PARENTS WITH MENTAL AND/OR SUBSTANCE USE DISORDERS AND THEIR CHILDREN

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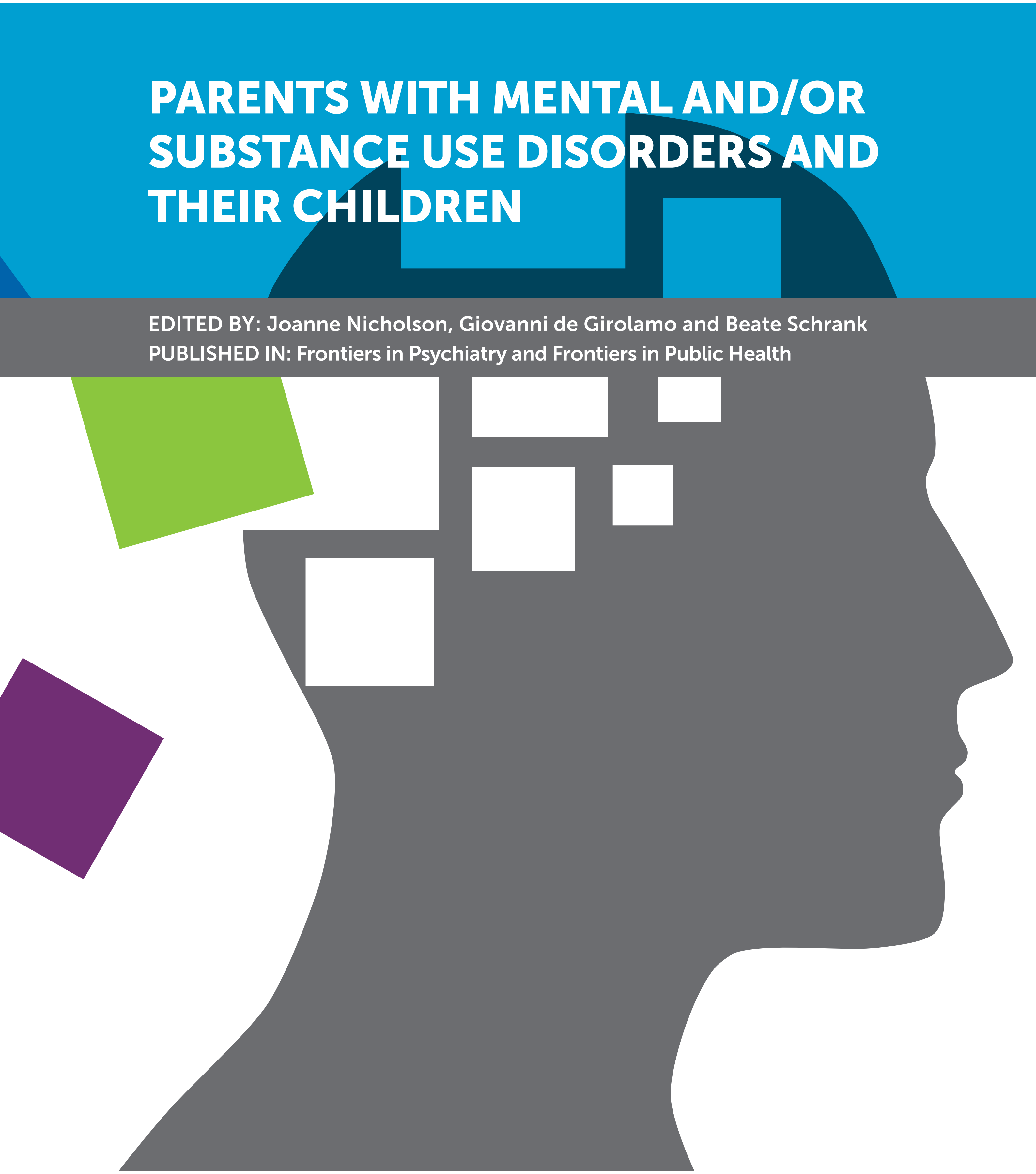

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PARENTS WITH MENTAL AND/OR SUBSTANCE USE DISORDERS AND THEIR CHILDREN

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EDITORIAL
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Editorial: Parents With Mental and/or Substance Use Disorders and Their Children

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Keywords: parents with mental illness, children of parents with mental illness, intergenerational transmission of mental illness, risk and protective factors, systems change, parents with substance use disorders, children of parents with substance use disorders

Editorial on the Research Topic

Parents With Mental and/or Substance Use Disorders and Their Children

INTRODUCTION

Families living with parental mental and substance use disorders face considerable biopsychosocial and, oftentimes, socioeconomic challenges, with complex pathways to mitigating risk, enhancing resilience in children and youth, and supporting recovery in adults who are parents. This special issue contains the latest knowledge on a) the prevalence of parental psychiatric disorders and children who may be affected, and relationships among risk and protective factors and outcomes for children, youth, and parents across the lifespan, including the perinatal period; b) intervention development, implementation, and testing at the individual practitioner, parent, child, or youth level, as well as recommendations for making change at the national level; and c) innovative measurement and methodological developments, including the protocols of studies currently underway. The special issue comprises 26 papers, representing the contributions of nearly 100 investigators from 15 countries in Europe, Asia, and North America. The research studies embrace diverse designs and methods; and both primary data collection with providers, parents, children, and youth, and secondary analyses of national and registry data sets. Several papers describe innovative research measures and methods, exciting developments in the field. Review and publication of the protocols of current studies serve to enhance research integrity and rigor, and guide next steps in future studies.

THE PREVALENCE OF PARENTAL MENTAL ILLNESS AND RELATED CHARACTERISTICS AND CONDITIONS

How many individuals in treatment in Adult Mental Health Services (AMHS) have children younger than 18 years of age who may need support or benefit from intervention? So far, data to answer this simple question have been sparse and inconsistent. Ruud et al. have obtained registry data from 23,167 receiving outpatient treatment in Norway at 107 AMHS and found that among

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them 36% had children under 18 years of age. Referrals to health and social services were made for children of one of three outpatients (with young children), while for a substantial proportion of these children it was unknown whether they had met or unmet needs. This finding suggests that the proportion of patients in treatment who have children who may need support is relevant, and requires appropriate actions.

Also, in Norway, Reedtz and co-investigators assessed a large sample (n = 422) of parents with mental illness receiving treatment and their young children (n = 581), and found that three quarters of them (76.2%; n = 526) were living with the ill parent. One third of the sample (32.5%; n = 170) lived with a single ill parent and with siblings, full-time or part-time. Younger children did not receive any information about their parents' disorders. Overall, this study confirms that a large percent of offspring spend a substantial amount of time with ill parents, and when parents are separated or divorced the contact with the ill parent may become particularly intensive, with an increased exposure to potentially stressful situations.

In their contribution, Moscoso et al. show that, in the family history of adolescents diagnosed with borderline personality disorder, there may be parental suicidal attempts, and the number of suicidal attempts is related to an increased severity of the adolescent's disorder. Children of parents who have attempted suicide, or completed suicide, are a particularly vulnerable population, who should receive targeted preventive interventions. Unfortunately, this rarely happens, even in countries with well-developed mental health services. This finding points to the need for a careful reconfiguration of services to enable effective response to potential users' needs.

An important study on a large sample of over 2,200 twins born in the United Kingdom in 1994–95 and followed up to the age of 18 has been conducted by Riches and collaborators. The authors assessed both the twins' mothers, when the children were ages 10 and 12, and the children themselves at ages 12 and 18. They assessed a variety of potential protective factors in relation to the development of sub-clinical psychotic phenomena among those children of mothers suffering from psychotic disorders. The authors found that good cognitive functioning, living in a more socially cohesive neighborhood and higher levels of perceived social support were independently protective against early psychotic symptoms among children of mothers with psychosis at ages 12 and 18. This underscores the importance of psychosocial factors in buffering the biological risk associated with having a mother with a severe psychotic disorder.

Three papers deal with perinatal issues affecting mother and child emotional life. Aktar et al. reviewed the literature analyzing early exposure to parental psychopathology, during the pregnancy and in the first *post-partum* year, and its association with child psychological functioning beyond the *post-partum* period, up to adulthood years. They conclude that early interventions targeted in particular to mothers showing severe emotional disturbances in the perinatal period may have beneficial effects on the child for periods which extend to adult life. Once again, the importance of early and timely interventions is confirmed, to effectively intervene with mental disorders at their onset, stopping the chain of intergenerational transmission of these pathological conditions.

Risk factors in the perinatal period and their effects on newborns were the focus of two studies. Ichikawa and colleagues have evaluated the association between low levels of prenatal alcohol exposure (PAE) and children's health. They assessed a large sample of children under 18 years old who have siblings (n = 1,600) and their mothers. Children were evaluated with the CBCL, while mothers' PAE was assessed retrospectively. Low PAE was associated with children's anxiety, internalizing problems and overall health problems, pointing to the importance of screening mothers for PAE to adopt timely preventive interventions and increase the odds of healthy child development.

Sheeba B et al. screened 280 pregnant mothers attending an antenatal clinic in Bangalore, India. Over 1/3 of the sample were found to screen positive for prenatal depression, and the risk was much higher for women exposed to domestic violence or to stressful life events, again highlighting the need for the delivery of appropriate screening interventions targeting emotional well-being among pregnant women. Screening and assessment can promote the implementation of timely interventions, with a beneficial double effect on the mother and on the newborn.

