's captain, feels unwell and can no longer meet the needs of his shipmates. This passage conjures the mental illness of a parent who has difficulty responding adequately to their child's needs. The captain also experiences many negative emotions (e.g., shame or guilt) and hides in his room rather than inform the crew of his situation. The captain is concerned about the possible reaction of the ship's company to his inability to function as usual, a situation recalling the stigma and shame associated with parental mental illness in families (10, 69).

Two previous pilot studies assessed the evaluation of the first version of the book and highlighted suggestions for improvement (70). Some changes have been made to enhance its acceptability for children ages 7 to 11. This study evaluates the second version of this book. The present study aims to evaluate the IPT-based book *Le Trésor de l'Île Rouge* targeting elementary school-aged children with a parent with a mental illness.

METHODOLOGY

Research Design

The formative evaluation examined children, parents and psychosocial workers perception of the acceptability, appropriateness and utility of the book. Qualitative data were collected by asking participants to respond to online open-ended questions after reading the book. Data triangulation between the perspectives gathered from children, parents and psychosocial workers was performed as a cross-validation modality to increase the validity and confidence level of research results (71). Involving children themselves in the assessment process is advocated as it provides more valid and accurate data based on their perspectives and experiences, which are often different from their parents or psychosocial workers (72–77).

Recruitment and Procedures

Between August 2020 and September 2021, participants were recruited from two community organizations (*La Boussole* and *L'Apogée*) that provide help and support services for the relatives of individuals with a mental illness. The organisations' management promoted the research to families using their services and to psychosocial workers. To be eligible, families had to have a child aged 7 to 11 years old and a parent with a mental illness. For psychosocial workers, the single inclusion criterion was to work with families with parental mental illness. Participants had 3 weeks to read the book and complete the online confidential formative evaluation questionnaire.

TABLE 1 | Strategies and activities of the book.

Chapter Title	Description and activities	Psychoeducational goals and strategies
Captain Philou and his new crew	Captain Philou puts together his new crew to find the Red Island Treasure and the crew members use all their strengths to battle enemy pirates.	Use your strengths (personal and family).
The storm rages on	The crew must overcome a terrible storm, which makes the pirates experience a lot of emotions and jeopardizes the operation of the ship.	Share your emotions with others, otherwise they will grow inside you.
The island of a thousand words	Poor communication between the crew members causes the boat to crash on an island.	Use your communication skills. Work together to overcome challenges.
The rotten fish	The captain is not feeling well. A pirate must take on his responsibilities. He has too many tasks and the ship is attacked by sharks.	Ask for help when you need it. Recognise and talk about your limits and needs.
The red ants	The pirates arrive on the Red Island. Excitement is running high, and pirates experience all kinds of emotions. They make a plan to overcome the obstacles they will encounter on their way.	Breathe gently through your belly to calm yourself. Use the problem-solving technique.
A celebration	The crew hikes over the Red Island and faces multiple dangers. They find the brother of a teammate and the treasure. A banquet is held to celebrate.	Emphasize the importance of doing fun activities as a family.
Captain's questions	Reading comprehension questions: 5 multiple choice questions related to the story and strategies taught.	Validation and learning integration of coping strategies.
Pirate-Active	Body percussion activities: Pirate dances and related reflection questions.	Get the children moving and make connections to the theoretical content.
Emotional-Pirate	Feelings tools: Mood thermometer and emotion display to evaluate feelings.	Encourage identification of emotions and generate discussion on the subject.
Tips from Diane La Sage	Emotional regulation skills: 3 tips for sharing and calming your emotions. Space to write down ideas.	Encourage sharing of emotions and healthy strategies to soothe emotions.
Quiz: Which pirate are you most like?	Identifying personal strengths: Quiz with questions and captions to determine which pirate child resembles.	Explore and reflect on personal strengths.
Special recipes from Jeanne La Borgne	Parent-child activities: 4 recipes related to the story about the pirates.	Promote quality time between the child and a significant adult.
My pirate team	Social support activity: 1 proximity circle exercise and 1 fun pirate drawing activity.	Identify people who can meet the child's needs.
The pirate apprentices' corner	Self-esteem drawing activity: Invitation to draw the crew's adventures with the chance to post the drawing on the book's website.	Promote children's self-esteem and their integration of information and reinforce the dynamic aspect of the book.

Although parents could help their child technically (e.g., write down the answers or rephrase the questions), they were told it was important for children to give their own answers when completing the questionnaire. A research assistant offered to assist children and parents as needed, but this help was not requested. Participating families were given a financial compensation of $25 in the form of a voucher.

Sample

In total, 22 participants completed the online questionnaire, including eight parents (five mothers and three fathers), eight children (four girls and four boys) and six psychosocial workers. The children's explicit consent was obtained as part of their participation. Three children were 7–8 years old; two children are 9–10 years old, and three children were eleven years old ($M = 9$ years). The participating parents were aged between 36 and 45 years old ($M = 40$ years). Four families were biparental, two were blended or uniparental. Two children had an attention deficit hyperactivity disorder and were being treated by a child psychiatrist. All participating children were receiving specific support services from a community organization for the family members of a person with a mental illness. Four parents had a diagnosis—mainly of mood or anxiety disorders—and four were the partner of a parent with a diagnosis. All four parents with a diagnosis had a comorbid mental illness with one or more co-occurring illnesses. Five parents had a university degree, two had a college degree and one parent reported having a high school diploma. All six psychosocial workers were woman. Four of them were under 35 years and two were between 40 to 45 years old. Five had a university degree and one indicated having a college degree. The majority had one to 2 years of experience in their workplace.

Formative Evaluation Questionnaire

To gather information concerning the book's acceptability, appropriateness and utility for children aged 7 to 11 years old with parent with a mental illness, participating children, parents, and psychosocial workers were asked open-ended questions about their overall appreciation of the book and its features. Examples of questions for children are the following: "Did you enjoy the book? Why?"; "What did you like most and least?";

"What strategy did you find most useful? Why?". Most of the questions asked to the parents and psychosocial workers were identical. For examples: "Did you enjoy the book?"; "Do you think this book was helpful and useful?"; "Would you recommend this story to families with school-aged children?". Psychosocial workers were asked one additional specific question: "Would you like to use this tool in your practice with children aged 7–11 and their families?". Parents were also questioned about their perception of their child's reading experience, with one question: "In your opinion, did your child like the book?".

Then, four specific open-ended questions were asked to psychosocial workers concerning the content's readability and appropriateness to the children's age and developmental level: "Do you think this book is adapted to the developmental capacities of the target clientele?"; "Do you think some aspects are difficult for children this age to understand?"; "Generally speaking, do you think the information is well explained?"; "In your opinion, is this book fun and enjoyable to read for children of this age group?." Similarly, parents were invited to indicate in a few lines whether they considered the book to be appropriate for children the same age as their own and to justify their answers.

Analysis Strategies

The qualitative data obtained have been transcribed and were processed using N'Vivo software (78). General categories were created to group the data according to the questions asked and the book's features. A triangulation of the data obtained from all respondents was also carried out (79). Excerpts of responses were selected for a concrete illustration of each category. Participants were assigned fictitious names. Recurrences or discrepancies in the responses of the various participants (parents, children, psychosocial workers) were highlighted.

RESULTS

Evaluation of the Book in General

All children, parents and psychosocial workers who participated in the study reported they liked the book. More specifically, all parents affirmed their children appreciated it, as testified by this father of a 7-year-old girl: "*My daughter really liked the book. When she started reading, she would take it with her in the car, even when we went to the grocery store. Which actually surprised me because she's hardly ever done that with other books. She practically finished reading the book before I did.*" Furthermore, participating parents reported that the book was a helpful and useful tool for their child and family. Among the most relevant elements underlined, they mentioned accessible content, problem-solving strategies and understanding of experienced emotions, as the following examples demonstrate:

> "*It makes them see that sometimes there are difficulties, but that it's always possible to overcome them together.*" (Anna, Mother of an 8-year-old girl)
> "*The children realised that with good strategies they could solve their problems easily.*" (Jade, Mother of an 8-year-old girl and an 11- year-old boy)
> "*In 2021, I think boys need to keep up with the times and this is a great way to show them that they can be close to their emotions even if they want to be strong all the time.*" (Layla, Mother of two boys aged 8 and 10)

The psychosocial workers all agreed the book was helpful and useful for 7- to 11-year-old children and their families. The two main reasons for the consensus were teaching strategies and use of the story as a therapeutic tool. In this regard, psychosocial workers reported that the book allowed children to access varied, concrete and simple tools. In their opinion, the proposed strategies are relevant to any child in the 7-to-11 age group and can be applied in a variety of situations to help them overcome difficulties. A few psychosocial workers and one parent also reported that using a story to address sensitive topics (e.g., difficulties or emotions) and teach positive coping strategies can improve children's attention, illustrate content and offer a playful setting for learning and expressing experiences. The following are some of their observations:

> "*This puts the strategies one must use to be a self-benevolent and emotionally attentive human being into a more amusing context.*" (Layla, Mother of two boys ages 8 and 10)
> "*I think this book has great potential for youth intervention. (. . .) I believe this book is helpful for any 7- to 11-year-old child because it offers strategies for communication, emotional regulation and self-management that may be relevant to them in the future. It also gives them the tools to remain calm in difficult situations. (...) The fact is that children can identify a situation in the book and receive a tool to solve it directly in the story. They can then apply it to themselves afterwards.*" (Florence, psychosocial worker)

Thus, all psychosocial workers agreed they would recommend the book to families with school-aged children and aimed to use it in their clinical practice with this clientele.

Evaluation of the Book's Features

Families and psychosocial workers were asked to comment on their appreciation of specific features (e.g., story, complementary activities, characters, illustrations) of the book. The following sub-sections describe the perspectives of participants regarding the book's features.

Story and Characters

In general, all the families and most of the psychosocial workers reported they appreciated the story. The participants considered it interesting, funny, captivating and imaginative. Jess (psychosocial worker) explained: "*There are many twists and turns, and the adventure style is appealing.*"

Parents especially appreciated the parallels between the situations in the story and those in family life, as the following excerpts show: "*Nice parallel to life in general. I liked that it shows both the good times and the not-so-good times since these are a part of family life.*" (Evelyne, Mother of 11-year-old girl); "*Somehow, I saw myself in the role of Captain Philou. The book uses fantasy to teach life values that I think I and my children need. In fact, I asked my 13-year-old son to read it too.*" (Donovan, Father of 7-year-old girl). This father also praised the way the six chapters combined theory and practice "*in a very simple and understandable way*".

Finally, one psychosocial worker seemed less appreciative of the story, which she deemed overly literal and redundant: "*I didn't feel there was a narrative, but rather a snapshot of each event (e.g., a challenge arises, you find solutions, you apply a solution, it works, you move on to the next challenge). It would have been interesting to have a larger narrative framework, which would create better flow in the story and smoother interactions between the characters*." (Valerie, psychosocial worker)

In another vein, many children, psychosocial workers and parents found the characters to be well described, diverse, colorful, original, and interesting. Mathieu, a father of two children 8 and 9 specified: *"The characters are endearing and relatable to children."* One social worker, on the other hand, commented that the characters were presented in an idealized fashion (e.g., the story failed to mention their bad habits, problems or faults). Another suggested that more connections could have been made between the reactions of the characters and mental health issues.

Illustrations, Visuals and Format

Children, parents and psychosocial workers reported liking the visuals, colors and illustrations of the book. These features are eye-catching and help make the book more dynamic. A psychosocial worker also noted that the color-highlighted tips and tools make it easy to identify the strategy used in the particular situation. Florence (psychosocial worker) explained: *"I think the book is super well done. The illustrations are very beautiful. (...) I really like the choice of colours."*

Most children, parents and psychosocial workers really liked the book's format. Psychosocial workers specifically liked the division of chapters and sections and the balance between text and drawings, which made the book easy and pleasant to read and allowed for reading one chapter per day. However, two felt it contained too much information (e.g., a lot of details on the instruction page) and was "*a little too long and busy*". As well, two said the activities should be presented after each chapter related to them, while another suggested the exercises be detachable so children could use them more readily.

Strategies Presented

When asked about the most valued features of the book, parents and psychosocial workers named the adaptive coping strategies, which were considered relevant, accessible to all and easy to understand.

> *"These are good strategies that are the basis of what we're taught to promote mental health."* (Evelyne, Mother of 11-year-old girl)
> *"I don't usually let children know how I feel about a difficult situation because I think I'm protecting them. But this book helped me realise how important it is to communicate with them and how to do this."* (Donovan, Father of a 7-year-old girl)

The taught strategies were also praised for being well illustrated, clearly explained for the 7- to 11-year-old age group and easily applicable in various situations. Because strategies were appropriate to a given context, the participants felt children could more easily identify the type of situation calling for the tools presented. Two psychosocial workers also commented on the diversity of the proposed learning methods (e.g., dance, drawings, sharing, games, etc.), which meant more children could be reached based on their learning styles and interests. Judith (psychosocial worker) specified: *"Excellent strategies, varied and accessible to all. They are well illustrated and explained for all ages."*. One psychosocial worker, on the other hand, felt that strategies should not be presented as instructions, since this interrupted the storyline and could cause children to lose interest: "*At times, I didn't feel like I was reading a children's story anymore, but a document explaining a strategy.*" (Valerie, psychosocial worker)

The majority of children stated they had used or were thinking of using a strategy from the book in the near future. Among the strategies they had used or were considering using in the near future, the breathing technique was named most often. Half the children mentioned communication techniques (e.g., talking about their emotions, using "*I*" instead of "*you*", looking at people when speaking). Also, Angelique (age 7) responded: "*Asking someone for help when you need it. I used this technique at school.*" Additionally, some parents intended to use, or reported having used, some of the strategies presented, as evidenced in the following:

> *"We've already experimented them, and it has helped us during conflicts.*" (Jade, Mother of an 8-year-old girl and an 11-year-old boy);
> *"They're great [strategies] and my kids are already using them."* (Mathieu, Father of a 9-year-old girl and an 8-year-old boy).

At the same time, parents and children identified the strategies they perceived as particularly useful and helpful. Some named self-regulation strategies (e.g., self-soothing or self-calming), others the belly-breathing technique or the tips for sharing feelings (e.g., the importance of and ways to facilitate communication about feelings) and promoting effective communication, including "*speaking in the first person*". The following comments reflect the strategies participants found helpful:

> *"Talking about how you feel because it helps a lot.*" (Lea, age 11)
> *"The way we communicate. We lived for several years with a dad who blamed us for his bad behaviour, so the kids see from the book that that this isn't a good way to express themselves.*" (Layla, Mother of two boys, ages 8 and 10)

For the father of a 7-year-old girl, the problem-solving technique was the most relevant: "*As long as we're alive, we'll have problems. I think that mastering this technique early on will better prepare children for life*."

Complementary Activities

All respondents described the activities as relevant, fun and interesting. Some parents and psychosocial workers emphasized that the activities improved the children's understanding of the story, allowed them to integrate the book's content, and made the story more interactive and dynamic.

> *"There were fun games. (..) I hope you write more books like this because it explains to people how to relax and gives them activities to do when they're bored."* (Rose-Anne, age 8)
> *"The games were fun and helped us get to know each other better, develop new strategies and laugh together."* (Jade, Mother of an 8-year-old girl and an 11-year-old boy)

Conversely, one father criticized the complementary activities as the least interesting aspect of the book: "*I liked the rest of the book less, even though it contains important information. I lost interest in reading after chapter 6. I found this part much more theoretical.*" Accordingly, he suggested the activities be separate from the book.

According to three psychosocial workers, Captain's reading comprehension questions are important because they allow for consolidating transmitted information, reinforcing what has been learned, and revisiting certain passages of the story with the children; as well, they make the reading experience more interactive and playful. These workers also recommend adding specific questions regarding the characters' reactions or general questions about problems experienced and ways to avoid certain situations or normalize the behaviors of persons with a mental illness instead of focusing solely on the tips and solutions in the questions asked. One worker, however, indicated this learning modality might not be of interest to all children, as it could prolong the reading.

In general, most child participants said they very much enjoyed the body percussion activities in the section *Pirate-Active*, and the psychosocial workers stated they were an interesting addition to the book. Only one child and one psychosocial worker reported they liked these activities least. Two workers said the activities allowed children to move and become aware of their physical sensations, making the book more dynamic. Only one mentioned she was not convinced of the activities' relevance or children's participation. Some psychosocial workers observed that the activities involved several persons (e.g., a few children together, a sibling), whereas a child might not have access to several persons willing to participate. Alternatives were therefore proposed to be added to the book, for example, a breathing or communication activity.

Furthermore, almost every child reported they enjoyed the pirates' emotional tools—including the pirate's mood thermometer—provided in this section. Only one said he liked them more or less. Parents, for their part, said they liked the way the book dealt with emotions and offered tips on attending to emotions and learning to regulate them. A father of a 11-year-old girl explained: *"I liked that it talks about emotions and gives tips on how to learn to work on those emotions."*

Most of the psychosocial workers found these tools interesting and relevant, notably, because they provided children with visual tools to recognize and identify their moods and emotions. Florence specified: *"I think this activity is highly relevant since it puts a visual image on how the child is feeling."* A few of them stated that they would have liked to see other features in this section (e.g., the breathing technique, a greater variety of emotions, questions allowing the child to draw parallels between the challenges experienced by the characters and their own life).

Overall, the majority of children reported they greatly enjoyed the tips and tools for sharing and regulating emotions presented in the section *Tips from Diane La Sage*, while psychosocial workers perceived this section as interesting, relevant and instructive. The reinvestment of the tools presented in the book and their connection with the child's experiential life were two more positives aspects mentioned. Sophie mentioned: *"It's good to have activities that refer to the child; it allows children to transfer elements of the story into their own life.".*

One psychosocial worker especially appreciated the first tip (*calm your emotional storm*) because it offered children various ways to regulate their emotions, allowing each to choose the solution best suited to a particular situation. However, one psychosocial worker suggested an activity be included motivating children to reflect on the presence of some of the characters' behaviors in people in their own lives (e.g., the captain's refusal to get out of bed or Louis Le Curieux's anxiety).

Besides, most child participants reported they greatly appreciated the quiz (*Which of the pirates are you most like?*). Psychosocial workers considered it to be playful, interactive and interesting for opening discussion and encouraging introspection (e.g., identifying personal strengths). However, one proposed the following change: "*It's pretty obvious what answer they have to give to look like the pirate of their choice. A simple key might make identification of the pirate less obvious.*"

Most of the children also greatly enjoyed the cooking activities (*Jeanne La Borgne's special recipes*) aimed at promoting quality time between parents and children; only one child reported having little interest. Some psychosocial workers believed these activities could help children develop skills, self-confidence, and good life habits; they reported that cooking was an interesting and fun way to encourage special parent-child moments. One person in particular commented: "*Nice idea, well presented with "pirate" ingredients. A nice activity that's not often suggested to children and their parents.*" (Marianne, psychosocial worker). Conversely, one psychosocial worker did not find these activities necessary for understanding the book and the tools, while another questioned the relevance of the recipes. A few suggested that changes be made (e.g., use simpler and inexpensive ingredients).

In addition, children report enjoying the *My pirate team* activity, which focuses on identifying their support network. The psychosocial workers also approved this activity because the support network is "*super important to focus on*". In general, they appreciated parental involvement in this activity, as most mentioned it was an excellent idea.

Then, child participants were highly enthusiastic about the drawing activity in *Pirate apprentices' corner* aimed at promoting children's self-esteem and reinforcing the dynamic aspect of the book. Only one child reported being uninterested. Two psychosocial workers considered this section relevant, but non-essential. Psychosocial workers, in fact, felt that drawing had several possible benefits for children because it fostered a sense of pride and the integration of content and encouraged children's participation.

Content Readability and Adaptation to Child's Age

From parents' perspectives, the strategies presented allowed children to understand how to find solutions without pushing them too hard. Josiane, a mother of a 9 year old girl and an 8 year old boy stated: *"It's easy to understand and fun at the same time."* In this regard, psychosocial workers believe children should be able to understand the information conveyed in the book. They reported that the book was, on the whole, easy to read and understand thanks to simple words, clear and playful explanations and a good number of illustrations. Judith, a psychosocial worker, explained: *"The book is still quite a long read, but I think the chapters and interactive aspect should hold children's interest and keep parents involved as well."*

Nevertheless, a minority of parents and psychosocial workers stated that some of the terminology used could be difficult to understand, especially for younger children. In this regard, some psychosocial workers reported that the children may need support to understand certain concepts presented in the book (e.g., the usefulness and origin of emotions) and to do the activities (e.g., body percussion activities), despite the fact that the book is generally easy to read. This limitation was noted by a mother who participated in the study with her 9-year-old son: "*There were a few words I had to explain to him, but generally he understood the story very well. He has a lot of trouble with reading comprehension, so I had to help him make the connections with real life because he wasn't making the connections himself.*"

Overall, only, one parent and one psychosocial worker suggested that some activities may be less adapted to a child's age (e.g., special recipes section) or gender (e.g., dance activities). Furthermore, two psychosocial workers expressed concerns about the book's readability and adaptation for school-aged children. Among other things, they found that the story was somewhat "childish" considering the target audience (particularly for 11-year-old children) and did not sufficiently consider the preferences of all school-aged children. With this in mind, one worker suggested that a similar bibliotherapy be created with a different theme (e.g., sports). This way, some children "*might get a little more out of the story*". However, she believed there was little chance a child would fail to enjoy this type of reading.

DISCUSSION

The purpose of the present study was to conduct a formative evaluation of an IPT-based book (*Le Trésor de l'Île Rouge)* targeting COPMI. The evaluation documents the perspective of families with parental mental illness and of psychosocial workers toward the acceptability, appropriateness and utility of the bibliotherapy. Results suggest the book is useful and well-adapted for 7- to 11-year-old children with a mentally ill parent and their families.

First, the results of our study support the findings of previous research indicating that bibliotherapy such as the one evaluated in the current study may help children learn concrete strategies and tools to cope with the personal and familial challenges they face on a daily basis (9). Children are introduced to different coping strategies and tools for dealing with adversity thanks to the adventures and challenges faced by the characters in the story. Through bibliotherapy, children can identify with characters and situations, create connections to their experiences and discover new and healthy adaptive coping strategies to use in everyday life (53, 54, 58). In this regard, almost all parents and children in this study reported they had used or intended to use a strategy taught in the book. Also, all participating psychosocial workers underlined they would use the book in their practice and recommend it to COPMI and their families.

Second, results show the book's use of a story and dynamic activities is relevant for helping families initiate discussions on sensitive subjects like parental mental health, a finding consistent with the recognized effects of bibliotherapy (52). Indeed, bibliotherapy is frequently used in interventions to help create a safe space for dialogue with children, facilitate discussion of sensitive and emotional content, and encourage reflection and exploration of children's experiences (53). Furthermore, the parents in our study reported they particularly appreciated the parallels between situations in the book and those in everyday life. The literature as well identifies this as a benefit of using bibliotherapy (52–54, 58). Indeed, stories can depict real aspects of family and community life for children, thereby strengthening family relationships (57).

Third, participants say they appreciate the various, concrete and simple coping strategies (e.g., tips for social-emotional regulation and problem-solving) offered throughout the book. They view the strategies proposed as relevant for helping school-aged COPMI respond to challenges (e.g., family conflicts, communication problems, stress, feelings of isolation). They recognize that learning these strategies may help foster better interpersonal relationships, particularly regarding parents and family members, and improve their social support network and well-being. This further supports the relevance of the recommendations for targeting these psychosocial factors in preventive interventions for families with parental mental illness (29, 30, 60).

Clinical Implications

Results on the whole underline the relevance and acceptability of the book *Le Trésor de l'Île Rouge* for elementary school-aged COPMI and their families. This high-risk context where many potential consequences can negatively impact a child's well-being (1) necessitates the development and evaluation of tools and psychoeducational interventions to prevent mental health problems or psychological distress in these children. To our knowledge, this is the first formative evaluation of a bibliotherapy targeting school-aged COPMI. In light of the results of the present study, it may be viewed as a promising psychoeducational prevention tool.

Besides, some elementary school-aged children living with parental mental illness already present symptoms of mental illness or experience significant distress. These children are in dire need of psychosocial help and support. Many parents who have a mental illness may worry about their children's mental health and look for tools or psychosocial support services to help them cope (37). These families may face important additional challenges owing to the current pandemic and the limited capacity of mental health services to meet their needs (38, 39); such challenges can include obstacles to accessing care and services (e.g., long waiting lists and the absence of dedicated care pathways and intervention tools adapted to their needs and family issues) (40). In this context, bibliotherapy like the one in this study could offer a useful form of support for children and families who are waiting for psychosocial services or hesitate to accept help from a professional.

Moreover, according to some authors (52), bibliotherapy can also be an effective universal prevention tool for promoting mental health in primary school children. The results of the present study underscore the relevance of combining storytelling and activities adapted to the developmental level of primary school children. The book captures their attention and facilitates learning. The use of tools and adapted activities can promote children's understanding of content, facilitate discussions on sensitive issues, normalize their experiences and reinforce their interest (42, 52). Therefore, a book like *Le Trésor de l'Île Rouge* may help parents, psychosocial workers and teachers to accompany children coping with difficult everyday situations and allow them to recognize and fulfill their potential. The book can be used in a variety of ways: individually by the child, together as a family (parent-child or sibling), or in the classroom (teacher-led individual or group educational activity).

Overall, results support the importance of adapting tools and interventions to the specific age group and developmental level of targeted children, as many researchers suggest (42, 51). More broadly, the involvement of children themselves in the evaluation of interventions ensures the evaluations are accessible, relevant and appreciated, which then ensures that the services made available to them are actually used (50). Results show that the book was favorably viewed and well-appreciated by families and psychosocial workers, who also praised the book's format, illustrations and characters. According to participants, the content is clearly written, playful, well-illustrated and easy to understand; child participants were able to understand and retain the information conveyed. It could be assumed the book's readability and suitability for elementary school-aged children influenced their appreciation of it. Yet the children's obvious enjoyment proves that it was, in fact, a good choice for them. As stated above, the use of tools and adapted activities can promote children's understanding of content, facilitate discussions on sensitive topics, normalize their experiences and reinforce their interest, according to some authors (42). In consequence and considering that parents also appear to appreciate this book, it's possible children and their parents will be more inclined to use it on a daily basis, for example, to spend time together as a family or use the tools included in the book. Because there is often a high attrition rate in interventions targeting families with parental mental illness and maintaining engagement is a well-known challenge (21), offering an adapted bibliotherapy as an intervention modality could help increase the retention and participation of children and families. Ultimately, an intervention that aligns with the characteristics of the targeted clientele and responds to their needs makes for a smoother implementation process (48) and ensures that the services made available are actually used (49).

Strengths and Limitations of the Study

To our knowledge, this study is the first of its kind to validate the acceptability, appropriateness and utility of a bibliotherapy for COPMI. Generally speaking, the effects of preventive interventions are usually evaluated without considering their implementation or first documenting their acceptability, adaptation or satisfaction by those concerned (47). In the current study, data were collected and analyzed from three different types of respondents (children, parents and psychosocial workers), therefore offering different perspectives and increasing results validity (71).

Additionally, an important strength of the study lies in that scarce empirical data are available regarding the satisfaction of children in this particular age group living with parental mental illness. Children's views of the content and modalities proposed in intervention programs developed to support them are rarely collected, despite the importance (72) and numerous benefits (73) of doing so. Their views may often differ from those of parents or psychosocial workers (44). Data from research using this type of design are known to be more valid and accurate because they come from the children themselves, the primary target audience (73, 74). Children are in the best position to share their perspectives (ideas, opinions, suggestions) and experiences (75, 76). This formative evaluation contributed to the development of a bibliotherapy adapted to children's preferences, needs and developmental level.

The project also has certain limitations. First, the small number of participants limits generalisability of results. Further evaluation with a larger number of participants is therefore recommended to verify whether similar results are obtained when the book is offered to larger samples and more diverse clienteles. This broader study would ensure representativeness of results and increase the potential for generalization. Second, variables were measured using measurement instruments designed by the research team, which nevertheless met the research objectives. Finally, certain biases may have interfered with the validity of the results obtained, such as participants' reactivity (e.g., trying to please, avoiding awkward responses or criticism) or participants' expectations (e.g., social desirability), particularly among children (73).

CONCLUSION

In conclusion, this research has documented the satisfaction of children, parents and psychosocial workers with regard to a bibliotherapy targeting elementary school-aged COPMI. Overall, results show that families and psychosocial workers had a favorable opinion of the book, perceiving it to be relevant, useful and helpful. Thus, it appears well adapted to the developmental level of school-aged children. This research highlights the importance of adapting the content and modalities of interventions to children's interests, preferences, needs and developmental level. Future research should focus on promoting children's participation in the research process. It's important to develop a research culture where children's voices are heard and their involvement in the activities and decisions affecting them is an integral part of everyday research practice. Furthermore, psychosocial workers should be made aware that children, even school-aged children, require adaptive strategies to cope with the stressors they encounter. Bibliotherapy resources such as *Le Trésor de l'Île Rouge* could be made available in settings frequented by children and parents, including schools and community organizations in a perspective of universal prevention. Such resources could also be offered to parents with a mental disorder who are eager to find tools supporting family resilience.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservations. Further inquiries should be sent to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Committee for Research Involving Humans, University of Quebec in Outaouais, Saint-Jérôme (Québec), Canada. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

KV and GP undertook and led the development of the research design and methodology, with contributions from AV. KV and GP undertook the literature review and led the data collection, with contributions from AV. KV led the data analysis and interpretation of findings, with contributions from GP and AV. KV and GP wrote the manuscript with editing/contributions from AV. All authors contributed to the article and approved the submitted version.

ACKNOWLEDGMENTS

This project was approved by the Research Ethics Committee of the Université du Québec en Outaouais (# 2017-63, 2509). The authors thank the Quebec Population Health Research Network (QPHRN) for its contribution to the financing of this publication. We are also grateful for the financial support that we received from the Centre de recherche universitaire sur les jeunes et les familles (CRUJEF), that helped with the data collection fees, as well as the one from Université du Québec en Outaouais that helped to cover the linguistic review. We thank the children, parents and psychosocial workers as well as the community organizations *La Boussole* and *L'Apogée* for their invaluable participation in this project. Many thanks to the Laboratory LaPProche for their support and advice. Another special thanks to Camille Ballick and Eugenia Drolet for their linguistic review of the manuscript.

REFERENCES

1. Reupert AE. J Maybery D, Kowalenko NM. Children whose parents have a mental illness: prevalence, need and treatment. Med J Aust. (2012) 1:7–9. doi: 10.5694/mjao11.11200
2. Association AP. *Diagnostic and Statistical Manual of Mental Disorders*. 5ed. Arlington: Elsevier-Masson (2015).
3. Brockington I, Chandra P, Dubowitz H, Jones D, Moussa S, Nakku J, et al. WPA guidance on the protection and promotion of mental health in children of persons with severe mental disorders. World psychiatry: official journal of the World Psychiatric Association (WPA). (2011) 10:93–102. doi: 10.1002/j.2051-5545.2011.tb00023.x
4. Sell M, Daubmann A, Zapf H, Adema B, Busmann M, Stiawa M, et al. Family functioning in families affected by parental mental illness: parent, child, and clinician ratings. *Int J Environ Res Public Health.* (2021) 18:79–85. doi: 10.3390/ijerph18157985
5. Beardslee W, Gladstone T, O'Conner E. Transmission and prevention of mood disorders among children of affectively parents: a review. *J Am Acad Child Adolesc Psychiatry.* (2011) 50:1098–109. doi: 10.1016/j.jaac.2011.07.020
6. Gladstone TRD, Beardslee WR, Diehl A. The impact of parental depression on children. In: Reupert A, Maybery D, Nicholson J, Gopert M, Seeman MV, editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. Cambridge University Press (2015). p. 117–26. doi: 10.1017/CBO9781107707559.013
7. Aldrige J, Becker S. *Children Caring for Parents With Mental Illness: Perspectives of Young Carers, Parents and Professionals*. Bristol: The Policy Press (2003).
8. Cooklin A. Promoting children's resilience to parental mental illness: engaging the child's thinking. *Adv Psychiatr Treat.* (2013) 19:229–40. doi: 10.1192/apt.bp.111.009050
9. Riebschleger J, Costello S, Cavanaugh DL, Grové C. Mental health literacy of youth that have a family member with a mental illness: outcomes from a new program and scale. *Front Psychiatry.* (2019) 10:2. doi: 10.3389/fpsyt.2019.00002
10. Reupert A, Gladstone B, Helena Hine R, Yates S, McGaw V, Charles G, et al. Stigma in relation to families living with parental mental illness: an integrative review. *Int J Ment Health Nurs.* (2021) 30:6–26. doi: 10.1111/inm.12820
11. Tabak I, Zablocka-Zytka L, Ryan P, Poma SZ, Joronen K, Vigano G, et al. Needs, expectations and consequences for children growing up in a family where the parent has a mental illness. *Int J Ment Health Nurs.* (2016) 25:319–29. doi: 10.1111/inm.12194
12. Hosman CM, van Doesum KT, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. the scientific basis to a comprehensive approach. *AeJAMH.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
13. Bell M, Bayliss DM, Glauert R, Harrison A, Ohan J. Children of parents who have been hospitalised with psychiatric disorders are at risk of poor school readiness. *Epidemiol Psychiatr Sci.* (2019) 28:508–20. doi: 10.1017/S2045796018000148
14. Goodman SH, Rouse MH, Connell AM, Broth MR, Hall CM, Heyward D. Maternal depression and child psychopathology: a meta-analytic review. *Clin Child Fam Psychol Rev.* (2011) 14:1–27. doi: 10.1007/s10567-010-0080-1
15. Dean K, Stevens H, Mortensen PB, Murray RM, Walsh E, Pedersen CB. Full spectrum of psychiatric outcomes among offspring with parental history of mental disorder. *Arch Gen Psychiatry.* (2010) 67:822–9. doi: 10.1001/archgenpsychiatry.2010.86
16. Siegenthaler E, Munder T, Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:8–17. doi: 10.1016/j.jaac.2011.10.018

17. Christiansen H, Anding J, Schrott B, Rohrle B. Children of mentally ill parents-a pilot study of a group intervention program. *Front Psychol.* (2015) 6:1494. doi: 10.3389/fpsyg.2015.01494
18. Kato N, Yanagawa T, Fujiwara T, Morawska A. Prevalence of children's mental health problems and the effectiveness of population-level family interventions. *J Epidemiol.* (2015) 25:507–16. doi: 10.2188/jea.JE20140198
19. Reupert AE, Cuff R, Drost L, Foster K, van Doesum KTM, van Santvoort F. Intervention programs for children whose parents have a mental illness: a review. *Med J Aust.* (2012) 1:18–22. doi: 10.5694/mjao11.11145
20. Hodgins S, Lane C, Lapalme M, LaRoche C, Lavoie F, Kratzer L, et al. (editors). *A Critical Review of the Literature of Children at Risk for Major Affective Disorders*. Ottawa, ON: The Strategic Fund for Children's for Children's Mental Health (1994).
21. van Doesum KT, Hosman CM. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: II Interventions. *AeJAMH.* (2009) 8:264–76. doi: 10.5172/jamh.8.3.264
22. Weissman MM, Wickramaratne P, Gameroff MJ, Warner V, Pilowsky D, Kohad RG, et al. Offspring of depressed parents: 30 years later. *Am J Psychiatry.* (2016) 173:1024–32. doi: 10.1176/appi.ajp.2016.15101327
23. Beardslee W, Keller M, Lavori P, Staley J, Sacks N. The impact of parental affective disorder on depression in offspring: a longitudinal follow-up in a nonreferred sample. *J Am Acad Child Adolesc Psychiatry.* (1993) 32:723–30. doi: 10.1097/00004583-199307000-00004
24. Piché G, Bergeron L, Cyr M. Transmission intergénérationnelle des troubles intériorisés: Modèles théoriques et recherches empiriques. *Can Psychol.* (2008) 49:309–22. doi: 10.1037/a0013995
25. Drost LM, van der Krieke L, Sytema S, Schippers GM. Self-expressed strengths and resources of children of parents with a mental illness: A systematic review. *Int J Ment Health Nurs.* (2016) 25:102–15. doi: 10.1111/inm.12176
26. Foster K, O'Brien L, Korhonen T. Developing resilient children and families when parents have mental illness: a family-focused approach. *Int J Ment Health Nurs.* (2012) 21:3–11. doi: 10.1111/j.1447-0349.2011.00754.x
27. Gladstone BM, Boydell KM, Seeman MV, McKeever PD. Children's experiences of parental mental illness: a literature review. *Early Interv Psychiatry.* (2011) 5:271–89. doi: 10.1111/j.1751-7893.2011.00287.x
28. Lewandowski RE, Verdeli H, Wickramaratne P, Warner V, Mancini A, Weissman M. Predictors of positive outcomes in offspring of depressed parents and non-depressed parents across 20 years. *J Child Fam Stud.* (2014) 23:800–11. doi: 10.1007/s10826-013-9732-3
29. Marston N, Stavnes K, Van Loon LMA, Drost LM, Maybery D, Mosek A, et al. A content analysis of Intervention Key Elements and Assessments (IKEA): what's in the black box in the interventions directed to families where a parent has a mental illness? *Child Youth Serv.* (2016) 37:112–28. doi: 10.1080/0145935X.2016.1104041
30. Tapias E, Coromina M, Grases N, Ochoa S. Psychological treatments with children of parents with mental illness: a systematic review. *Child Youth Care Forum.* (2021) 50:1107–30. doi: 10.1007/s10566-021-09608-2
31. Bettis AH, Forehand R, Sterba SK, Preacher KJ, Compas BE. Anxiety and depression in children of depressed parents: dynamics of change in a preventive intervention. *J Clin Child Adolesc Psychol.* (2018) 47:581–94. doi: 10.1080/15374416.2016.1225503
32. Zalewski M, Goodman SH, Cole PM, McLaughlin KA. Clinical considerations when treating adults who are parents. *Clin Psychol.* (2017) 24:370–88. doi: 10.1111/cpsp.12209
33. Piché G, Villatte A, Bourque S. *Trouble mental chez le parent : Enjeux familiaux et implications cliniques*. Quebec: Presses de l'Université Laval (2021).
34. Niemelä M, Kallunki H, Jokinen J, Räsänen S, Ala-Aho B, Hakko H, et al. Collective impact on prevention: let's talk about children service model and decrease in referrals to child protection services. *Front Psychiatry.* (2019) 10:64–70. doi: 10.3389/fpsyt.2019.00064
35. van Santvoort F, Hosman CM, van Doesum KT, Janssens JM. Effectiveness of preventive support groups for children of mentally ill or addicted parents: a randomized controlled trial. *Eur Child Adolesc Psychiatry.* (2014) 23:473–84. doi: 10.1007/s00787-013-0476-9
36. Grove C, Reupert A, Maybery D. Gaining knowledge about parental mental illness: how does it empower children? *Child Fam Soc Work.* (2015) 20:377–86. doi: 10.1111/cfs.12086
37. Afzelius M, Plantin L, Östman M. Families living with parental mental illness and their experiences of family interventions. *J Psychiatr Ment Health Nurs.* (2018) 25:69–77. doi: 10.1111/jpm.12433
38. Gagné M-H, Piché G, Clément M-È, Villatte A. Families in confinement: a pre–post COVID-19 study. *Couple Fam Psychol: Res and Pract.* (2021). doi: 10.1037/cfp0000179
39. Furlong M, McGilloway S, Mulligan C, Killion MG, McGarr S, Grant A, et al. Covid-19 and families with parental mental illness: crisis and opportunity. *Front Psychiatry.* (2021) 12. doi: 10.3389/fpsyt.2021.567447
40. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
41. Thanhäuser M, Lemmer G, de Girolamo G, Christiansen H. Do preventive interventions for children of mentally ill parents work? results of a systematic review and meta-analysis. *Curr Opin Psychiatry.* (2017) 30:283–99. doi: 10.1097/YCO.0000000000000342
42. Garber J, Frankel SA, Herrington CG. Developmental demands of cognitive behavioral therapy for depression in children and adolescents: cognitive, social, and emotional processes. *Annu Rev Clin Psychol.* (2016) 12:181–216. doi: 10.1146/annurev-clinpsy-032814-112836
43. Van Doesum K, Maia T, Pereira C, Loureiro M, Marau J, Toscano L, et al. The impact of the "Semente" program on the family-focused practice of mental health professionals in Portugal. *Front Psychiatry.* (2019) 10:305. doi: 10.3389/fpsyt.2019.00305
44. Grové C, Reupert A, Maybery D. The perspectives of young people of parents with a mental illness regarding preferred interventions and supports. *J Child Fam Stud.* (2016) 25:3056–65. doi: 10.1007/s10826-016-0468-8
45. Reupert A, Goodyear M, Eddy K, Alliston C, Mason P, Maybery D, et al. Australian programs and workforce initiatives for children and their families where a parent has a mental illness. *AeJAMH.* (2009) 8:277–85. doi: 10.5172/jamh.8.3.277
46. Craig P, Ruggiero E, Frohlich KL, Mykhalovskiy E, White M. *Taking Account of Context in Population Health Intervention Research: Guidance for Producers, Users and Funders of Research*: National Institute for Health Research (2018).
47. Loechner J, Starman K, Galuschka K, Tamm J, Schulte-Korne G, Rubel J, et al. Preventing depression in the offspring of parents with depression: a systematic review and meta-analysis of randomized controlled trials. *Clin Psychol Rev.* (2018) 60:1–14. doi: 10.1016/j.cpr.2017.11.009
48. Chen HT. *Practical Program Evaluation: Assessing and Improving Planning Implementation, and Effectiveness*. London: Sage Publishing (2005).
49. Collins E, Clark H. *Supporting Young People to Make Change Happen: A Review of Theories of Change*. ActKnowledge and Oxfam Australia. (2013).
50. Patton MQ. *Developmental evaluation: Applying complexity concepts to enhance innovation and use*. New York, NY: Guilford Press (2010).
51. Newman L, Birleson P. Mental health planning for children and youth: is it developmentally appropriate? *Australas Psychiatry.* (2012) 20:91–7. doi: 10.1177/1039856211432479
52. Mendel MR, Harris J, Carson N. Bringing Bibliotherapy for Children to Clinical Practice. *J Am Acad Child Adolesc Psychiatry.* (2016) 55:535–7. doi: 10.1016/j.jaac.2016.05.008
53. Gervais J, Bouchard S, Gagnier N. *Using Stories to Prevent Anxiety Disorders in a School Context : Dominique's Handy Tricks Program*. IntechOpen (2011).
54. Goddard AT. Children's books for use in bibliotherapy. *J Pediatr Health Care.* (2011) 25:57–61. doi: 10.1016/j.pedhc.2010.08.006
55. Montgomery P, Maunders K. The effectiveness of creative bibliotherapy for internalizing, externalizing, and prosocial behaviors in children: a systematic review. *Child Youth Serv Rev.* (2015) 55:37–47. doi: 10.1016/j.childyouth.2015.05.010
56. Moldovan R, Cobeanu O, David D. Cognitive bibliotherapy for mild depressive symptomatology: randomized clinical trial of efficacy and mechanisms of change. *Clin Psychol Psychother.* (2013) 20:482–93. doi: 10.1002/cpp.1814

57. Pulimeno M, Piscitelli P, Colazzo S. Children's literature to promote students' global development and wellbeing. *Health Promot. Perspect.* (2020) 10:13. doi: 10.15171/hpp.2020.05
58. Heath MA, Smith K, Young EL. Using children's literature to strengthen social and emotional learning. *Sch Psychol Int.* (2017) 38:541–61. doi: 10.1177/0143034317710070
59. Tussing HL, Valentine DP. Helping adolescents cope with the mental illness of a parent through bibliotherapy. *Child Adolesc Soc Work J.* (2001) 18:455–69. doi: 10.1023/A:1012992116742
60. Reupert AE, Maybery D. "Knowledge is power": educating children about their parent's mental illness. *Soc Work Health Care.* (2010) 49:630–46. doi: 10.1080/00981380903364791
61. Bearslee W, Wright E, Gladstone T, Forbes P. Long-term effects from a randomized trial of two public health preventive interventions for parental depression. *J Fam Psychol.* (2007) 21:703–13. doi: 10.1037/0893-3200.21.4.703
62. Piche G, Villatte A, Habib R, Vetri K, Beardslee W. FAMILLE+: A Multifamily Group Program for Families with Parental Depression. *J Parent Fam Mental Health.* (2020) 5:1–4. doi: 10.7191/parentandfamily.1014
63. Piché G, Vetri K, Villatte A, Habib R. Évaluation pilote d'un programme d'intervention préventive ciblant les familles comprenant un parent ayant vécu un trouble dépressif. *Revue de Psychoéducation.* (2022) (in press).
64. Withnell N, Murphy N. *Family Interventions in Mental Health*. New York, NY: McGraw-Hill Education (2012).
65. Mufson L, Dorta KP, Moreau D, Weissman MM. *Interpersonal Psychotherapy for Depressed adolescents*. 2nd ed. New York, NY: Guilford Press (2004).
66. Young JF, Kranzler A, Gallop R, Mufson L. Interpersonal psychotherapy-adolescent skills training: Effects on school and social functioning. *School Ment Health.* (2012) 4:254–64. doi: 10.1007/s12310-012-9078-9
67. Dietz LJ. Family-based interpersonal psychotherapy: an intervention for preadolescent depression. *Am J Psychother.* (2020) 73:22–8. doi: 10.1176/appi.psychotherapy.20190028
68. Mahan RM, Swan SA, Macfie J. Interpersonal psychotherapy and mindfulness for treatment of major depression with anxious distress. *Clin Case Stud.* (2018) 17:104–19. doi: 10.1177/1534650118756530
69. Villatte A, Piché G, Benjamin S. Perceived support and sense of social belonging in young adults who have a parent with a mental illness. *Front Psychiatry*. (2022) 12:1–15. doi: 10.3389/fpsyt.2021.793344
70. Vetri K. *L'adaptation d'un dispositif de bibliothérapie pour les enfants âgés de 7 à 11 ans vivant avec un parent ayant un trouble mental*. Saint-Jérôme, QC: Université du Québec en Outaouais (2022).
71. Gibson CB. Elaboration, Generalization, Triangulation, and Interpretation. *Organ Res Methods.* (2016) 20:193–223. doi: 10.1177/1094428116639133
72. Bee P, Bower P, Byford S, Churchill R, Calam R, Stallard P, et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. *Health Technol Assess.* (2014) 18:100–250. doi: 10.3310/hta18080
73. Kennan D, Dolan P. Justifying children and young people's involvement in social research: assessing harm and benefit. *Ir J Sociol.* (2017) 25:297–314. doi: 10.1177/0791603517711860
74. Broström S. Children's participation in research. *Int J Early Years Educ.* (2012) 20:257–69. doi: 10.1080/09669760.2012.715407
75. Kellett M. Empowering children and young people as researchers: overcoming barriers and building capacity. *Child Indic Res.* (2011) 4:205–19. doi: 10.1007/s12187-010-9103-1
76. Nabors LA, E-Libro C. *Research Methods for Children*. Hauppauge: Nova Science Publishers, Inc (2013).
77. Brady B. Developing children's participation: lessons from a participatory IT project. *Child Soc.* (2006) 21:31–41. doi: 10.1111/j.1099-0860.2006.00024.x
78. International Q. *N'Vivo* (2020).
79. Thomas DR, A. general inductive approach for analyzing qualitative evaluation data. *Am J Eval.* (2006) 27:237–46. doi: 10.1177/1098214005283748

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 28 April 2022
doi: 10.3389/fpsyt.2022.781556

Integrated Family Approach in Mental Health Care by Professionals From Adult and Child Mental Health Services: A Qualitative Study

Hanna Stolper[1]*, *Karin van Doesum*[2] *and Majone Steketee*[1]

[1] *Department of Psychology Education and Child Studies, Erasmus University Rotterdam, Rotterdam, Netherlands,*
[2] *Department of Clinical Psychology, Radboud University Nijmegen, Nijmegen, Netherlands*

Objective: A multiple case-study in which each case was evaluated by adult and child mental health professionals who used an integrated family approach in their treatments. In this approach, treatment focuses on the mental disorders of the parents as well as on the development of the young child and family relationships. This study evaluated the experiences of professionals from adult and child mental health services using this approach. The aim of the study is identifying key elements of this approach, processes involved in treatment, and barriers to its success, with the aim of contributing to the development of practice based integrated mental health care for the whole family.

Background: Parental mental disorders have an impact on parenting and child development. To stop detrimental cascade effects and prevent parent and child from being caught up in the intergenerational transmission of psychopathology, an integrated family approach in mental health care is needed. Methods: A qualitative case study design using a grounded theory approach. Data were collected through 19 group interviews of professionals ($N = 37$) from adult and infant mental health teams who worked together in the treatment of a family.

Results: Professionals from the two services were comfortable coping with complexity and felt supported to perform their treatments by staying in touch with each other in multi-disciplinary consultations. They indicated that by attuning the treatment components to each other and tailoring them to the capabilities of the family, their treatments had more impact. A flexible attitude of all involved professionals and commitment to the interest of all family members was essential.

Conclusion: According to professionals, treatment with an integrated family approach in mental health care is of value for families by addressing the distinct roles, positions and relationships, by implementing a flexible complementary treatment plan, and by empowering professionals by multi-disciplinary consultations.

Keywords: integrated family approach, adult and child mental health services, parental mental disorder, infants and early childhood, transmission of psychopathology, family focused practice, qualitative study

Edited by:
Anja Wittkowski,
The University of Manchester,
United Kingdom

Reviewed by:
Karen Wright,
University of Central Lancashire,
United Kingdom
Roser Granero,
Universitat Autònoma de
Barcelona, Spain

Correspondence:
Hanna Stolper
h.stolper@jeugdggz.com

Specialty section:
This article was submitted to
Child and Adolescent Psychiatry,
a section of the journal
Frontiers in Psychiatry

Received: *22 September 2021*
Accepted: *28 March 2022*
Published: *28 April 2022*

Citation:
Stolper H, van Doesum K and Steketee M (2022) Integrated Family Approach in Mental Health Care by Professionals From Adult and Child Mental Health Services: A Qualitative Study. Front. Psychiatry 13:781556. doi: 10.3389/fpsyt.2022.781556

INTRODUCTION

In the Netherlands, an integrated family approach in mental health care has been developed that involves the parent with a mental disorder as well as the young child and their relationships within the family in order to prevent the intergenerational transmission of psychopathology from the parent to the child. Epidemiological research provides evidence that children of parents with a mental disorder are at serious risk of developing a mental disorder of their own during their lifetime (1, 2). The prevalence of mental disorders in such children ranges from 41 to 77% for the whole diagnostic spectrum (3). The association between mental disorders in parents and their development among children is complex and depends on a number of interacting risk and protective factors (3–5). The risks are more prominent during pregnancy and early life due to the vulnerability of the fetus and the great dependence of the infant on the environment. In this crucial phase of brain development, the brain is vulnerable to stress, and the exposure to stress early in life is associated with later psychopathology (6, 7). The establishment of a secure attachment relationship with caregivers is another critical development during the first year of life, which provides an important foundation for subsequent cognitive and social emotional development [for instance, see (8)]. In addition, a secure attachment style is mentioned as a protective factor against the development of psychopathology (9–11). Therefore, mental health care that integrates treatment of parents with a mental disorder and their young children is needed (5).

Parenting is likely to be more challenging for parents with a mental disorder because there is a risk of reactivating or exacerbating the symptoms (12–14). Although parenthood can be a motivator for parents with mental disorders to manage their symptoms more effectively (15), there is a great deal of evidence that suggests parenting can be affected by a mental disorder (13, 16, 17). For instance, depression is associated with such parental behaviors as unresponsiveness, intrusiveness, hostility, and a high level of expressed negative emotions (18).

According to Sameroff's (19) transactional model, the bidirectional nature of the parent-infant relationship over time affects both parent and child. For instance, if the parent is unpredictable in emotional availability and stimulation, the young child is at risk of developmental delays, insecure attachment styles, and challenging behavior. This in turn will make parenting more stressful and less satisfying with the additional risk of aggravating the parents' psychiatric symptoms. Therefore, in addition to the treatment of parental mental disorders, mental health services should include parenting and the evolving parent-child relationship to prevent and repair negative parent-infant interaction patterns (20, 21).

To prevent parents and children from negative cascade effects and being caught up in the cycle of intergenerational transmission of mental disorders and adverse outcomes, a paradigm shift from an individual model to a family centered model in psychiatry is needed (13, 22). However, treatment with a focus on the whole family is not common in adult mental health services for various reasons (13, 23). Worldwide, mental health services for adults and children are mostly separated, and professionals are educated in one of these two areas of clinical practice. An individual perspective on mental disorders and symptoms in adult mental health care results in a limited view that excludes the role of patients as parents, their dependent children, and the developing parent-child relationship (24, 25). In addition, child mental health services focus one-sidedly on the problems of the child and do not incorporate the mental health problems of the parents in treatment (26, 27). The split between adult and child mental health services makes it more difficult to meet the needs of parents, their young children, and the evolving relationship at the same time. Collaboration between adult and child mental health services is recommended by several researchers (22, 23, 28, 29).

An integrated family approach in the practice of mental health care involves multi-disciplinary treatment for the family as a whole. Treatment using this approach offers parents with a diversity of mental disorders and their young children a combined treatment addressing current problems in different domains within the family, namely the mental disorder(s) of the parent(s), the partner relationship, parenthood and family life, the parent-child relationship, and the child's mental or relational disorder. The aim of this collaborative integrated family approach is to increase the quality and efficiency of the treatment for parents and their young children, to improve their relationships, and to ameliorate the risk of intergenerational transmission of psychopathology or other adverse outcomes.

This combined treatment is conducted by professionals from adult and child mental health services, who collaborate closely by tailoring treatment components to the capabilities of the family, and their social and socio-economic context. These professionals carried out their treatments according to their own expertise, parallel or integrated, but always in tune with the other treatments. Therefore, they meet up regularly in a multidisciplinary consultation.

A prominent feature of families considered for integrated treatment is a complexity of interacting problems which often result in negative cascading effects. Therefore, treatment using an integrated family approach is not conducted according to a standard fixed program, but is tailored to the specific needs of the individual family. For instance, families in which there are conflicts between the parents receive couples therapy, while in other families there is a need for home treatment because of unpredictable, or lack of, daily routines. Professionals jointly determine in their multidisciplinary consultations which targets of intervention should be prioritized to initiate positive cascade effects, in which sequence, in which timeframe, and by which professionals. Expertise from adult and children's mental health services is brought together for the benefit of the whole family.

The integrated family approach is closely related to family-focused practice (FFP), which is an umbrella term for preventive and supportive interventions within adult mental health care services in which attention is paid to the family members of the patient, especially the children (22). An example of this approach is psycho-education regarding a parent's mental disorder for both the partner and the children. The difference between FFP and the integrated family approach is that the latter incorporates the parent with the disorder, the young child, and their relationships within the family in their treatment, and therefore integrates treatments by adult and child mental health services professionals. However, the aims of treatment using the integrated family approach and FFP are identical, namely the prevention of intergenerational transmission of psychopathology.

There are no studies whereby such integrated treatment and collaboration between professionals from adult and child mental health services has been evaluated. Thus, the main aim of this study is to gain insight into this approach by identifying which key elements of this approach and which processes that occur during treatment contribute to treatment success and what barriers exist to treatment success. In addition, we hope to inspire and motivate professionals and mental health services to treat the family in an integrated way so that families as a whole benefit from treatment. This study is part of a broader study that also examines the experiences of the parents and the effects of this treatment; however, the data collection for this has not yet been completed. The research questions posed in this study were: First, what do professionals indicate as key elements of success in an integrated family approach, and which processes amongst professionals emerged, in treatment of parents and their young children? Second, what do professionals identify as the benefits for the whole family? Third, what challenges or barriers did professionals experience that posed a threat to the success of treatment?

METHODS

Design

In this multiple case-study of 19 families, we evaluated with professionals of adult mental health service (AMHS) and child and adolescent mental health service (CAMHS) their experiences with the use of an integrated family approach in these 19 cases. We adopted a qualitative design using a grounded theory approach. This approach is suitable when no previous research has been done about the object of study (30). As far as we know there is a lack of knowledge of experiences of professionals using a multi-disciplinary family approach involving both adult and child mental health services. The grounded theory approach is an inductive method to derive theory through systematic collection and analysis of data (31). In line with the interpretive grounded theory approach, researchers were engaged with and played an active role in interpreting the data, resulting in a theory about key elements, and processes involved in an integrated family approach grounded in the data (32), as well as barriers to this success.

We conducted group interviews, with the professionals ($N = 37$) who were involved in these 19 cases. Group interviews have the potential to create a process among professionals wherein they can share and compare their views and experiences in a conversation (33). This can lead to a reflective stance of the professionals and increases the chance of more differentiated and deepening information about experiences with integrated family treatment and reflecting on factors contributing to and obstructing successful treatment compared to individual interviews.

Data Sampling

The data were collected through group interviews with professionals who were actually involved in the treatment of the 19 selected families. Each group interview focused on one of the 19 cases. In the group interview, the treatment the family received was evaluated by all professionals who played a substantial role in that treatment. Ethics approval was granted by the Medical Ethics Review Board at the University Medical Centre of Utrecht in the Netherlands (18-186/C). Parents of the families were asked by their therapist if they would be willing to participate in the study. The selection of the 19 family cases was based on the following criteria: adult patients with a mental disorder according to the DSM-5 and a young child up to 6 years, with relational problems or other disorders according to the DSM-5. The first 19 families were included whose treatment has been completed when the study has started and who had given their informed consent to participate. All of these families had complex problems in different domains that were interrelated and mutually influencing each other.

Procedure

Group interviews were semi-structured, lasted about 75 min, and chaired by a moderator, the primary researcher or a research assistant, and an assistant. The number of professionals in each group interview was dependent on how many professionals were involved in the treatment of the particular family, varying from two to five. Prior to the group interview, the professionals were informed about the aim and the topics of the group interview. In preparation for the group interview, the electronic case file for the parent and child were studied by the researchers and displayed in a compact chronological overview in a timeline. The moderator presented this at the start of the group interview as a warmup for the professionals. Most of the time, this was the start of a spontaneous discussion between group members.

Interview questions were open, not based on the literature but guided by topics related to an integrated family approach. These were: (theoretical) considerations about the shift from an individual to a family approach in treatment, the efficacy of the whole treatment, which factors in the treatment were helpful and whether any challenges were experienced, the contribution of the regularly multi-disciplinary consultation, the influence of the other colleagues, and critical thoughts about and satisfaction with the treatment.

After finishing each group interview, the moderator and the assistant shared their impressions in a debriefing about atmosphere, content, observations, and differences with previous group interviews. Group interviews and debriefings were audio taped and transcribed and the text was proofread.

The method of constant comparison between the different group interviews was used during the process of data sampling and a few interview questions were added to explore specific topics in more detail. For example, when some professionals were spontaneously talking about what they learned from working with this approach and what it means to them personally, a question about learning was added because it seems an interesting topic. Another adjustment in the interview guideline was made because to understand which processes contributed in the cases in which an integrated family approach did not work out well.

Data Analysis

The transcripts of the group interviews were anonymized, which means that each professional was provided with a code indicating their discipline, so the analysis process was not influenced by remembering the person. Data were analyzed using grounded theory following the analysis stages including open, axial, and selective coding (31). Atlas-ti 8 software was utilized for coding. During the open coding phase, codes were attached to quotes that could be meaningful in light of the research questions. Subsequently, this process was iterated by comparing new information with old data. In the next phase of axial coding, themes and categories emerged through linking various codes together. These categories provided the building blocks for developing grounded theory. The coding of the transcripts was done by at least two independent researchers (14 by the primary researcher and research assistants and five by two research assistants), who afterwards compared and discussed their coding to achieve consensus. Reliability of coding was assessed by estimating the degree of consensus upon completion of the coding of each transcript. In case of disagreement in the interpretation, when coders could not reach consensus about a part of the text, the primary researchers decided how to code the text. The process of coding includes memoing and writing a debriefing after finishing coding for each group interview. The memos contained information about shared emergent ideas about, perceptions of, and relationships identified within the data, and were used in searching for patterns to achieve a grounded theory.

After 16 group interviews, no new information emerged, but given the wide diversity of the cases three more group interviews were added to control for possible bias. Data sufficiency (34) was achieved when no significant new information emerged during the final group interviews and no new codes emerged during the analysis. The text of the interviews and all codes were read, reread, and compared. From the data, six superordinate categories and nine subordinate categories formed the basis of the emergent theory about key elements and processes which contributed to or impeded the success of a multi-disciplinary integrated family approach conducted by adult and child mental health services.

RESULTS

The treatment of 19 families was evaluated in group interviews with all professionals who were involved in a substantial part of the treatment of that particular family, except in one group interview. In that one case, a professional who was intended to participate in the interview but was unable to attend at the last minute. We decided to interview her by phone. Of the 17 group interviews, 14 were done in-person, but due to the Covid-19 lockdown, three were online. No difference was found in the quality from the data of the live and online meetings. In two of the 19 evaluations it was impossible to get all the involved professional together. In these two cases, all these professionals have filled in a questionnaire with the same content as the interview topics. **Table 1** provides some information about features of the parents and children in the 19 families and how many professionals attended each group interview. Some professionals participated in several interviews because they were involved with various cases. From this table it can be seen that the members of the families who were evaluated are heterogenous regarding their DSM-5 classifications, ages, and duration of treatment they received. Comorbidity is present in 68% of the parents and 37% of the children. We tried to follow the daily practice of complex cases treated by mental health services very closely. In clinical practice there is wide variety in the phenomenology of mental disorders (35) and the contexts of the patients and families. Therefore, this study did not focus on a specific classification to avoid the false impression of a homogeneous group that could have been treated in a uniform way.

The analysis of the data showed that according to professionals, an integrated family approach in mental health care generates value for families, although there were also challenging issues that could pose a threat to treatment success. We found three different important key elements and three processes amongst professionals which led to enhanced quality of treatment and improved outcomes for the family (see **Figure 1**). The three key elements are: first, the *family is seen as a whole and the distinct roles, positions and interrelationships of the family members are addressed in treatment* by the different services; second, the *treatment plan is flexible, complementary, and tailored*; and third, there is *multi-disciplinary consultation on a regular basis.* The three processes amongst professionals which led to enhanced quality of treatment because of their consultations were: *being comfortable in coping with complexities of problems, learning, pleasure* and *satisfaction*.

The main challenging issues in collaboration that pose a threat to the intended benefits are differences between the AHMS and CAMHS professionals in therapeutic concepts, organization policy, loyalties, need to exchange information, and differences in ideas about realistic targets.

Key Elements Within an Integrated Family Approach That Contribute to Benefit of the Family

Focus on the Whole Family

Professionals considered it to be of value that in the different treatments by the two services (CAHMS and AHMS), the focus was on the whole family, *addressing distinct roles, positions, and relationships*. It especially benefited parents who were able to distinguish between their roles in treatment: The role of an individual with a mental disorder, a parent of a child, and partner or ex-partner. In an integrated family approach, all distinct roles of the adult were addressed. In the treatment of the parent-child relationship at CAMHS, the vulnerability of parenthood and the fear of losing their child through outplacement impacts parents' behavior. While in individual treatment of the mental disorder parenthood is not the focus, this makes the parent more able to work on their own issues in the therapeutic relationship

TABLE 1 | Features of parent and child which treatment was evaluated by professionals in 19 group interviews.

N	Parent			Child			Treatment period[2]	Professionals[3] in group interview	
	DSM-5	**C**	**Age[1]**	**DSM-5**	**C**	**Age[2]**		**AMHS**	**CAMHS**
01	Personality disorder	1	23	Parent-child relational problem	0	24	10	1	1
02	Personality disorder	4	32	Post-traumatic stress disorder	2	1	24	1	1
03	Personality disorder	1	43	Parent-child relational problem	0	9	18	1	1
04	Personality disorder	1	30	Post-traumatic stress disorder	1	24	27	2	1
05	Personality disorder	1	36	Unspecified neurodevelopmental disorder	1	52	15	1	1
06	Personality disorder	2	18	Unspecified neurodevelopmental disorder	1	24	16	2	2
07	Bipolar disorder	1	33	Parent-child relational problem	0	3	13	3	2
08	Depressive disorder	0	30	Parent-child relational problem	0	60	9	1	2
09	Depressive disorder	1	26	Parent-child relational problem	0	7	20	1	1
10	Depressive disorder	0	39	Parent-child relational problem	0	12	14	1	2
11	Anxiety disorder	0	32	Parent-child relational problem	0	10	8	2	2
12	Anxiety disorder	2	28	Parent-child relational problem	0	4	10	2	1
13	Autism spectrum disorder	2	33	Autism spectrum disorder	1	24	16	1	2
14	Autism spectrum disorder	0	44	Parent-child relational problem	0	11	33	1	2
15	Autism spectrum disorder	1	33	Parent-child relational problem	0	48	15	1	3
16	Post-traumatic stress disorder	0	37	Unspecified neurodevelopmental disorder	1	60	30	1	2
17	Post-traumatic stress disorder	0	38	Parent-child relational problem	1	8	15	3	1
18	Post-traumatic stress disorder	2	27	Parent-child relational problem	0	8	20	2	2
19	Other specified trauma and stressor-related disorder	1	30	Parent-child relational problem	0	12	8	2	1

C, comorbidity disorders; [1] amount in years; [2] amount in months; [3] categories of professionals: psychiatrist, psychotherapist, psychologist (clinical psychologist, general psychologist), group therapist, family or couples' therapist, nurse (nurse specialist, community psychiatric nurse), professional in home treatment.

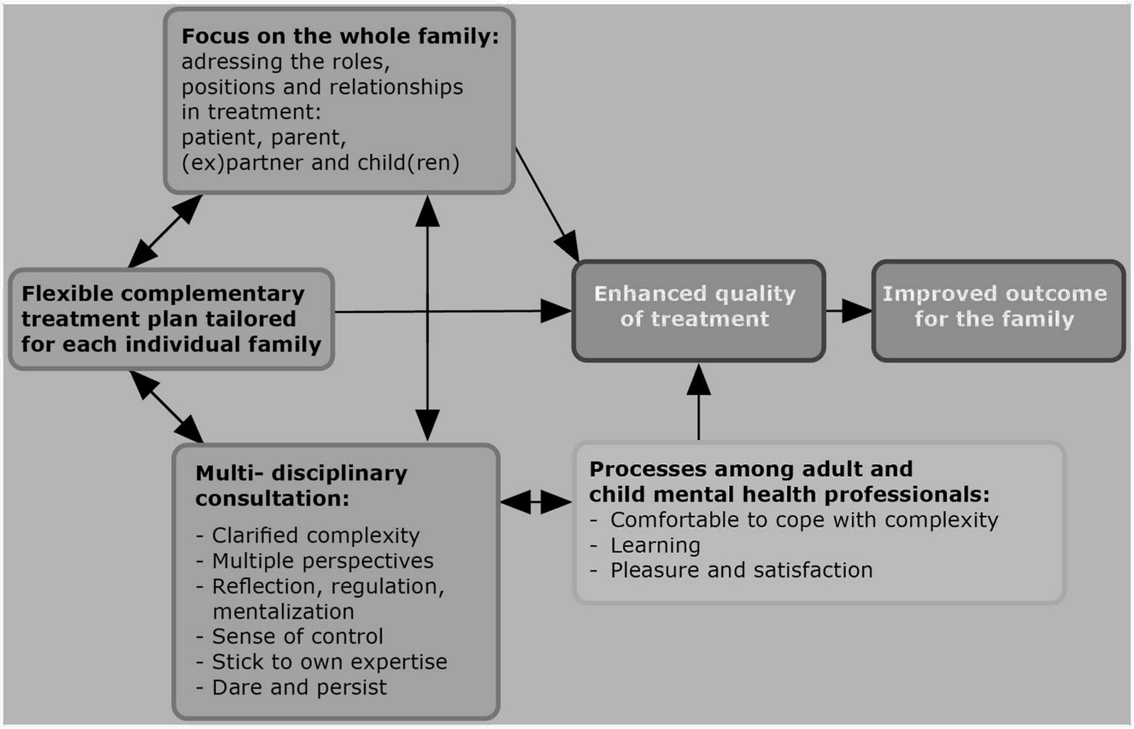


FIGURE 1 | Grounded theory about key elements, processes and benefits of an integrated family approach according to professionals.

without being anxious about consequences for their parenthood. A psychotherapist (CAMHS) who worked with the parent-child relationship described an example of this as follows:

> *I have always felt that she was more socially desirable with me. With you [nurse AMHS] she could also express anger. With me she suppressed that very much from, I suppose, the fear of "what if I show it here?" The consequences for her, whether she could raise her child. And so, it was a good thing that it could all be there.*

Flexible Complementary Treatment Plan Tailored for Each Individual Family

The direct benefit that professionals experienced was that they were better able to "*grasp the whole picture*" of the family and the environment because the focus is on the entire system.

In a family of two children aged 3 and 6 years, all members were struggling with autism or ADHD. There were housing and financial problems, and they experienced a lot of stress due to their lack of overview and planning skills. The involved professionals attuned their treatments to each other and prioritized what was most necessary so that the family was not overburdened. The nurse of the mother said:

> *Tuning in [with each other] also makes sure that you get an overall picture. I think that's really important. Because if something is going to happen in the treatment of the child, it has an impact on the treatment of the mother. And the way I work with mother affects the treatment of the child. And certainly, if patients don't tell you, if you don't ask about it, it's just possible that you [colleague CAMHS] would have started a whole treatment and I would never have known about it through the mother.*

By attuning the treatment components to each other and tailoring them to the capabilities of the family, an integrated family approach enables a better-suited and improved outcome of treatment. Flexibility and timing were mentioned as essential elements. Sometimes temporizing is needed to give the parent time to become open to learning and taking advantage of an intervention. If this succeeds, therapeutic processes become complementary and reinforce each other, resulting in improved outcome. In one of the families, it took a year before the parents were motivated and there was enough space to start couple-relations therapy. The couples' therapist said:

> *It was a good timing when I came in, and I also noticed it was very helpful that we were doing it together as well as each of us doing their own part. Because I wouldn't have been able to do anything with this couple if she [the mother] didn't get healthier and it didn't calm down at home with the daughter.*

The psychologist at AMHS agreed: "*I think we have been achieved this with the flexibility of all the different treatments.*" The psychotherapist at CAHMS added that although their intervention had started with a focus on parent-child interaction, this was not appropriate, and was redirected to a home treatment with a more practical approach aimed at parenting issues and cooperation as parents.