This set of papers highlights the role played by a parental mental disorder in the overall functioning of the family system, and once again underscores the need for appropriate mental health prevention, intervention and promotion. Even simple approaches can have an enduring effect contributing to children's outcomes over the life span.

INTERVENTION DEVELOPMENT, IMPLEMENTATION, AND TESTING

A number of contributions to this special issue represent efforts to develop, implement, and test interventions involving practitioners, as well as parents and children or youth. In Portugal, van Doesum and colleagues targeted professionals from a hospital psychiatric service received training in the Child Talks intervention. Professionals' reports regarding attitudes, knowledge, confidence, and organizational support improved significantly, as measured by the Family-Focused Mental Health Practice Questionnaire prior to training and 10 months later. In Hafting et al.'s paper, the authors integrate findings from a series of studies on the capacity of General Practitioners (GPs) to identify and provide support to the children of ill and substance-abusing parents. Given their finding that both parents and GPs are ambivalent about addressing these issues during consultation visits, the authors make recommendations for GPs to gain knowledge about the family situation.

Norwegian colleagues, Lauritzen et al., describe 5-year follow-up findings from a study in which two interventions were implemented to support healthcare professionals in identifying and providing support for children of patients within AMHS. While some changes in clinical practice were found, the authors conclude that practice change is a very time-consuming process. In the USA, Nicholson and Valentine interviewed key informants as the first step in designing a model of parent-peer supports for parents with psychiatric disorders. While peer supports are burgeoning in the USA in other domains, the authors' findings raise key issues in practice with parents, including the training needs and organizational infrastructure to support both parent-peer specialists as well as parents themselves.

Children and adolescents living with parental mental illness, their families and professionals who work with them were engaged in co-producing an 8-week group intervention, Young SMILES, for 6- to 16-year olds and their parents, as described by Gellatly and collaborators. This leading UK effort lays the groundwork for meeting the needs of a broad range of ages and levels of need, and for rigorous research to evaluate the impact. Reupert and colleagues target young adults aged 18 to 25 years, with a parent with a mental health and/or substance use disorder, in laying the groundwork for the development for an online group intervention in Australia, "mi.spot." They suggest implementation considerations and directions for future testing. Potijk and colleagues in the Netherlands addressed parents' hesitancy to let children participate in preventive programs through a pilot study in which a psychoeducation program on parenting and mental illness for parents was developed. They make recommendations regarding implementation as well as for future research. Importantly, mothers' identity is placed in the context of a personal recovery paradigm by Australian authors, Hine et al. The authors suggest that services may promote recovery by supporting the enhancement of their self-concept associated with mothering, in addition to attending to other attributes and roles.

Systems-level change is addressed by Isobel et al. in their study of the stories of change provided by an international sample of systems' change experts from 16 countries. While the issues facing children, parents and families may share similarities across countries, the pathways to systems-level change among countries have unique aspects as well as areas of overlap. This paper provides an interesting global perspective. In Finland, the Let's Talk model was implemented in three municipalities, supporting stakeholders and families in working together to enhance outcomes for children. Niemela and colleagues compared data from these municipalities with national data, and demonstrated a significant decrease in referrals to child protections services.

RESEARCH INNOVATION

This issue also provides insight into cutting-edge research endeavors in the field. *Postpartum* psychiatric disorders such as depression, anxiety, or psychosis occur in about 15% of mothers and negatively affect their caregiving behavior at an early stage of child development. In response to shortcomings of existing measurement tools, Heinisch et al. have developed an adapted method to assess maternal sensitivity based on methods recently approved in attachment research. Initial application of the new measurement tool showed different deficits in mothers' caregiving behavior depending on diagnosis. At the same time, many mothers were well capable of behaving in sensitive manner toward their child despite their illness. The new method is open for use with pending reliability testing.

Children of parents with a mental illness may be challenging to reach for research and interventions. Using a case study analysis, Grove explores social networking sites as a medium to include at-risk youth and their families by removing accessibility barriers. Ethical considerations and limitations of this promising means of engagement are discussed. Riebschleger and colleagues provide further validation of the Knowledge of Mental Illness and Recovery (K-MIR) scale to examined mental health literacy levels and coping outcomes for youth, which they used before and after a school-based mental health literacy program, Youth Education and Support (YES). Their study also shows a positive effect of the intervention on mental health literacy and coping strategies.

Research currently underway provides promise of exciting future findings. Thorup and co-investigators introduce the follow-up of their large registry based cohort study, which will assess 522 children born to parents with schizophrenia, bipolar disorder and controls as they turn 11 years of age. This study will determine whether the children at familial risk reveal delayed developmental courses, but catch up at age 11, or whether the discrepancies between the groups have grown even larger. It will also shed light on aspects of resilience and neurobiological outcomes.

The Village project is described by Christiansen et al., in which they seek to improve child development and wellbeing outcomes for children of parents with a diagnosed mental illness. The project encompasses co-development, implementation, and evaluation of a practice approach to the early identification and collaborative care, through establishing child-focused support networks and using innovative open innovation science (OIS) approaches.

Christiansen and colleagues describe the part I protocol for a randomized controlled trial comparing parental cognitive behavioral therapy/CBT with CBT + Parenting Program for parents with a mental disorder, flanked by four add-on projects that apply behavioral, psychophysiological, and neuro-imaging methods to examine potential moderators and mediators of risk transmission. This large, multicenter study provides opportunity to test the components of the transgenerational transmission of mental disorders. The part II protocol for the study is provided by Stracke and colleagues.

Steardo and co-investigators are conducting a *randomized controlled trial* for pregnant women with high *postpartum* depression scores. Women will be randomized to receive a psychoeducational intervention or a best practice control condition. The authors will not only develop an informative package for pregnant women and promote a screening program for women with *postpartum* depression, it will also identify factors associated with a higher risk of developing perinatal or postnatal depression, and evaluate the effectiveness of the psychoeducational intervention for reducing symptoms during pregnancy.

Finally, Reedtz and an international group of colleagues introduce the creation of a research and intervention model to promote large-scale implementation and evaluation of generic, brief interventions for children of parents with mental disorders. This multi-site international protocol allows for assessment of both minor children aged 6 to 18 years and their parents, and draws from both electronic patient journals as well as primary data collection using standardized measures. Child Talks+, developed in the Netherlands, will be implemented in participating countries.

CONCLUSION

The papers in this special issues represent a significant contribution to building the evidence base of effective interventions for family members who are living with or who are parents with mental and substance use disorders, and lay the groundwork for future innovation in research, screening and assessment, intervention development, and testing. The evidence may be applied to effect innovation or change in policy and practice, and suggests the need for widespread knowledge translation and dissemination, highlighting the significant potential to improve public mental health in countries throughout the world.

AUTHOR CONTRIBUTIONS

JN, GdG, and BS contributed substantially to the development of this special issue, and to the review and editing of all papers included. JN, GdG, and BS actively contributed to the preparation and review of the editorial manuscript.

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ORIGINAL RESEARCH
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Adult Mental Health Outpatients Who Have Minor Children: Prevalence of Parents, Referrals of Their Children, and Patient Characteristics

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Background: A strong connection exists between parental mental illness and lifetime mental health risk for their children. Thus, it is important to determine, when parents attend for treatment for their illness, the prevalence and characteristics of parents with a mental illness and identify referral actions for their children. Previous studies indicate that 12–45% of adult mental health service patients are parents with minor children. There is a need for studies with larger sample sizes that investigate the prevalence and characteristics of parents, and factors associated with referral actions for their children.

Method: Data on 23,167 outpatients was drawn from a national census study across 107 Norwegian adult mental health outpatient clinics during 2 weeks in April 2013. Clinicians identified various socio-demographic characteristics of patients who were parents and referral actions for their children.

Results: Eight thousand thirty-five (36%) of outpatients had children under 18 years. Thirty-one percent were provided with referrals for their children and 58% were reported to have children with no need for referral. Three percent were reported to have children with unmet needs who were not referred. There were missing data on children's needs and referral actions for 8% of parents. Patients who care for minor children were more likely to be refugees, and less likely to be single, male, not own a house/apartment, and have a schizophrenia spectrum illness or substance use disorder. Children were more likely to be referred when their parent was single, with no income from paid work, low education, not owning house/apartment, poor family network, long outpatient treatment, and an individual care plan; and less likely for men with a moderate or less severe mental illness. Children were referred to child protection agencies, child and adolescent mental health services and school psychological/pedagogic services.

Discussion: The prevalence of outpatients with children is similar to other studies. Referrals were made for children of one third of outpatients with minor children. Needs and referrals of children was unknown for one in ten outpatients. Mental health outpatient clinics must improve procedures to identify parenting status and ascertain and act on children's needs.

Keywords: prevalence of parents with mental illness, children of parents with mental illness, mental health outpatients, patient characteristics, needs of care, referrals

INTRODUCTION

This paper presents data from a national census of patients in adult mental health outpatient clinics in Norway. It provides prevalence of outpatients who care for children under the age of 18 years, prevalence of parents whose children are considered by clinicians to require further support and are referred to external agencies, and how parents' socio economic and clinical characteristics influence prevalence and type of referrals made by clinicians for the children. This information can be used by policy makers and managers to strengthen policy, as well as to support clinicians to better identify parents, determine their children's needs and refer the children to appropriate services.