With another family with a baby and a mother with a personality disorder embroiled in a complicated divorce, both psychologists from AMHS and CAMHS adapted their goals and tempo of treatment to the situation. The psychologist at AMHS answered the question of whether she was affected by the treatment conducted by her colleagues as follows:

> *I have become less ambitious in my attitude toward the mother. If this is the reality in which she lives and if this is what is going on than a number of things cannot be done now. The mother was also ambitious in terms of education and work. And because I had this contextual information I thought: "Hey, wait a minute, if all this is going on then she has to be more careful on certain points."*

The psychologist at CAHMS working with another family said: "*We knew from each other what we were doing. I really thought that was an added value.... I knew when the trauma treatment of the mother was going to start and that she needed space for it.*"

Professionals at CAMHS especially emphasized the importance of considering the information from their colleagues about the impact of the mental disorder and capacities of the parent(s) and adjusting their treatment accordingly. A psychotherapist said:

> *It was, because in our regular consultation you [nurse AMHS)] told me about her history, that I recognized that I was involved with someone who had a serious chronic mental illness. That did help me to be more careful.... It helped me to determine what I could do and could not.*

Multi-Disciplinary Consultation

Multi-disciplinary consultations, in which the information provided by all professionals was brought together, and *clarified the complexity of the family*. All of them had problems in different domains, including the parental mental disorder, the young child's individual problems, and the parent-child relationship. In most of them there were also problems in other facets of the family (the other parent, the partner relation, the other children) or the environment (e.g., financial, housing). With all this shared information and *multiple perspectives,* professionals were able to develop a joint vision, recognize patterns in processes in treatment, and were more able to prioritize and stay on the same track.

In a family with complicated relations and loyalties, the psychotherapist from AHMS became aware through consultation of how she was taken in by idealizing the partner of her patient and that the family dynamic was more complicated than she could see from her position as a therapist who focused on the individual mental health problems.

> *I think that if you do it together, you will get a better understanding of such a complicated [family] system.*

It turned out that professionals were easily overwhelmed by the complexity of the families which exceeded their own knowledge and expertise. In one case the mother and her baby were referred, but at the start of the treatment it turned out that the father also was suffering from a mental disorder, and they experienced problems in their partner relationship. Three mental health services (two AMHS and CAHMS), the child protection system, and social services were involved. The grandparents participated in the care of the baby. In the group interview five professionals participated. The couple therapist said:

> *There was so much going on. A lot was needed. You could never have done this on your own.... You'd be pulled into a hundred pieces.... All those different contexts and different people seeing different people, saying different things.*

An important process that emerged was the *regulation* of emotions professionals experienced within themselves through the knowledge that another colleague was treating the problems in a different but related domain. Knowing that all risks and problems in the family were addressed by colleagues gives professionals *a sense of control* and supports professionals *in keeping focus on their own expertise and targets.*

A single mother with borderline personality disorder (BPS) and multiple traumas with hardly any support system was referred for treatment after delivery. The involved nurse motivated her patient to contact a colleague from CAMHS because she was concerned about the impact of the mental disorder on the development of the baby and the parent-child relationship. She felt that working with the baby was beyond her expertise and the therapeutic alliance too fragile to allow her to do that:

> *She [the mother] had quite a big emotion regulation disorder. And with the baby there, that was a concern for me. So, it was good to know that there was a child therapist involved, who knows much more than I about mothers and infants. … That gave me more peace of mind … When I saw her [the mother] I always thought about that baby. But I don't have to say anything about my concerns about the baby. I could not do that, I know my colleague does. … If I would have said something about it, I would have lost her [drop out].*

The same experience was brought up by professionals at CAMHS. A mother and her baby were referred for parent-child therapy, after hospitalization of the mother because of severe depression and suicidality. She was involved in a complicated divorce. The involved psychologist said:

> *I found it quite complex, their intention to divorce, and it was important for me that you [family therapist] took that piece. It gave me space to really work on that parent-child relationship. But this is also the case with the individual therapy of the mother. I knew she had really been very depressed and suicidal, and it was important to know that this was monitored by my colleague.*

The multi-disciplinary consultations provide professionals *space to think* about the meaning of the problems, to *understand* how they are interrelated, and to think about what the best port of entry for treatment will be to elicit change. A mother with a major depression and her baby had been referred for treatment with an integrated family approach after a crisis period. The professionals felt much pressure from the parents to achieve rapid improvement, which was not a realistic expectation given the severity and complexity of the problems. In the multi-disciplinary consultation this was discussed and the reflection about the meaning of it led to a better understanding. The psychotherapist from CAMHS mentioned this as follows:

> *Our regular consultation contributed to a better understanding of the mother's process … her desire to solve things quickly. Her trouble with tolerating that it doesn't work like that. Something did happen there [in the consultation] that we could understand her better and so it gave us a chance to talk about it with her.*

The multi-disciplinary consultation supported professionals in *maintaining their reflective stance* in challenging processes as splitting between the therapists, and handling a fragile or complicated therapeutic relationship by sharing their feelings. In one case, the treatment of the child was on hold because the mother was angry and did not trust the psychotherapist at CAMHS anymore. She was splitting the therapists into good and bad, but this did not lead to disruption between the collaborating colleagues because they discussed this in their consultation. The psychotherapist at AMHS made this a topic in the therapy with her patient. This was important in enabling the psychotherapist at CAMHS to stay involved until she could continue the treatment.

In cases in which professionals experienced powerlessness, friction between them emerged easily, which provoked the risk that these feelings would be transformed into anger at the other colleagues, as stated by the parent-child therapist: "*I think the risk in this case is that you say, 'Oh, the other person [colleague] is not doing it right. That's the one who has to fix it.' We all didn't get it solved*."

Another important result mentioned by the professionals was that they were *daring* to be more directive and confronting in their own treatment because of the involvement of their colleague of the other service. A nurse from AMHS said that she dared to discuss difficult things that she would otherwise suppress because of the vulnerability of the mother, knowing that the stability of the young child is guaranteed by her colleague. "*Well, especially that collaborating, … yes, I tackled more things, dared more. … And I think that otherwise I would have thought 'that's not possible, because there's a baby'. … I think I wouldn't have discussed certain things*." The psychotherapist at CAMHS in turn dared to confront more because she saw that her colleague was doing so and it did not lead to disruption in the relationship.

The multi-disciplinary consultation performs a regulatory function when it comes to being *persistent* despite slow progress. The problems, resulting from accumulating and interrelated risk factors in different domains resulting in negative cascade effects, are not easy to change and generally take a long period of treatment, sometimes several years. A mother with a personality disorder lacked feelings of bonding with her 9-month-old baby. She was convinced that her baby did not need her and so absorbed in her own concerns that she was constantly searching for confirmation of her motherhood in the baby's behavior. Consequently, she was not emotionally available enough to the baby, who in turn was avoidant toward her mother which was again a confirmation for the mother that she was superfluous. At AMHS, individual psychotherapy with the mother was conducted in which her own attachment history was an important topic and EMDR was part of her treatment. At CAMHS, videos confirmed to the mother that her baby was avoidant of her (not due to autism, but an attachment issue). She could discuss her feelings about the baby with the therapist and with help she could be emotionally available to her child and sensitive to the child's needs. There were also a few sessions with the parents and both therapists. It took one and a half year to change the beliefs of the mother and to change the patterns in the parent-child interaction. The psychotherapist who addressed the parent-child relationship gave her reflection on this collaboration with her colleague: "*And that we [colleagues] had contact and that we could share encouraged me to keep going, not to give up. Because it was sometimes very difficult, there was so little progress, especially in the first year*."

Processes Among Adult and Child Mental Health Professionals That Contribute to Benefit of the Family

The key elements identified above generate processes in the professionals which enable them to enhance their functioning.

Comfortable to Cope With Complexity

As illustrated in the above section, by joining the multi-disciplinary consultations, processes among professionals occur in which they felt regulated and calm, making them comfortable in coping with the complexity of the problems in these families.

Learning

A specific process mentioned by professionals was learning from colleagues with another area of expertise, as described by a nurse (AMHS): "*I received something extra, because I've also seen your work, and I really liked that. …I've learned a lot more about attachment and about [parent-infant] regulation.*"

In one of the group interviews, the AMHS professional brought up that she had felt uncomfortable giving feedback to her colleague at CAMHS about her treatment which did not match with this mother. The parent-child therapist's reaction was that this was indeed "*hard to take*", but it had helped her.

Pleasure and Satisfaction

In a majority of the group interviews, it was mentioned that the collaboration provides more pleasure and job satisfaction, wherein the following aspects were mentioned: "*It becomes more lively,*" "*The children are in the picture,*" "*It provides multiple perspectives,*" "*More involvement,*" "*More sense of connection,*" "*It is motivating*", "*That we all did it together, good allocation of tasks,*" and "*More things succeed that otherwise would not succeed.*" Especially professionals in AMHS got a broader view: "*I enjoyed viewing the case together and from different perspectives, discussing the child's interests, possibilities of treatment for the parent(s), the 'stuck in between' position of parent-child therapist, and searching for a helpful solution*."

Enhanced Quality of Treatment and Improved Outcome for the Families

Overall, professionals indicated that by using an integrated family approach, involving the three key elements, together with their strengthened functioning, both the quality of treatment and the outcome for the family is improved. They perceived treatment as having improved impact with their treatments compared to working separately. The following results were reported: reduction of symptoms of the parental mental disorder, and improvement of quality of the parent-child relationship, partner relationship and family relationships.

The professionals discussed this point and concluded that treatments with an integrated family approach carried out by adult and child mental health services needs time and effort involving multi-disciplinary consultation, but in the end will be more efficient and produce an improved outcome for the family. A nurse treating the mother in a complex situation with problems in all domains made this point as follows:

> *What I learned from it is that I continue focus on collaboration and communicating about when you run into things. Even though there is not always time or opportunity to do so. I don't think we would have gotten that far without this collaboration.*

A therapist of a mother expressed her view of the efficiency of collaborating with professionals with different areas of expertise as follows: "*When I started treatment that way with you [colleagues at CAMHS], I had a lot more background information as well. I didn't have to find out the same things you did.*"

Not surprisingly, many professionals mentioned the *preventive impact* of working simultaneously with the adult and the parent-child relationship, because this is an important goal of this integrated family approach. With prevention, mostly the impact on the development of the child is considered: "*… and where we hope that there will be a solid foundation [for the infant], I think we can be satisfied that we have made a nice contribution with a lot of work, and with a lot of waiting and acceptance of pace*".

In some group interviews, it appeared that in general there is more attention from those at AMHS for the children of the patient because of the existing collaboration. In addition, it was mentioned that more families could benefit from this integrated family approach.

One participant, a psychiatrist (AMHS), mentioned that an eye opener for her was recognizing the importance of looking beyond the individual to the whole context. They jointly chose a different port of entry in treatment than the guidelines prescribe. Instead of treating the chronic traumas of the mother, they chose parenting and the parent-child relationship as the port of entry in line with the request for help, and she was surprised that the duration of the treatment was shorter and the outcome of the treatment was better than she had expected at the start:

> *And that is so miraculous to me, that if you go beyond the individual and just go to the [family and social] context, treatment can have a completely different focus, as a result of which something happens to the individual which has completely surprised me. […]*
>
> *Well, I also think that from an individual viewpoint and just follow the guideline then "We have to do this and this and this," while if you take a contextual viewpoint then there are so many more factors you can act on, which can also lead to a lot of change in the individual. That is what I have learned from this case.*

Challenges in Collaboration by Adult and Child Mental Health Services

Professionals experienced various challenges in carrying out their treatments with an integrated family approach involving both adult and child mental health services. The challenges mentioned by professionals are related to the traditional differences in therapeutic concepts and organizational policy between the two services and loyalty of professionals to their own service and patient.

In a few cases the professionals struggled during treatment with the differences in the therapeutic concepts whereby on the one hand from an individual point of view the adult patient is seen as an autonomous individual with their own responsibility and on the other hand from a family approach perspective the adult is seen as a young parent with a dependent infant. For instance, professionals at AMHS emphasized patients' own responsibility for not showing up for a therapy session and the importance of their accepting the consequences of that: if that is repeated, the therapy will be terminated. Professionals at CAMHS were more likely to reach out in the interest of the young child.

There was a particular group interview regarding a case of a 18 years old mother who, on the one hand, was eager for trauma treatment, but on the other hand, did not attend her appointments regularly. The professionals discussed this, and they conceptualized the problem differently from each other. The therapists from AMHS defined it as a motivational problem: "*She had not really committed to her treatment*". The professionals from CAMHS defined her behavior as a product of her being a mother who developmentally was still a teenager, with planning problems which required more support from professionals to get the therapy started, for example through home visits. In their multi-disciplinary consultation, they struggled to decide regarding the port of entry. The professionals from CAHMS preferred to give priority to treating the traumas of the mother based on the idea that she was so preoccupied with her traumatic experiences that it impeded her from connecting with her child. The professionals from AMHS, in contrast, experienced that it was beyond their control to get the mother to show up for her sessions. Because there was a problem with parenting and the parent-child relationship, this is arguably a better port of entry. They all experienced a lot of powerlessness being caught in the dilemma between ending treatment because there did not seem to be a good context for treatment and on the other hand feeling motivated to help dependent young children.

In another case, the professional from AMHS mentioned that if she had involved the mentor of the mother-child home in an earlier stage, she would have had information about the abuse that was going on, but it was against the therapeutic concept to do so in an early stage of treatment:

> *She [mentor] said, "There had been abuse for some time, why didn't you respond?" And I thought: but I didn't know that at all. … In hindsight, I thought: maybe I should have had this mentor in the room much earlier and looked more closely at "How is it really going?" or "What is your impression as a third party?" With us, a third party [outside the organization] comes in very hard. In general, I agree with that … we think the patient's own control is important… but with this mother I think afterwards "She [the mentor] had so much information."*

The loyalty to one's "own service" in this case was particularly focused on by the professionals from AMHS. On the one hand, they wanted to be loyal to the rules of the service they worked for, and on the other hand, they felt the pressure of the interests of the children that the colleagues from CAIMHS pointed out.

Different views regarding what were realistic targets in treatment are also mentioned in a case where the treatment at AMHS and CAMHS started at various times. The professional who was just entering treatment was more optimistic than the one who had already had a long-term therapeutic involvement with the family, which caused some strain in their joint effort.

Another point particularly brought up by professionals at CAMHS was they felt a greater need to exchange information compared to their colleagues at AMHS because they needed to know what they could expect and ask from the parent. This dynamic was confirmed by the colleague at AMHS who felt no need to get frequent information about the progress of the parent-child therapy. It was sufficient to know that the child was seen in treatment. Moreover, the professionals from AMHS felt a great responsibility to protect the privacy of their patient, which sometimes prevented them from sharing information easily, despite parental consent to do so.

The collaborative relationship of professionals in treatments including parent and child can fall under pressure due to loyalties, especially if there are young children. The dependency and vulnerability of the young child easily leads to identification with the child, resulting in blaming the parent, who is the patient of the therapist at AMHS. If this dynamic is not addressed, it will isolate the therapist in a bond with the patient and withdrawal from joint treatment may occur. In one case this did occur. The therapist of the mother felt her patient was judged and she had to defend her: "*It seems to me that the mother's problems were blamed on her, that she shouldn't do certain things as a mother. While I think yes, she is also a patient.... The children had to be protected, but that atmosphere was too much for me.*"

All these mentioned challenges interfere with fruitful collaboration, and clarify what conditions are needed to prevent this from happening. Professionals struggling with these challenges highlighted the importance of commitment of all involved professionals to the family approach, taking more time for reflection, and ensuring that the interests of all family members are represented in the multi-disciplinary consultation.

DISCUSSION

The analysis of the data showed that according to the professionals, an integrated family approach in mental health care generates value for parents with mental disorders and their young children. This study revealed three important key elements that led to enhanced quality of treatment and improved outcomes for the families. The first key element is that the family is seen as a whole, and the distinct roles, positions and interrelationships of the family members are addressed in treatment by the different services. The second key element is the flexible, complementary, and tailored treatment plan, and the third key element is the multi-disciplinary consultation on regular basis.

The focus on *the family as a whole* provides information about the parental mental disorder, functioning in the parental role and family relationships, and the functioning of the child. This complementary information helped professionals to grasp the whole picture. Sharing information and observations in the *multi-disciplinary consultations* made it easier to recognize and understand patterns and themes. This contributed to making a better assessment of which goals were viable and how they could tailor their treatment to each individual family. *Flexibility in attuning their complementary treatments* to each other and *tailoring them to the capabilities of the family members* enabled the whole treatment to be applied better to the needs of the whole family. Because of these key elements in an integrated family approach, the professionals perceived their quality of treatment as enhanced, resulting in improved outcomes. The mentioned key elements prevent fragmentation of treatment, which is mentioned as an important barrier in providing integrated care (36, 37), and overburdening of the family.

As a result of these key elements, *processes* emerged in which professionals felt more comfortable in coping with the complexity of the problems in the family. They experienced a sense of control and regulation in conducting their treatment while facing a lot of problems in different, related domains in family life and the environment. They experienced support from their colleagues, knowing that they were working on related domains. This study showed that the multi-disciplinary consultation plays an important part in this. The process of working together resulted in professionals being able to keep their focus on their own expertise and targets, to maintain their reflective stance, and to think about and understand challenging processes in the therapeutic relationship and the dynamics between the therapists involved. The capacity to understand behavior of self and others in terms of internal states such as feelings, desires, and needs is called mentalization (38). Maintaining the mentalizing stance of collaborating professionals within a team is mentioned by Nijssens et al. (39) as an important capacity in challenging interactions in treatment, through which "coherence and consistency of the treatment" remains ensured (p. 85). Furthermore, professionals felt encouraged to be more directive and confrontational in their treatments and holding on and moving forward despite the treatment path being "long and bumpy." In addition, professionals expect prevention of adverse outcomes, especially for the young child, which makes them consider their work to be more meaningful. In most evaluations, professionals told us they felt more pleasure and satisfaction in working together in the treatment of the whole family and learning from each other. All these benefits for the professionals indicate an indirect *benefit for the family* that received their treatment of better regulated and more focused professionals. The professionals' feelings of self-efficacy is one of the identified facilitators in providing integrated care (36).

Another benefit for the family mentioned by professionals is in addressing the distinct roles of patient, parent, and (ex)partner in different treatments. The impact of the mental disorder is not limited to the individual functioning of the adult, but also affects the relationship with the children and the partner and, conversely, the context of the family may affect the course of symptoms. The involvement of professionals from both AMHS and CAMHS in the treatment honored the distinct roles and relationships in family life and allowed the parent to feel more comfortable about paying attention to their functioning as an individual and in relationships within the family.

Our findings about the importance and benefit of addressing the distinct roles and positions in the family are in line with previous research of family focused practice. Parents often have concerns about the effect of their mental disorder on their parenting and relationship with, and development of their children (40), but do not easily bring this up in their own treatment for fear of losing custody of their child (41). In addition, mental health professionals at AMHS are not trained to treat parenting and child development issues (42). These barriers experienced by parents and professionals can be overcome by adopting an integrated family approach.

The novel element in this study is the integration of multi-disciplinary treatments conducted by professionals from two services, AMHS and CAMHS, targeting the whole family. However, although this offers many opportunities to increase the quality and impact of total treatment, it is also more complicated, particularly because this entailed a team around the family in which professionals had not previously worked together and did not share the same therapeutic views (37, 43).

Besides the benefits, we also found *challenges* in conducting treatment of the whole family which pose a threat to the collaboration and a joint focus on the treatment of involved professionals of AMHS and CAHMS. These challenges are differences in therapeutic concepts, organization policy, loyalties, need to exchange information, and ideas about realistic targets. Sharing information between professionals from both services in a multi-disciplinary consultation is one of the key elements of treatment embracing an integrated family approach. If a professional is reluctant to do so due to personal values, their colleagues are limited in their ability to tailor the treatment to the capabilities and circumstances of the family. In our sample, we saw a tendency for CAMHS professionals being more in need of information regarding making decisions about how to direct their treatment than their colleagues at AMHS. This is a direct consequence of the different goal statements of the two services. Treatment of the young child is impossible without the parent (44), whereas treatment for the adult could be done without involving the children.

Another challenging factor was the different therapeutic concepts and policies between the services. This is related to the traditional difference in focus between CAMHS and AMHS, and the responsibility felt by the professional. On the one hand, the individual focus of AMHS views the patient as autonomous and with responsibility for their own lives. On the other hand, the focus of CAMHS is on the child and the parent-child relationship, and the professional feels a certain responsibility for the development of the child. This is not in a legal sense but arises from a sense of commitment to the vulnerability and interests of the young child. These differences pose a challenge and can potentially give rise to conflicting views on issues regarding port of entry and termination of the treatment. The loyalty of the professional to their own patient and own service is easily triggered by controversy and may keep them away from being committed to the whole family.

The above-mentioned processes which threaten collaboration between AMHS and CAMHS are in line with previous research (45, 46).

LIMITATIONS

Some limitations of our study concern the sample of families whose treatment was evaluated by the professionals. First, the recruited professionals were from two mental health services in a small part of the Netherlands. This is because there are no other mental health organizations with a comparable liaison between AMHS and CAMHS embracing an integrated family approach wherein professionals are facilitated to do so. Second, we included all professionals involved in the treatment of families that occurred during our study, and we finished data collection after data sufficiency, which possibly results in bias in terms of the specific characteristics of the sample. Although there was sufficient diversity among the included families, we cannot exclude bias partly because the study took place in a geographically small area. Third, professionals and researchers are part of the same organization, and in certain cases, they know each other, which carries the risk of their not being willing to speak out openly. Fourth, in this study we have only used one method and one source, the experiences of professionals, which is a limited foundation for a grounded theory approach presenting a concept of a theory about an integrated family approach in mental health care. This paper is part of a broader study, with the goal to build a model grounded in data of different sources as the literature, experiences of patients, observations, and measurements of treatment outcomes. In the additional studies different methods will be used to establish a more elaborate theory.

Implications for Clinical Practice

This exploratory evaluation suggests that treatments using an integrated family approach carried out by adult and child mental health services working together is of value for the families involved by empowering professionals from the different services to collaborate with one another. The experiences of professionals in this study were helpful for informing the management and the workforce of mental health services about which key elements and processes are associated with an integrated treatment approach will benefit parents and children and their relationships. By paying attention to the challenges and threats that often emerge in these treatment programs, they are better able to create conditions in which professionals will be family focused in their treatment.

An interesting finding is the great benefit of the *multidisciplinary consultation* of involved professionals, on the one hand the conducting of complementary well-tailored care for each individual family and on the other hand for professionals to be able to cope with complexity in the family. These findings may seem contradictory considering the current call for patient involvement in shared decision making (SDM). SDM is a concept wherein decisions are made based on interaction with the patient, regarding which treatment has the best evidence and is most appropriate with respect to patients' values and preferences (47, 48). However, multi-disciplinary consultation and SDM do not exclude each other. The multi-disciplinary consultation enabled the professionals to see all aspects of the whole family. Because of this, the professionals are better able to assist patients and parents to make shared decisions.

CONCLUSIONS

The experiences of professionals in this study indicate that using an integrated family approach in treatment could be of benefit for the families involved, although there are challenging issues that could pose a threat to it. The key elements that provide benefits for the family are the focus on the whole family, flexible complementary tailored treatment, and multidisciplinary consultation. Professionals indicate that because of these key elements, they were more comfortable coping with the complexity of problems in the families they treated, and they perceived that this led to better quality of treatment and resulted in improved outcomes for the family. The key elements providing benefits and the challenging issues can be understood as a recommendation to the managers to enable professionals of adult and child mental health services to collaborate and discuss their issues, all for the benefit of families.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation, and on request by email to the first author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Medical Ethics Review Board at the University Medical Centre of Utrecht in Netherlands. Written informed consent to participate in this study was provided by the participants and participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

HS put the article into writing and collected the data and conducted the data analysis with MS. All authors contributed to the article in having read and commented on the manuscript text and approved the final version of the article.

ACKNOWLEDGMENTS

We would like to thank the professionals who took their time and were open about sharing their individual experiences, the parents of the families who gave their permission to the professionals and researchers to speak about their treatment. We thank the research team that carried out the research: Anne Lynn Bijl, Trijnie Beens, Petra Henselmans, Lieke Imandt, Elvera de Kievit, Juliët Thiele, and Naomi Weezenaar. We would also like to thank the Dimence Groep that encouraged and facilitated multi-disciplinary treatment using an integrated family approach and made this research possible.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.781556/full#supplementary-material

Supplementary Figure 1 | Grounded theory flow chart (49).

REFERENCES

1. Havinga P, Boschloo L, Bloemen A, Nauta M, De Vries S, Penninx B, et al. Doomed for disorder? high incidence of mood and anxiety disorders in offspring of depressed and anxious patients: a prospective cohort study. *J Clin Psychiatry.* (2017) 78:17. doi: 10.4088/JCP.15m09936
2. Rasic D, Hajek T, Alda M, Uher R. Risk of mental illness in offspring of parents with schizophrenia, bipolar disorder, and major depressive disorder: a meta-analysis of family high-risk studies. *Schizophr Bull.* (2014) 40:28–38. doi: 10.1093/schbul/sbt114
3. Hosman CMH, van Doesum KTM, van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. *Aust e-J Adv Mental Health.* (2009) 8:250–63. doi: 10.5172/jamh.8.3.250
4. Rutter M, Sroufe LA. Developmental psychopathology: concepts and challenges. *Dev Psychopathol.* (2000) 12:265–96. doi: 10.1017/S0954579400003023
5. Stolper H, Doesum K, Van, Steketee M. How to support parents of infants and young children in mental health care : a narrative review. *Front Psychol.* (2021) 12:1–14. doi: 10.3389/fpsyg.2021.745800
6. Agorastos A, Pervanidou P, Chrousos GP, Baker DG. Developmental trajectories of early life stress and trauma: a narrative review on neurobiological aspects beyond stress system dysregulation. *Front Psychiatry.* (2019) 10:1–25. doi: 10.3389/fpsyt.2019.00118
7. Van den Bergh BRH, van den Heuvel MI, Lahti M, Braeken M, de Rooij SR, Entringer S, et al. Prenatal developmental origins of behavior and mental health: the influence of maternal stress in pregnancy. *Neurosci Biobehav Rev.* (2017) 117:26–64. doi: 10.1016/j.neubiorev.2017.07.003
8. Berlin LJ, Cassidy J, Appleyard K. The influence of early attachments on other relationships. In: Cassidy J, Shaver PR, editors. *Handbook of Attachment: Theory, Research, and Clinical Applications.* New York, NY: The Guilford Press (2008). p. 333–47.
9. McGoron L, Gleason MM, Smyke AT, Drury SS, Nelson CA, Gregas MC, et al. Recovering from early deprivation: attachment mediates effects of

caregiving on psychopathology. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:683–93. doi: 10.1016/j.jaac.2012.05.004

10. Schechter DS, Willheim E. Disturbances of attachment and parental psychopathology in early childhood. *Child Adolesc Psychiatr Clin N Am.* (2009) 18:665–86. doi: 10.1016/j.chc.2009.03.001
11. Seifer R. Young children with mentally ill parents: resilient developmental systems. In: Luthar SS, editor. *Resilience and Vulnerability: Adaptation in the Context of Childhood Adversities*. New York, NY: Cambridge University Press (2003). p. 29–49.
12. Aktar E, Qu J, Lawrence PJ, Tollenaar MS, Elzinga BM, Bögels SM. Fetal and infant outcomes in the offspring of parents with perinatal mental disorders: Earliest influences. *Front Psychiatry*. (2019) 10:391. doi: 10.3389/fpsyt.2019.00391
13. Falkov A, Goodyear M, Hosman CM, Biebel K, Skogøy BE, Kowalenko N, et al. A systems approach to enhance global efforts to implement family-focused mental health interventions. *Child Youth Serv.* (2016) 37:175–93. doi: 10.1080/0145935X.2016.1104104
14. Christiansen H, Reck C, Zietlow AL, Otto K, Steinmayr R, Wirthwein L, et al. Children of mentally III parents at risk evaluation (COMPARE): design methods of a randomized controlled multicenter study - part I. *Front Psychiatry*. (2019) 10:128. doi: 10.3389/fpsyt.2019.00128
15. Schrank B, Moran K, Borghi C, Priebe S. How to support patients with severe mental illness in their parenting role with children aged over 1 year? A systematic review of interventions. *Soc Psychiatry Psychiatr Epidemiol.* (2015) 50:1765–83. doi: 10.1007/s00127-015-1069-3
16. Laulik S, Chou S, Browne KD, Allam J. The link between personality disorder and parenting behaviors: a systematic review. *Aggress Violent Behav.* (2013) 18:644–55. doi: 10.1016/j.avb.2013.07.017
17. Christie H, Hamilton-Giachritsis C, Alves-Costa F, Tomlinson M, Halligan SL. The impact of parental posttraumatic stress disorder on parenting: a systematic review. *Eur J Psychotraumatol.* (2019) 10:1550345. doi: 10.1080/20008198.2018.1550345
18. Beardslee WR, Gladstone TRG, O'Connor EE. Transmission and prevention of mood disorders among children of affectively ill parents: a review. *J Am Acad Child Adolesc Psychiatry.* (2011) 50:1098–109. doi: 10.1016/j.jaac.2011.07.020
19. Sameroff AJ. Ports of entry the dynamics of mother-infant interventions. In: Sameroff AJ, McDonough SC, Rosenblum LK, editors. *Treating Parent-Infant Relationship Problems: Strategies for Intervention*. New York, NY: The Guilford Press (2004). p. 3–28.
20. Barlow J, McMillan A, Kirkpatrick S, Ghate D, Barnes J, Smith M. Health-led interventions in the early years to enhance infant and maternal mental health: a review of reviews. *Child Adolesc Ment Health.* (2010) 15:178–85. doi: 10.1111/j.1475-3588.2010.00570.x
21. Forman DR, O'Hara MW, Stuart S, Gorman LL, Larsen KE, Coy KC. Effective treatment for postpartum depression is not sufficient to improve the developing mother–child relationship. *Dev Psychopathol.* (2007) 19:585–602. doi: 10.1017/S0954579407070289
22. Foster K, Maybery D, Reupert A, Gladstone B, Grant A, Ruud T, et al. Family-focused practice in mental health care: an integrative review. *Child Youth Serv.* (2016) 37:129–55. doi: 10.1080/0145935X.2016.1104048
23. Gatsou L, Yates S, Hussain S, Barrett M, Gangavati S, Ghafoor R. Parental mental illness: incidence, assessment and practice. *Mental Health Practice.* (2016) 19:25–7. doi: 10.7748/mhp.19.5.25.s18
24. Reupert A, Price-Robertson R, Maybery D. Parenting as a focus of recovery. *Psychiatr Rehabil J.* (2017) 40:361–70. doi: 10.1037/prj0000240
25. Krumm S, Checchia C, Kilian R, Becker T. Mental health nurses' and psychiatrists' views on addressing parenthood issues among service users. *Int J Ment Health Nurs.* (2019) 28:425–35. doi: 10.1111/inm.12525
26. Campbell TCH, Reupert A, Sutton K, Basu S, Davidson G, Middeldorp CM, et al. Prevalence of mental illness among parents of children receiving treatment within child and adolescent mental health services (CAMHS): a scoping review. *Eur Child Adolesc Psychiatry.* (2020) 30:997–1021. doi: 10.1007/s00787-020-01502-x
27. Wesseldijk LW, Dieleman GC, van Steensel FJ, Bartels M, Hudziak JJ, Lindauer RJ, et al. Risk factors for parental psychopathology: a study in families with children or adolescents with psychopathology. *Eur Child Adolesc Psychiatry.* (2018) 27:1575–84. doi: 10.1007/s00787-018-1156-6
28. Brockington I, Chandra P, Dubowitz H, Jones D, Moussa S, Nakku J, et al. WPA guidance on the protection and promotion of mental health in children of persons with severe mental disorders. *World Psychiatry.* (2011) 10:93–102. doi: 10.1002/j.2051-5545.2011.tb00023.x
29. Stein A, Harold G. Impact of parental psychiatric disorder physical illness. In: Thapar A, Pine DS, Leckman JF, Scott S, Snowling MJ, Taylor EA, editors. *Rutter's Child and Adolescent Psychiatry.* 6th ed. Wiley & Sons (2015). p. 352–63.
30. Chun Tie Y, Birks M, Francis K. Grounded theory research: a design framework for novice researchers. *SAGE Open Med.* (2019) 7:2050312118822927. doi: 10.1177/2050312118822927
31. Corbin J, Strauss A. *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory*. Thousand Oaks: Sage Publications (2014).
32. Sebastian K. Distinguishing between the types of grounded theory: classical, interpretive and constructivist. *J Soc Thought.* (2019) 3:1–9. Available online at: https://ojs.lib.uwo.ca/index.php/jst/article/view/4116/6633
33. Morgan DL, Krueger RA, King JA. *The Focus Group Kit*. Vol. 1–6. Thousand Oaks: Sage Publications, Inc. (1998).
34. Suri H. Purposeful sampling in qualitative research synthesis. *Qual Res J.* (2011) 11:63–75. doi: 10.3316/QRJ1102063
35. Allsopp K, Read J, Corcoran R, Kinderman P. Heterogeneity in psychiatric diagnostic classification. *Psychiatry Res.* (2019) 279:15–22. doi: 10.1016/j.psychres.2019.07.005
36. Nooteboom LA, Mulder EA, Kuiper CH, Colins OF, Vermeiren RR. Towards integrated youth care: a systematic review of facilitators and barriers for professionals. *Administr Policy Mental Health Mental Health Serv Res.* (2021) 48:88–105. doi: 10.1007/s10488-020-01049-8
37. Zonneveld N, Raab J, Minkman MMN. Towards a values framework for integrated health services: an international delphi study. *BMC Health Serv Res.* (2020) 20:1–13. doi: 10.1186/s12913-020-5008-y
38. Fonagy P, Gergely G, Jurist EL, Target M. *Affect Regulation, Mentalization, and the Development of the Self.* New York, NY: Other Press (2002).
39. Nijssens L, Luyten P, Bales DL. Mentalization-based treatment for parents (MBT-P) with borderline personality disorder and their infants. In: Midgley N, Vrouva I, editors. *Minding the Child*. London: Routledge (2013). p. 93–111.
40. Van der Ende PC, Van Busschbach JT, Nicholson J, Korevaar EL, Van Weeghel J. Strategies for parenting by mothers and fathers with a mental illness. *J Psychiatr Ment Health Nurs.* (2015) 23:86–97. doi: 10.1111/jpm.12283
41. Dolman C, Jones I, Howard LM. Pre-conception to parenting: a systematic review and meta-synthesis of the qualitative literature on motherhood for women with severe mental illness. *Arch Womens Mental Health.* (2013) 16:173–96. doi: 10.1007/s00737-013-0336-0
42. Maybery D, Reupert A. Parental mental illness: a review of barriers and issues for working with families and children. *J Psychiatr Ment Health Nurs.* (2009) 16:784–91. doi: 10.1111/j.1365-2850.2009.01456.x
43. Maybery D, Foster K, Goodyear M, Grant A, Tungpunkom P, Skogoy BE, et al. How can we make the psychiatric workforce more family focused? In: Reupert A, Maybery D, Nicholson J, Göpfert M, Seeman VM, editors. *Parental Psychiatric Disorder: Distressed Parents and Their Families*. New York, NY: Cambridge University Press (2015). p. 301–11.
44. Zeanah CH, Zeanah PD. Infant mental health, the clinical science of early childhood. In: Zeanah CH, editor. *Handbook of Infant Mental Health*. Guilford Publications (2018). p. 5–24.
45. Robson J, Gingell K. Improving care for families where children and parents have concurrent mental health problems. *Child Adolesc Ment Health.* (2012) 17:166–72. doi: 10.1111/j.1475-3588.2011.00630.x
46. Auschra C. Barriers to the integration of care in inter-organisational settings: a literature review. *Int J Integr Care.* (2018) 5:1–14. doi: 10.5334/ijic.3068

47. Elwyn G, Dehlendorf C, Epstein RM, Marrin K, White J, Frosch DL. Shared decision making and motivational interviewing: achieving patient-centered care across the spectrum of health care problems. *Ann Fam Med.* (2014) 12:270–5. doi: 10.1370/afm.1615
48. Slade M. Implementing shared decision making in routine mental health care. *World Psychiatry.* (2017) 16:146–53. doi: 10.1002/wps.20412
49. *Disaster Reintegration Model: A Qualitative Analysis on Developing Korean Disaster Mental Health Support Model - Scientific Figure on ResearchGate.* Available online at: https://www.researchgate.net/figure/Grounded-Theory-Flow-Chart_fig1_323282067 (accessed April 01, 2022).

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 16 May 2022
doi: 10.3389/fpsyt.2022.815018

A Feasibility Randomized Controlled Trial of a Parenting Intervention Offered to Women With Severe Mental Health Problems and Delivered in a Mother and Baby Unit Setting: The IMAgINE Study Outcomes

Anja Wittkowski[1,2,3]*, *Richard Emsley*[4], *Penny E. Bee*[5], *Elizabeth Camacho*[6], *Rachel Calam*[1], *Kathryn M. Abel*[1,3], *Paula Duxbury*[7], *Paula Gomez*[2], *Kim Cartwright*[7] and *Holly E. Reid*[1]

[1] Division of Psychology and Mental Health, School of Health Sciences, Faculty of Biology, Medicine and Health, The University of Manchester, Manchester, United Kingdom, [2] Department of Clinical Psychology, Laureate House, Wythenshawe Hospital, Greater Manchester Mental Health NHS Foundation Trust, Manchester, United Kingdom, [3] Manchester Academic Health Science Centre, The University of Manchester, Manchester, United Kingdom, [4] Department of Biostatistics and Health Informatics, Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, United Kingdom, [5] Division of Nursing, Midwifery and Social Work, School of Health Sciences, Faculty of Biology, Medicine and Health, The University of Manchester, Manchester, United Kingdom, [6] Division of Population Health, Health Services Research and Primary Care, Faculty of Biology, Medicine and Health, School of Health Sciences, The University of Manchester, Manchester, United Kingdom, [7] Department of Research and Innovation, Greater Manchester Mental Health NHS Foundation Trust, Manchester, United Kingdom

Edited by:
Andrew T. Olagunju,
McMaster University, Canada

Reviewed by:
Amritha Bhat,
University of Washington,
United States
Seong Leang Cheah,
Independent Researcher, Sydney,
NSW, Australia

***Correspondence:**
Anja Wittkowski
anja.wittkowski@manchester.ac.uk

Specialty section:
This article was submitted to
Public Mental Health,
a section of the journal
Frontiers in Psychiatry

Received: *14 November 2021*
Accepted: *28 March 2022*
Published: *16 May 2022*

Citation:
Wittkowski A, Emsley R, Bee PE, Camacho E, Calam R, Abel KM, Duxbury P, Gomez P, Cartwright K and Reid HE (2022) A Feasibility Randomized Controlled Trial of a Parenting Intervention Offered to Women With Severe Mental Health Problems and Delivered in a Mother and Baby Unit Setting: The IMAgINE Study Outcomes.
Front. Psychiatry 13:815018.
doi: 10.3389/fpsyt.2022.815018

Background: Approximately 1–2% of mothers may experience severe mental illness (SMI) requiring admission to an inpatient Mother and Baby Unit (MBU). MBUs aim to provide mental health assessment and treatment and strengthen the mother-infant relationship, essential for infant development. Whilst MBUs offer various interventions, they do not routinely offer structured parenting interventions. The Baby Triple P Positive Parenting Program (BTP) was developed to enhance parenting competence, psychological coping and the quality of partner and other social support. Guided by lived experience consultation, we aimed to determine the feasibility and acceptability of delivering BTP plus Treatment as Usual (TAU) in this setting.

Method: A multi-site, parallel-group, single-blind pilot randomized controlled trial (registration: ISRCTN12765736) comparing BTP+TAU to TAU in participants, recruited from two MBUs in England. The Baby Triple P intervention consisted of eight parenting sessions, with the final four being delivered over the telephone following MBU discharge. Feasibility outcomes were participant intervention engagement and study retention. Clinical outcomes including maternal parenting competence, bonding and mental health outcomes were assessed at baseline, post-baseline/intervention (10 weeks) and six-month follow-up. Data were analyzed using descriptive statistics and linear regression models. An economic feasibility analysis was also conducted.

Results: Thirty-seven of the 67 eligible participants consented; 34 were randomized (16 to BTP+TAU and 18 to TAU), of whom 20 were retained at post-intervention data collection and 21 at six-month follow-up. Twelve participants (75%) completed the intervention, which was rated as highly acceptable. Clinical outcomes signaled potential improvements in maternal parenting competence, bonding, mood and mental health symptomatology in participants who received the intervention. Healthcare resource use and EQ-5D-5L questionnaires were well-completed by participants. Delivering BTP in this setting is estimated to cost £443-822 per participant.

Conclusions: This is the first trial of a parenting intervention in a MBU setting. BTP is feasible and acceptable to mothers with SMI, with a promising signal for treatment efficacy. Although minor modifications may be required for the collection of observer-rated measures post-MBU discharge, the findings indicate that a larger, definitive trial could be conducted, especially if the setting is extended to include perinatal mental health community settings.

Keywords: mothers, intervention, perinatal, severe mental illness (SMI), parenting, inpatient admission, feasibility and acceptability

INTRODUCTION

Approximately 10–20% of women develop mental health difficulties during pregnancy or the first year of having a baby, with an estimated 1% experiencing severe mental illness requiring specialist psychiatric services (1–3). Severe mental illness (SMI) in the perinatal period refers to severe and incapacitating depression, psychosis, schizophrenia, bipolar disorder, schizoaffective disorder and postpartum psychosis (2, 3). Within the literature, it has long been established that maternal mental illness has a significant, detrimental impact on the woman, her family, and her developing child [e.g., (4–10)]. The quality of the mother-infant interaction in women experiencing SMI or severe mental health difficulties can be poorer compared to mothers experiencing affective disorders [e.g., (11, 12)]. For example, Wan et al. (11) observed mothers with schizophrenia to be less responsive and sensitive toward their infants and the infant in turn were more avoidant.

Mother and Baby Units (MBUs) offer an inpatient setting for the treatment of mothers experiencing severe mental illness in the perinatal period whereby mothers are admitted jointly with their babies [e.g., (1, 2, 13)]. However, despite improvements in mental health in mothers admitted to these units, research has found that the treatment of the mother's symptoms does not necessarily always translate into more positive and attuned interactions with her baby (12). Early interventions may be a good solution to promote nurturing environments and parent and baby interaction because the potential benefits of these interventions in the general population have been largely accepted, especially in relation to child outcomes (14–19).

The Triple P system of interventions is a major contribution to the parenting intervention research with solid theoretical, scientific and clinical foundations (20–25). Triple P interventions are aimed at contributing to the healthy development of children by enhancing parental knowledge and resourcefulness regarding positive parenting practices. Given the substantial long-term negative effects of early adverse experiences, and the capacity of positive relationships to buffer or modulate these effects, an expansion of Triple P interventions has resulted in an intervention to address parental practices and needs in families expecting a baby (23). Baby Triple P is a positive parenting intervention aimed at preparing parents for their transition into parenthood by providing them with knowledge and skills to promote secure attachment with a new baby, to improve the quality of partner support alongside wider social support and to increase coping resources to reduce parental distress (26).

First-time parents rated this universal intervention to be acceptable (27). There is also evidence from studies with non-psychiatric parents that Baby Triple P has reduced infant distress in terms of inconsolable crying in six-month-old infants who also appeared to be more content in contrast to infants in the control group (28). At 2 years of corrected age, pre-term children were also found to have significantly better cognitive function, motor and symbolic communication skills compared to their control group (29). Furthermore, the acceptability and feasibility of Baby Triple P was explored in a sample of mothers (76.9% were primiparous) with postnatal depression in order to see if it could be beneficial within a mental health context (30). Of the 27 women randomized to treatment as usual or the intervention in this pilot trial, all 12 women who received Baby Triple P rated the intervention as highly acceptable and all of them were retained until the final follow up. Although this study was underpowered for analysis of effect, the results were in the predicated direction post-intervention in terms of reported levels of depression, happiness, self-regulation and subjective bonding. Acceptability and applicability in a more severe mental illness context were further examined in two related studies, using Q-methodology to explore the views and attitudes of mothers with SMI admitted to a MBU as well as MBU staff (31, 32). Mothers believed that a parenting intervention like Baby Triple P would be beneficial to them. They also deemed the MBU environment to be suitable for its delivery (31). This view was shared by MBU staff who also endorsed that this type of intervention would be feasible within the MBU setting and acceptable to mothers, regardless of their personal situation (32).

The preventive focus of the Baby Triple P programme on strengthening the mother and baby relationship or bond as well as on reducing maternal stress and increasing social support could be beneficial, specifically for women presenting with perinatal mental health problems. Despite the availability of psychiatric interventions for mothers experiencing severe mental health difficulties, no structured parenting interventions are routinely offered within these specialist perinatal settings.

The aim of this study was to evaluate the acceptability of the Baby Triple P parenting intervention in mothers with severe mental health problems in a MBU setting and the feasibility of recruiting, engaging and retaining women in this study with a view to evaluating it in a full-scale randomized controlled trial (33). In particular, this study aimed to (1) establish the suitability and acceptability of the study procedures for mothers experiencing severe mental health problems admitted to a Mother and Baby Unit, (2) determine whether there were any signals that the intervention might improve maternal and infant outcomes and (3) identify key drivers of cost associated with the intervention.

METHODS

Design and Study Setting

This study used a multisite, parallel-group single-blind (outcome assessors) randomized controlled trial (RCT) design to compare Baby Triple P plus treatment as usual (BTP+TAU) with TAU only in a MBU setting, with one MBU located in the Northwest (Site 1) and one in the Midlands (Site 2), in the UK. These MBUs had a capacity of 10 and 9 beds, respectively, and were comparable in serving a largely urban and ethnically and socio-economically diverse group of female service users and their families.

A mixed-methodology approach was used to establish the feasibility and acceptability of the intervention and study procedures [see (33), for the study protocol]. Acceptability was explored in depth through participant and MBU staff interviews and these qualitative findings will be published elsewhere (34).

Ethical Approvals and Research Governance

This trial was supported by the NIHR Research for Patient Benefit Programme (NIHR RfPB, grant number PB-PG-1014-3505) and sponsored by The University of Manchester. Study approvals were granted by the NHS National Research Ethics Service (NRES) *via* the Northwest–Greater Manchester South Research Ethics Committee (REC) (16/NW/0510), the Health Research Authority (HRA) (IRAS project number 188486, protocol number 16233) and the Research and Innovation departments of both NHS trusts overseeing the two participating MBUs (Greater Manchester Mental Health NHS Foundation Trust and Birmingham and Solihull Mental Health NHS Foundation Trust). Furthermore, an independent Trial Steering and Data Monitoring Committee and a Patient and Public Involvement (PPI) group supported this study through regular meetings throughout the feasibility trial's duration.

Participant Inclusion and Exclusion Criteria

Women jointly admitted with their babies to one of the two participating MBUs were screened against the following eligibility criteria: Participants had to: (1) be aged ≥18 years, (2) have at least one infant aged <12 months or be in the third trimester of pregnancy and expected to reside on the MBU following delivery and (3) be proficient in English to provide written informed consent and/or participate in the study assessments, interviews and, if allocated, in the intervention.

Participants were not eligible for this study if they had any of the following characteristics: (1) they experienced significant psychiatric symptoms that compromised their ability to concentrate on assessments or intervention sessions, (2) they showed severe personality disorder traits including self-harming behaviors or (3) their infants were removed from their care on a non-temporary basis. Participants were also excluded from study participation if their discharge from the MBU was scheduled within seven days of them expressing interest in the study because they would be unable to complete the initial four sessions within a week if randomized to the intervention.

Recruitment

Recruitment was conducted until April 2018 (commencing November 2016 at Site 1 and from March 2017 at Site 2). Recruitment methods involved MBU staff identifying participants who met the eligibility criteria and were willing to be approached by the research team to receive information about the study. A "Consent to Approach" form was used to document potential participants' consent to be contacted by a member of the research team. Each participant provided written, informed consent and their continued consent was sought regularly by the project manager prior to each assessment. The full recruitment procedure is detailed in Wittkowski et al. (33).

Randomization

Participants were asked to complete the first set of outcome measures at baseline before they were randomly allocated to either BTP+TAU or TAU only. The randomization list was held by the Manchester Academic Health Science Centre Clinical Trials Unit (MAHSC-CTU), subsequently known as the Manchester CTU. The allocation ratio was 1:1 with randomized permuted blocks of size 4 and 6.

The Baby Triple P Intervention

The BTP programme consisted of eight sessions, which were delivered by trained facilitators: a clinical psychologist at Site 1 and an occupational therapist at Site 2. A full description of the sessions is presented in **Table 1**. Participants allocated to receive this intervention were given the BTP workbook (26) to keep. They were advised to share it with their partners and, if desired, other family members, but not to share it with other mothers on the MBU to avoid contamination of outcomes. All participants adhered to this request.

TABLE 1 | Session content summary of the baby triple P positive parenting programme.

Session number/Theme	Content covered in session	Strategies
Session 1—Positive parenting	• Aims of positive parenting. • Factors that impact child development • Strategies for promoting healthy development. • Strategies to promote secure attachment and healthy interactions with baby. • Goal setting for first 12 months as a parent.	Communication strategies to show affection to baby.
Session 2—Responding to your baby	• Responding to baby • Teaching of new behaviors and skills	Praising baby, show attention, providing interesting/novel activities and setting routines.
Session 3—Survival skills	• Identification of unpleasant emotions and how they affect parenting. • Identification of unhelpful ways to think about parenting (parenting traps) • Expectations of transition to parenthood. • Common experiences when having a new baby.	Coping skills, settling techniques, relaxation and stress management techniques, establishing boundaries, coping plans development, though identification, social support.
Session 4—Partner support	• Common experiences in couples in transition to parenthood. • Identification of unhelpful ways of thinking about relationship. • Communication skills for maintaining relationship wellbeing.	Communication, constructive feedback, support for each other, problem solving approach, sharing task and activities.
Sessions 5 to 8—Implementing parenting routines	• Prompting self-evaluation, • Goal-setting and planning for areas of future change. • Identifying obstacles and risks and strategies to address them.	All as indicated above.

Adapted from Tsivos et al. (30).

Intervention Fidelity and Process Evaluation

Both facilitators were trained by an accredited BTP trainer to deliver the intervention, which was supported by a facilitator manual. In addition, the facilitators recorded a log of the amount of their time spent on delivering the intervention. To ensure fidelity of the intervention delivery, both facilitators also completed BTP specific checklists following each session and discussed intervention delivery and its challenges in regular peer assisted supervision and support sessions [for further details of this supervision model, see (35, 36)]. Furthermore, five sessions were digitally recorded and assessed by an independent and experienced Triple P therapist and supervisor, who confirmed that BTP sessions were delivered with high content fidelity and high process quality.

Treatment as Usual (TAU)

TAU consisted of case management using a care programme approach provided by allocated MBU psychiatric staff including consultant psychiatrists, nurses and nursery nurses and pharmacological interventions as well as non-parenting psychological interventions (e.g., CBT for depression). TAU varied according to patient needs, MBU capacity and staff availability, but excluded any parenting interventions. The variability of psychosocial and psychological interventions offered in MBUs in the UK has been documented elsewhere [see (37, 38)]. As both MBUs admitted women from anywhere in England and Wales based on bed availability, post-discharge care varied and depended on local service provision. Hence, TAU following MBU discharge included multidisciplinary team management offered by perinatal community mental health teams (CMHTs), where available, or by crisis or home treatment teams, non-perinatal CMHTs or Improving Access to Psychological Therapies (IAPT) teams.

Outcome Measures

The primary outcome was the feasibility of recruiting and retaining participants to the study and the intervention. The feasibility of BTP delivery was assessed *via* engagement with the intervention (i.e., percentage of sessions attended) and acceptability were derived from participants' satisfaction with the intervention, which was assessed *via* the *Client Satisfaction Questionnaire* (CSQ) (39).

Secondary outcomes, collected to identify signals of effectiveness and key drivers of cost-effectiveness, are summarized in **Table 2**. The suitability and acceptability of outcome measures were informed by data completeness analysis (i.e., number of items responded by active participants). Outcome data were collected by research assistants blind to the allocation arm at three time points during the study: Time 1 (baseline), Time 2 (10 weeks post-baseline) and Time 3 (6 months post-baseline).

Procedure

Full details of the procedure are reported elsewhere (33). After consenting to the study, participants completed the baseline assessment measures and MBU staff were asked to complete relevant observer-rated measures. Participants were randomly allocated to continue with TAU alone or to receive the intervention in addition to TAU during their MBU admission. Participants allocated to the intervention were usually offered weekly sessions. At each site, the project manager, who was not blind to the allocation, offered session reminders to participants and checked ongoing consent prior to each follow up assessment. These assessments were typically conducted in participants' homes because most participants were discharged from the MBU by this stage. All relevant health and safety procedures were followed.

After study completion, all participants were offered £30 as a reimbursement for their time and contributions alongside a certificate of completion and a list of useful contacts or

TABLE 2 | Overview of outcome measures used.

Outcome measure	What is being measured	Score interpretation	Completed by	Time 1	Time 2	Time 3
Family Background Questionnaire [FBQ, (40)] and *Maternal Social Support* (41)	Sociodemographic characteristics including social support	FBQ—n/a, mostly descriptive MSS: higher scores indicate better perceived social support. Cut off scores suggest <18=low, 19–24 = medium and >24 = adequate levels of support	Participants	☑		
Maternal Efficacy Questionnaire [MEQ, (42)]	Maternal self-efficacy	Higher scores indicating higher maternal efficacy	Participants	☑	☑	☑
Brief Depression, Anxiety and Stress Scale [DASS-21, (43–45)]	Subjective mood and stress	Higher scores indicate worse mood or higher stress Cut off scores are: 1. 0–4 = normal, 5–6 = mild, 7–10 = moderate, 11–13 = severe and ≥14 extremely severe levels of depression 2. 0–3 = normal, 4–5 = mild, 6–7 = moderate, 8–9 = severe and ≥10 = extremely severe levels of anxiety 3. 0–7 = normal, 8–9 = mild, 10–12 = moderate, 13–16 = severe and ≥17 extremely severe levels of stress	Participants	☑	☑	☑
Brief Symptom Inventory [BSI, (46)]	Psychiatric symptom presence and severity	Scores exceeding 63 indicate clinical significance and increased psychopathology.	Participants	☑	☑	☑
Postpartum Bonding Questionnaire [PBQ, (47, 48)]	Subjective mother-baby relationship and bond	Lower scores indicate better perceived bonding, and higher scores indicate poorer bonding and higher maternal psychopathology. Cut of scores are: 1. <11 = high bond and ≥12 low bond (PBQ Impaired Bonding) 2. <16 = normal mother-infant relationships and scores from 17–35 indicate high mother-infant relationship disorders (PBQ Rejection and Pathological Anger) 3. 1–9 = low infant-focused anxiety and ≥10 = high infant-focused anxiety (PBQ Infant-focused Anger) 4. 1–2 = low maternal pathological anger and ≥3 = high maternal pathological anger (PBQ Incipient Abuse)	Participants	☑	☑	☑
Five-Level EQ-5D [EQ-5D-5L, (49–51)]	Health status, used to calculate quality adjusted life years (QALYs)	All five dimensions have five response levels. Lower scores indicate better health and higher scores indicate worse health	Participants	☑	☑	☑
Health and Social Care Resource Use Questionnaire	Capturing resource use during the study period	N/A	Participants		☑	☑

(Continued)

TABLE 2 | Continued

Outcome measure	What is being measured	Score interpretation	Completed by	Time 1	Time 2	Time 3
Clinical Global Impression Scale [CGI, (52)]	Improvement from admission to discharge	This scale includes 3 factors (severity of mental illness, improvement since admission, efficacy of treatment with medication compared to severity of side effects). High and low scores indicate worse and better mental health respectively. Score interpretations state: 1. Minimum score = 1 (normal), maximum score = 7 (among the most extremely ill patients) (Severity of mental illness) 2. Minimum score = 1 (very much improved), maximum score = 7 (very much worse) (Improvement since admission) 3. Minimum score = 0 (marked improvement, no side-effects), maximum score = 4 (unchanged/ worse, side effects outweigh therapeutic effects). (Efficacy of treatment with medication compared to severity of side effects)	MBU staff	☑	☑	Could no longer be rated by MBU staff
Brief Psychiatric Rating Scale [BPRS, (53, 54)]	Psychiatric symptom severity	7-point Likert scale for 18 factors (minimum score = 18, maximum score = 126). Lower scores indicated better mental health and higher scores indicated worse mental health. Cut-off scores state: 18–31= mildly ill, 32–41= moderately ill, 42–53= markedly ill, >53 = severely ill.	MBU staff	☑	☑	Could no longer be rated by MBU staff
Louis MACRO (Mother and Child Risk Observation) Measure (55)	Infant wellbeing and mother-baby-relationship	Higher scores indicate better status and lower risk.	MBU staff	☑	☑	Could no longer be rated by MBU staff

organizations for additional support. After the last follow-up assessment was completed, participants in the TAU only condition were also offered the BTP workbook.

Data Analysis

The statistical and economic analyses were described in the protocol (33) and follow the intention-to-treat principle. Analyses were conducted in STATA (56) and SPSS (57).

We report participant flow using the CONSORT Statement for Pilot and Feasibility Studies (58), and descriptive summaries were generated for the outcome measures. A linear regression model was used to estimate the effect of treatment allocation on the self-reported outcomes at post-intervention (i.e., 10 weeks post-baseline, Time 2) and at 6-month follow-up (Time 3) separately, adjusting for outcome measures at baseline. Adjusted mean differences, bias corrected bootstrap standard errors and 95% confidence intervals (CIs) are reported as well as Cohen's *d* standardized effect sizes (calculated from the adjusted mean differences and the pooled standard deviation at baseline) and their corresponding 95% CIs.

Resource use data were collected from an NHS and social care perspective. The currency used was GBP (£) and price year 2018. Descriptive summaries were generated for the EQ-5D-5L and healthcare resource use data. Utility values were derived using the crosswalk methodology (50) as currently recommended by NICE (51) and quality adjusted life years (QALYs) were calculated using an area under the curve approach. The cost of delivering the Baby Triple P intervention was estimated based on the number of hours that facilitators reported spending on delivering the intervention, and the respective unit cost (59) of the NHS grade that each facilitator was employed on. The cost of training the facilitators, based on the employment grade of the person who delivered the training and duration of training, was also included.

RESULTS

Participant Characteristics

The two groups of participants were comparable across most demographic and clinical characteristics (see **Table 3**) and reflect the wider urban MBU population as well. On average participants were 29 years old and their partners slightly older at 33 years old. Their infants, of which 55% were girls, were mostly the result of planned (59%) but complicated (62%) pregnancies. The infants had an average age of 14.62 (SD = 10.31) weeks. The mothers were mostly British (68%), primiparous (59%), married or cohabiting (61% and 29%, respectively) and rated their partner and wider social support to be moderate (20.81, SD = 3.41).

Although infants in the TAU group were on average 2 weeks older than those in the BTP+TAU group at baseline, there were no other differences between the two groups which suggests that randomization was performed successfully.

In terms of their mental health, most participants reported previous mental health difficulties (85%) and were currently taking medication (97%), predominantly for affective diagnoses with anxiety (72%). The most common diagnoses were depression (21%) and anxiety with an affective disorder (21%) but only 6% received a diagnosis of an anxiety disorder only. The other most common diagnoses were bipolar disorder (15%) and personality disorder with affective disorder (15%). Postpartum psychosis was diagnosed in 12% of the participants. A higher percentage of participants from the TAU only group received a diagnosis of postpartum psychosis (16%) and of personality disorders with affective disorders (16%) compared to the BTP+TAU group (6 and 6% respectively). Further diagnostic and psychosocial information can be found in **Table 3** and **Supplementary Table 1**.

Changes to the Study Protocol

As the original grant submission for this study predated the publication of this CONSORT statement, we analyzed the data in accordance with our original funding submission and published protocol [see (33)]. However, there were some changes to the original protocol (see **Supplementary Table 2** for full details). Based on preliminary data on the capacity (19 mothers and babies per unit per month) and turnover [average admission duration of approximately seven weeks; (37)] of the two participating MBUs, we anticipated a potential pool of 209 women to be admitted and aimed to recruit approximately 60 to this feasibility trial. During the study recruitment period, however, the admission rate was considerably lower, with women being admitted for longer with an average of about nine weeks. As the amount of available data was low, analyses were conducted using all available values rather than performing multiple imputation. NHS site was not included as a covariate along with group and baseline measures, due to the low number of participants recruited at Site 2. Due to the low number of participants assessed at post-baseline with observer-rated measures, regression analysis was also not conducted using observer-rated measures, except for the CGI (52). Originally, research staff were going to complete observer-rated measures, but during the study the research team and trial steering committee agreed that, despite training in questionnaire administration, research assistants lacked the expertise of clinical psychiatric and nursing staff to adequately rate symptoms of mental health. Finally, as one of the sites had stopped administering the Health of the Nation Scale (60) as a routine outcome measure, it was also not used in any analyses.

Although two blind breaches per site occurred, in all four cases it was possible for another trained assessor to undertake these assessments instead. Hence, research assistants who undertook data collection remained blind to the participants' allocation arm.

Feasibility of Recruitment and Retention

The flow of participants through the study is summarized in **Figure 1**. All 165 women (100%) admitted to one of the two MBUs during the recruitment period were screened, but 98 (59%) were not eligible. Of the 67 eligible women, 37 (55%) consented to take part in the study. However, two participants were discharged before randomization took place and one participant was discharged following randomization, making all three ineligible. The remaining 34 participants (roughly 52% of the 65 eligible participants) were randomized to receive either TAU only ($n = 18$) or BTP+TAU ($n = 16$). As can be seen in

TABLE 3 | Demographic, psychiatric, psychosocial and delivery-related characteristics of the participants, their infants and partners.

	Total (*n* = 34)	**BTP + TAU (*n* = 16)**	**TAU Only (*n* = 18)**
Maternal characteristics			
Mean age (years) (SD)	29.3 (4.1)	29.3 (0.98)	29.3 (4.4)
Perceived severity of current psychological difficulties (M and SD, from a 1 to 10 scale)	6.5 (1.66)	6.7 (1.6)	6.3 (1.6)
Currently taking medication (yes%)	97% (*n* = 33)	94% (*n* = 15)	100% (*n* = 18)
Mean length of stay in MBU in weeks (SD)	9.2 (4.5)	9.5 (5.2)	9.1 (3.9)
Mental health history - previous psychological difficulties (yes%)	85% (*n* = 29)	87% (*n* = 14)	83% (*n* = 15)
Affective disorders (% of sample with previous mental health difficulties)	13% (*n* = 4)	–	26% (*n* = 4)
Affective disorders + anxiety (% of sample with previous mental health difficulties)	72% (*n* = 21)	86% (*n* = 12)	60% (*n* = 9)
Other (% of sample with previous mental health difficulties)	6% (*n* = 2)	–	13% (*n* = 2)
Did not specify (% of sample with previous mental health difficulties)	10% (*n* = 2)	14% (*n* = 2)	–
Difficulties occurring during previous pregnancies (yes, % of sample with previous mental health difficulties)	17% (*n* = 5)	14% (*n* = 2)	20% (*n* = 3)
Psychiatrist diagnosis			
Postpartum psychosis	12% (*n* = 4)	6% (*n* = 1)	16% (*n* = 3)
Bipolar disorder	15% (*n* = 5)	19% (*n* = 3)	11% (*n* = 2)
Depression with psychotic features	12% (*n* = 4)	6% (*n* = 1)	16% (*n* = 3)
Depression	21% (*n* = 7)	31% (*n* = 5)	11% (*n* = 2)
Anxiety (including GAD and PTSD)	6% (*n* = 2)	12% (*n* = 2)	0% (*n* = 0)
Anxiety (including GAD, PTSD and OCD) and affective disorders	21% (*n* = 6)	12% (*n* = 2)	16% (*n* = 4)
Personality disorder and affective disorders	15% (*n* = 5)	6% (*n* = 1)	16% (*n* = 4)
Schizophrenia	2% (*n* = 1)	6% (*n* = 1)	0% (*n* = 0)
Infant characteristics			
Mean age (weeks)	**14.6 (10.3)**	**13.6 (2.5)**	**15.5 (2.5)**
Gender (Female %)	55% (*n* = 19)	63% (*n* = 10)	50% (*n* = 9)
Relationship status (%)			
Married	61.8% (*n* = 21)	50% (n = 8)	72% (*n* = 13)
Living together	29.4% (*n* = 10)	50% (n = 8)	11% (*n* = 2)
Single	8.8% (*n* = 3)	0% (*n* = 0)	17% (*n* = 3)
Parity (%)			
First time parent	59% (*n* = 20)	63% (n = 10)	50% (*n* = 10)
Two children	29% (*n* = 10)	37% (n = 6)	20% (*n* = 4)
Three children	6% (*n* = 2)	–	11% (*n* = 2)
Over three children	6% (*n* = 2)	–	11% (*n* = 2)
Ethnicity (%)			
British	68% (*n* = 24)	75% (*n* = 12)	61% (*n* = 11)
Other white background	18% (*n* = 6)	13% (*n* = 2)	20% (*n* = 4)
Asian British	12% (*n* = 4)	6% (*n* = 1)	16% (*n* = 3)
Other mixed background	3% (*n* = 1)	6% (*n* = 1)	–
Education level (%)			
No qualifications	6% (*n* = 2)	6% (*n* = 1)	6% (*n* = 1)
GCSEs, CSEs or O-levels	18% (*n* = 6)	19% (*n* = 3)	17% (*n* = 3)
A levels/BTEC	18% (*n* = 6)	19% (*n* = 3)	17% (*n* = 3)
Trade/apprenticeship	15% (*n* = 5)	25% (*n* = 4)	6% (*n* = 1)
University degree	24% (*n* = 8)	25% (*n* = 4)	22% (*n* = 4)
Postgraduate degree	9% (*n* = 3)	–	17% (*n* = 3)
Other	12% (*n* = 4)	6% (*n* = 1)	17% (*n* = 3)
Family income (%)			
Upper-middle – High	38% (*n* = 13)	44% (*n* = 7)	33% (*n* = 6)
Middle	38% (*n* = 13)	38% (*n* = 6)	39% (*n* = 7)
Low-middle – Low	24% (*n* = 8)	19% (*n* = 3)	28% (*n* = 5)

(Continued)

TABLE 3 | Continued

	Total (n = 34)	BTP + TAU (n = 16)	TAU Only (n = 18)
Reported financial issues in the last 12 months	15% (n = 5)	13% (n = 2)	28% (n = 5)
Maternal employment (%)			
Full-time	9% (n = 3)	–	17% (n = 3)
Part-time	6% (n = 2)	6% (n = 1)	6% (n = 1)
Home-duties	15% (n = 5)	–	28% (n = 5)
Maternal leave	50% (n = 17)	63% (n = 10)	39% (n = 7)
Unemployed	21% (n = 7)	31% (n = 5)	11% (n = 2)
Partner characteristics			
Mean partner/husband age (years)	33 (6.8)	33 (7)	32 (6.6)
Previous diagnosis of depression (yes)	3% (n = 1)	6% (n = 1)	–
Partner's education (%)			
No qualifications	3% (n = 1)	6% (n = 1)	–
GCSEs, CSEs or O-levels	24% (n = 8)	13% (n = 2)	33% (n = 6)
A levels/BTEC	12% (n = 4)	13% (n = 2)	11% (n = 2)
Trade/apprenticeship	15% (n = 5)	25% (n = 4)	6% (n = 1)
University degree	24% (n = 8)	25% (n = 4)	22% (n = 4)
Other	24% (n = 8)	31% (n = 5)	17% (n = 3)
Partner employment (%)			
Full-time	68% (n = 23)	88% (n = 14)	50% (n = 9)
Part-time	6% (n = 2)	–	11% (n = 2)
Home-duties	6% (n = 2)	–	11% (n = 2)
Unemployed	15% (n = 5)	13% (n = 2)	17% (n = 3)
Pregnancy characteristics (%)			
Planned pregnancy (yes %)	59% (n = 20)	63% (n = 10)	53% (n = 10)
Complications during pregnancy (yes %)	62% (n = 21)	56% (n = 9)	67% (n = 12)
Vaginal delivery	53% (n = 18)	50% (n = 8)	53% (n = 10)
Induced labor	21% (n = 7)	19% (n = 3)	20% (n = 4)
Assisted delivery			
Forceps	15% (n = 5)	25% (n = 4)	5% (n = 1)
Ventouse	3% (n = 1)	–	5% (n = 1)
Episiotomy	12% (n = 4)	13% (n = 2)	10% (n = 2)
Fetal Monitoring	12% (n = 4)	6% (n = 1)	17% (n = 3)
Emergency Cesarean	12% (n = 4)	13% (n = 2)	10% (n = 2)
Planned Cesarean	18% (n = 6)	19% (n = 3)	17% (n = 3)
Other	3% (n = 1)	–	5% (n = 1)
Maternal social support (mean/SD)	20.8 (3.4)	20.4 (4.9)	20.6 (4.2)
Feels supported by friends	3.6 (1.3)	3.5 (1.3)	3.7 (1.3)
Feels supported by family	4.1 (1.2)	4.3 (1.2)	3.9 (1.3)
Feels supported by husband/partner	4.5 (0.9)	4.7 (0.5)	4.2 (1.1)
Experiences high level of conflict with husband/partner	3.4 (1.0)	3.5 (1.0)	3.2 (1.2)
Feels being controlled by husband/partner	4.0 (1.5)	4.5 (1.1)	3.6 (1.8)
Feels loved by husband/partner	4.2 (1.2)	4.4 (1.0)	4.0 (1.4)

Values in bold indicate significant differences between groups at $p < 0.05$.