Parental mental illness impacts on the functioning of the family unit and poses a risk to the healthy development of children. Compared to other children, those growing up with a parent with a mental illness are at risk of a range of adverse behavioural, developmental and emotional outcomes (1–3). Bell et al. (4) found that children of parents who had been hospitalized for a mental illness were much less likely to be school ready. Difficulties with schooling have also been shown in Sweden where Hjern et al. highlighted that twice as many children whose parents had been hospitalised for a mental illness had lower school results and were not able to start secondary education, compared to children without parental mental illness (5). However, it was also estimated that most of the risk was associated with three contributing social factors (needing social support, low parental education level, parental separation/divorce), or an interaction between parental illness and social problems. Foster et al. (6) identified parental functioning and parental mental illness among risk/protective factors for children according to the child.

Evidence suggests that children whose parents have a mental illness have almost double the chance of developing a mental illness themselves (5, 7). In a Swedish study of 535,000 children up to 30–35 years of age, those who grew up with substance abuse and/or mental illness in the family and who also required social welfare were identified as an extreme high-risk group. This group of children reported considerably higher levels of mortality, mental illness, substance abuse, criminality, and were recipients of social welfare benefits, while having low workforce participation (5). In Norway it is estimated that 25% of mental illnesses among adolescents, and 10% of early death, is associated with parental mental illness (8).

A recent systematic literature review found that between 12 and 45% of all patients attending adult mental services were parents (9). Four studies in the review found prevalence rates from 36 to 38% (10–13), suggesting that a substantial minority (one third) of patients using adult mental health services are parents (9). However, a limitation of these audit-style studies (12, 14) is that they commonly focus on a single regional adult mental health service with modest sample sizes. Other studies have assumed an epidemiological approach that projects findings from a representative sample to approximate the whole population (15) or utilised already collected national data sets (16). In Norway it has been estimated that 23.1% of children have a parent with a moderate or severe mental illness that may affect their daily life (8). Among children born in Sweden 1987–1989, 7.8% of minor children have a parent admitted to hospital due to psychiatric illness and/or substance abuse. Further, a survey from 2007 to 2011 found that parental mental health problems, not sufficiently severe to require hospitalizations, were relatively common, with 18.1% of parents to children aged 10–18 years reporting nervousness, anxiety or worry (5).

To the best of our knowledge only one study has examined whole-of-service data for parental mental health prevalence. In the early 2000s, Maybery et al. (16) found that in one year, 20% of adult mental health patients in the Australian state of Victoria were parents. In a follow up study, the authors examined state-wide adult mental health outpatient data over a 4 years period and found that 19–20% of almost 60,000 outpatients were parents (17). While the study provided valuable prevalence data, it failed to go further and illustrate information about the parents (such as gender, diagnosis) nor about the needs of their children. Consequently, there is a lack of whole of population knowledge about the prevalence of adult patients in mental health or addiction services who are parents caring for children. Such information would provide important data that can be used to inform service policy, intake procedure, and professional development of clinicians.

When assessing the risks to such children, it is essential to obtain information about the parent, including diagnosis, gender, socio economic context, and family networks, as well as other circumstances which may influence children's experiences and outcomes. For example, parents who have an anxiety disorder are less likely to grant their children autonomy and more likely to demonstrate lower levels of sensitivity (18), while children of parents with psychosis might be directly involved in a parent's delusions (19). After controlling for other risk factors at birth, Hammond et al. (20) found that two thirds of infants of mothers with a psychotic disorder were reported to child protection services. This was four times that of infants of mothers without a mental illness. Infants of mothers with a mood disorder were twice as likely to be referred to child protection services compared to infants of mothers without a mental illness.

The family environment, including the presence of marital discord, the presence or absence of the other parent and the availability of social support to the family may also influence the level of risk exposure to the children (21). There are also differences between the experiences and needs of mothers and fathers with a mental illness, in terms of custody arrangements and who they call on for help (22). These family circumstances have important implications for the development of risk-assessment tools and case-related decision making and accordingly need to be identified from the outset.

Other studies have considered parent and family variables when determining prevalence. A 4-year census in one Australian adult mental health service identified that a majority of female outpatients were parents (14). Nearly half were married, with around a third separated or divorced. Approximately half of parents reported a good level of social support. The most prevalent diagnoses were mood and psychotic disorders, followed by anxiety disorders. Approximately 60% of dependent children lived with parents, including 34–56% of children whose parents had a psychotic disorder. Around a third of children were identified as having child protection involvement (14). These findings are broadly consistent with those from another Australian adult cross-sector service census, with the majority of identified parents being female and over half of being single (23). In that audit, the most common parental diagnoses were psychotic and mood disorders, with ∼40% of parents living with their children or another family member (23). Both audits however were limited by moderate sample sizes (average yearly sample size around 800 limited to specific catchment areas).

It is important that adult mental health services are responsive to the needs of children in these families, which at a minimum would involve identifying children, assessing their needs, and as required, referring them on to appropriate services (24–26). However, little is known about clinicians' actions in adult mental health outpatient services in relation to detection, follow up and referral of patients' children. Some small-scale qualitative studies (25, 27, 28) have shown that clinicians working in adult mental health services struggle to balance the needs of both parents and children and do not routinely refer children on to appropriate services. However, further research is required to generalise and extend these findings.

Research has clearly demonstrated the benefits of family focused practice for parents with a mental illness, their children and other family members (29, 30). Two controlled trials found that family focused approaches significantly improved mental health outcomes for parents (31, 32). A meta-analysis found that family focused preventive interventions reduced the risk of children developing the same mental illness as their parent by 40% (30). Consequently, to inform mental health service policy and practice it is important to identify the number of outpatients attending adult mental health services who are parents with minor children.

CONTEXT OF THE STUDY

In Norway, specialized mental health services are organised in general hospital services across 19 health trusts. The division of mental health and addiction services in each health trust has inpatient and outpatient services for children and adolescents, adults, and older persons. Community mental health centres for adults include all outpatient mental health clinics, mobile teams, day units and almost half of the mental health inpatient beds in Norway (33–35). Almost all mental health services are public, but a few hospitals and CMHCs are owned by private trusts.

The Norwegian Health Personnel Act of 2010 requires health personnel to ascertain whether a patient has minor children and if so, to record this in their patient record. The law also stipulates that clinicians should talk with the patient about their children's needs and offer to give information and guidance. Within the limits of confidentiality, clinicians might invite children to be involved in conversations about their parents' illness, treatment, and the possibility of visiting the service treating their parent. In addition, the law stipulates that clinicians assess the needs of the children and refer children to relevant services such as child protection agency, CAMHS, educational-psychological services in schools and family counselling offices as required (36).

AIMS AND RESEARCH QUESTIONS

The study aimed to determine the number and characteristics of adult outpatients who care for children under the age of 18, from a national outpatient census in mental health services in Norway. A further aim was to identify the prevalence of outpatients with minor children where the clinician identified a need for referral of children to an external agency.

The paper addresses the following research questions: 1. What is the prevalence of outpatients in adult mental health clinics who care for children under the age of 18 years? 2. What are the socio-demographic and clinical characteristics of these outpatients, compared to outpatients who do not care for minor children (in terms of diagnosis, gender, age, marital status, income, education level, housing, refugee status, and country of birth)? 3. For what prevalence of outpatients with minor children do clinicians identify a need for referral of children to an external agency for children, and what agencies are the children referred to? 4. What patient characteristics are associated with referral of children to external agencies?

MATERIALS AND METHODS

Design

The design of the census was a cross-sectional study of outpatients seen by all outpatient clinics and mobile teams in adult Norwegian mental health services during 2 weeks in April 2013. The census was commissioned and financed by the Norwegian Directorate of Health. The work was undertaken by the SINTEF research foundation. The study was approved by the Regional Committee for Medical and Health Research Ethics (reg.no. 2012/848).

Sample

The sample consisted of data on 23,167 adult outpatients seen by 107 of the 110 mental health outpatient clinics in Norway. The prevalence of outpatients included was 60% based on data from the National Patient Register indicating that the total number of outpatients during the 2 weeks was 38,904. The clinics that did not participate were small and cited a lack of time for not participating; these comprised 1% of all outpatient consultations during the 2 weeks.

Variables

The census form included outpatients' socio-demographic data (gender, age, and marital status, main source of income, highest education, housing situation, refugee status, and country of birth), main mental diagnoses using ICD-10 (for substance use disorders secondary diagnosis is also included, as this is often listed as a secondary diagnosis for patients with both a mental and a substance use disorder), and the following questions about patients' children: (1) Does the patient care for children under 18 years? (yes/no/unknown). (2) If yes, number of children. (3) Have measures been taken to follow up any of the children? (available response being; yes; no and no need to refer; no and a need to refer; unknown). (4) What agencies were children referred to (possible to mark more than one of the listed agencies).

Data Collection

All outpatients who had one or more consultations in 2 weeks (15–28 April 2013) were targeted. Several months prior to the census, service managers and clinicians received information about the census and the data collection procedures. Data were collected on hard copy forms. The clinicians completed one anonymous form for each outpatient. They were encouraged to invite the patients to participate in filling in the form, and 57% of the patients participated. The completed forms were returned to a data collection company, who scanned the forms and delivered data files to the project team.

Data Analyses

Descriptive statistics, chi square testing and logit regressions were computed with STATA 15.