Figure 1, a greater number of participants were recruited at Site 1 ($n = 27$) than at Site 2 ($n = 7$).

In terms of study retention, 21 of the 34 eligible participants (approximately 62%) completed the final follow-up assessment, but retention rates varied considerably between the groups from baseline to the two assessment timepoints (e.g., 75 and 63% for BTP+TAU and 44–61% for TAU). Only four participants from the BTP+TAU group did not complete assessments at Time 2, in contrast to 10 from the TAU only group. However, at Time 3 three TAU participants agreed to questionnaire completion again, while only one participant in the other group did so. From Time 1 to Time 2, 75% of BTP+TAU

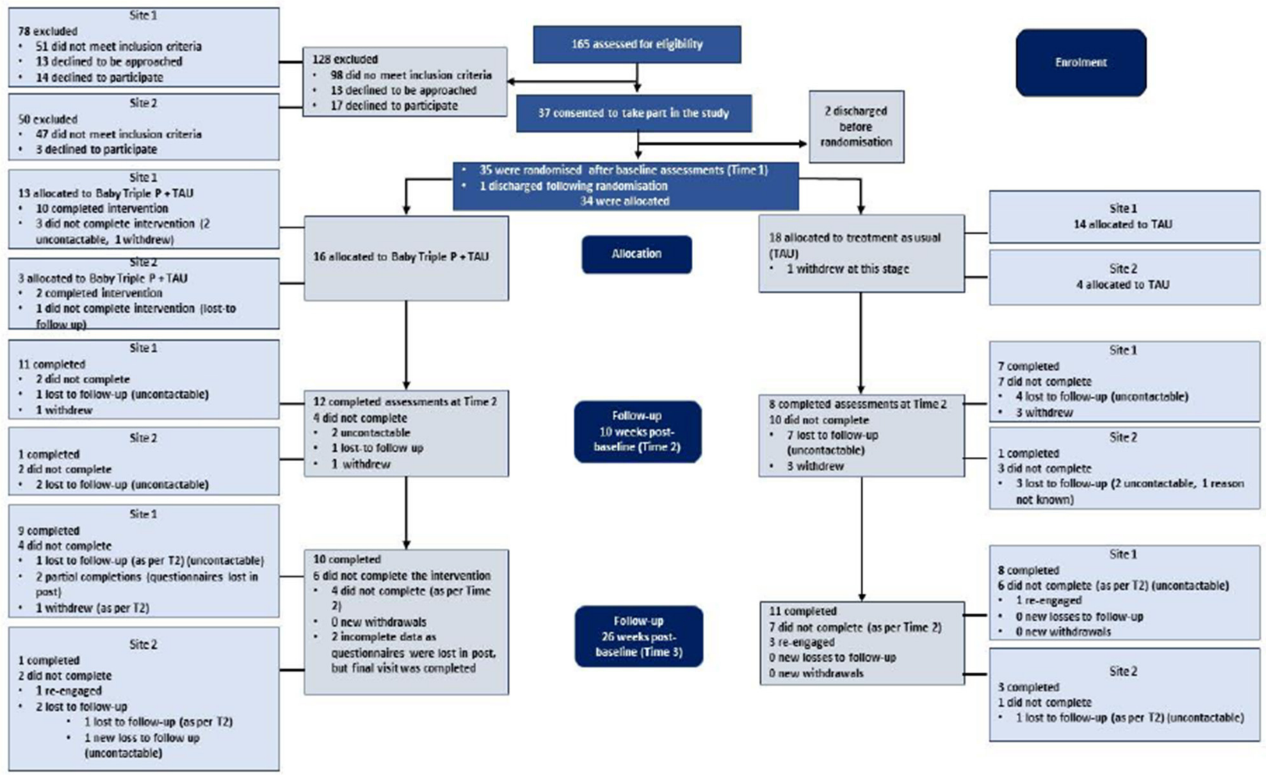


FIGURE 1 | Consort diagram.

participants were retained in the study. At the final follow up, all 12 of the participants who completed the intervention agreed to assessments visits. However, two participants requested to complete the questionnaires afterwards and their self-report data, with the exception of the EQ-5D-5L (49, 51), were subsequently lost in the post. Thus, full data from 62.5% of participants were available for analysis. In contrast, only 44% of TAU only participants remained engaged with the study at Time 2 but, with three participants returning to complete measures at Time 3, 61 % were retained in the end.

Baby Triple P Engagement and Acceptability

Of the 16 participants allocated to receive BTP+TAU, 93% ($n = 14$) completed the four core sessions which were almost always delivered during their MBU admission. The first four sessions are essential, whilst the remaining four sessions are for parents to practice their skills and to problem solve; these final sessions are typically delivered *via* the telephone. The data revealed that 81% continued to engage with the intervention at Session 5 which then dropped to 75% for the remaining three sessions. Most participants would have been discharged from the MBU at this stage of the intervention. In total, 75% ($n = 12$) completed the intervention.

Ten of these 12 participants returned the *Client Satisfaction Questionnaire* (CSQ) (39) by post. Their overall mean score was 75.33, out of a total possible score of 91, which indicates high acceptability of the intervention (see **Supplementary Table 3** for further details). Responses for individual domains indicate that participants particularly valued the quality of the programme. They were also satisfied with the overall delivery of the intervention and that they were particularly satisfied with the progress of their baby.

A more detailed analysis of the intervention's acceptability alongside the study procedures, based on interviews with study participants and MBU ward staff, will be reported elsewhere [see (34)].

Serious Adverse Events and Contamination

No research related serious adverse events were recorded for any participants during the study period, suggesting that the parenting intervention was safe. All participants adhered to our requests of restricting any sharing of BTP learning and materials, such as the workbook, with their partners and/or family. No contamination issues were reported or observed.

Secondary Outcome Measures

Of the participants who were retained in the study, completion of the assessment measures was very high for all five outcomes collected across the three assessments: MEQ (91%), DASS-21 (97%), PBQ (97%), BSI (94%) and EQ-5D-5L (100%). However, one participant in the BTP+TAU group completed the EQ-5D-5L only at the Time 2 assessment, but not the other self-report questionnaires. We did not identify any patterns in the missing data that indicated unacceptability of certain items or specific questionnaires. MBU staff completed observer-rated measures for 94% of participants at Time 1. However, their completion rate dropped to 20% at Time 2, which typically fell outside participants' average MBU admission of about 9 weeks, because MBU staff no longer had contact with study participants following discharge and could not report on their outcomes. Consequently, it was decided to not seek further information from staff at Time 3.

Between-Group Effect Sizes

The mean total scores for the clinical outcome measures at baseline are presented in **Table 4** and group differences following treatment and at follow-up are detailed in **Table 5**. Based on cut-off scores from the DASS (45), at baseline participants across both groups rated their symptoms of depression as severe, anxiety as extremely severe, and stress as moderate (see **Table 4**). Based on cut-off scores proposed by Brockington et al. (48), mean PBQ total scores as well as mean PBQ scores regarding impaired bonding indicate psychopathology in both groups. Although *t*-scores for BSI Global Severity Index suggest that this sample was below the cut-off score (i.e., *t*-score of 63) for identification of psychiatric disorders (61), participants' BSI scores appear to be higher than other clinical samples [e.g., (62)]. At baseline, MBU psychiatrists rated the participants as being moderately to severely ill [according to their CGI and BPRS assessments (54)]. Mothers allocated to the BTP+TAU group were also assessed as presenting with more risky behaviors related to baby care than the mothers from the TAU only group as assessed by Louis MACRO total scores and the Louis MACRO subscales scores regarding emotional care, parenting and mother's mental state (see **Table 4**).

The adjusted mean differences between the groups indicate higher levels of improvement (i.e., higher scores in MEQ and lower scores on all other measures) for the intervention group than for the TAU only group at post-intervention (see **Table 5**). From baseline to post-intervention, large effect sizes (63) were observed for the DASS total scores as well as the DASS stress subscale scores. Improvements were also evident in terms of symptomatology: large effect sizes were noted for participant-completed BSI positive symptom Total and Distress scores, interpersonal sensitivity, depression, hostility and psychoticism. Although only evidenced by a medium effect size, improvements in mental state, as assessed by the psychiatry staff rated CGI, were also greater for the BTP+TAU group compared to the TAU only group. Improvements were also noted in mothers' perceptions of their parent-baby bond: medium effect sizes were noted for the overall PBQ total score as well as for the rejection and anger subscale both at post-intervention and six-month follow-up.

Although overall effect sizes seem to reduce from post-intervention to final follow-up, inspection of mean values for each group across assessment points indicate greater and sustained improvements in all available outcome measures for the BTP+TAU group compared to the TAU only group. However, the small sample size as well as the large range in confidence interval values could suggest imprecision of the effect of the differences between groups.

The potential benefits of the intervention were explored further: individual scores from self-reported measures were also assessed for clinical significance changes by calculating Jacobson and Truax's (64) reliable change index (RCI) from baseline to post-intervention and follow-up scores. We then summarized the number of participants with a reliable change indicating improvement (a score of > 1.96) in each treatment group. Results from this reliable change index analysis indicated that more participants in the BTP+TAU group showed clinically significant improvements from baseline to follow-up assessments in almost all of the self-reported measures compared to the TAU only participants (see **Supplementary Table 4**). The findings indicated that those in the intervention group improved more quickly from baseline to Time 2, especially in terms of self-reported symptoms of anxiety, depression and stress (DASS) and other symptoms (BSI), and these improvements were also evident from baseline to final assessment. Both groups gained in perceived maternal competence (MEQ) but only participants in the intervention group showed improvements in how they rated their overall bonding with their infants (e.g., 0% vs. 36% and 0% vs. 40% to Time 2 and Time 3, respectively).

Economic Data

Utility values and QALYs derived from the EQ-5D-5L are summarized in **Table 6**. Over the whole follow-up period the intervention group had higher utility values on average than the TAU only group.

The cost breakdown of delivering BTP in a MBU setting is summarized in **Table 7**. Training was delivered once in each Site over 3 days (assuming 7.5 h per day, this equates to 22.5 h). The total cost (including training) for Site 1 was estimated to be £443 per participant, based on an NHS Band 8a Clinical Psychologist delivering the training (£63/h, 64) and the 69 h delivering the intervention to 13 mothers. The total cost for Site 2 was estimated to be £822 per participant, based on an NHS Band 7 Occupational Therapist delivering the training (£53/h, 64) and the 24 h delivering the intervention to three mothers. The average cost across both sites was £514 per participant. Due to low recruitment at Site 2, the training was more expensive per participant who received the intervention at that MBU. Data regarding healthcare resource use, reported in **Supplementary Table 5**, suggest that there were differences between the groups, with greater resource use for the TAU only group for all services except for nurses. The healthcare resource use data were complete in almost all cases examined and the quality of the data were good.

The details of the index MBU admission were recorded for 22 out of 34 participants. The mean length of stay was 62 days (95% CI 39 to 85; $n = 10$) in the TAU group and 64 days (95% CI 47 to 82; $n = 12$) in the BTP+TAU group. The unit cost per MBU bed per night was £729 (65). The mean cost of the index MBU admission was £46,778 (95% CI 34,084 to 59,471) in the intervention group and £45,417 (95% CI 28,731 to 62,103) in the control group. Two participants were re-admitted to an MBU following index admission (both from the TAU group), totalling an additional 37 days of inpatient care in an MBU (costing £26,973); this may be a key driver of cost.

TABLE 4 | Self-reported and observer-related outcomes at baseline.

	BTP + TAU ($n = 16$)		TAU only ($n = 18$)		Total ($n = 34$)	
	M	***SD***	***M***	***SD***	***M***	***SD***
Self-report measures						
MEQ	52.44	10.58	52.78	10.81	52.62	10.54
DASS total	83.13	18.17	81.56	32.8	82.29	26.55
DASS depression	25.06	10.88	25	13.99	25.02	12.43
DASS anxiety	22.31	8.8	20.72	13.65	25.03	12.44
DASS stress	26.44	10.09	28.11	11.47	21.47	11.48
PBQ total	46.56	26.77	42	25.17	27.32	10.72
PBQ impaired bonding	18.81	12.85	19.22	12.3	19.03	12.37
PBQ rejection and pathological anger	13	8.07	11.56	8.07	12.24	7.98
PBQ infant focused anger	7.25	4.63	7.94	3.78	7.62	4.15
PBQ risk of abuse	2.19	3.97	3.28	3.98	1.5	3.51
BSI global Severity Index *t*-score (raw score)	58 (2.05)	6.66 (0.66)	60.12 (2.25)	10.61 (0.93)	59.09 (2.15)	8.85 (0.80)
BSI positive Symptom Total *t*-score (raw score)	59.38 (42.18)	7.92 (7.25)	60.88 (42.41)	11.22 (10.54)	60.15 (42.30)	9.64 (8.96)
BSI positive Symptom distress *t*-score (raw score)	55.81 (2.54)	8.06 (0.57)	56 (2.53)	10.69 (0.74)	55.91 (2.54)	9.36 (0.65)
BSI Somatisation	57.19 (1.62)	7.62 (0.75)	56.88 (1.69)	9.86(1.09)	57.03 (1.68)	8.71 (0.92)
BSI obsessive-compulsive	59.5 (2.54)	7.45 (0.91)	60.47 (2.60)	9.42 (0.97)	60 (2.57)	8.4 (1.08)
BSI interpersonal sensitivity	58.5 (2.48)	9.97 (1.12)	60.41 (2.63)	9.93(1.08)	59.48 (2.56)	9.84 (1.08)
BSI depression	57.38 (2.57)	9.11 (1.13)	56.12 (2.51)	10.43 (1.15)	56.73 (2.54)	9.68 (1.12)
BSI anxiety	56.19 (2.44)	5.38 (0.72)	55.71 (2.35)	8.6(1.15)	55.94 (2.39)	7.11 (0.89)
BSI hostility	54.81 (1.45)	8.73 (1.06)	58.59 (1.85)	9.65(1.22)	56.76 (1.66)	9.27 (1.15)
BSI phobic anxiety	59.56 (2.12)	6.83 (1.01)	62.35 (2.62)	8.54(1.19)	61 (2.38)	7.77 (1.12)
BSI paranoid ideation	53.81 (1.58)	8.95 (1.04)	58.06 (2.04)	10.84 (1.31)	56 (1.82)	10.05 (1.19)
BSI psychoticism	55.5 (1.71)	6.35 (0.78)	57.94 (2.07)	11.92 (1.15)	56.76 (1.89)	9.56 (0.99)
Staff-rated measures						
Louis MACRO total	**16.97**	**1.32**	**18.08**	**1.42**	**17.6**	**1.47**
Louis MACRO safety	3.81	0.22	4.52	2.57	4.19	1.91
Louis MACRO physical care	3.75	0.22	3.84	0.29	3.8	0.26
Louis MACRO emotional care	**3.1**	**0.44**	**3.47**	**0.49**	**3.3**	**0.5**
Louis MACRO parenting	10.65	0.63	11.23	0.74	10.97	0.74
Louis MACRO infant characteristics	**3.41**	**0.4**	**3.59**	**0.52**	**3.51**	**0.47**
Louis MACRO mother's mental state	**3.11**	**0.41**	**3.35**	**0.65**	**3.24**	**0.56**
BPRS total	59.56	19.11	58.72	16.08	59.12	17.3
CGI severity	4.93	0.92	5	0.84	4.9	0.86

17 participants were assessed for BSI scores in the TAU group; 15 participants were assessed for Louis MACRO scores in the BTP + TAU group. Values in bold indicate significant difference at $p < 0.05$.

DISCUSSION

Feasibility, Acceptability and Satisfaction Indicators

This is the first feasibility trial to evaluate the feasibility and acceptability of a structured parenting intervention like Baby Triple P in mothers with severe mental health difficulties. The findings indicate that mothers who were MBU inpatients found the intervention and its delivery in this setting acceptable. In addition, the individually randomized trial design, including the randomization procedure, was found to be feasible to be scaled up to a fully powered RCT to evaluate the effectiveness and cost-effectiveness of BTP in this setting. However, recruitment was lower than anticipated due to lower MBU admissions so the optimal approach to recruitment may need to be revisited.

In terms of outcome measure suitability, the high rates of participants answering all items as well as the low number of items being omitted suggest that outcome measures were acceptable and user-friendly. Furthermore, no specific patterns of items being left out were identified which indicates that the questions raised in the measures were acceptable for this population. Overall, the excellent rate of data completion (of >95% across all three assessment points) could be an indicator of our patient-reported outcomes (i.e., self-reported measures) being suitable for gauging differences in a future trial. Completion of observer-rated measures by MBU staff was also excellent during participants' admission for the first assessment time point (94%). However, at subsequent assessment times MBU staff could not complete those measures consistently because participants had been discharged. In a future RCT, the possibility

TABLE 5 | Differences between groups after treatment.

Post-intervention (10 weeks after baseline; Time 2)	**BTP + TAU (*N* = 11)***		**TAU only (*n* = 8)**		**Adjusted mean difference**	**SE**	**BCa 95% CI**		**Cohen's *d***
	M	***SD***	***M***	***SD***					
MEQ	64.45	7.92	60.63	13.60	4.27	4.04	−2.52	10.54	0.38
DASS total	51.45	29.73	74.25	29.69	−24.55	13.77	−52.02	5.96	−0.83
DASS depression^s	14.55	12.36	22.25	12.85	−8.25	5.21	−17.38	1.04	−0.65
DASS anxiety^s	15.27	10.13	16.88	11.23	−4.09	5.28	−13.75	8.73	−0.38
DASS stress^s	19.82	10.45	29.38	9.40	−10.08	4.42	−18.16	−0.25	-1.02
PBQ total	20.27	23.44	29.00	27.21	−14.01	8.54	−32.20	4.39	−0.55
PBQ impaired bonding^s	9.45	10.96	13.63	13.06	−5.30	3.67	−13.90	2.44	−0.44
PBQ rejection and pathological anger^s	6.00	6.87	8.38	8.28	−3.53	2.16	−7.92	1.43	−0.46
PBQ infant focused anger^S	3.91	3.51	5.25	4.06	−1.12	1.57	−4.48	2.36	−0.29
PBQ Incipient abuse^s	0.91	3.02	1.50	3.51	0.71	0.62	−0.21	1.49	0.22
BSI global Severity Index (*t*-scores)	46.09	13.03	52.25	14.09	−7.32	4.48	−17.99	3.28	−0.73
BSI positive Symptom Total (*t*-scores)	47.55	13.70	53.88	14.82	−9.66	3.19	−16.71	−3.33	–0.92
BSI positive Symptom distress (t-scores)	42.73	10.21	50.75	12.65	−6.35	4.53	−14.30	2.65	−0.76
BSI somatisation^s	48.18	9.83	49.50	11.67	−3.70	4.90	−12.02	4.66	−0.47
BSI obsessive-compulsive^s	50.55	13.71	52.00	9.65	−3.23	5.61	−14.44	6.24	−0.36
BSI interpersonal^s sensitivity	49.55	10.43	57.75	9.93	−7.85	3.76	−14.90	−0.95	−1.04
BSI depression^s	44.73	13.03	52.25	14.26	−7.98	4.66	−17.13	−0.04	−0.80
BSI anxiety^s	43.91	11.11	49.25	11.35	−5.98	3.88	−13.59	2.33	−0.72
BSI hostility^s	46.00	10.42	52.50	10.72	−6.79	3.78	−14.51	0.81	−0.87
BSI phobic anxiety^s	50.91	11.71	56.50	14.02	−2.64	5.80	−11.45	4.57	−0.28
BSI paranoid ideation^s	46.55	10.43	48.00	12.31	−4.48	3.32	−11.16	3.49	−0.54
BSI Psychoticism^s	45.82	8.38	54.88	15.29	−9.87	2.87	−15.67	−4.11	−1.14
Clinical global impression	1.38	0.51	1.64	0.67	−0.23	0.25	−0.74	0.22	−0.48

Six-month Follow-up (Time 3)	**BTP + TAU (*N* = 10)**		**TAU only (*n* = 11)**		**Adjusted mean difference**	**SE**	**95% CI**		**Cohen's *d***
	M	***SD***	***M***	***SD***					
MEQ	64.90	11.32	61.80	9.77	2.38	0.18	0.36	0.98	0.22
DASS total	43.60	43.44	56.55	33.29	−18.09	16.28	−49.27	19.31	−0.47
DASS depression^s	12.30	12.91	16.45	13.57	−5.91	5.99	−16.78	4.39	−0.45

(Continued)

TABLE 5 | Continued

Six-month Follow-up (Time 3)	BTP + TAU (*N* = 10)		TAU only (*n* = 11)		Adjusted mean difference	SE	95% CI		Cohen's *d*
	M	***SD***	***M***	***SD***					
DASS anxiety[s]	10.20	10.56	14.64	12.98	−7.48	5.65	−18.07	2.25	−0.63
DASS stress[s]	15.20	13.41	19.00	11.51	−5.89	5.31	−15.39	4.71	−0.47
PBQ total	20.50	17.89	34.36	32.32	−16.23	10.48	−35.31	4.19	−0.61
PBQ impaired bonding[s]	9.60	8.06	16.00	14.62	−5.67	4.24	−14.03	3.00	−0.47
PBQ rejection and pathological anger[s]	5.10	4.98	9.91	9.54	−5.30	2.78	−11.69	1.39	−0.69
PBQ infant focused anger[s]	4.80	4.52	5.64	4.63	−0.05	2.06	−4.58	5.16	−0.01
PBQ incipient abuse[s]	1.00	2.83	2.82	4.62	0.50	0.53	−0.47	1.79	0.13
BSI global Severity Index (t-scores)	47.30	16.83	49.82	12.95	−4.36	5.38	−14.25	7.65	−0.29
BSI positive Symptom Total (t-scores)	48.10	15.79	50.82	12.64	−3.29	4.39	−11.03	5.47	−0.23
BSI positive Symptom distress (t-scores)	48.10	19.30	48.36	12.69	−5.60	6.13	−17.74	9.34	−0.35
BSI somatisation[s]	49.60	12.02	51.27	12.02	−2.94	5.42	−12.77	7.26	−0.24
BSI obsessive-compulsive[s]	52.30	11.13	51.00	10.99	0.32	5.16	−8.54	8.54	0.03
BSI interpersonal sensitivity[s]	48.60	15.06	52.36	14.07	−2.99	5.37	−12.79	6.88	−0.21
BSI depression[s]	45.20	16.72	47.27	11.12	−4.06	5.06	−13.84	7.08	−0.29
BSI anxiety[s]	42.90	12.26	48.18	10.42	−8.40	4.08	−17.02	1.42	−0.74
BSI hostility[s]	47.90	15.18	50.64	10.41	−4.16	4.54	−12.49	4.48	−0.32
BSI phobic anxiety[s]	51.60	14.21	57.09	13.85	−3.48	5.88	−14.51	6.92	−0.25
BSI paranoid ideation[s]	47.30	14.39	47.82	12.59	−2.30	5.03	−12.80	11.07	−0.17
BSI psychoticism[s]	44.70	12.46	50.64	13.60	−5.64	4.82	−15.44	6.53	−0.43

**One participant at post-intervention assessment remained in the study but only completed the EQ-5D.*

[s] *Indicates measure is a subscale. DASS stress difference at post-intervention significant at p = 0.04; BSI psychoticism difference significant at p = 0.02; BSI Positive symptom total significant at p = 0.01MEQ: Higher scores indicate higher maternal efficacy; DASS, PBQ AND BSI: lower scores indicate lower psychopathology; lower scores on the CGI indicate better mental health status.*

10 participants from the TAU only group completed MEQ measure at six-month follow-up assessment.

Cohen's d: 0.2 = small effect, 0.5 = medium effect, 0.8 = large effect (63).

TABLE 6 | Summary of EQ-5D data.

	Mean (95% CI)	
	BTP+TAU (*n* = 16)	TAU (*n* = 18)
Utility at Time 1	0.57 (0.48, 0.66) *n* = 16	0.55 (0.42, 0.68) *n* = 18
Utility at Time 2	0.70 (0.55, 0.85) *n* = 11	0.54 (0.26, 0.83) *n* = 8
Utility at Time 3	0.72 (0.55, 0.76) *n* = 10	0.63 (0.46, 0.79) *n* = 11
QALYs (baseline to week 26)	0.37 (0.31, 0.43) *n* = 8	0.27 (0.15, 0.40) *n* = 8
Net QALYs (95% CI)	0.10 (−0.02 to 0.22) *n* = 16	

TABLE 7 | Summary of costs for training and intervention delivery.

	Cost of delivering intervention*	Cost of training**	Total cost	Cost/ participant
Site 1 (*n* = 13)	£4,347	£1,418	£5,765	£ 443
Site 2 (*n* = 3)	£1,272	£1,193	£2,465	£ 822
Overall (*n* = 16)	£5,619	£2,611	£8,230	£ 514

**Total cost based on number of full hours of care provided (69 in Site 1, 24 in Site 2).*
***Number of hours of training (3 days at 7.5 per day = 22.5 h) x unit cost of trainer and trainee's time.*

of including observer-rated measures that can be administered by research staff should be considered alongside a stronger request for all self-reported measures to be completed during the follow-up appointments, even if online. Online or remote data collection has increased in use over the last 2 years to minimize infection risks; hence, tried and tested methods could be used in a future trial.

Although 100% of women admitted during the recruitment window were screened for study eligibility, the available pool to recruit from was lower than expected, potentially reflecting the early stages of the transformation of the perinatal mental health service provision in the UK (66). Furthermore, there was a difference in the recruitment rate between the two sites: the site with lower recruitment admitted more women with complex problems during the recruitment window and so they could not be included in the study. In addition, an increase in complex problems often meant longer admissions and a slower turnover of potential participants being admitted to that MBU. However, the diagnoses reported for this total sample are similar to those reported in previous MBU surveys in the UK (37, 67–69). Finally, although our inclusion criteria were relatively broad, women had to continue to reside on the ward for at least another week after expressing interest in the study to be eligible. Thus, only about 41% of all women admitted were eligible for study inclusion. Recruitment from more MBUs and possibly the inclusion of participants from recently developed perinatal CMHTs (66) would ensure better recruitment opportunities in future trials, potentially an even more diverse ethnic representation. The inclusion of participants who are not proficient in English should also be considered in any future study to test interventions for diverse groups of parents with mental health problems. In addition, the involvement of much younger mothers (≥16 years) could also be considered. As seen in this study, three mothers were eligible for study inclusion, but their relatively quick improvements meant they were discharged from the MBU much faster than originally anticipated by staff. If a future trial recruited from inpatient and outpatient perinatal mental health services, the timeframe of service use could be revised substantially, given the expectation that women discharged from MBUs are seen within relevant local perinatal mental health services.

Our intervention retention rate of 93% after the four core sessions of BTP was excellent and higher for our sample of mothers with severe mental health problems compared to a sample of mothers with postnatal depression [intervention retention rate of 85%; (30)] or a sample of non-psychiatric, healthy parents of premature babies [80%; (29)]. The completion of those crucial core Triple P sessions could be seen to constitute programme completion, because the remaining four sessions are designed for parents to practice their learnt parenting skills. Our retention rate of 75% at the end of the intervention was also higher compared to parents of premature babies [66%; (29)] and first-time, healthy mothers [40%; (28)], but not as promising as that of 85% for mothers with postnatal depression (30). Overall, our intervention attrition rate of 13% compares positively with attrition rates from Triple P interventions for parents of older children (>2 years) as attrition rates across several studies reached 19.5% [see meta-analysis by Nowak and Heinrichs (70)].

Clinical Outcome Indicators

Although this study was not powered to detect statistically significant differences between the treatment groups, we identified signals of effectiveness. At Time 2 following the intervention, greater improvements were noted for the BTP+TAU group than the TAU group on all of the measures. The biggest differences (i.e., large effect sizes) were for participant-reported depression, anxiety and stress (i.e., DASS-21 scores) and for positive symptom total scores on the BSI which measures psychiatric symptom presence and severity. At Time 3 the between group differences had attenuated for almost all measures except the PBQ which evaluates the mother-baby bond and relationship. Considering that there is evidence to suggest that early remission of postnatal psychiatric difficulties can mitigate the effects of maternal SMI on child development (71), the results from the present study could be worth exploring in a full RCT. These findings may also suggest that the expected mechanism of action of this parenting intervention in relation to strengthening the mother-infant relationship may be more evident over a longer period. Accurately capturing the changes in the parent-infant relationship across a longer time period, potentially without relying solely on self-report measures [e.g., (72)], would be crucial for any future trial.

Economic Outcome Indicators

The quality and completeness of data from the resource use and EQ-5D-5L questionnaires were good. There was a net QALY gain in the BTP+TAU group compared with the TAU only group, which may signal additional health benefits associated with BTP. This difference was not statistically significant, but the sample size was small and the study was not powered to detect differences in QALYs. The cost to deliver BTP was modest; however, it may be possible to explore whether BTP could be delivered over fewer sessions or by lower salaried professionals without reducing the fidelity of the intervention. The resource use questionnaire is well designed for data collection when administered in an interview format by field researchers as done here. It may be necessary to modify the design if a different mode of completion was used in a full RCT. A key driver of healthcare costs among the study participants was MBU readmission. Two participants from the TAU only group were readmitted to their MBUs which was associated with a cost of £26,973. None of the participants randomized to BTP+TAU was readmitted. If the addition of this parenting intervention to usual care was associated with a lower likelihood of readmission to a MBU this would be an important outcome. However, given the small number of participants included in this analysis, it is possible that this was observed by chance.

Challenges of Undertaking This Study in a MBU Setting and Limitations

Although acute psychiatric wards have been found to be complex and challenging environments for patients as well as staff, staff supported the consent, recruitment and other study procedures at both MBUs. The longer admissions for the women recruited to this study may indicate more complex mental health presentations, but it is important to note that those women allocated to receive the intervention engaged extremely well, especially during their inpatient admission. However, other challenges should be acknowledged. Two MBUs, comparable in terms of size and other relevant factors, were used to enhance the generalisability of the findings, but only one site almost met the anticipated recruitment rate whilst the other one did not. During this study admissions to the lower recruitment site varied and that MBU was not always operating at full capacity. Over the last few years MBUs in the UK have been part of a changing service landscape with the development of perinatal mental health services including community service provision. It is possible that those developments impacted one MBU more than the other.

The collection of staff-rated measures proved challenging after participants were discharged from the MBU and several measures could not be included. Some of these measures were chosen because they were routinely collected outcomes at both MBUs during the study planning phase (e.g., HONOS and LouisMACRO). For any future trial, observer-rated measures should be chosen carefully so that research staff could complete these. The transformation of perinatal mental health services in the UK may ensure that women will be referred to perinatal CMHTs following MBU-discharge which means that routine outcome measures used within these services could be considered and the woman's care coordinator could be asked to assist with the completion of measures. It would also be sensible to extend the trial to include such services in order to omit the exclusion criterion of ward presence for a period of seven days following expressing an interest in the study; we had to exclude three participants for this reason. Furthermore, due to its established training structure, the intervention can easily be offered by various members of an inpatient or outpatient perinatal mental health team, including nursery nurses. BTP was developed for the infant's parents so delivery does not have be restricted to the mother but can successfully be offered to fathers as well as other significant others in the case of single parents. As many countries may have different criteria for the admission of a mother to a psychiatric unit, sometimes without the ability for a joint mother and baby admission, it would be useful to explore the benefits of this type of parenting interventions in other countries as an adjust to the mother's usual mental health care. Finally, the inclusion of a video-based assessment of the parent-infant interaction and relationship should be considered to assess potential attachment behaviors shown by the infant and to have a more objective measure of the parent-child interaction, rated by trained and accredited coders. The intervention clearly supported mothers bonding with their infants and this aspect warrants further investigation, especially in relation to the infant's psychological development and mental wellbeing.

When interpreting the current findings, some limitations need to be considered. The study sample size was relatively small and admission rates to both MBUs were lower than anticipated. Uptake and engagement rates of mothers need to be explored further in an outpatient mental health setting. Whilst no contamination issues were reported by participants or observed by MBU staff, this possibility remains. However, as the prevalence of maternal mental illness is reportedly rising (73), there is a need to explore the benefits of a range of interventions that can improve the mental health of mothers as well as their bonding with their infants.

Conclusions

This preliminary study indicates that a parenting intervention like Baby Triple P can be delivered and implemented successfully in acute psychiatric inpatient settings like a Mother and Baby Unit to service users who appeared to benefit from what the intervention has to offer to them and their infants. We identified that this intervention and our study procedures could be delivered safely. There was good retention in both treatment groups and an exceptionally high level of completion of self-report outcome measures. Some modifications may need to be made to the collection of observer-rated outcomes but overall the findings suggest that the study procedures are feasible for a future large-scale trial of a structured parenting intervention that would complement other therapeutic approaches perfectly and could easily be implemented into existing perinatal mental health services.

DATA AVAILABILITY STATEMENT

The raw data can be made available by the authors on request.

ETHICS STATEMENT

This trial was supported by the NIHR Research for Patient Benefit Programme (NIHR RfPB, grant number PB-PG-1014-3505) and sponsored by the University of Manchester. Study approvals were granted by the NHS National Research Ethics Service (NRES) *via* the Northwest–Greater Manchester South Research Ethics Committee (REC) (16/NW/0510), the Health Research Authority (HRA) (IRAS project number 188486, protocol number 16233) and the Research and Innovation departments of both NHS trusts overseeing the two participating MBUs (e.g., Greater Manchester Mental Health NHS Foundation Trust and Birmingham and Solihull Mental Health NHS Foundation Trust). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

AW conceived of the idea and developed it into a proposal with the help of RC, RE, PB, EC, and KA. RE led on the statistical analysis and EC on the health economic evaluation. KC and PD supported trial coordination, while HR and PG acted as research assistants, involved in data collection or data analysis. All authors contributed to the development of this manuscript and approved its publication.

FUNDING

This feasibility trial represents independent research, which was funded by the National Institute for Health Research (NIHR) under its Research for Patient Benefit (RfPB) Programme (PB-PG-1014-35059). The work was also supported by the Greater Manchester Mental Health NHS Foundation Trust (GMMH) and the Birmingham and Solihull Mental Health NHS Foundation Trust (BSMH). The Open Access publication was made possible by the support of the University of Manchester. RE is supported by the NIHR Biomedical Research Centre at South London and Maudsley NHS Foundation Trust and King's College London and an NIHR Research Professorship (NIHR300051).

ACKNOWLEDGMENTS

We would like to express our sincere gratitude and thanks to the women who took part in this study and our service user reference group whose ideas and suggestions we valued highly and who offered us their ongoing support and advice throughout the study. We are also grateful for the support offered by Brigid Corrigan and Catherine Cross from GMMH and to collaborators at BSMH, namely Claire Hooper, Mary McGuinness, Giles Berrisford and Linda Everard. Thank you also to Rangeni Zinyama, Nadia Starkova and Blair Davis from BSMH and to Grace Bamber from GMMH for supporting the data collection. Finally, we greatly appreciated the support from the Manchester CTU and in particular Tracey Rose as well as various members of our Trial Steering Committee, chaired by Andrew Gumley, as well as the assistance offered by Gokce Cokamay Yilmaz and Zoyah Sheikh from GMMH's Perinatal Mental Health and Parenting (PRIME) Research Unit in finalizing this manuscript.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.815018/full#supplementary-material

REFERENCES

1. Howard LM. Khalifeh H. Perinatal mental health: a review of progress and challenges. *World Psychiatry.* (2020) 19:3. doi: 10.1002/wps.20769
2. National Institute for Health and Clinical Excellence. *NICE Clinical Guideline 192: Antenatal and Postnatal Mental Health: Clinical Management and Service Guidance*. (2014). Available from: https://www.nice.org.uk/guidance/cg192
3. National Institute for Health and Clinical Excellence. *NICE Clinical Guideline 192: Antenatal and Postnatal Mental Health: Clinical Management and Service Guidance*. (2020). Available from: https://www.nice.org.uk/guidance/cg192.
4. Hipwell AE, Goossens FA, Melhuish EC, Kumar R. Severe maternal psychopathology and infant–mother attachment. *Dev Psychopathol.* (2000) 12:157–75. doi: 10.1017/S0954579400002030
5. Oyserman D, Mowbray CT, Meares PA, Firminger KB. Parenting among mothers with a serious mental illness. *Am J Orthopsychiatry.* (2000) 70:296–315. doi: 10.1037/h0087733
6. Grace SL, Evindar A, Stewart DE. The effect of postpartum depression on child cognitive development and behavior: a review and critical analysis of the literature. *Arch Womens Ment Health.* (2003) 6:263–74. doi: 10.1007/s00737-003-0024-6
7. Hay DF, Pawlby S. Prosocial development in relation to children's and mothers' psychological problems. *Child Dev.* (2003) 74:1314–27. doi: 10.1111/1467-8624.00609
8. Goodman SH, Rouse MH, Connell AM, Broth MR, Hall CM, Heyward D. maternal depression and child psychopathology: a meta-analytic review. *Clin Child Fam Psychol Rev.* (2011) 14:1–27. doi: 10.1007/s10567-010-0080-1
9. Fraser A, Macdonald-Wallis C, Tilling K, Boyd A, Golding J, Smith GD, et al. Cohort profile: the avon longitudinal study of parents and children: ALSPAC mothers cohort. *Int J Epidemiol.* (2013) 42:97–110. doi: 10.1093/ije/dys066
10. Stein A, Pearson RM, Goodman SH, Rapa E, Rahman A, McCallum M, et al. Effects of perinatal mental disorders on the fetus and child. *Lancet.* (2014) 384:1800–19. doi: 10.1016/S0140-6736(14)61277-0
11. Wan MW, Appleby L, Webb R, Abel KM. What Predicts poor mother-infant interaction in schizophrenia. *Psychol Med.* (2007) 37:537–46. doi: 10.1017/S0033291706009172
12. Murray L, Halligan S, Cooper P. Effects of postnatal depression on mother-infant interactions, and child development. In: Bremner JG, Wachs TD, editors. *The Wiley- Blackwell Handbook of Infant Development. Volume II: Applied and Policy Issues*. Hoboken: Wiley (2010). p. 192–220. doi: 10.1002/9781444327588.ch8

13. Gillham R, Wittkowski A. Outcomes for women admitted to mother and baby unit: a systematic review. *Int J Womens Health.* (2015) 7:459–76. doi: 10.2147/IJWH.S69472
14. *The Marmot Review: The Fair Society, Healthy Lives*. (2010). Available at: https://www.instituteofhealthequity.org/resources-reports/fair-society-healthy-lives-the-marmot-review/fair-society-healthy-lives-full-report-pdf.pdf (accessed September 8, 2021).
15. Allen G. *Early Intervention: The Next Steps, An Independent Report to Her Majesty's Government by Graham Allen MP*. London: The Stationery Office (2011).
16. Munro E. *The Munro Review of Child Protection: Final Report, a Child-Centred System*. London: The Stationery Office (2011).
17. *National Institute for Health and Care Excellence, ICE Public Health Guidance 40: Social and Emotional Wellbeing: Early Years*. (2012). Available from: https://www.nice.org.uk/guidance/ph40/resources/social-and-emotional-wellbeing-early-years-pdf-1996351221445 (accessed September 8, 2021).
18. Leadsom A, Field F, Burstow P, Lucas C. *The 1001 Critical Days: The Importance of the Conception to Age Two Period*. London: A Cross Party Manifesto (2013).
19. Bauer A, Parsonage M, Knapp M, Iemmi V, Adelaja B. *The Costs of Perinatal Mental Health Problems*. London: Centre for Mental Health (2014).
20. Sanders MR. Triple P-positive parenting program: towards an empirically validated multilevel parenting and family support strategy for the prevention of behavior and emotional problems in children. *Clin Child Fam Psych.* (1999) 2:2.
21. Sanders MR, Markie-Dadds C, Turner KMT. *Theoretical, Scientific and Clinical Foundations of the Triple P- Positive Parenting Program: A Population Approach to the Promotion of Parenting Competence*. Queensland: The Parenting and Family Support Centre (2003) 1–24.
22. Turner KMT, Sanders MR. Dissemination of evidence-based parenting and family support strategies: learning from the triple P–positive parenting program system approach. *Aggress Violent Behav.* (2006) 11:176–93. doi: 10.1016/j.avb.2005.07.005
23. Sanders MR. Development, evaluation and multinational dissemination of triple P-positive parenting program. *Annu Rev Clin Psychol.* (2012) 8:345–79. doi: 10.1146/annurev-clinpsy-032511-143104
24. Sanders MR, Kirby JN, Tellegen CL, Day JJ. The triple P-positive parenting program: a systematic review and meta-analysis of a multi-level system of parenting support. *Clin Psychol Rev.* (2014) 34:337–57. doi: 10.1016/j.cpr.2014.04.003
25. Li N, Peng J, Li Y. Effects and moderators of triple p on the social, emotional, and behavioral problems of children: systematic review and meta-analysis. *Front Psychol.* (2021) 12:709851. doi: 10.3389/fpsyg.2021.709851
26. Spry C, Morawska A, Sanders MR. *Baby Triple P Group Workbook*. Milton, Australia: Triple P International Pty Ltd (2011).
27. Spry C. *The Baby Triple P Project: Effects of a Parenting Intervention to Promote a Successful Transition to Parenthood.* (PhD thesis). (Queensland (Australia)).: The University of Queensland (2013).
28. Popp L, Fuths S, Schneider S. The relevance of infant outcome measures: a pilot-RCT comparing baby triple P positive parenting program with care as usual. *Front Psychol.* (2019) 10:2425. doi: 10.3389/fpsyg.2019.02425
29. Colditz PB, Boyd RN, Winter L, Pritchard M, Gray PH, Whittingham K, et al. A randomized trial of baby triple P for preterm infants: child outcomes at 2 years of corrected age. *J Pediatr.* (2019) 210:48–54. doi: 10.1016/j.jpeds.2019.01.024
30. Tsivos ZL, Calam R, Sanders MR, Wittkowski A. Interventions for postnatal depression assessing the mother–infant relationship and child developmental outcomes: a systematic review. *Int J Womens Health.* (2015) 7:429–47. doi: 10.2147/IJWH.S75311
31. Butler H, Walker S, Hare DJ, Wieck A, Wittkowski A. The acceptability and feasibility of the baby triple p positive parenting programme on a mother and baby unit: Q-methodology with mothers with severe mental illness. *Arch Womens Ment Health.* (2014) 17:455–63. doi: 10.1007/s00737-014-0429-4
32. Butler-Coyne H, Walker S, Hare DJ, Wieck A, Wittkowski A. Acceptability of a positive parenting programme on a mother and baby unit: Q-methodology with staff. *J Child Fam Stud.* (2017) 26:623–32. doi: 10.1007/s10826-016-0564-9
33. Wittkowski A, Cartwright K, Emsley R, Bee P, Calam R, Cross C, et al. Enhancing maternal and infant wellbeing: study protocol for a feasibility trial of the baby triple P positive parenting programme for mothers with severe mental health difficulties (the IMAGINE study). *Trials.* (2018) 19:1–15. doi: 10.1186/s13063-018-2869-z
34. Wittkowski A, Duxbury P, Bee P, Calam R, Emsley R, Camacho E, et al. The acceptability of a parenting intervention to mothers with severe mental health problems and staff in a mother and baby unit setting: The IMAgINE study (in preparation).
35. McPherson KE, Schroeter B. Promoting program fidelity through peer-assisted supervision and support. In: Sanders MR, Mazzucchelli TG, editors. *The Power of Positive Parenting: Transforming the Lives of Children, Parents, and Communities Using the Triple P System*. Oxford University Press (2018) p. 405–15 doi: 10.1093/med-psych/9780190629069.003.0037
36. Owens CR, Haskett M, Norwalk K. Peer assisted supervision and support and providers' use of triple P-positive parenting program. *J Child Fam Stud.* (2019) 28:1664–72. doi: 10.1007/s10826-019-01385-w
37. Wittkowski A, Santos N. Psychological and psychosocial interventions promoting the mother-infant interaction on mother and baby units in the United Kingdom. *JSM Anxiety Depress.* (2017) 2:1022.
38. Garrett C, Turner B, Wittkowski A. Psychological and psychosocial interventions for improving the mental health of women admitted to mother and baby units in the United Kingdom. *Womens Health Res.* (2018) 2:29–41. doi: 10.1057/whr0000009
39. Sanders MR, Markie-Dadds C, Turner KMT. *Practitioners Manual for Standard Triple P*. Brisbane: Families International Publishing (2001).
40. Sanders MR, Morawska A. *Family Background Questionnaire*. Brisbane: Parenting and Family Support Centre. (2010). doi: 10.1037/t69619-000
41. Webster J, Linnane JWJ, Dibley LM, Hinson JK, Starrenburg SE, Roberts JA. Measuring social support in pregnancy: can it be simple and meaningful? *Birth.* (2000) 27:97–101. doi: 10.1046/j.1523-536x.2000.00097.x
42. Teti DM, Gelfand DM. Behavioral competence among mothers of infants in the first year: the mediational role of maternal self-efficacy. *Child Dev.* (1991) 62:918–29. doi: 10.1111/j.1467-8624.1991.tb01580.x
43. Lovibond PF, Lovibond SH. The structure of negative emotional states: comparison of the Depression Anxiety Stress Scales (DASS) with the beck depression and anxiety inventories. *Behav Res Ther.* (1995) 33:335–43r. doi: 10.1016/0005-7967(94)00075-U
44. Henry JD, Crawford JR. The short-form version of the Depression Anxiety Stress Scales (DASS-21): construct validity and normative data in a large non-clinical sample. *Br J Clin Psychol.* (2005) 44:227–39. doi: 10.1348/014466505X29657
45. Gomez F. *A Guide to the Depression, Anxiety and Stress Scale (DASS 21).* Sydney: Central and Eastern Sydney Primary Health Networks (2016).
46. Derogatis LR, Melisaratos N. The brief symptom inventory: an introductory report. *Psychol Med.* (1983) 13:595–605. doi: 10.1017/S0033291700048017
47. Brockington I, Oates J, George S, Turner D, Vostanis P, Sullivan M, et al. A screening questionnaire for mother-infant bonding disorders. *Arch Womens Ment Health.* (2001) 3:133–40. doi: 10.1007/s007370170010
48. Brockington IF, Fraser C, Wilson D. The postpartum bonding questionnaire: a validation. *Arch Womens Ment Health.* (2006) 9:233–42. doi: 10.1007/s00737-006-0132-1
49. Herdman M, Gudex C, Lloyd A, Janssen MF, Kind P, Parkin D, et al. Development and preliminary testing of the new five-level version of EQ-5D (EQ-5D-5L). *Qual Life Res.* (2011) 20:1727–36. doi: 10.1007/s11136-011-9903-x
50. Van Hout B, Janssen MF, Feng YS, Kohlmann T, Busschbach J, Golicki D, et al. Interim scoring for the EQ-5D-5L: mapping the EQ-5D-5L to EQ-5D-3L value sets. *Value Health.* (2012) 15:708–15. doi: 10.1016/j.jval.2012.02.008
51. *National Institute for Health and Care Excellence, EuroQoL group: Position Statement on Use of the EQ-5D-5L Value Set for England*. (2019). Available at: https://www.nice.org.uk/about/what-we-do/our-programmes/nice-guidance/technology-appraisal-guidance/eq-5d-5l (accessed September 8, 2021).

52. Guy W. Clinical Global Impression Scale (CGI). *ECDEU Assessment Manual for Psychopharmacology - US Department of Health, Education, and Welfare Publication*. Rockville: National Institute of Mental Health (1976).
53. Lukoff D, Nuechterlein KH, Ventura J. Manual for the expanded brief psychiatric rating scale. *Schizophrenia Bull.* (1986) 12:594–602. doi: 10.1093/schbul/12.4.578
54. Leucht S, Kane JM, Kissling W, Hamann J, Etschel EV, Engel R. Clinical implications of brief psychiatric rating scale scores. *Br J Psychiat.* (2005) 187:366–71. doi: 10.1192/bjp.187.4.366
55. Louis A, Condon J, Shute R, Elzinga R. The development of the Louis MACRO (Mother and Child Risk Observation) forms: assessing parent-infant-child risk in the presence of maternal mental illness. *Child Abuse Neglect.* (1997) 21:589–606. doi: 10.1016/S0145-2134(97)00027-6
56. StataCorp. *Stata Statistical Software: Release 15*. College Station, TX: StataCorp LLC (2017).
57. IBM Corp. *Released 2015. IBM SPSS Statistics for Windows, Version 23.0*. Armonk, NY: IBM Corp.
58. Eldridge SM, Chan CL, Campbell MJ, Bond CM, Hopewell S, Thabane L, et al. CONSORT 2010 statement: extension to randomised pilot and feasibility trials. *BMJ.* (2016) 355:i5239. doi: 10.1136/bmj.i5239
59. Curtis LA, Burns A. *Unit Costs of Health and Social Care.* Kent: Personal Social Services Research Unit. (2018).
60. Wing JK, Beevor AS, Curtis RH, Park SB, Hadden S, Burns A. HoNOS: Health of the Nation Outcome Scales. Research and development. *Brit J Psychiat.* (1998) 172:11–8. doi: 10.1192/bjp.172.1.11
61. Handal PJ, Gist D, Gilner FH, Searight HR. Preliminary validity for the Langner symptom survey and the brief symptom inventory as mass-screening instruments for adolescent adjustment. *J Clin Child Psychol.* (1993) 22:382–6. doi: 10.1207/s15374424jccp2203_9
62. Dawson G, Frey K, Panagiotides H, Yamada E, Hessl D, Osterling J. Infants of depressed mothers exhibit atypical frontal electrical brain activity during interactions with mother and with a familiar, nondepressed adult. *Child Dev.* (1999) 70:1058–66. doi: 10.1111/1467-8624.00078
63. Coe, R. *It's the Effect Size, Stupid: What Effect Size is and Why it is Important (Conference presentation).. British Educational Research Association Annual Conference, Exeter, United Kingdom*. (2002). Available at: https://f.hubspotusercontent30.net/hubfs/5191137/attachments/ebe/ESguide.pdf
64. Jacobson NS, Truax P. Clinical significance: a statistical approach to defining meaningful change in psychotherapy research. *J Consult Clin Psych.* (1991) 59:631–48. doi: 10.1037/0022-006X.59.1.12
65. Department of Health (DH). *Archived Reference Costs*. (2020). Available from: https://webarchive.nationalarchives.gov.uk/ukgwa/20200501111106/https://improvement.nhs.uk/resources/reference-costs/ (accessed October 21, 2021).
66. National Collaborating Centre for Mental Health. *The Perinatal Mental Health Care Pathways. Full Implementation Guidance*. London: National Collaborating Centre for Mental Health (2018).
67. Wittkowski A, Harvey J, Wieck A, A. Survey of instruments used for the psychological assessment of patients admitted to mother and baby inpatient facilities. *Arch Womens Ment Health.* (2007) 10:237–40. doi: 10.1007/s00737-007-0195-7
68. Elkin A, Gilburt H, Slade M, Lloyd-Evans B, Gregoire A, Johnson S, et al. A national survey of psychiatric mother and baby units in England. *Psychiat Serv.* (2009) 60:629–33. doi: 10.1176/ps.2009.60.5.629
69. Turner B, Garrett C, Wittkowski A. Male partners of women admitted to mother and baby units in the united kingdom: a survey of available psychosocial and psychological interventions. *Womens Health Res.* (2017) 1:25–36. doi: 10.1057/whr0000004
70. Nowak C, Heinrichs NA. Comprehensive meta-analysis of triple p-positive parenting program using hierarchical linear modeling: effectiveness and moderating variables. *Clin Child Fam Psychol Rev.* (2008) 11:114r. doi: 10.1007/s10567-008-0033-0
71. Balbernie R. Circuits and circumstances: the neurobiological consequences of early relationship experiences and how they shape later behaviour. *J Child Psychother.* (2001) 27:237–55. doi: 10.1080/00754170110087531
72. Wittkowski A, Vatter S, Muhinyi A, Garrett C, Henderson M. Measuring bonding or attachment in the parent-infant-relationship: a systematic review of parent-report assessment measures, their psychometric properties and clinical utility. *Clin Psychol Rev.* (2020) 82:101906r. doi: 10.1016/j.cpr.2020.101906
73. Abel KM, Hope H, Swift E, Parisi R, Ashcroft DM, Kosidou K, et al. Prevalence of maternal mental illness among children and adolescents in the UK between 2005 and 2017: a national retrospective cohort analysis. *Lancet Public Health.* (2019) 4:e291–300. doi: 10.1016/S2468-2667(19)30059-3

Author Disclaimer: The views expressed are those of the authors and not necessarily those of the NHS, the NIHR or the Department of Health.

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

METHODS
published: 15 June 2022
doi: 10.3389/fpsyt.2022.805781

Virtual Community Engagement Studio (V-CES): Engaging Mothers With Mental Health and Substance Use Conditions in Research

Yaara Zisman-Ilani[1,2]*, Jennifer Buell[3], Shayna Mazel[3], Shannon Hennig[4] and Joanne Nicholson[3]

[1] Department of Social and Behavioral Sciences, College of Public Health, Temple University, Philadelphia, PA, United States, [2] Department of Clinical, Educational and Health Psychology, Division of Psychology and Language Sciences, University College London, London, United Kingdom, [3] Institute for Behavioral Health Schneider Institutes for Health Policy, Heller School for Social Policy and Management, Brandeis University, Waltham, MA, United States, [4] Maternal Mental Health Research Collaborative, Calgary, AB, Canada

Active engagement of community stakeholders is increasingly encouraged in behavioral health research, often described as a co-production approach. Community stakeholders (e.g., patients, providers, policy makers, advocates) play a leading role together with research investigators in conducting the various phases of research, including conceptualization, design, implementation, and the interpretation and dissemination of findings. The concept of co-production has promising benefits for both the target population and the research outcomes, such as producing person-centered interventions with greater acceptability and usability potential. However, it is often the case that neither researchers nor community members are trained or skilled in co-production methods. The field of behavioral health research lacks tools and methods to guide and promote the engagement of diverse stakeholders in the research process. The purpose of this methods paper is to describe the Virtual Community Engagement Studio (V-CES) as a new method for engaging vulnerable populations like mothers with mental health and substance use conditions in research. We piloted the method in collaboration with the Maternal Mental Health Research Collaborative (MMHRC), focusing on one of the most vulnerable, under-researched populations, mothers coping with mental health and/or substance abuse disorders. Our pilot included mothers and providers who work with them as Community Experts to inform all phases of research design and implementation, and the interpretation and application of findings. The aim of this article is to describe the V-CES as a powerful tool that supports the engagement of mothers with mental health and/or substance use disorders and other community stakeholders in research, to provide examples of its use, and to make recommendations for future use, based on lessons learned. The V-CES toolkit is available for use with this target population as well as others.

Keywords: community engagement, co-production, parents with mental illness, mothers, mental health, substance use disorder

OPEN ACCESS

Edited by:
Bolanle Adeyemi Ola,
Lagos State University, Nigeria

Reviewed by:
Domenico De Berardis,
Mental Health Center (CSM) and Psychiatric Service of Diagnosis and Treatment (SPDC), Italy
Anna Franceschini,
Public Health Unit, Italy
Julia Elena Marquez-Arrico,
University of Barcelona, Spain

***Correspondence:**
Yaara Zisman-Ilani
yaara@temple.edu

Specialty section:
This article was submitted to Addictive Disorders, a section of the journal Frontiers in Psychiatry

Received: 30 October 2021
Accepted: 20 May 2022
Published: 15 June 2022

Citation:
Zisman-Ilani Y, Buell J, Mazel S, Hennig S and Nicholson J (2022) Virtual Community Engagement Studio (V-CES): Engaging Mothers With Mental Health and Substance Use Conditions in Research. Front. Psychiatry 13:805781. doi: 10.3389/fpsyt.2022.805781

INTRODUCTION

The onset of COVID-19 and its emotional, social and psychological implications, particularly for mothers of childbearing age (e.g., social isolation, working at home with children, difficulty accessing treatment, loss of employment) have been found to be associated with a surge in use of substances (e.g., opioids, cannabis, alcohol), anxiety and depression (1, 2). Mothers of childbearing age with pre-existing conditions of mental illness are at a particular higher risk for developing a substance use comorbidity (3, 4). Unfortunately, this vulnerable group of women is often less engaged in mental health services and substance use treatment programs, and even less engaged in research projects that presumably target their needs and challenges (5, 6). The low rates of engagement in research among this group directly affects the quality of the services and care they receive, as new interventions and programs are being developed without the critical input of this vulnerable group of potential end-users or beneficiaries.

Co-production or co-design approaches are offered as processes for tailoring interventions and treatment programs, making them more relevant to the lived experience of the target population and, as a result, potentially increasing service/treatment engagement and effectiveness (7, 8). In a co-production process, community stakeholders (e.g., patients, providers, policy makers, advocates) and research investigators partner in conducting the various phases of research, including conceptualization, design, implementation, and dissemination. A co-production approach can potentially lead to more meaningful and impactful programs, interventions and outcomes for both patients and researchers (9). Co-production is also associated with benefits to researchers, such as enhanced acceptability and feasibility of methods and procedures, enhanced relevance of outcomes in terms of meaning and impact for patients, and enhanced sustainability of interventions (10, 11). Similar to other engagement approaches, such as shared decision making (12, 13), benefits for patients/participants in co-production include improved quality of care and outcomes, and feeling valued and empowered by sharing their experiences and expertise on behalf of "a greater good" – that is, informing research and practice approaches that may improve the lives of others. Patients/community stakeholders may also benefit from the sense of mastery that emerges from being "in the driver's seat," not just as passive patients or research subjects, but as significant decision-makers (10).

Despite the promising potential, the use and implementation of co-production in practice is limited (14). Researchers may be challenged in engaging community members (e.g., patients, providers, policy makers, and advocates) as partners in research endeavors rather than, or in addition to, as participants (15). Many researchers are not trained or skilled in identifying, recruiting, convening and engaging community stakeholders or preparing them for participation in research in an advisory capacity or as contributing members of a research team. At the same time, mothers with mental or substance use disorders may not feel comfortable participating, may be distrusting of researchers, or may have concerns that there could be possible legal or social services consequences to their involvement, due to stigma and the sensitive nature of maternal mental illness and substance use (16).

The COVID-19 pandemic has given rise to the rapid digitalization of remote mental health via telemedicine and digital psychiatry approaches (17, 18). Attention is also being given to the development of virtual research methods, with the Patient-Centered Outcomes Research Institute (PCORI) in the United States soliciting projects via its Eugene Washington Engagement Award program to develop and enhance community engaged research approaches in the virtual era imposed by COVID-19 pandemic. This article describes the results of a PCORI engagement award to develop the Virtual Community Engagement Studio (V-CES) method for virtually engaging this vulnerable target population, mothers of childbearing age with mental health and/or substance use disorder, across the research life cycle. We will describe the V-CES method and its application including feedback from participants, lessons learned, and recommendations.

MATERIALS AND EQUIPMENT

V-CES Background

The V-CES was developed based on the community engagement studio (CES) model (19–21) as an interactive method to facilitate co-production in behavioral health research with vulnerable populations, in our case, mothers with mental health and/or substance use disorders. The end goal was to engage community stakeholders as experts with researchers in various steps of behavioral health research to provide input to inform and improve recruitment procedures, data collection, ethical considerations, the choice of outcomes, and the interpretation and dissemination of findings. The V-CES is inspired by the CES approach that focuses on supporting the involvement of community members with researchers to inform next steps in research and treatment innovation (19, 20). The original CES was developed by Joosten and colleagues at Vanderbilt University to recruit and train stakeholders and prepare researchers to participate in an in-person meeting (19). The V-CES and the original in-person CES model describe research efforts with people rather than on people, similar to existing community participatory based research (CBPR) or Cooperative Inquiry (CI) approaches. Since we adapted the CES method during and in response to the COVID-19 pandemic, the community engagement studio takes place in a virtual/digital medium (i.e., an online video conference space), which requires consideration of the differences from engaging with community stakeholders in-person. Conducting the CES virtually provides opportunities to facilitate and broaden the engagement of diverse mothers (e.g., race, ethnicity, disability) in diverse geographic areas (e.g., rural, urban), time zones, and living situations (e.g., alone or with extended family), with diverse responsibilities (e.g., caring for young children at home) that impact on scheduling (e.g., during school hours or after school) with researchers from different institutes or universities, states or countries, and areas of research.

V-CES Structure

The V-CES method includes two components: The core V-CES team, which is at the center of the V-CES activity, and the operational team, working behind the scenes, that administratively supports the operation and execution of the V-CES. Importantly, both components reflect a commitment to a partnership between the researcher and the target population (i.e., mothers with mental and/or substance use disorder). The core V-CES team is composed of three types of members: researchers, Community Experts, and Facilitators. The V-CES is implemented by the operational team that includes a Community Navigator, Science Navigator, and a Manager. The operational team is responsible for implementing the V-CES, provides coaching and support to both researchers and Community Experts, and manages administrative aspects such as logistics and resource preparation, including video conference platform, V-CES recruitment, and the solicitation of feedback from V-CES participants.

The V-CES Core Team

Researchers

The researcher identifies a theme or topic area and prepares a short presentation to discuss with the V-CES team in preparation for the V-CES. The V-CES team recommends changes to the presentation as needed to improve clarity and ensure the language and tone are appropriate and sensitive to the Community Experts' characteristics and experiences. Researchers are guided to avoid jargon, technical terms and acronyms and encouraged to use plain language. The researcher's opening V-CES presentation serves to elicit feedback from the Community Experts on how best to move forward with a research project. After making the presentation, the researcher's role is primarily to listen, asking and answering questions for clarification. Participants may want to know, for example, why a researcher is interested in a particular topic.

Community Experts

Community members, be they mothers, family members, peer specialists, service providers or advocates, are considered experts by experience and are the key to the success of the V-CES. Ideally, they represent diverse backgrounds and are connected to the community in various ways. For example, a care manager or peer specialist who works with mothers in metal health/substance use treatment may have a very different, but equally valuable, perspective from a mother currently in recovery with a similar condition. Generally, Community Experts should have good verbal communication and listening skills, a desire to learn about research, and a willingness to share their experiences. Accommodations can be made to support the engagement of participants whose skills may be compromised by a health condition or disability.

Facilitator

As recommended by Joosten et al. (19) the Facilitator's task is to create a comfortable, safe environment that allows for open and frank discussion and to guide the conversation among researchers and Community Experts. A skilled Facilitator does not interject their opinions or biases into the conversation. The Facilitator should have professional and/or lived experience working within the target population community and possess the ability to balance the differences in power that can naturally occur when researchers and community members come together. The Facilitator's responsibilities include explaining (and keeping) discussion ground rules (e.g., be concise, don't interrupt, and maintain confidentiality), keeping the discussion on track, using the predefined questions as the discussion framework, and guiding the discussion, only interjecting their own opinion and personal observations with intention and purpose.

The V-CES Operational Team

Community Navigator

A boundary spanner with familiarity with the target community, experience with academic-community partnerships, and understanding of principles of community engagement is a good candidate for the Community Navigator role. The navigator should have experience building rapport and trusting relationships with key community leaders. Specific responsibilities include helping to identify, orient, and support Community Experts who participate in the V-CES; coaching the researchers on communicating with Community Experts with personal or professional experience with maternal mental health and/or substance use; and developing and maintaining mechanisms to communicate with community partners, increase interaction between community partners and researchers, and track the development of research-community partnerships. Hiring a Community Navigator from the community puts into practice fundamental principles of community engagement such as mutual benefit, respect and community capacity building (19). A respected community member is likely to have access to networks unfamiliar to someone who works in an academic/research setting.

Science Navigator

The Science Navigator provides guidance on identifying and recruiting participating researchers, and coaches them on communicating effectively with non-researchers and engaging Community Experts as consultants, rather than as research subjects (19). The Science Navigator benefits from having experience in patient-centered outcomes research, community-engagement, comparative effectiveness and community-based participatory research (19). Specific responsibilities include helping to identify, orient, and support researchers who participate in the V-CES; coaching the researchers on communicating effectively with non-researchers and on engaging Community Experts as consultants; and encouraging Researchers to consult Community Experts who would like to remain involved as the research project develops.

Manager

The Manager works with the Community Navigator and Science Navigator to reach out to selected researchers and Community Experts, securing the time and access code to virtual sessions (e.g., via ZOOM, SKYPE, TEAMS) for the V-CES, and preparing necessary materials to assist, plan, and implement the V-CES. Specific responsibilities include managing logistics such as securing a virtual space and time for the V-CES that are convenient for the Community Experts; making sure the appropriate documentation is completed for each V-CES, including capturing the Community Expert feedback from each session; and the completion of evaluation surveys and forms needed to process payments or stipends for participation.

METHODS

Step by step procedures for implementing a V-CES are described below, using examples from the application of the V-CES method with researchers and the target population of mothers with mental and/or substance use disorders and providers who work with them in the community. We implemented four V-CES's during the COVID-19 pandemic with a total of 19 Community Experts (i.e., mothers and providers). The first two sessions were conducted in April 2020 and included 16 participants: researcher, facilitator, manager and 13 Community Expert, White women, ages 25 to 45, from four states in the US. A third V-CES was held in March 2021 and included 6 participants: researcher, facilitator, manager and 3 Community Experts, White women from the Massachusetts area. A fourth V-CES was conducted in May 2021 with 8 participants: researcher, facilitator, manager and 5 Community Experts, White women, one identifying as Hispanic/Latina, ages 25 to 55 from six states in the US. The V-CES procedures were reviewed by the Brandeis University Institutional Review Board and deemed to be exempt from consideration as Human Subjects Research. Community Experts received gift cards in the amount of $150 US.

Recruitment

Recruitment of Researchers

The Operational Team was responsible for the recruitment process. The V-CES Manager centralized the recruiting process with the help of the Community and Science Navigators. We used two strategies to recruit researchers and Community Experts for the V-CES sessions. Researchers were recruited by the Science Navigator from a mapped pool of researchers who focus on maternal mental health and/or opioid use/recovery. Interested researchers were invited to submit a paragraph describing their research and plans for community engagement including a summary of the problems or questions their project would address, target population, stage of research, and questions they wanted to propose to Community Experts along with feedback needed. The V-CES team chose four research projects problems or questions that were most likely to benefit from input from or be of interest to available Community Experts.

Recruitment of Community Experts

To recruit mothers with mental and/or substance use disorders as Community Experts, we reached out through the Maternal Mental Health Research Collaborative (MMHRC) listserv and social media platforms. First, we emailed a survey to the MMHRC listserv. The survey was designed to query mothers who would be interested in participating as Community Experts regarding contact information and availability as well as basic personal and/or professional mental health and/or substance use experiences to help build an appropriate Community Expert pool. We published the survey link online to reach potential participants who were members of the MMHRC Facebook group, a social media initiative for reaching mothers coping with maternal mental health conditions and/or substance abuse. The Manager or the Community Navigator contacted potentially interested Community Experts to set up an additional screening conversation via telephone or online platform. The purpose of this additional screening step was to confirm potential Community Experts' interest, availability, and comfort level participating in an online group discussion on challenges facing mothers and how to improve research on maternal mental health and/or substance use. An important part of the screening process was also ensuring that Community Experts have a relatively quiet and private place where they feel comfortable talking about sensitive topics, and the technology tools and skills to participate virtually.

Preparation of Participants

Preparing Community Experts and researchers for the V-CES and buttressing their sense of agency is an important step in the V-CES process. Prior to the V-CES, the team emailed the V-CES participants a guide that provided a general description of process and the role of the Community Experts and researchers; and an online survey that captured general background information. Participants were asked to review and complete these materials prior to the V-CES. Participants were encouraged to contact the Operational Team with any comments and/or questions. We prepared a series of videos of researchers addressing questions provided by mothers regarding research – Research 101 for Mothers (https://research4moms.com/research-101/). Mothers provided video clips for researchers regarding research participation (https://research4moms.com/research-101/). These videos were available to participants who requested further information about the project.

The V-CES Process

In the introduction of a V-CES meeting, Community Experts and the researcher were informed by the Facilitator that the Science Navigator would take notes. Participants were encouraged to have their video cameras on, if they were comfortable. The V-CES Manager and/or the Facilitator asked for participants' permission to record the meeting. This recording was only used as a reference for notes and would never be shared without permission from participants. The steps in conducting the V-CES included: greetings and introductions, providing a brief overview of the purpose of the meeting and the process of discussion and communication ("discussion roles"), and the researcher's presentation. The Facilitator kept the conversation on track, making sure everyone's voice was heard and the two to three research topics were addressed. The Science Navigator took notes throughout the discussion and, finally, the Community Navigator thanked everyone for their participation and explained next steps.

V-CES Follow-Up

The V-CES follow up materials provided the researcher important feedback from the Community Experts and facilitated further collaboration between the researcher and the Community Experts.

Follow-Up for the Researcher

A summary report including the Science Navigator's notes and verbatim written comments from the Community Experts was shared with the researcher within one week of the V-CES. We highlighted specific recommendations as related to the topics that were discussed during the meeting (21). The researcher also received a one-page Continuing Community Engagement Guide that suggested ways for the researcher to maintain appropriate communication with the Community Experts who reported interest in serving as consultants as the research project develop.

Follow-Up for the Community Experts

We notified participants of any changes, adjustments and improvements to the research made as a result of their input. Items shared as follow-up could include updated outreach materials, policy and procedural changes or significant accomplishments of the study due to advice received during the V-CES. If possible, it would be important to provide Community Experts periodic updates on the project as well as any findings published or disseminated by the researcher (21). Community Experts participating in the V-CES sessions contributed to and had opportunity to review tip sheets for researchers and mothers developed as part of the project (See **Supplementary Material**). The V-CES toolkit is also provided as **Supplementary Material**.

RESULTS

Recruitment via Social Media

Our recruitment advertisement via the MMHRC listserv and Facebook group resulted in 91 mothers contacting us. We were able to strategically select an average of 5 mothers for each of the V-CES sessions and followed specific email invitations with a screening call by the V-CES Manager. We recruited more participants than needed in case a potential participant had a last-minute schedule conflict or childcare challenge.

Community Experts' Feedback

Community Experts who participated in the V-CES reported positive experiences. They were grateful for the opportunity to impact research and wanted more opportunities to participate in such co-production initiatives in the future. For example, one participant shared she felt *"... being heard and listened to [by the researcher]. I appreciate the time that was put into this"* (V-CES March 21). Another participant shared that she *"... like the fact that the researcher is trying to reach out to women during the first year after giving birth. I know it's a difficult time for many women, but so critical to understand for others"* (V-CES May 21).