RESULTS

Prevalence of Outpatients Who Care for Children Under the Age of 18

Of the 23,167 registered outpatients, information on gender and children was given for 22,398 (97%) of patients. A total of 8,035 (36%) of these had children under age 18, with 5,729 (71%) being female and 2,306 (29%) male.

Characteristics of Outpatients Who Care for Children Under the Age of 18

Bivariate chi square analyses of associations between the patient characteristics in **Table 1** show significant relationships between a higher prevalence of those caring for children under 18 and most of the variables under investigation: being female, age 30–49 years, higher education, income from paid work, having a spouse/partner, living with spouse/partner, owning a house/apartment, having good networks with family, and friends, fewer consultations since start of treatment, being a refugee, born outside Norway, not living in a large municipality, not having an individual care plan (required for anyone in need of long-term and coordinated services), not being on community treatment order, and having a diagnosis of moderate or less severe mental illness like anxiety disorders and moderate depression. Some of the same patterns are found in a more detailed analysis in **Table 2** with a focus especially on gender and main diagnosis.

In a logistic regression analysis of associations between the odds ratio for caring for children under 18 and the same patient characteristics (**Table 3**), most of the patient characteristics showed a significant association with the same direction as in the bivariate analyses, but with various odds ratios. The significant odds ratios were highest for age groups 30–49 and refugees; and lowest for being single, male, not having own house/apartment, and having a schizophrenia spectrum illness, or substance use disorder.

Prevalence of Outpatients With Minor Children Being Referred

The data on the number of children identified by clinicians to require referral to an external agency is shown in **Table 4**.

The clinicians answered the question on whether measures have been taken for referral for the children for 7,405 (92%) outpatients. Of these, 2,488 (31%) were reported to require referral to an agency. Of the 4,917 (61%) outpatients with children reported as not being referred, 247 (3%) were still reported to need a referral. This indicates that for 34% of the parents a referral of their children was identified as required, that some of these were not referred, and that children of 58% of the parents were reported to not require a referral. The need for referral is unknown for children of 630 (8%) of outpatients with minor children, including those who responded with "Do not know" and those with missing answers. Patients were involved in filling in the form in 61% of cases where the patient was a parent ($n = 8{,}035$). Sixty-three percent of female patients contributed to filling in the form compared to 59% of the male patients. If the parent was involved in filling in the form, 2% of the answers regarding measures taken for referrals were "Don't know" compared to 6% if the parent was not involved.

Information on the referral agencies is presented in **Table 5**. Almost half the outpatients with referred children (45%) had children who were referred to child protection agencies, closely followed by child and adolescent mental health services (39%) and educational-psychological services in the school system (35%). The most common combinations of services involved in follow up of the children are adult mental health outpatient clinics together with family counselling agencies ($n = 388$) or child protection agencies ($n = 304$).

Characteristics for Outpatients With Minor Children Being Referred

Bivariate chi square analyses of associations between the patient characteristics and whether measures were taken

TABLE 1 | Bivariate analyses of the association between patient characteristics and outpatients ($N = 22,847$) who care for children under 18.

	No children	Have children	Missing data	Total	% with children	Chi-test (x^2)
Gender						x^2 360.66 (1) $p < 0.001$
Female	8,425	5,729	262	14,416	40	
Male	5,938	2,306	187	8,431	28	
Age group						x^2 4087.50 (6) $p < 0.001$
18–23 years	3,258	249	69	3,576	7	
24–29 years	2,919	1,029	74	4,022	26	
30–39 years	2,603	2.965	81	5,649	53	
40–49 years	1,935	2,666	93	4,694	58	
50–59 years	1,805	765	52	2,622	30	
60–69 years	850	95	20	965	10	
70 years and above	672	59	31	762	8	
Education						x^2 506.11 (2) $p < 0.001$
High education	2,432	2,251	60	4,743	48	
Medium education	6,289	3,597	131	10,017	36	
Low education	5,772	2,267	325	8,364	28	
Main source of income						x^2 784.99 (2) $p < 0.001$
Income from labour	3,164	2,922	77	6,163	48	
Health related benefit	7,762	4,179	175	12,116	35	
Other economic support	3,567	1,014	264	4,845	22	
Marital status						x^2 4533.88 (2) $p < 0.001$
Married/cohabitant/partner	3,712	5,203	106	9,021	58	
Separated/divorced/widower/widow	1,624	1,355	64	3,043	45	
Single/unmarried	9,061	1,466	190	10,717	14	
Household						x^2 4258.02 (2) $p < 0.001$
Alone with or without children	7,033	2,704	172	9,909	28	
Spouse/cohabitant	3,596	5,090	98	8,784	59	
Other household	3,705	201	79	3,985	5	
Accommodation						x^2 2614.39 (3) $p < 0.001$
Own house	5,156	5,306	125	10,587	51	
Rented house/apartment, private market	4,298	1,868	95	6,261	30	
Rented house/apartment, local authorities	1,466	341	27	1,834	19	
Other	3,305	361	72	3,738	10	
Network family						x^2 79.72 (3) $p < 0.001$
Very good	4,169	2,642	113	6,924	39	
Good	6,325	3,561	124	10,010	36	
Poor	1,998	979	43	3,020	33	
Very poor	715	251	30	996	26	
Network friends						x^2 178.45 (3) $p < 0.001$
Very good	3,013	2,078	65	5,156	41	
Good	6,531	3,881	152	10,564	37	
Poor	2,180	996	58	3,234	31	
Very poor	825	238	22	1,085	22	
Number of consultations since start of treatment						x^2 145.81 (7) $p < 0.001$
<3	1,567	847	72	2,486	35	
3–5	1,551	1,009	37	2,597	39	
6–9	1,601	1,007	40	2,648	39	
10–19	2,345	1,381	70	3,796	37	
20–39	2,321	1,355	47	3,723	37	
40–99	2,155	1,204	43	3,402	36	
100–199	1,064	484	28	1,576	31	
200 or more	725	185	12	922	20	

(Continued)

TABLE 1 | Continued

	No children	Have children	Missing data	Total	% with children	Chi-test (x^2)
Asylum seeker						x^2 0.67 (1) $p = 0.4129$
No	14,426	8,071	513	23,010	36	
Yes	67	44	3	114	40	
Refugee						x^2 36.22 (1) $p < 0.001$
No	14,093	7,768	496	22,357	36	
Yes	400	347	20	767	46	
Born outside Norway						x^2 94.51 (1) $p < 0.001$
No	12,973	6,903	466	20,342	35	
Yes	1,520	1,212	50	2,782	44	
Having an individual care plan						x^2 411.4 (1) $p < 0.001$
No	12,329	7,608	471	20,408	38	
Yes	2,164	507	45	2,716	19	
Community treatment order						x^2 193.3 (1) $p < 0.001$
No	13,866	8,019	500	22,385	37	
Yes	627	96	16	739	13	
Living in a large municipality						x^2 244.16(1) $p < 0.001$
No	10,680	6,709	413	17,802	39	
Yes	3,813	1,406	103	5,322	27	
Diagnoses						x^2 1013.38(7) $p < 0.001$
Personality disorders	1,118	640	33	1,791	36	
Substance use disorders (as first or second diagnosis)	1,058	241	27	1,326	19	
Schizophrenia etc.	1,902	295	52	2,249	13	
Affective disorders	3,982	2,690	94	6,766	40	
Anxiety disorders	3,128	2,455	90	5,673	44	
Behavioural syndromes	552	221	14	787	29	
Behavioural and emotional disorders	557	418	16	991	43	
Other mental illness	1,320	619	27	1,966	32	

to refer children are shown in **Table 6**. There are highly significant associations for gender, age group, level of education, main source of income, marital status, type of household, accommodation, networks of family, network of friends, number of consultations since start of treatment, having an individual care plan, being under community treatment order, and main diagnosis. There are no significant associations for the size of the municipalities or for being part of a minority group (asylum seeker, refugee, born outside Norway). The pattern of significant differences shows that many indicators on lower socio-demographic status as well as severe mental illness and substance use disorder are associated with referrals of children.

Results of a logistic regression of the association between patient characteristics and odds ratios for referral of children is shown in **Table 7** for the 6,634 outpatients with non-missing data for the independent variables in the logistic regression and who had children who did or did not receive a referral. The significant odds ratios were highest for age groups 30–59, lower education, being single, not having income from paid work, not owning house/apartment, poor family network, having had many outpatient consultations, and having an individual care plan. The significant odds ratios were lowest for being male and for having moderate or less severe mental illness.

DISCUSSION

In the current study one third of adult outpatients from 107 Norwegian mental health outpatient clinics cared for children under 18 years of age. One third of those parents had children who required a referral, six out of 10 had children not requiring a referral, and for one in 10 parents, the needs of their children was unknown or not reported. Children of three of 10 outpatient parents were reported to have been referred to relevant services. Patient characteristics associated with referral actions for their children were low education, being single, not owning a house/apartment, having a poor family network, having an individual care plan, being female, and having moderate or a less severe mental illness.

Prevalence of Outpatients Who Care for Children Under 18

Thirty-six percent of the outpatients in this study cared for children under 18 years of age. This falls within the range of 36–38% found in four previous studies identified in a systematic review of adult mental health services (9). Importantly, this study is the first of its kind internationally to illustrate "whole of country" population data and represents a significant step forward in parent prevalence statistics. The results provide a

TABLE 2 | Gender, diagnosis and prevalence of outpatients ($N = 8,035$) with care for children under 18 among sociodemographic subgroups.