Community Experts felt empowered by their research co-production experience and wanted to continue their involvement in the future to impact and disseminate research on maternal mental health and substance use issues:

> *"I loved that this researcher really wanted to figure out how we can add supports and not let these moms slip through the cracks. The researcher is motivated by identifying this vulnerable population and providing them with support"* (V-CES May 21)
> *"I liked the connections...I liked the diversity of professionals...I liked the empathy... I liked the desire to improve services, I liked that [the researcher] wanted to know what we think is working and not working and where we would like to see services go in the future."* (V-CES May 21)
> *"It was so helpful having all 3 experts from different geographic locations and within different medical communities - rural, large city, and small city; but we all had similar experiences. This says so much about how change needs to be widespread and proves to be a big challenge."* (V-CES March 21)

Participation in research co-production also has the potential to impact participants' recovery:

> *"Getting new mothers to talk about their substance use to begin with. It's a huge step for these moms to come forth to talk about addiction, or even admit they are experiencing addiction. It's scary and full of judgement from others. The stigma around addiction and mental health needs to be educated with the entire community. Hopefully moms will see the benefit of this program and trust the process"* (V-CES May 2021)

Last, Community Experts provided essential feedback for research conducted with mothers with mental health and substance use:

> *"I think that the idea of a flyer seems very non-threatening along with all the other paperwork that gets sent home with a new mom when she's discharged from the hospital. I had an entire folder. As we talked about the questions, it was really important to make the surveys more conversational in tone."* (V-CES May 21)
> *"I would say disclosure about substance use or any mental health that the participant maybe going through [is an issue]. As a mom, they [research participants] may feel judged or would be scared that something may happen to their baby if anyone knows what they are going through. Another challenge may be retention, having a person fill out a survey every month could be challenging, but I think with good incentives, it may make it a little easier."* (V-CES May 21)
> *"It's a huge step for these moms to come forth to talk about addiction, or even admit they are experiencing addiction. It's scary and full of judgement from others. The stigma around addiction and mental health needs to be educated with the entire community. Hopefully moms will see the benefit of this program and trust the process."* (V-CES May 21)

Researchers' Feedback

We learned that even when working with researchers who have previous community engagement experience, it was important and necessary to coach researchers on how to engage with Community Experts effectively. We found that for a successful and collaborative conversation Community Experts want: 1). to understand the researcher's motivation, so researchers should be willing and able to talk about their commitment to the topic, professionally and perhaps, personally; 2). to know how their input will specifically impact the research project and then the broader community; and 3). to feel heard as knowledgeable consultants. We had researchers create brief video overviews to introduce themselves and their interests, provided to participants prior to the event. The team reviewed their brief presentations in advance of the V-CES and provided feedback. Researchers were challenged by having a conversation in plain language with mothers and stakeholders whose perspectives they were hoping to solicit. They benefitted from coaching prior to V-CES sessions, as well as guidance and direction during sessions.

researchers reported that their perception about the role of patient or community stakeholders in their research changed as a result of the V-CES:

> *"I'm thinking about their interactions with the medical community and stigmatization and 'othering' they talked about."* (V-CES April 2020).
> *"They [Community Experts] had a fantastic understanding of the recruitment process."* (V-CES May 2021).

DISCUSSION

This article describes the efforts and steps taken to develop and implement V-CES, a co-production engagement method to involve patients and community stakeholders in the design and implementation of research projects, and the interpretation and dissemination of findings. The V-CES method is based on the CES model and was developed in response to the barriers and challenges in community engaged research caused by the breakout of the COVID-19 pandemic. Our experience demonstrates the potential contribution to V-CES to improve research engagement and relevancy in the virtual space post-pandemic, allowing for increased diversity of participants, communities, and service contexts. For researchers and Community Experts, in our case mothers with mental health and substance use, V-CES participation provided a "win-win" scenario, with the potential to improve recruitment efforts and make research outcomes more personalized, meaningful, and relevant.

Our specific V-CES pilot had three main limitations. First, by nature, the V-CES excludes populations without access to the internet or those who are not comfortable using online platforms. Therefore, participants were those with stable internet access, who felt comfortable using a virtual platform (Zoom) and were able to be in a safe, convenient location during their participation in a V-CES. As with other virtual remote approaches, it is important to recognize that the V-CES may be less accessible or effective for individuals who have no or limited access to the internet and those who prefer in-person interaction for many reasons (22). One benefit of the V-CES, compared to other digital-virtual approaches, is the existence of an in-person model, the original CES, that allows for the inclusion of populations who do not have or may benefit less from a virtual model.

Second, most Community Experts were White from Northeastern and Midwestern states. Recruiting a racially diverse group was difficult because a primary source for recruitment was the MMHRC Facebook group page, where most members are Northeastern/Midwestern US white women (3). Future use of the V-CES method should purposefully address diverse Community Expert populations. Last, due to the nature of the study (co-production participatory design), we did not collect information about what kind of mental health issues and/or SUDs recruited mothers experience, which may limit the replicability of the method in different subpopulations.

To summarize, the V-CES is a potentially useful approach for operationalizing co-production processes virtually, which is beneficial during emergencies (i.e., COVID-19) (23) but also for those living in rural areas, lacking transportation, or balancing work schedules and responsibilities at home, or for those who are experiencing barriers to "classical participation"(24). While the V-CES model may well be useful to researchers and Community Experts implementing co-produced research in other health domains, future studies are required to contribute to the growing literature on the science of engagement. For example, our team has been awarded a PCORI engagement award to implement and further evaluate the V-CES with mothers with intellectual and developmental disabilities and behavioral health conditions, with community stakeholders, and researchers. The V-CES toolkit and tip sheets are available as **Supplementary Material** and on the MMHRC website (https://heller.brandeis.edu/ibh/affiliates/mmhrc/about.html). We welcome its further use, implementation, and evaluation by community stakeholders and researchers, and look forward to receiving feedback for further improvements and future studies.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available to protect the participants' privacy. Requests to access the datasets should be directed to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the V-CES procedures were reviewed by the Brandeis University Institutional Review Board and deemed to be exempt from consideration as Human Subjects Research. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

FUNDING

Research reported in this article was funded through two Patient-Centered Outcomes Research Institute® (PCORI®). Awards: #EAIN-00147 and #8285-BU. The views and statements presented in this article are solely the responsibility of the authors and do not necessarily represent the views of the Patient-Centered Outcomes Research Institute® (PCORI®), its Board of Governors or Methodology Committee.

ACKNOWLEDGMENTS

We would like to thank the researchers and Community Experts who participated in the development and implementation of the Virtual Community Engagement Studio method and provided feedback on the process and resources produced. We would also like to acknowledge the contribution of the Vanderbilt University team in developing the original Community Engagement Studio method (https://victr.vumc.org/community-engagement-studio/). We respect their commitment, and appreciate the support of the Patient Centered Outcomes Research Institute to the process of engaging diverse stakeholders in research.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.805781/full#supplementary-material

REFERENCES

1. Zaami S, Marinelli E, Varì MR. New trends of substance abuse during COVID-19 pandemic: an international perspective. *Front Psychiatry.* (2020) 10:700. doi: 10.3389/fpsyt.2020.00700
2. Melamed O, Hauck T, Buckley L, Selby P, Mulsant B. COVID-19 and persons with substance use disorders: inequities and mitigation strategies. *Subst Abus.* (2020) 41:286–91. doi: 10.1080/08897077.2020.1784363
3. Mazel S, Zisman-Ilani Y, Hennig S, Garnick D, Nicholson N. Virtual engagement in a social media community of mothers with substance use disorders: content analysis. *JMIR Form Res.* (2021) 5:e24353. doi: 10.2196/24353
4. Luciano A, Nicholson J, Meara E. The economic status of parents with serious mental illness in the United States. *Psychiatr Rehabil J.* (2014) 37:242–50. doi: 10.1037/prj0000087
5. Hodgkinson S, Beers L, Southammakosane C, Lewin A. Addressing the mental health needs of pregnant and parenting adolescents. *Pediatrics.* (2014) 133:114–22. doi: 10.1542/peds.2013-0927
6. Priester MA, Browne T, Iachini A, Clone S, DeHart D, Seay KD. Treatment access barriers and disparities among individuals with co-occurring mental health and substance use disorders: an integrative literature review. *J Subst Abuse Treat.* (2016) 61:47–59. doi: 10.1016/j.jsat.2015.09.006
7. Lambert N, Carr S. "Outside the original remit": co-production in UK mental health research, lessons from the field. *Int J Ment Health Nurs.* (2018) 27:1273–81. doi: 10.1111/inm.12499
8. Bradley E. Carers and co-production: enabling expertise through experience? *Ment Heal Rev J.* (2015) 20:232–41. doi: 10.1108/MHRJ-05-2014-0016
9. Graham ID, McCutcheon C, Kothari A. Exploring the frontiers of research co-production: the integrated knowledge translation research network concept papers. *Heal Res Policy Syst.* (2019) 17:1–4. doi: 10.1186/s12961-019-0501-7
10. Farr M, Davies P, Andrews H, Bagnall D, Brangan E, Davies R. Co-producing knowledge in health and social care research: reflections on the challenges and ways to enable more equal relationships. *Humanit Soc Sci Commun.* (2021) 8:1–7. doi: 10.1057/s41599-021-00782-1
11. Hickey G. The potential for coproduction to add value to research. *Health Expect.* (2018) 21:693. doi: 10.1111/hex.12821
12. Salyers MP, Zisman-Ilani Y. Shared *Decision-Making and Self-Directed Care. In: The Palgrave Handbook of American Mental Health Policy.* Cham: Springer International Publishing. (2020) p. 197–228.
13. Zisman-Ilani Y, Barnett E, Harik J, Pavlo A, O'Connell M. Expanding the concept of shared decision making for mental health: a systematic and scoping review of interventions. *Ment Heal Rev J.* (2017) 22:191–213. doi: 10.1108/MHRJ-01-2017-0002
14. Paylor J, McKevitt C. The Possibilities and Limits of "Co-producing" Research. *Front Sociol.* (2019) 0:23. doi: 10.3389/fsoc.2019.00023
15. Lignou S, Capitao L, Hamer-Hunt JM, Singh I. Co-production: an ethical model for mental health research? *Am J Bioeth.* (2019) 19:49–51. doi: 10.1080/15265161.2019.1619877
16. Cairns J, Nicholls J. Co-production in substance use research. *Drugs Alcohol Today.* (2018) 18:6–16. doi: 10.1108/DAT-02-2018-0002
17. Gentile A, Torales J, O'Higgins M, Figueredo P, Castaldelli-Maia JM, De Berardis D, et al. Phone-based outpatients' follow-up in mental health centers during the COVID-19 quarantine. *Int J Soc Psychiatry.* (2022) 68:129–33. doi: 10.1177/0020764020979732
18. Gentile A, De Berardis D, Tomasetti C, Ventriglio A. Digital psychiatry in COVID-19 pandemic: an Italian perspective. *Psychiatry Res.* (2020) 292:113316. doi: 10.1016/j.psychres.2020.113316
19. Joosten Y, Israel T, Williams N, Boone L, Schlundt D, Mouton C, et al. Community engagement studios: a structured approach to obtaining meaningful input from stakeholders to inform research. *Acad Med.* (2015) 90:1646–50. doi: 10.1097/ACM.0000000000000794
20. Joosten YA, Israel TL, Head A, Vaughn Y, Villalta Gil V, Mouton C, et al. Enhancing translational researchers' ability to collaborate with community stakeholders: lessons from the community engagement studio. *J Clin Transl Sci.* (2018) 2:201–7. doi: 10.1017/cts.2018.323
21. Israel T, Joosten Y, Vaughn Y. Community Engagement Studio Toolkit 2.0. (2019). Available from: https://victr.vanderbilt.edu/pub/community/ (accessed Oct 26, 2021).
22. Husain MO, Gratzer D, Husain MI, Naeem F. Mental illness in the post-pandemic world: digital psychiatry and the future. *Front Psychol.* (2022) 12:567426. doi: 10.3389/fpsyg.2021.567426
23. Clark M, Vliet E. van, Collins M. Reflections from the COVID-19 pandemic on inequalities and patient and public involvement and engagement (PPIE) in social care, health and public health research. *Heal Expect.* (2021) 24:1547–50. doi: 10.1111/hex.13278
24. Noa C, Tal A, Zisman-Ilani Y. Digital community inclusion of individuals with serious mental illness: a national survey to map digital technology use and community participation patterns in the digital era. *JMIR Ment Heal.* (2021) 8:e28123. doi: 10.2196/28123

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 15 July 2022
doi: 10.3389/fpsyt.2022.817208

Developing and Initially Validating the Youth Mental Health Literacy Scale for Ages 11–14

Joanne Riebschleger[1], Christine Grové[2]*, Kimberly Kelly[3] and Daniel Cavanaugh[4]*

[1] School of Social Work, Michigan State University, East Lansing, MI, United States, [2] Department of Educational Psychology, Monash University, Melbourne, VIC, Australia, [3] Department of Counseling, Educational Psychology and Special Education, Michigan State University, East Lansing, MI, United States, [4] School of Nursing and Health Studies, University of Washington, Bothell, WA, United States

Despite rising rates of youth mental health disorders and suicides, most youth lack access to accurate, non-stigmatized mental health information. Instead, many describe people with mental illness as violent and incompetent. Mental health literacy aligns with resilience theory. It assumes that youth that have accurate mental health information will have less stigmatized views of mental illness and will be more likely to seek help earlier should mental health symptoms arise. Accurate, non-stigmatized mental health information is especially needed for Children of a Parent or other Family Member that has a mental illness (COPFMI) since they are more likely to acquire a mental illness than children who do not have a family member with a mental illness. COPFMI youth are in need of the same mental health information as general population youth but they can also benefit from knowing how to deal with a family member's mental health disorder. Based on many foundation studies and key stakeholder input from parents, educators, mental health providers, child welfare providers, and especially youth, an emerging Youth Mental Health Literacy (YMHL) scale was developed and validated for measuring the mental health literacy levels of youth ages 11–14. The scale provides a full scale score of youth mental health literacy. It has subscales of knowledge of mental illness and recovery; stigma, help seeking for self/others; coping with stress; and dealing with family mental health challenges. The validation study indicated support for a unidimensional structure for each of the refined subscales. The subscales showed suitable reliability as evaluated by several measures of internal consistency. While the scale needs further study with larger samples of youth, it is hoped that the scale can yield mental health literacy outcome data that can help mental health literacy programs to build evidence-based programs that may, in turn, help prevent, delay, or ameliorate mental health disorders among youth.

Keywords: mental health, scale, children, mental health literacy (MHL), youth, families

Edited by:
Hongmei Wang,
Zhejiang University, China

Reviewed by:
Sharinaz Hassan,
Curtin University, Australia
Lin Zhu,
Shanghai Health Development Research Center, China

****Correspondence:***
Joanne Riebschleger
riebsch1@msu.edu
Christine Grové
christine.grove@monash.edu

Specialty section:
This article was submitted to Public Mental Health, a section of the journal Frontiers in Psychiatry

Received: *17 November 2021*
Accepted: *02 June 2022*
Published: *15 July 2022*

Citation:
Riebschleger J, Grové C, Kelly K and Cavanaugh D (2022) Developing and Initially Validating the Youth Mental Health Literacy Scale for Ages 11–14. Front. Psychiatry 13:817208. doi: 10.3389/fpsyt.2022.817208

INTRODUCTION

The aim of this work is to report an early stage of development and validation of a new scale to measure mental health literacy among youth ages 11–14. The researchers set about to design a scale that could be used to measure mental health literacy (MHL) levels of youth drawn from general population youth and/or those that have a parent or other family member with a mental illness.

The idea was to find a scale that could be flexible for use across a range of emerging programs. For example, if a program was delivered to whole classrooms of middle school and high school student general population youth (1), MHL levels could be measured. Program providers targeting youth that have a parent or other family member with a mental illness (2) could also use the scale to measure MHL levels.

Our definition of youth mental health literacy includes all of the constructs/components of mental health literacy, as defined by Jorm (3): *knowledge* of possible mental health symptoms, *recovery* from mental illness, mental illness *stigma*; and *help-seeking* for self and others. This MHL definition was enhanced with the addition of two additional components. We added youth *coping* with stress as a definitional component of mental health literacy (4, 5). The second add-on component of MHL pertains to youth with a parent or other family member with a mental health disorder, specifically their interactions with *family* mental health concerns (6).

The inclusion of youth coping with stress as a part of MHL is based on rapidly rising rates of youth mental health disorders, often preceded and/or accompanied by persistently high levels of youth reported stress (4, 5). Further, a general population mental health literacy program that assessed youth reported stress regularly found youth reported moderate to very high levels of stress throughout the program (6). Since many young people have a family member with a serious mental illness such as a parent and/or sibling, we also included an additional mental health literacy subscale called family, i.e., dealing with family mental health concerns (7).

In this study, youth is defined as ages 11–18; young adulthood includes people ages 19–22. Study participants herein are ages 11–14, aligning with grades 7 and 8th grades, or pre-secondary school enrollment. Mental illness is defined herein as an axis I disorder, as explicated by the American Psychiatric Association (8). Common mental illnesses include anxiety, depression, and substance abuse.

Youth Mental Health

There are significant risks associated with youth acquiring a mental illness. In the USA, suicide is now the second leading cause of death of people ages 10–34 (9). The CDC revealed a 40% increase in youth reports of feeling persistently sad/hopeless in the pre-COVID time frame of 2009 to 2019. Nearly one in six youth revealed making a suicide plan in the past year, yielding a 44% increase from 2009 to 2019 (10). The World Health Organization or WHO (11) noted, "Mental health disorders account for 16% of the global burden of disease and injury among people ages 10–19" (p. 1). WHO (11) cited the work of Kessler et al. (12) to explain that "half of all mental disorders begin by age 14 but most cases go undetected and untreated" (p. 1). Symptomatic children and adolescents that have not received mental health support or treatment are at risk of substance abuse, violence exposure, poor sexual health, and increased disability-adjusted life years (13). Those youth that continue to experience mental health disorders into adulthood are more likely than adults without a mental health disorder to experience a shortened life span (14), increased physical illness (15), and increased criminal justice involvement (16). People who experience mental illness also face an increased likelihood of being a victim of violent crime (17, 18). They are more likely to experience stigma and discrimination in employment, housing, public services, and interpersonal relationships (19). Suicide and self-harm are particular risks for people with a mental illness (20).

Children of a Parent or Other Family Member With Mental Illness (COPFMI)

Those at particular risk for acquiring a mental health disorder include children/youth that have a parent or other family member with a mental illness (COPFMI). Nearly 24% of mental health patients in Australian adult services have one or more minor children (21). Campbell et al. (22) reported that many children and adolescents receiving mental health treatment have a parent with a mental illness, specifically 36% of their mothers and 33% of their fathers. This has led to calls for increasing child, parent, and family mental health promotion and prevention programs (3, 23, 24). For example, foundation work conducted by Nicholson (25) recommended support for parents, especially mothers, to reduce parents' mental health symptom levels and developmental risks of their children.

Youth with a parent or other family member with a mental illness often experience family separations, early caregiving, frequent moves, school changes, and high rates of worrying about family and personal well-being (26). Mental health stigma is associated with higher rates of out of home placements for children with a parent with a mental illness (27). Parental mental illness can affect children in many ways, including becoming the caregiver for parents, siblings, and other family members (2). COPFMI have lower levels of academic achievement, social functioning, and school participation (28, 29). Youth living with a parent with a mental illness may experience a challenging family environment that can include parenting difficulties, emotional vulnerability, high stress reactivity, and child feelings of guilt, shame, and loneliness (28, 29). COPFMI are more likely to acquire mental illnesses than children without COPFMI experience (26, 30), especially depression and anxiety.

Risk and Resiliency

The CDC, as well as WHO, use a risk and resiliency theoretical foundation to consider the process of people acquiring health and mental health disorders. The guiding theory for the YMHL scale development process is similarly based on risk and resiliency, within a frame of youth development. The theory assumes that youth development can be negatively impacted when youth are exposed to risks such as child-parent role reversal, trauma, losses, physical neglect, emotional neglect, bullying, violence, and homelessness. However, risk and resiliency theorist Masten (31) purports that many youth meet, and even exceed, developmental tasks despite exposure to risk factors. Masten (31) says young people can benefit from the application of resilience promoting and ameliorating factors that include coping skills acquisition, social support, community resources, and access to accurate knowledge for dealing with a situational risk. The latter fits especially well with health literacy whereby a patient and/or family members learn about preventing, ameliorating, and/or managing the severity of a health disorder.

Mental Health Literacy

Mental health literacy is similar to health literacy; individuals and/or family members acquire accurate, non-stigmatized information about a disorder and its treatment (3, 6, 32). Mental health knowledge can contribute toward increased resilience among young people, including those with a parent or other family member with a mental illness (33). Mental health literacy program outcomes can include youth who are more likely to recognize mental health stigma and how to respond to stigma events (7). People with accurate, non-stigmatized mental health information are more likely to seek help earlier for what they suspect may be mental illness symptoms. This is important because the present average delay from symptoms arising to getting mental health services is 8.2 years for those with mood disorder (34). Within this 8.2 year time frame, it took an average of 6.9 years to recognize that a disorder may be present. Thompson et al. (34) also indicated that there is a 1.3 year delay between recognizing the mental health problem, seeking help, and making first treatment contact.

Knowledge of mental illness symptoms could decrease the time span between recognizing disorders and seeking help (1, 35). Bale et al. (32), as well as Riebschleger et al. (7) report that individuals armed with mental health literacy information are more likely to understand that mental illness is common and mental health treatment is usually effective. Those with higher levels of mental health literacy are less likely to hold stigmatized views of mental illness and are more likely to seek help for mental health symptoms of one's self and others (7, 32). Earlier help-seeking among youth could mean that more severe mental illness symptoms and the social and economic secondary effects associated with experiencing mental health stigma may not develop or may be less severe if they do develop. Additionally, earlier help-seeking may reduce feelings of isolation, lower school performance, overall stress levels, and suicidal thoughts (35, 36).

Youth with a parent or other family member with a mental illness that have higher levels of mental health literacy may be likely to state that a family member's level of illness is not their fault. For example, youth are less likely to think about what they should, or should not have done to keep another person in the family from "getting worse" (2, 6, 36). In fact, they are also more likely to report that mental health disorders are health challenges that are nobody's fault. They are more likely to know how to talk about mental illness, recognize that recovery is possible, and state that there is hope for their future. They are more likely to know how to seek help for family and peer mental health crises. They are more likely to know when to help out in the family and when they can relax and "be a kid." (37).

Mental Health Literacy Programs

A number of mental health literacy interventions are emerging or developed (38). For example, Mental Health First Aid helps prepare family caregivers and emergency responders to engage in best practices during mental health emergencies (3). Australian programs Be You and Beyond Blue target mental health in early learning services and schools toward building a positive, inclusive, and resilient community (39). Kutcher et al. (1) built a mental health education program for Canadian high school students. Two key purposes are to increase youth ability to seek help earlier for mental health symptoms and to reduce mental health stigma. Family Talk is a program developed by Beardslee et al. (38) that is under development in a number of European countries (40, 41). Family Talk programs help parents with a mental illness to be able to talk to their children about their mental health disorder (41). The idea is to open communication about mental illness among family members. The Youth Education and Support program by Riebschleger et al. (7) especially targets small groups of school youth, some of whom likely have a family member with mental illness. The program delivers 10 activity-based sessions of mental health literacy content to youth ages 11–16.

A Need for Youth Mental Health Literacy Scales

There are emerging and developed mental health literacy programs that educate about mental health constructs/components (7, 42–44). However, the programs are usually not sufficiently evaluated because of a limited availability of psychometrically valid mental health literacy scales (45–48). Measurement of outcomes of mental health literacy programs will not be able to move toward evidence-based practices until they have access to sound mental health literacy scales. Programs that are not evidence-based face potential barriers to implementation and funding (49).

O'Connor and Casey's (50) review of the literature found that there are few appropriate mental health literacy assessment tools to evaluate mental health literacy programs for young people. They found no available mental health literacy measures covered all aspects of mental health literacy as described by Jorm (3), i.e., knowledge of mental health and recovery; mental illness stigma; and help-seeking/giving. Wei et al. (47) conducted a scoping review of available tools to find that more measures have been developed but most covered only some aspects of MHL and few had undergone strong psychometric validation. Another more in-depth review found that some MHL scales had undergone more rigorous evaluation (48). However, the scales developed for middle and high school students, did not appear to cover all components of MHL (48). Newer scales have been published to evaluate mental health literacy for adults but have too high of a reading level for young people (50, 51). Members of the authors' research team published an earlier Knowledge of Mental Illness and Recovery scale (7) but it did not have new constructs of coping and family.

There is a particular need for general population youth mental health literacy scales that include all of the Jorm (3) mental health literacy components of knowledge of common mental illnesses, recovery from mental illness, mental illness stigma, help seeking for self, and help seeking for others. However, it is important to also measure specific COPFMI ways of dealing with family members' mental illness, such as knowing how to seek help for a family member's mental illness crisis; when to help or not help out at home; and how and when to talk to family members and others about mental health (52). Scale authors can consider including a family component since one of four children may have a parent with a mental illness (21). When siblings and other family members are considered as well, the likelihood of a mental health literacy program participant youth having someone in the family that has a mental illness is even higher. It is also important to consider adding questions on coping with stress as recommended by youth providing input to the development of the scale herein.

There is a need for scales that have reading levels consistent with the reading levels of the youth. For example, for youth ages 11–14, the scale reading level would need to be about ages 9–10 in order to include slower readers. The questions should align with youth-reported mental health perspectives which tend to be behavioral (What do I see people doing and saying?) and not diagnostic (What are the symptoms and diagnoses?) (28). The scale should challenge the youth to consider the accuracy of their cognitive assumptions about mental illness, particularly if they hold stigmatized views of mental illness. Unfortunately, there appear to be limited scales that meet all of the needed criteria, especially for youth ages 11–14.

MATERIALS AND METHODS

A Strong Research Foundation

The Youth Mental Health Literacy Scale content was drawn from many years of research devoted to identifying mental health literacy needs of children and youth. This included research targeting how children and youth with a family member with a mental illness describe their information needs. A wide array of stakeholders informed the scale developers of mental health literacy needs of youth, including children, youth, parents, mental health services providers, child welfare providers, and educators. In addition, authors Riebschleger and Grové spent a good deal of time talking to children and youth about mental illness within the administration of the Youth Education and Support program (6, 53) and a video MHL program for COPFMI youth (2).

Riebschleger et al. (52) surveyed COPFMI experts specifically about what kind of content would be needed for a scale to develop mental health literacy. We then conducted an intensive literature review guided by the overarching research question, "What do children and youth need to know about mental illness?" (36). The data collection included examining literature about the mental health literacy needs of children and youth with a family member with mental illness. Riebschleger et al. are also engaged in developing a youth informed mental health literacy website for adolescents (https://mhiteens.org/). The entire content was generated by youth and young adult suggestions for the recommended content. We also had a youth advisory group that continued to review the mental health literacy content and to make continued suggestions for youth mental health literacy content. The youth suggestions were included in the development of scale constructs.

Across the research projects, we learned: (1) general population *and* COPFMI youth need accurate non-stigmatized mental health information; (2) parents with a mental illness do not often talk to their children about their disorders; (3) children ages 11–16 report experiencing high levels of stress on a regular basis; (4) most children ages 11–16 could identify depressive symptoms but knew little about other disorders; and (5) many children and youth described people with mental illness as looking physically "different"; being violent, dangerous, cognitively incompetent; and unlikely to have "a good life." Few youth knew the word stigma or its meaning. Some thought people with mental illness were physically ill; several thought they used wheelchairs. Additionally, children with a family member with a mental illness did not seem to know any more about mental illness than general population youth. They knew the most about depression but very few knew about schizophrenia or bipolar disorder. Simply put, youth levels of mental health literacy were low across all of the studies.

Constructs and Instrumentation

Most of the content needed by COPFMI were the same as general population youth. However, there appeared to be a need to include additional understanding of how to deal with a family member's mental illness, e.g., seeking help for mental illness crises, understanding that mental illness is nothing to be ashamed of, and articulating that mental illness is no one's fault. Many of the COPFMI youth blamed themselves for the symptoms of a person with mental illness (7). The researchers drew on literature and stakeholder needs assessment to determine that a youth mental health literacy scale should cover all of the Jorm (3) constructs, with additional coping and family subscales. Another consideration was that the scale would likely be useful for assessing the outcomes of educational mental health literacy interventions with 11–14 year old middle school youth and for special programs for youth with a family member with a mental illness.

The research team developed items to align with five components: knowledge about mental illness, knowledge of recovery, stigma, help seeking for self, and help for others. These items covered all of the Jorm (3) MHL content areas. Given the rapid rise in stress levels reported by youth and evidence of a connection between stress and the development of anxiety and depression (4, 5), the component "coping with stress" questions was included. In addition, scale questions about living and responding to mental illness behaviors of a family member were developed; they comprise a family subscale. The response options were a closed-format type with a response set of three choices consisting of one correct answer and two incorrect answers, as recommended by a scale development expert. The resultant draft YMHL scale consisted of 74 multiple choice questions with subscales that were given full names followed by abbreviations, i.e., knowledge of MI (K-MI), knowledge of MI recovery (K-R), stigma (S), help seeking for self (HS-S), help seeking for others (HS-O), coping (C), and family subscale (F). The questions were graded correct/incorrect according to an answer key that underwent review by the item developers and the consulting project psychometricians.

The *knowledge* of mental illness subscale combines mental illness and recovery constructs. Subscale question responses focus on mental illness presented as a health disorder that often responds to active treatment. Subscale questions ask about the prevalence of mental illness, common mental illnesses (especially depression, anxiety) and recovery strategies (counseling, medications, social support, healthy habits). Questions identify that mental illness can have a genetic component.

The *stigma* subscale focuses on the discrimination and inaccurate labeling of people with mental illness. Subscale questions ask about people making fun of people with mental illness and experiences of being judged for having a mental illness (assumed incompetent, weak, violent). There is a question about media and social media emphases on mental illness associated with incompetence, weakness, and violence. Some questions focus on the societal tendency to blame people for "causing" the mental illness of selves or others. One question asks about people feeling embarrassed about their mental illness and/or the mental illness of a family member. Other subscale questions report the facts of mental illness: people with mental illness are as intelligent as others in the general population, more likely to be a violence survivor than a perpetrator, able to make decisions, and are usually able to work. People with mental illness can have good lives. They can also be okay parents too (so one should not assume parents with mental illness are neglectful or abusive).

The *help* subscale combines help seeking for self and help giving for others. There is a strong emphasis on talking to adults that may be able to help. There is a specific subscale question on the importance of telling trusted adults at school (and other places) about someone having thoughts of suicide. This is presented as a life threatening situation that youth should not keep secret even if the person might not like them telling. Several questions address how and when to help out at home when a family member has a mental illness and how to support a friend with mental illness.

The *coping* subscale addresses stress management. The questions ask about useful ways to manage stress (talking to someone, exercise, nutrition, relaxation, deep breathing, listening to music, journaling, and positive activities selected by the youth). Negative stress management is described as behaviors that are usually not useful, i.e., yelling at family, friends, and others; breaking things; and using alcohol and illicit drugs.

The *family* scale was built into the instrument to provide flexibility of program evaluation. The family questions are spread across knowledge, stigma, help, and cope subscales. The logic is that family situations can be part of mental health literacy. Professionals delivering MHL programs in general education may or may not want to employ the family subscale as part of their programming evaluation. However, the professionals delivering emerging MHL programs for COPFMI youth are likely to find the family subscale important as one way to measure pre and post intervention outcomes.

Procedures

Prior to scale administration, we obtained research approval from the Michigan State University Institutional Review Board, and organizational approval from the schools, followed by parent consent, and then youth assent. The draft YMHL scale was administered to youth ($n = 178$) enrolled in biology, gymnastics, and psychology classes located in two middle schools and one high school. The schools were located in a midwestern state. The present research focuses on YMHL scale development for middle school students ages 11-14, and uses data collected from the younger youth ($n = 85$). This validation sample is smaller than the original plan due to the March 2020 school closures in response to the coronavirus pandemic. Co-Vid continues to make collection of data in schools difficult as schools move back and forth between online and in person formats per fluctuating Co-Vid infection rates.

Analysis

We used a multi-step process to refine the initial item sets for each of the subscales and examine the psychometric properties of the subsequently refined seven subscales using the pilot study data collected from middle school students. Scale refinement was guided by results obtained from item- and subscale-level descriptive statistics, confirmatory factor analysis (CFA), and Mokken Scaling Analysis (MSA), a nonparametric item response theory model (54–56). To evaluate the structural validity of each of the seven subscales, we used both Mokken Scaling Analysis and Rasch modeling (57). Subscale reliabilities were assessed using several internal consistency measures.

Subscale refinement proceeded first with initial item- and scale-level descriptive analyses to assess item scalability and individual youth response quality. Response patterns across all subscale items were examined, and youth with unusual patterns were flagged. Unusual patterns may suggest that individual respondents are not interacting with the instrument in an expected way (i.e., lack of attention, reading level issues) but may indicate that items are not functioning properly. To assess whether the unusual pattern is specific to a respondent rather than to an issue with the item, patterns were examined for multivariate outliers. In the few cases where this was found, respondents with unusual patterns were flagged for later consideration, since later item refinement steps consequently impact the flagged respondent's response pattern. To make a judgment about item functioning, unusual response patterns within an item were also considered. Tetrachoric correlations between item pairs within subscales were used to identify items that did not fit with the remaining subscale items and items with negative correlations were removed from the subscale. All items that all youth got wrong, along with items that all got right, were eliminated, and items exhibiting little variation were flagged for consideration following later refinement steps. Missing data patterns of the youths' responses were examined, and those missing half or more of the responses were removed from the validation sample. Following these steps, the validation sample of each of the subscales consisted of a set of positively correlated items for youth who had data for more than half of the possible number of responses.

Several techniques of MSA were used to further guide item refinement, examine subscale dimensionality, and evaluate response patterns. In contrast to factor analytic techniques, this approach makes few assumptions about the data and does not require large datasets. To flag unusual items or responses in the validation sample, results of Guttman errors and automated item selection procedure (AISP) were examined. Guttman errors are person-specific and used to flag respondents who provide an exceptionally high number of unexpected responses given their responses on remaining items. The unidimensionality of each subscale was examined separately using an automated item selection procedure (AISP) that separates items into like groups, much like what is produced by an exploratory factor analytic approach. The results of this procedure were examined for items inconsistent with a unidimensional scale. The items flagged in the AISP were excluded from subsequent analyses. Following AISP, resultant item sets were tested for local independence, monotonicity, and invariant item ordering. Following further refinement suggested by the tests of these scale properties, the dimensionality of the items and subscales were examined using the homogeneity coefficient (58) typically used with the Mokken scaling approach. The item-specific homogeneity coefficient, an indicator of item scalability, provides a measure of correlation of the item with the remaining items in the subscale. The scale homogeneity coefficient measures the degree to which the total score accurately reflects person ranking on the construct purported to be measured by the subscale. Interpretation of the homogeneity coefficients were guided by rules of thumb developed by Mokken (54).