	Number of patients with children	Patients with children (%)	Type of household (%) with or without children			Poor family network (%)	Poor friend network (%)
			Alone	Spouse	Other		
Gender							
Female	5,729	40	41	43	15	17	16
Male	2,306	28	47	31	21	18	24
Diagnoses							
Personality disorders	635	37	49	38	13	30	29
Substance use disorders (as first or second diagnosis)	240	19	61	19	20	27	30
Schizophrenia etc.	293	13	67	14	18	19	29
Affective disorders	2,669	40	42	44	14	16	16
Anxiety disorders	2,429	44	36	49	15	15	15
Behavioural syndromes	219	29	33	37	30	12	13
Behavioural and emotional disorders	412	43	43	36	21	15	13
Other mental illness	613	32	36	37	27	15	18

strong rationale for intake systems to identify parenting status, incorporate parenting roles, and responsibilities in treatment plans, and assess and address the needs of patients' children. Overall, these data confirm that a significant minority of patients in adult mental health services are parents with children under the age of 18.

Characteristics of Outpatients With Care for Minor Children

The results also provide greater certainty about the sociodemographic characteristics of parents and children in mental health services. Sociodemographic patient characteristics associated with higher odds ratios for caring for children under 18 were being age groups 30–49 and refugee, and characteristics associated with lower odds ratios were being male, single, not having own house/apartment, poor network of friends and not living in a large municipality. Forty percent of females and 28% of males cared for minor children. In terms of gender, others have shown somewhat similar results, with between 34 and 59% of all female patients recorded as mothers and 25–39% of all males as fathers (10, 13). A 4-year census in one Australian adult mental health service found nearly half of parents (41.2–45.0%) were married, with around a third (30.8–36.2%) separated or divorced. Approximately half of parents (43.9–52.9%) reported a good level of social support. Having a good level of support and network of friends is important for those with a mental illness and especially for the well-being of parents and children (37). A Norwegian study (38) found that the prevalence of refugees was higher among the outpatients in a mental health clinic than in the population in the catchment area, but still concluded that the outpatient clinic was probably underused by refugees based on much higher self-reported mental health problems among refugees that others in an epidemiological study in the area. The higher odds ratio for refugees being parents in the current study may be due to a higher prevalence of refugees being parents to minors, as shown in **Table 1**.

Clinical patient characteristics associated with lower odds ratio for caring for children under 18 were longer outpatient care, having an individual care plan, and having a severe mental illness or a substance use disorder. These data would indicate that those with a severe illness by and large are not presenting to adult mental health services with children or are not disclosing that they have children. It might also be that those with severe mental illness are more often hospitalized and/or cared for by the community care teams than by mental health outpatient clinics, and that more parents with severe mental illness might have lost custody of their children and have little contact with the children. The fear of the involvement of child protection agency might dissuade patients who have a severe illness from disclosing their parenting status (39) and the data highlighting referral patterns in this study in some ways substantiates these fears.

Forty-four percent of parents were reported to have an anxiety disorder, 40% an affective disorder, 43% a behavioural and emotional disorder, and 37% a personality disorder. In Australia, Fernbacher et al. (23) found that 42% of parents in an outpatient service had schizophrenia, 23% depression, and 13% bipolar disorder. In the 4-year census in one Australian adult psychiatric service (14) the most prevalent diagnoses were mood (35–42%) and psychotic (22–35%) disorders, followed by anxiety (11–20%) disorders. Overall, it appears that many parents in adult mental health services have an affective disorder. This is important information that can be used to inform treatment as well as continued professional development for clinicians regarding family focused practice.

Prevalence of Outpatients With Minor Children Being Referred

For 92% of the outpatients with minor children the clinicians had identified if children required a referral to an external agency, and the need for referral was not ascertained for 8% of outpatients with minor children. Children of 61% of outpatient parents were not considered by clinicians to require a referral, even though

TABLE 3 | Logistic regression of the association between patient characteristics and whether the outpatients (*N* = 22,847) care for children under 18.

	Odds ratio	**Std. Err.**	**z**	$P > z$	**95% Confidence interval**	
Gender						
Male	0.676	0.028	−9.37	0.00	0.623	0.734
Age group						
18–23 years	1.000	(base)				
24–29 years	3.029	0.274	12.25	0.00	2.537	3.617
30–39 years	8.114	0.724	23.46	0.00	6.812	9.665
40–49 years	7.821	0.730	22.02	0.00	6.512	9.392
50–59 years	1.666	0.168	5.07	0.00	1.367	2.029
60–69 years	0.349	0.051	−7.21	0.00	0.262	0.464
70 years and above	0.213	0.037	−8.80	0.00	0.151	0.300
Education						
High education	1.000	(base)				
Medium education	1.033	0.050	0.67	0.51	0.939	1.136
Low education	1.167	0.065	2.75	0.01	1.046	1.303
Income						
Income from labour	1.000	(base)				
Health related benefits	0.857	0.039	−3.36	0.00	0.784	0.938
Other economic support	1.062	0.071	0.89	0.37	0.930	1.211
Marital status						
Married/cohabitant/partner	1.000	(base)				
Separated/divorced/widow/widower	0.842	0.117	−1.23	0.22	0.641	1.106
Single/unmarried	0.168	0.023	−12.99	0.00	0.128	0.220
Type of household						
Alone with or without children	1.000	(base)				
Spouse/cohabitant	0.832	0.113	−1.35	0.18	0.637	1.086
Other type of household	0.298	0.031	−11.67	0.00	0.243	0.365
Accommodation						
Own house	1.000	(base)				
Rented house/apartment, private marked	0.647	0.031	−9.10	0.00	0.589	0.710
Rented house/apartment, local authorities	0.564	0.049	−6.60	0.00	0.476	0.669
Other accommodation	0.638	0.058	−4.92	0.00	0.534	0.763
Poor network family	0.906	0.048	−1.88	0.06	0.817	1.004
Poor network friends	0.820	0.043	−3.79	0.00	0.740	0.909
Consultations 20 or more since start	0.870	0.035	−3.49	0.00	0.805	0.941
Refugee	1.399	0.163	2.87	0.00	1.112	1.759
Born outside Norway	1.156	0.074	2.27	0.02	1.020	1.311
Has and Individual care plan	0.731	0.051	−4.53	0.00	0.638	0.837
Community treatment order (CTO)	0.838	0.120	−1.24	0.22	0.633	1.109
Living in large municipality	0.681	0.032	−8.20	0.00	0.622	0.747
Main diagnoses						
Personality disorders	1.000	(base)				
Substance use disorders (as first or second diagnosis)	0.627	0.068	−4.31	0.00	0.507	0.775
Schizophrenia spectrum disorders	0.406	0.041	−8.90	0.00	0.333	0.495
Affective disorders	1.047	0.074	0.65	0.52	0.911	1.204
Anxiety disorders	1.038	0.075	0.52	0.60	0.901	1.197
Behavioural syndromes	0.718	0.088	−2.71	0.01	0.565	0.912
Behavioural and emotional disorders	1.528	0.164	3.95	0.00	1.238	1.886
Other mental illness	1.083	0.101	0.85	0.39	0.902	1.301
Constant	0.712	0.129	−1.88	0.06	0.500	1.015

TABLE 4 | Reported needs and whether measures have been taken to follow up of the children of outpatients ($N = 8{,}035$) with care for children under 18.

	Number of patients	% of patents
Yes child/ren have needs	2,488	31
No, but have needs	247	3
No, but does not have any needs	4,670	58
Do not know	257	3
No answer	373	5
Number of outpatients with responsibility for children under age 18	8,035	100

TABLE 5 | Prevalence of outpatients ($N = 2{,}488$) where children are reported to be followed up by various services or agencies for children*.

	Number of patients	% of patients
Child protection agency	1,128	45
Family counselling office	165	7
Educational-psychological service/ school	861	35
Child and adolescent mental health service	975	39
Adult mental health outpatient clinic	83	3
Other	544	22
No answer	46	2

More than one agency may have been marked for each outpatient.

3% of these were still provided with a referral to an external agency. In Norway health personnel are required to ascertain whether a patient has minor children, talk with the patient about their children's needs and offer to give information and guidance. But there are no standard procedures or rules for referral of the patients' children. There may be capacity problem in the child and welfare services, as well as variations in resources and availability of such services. However, as shown in **Table 6**, we did not find significant differences between large and small municipalities in the probability of measures taken to follow up children.

Clinicians' actions did not necessarily align with their stated beliefs, an incongruence that has also been highlighted elsewhere (27). Altogether 34% of outpatients with minor children had children who had been referred. For 58% of outpatients, their children were identified as not requiring any further referral or support. In one Australian audit around a third (28–39%) of all children had been identified as requiring the involvement of child protection services (14), slightly less than what was found in the current study (45%). Both figures however indicate sizable referrals to child protection agencies which highlights the need for collaborative service delivery models but also the important role that child protection plays in the lives of these families.

Given the range of risk and protective factors for these children, not all children whose parent has a mental illness will be adversely affected (40), nor will all children in the same family be affected in the same way. Nonetheless, given the high risks for children associated with parental mental illness it is incumbent on clinicians to provide appropriate assessment and monitor their needs over time. It is not clear from the present study whether clinicians undertake such an assessment or whether, as some studies have shown (27), they did not always acknowledge children's needs and accordingly did not follow through on appropriate referrals. Future open-ended survey responses and/or qualitative research may be needed to identify barriers and facilitators to these clinical decisions. In a Norwegian cross-sectional study including a questionnaire to both patients with children and to clinicians in mental health services, 95% of the clinicians answered that they had talked with the patient about the situation with their children, while 71% of patients said that they had such a conversation with a clinician (41).

Characteristics for Outpatients With Minor Children Being Referred

Sociodemographic patient characteristics associated with higher odds ratio for children being referred were being female, single, low education, not owning a house/apartment, not receiving income from paid work, and a poor family network. Maybery et al. (16) argue that risks for children escalates when the parent is single, without a support network, experiences housing insecurity and lives in poverty. So, it is perhaps not surprising that these children were more likely to be referred than other children. However, the specific reasons for why these children were referred still remains unclear.

Having an individual care plan was associated with higher odds ratio for children being referred, while moderate to less severe mental illness was associated with lower odds ratio for children being referred. Though the odds ratio of children being referred were parents having a moderate to less severe psychiatric illness, the established individual care plan suggests that these are families with multiple problems, with a need for coordinated support. The poorer family network might also be an important explanation for the higher need for other types of support.

For 52% of the patients with minor children, children had been referred to child protection agencies (45%) and family counselling offices (7%). This is not surprising as the sociodemographic characteristics of the patients with children being referred might indicate a need for economic and practical help and social support as well as help to cope with the parental illness. Many children (39%) had been referred to the child- and adolescent services, indicating that the children needed treatment for their own mental and/or behavioral problems.

For one third (35%) of the families, children had been referred to educational-psychological services/school, which confirms the need for support found in a Swedish study. Hjern et al. (5) argue that both parents' illness and substance abuse negatively impact school performance in their children, with social factors also having an important impact, and preventive general interventions in schools should be establish for such children. This would apply to children with parental illness, as well as children with separated/divorced parents or parents with low educational levels.

Some explanatory variables in the logistic regression in **Table 7** are correlated, but not to the extent that this is problematic. The highest correlation is between the variable

TABLE 6 | Bivariate analyses of characteristics of outpatients with children ($N = 8{,}035$)* and whether measures have been taken to follow up their children.

	No follow up	Follow up	Missing	Total	% Follow up	Chi-test (X^2)
Gender						x^2 127.13 (1) $p < 0.001$
Female	3,479	1,977	273	5,729	35	
Male	1,695	511	100	2,306	22	
Age group						x^2 59.88 (6) $p < 0.001$
18–23 years	169	67	11	247	27	
24–29 years	673	305	43	1,021	30	
30–39 years	1,839	959	135	2,933	33	
40–49 years	1,661	847	138	2,646	32	
50–59 years	530	213	21	764	28	
60–69 years	79	11	4	94	12	
70 years and above	55	3	1	59	5	
Education						x^2 148.97 (2) $p < 0.001$
High education	1,631	508	89	2,228	23	
Medium education	2,303	1,111	150	3,564	31	
Low education	1,240	869	134	2,243	39	
Main source of income						x^2 154.09 (2) $p < 0.001$
Income from labour	2,107	658	126	2,891	23	
Health related benefit	2,460	1,494	187	4,141	36	
Other economic support	607	336	60	1,003	33	
Marital status						x^2 197.22 (2) $p < 0.001$
Married/cohabitant/partners	3,587	1,319	250	5,156	26	
Separated/divorced/widower/widow	723	564	51	1,338	42	
Single/unmarried	807	580	66	1,453	40	
Household						x^2 180.19 (2) $p < 0.001$
Alone with or without children	1,472	1,094	110	2,676	41	
Spouse/cohabitant	3,498	1,304	243	5,045	26	
Other household	133	56	8	197	28	
Accommodation						x^2 186.72 (3) $p < 0.001$
Own house	3,621	1,386	247	5,254	26	
Rented house/apartment, private marked	1,044	732	73	1,849	40	
Rented house/apartment, local authorities	149	182	7	338	54	
Other	216	124	17	357	35	
Network family						x^2 100.54 (3) $p < 0.001$
Very good	1,817	685	115	2,617	26	
Good	2,266	1,097	164	3,527	31	
Poor	527	404	36	967	42	
Very poor	129	114	5	248	46	
Network friends						x^2 41.07 (3) $p < 0.001$
Very good	1,430	543	82	2,055	26	
Good	2,430	1,244	173	3,847	32	
Poor	581	359	40	9,80	37	
Very poor	144	83	10	237	35	
Number of consultations since start of treatment						x^2 109.68 (7) $p < 0.001$
Less than 3	578	215	48	841	26	
3–5	684	259	51	994	26	
6–9	670	274	56	1,000	27	
10–19	912	374	78	1,364	27	
20–39	856	435	50	1,341	32	
40–99	720	437	37	1,194	37	
100–199	271	199	11	481	41	
200 or more	80	100	4	184	54	

(Continued)

TABLE 6 | Continued

	No follow up	Follow up	Missing	Total	% Follow Up	Chi-test (X^2)
Asylum seeker						x^2 0.59 (1) $p = 0.4413$
No	5,148	2,472	372	7,992	31	
Yes	26	16	1	43	37	
Refugee						x^2 2.35 (1) $p = 0.1252$
No	4,942	2,395	357	7,694	31	
Yes	232	93	16	341	27	
Born outside Norway						x^2 272 (1) $p = 0.0993$
No	4,427	2,093	316	6,836	31	
Yes	747	395	57	1199	33	
Having an individual care plan						x^2 154.62 (1) $p < 0.001$
No	4,971	2,198	362	7,531	29	
Yes	203	290	11	504	58	
Community treatment order						x^2 13.11 (1) $p < 0.001$
No	5,128	2,441	370	7,939	31	
Yes	46	47	3	96	49	
Living in a large municipality						x^2 0.49(1) $p = 0.4856$
No	4,280	2,042	322	6,644	31	
Yes	894	446	51	1,391	32	
Diagnoses						x^2 107.14(7) $p < 0.001$
Personality disorders	353	261	21	635	41	
Substance use disorders (as first or second diagnosis)	110	119	11	240	50	
Schizophrenia etc.	166	115	12	293	39	
Affective disorders	1,769	785	115	2,669	29	
Anxiety disorders	1,648	661	120	2,429	27	
Behavioural syndromes	152	55	12	219	25	
Behavioural and emotional disorders	235	161	16	412	39	
Other mental illness	385	191	37	613	31	

**Those who receive follow up (n = 2,488) compared to those who do not (n = 5,174) or we do not know (n = 373).*

accommodation and marital status. If accommodation is removed from the regression, the size of the coefficient of marital status is the same as when accommodation is included.

In this study, if the patient did not participate in filling in the form, the question on whether children are referred was more often (6%) answered with unknown compared to if the patient participated in filling in the form (2%). This supports the importance of clinicians involving the patient in assessing the needs of their children.

Strengths and Limitations

A major strength of the study is the large census-level sample from almost all the psychiatric outpatient clinics in one country. As the data was registered on anonymous forms and did not require written consent from the patients, there is no obvious reason to expect that the material is skewed due to subgroups being less reluctant to participate. Some clinicians did however indicate that completing the form was time-consuming and that they did not have time to include all their patients. A limitation is that the clinicians might have only included the less complicated cases to save time. It is also possible that outpatients who missed their appointed consultations were less likely to be included. An important limitation of the study compared with epidemiological studies is that only those who receive treatment were included. The prevalence numbers presented are valid for outpatients in specialist mental health treatment and not for the population suffering from mental illness in general. Finally, the results may not be representative of patients in other types of mental health services and in other countries.

The amount of missing data is a limitation, as is the small amount of information collected regarding patients' children. Information was only collected on whether children were considered to have needs and whether these needs warranted a referral to external agencies. It was not possibility to examine the quality and thoroughness of these assessments. Information was not collected on what children's needs were, nor whether referral actions were appropriate, implemented and ultimately successful at addressing children's needs. Future research could explore these factors.

CONCLUSIONS AND IMPLICATIONS

Conclusions

The prevalence of outpatients in adult psychiatric clinics who care for children under the age of 18 years is 36% in this

TABLE 7 | Logit regression of the association between patient characteristics and whether minor children of outpatients ($N = 6634$)* are being referred.