Factor analytic techniques were also used to explore the dimensional structure of each subscale. Principal components and principal axis factoring were used to explore the number of dimensions for each of the refined subscales. A confirmatory factor analysis model that used the refined subset of items was also estimated for each subscale, and the factor loadings and various model fit statistics were examined to determine the level of agreement between what MSA suggested and what the CFA results indicated regarding the dimensionality of each subscale. The results from the CFA were treated as complementary to the MSA results, rather than as a primary approach to assessing dimensionality due to the strong assumptions of this approach. To estimate a factor analysis model using binary data, one must assume that the binary scores are discretized versions of latent continuous variables, and that the underlying continuous variables have a multivariate normal distribution (59). Because the YMHL is a newly developed scale, we felt that invoking such strong assumptions would be unwarranted at this early stage. Therefore, the MSA results were afforded more weight than the CFA results in subscale refinement decisions. However, because a factor analytic approach is more common in the literature than MSA, we chose to include information about how the scale refinement suggested by MSA compared to that suggested by FA results where relevant.

Items and responses that were flagged by descriptive analyses were noted but retained for use in the MSA and confirmatory factor analysis. These analyses were used as the principal method for informing the refinement of each subscale to a subset of items that work together to measure the dimension of youth mental health literacy targeted by that subscale. The reduced item set for each subscale was then scaled using the Mokken approach, followed by scaling with the alternative approach of Rasch modeling (57) if the data met the strong assumptions of this approach.

Like MSA, the Rasch approach can be used to produce a measure of the construct or latent trait (e.g., knowledge of mental health, stigma) for a respondent. This parametric scaling technique, appropriate for binary items, shares the assumptions of MSA (local independence, monotonicity, and invariant item ordering), but with a more stringent conceptualization of the nature of the underlying construct as a quantitative rather than the ordinal characterization of Mokken scaling. As such, the Rasch approach requires additional stricter assumptions about the distribution of the data in order for the model to be able to make more finely-grained measured distinctions between respondents. Whether the data meet the Rasch modeling assumptions is determined by a number of tests and measures, including model fit and person fit statistics, tests of item infit and outfit, and an assessment of subgroup homogeneity of scores. Assessment of these assumptions proceeds in an iterative manner, as ill-fitting items and persons are removed from the dataset and the reduced dataset to analyzed to detect additional misfitting items and/or persons. A final model is one that is judged to produce acceptable fit statistics with the reduced set of items and persons. The restrictive assumptions of Rasch approach can frequently result in a sizeable deletion of both items and persons and consequently produce a scale with unacceptable reliability (60). In the instances when the data can support a Rasch analysis, the model offers the advantage of more detailed construct measures.

Following scaling, the reliability and the distribution of scores of each subscale were examined. Estimates of the internal consistency of a scale only make sense if the scale is unidimensional; this requirement was assessed prior to determining reliability. Several indices of internal consistency were computed. The accuracy of the usual measure, Cronbach's α, relies on the strict assumptions of Classical Test Theory, which are rarely met in applied contexts. Given this weakness, the estimate is augmented with the calculation of the estimate's bootstrapped 95% confidence interval (61), as well as an additional measure of internal consistency. This alternative internal consistency measure, ω [coefficient omega (62)], relaxes the strict assumptions required by Cronbach's α and has been shown to be a better estimator of reliability (63). Conceptually, ω can be considered as an estimate of the amount of variance of subscale items that is accounted for by a single (unidimensional) latent trait or construct. The values of both α and ω are on the same numerical reliability scale.

Several other descriptive analyses were performed on the refined subscales following scaling. The distribution of the total scores were determined and investigated. The average difficulty and discrimination were estimated for each subscale. The acceptability of the score distribution was considered subjectively as being suitable for separating the youth along the dimensions of MHL. In order for some separation to occur, the scores would need to have enough variation to suggest groupings. Related to meeting this goal, a suitable scale would also produce a score distribution that does not exhibit floor or ceiling effects. The presence of these effects was assessed for the youths' scores resulting from each refined subscale.

TABLE 1 | Validation sample descriptives $n = 80$.

	Percent
Gender	
Female	53.8
Male	42.5
Not reported	3.7
Race/Ethnicity	
White	58.8
Asian, Indian, American Indian, or Alaskan Native	20.1
Black	5.0
Latinx	3.8
Mixed Race	3.8
Not reported	8.8
Grade	
7th	32.5
8th	67.5
	Mean (sd)
Age	12.96 (0.75)

RESULTS

The initial subscale refinement analysis indicated several important findings about the original item sets for each of the subscales. Item-level descriptive statistics were reviewed for each subscale, and these results were used to identify items with substantial overlap that behaved similarly in terms of response patterns. This information, coupled with in-depth review by the item developers reduced the total number of items from 74 to 60. Correlation results suggested that two pairs of subscales could be combined. The Knowledge (K) subscale is a combination of knowledge of MI (K-MI) and knowledge of MI recovery (K-R). The Help-Seeking (HS) subscale is a combination of the help seeking for self (HS-S) and help seeking for others (HS-O) subscales. The items that had negative correlations with the remaining items were removed. Following these refinements, the remaining items were 10 for K, 10 for S, 14 for HS, 13 for C, and 13 for F. The dataset was reduced to include only respondents who provided more than half of the responses for each of the subscales, resulting in a validation data set ($n = 80$) described in **Table 1**.

Each subscale was examined for dimensionality using the AISP, and the responses of the youth were assessed for unusual patterns. The AISP analysis indicated that some items within all but the F subscale were not grouping as expected if the subscale were providing a unidimensional measure of the subscale's construct. Using the lowest threshold value for indicating item grouping, item elimination was accomplished iteratively, as items one by one were excluded and the analysis rerun with the reduced item set until AISP results indicated a unidimensional group of items. In making a final decision about elimination of an item, we also considered the results of tests for local independence, monotonicity, and invariant item ordering; all item subsets suggested by the AISP analysis met these tests. The subscale items were also examined with factor analytic models, and the results did not depart from the dimensionality findings using the AISP. Examination of responses using the number of extreme Guttman errors found no more than 10 across the subscales. Given more scale refinement was expected in the scaling step, it was decided to retain high-error respondents and reevaluate their status later in the validation process. Following the refinements given MSA results, the subscales K and H were reduced by 1 item, the S subscale was reduced by 2 items, the C subscale was reduced by 4 items, and the F subscale remained at 13 items.

The structural validity of the five subscales was examined first using MSA. The reduced sets of subscale items were used as the starting set for the MSA scaling procedures. Coefficients of homogeneity for each item, item pair, and the subscale were computed to determine the scalability of each item. Items not exhibiting acceptable levels of homogeneity (minimum threshold of 0.3, as suggested by 52) were excluded from further analyses, and analyses were rerun iteratively after item removal. This was followed by an evaluation of the local independence among items; violating items were removed and local independence was reassessed iteratively until this yielded a set of items that were related through the construct only. Only the S subscale had two items that were flagged at this stage; both were removed. The relationship between the endorsement of items of differing levels of challenge to the measure of the construct was confirmed by evaluating the monotonicity of the scale; all items within each of the subscales conformed with the expected relationship. AISP was rerun to confirm that the resultant item set of each subscale formed a one-dimensional measure. An evaluation of the degree of invariance of the item ordering across different levels of the measure followed. The Guttman errors of respondents were reassessed with the resultant subscale item sets to identify possible idiosyncratic response patterns. The number of respondents exhibiting a high number of Guttman errors ranged from two on the HS subscale to eight on the C subscale. Because the distribution of Guttman errors did not show a concentrated cluster of respondents with high numbers of errors and given the already relatively small sample size, it was decided to retain this small number of youth in the validation sample.

The iterative approach described above was used to produce a final item set for each subscale that did not produce significant violations. After applying the MSA scaling approach, the number of items distributed across the subscales were as follows: a K subscale of nine items, a S subscale of six items, a 13-item subscale for HS, a nine-item C subscale, in addition to a 13-item F subscale measuring the family component across the four domains of knowledge, stigma, help-seeking, and coping. Using the homogeneity coefficient rules of thumb developed by Mokken (54), scalability can be classified as strong, moderate, or weak, based on the inequalities $H > 0.5$, $0.4 \leq H < 0.5$, and $0.3 \leq H < 0.4$, respectively. Using these rules of thumb as a guide, the subscales for S, C, and F exhibited strong scalability while the K and HS subscales exhibited moderate scalability. The item scalabilities, item-total correlations, and scalabilities of the subscales are shown in **Table 2**. Given that all five subscales also met the unidimensionality, monotonicity and local independence

TABLE 2 | Item scalabilities and item-total (point-biserial) correlations.

Subscale (*items*)	***H***														
Knowledge ($i = 9$)	0.43	Item	1	11	14	15	22	40	42	60	72				
		H_i	0.35	0.92	0.51	0.52	0.47	0.37	0.55	0.29	0.35				
		r_{bs}	0.59	0.61	0.48	0.57	0.70	0.57	0.71	0.53	0.54				
Stigma ($i = 6$)	0.58	Item	25	27	32	48	51	61							
		H_i	0.54	0.80	0.72	0.58	0.59	0.42							
		r_{bs}	0.66	0.70	0.70	0.70	0.65	0.62							
Help ($i = 13$)	0.44	Item	35	36	37	45	49	53	55	57	58	59	64	66	67
		H_i	0.55	0.39	0.38	0.68	0.42	0.35	0.50	0.37	0.36	0.58	0.45	0.43	0.41
		r_{bs}	0.60	0.57	0.52	0.71	0.60	0.55	0.73	0.58	0.55	0.73	0.68	0.64	0.46
Coping ($i = 9$)	0.69	Item	8	33	34	39	45	51	52	54	55				
		H_i	0.70	0.76	0.73	0.73	0.88	0.73	0.56	0.67	0.58				
		r_{bs}	0.57	0.75	0.72	0.63	0.78	0.78	0.66	0.75	0.68				
Family ($i = 13$)	0.53	Item	36	37	38	49	51	53	55	59	62	64	66	68	69
		H_i	0.51	0.46	0.84	0.50	0.53	0.43	0.55	0.84	0.90	0.42	0.43	0.51	0.47
		r_{bs}	0.69	0.62	0.75	0.64	0.71	0.59	0.72	0.75	0.80	0.55	0.60	0.59	0.53

TABLE 3 | Subscale descriptives.

Subscale	**Initial item set**	**Final item set**	**Score mean (SD)**	**Reliability**		**Difficulty Mean (SD)**	**Discrimination mean (SD)**
				Cronbach's α 95% *CI*	**McDonald's ω 95% *CI***		
Knowledge	18	9	6.98 (1.92)	0.73 (0.55, 0.86)	0.78 (0.71, 0.90)	0.78 (0.20)	0.59 (0.08)
Stigma	14	6	4.74 (1.47)	0.72 (0.51, 0.84)	0.78 (0.63, 0.91)	0.79 (0.16)	0.67 (0.03)
Help-seeking	16	13	10.99 (2.68)	0.84 (0.70, 0.93)	0.98 (0.83, 1.00)	0.85 (0.10)	0.61 (0.08)
Coping	11	9	7.55 (2.14)	0.85 (0.73, 0.92)	0.89 (0.8, 0.95)	0.84 (0.13)	0.70 (0.07)
Family	22	13	11.16 (2.75)	0.87 (0.71, 0.94)	0.94 (0.68, 1.00)	0.86 (0.09)	0.66 (0.08)

assumptions of Mokken scale analysis, this suggests that the sum score for each subscale can be used to reliably order respondents on the construct measured by the subscale.

Each one of the subscale item sets were then scaled using a Rasch modeling approach. For all the subscales, model and person fit indicators suggested that this parametric scaling approach was not appropriate with the set of subscale items and the sample of youth. An iterative approach to scale refinement using the Rasch model was used, but the results of each subsequent analysis flagged multiple items and persons and produced statistics indicating misfit even after prior misfitting items were removed from the analysis. No suitable subset of items was found to have acceptable Rasch model fit for any of the subscales.

Various characteristics of the subscales, including estimates of internal consistency are shown in **Table 3**. In addition to the point estimate of Cronbach's α, bootstrapped 95% confidence intervals are provided to give an idea of the level of uncertainty of this point estimate. All confidence intervals include 0.8, the typical reliability target generally desirable for measurement scales. Coefficient ω produced slightly higher reliability estimates and also indicated acceptable reliability given the 95% confidence intervals.

The distribution and descriptive statistics of the subscales show a fair amount of variability among scores. The K and S subscales showed the least score variation, while the HS and F subscales show the most variation in scores. The means and distributions of the scores for all subscales indicate that the youth found the subscales to be a bit on the easy side, and generally showed that the youth were able to correctly answer a majority of the items. The subscales also look similar with respect to the average item difficulty (proportion of respondents correctly answering an item) with all having an average difficulty above 0.77 with moderate variation. The frequency distributions of subscale scores are all negatively skewed, which is consistent with the mean difficulties exhibited by all subscales. A ceiling effect was seen for the C (44%), HS (31%), and F (34%) subscales, while a floor effect is not an issue for any subscale. Item discrimination, the measure of the extent an item can distinguish between low and high scorers, was averaged across the items within a subscale; all subscales showed acceptable item discrimination values.

DISCUSSION

Overview

This paper explored the development of a new scale to measure youth mental health literacy in youth ages 11–14. The scale includes all of the Jorm (3) components of knowledge of common mental health disorders, recovery strategies, mental illness stigma, as well as help seeking for self and others in the event of possible mental illness symptoms. This scale could be useful for the general population. We also included coping and family subscales. Since it is possible that children with a family member with a mental illness may comprise almost one of four youth (21), it is likely that the family information will be useful for many within the general population at present and in the future. To our knowledge, this is one of the first youth mental health literacy scales with a reading level appropriate for youth ages 11–14 that covers the full range of mental health literacy components.

The YMHL scale development aligns with calls to develop youth mental health literacy programs (1, 33), particularly given the rising rates of youth mental illness symptoms and suicide (9, 11). The assumptions underlying the need for mental health literacy interventions, and accompanying scales, are that youth can benefit from the application of risk-reducing and resilience-promoting developmental resources. These assumptions are consistent with Masten's (31) risk and resiliency theory. These resources include coping skills, social support, community resources, and access to accurate knowledge for dealing with developmental risks. The latter fits especially well with health literacy aiming to prevent, ameliorate, or manage a health disorder. Standardized scales need to be part of the measurement of the extent of effectiveness of youth mental health programs. The idea is to help move these programs toward becoming evidence-based. Evidence-based programs are more likely to be further funded, tested, revised, and disseminated.

The subscales developed and initially validated provide a means to produce such evidence. The instruments went through a rigorous development process, in which the resultant items were vetted using a variety of psychometric approaches to validate the scales and provide information about the nature of these measures. Using the validation dataset, we found that the five different aspects of youth mental health literacy could be measured with some assurance of the validity of the use of resultant subscales, and with acceptable reliability given our sample data. There is some evidence that the subscales can distinguish between more clustered groups of respondents, but due to the nature of the items and responses, a more finely-grained measure using a Rasch model scaled version of the subscales was not possible.

Limitations and Strengths

The greatest limitation of this study is a small sample restricted by the onset of the COVID-19 and the need to cut the sample into youth ages 11–14 and youth 15–18 to accommodate developmental knowledge differences. The ages 11–14 scale herein was developed with data drawn from youth attending one of two middle schools in one Midwestern state. Clearly, the scale is newly emerging and requires further testing with more rigorous designs, increased geographic diversity, and especially, larger samples. Larger samples would also allow us to compare results that we obtained using what is likely an inadequate sample size for Mokken scaling analysis and the Rasch model. While we anticipate that Rasch analysis would likely yield the same results with a bigger sample, we would be interested to see if Mokken scaling applied to a larger sample may show more score variability. While the small sample size our results are based on warrants a cautious application of our findings regarding the five subscales, these findings form a basis for continued work on measuring mental health literacy. It is also not known to what extent the scale is useful for particular youth mental health programs so this is an important consideration. The scale needs to align with the YMHL program aims and content.

The greatest strength of the scale lies in its strong foundation studies and stakeholder inclusion. The content was built from findings of numerous needs assessments conducted across many years. Youth input was regularly included in content development. Other stakeholder input was drawn from parents, educators, child welfare professionals, and mental health services providers (2, 6, 7, 36, 64). Family content was drawn from input provided by youth that have a family member with a mental illness, a survey of international researchers in family mental health, and an intensive literacy study intended to flesh out the mental health literacy needs of youth with a family member with a mental illness (36, 37, 52).

Recommendations

The scale development process leads to recommendations for future research, practice, and policy. Future research should seek a larger sample, more geographically diverse settings, and data should be collected within more rigorous designs including those with control groups and random, or at least wait-list control, sampling. There is likely to be future needs to develop valid and reliable scales for an array of age ranges, e.g., children, young teens, older teens, young adults, and adults. For example, a YMHL scale for ages 15–18 will be forthcoming.

In order to achieve subscales capable of making more finely-grained measures and achieve more detailed distinction of differences in YMHL, future research will also draft and test additional items to be included and vetted in additional validation efforts. The items will aim to provide more variety of challenging questions as well as those targeted at the upper middle of the distribution of YMHL levels. The goal continues to produce an instrument that accurately measures the dimensions of YMHL yet does not present the burden of a large number of additional items.

The authors have requests for access to the YMHL scale from researchers across a number of nations; many of these require translation to a language other than American, Australian or British English (this may also require some translation). It is recommended that the scale translation process be rigorous. The translation of an instrument measuring constructs such as those here presents a formidable challenge for accurate translations. It is well-known and understood that a word for word translation of an instrument is an unacceptable practice. Translation must necessarily be that of the ideas and spirit of each of the items in a contextually accurate manner. The steps of translation must be iterative, and must incorporate a team approach that should include input from the target population of the scale. Furthermore, each translated scale becomes a brand-new scale that must be validated before use.

Recommendations for practice are to continue to develop, test, and revise mental health literacy programs for children, youth, and families. Programs need to continue to acquire and maintain evidence-based practice status. Key stakeholder input is important to further program development processes. Input from parents, educators, mental health, and especially youth, should be gathered and applied to program planning, content, delivery, and evaluation processes. Programs could be delivered in a number of settings but schools may be the best place to access children and youth. Future program development could focus on the specific mental health literacy knowledge needs of diverse groups and cultures. COPFMI mental health literacy programs should be a high priority, especially given youth higher levels of risk for acquiring a mental illness and because many of them live day to day with a family member with a mental illness.

Recommendations for policy are to include mental health literacy programs within the prevention and health promotion arm of a mental health services continuum. Educators, mental health providers, and other community services providers should advocate for funding for mental health literacy programs and research testing. It is even possible that mental health programming would be a regular part of educational programming across all levels of students. That could reduce mental illness stigma and promote earlier help seeking.

SUMMARY

The Youth Mental Health Literacy Scale for ages 11–14 is designed to measure mental health literacy among general population youth and those with a family member with a mental illness. We will continue to work on refining this scale with larger samples and continued stakeholder input. The scale is intended to yield youth mental health literacy outcome data that can help mental health literacy programs to build evidence-based programs that, in turn, may help prevent, delay, or ameliorate mental health disorders among youth. Perhaps 1 day mental health literacy will be a common health learning activity in schools and communities. It is hoped that the YMHL scale can help play a role toward that shifting paradigm.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Michigan State University Institutional Review Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

JR served as primary investigator for developing the scale based on 12 years of research, wrote scale questions and led the data collection for scale testing, and coordinated the entire manuscript and wrote much of background, part of the methods, and most of the discussion section. CG worked on needs assessment studies and helped write scale questions and wrote part of the background section and edited the manuscript. KK developed statistical findings and wrote most of the methods section and all of the findings section, as well as a small part of the discussion section. DC helped write scale questions and the collect scale testing data and wrote part of the background section and engaged in editing. All authors contributed to the article and approved the submitted version.

FUNDING

JR salary was partially supported by the USDA National Institute of Food and Agriculture and Michigan State University AgBioResearch.

ACKNOWLEDGMENTS

The authors thank Dr. Shane Costello from the Department of Educational Psychology at Monash University in Melbourne, Australia for his contributions in guiding early scale development and analyses.

REFERENCES

1. Kutcher S, Bagnell A, Wei Y. Mental health literacy in secondary schools: a Canadian approach. *Child Adolesc Psychiatric Clin.* (2015) 24:233–44. doi: 10.1016/j.chc.2014.11.007
2. Grové C, Reupert AE, Maybery DJ. Gaining knowledge about parental mental illness: how does it empower children? *Child Family Social Work.* (2015) 20:377–86. doi: 10.1111/cfs.12086
3. Jorm AF. Mental health literacy: empowering the community to take action for better mental health. *Am Psychol.* (2012) 67:231–43. doi: 10.1037/a0025957
4. American Association of Child and Adolescent Psychiatry (AACAP) (2019). "Stress management and teens," *Facts for Families, 66*, 1–3. https://www.aacap.org/AACAP/Families_and_Youth/Facts_for_Families/FFF-Guide/Helping-Teenagers-With-Stress-066.aspx
5. Milkkelsen HT, Haraldstad K, Helseth S, Skarstein S, Småstuen MC, Rohde G. Health-related quality of life is strongly associated with self-efficacy,

self-esteem, loneliness, and stress in 14-15-year-old adolescents: a cross-sectional study. *BMC.* (2020) 18:352–69. doi: 10.1186/s12955-020-01585-9

6. Riebschleger J, Tableman B, Rudder D, Onaga E, Whalen P. Early outcomes of a pilot psychoeducation group intervention for children of a parent with a psychiatric illness. *Psychiatr Rehabil J.* (2009) 33:133–41. doi: 10.2975/33.2.2009.133.141
7. Riebschleger J, Costello S, Cavanaugh DL, Grové C. Mental health literacy of youth that have a family member with a mental illness: outcomes from a new program and scale. *Front Psychiatry.* (2019) 10:2. doi: 10.3389/fpsyt.2019.00002
8. American Psychiatric Association. *Diagnostical and Statistical Manual of Mental Health Disorders (5th ed.)*. (2013). Author.
9. Centers for Disease Control and Prevention [CDC]. *Facts About Suicide*. (2021). Available at U.S. government site: https://www.cdc.gov/suicide/facts/index.html
10. United States Center for Disease Control (CDC) (2020). *Youth Risk Behavior survey: Data Summary and Trends Report 2009–2019*. Available at https://www.cdc.gov/healthyyouth/data/yrbs/pdf/YRBSDataSummaryTrendsReport2019-508.pdf
11. World Health organization [WHO]. *Adolescent Mental Health*. (2020). Available at: https://www.who.int/news-room/fact-sheets/detail/adolescent-mental-health
12. Kessler RC, Angermeyer M, Anthony JC, De Graaf R, Demyttenaere K, Gasquet I, et al. Lifetime prevalence and age-of-onset distributions of mental disorders in the World Health Organization's World Mental Health Survey Initiative. *World Psychiatry*. (2007) 6, 168–176.
13. Patel V, Flisher AJ, Hetrick S, McGorry P. Mental health of young people: a global public-health challenge. *The Lancet*. (2007) 369:1302–1313. doi: 10.1016/S0140-6736(07)60368-7
14. Colton CW, Manderscheid RW. Congruencies in increased mortality rates, years of potential life lost and causes of death among public mental health clients in eight states. *Prev Chronic Dis.* (2006) 3:1–14.
15. Ormel J, Von Korff M, Burger H, Scott K, Demyttenaere K, Huang YQ, et al. Mental disorders among persons with heart disease: Results from World Mental Health surveys. *Gen Hosp Psychiatry*. (2007) 29:325–34. doi: 10.1016/j.genhosppsych.2007.03.009
16. Soderstrom IR. Mental illness in offender populations: Prevalence, duty, and implications. *J Offender Rehabil.* (2007) 45:1–17. doi: 10.1300/J076v45n01_01
17. Desmarais SL, Van Dorn RA, Johnson KL, Grimm KJ, Douglas KS, Swartz MS. Community violence perpetration and victimization among adults with mental illnesses. *Am J Public Health.* (2014) 104:2342–9. doi: 10.2105/AJPH.2013.301680
18. Teplin LA, McClelland GM, Abram KM, Weiner DA. Crime victimization in adults with severe mental illness: comparison with the national crime victimization survey. *Arch Gen Psychiatry*. (2005) 62:911–21. doi: 10.1001/archpsyc.62.8.911
19. Corrigan PW, Watson AC. Understanding the impact of stigma on people with mental illness. *World Psychiatry*. (2002) 1:16–20.
20. Miller BF, Coffey MJ. Understanding suicide risk and prevention. *Health Affairs Policy Brief* (2021).
21. Maybery DJ, Reupert AE, Patrick K, Goodyear M, Crase L. Prevalence of parental mental illness in Australian families. *Psychiatric Bull*. (2009) 33:22–26. doi: 10.1192/pb.bp.107.018861
22. Campbell TCH, Reupert A, Sutton K, Soumya B, Davidson G, Middledorp CM, et al. Prevalence of mental illness among parents of children receiving treatment within child and adolescent mental health services (CAMHS): a scoping review. *Eur Child Adolesc Psychiatry*. (2019) 30:997–1021. doi: 10.1007/s00787-020-01502-x
23. Giannakopoulos G, Solantaus T, Tzavara C, Kolaitis. Mental health promotion and prevention interventions in families with parental depression: a randomized controlled trial. *J Affect Disord.* (2021) 278:114–121. doi: 10.1016/j.jad.2020.09.070
24. Jorm AF. We need to move from 'mental health literacy' to 'mental health action'. *Mental Health Prevent*. (2020) 18:200179. doi: 10.1016/j.mhp.2020.200179
25. Nicholson, J. Supporting mothers living with mental illnesses in recovery. In: Benders-Hadi N, Barber ME (eds). *Motherhood, Mental Illness and Recovery: Stories of Hope*. (2014). Springer International Publishing. p. 3–17.
26. Mowbray CT, Oyserman D. Substance abuse in children of parents with mental illness: Risks, resiliency, and best prevention practices. *J Prim Prev.* (2003) 23:451–82. doi: 10.1023/A:1022224527466
27. Powell RM, Nicholson J. Addressing risk factors among parents with serious mental illness: Commentary on Ostrow et al. *Psychiatric Serv (Washington, DC).* (2021) 72:466–7. doi: 10.1176/appi.ps.202000790
28. Gladstone BM, Boydell KM, McKeever P. Recasting research into children's experiences of parental mental illness: Beyond risk and resilience. *Soc Sci Med*. (2006) 62:2540–50. doi: 10.1016/j.socscimed.2005.10.038
29. Mordoch E, Hall WA. Children living with a parent who has a mental illness: a critical analysis of the literature and research implications. *Arch Psychiatr Nurs.* (2002) 16:208–16. doi: 10.1053/apnu.2002.36231
30. Serbin L, Karp J. The intergenerational transfer of psychosocial risk: Mediators of vulnerability and resilience. *Annu Rev Psychol.* (2004) 55:333–63. doi: 10.1146/annurev.psych.54.101601.145228
31. Masten AS. *Ordinary Magic*. New York, NY: Guilford Press (2014).
32. Bale J, Grové C, Costello S. A narrative literature review of child-focused mental health literacy attributes and scales. *Mental Health Prevent*. (2018) 12:26–35. doi: 10.1016/j.mhp.2018.09.003
33. Kutcher S, Wei Y, Costa S, Gusmão R, Sokauskas N, Sourander A. Enhancing mental health literacy in young people. *Eur Child Adolesc Psychiatry*. (2016) 25:567–9. doi: 10.1007/s00787-016-0867-9
34. Thompson A, Issakidis C, Hunt C. Delay to seek treatment for anxiety and mood disorders in an Australian clinical sample. *Cambridge Core.* (2012) 25:71–84. doi: 10.1375/bech.25.2.71
35. Robles CC, Bronstein J. Easing the path for improving health-seeking behavior in youth. *EClinicalMedicine.* (2020) 18:100256. doi: 10.1016/j.eclinm.2019.100256
36. Grové CJ, Riebschleger J, Bosch A, van der Ende P. Expert views of children's knowledge needs regarding parental mental illness. *Child Youth Serv Rev.* (2017) 79:249–55. doi: 10.1016/j.childyouth.2017.06.026
37. Riebschleger J, Onaga ET. Mental health consumer parents' recommendations for psychoeducation for their minor children. Special issue on parents with disability challenges. *Psychiatric Rehabil J.* (2014) 37:183–5. doi: 10.1037/prj000007
38. Beardslee WR, Solantaus TS, Morgan BS, Gladstone TR, Kowalenka NM. Preventive interventions for children of parents with depression: International views. *Med J Aust*. (2013) 199:S23–5. doi: 10.5694/mja11.11289
39. Beyond Blue. *Beyondblue*. (2019). Available at: http://www.beyondblue.org.au
40. Pihkala H, Cederström A, Sandlund M. Beardslee's preventive family intervention for children of mentally ill parents: a Swedish national survey. *Int J Mental Health Promot.* (2010) 12:29–38. doi: 10.1080/14623730.2010.9721804
41. Mulligan C, Furlong M, McGarr S, O'Connor S, McGilloway S. The Family Talk programme in Ireland: A qualitative analysis of the experiences of families with parental mental illness. *Front Psychiatry*. (2021) 12:783189. doi: 10.3389/fpsyt.2021.783189
42. Mcluckie A, Kutcher S, Wei Y, Weaver C. Sustained improvements in students' mental health literacy with use of a mental health curriculum in Canadian schools. *BMC Psychiatry.* (2014) 14:1–6. doi: 10.1186/s12888-014-0379-4
43. Perry Y, Petrie K, Buckley H, Cavanagh L, Clarke D, Winslade M, et al. Effects of a classroom-based educational resource on adolescent mental health literacy: a cluster randomised controlled trial. *J Adolesc.* (2014) 37:1143–51. doi: 10.1016/j.adolescence.2014.08.001
44. Watson AC, Otey E, Westbrook AL, Gardner AL, Lamb TA, Corrigan PW. Changing middle schoolers' attitudes about mental illness through education. *Schizophrenia Bull.* (2004). 30:563–572. doi: 10.1093/oxfordjournals.schbul.a007100
45. Wei Y, Kutcher S, Szumilas M. Comprehensive school mental health: an integrated "school-based pathway to care" model for Canadian secondary schools. *McGill J Educ.* (2011) 46:213–29. doi: 10.7202/1006436ar
46. Wei Y, Hayden JA, Kutcher S, Zygmunt A, McGrath P. The effectiveness of school mental health literacy programs to address knowledge, attitudes and help seeking among youth. *Early Interven Psychiatry*. (2013) 7:109–121. doi: 10.1111/eip.12010
47. Wei Y, McGrath PJ, Hayden J, Kutcher S. Mental health literacy measures evaluating knowledge, attitudes, and help-seeking: a scoping review. *BMC Psychiatry*. (2015) 15:291. doi: 10.1186/s12888-015-0681-9

48. Wei Y, McGrath PJ, Hayden J, Kutcher S. Measurement properties of tools measuring mental health knowledge: a systematic review. *BMC Psychiatry*. (2016) 16:297. doi: 10.1186/s12888-016-1012-5
49. van Doesum KTM, Riebschleger J, Carroll J, Grové CJ, Lauritzen C, Murdoch E, et al. Successful recruitment strategies for prevention programs targeting children of parents with mental health challenges: an international study. *Child Youth Serv*. (2016) 37:156–74. doi: 10.1080/0145935X.2016.1104075
50. O'Connor M, Casey L. The Mental Health Literacy Scale (MHLS): A new scale-based measure of mental health literacy. *Psychiatry Res*. (2015) 229:511–6. doi: 10.1016/j.psychres.2015.05.064
51. Reavley NJ, Mackinnon AJ, Morgan AJ, Jorm AF. Stigmatizing attitudes towards people with mental disorders? A comparison of Australian health professionals with the general community. *Aust N Z J Psychiatry*. (2014) 48:433–41. doi: 10.1177/0004867413500351
52. Riebschleger J, Grové CJ, Cavanaugh D, Costello SC. Mental health literacy content for children of parents with a mental illness: Thematic analysis of a literature review. *Brain Sci*. (2017) 7:1–19. doi: 10.3390/brainsci7110141
53. Marinucci A, Grové C, Allen K-A, Riebschleger J. Evaluation of a youth mental health literacy and action program: protocol for a cluster controlled trial. *Mental Health Prevent*. (2021) 24:200–216. doi: 10.1016/j.mhp.2021.200216
54. Mokken RJ. A theory and procedure of scale analysis. *De Gruyter*. (1971). doi: 10.1515/9783110813203
55. Sijtsma K, Meijer RR, van der Ark LA. Mokken scale analysis as time goes by: an update for scaling practitioners. *Pers Individ Dif*. (2011) 50:31–7. doi: 10.1016/j.paid.2010.08.016
56. Sijtsma K, Molenaar IW. *Introduction to Nonparametric Item Response Theory*. (2002). Newcastle upon Tyne, UK: Sage. doi: 10.4135/9781412984676
57. Rasch G. Probabilistic models for some intelligence and attainment tests. *Danish Inst Educ Res*. (1960).
58. Loevinger J. The technique of homogeneous tests compared with some aspects of 'scale analysis' and factor analysis. *Psychol Bull*. (1948) 45:507–29.
59. Mislevy RJ. Recent development in the factor analysis of categorical variables. *J Educ Stat*. (1986) 11:3–31.
60. Meijer RR, Sijtsma K, Smid NG. Theoretical and empirical comparison of the Mokken and the Rasch approach to IRT. *Appl Psychol Meas*. (1990) 14:283–98. doi: 10.1177/014662169001400306
61. Iacobucci D, Duhachek A. Advancing alpha: measuring reliability with confidence. *J Consum Psychol*. (2003) 13:478–87. doi: 10.1207/S15327663JCP1304_14
62. McDonald RP. *Test Theory: A Unified Treatment*. (1999). Mahwah, N.J.: L. Erlbaum Associates.
63. Revelle W, Zinbarg RE. Coefficients alpha, beta, omega, and the glb: Comments on Sijtsma. *Psychometrika*. (2009) 74:145–54. doi: 10.1007/s11336-008-9102-z
64. Grové C, Reupert A, Maybery D. The perspectives of young people of parents with a mental illness regarding preferred interventions and supports. *J Child Fam Stud*. (2016) 25:3056–65. doi: 10.1007/s10826-016-0468-8

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Advantages of publishing in Frontiers

OPEN ACCESS
Articles are free to read
for greatest visibility
and readership

FAST PUBLICATION
Around 90 days
from submission
to decision

HIGH QUALITY PEER-REVIEW
Rigorous, collaborative,
and constructive
peer-review

TRANSPARENT PEER-REVIEW
Editors and reviewers
acknowledged by name
on published articles

Frontiers
Avenue du Tribunal-Fédéral 34
1005 Lausanne | Switzerland

Visit us: www.frontiersin.org
Contact us: frontiersin.org/about/contact

REPRODUCIBILITY OF RESEARCH
Support open data
and methods to enhance
research reproducibility

DIGITAL PUBLISHING
Articles designed
for optimal readership
across devices

FOLLOW US
@frontiersin

IMPACT METRICS
Advanced article metrics
track visibility across
digital media

EXTENSIVE PROMOTION
Marketing
and promotion
of impactful research

LOOP RESEARCH NETWORK
Our network
increases your
article's readership