	Odds ratio	Std. Err.	z	$P > z$	95% Confidence interval	
Gender (male)	0.466	0.032	−11.060	0.00	0.407	0.533
Age group						
18–23 years	1.000	(base)				
24–29 years	1.316	0.237	1.53	0.13	0.925	1.873
30–39 years	1.986	0.343	3.97	0.00	1.416	2.785
40–49 years	1.952	0.345	3.78	0.00	1.380	2.761
50–59 years	1.702	0.332	2.73	0.01	1.161	2.495
60–69 years	0.449	0.175	−2.05	0.04	0.209	0.966
70 years and above	0.123	0.092	−2.79	0.01	0.028	0.535
High education	1.000	(base)				
Medium education	1.419	0.102	4.87	0.00	1.233	1.634
Low education	1.896	0.156	7.77	0.00	1.613	2.228
Income from labour	1.000	(base)				
Health related benefits	1.376	0.091	4.83	0.00	1.209	1.566
Other economic support	1.361	0.136	3.07	0.00	1.118	1.657
Married/cohabitant/partner	1.000	(base)				
Separated/divorced/widow/widower	2.534	0.512	4.60	0.00	1.705	3.766
Single/unmarried	2.056	0.423	3.51	0.00	1.374	3.076
Alone with or without children	1.000	(base)				
Spouse/cohabitant	1.427	0.284	1.79	0.07	0.966	2.107
Other type of household	0.562	0.125	−2.58	0.01	0.362	0.870
Own house	1.000	(base)				
Rented house/apartment, private marked	1.337	0.097	4.02	0.00	1.161	1.541
Rented house/apartment, local authorities	1.701	0.236	3.83	0.00	1.296	2.232
Other	1.439	0.233	2.25	0.03	1.047	1.978
Poor network family	1.447	0.112	4.79	0.00	1.244	1.683
Poor network friends	1.032	0.083	0.40	0.69	0.882	1.209
Consultations from start: 20 or more	1.281	0.075	4.23	0.00	1.142	1.436
Having an individual care plan	2.232	0.247	7.27	0.00	1.797	2.771
Community treatment order	1.121	0.297	0.43	0.67	0.667	1.884
Main diagnosis						
Personality disorders	1.000	(base)				
Substance use disorders (as first or second diagnosis)	1.306	0.239	1.46	0.14	0.913	1.870
Schizophrenia spectrum disorder	0.857	0.147	−0.90	0.37	0.612	1.198
Affective disorders	0.774	0.080	−2.48	0.01	0.633	0.947
Anxiety disorders	0.684	0.072	−3.64	0.00	0.557	0.839
Behavioural syndromes	0.606	0.116	−2.61	0.01	0.416	0.883
Behavioural and emotional disorders	1.174	0.172	1.09	0.27	0.880	1.565
Other mental illness	1.124	0.155	0.85	0.40	0.858	1.472
Constant	0.099	0.028	−8.05	0.00	0.056	0.174

*One thousand twenty-eight patients had missing values for one or more of the variables, see **Table 6**.*

study of a sample from a national census in Norwegian adult mental health clinics. This provides considerable certainty regarding parent prevalence as the statistics emanate from a whole of country data set. The findings also add important information about the characteristics of higher risk families. Significant odds ratios among psychiatric outpatients being parents with care for minor children were highest for age groups 30–49 and refugees; and lowest for being single, male, not having own house/apartment, and having a schizophrenia spectrum disorder. Of the 92% of the patients with minor children where clinicians had answered whether measures have been taken for referral for the children, 31% were reported to have children having been referred, 58% did not have children requiring a referral, and 3% had children who had not been referred in spite of being identified as requiring a referral. The need for referral was unknown for children of 8% of the outpatients with minor children. The agencies most referred to were child protection agencies (45%), CAMHS

(39%) and educational-psychological services in the school system (35%). The significant odds ratios for having children who required referral to an agency were highest for parents aged 30–59 who were single with low education, not having income from paid work, not owning a house/apartment, poor family network, having had many outpatient consultations, and having an individual care plan. The significant odds ratios were lowest for males and for having moderate or less severe psychiatric illness.

Service and Practice Implications

There are several practice and systems implications for the present results. Given the sizable minority of patients who are parents, adult mental health services require appropriate infrastructure systems and procedures to identify parenting status of patients, including those who may be pregnant. Treatment plans should address needs of parents and their children and include referral of parents and their children to early intervention services as appropriate. Referrals to child protection agencies should be carefully considered, once accurate and sensitive assessments of the family's strengths as well as vulnerabilities are made, including the family's personal and professional networks. Appropriate training could be offered to clinicians in adult mental health outpatient services, with particular attention on how mental illness impacts parenting and the range of services that families may be referred to.

Implications for Research

Future studies need to investigate more broadly the family circumstances for children whose parents have mental illness, the specific actions of clinicians and the outcome of referrals for children. Why clinicians provide referrals for the children of some of their patients, but not others, could be further investigated. The pathways of care, as parents and children navigate various mental health and other systems (including but not limited to adult mental health, child protection services, schools), could be documented to determine what happens for families, areas of duplication/gaps, and outcomes for different family members, over time.

AUTHOR CONTRIBUTIONS

TR, DM, and SO contributed conception and design of the study with input from AR, KF, BW, BS, and AG. SO collected the data, organized the database and performed the statistical analyses. DM and TR wrote the first draft of the manuscript. AR, KF, BW, BS, AG, and SO wrote sections of the manuscript. All authors contributed to manuscript revision, read and approved the submitted version.

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Identification of Children of Parents With Mental Illness: A Necessity to Provide Relevant Support

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Background: The main objective of this study was to identify and describe core life circumstances of children with mentally ill parents (COPMI) and their parents. Knowledge about COPMI aged 0–17 years is necessary, as assessment of the risk and protective factors in their lives provide solid background for preventive interventions.

Methods: Participants ($N = 422$) were parents of minor children ($N = 589$) receiving treatment in the clinic for psychiatric illness and substance abuse at the University Hospital of Northern Norway. Data was drawn from electronic patient journals.

Results: A total of 286 mothers and 136 fathers participated in the study, and 46.3% were single parents. Parents had 1–7 children ($M = 2.24$; $SD = 1.02$). Most parents had one diagnosis ($n = 311$, 73.7%), and mood disorders was the most frequent type of diagnosis. The largest proportion of parents had serious mental disorders ($n = 185$; 46.0%), and a large proportion of the sample was affected by disorders of moderate severity ($n = 156$; 38.8%). The mean age of the children was 8.6 years (SD = 4.97), and 432 children (74.6%) had one or more siblings. The large majority of children had access to adult resource persons other than the mentally ill parent ($n = 424$; 94%), but 6% of the children ($n = 27$) did not. About three quarters of the children (76.2%, $n = 526$) were living with the mentally ill parent ($n = 401$), and 170 children (32.5%) lived with a single parent with a mental health disorder and siblings, full time or part of the time. The odds that parents had informed their children about the treatment/hospitalization and condition was higher the older the child was ($p < 0.001$), and the youngest children rarely got necessary information about this.

Discussion: Risk and protective factors associated with the children's ages, access to resource persons, information about the parent's health problems and treatment are discussed in relation to different preventive steps for COPMI.

Keywords: parents, mental illness, mental health care services for adults, risk factors, children

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INTRODUCTION

Worldwide around onev in five minor children has a parent with a mental illness (1). In Norway it is estimated that 450,000 children have parents with a mental illness or substance use disorders (2). These children are at high risk of developing a mental illness themselves (3).

In a meta-analysis, (4) found that children of parents with a severe mental illness had a 50% chance of developing any mental illness, and 32% chance of developing a severe mental illness.

In Norway, it has been estimated that children of parents with a mental illness (COPMI) have double the risk of both short-term and long-term negative consequences compared to children of parents without mental illnesses (2). Elevated risk has been documented for COPMI across the diagnostic spectrum of mental disorders in parents, including schizophrenia (5) obsessive-compulsive disorder (6), depression (7, 8), substance abuse disorders (9), anxiety disorders (10), bipolar disorder (11), eating disorders (12), personality disorders (13) and suicide (14). The transmission of risk for psychopathology from parents to children is both diagnosis-specific such that children may develop the same mental illness as their parents, and general, such that children are at risk of developing a wide range of disorders (10).

In addition to hereditary components of mental illness enhancing the risk of mental illness among the offspring, parents' symptomatology may also has a social impact and the ways parents interact with their children is therefore highly significant (8). Psychopathology in a parent often impairs parenting skills, the quality of care they provide and the parent-child interaction (15, 16). Such impairments may in turn lead to reduced involvement with the child, as well as insensitivity, hostility, rejection, neglect and potential abuse (17). The failure in one or more aspects of parenting can lead to insecure attachment (18, 19), emotional dysregulation, negative emotionality and pathological coping strategies (17), as well as psychopathology in childhood, adolescence and adulthood (20).

Different characteristics of the parent's psychopathology predict increased risk for the COPMI, including the symptom burden, comorbidity, and the severity and duration of illness. Empirical studies have repeatedly found that there is a greater negative impact on children whose parents have co-morbid disorders and personality disorders, compared to children whose parents have single disorders (21). Comorbid mental disorders in a parent may lead to larger and more long-lasting functional impairments, poorer prognosis and treatment complications (22, 23). Brennan et al. (24) found that children of depressed mothers were at greater risk of developing behavioral problems if the depression was severe, and that severity had a significant relationship to aspects of the children's language development. Although there are several risk factors related to parent's psychopathology, some COPMI are very resilient and are not impacted adversely. To our knowledge, no systematic reviews or meta-analyses have been conducted to quantify resiliency factors, but qualities of child personality and temperament, quality of attachment between the child and primary caregivers, as well as social support in the family and social network, is believed to buffer against adverse outcomes for COPMI.

It seems evident that the child's age is an important factor contributing to the outcomes for children at risk, and that younger children are at higher risk. For 1- and 2-years old children, depression in parents has been associated with impaired cognitive development, more behavioral problems, lower IQ scores in late childhood, as well as elevated rates of affective disorders in adolescence (8, 25).

The family is the core arena of development for children. However, the family situation for COPMI may be characterized by family conflicts, violence and negative life events (17, 26). Research findings have shown that children from conflict-ridden families were viewed less favorably by their peers and had fewer friends (26). In addition, COPMI often have care responsibilities in the household that exceed their emotional and cognitive maturity (27). Therefore, the risk for negative outcomes may be higher for children who live alone with one parent with mental illnesses, compared with those who live with both one parent with a mental illness and one healthy parent. Studies have found that single parents report having more mental health problems and behavioral problems compared with married parents (28). In contrast, the presence of a supportive and caring parent who understands the suffering of the ill parent can act as a buffer against negative child developmental outcomes related to depression in the mother (29).

Social and emotional ties to people outside the family can also moderate the effect of mental illness in parents (30), and the children's social network therefore plays a major protective role. Children benefit from having access to stable, non-familial trusted adults, such as teachers and other educational staff, as well as other adults in the child or parents' social support network and friends (31, 32).

Though impaired parenting as a result of parental psychopathology is a very potent risk factor for the development of emotional and behavioral problems in children, especially in early childhood, key parental functions are also modifiable (15, 16). Siegenthaler et al. (4) reported that family-focused interventions reduced children's risk of acquiring their own mental health problems by 40%. Another recent systematic review and meta-analysis reported small, but significant and lasting effects related to interventions for mothers and infants, as well as for children and adolescents themselves (20). Psycho-education is a common component across programs for COPMI and their families (33). The aim of such efforts is often to strengthen children's knowledge of the parent's psychopathology, as well as to reduce feelings of guilt and shame related to parental psychopathology. In general, research gives some indications that most parents do not speak with their children about mental illness. For example, a British survey showed that 55% of parents without mental illnesses and with children aged 6 to 18 did not talk with their children about mental illnesses.[1] For COPMI, who are exposed to parental mental health symptoms on a daily basis, mental health literacy tend to be low (34), and they do not have access to accurate, non-stigmatized information about mental health disorders and treatments (35). Many COPMI do not seek help, neither from health care providers nor in their own network (36, 37). However, interviews of COPMI indicate that they want to be recognized as full-fledged members of the family by their parent's treatment providers, and to participate in the parent's therapeutic process to gain knowledge about the parent's illness and how to deal with it (36, 38). Many parents

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[1] Warren E. New Survey Shows That Mental Health is a "Nonversation" for Parents (2015). Available online at: http://www.time-to-change.org.uk/news/new-survey-shows-mental-health-%E2%80%98nonversation%E2%80%99-parents-0

with mental illness are also concerned about the effects their mental illness may have on their children. They often want help, advice and guidance about how to talk to their children about their troubles.

In spite of this, COPMI receive little attention within mainstream mental health services (1, 34). In Norway, systematic routines for identifying these children did not exist until a few years ago (39). Based on knowledge about the transmission of mental disorders across generations, Norwegian authorities adopted amendments to the Health Personnel Act and Specialized Health Services Act in 2010. These provisions require health personnel in adult mental health services to identify and fulfill the needs of COPMI. It has taken time to implement these legal amendments, and they have thus far not led to satisfactory changes in clinical practice (40, 41). One study showed that 56% of health personnel at a large Norwegian university hospital did not identify patients' children (39), and a 5-years follow-up study showed that 28% of the health personnel in the same clinic still did not identify patients' minor children (42). Another recent Norwegian study showed that only 17% of patients in two psychiatric hospitals were assessed completely with family assessment forms (43). These results indicate that although the law requires identification of COPMI and provision of support in Norway, there has not been sufficient systematic work around implementing new clinical practice related to this issue in adult mental health care. Similar findings were reported in a study from adult mental health services in New Zealand (44).

The lack of identification of COPMI has large implications for public health as it is a core prerequisite to intervene in the high risk group of COPMI. Routine identification of COPMI in adult mental health services will provide necessary information about the children, their family situation and needs, and hence form the basis for provision of necessary family support and necessary professional collaboration across services and service levels in the municipalities where the family lives.

THE NORWEGIAN COPMI PROJECT—THE PRESENT STUDY

The COPMI project is a longitudinal research project 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 (45). The project started in 2010, and involves a long-term strategy for changing clinical practice. The clinic initiated new procedures to identify COPMI by the use of *Family Assessment.* The *Family Assessment* form is an intervention for treatment providers to increase the identification of patients' minor children. The form consists of questions that the health personnel were required to collect as a result of legislative changes.

The main objective of the present study was to identify the children and basic life circumstances related to child development in families where the parent has a mental disorder, as well as to discuss how this information may be utilized to plan and strengthen professional collaboration in the provision of relevant support for COPMI.

These research questions were investigated:

(a) What are the demographic (gender, marital status, total number of children), and illness characteristics of the patients?
(b) What characterizes the children's gender, age, number of siblings, day care, and living arrangements, as well as the knowledge provided to them about parental mental illness and access to social supports?
(c) What is the relation between parent's diagnosis and disorder severity, and where the children live?
(d) Which factors influence whether or not the children get information about their parent's psychopathology?

METHODS

Participants

The participants were 422 parents with mental disorders who received treatment at a clinic for psychiatric disorders and substance abuse at the University Hospital of North Norway (UNN). If the parent had more than one child, one assessment form per child was filled out. A total of 581 minor children were assessed.

Data Material

The data consisted of information gathered by health personnel by using an adapted version of the information form "Family Assessment" (45). This is a standardized information form, designed to gather information about the child's gender, age, siblings, parental access, residence, and other caregivers who cared for the child during the parent's illness, as well as where the child was during the day and whether the child had received information about the parent's psychopathology. The form contained two questions about whether or not the children had been given information about the parent's treatment/hospitalization and the parent's condition. The response options for these questions were "yes," "partially," and "no." In addition, ordinary electronic patient journals were assessed to gather information on the patient's gender, marital status, the total number of children, and diagnoses.

Procedure

The information form was implemented as a compulsory routine for all staff in the participating departments and it was integrated in the electronic patient record. Under Norwegian law, collecting this information does not require consent.

The treatment provider filled in the information form in the electronic patient record during the conversation with the parent about the children. The treatment providers were psychologists, specialist psychologists, clinical social workers, clinical social educators, nurses, psychiatric nurses, activity therapists, doctors, psychiatrists, and social workers.

Analysis

All analyses were performed using IBM SPSS Statistics 24. Descriptive frequency analyses were used to describe the sample. A chi-squared test was used to evaluate the relation between the parent's gender, whether the child lived alone with a parent with a mental illness or addiction, who cared for the child during any hospitalization, and whether the child had received information about the parent's illness. A chi-squared test was also used to look at the relationship between family composition and whether the child had other adult resource persons (i.e., the mentally healthy parent, a step parent, teachers, relatives, family friends, and/or neighbors).

Multivariate hierarchical logistic regression analyses were conducted to investigate the odds of having received information about the mental condition of the parent or about the parent being in treatment for children who lived with a single parent with a mental illness compared to other children. Multivariate hierarchical logistic regression analyses were also conducted to examine the relationships between information provided (about the mental condition and treatment), and the children's age, gender, and parental diagnosis. Interaction effects between the parental diagnosis and child age and gender were tested.

In order to compare whether the parent's diagnoses were connected to the various dependent variables (information about the disorder, receiving health care, living arrangements) in logistic regression analyses, the diagnosis variable was re-coded into dummy variables. Diagnoses that belonged to the same chapter in ICD-10 (46) were coded into the same dummy variables. Diagnostic categories with $n < 15$ were excluded in these analyses. This was true for F00–F09 Organic, including symptomatic, mental disorders ($n = 5$), F50–F59 Behavioral syndromes associated with physiological disturbances and physical factors ($n = 5$), F70–F79 Mental retardation ($n = 1$), F80–F89 Disorders of psychological development ($n = 0$), F99 Unspecified mental disorder ($n = 0$), F90–F98 Behavioral and emotional disorders with onset usually occurring in childhood and adolescence ($n = 14$), and R-/Z diagnoses ($n = 11$). In a regression analysis where the children's age was one of the variables, age was categorized in groups. In order to use the variables "has your child received information about your condition?" and "has your child received information about your health care/hospitalization?" as independent variables in a binary logistic regression, these were coded as dichotomous variables, where the response option "partially" was coded as "yes."

The parents' primary diagnoses were categorized by their presumed severity. However, the distinction between serious and milder mental disorders is not firmly established in the discipline. The data set also did not contain information about daily functioning. The divisions used by Kessler et al. (22) was therefore adopted for this purpose, but without the functional goals. The diagnosis was coded as serious if it fulfilled one of the following criteria; assumed disability or significant limitations as a result of illness, drug addiction or if the disorder typically leads to 30 or more days per year in which the person is not able to maintain their roles socially, in the household, as an employee or as a partner. The diagnosis was coded as moderate if it did not meet any of the above criteria, but was considered to have a moderate impact on the aforementioned roles. All other disorders were classified as mild.

Logistic regression was used to examine the relationship between diagnoses and whether or not the child had received information about the parent's treatment and condition. To be able to compare the odds that the child had received information depending on the type of diagnosis, the diagnosis variable was recoded into dummy variables. Diagnoses that belonged to the same chapter in ICD-10 were coded in the same dummy variable, and diagnostic categories with $n < 15$ were excluded in these analyses.

RESULTS

Characteristics of the Parents/Patients

Gender

The sample ($N = 422$) consisted of 286 mothers (67.7%) and 136 fathers (32.3%). The proportion of mothers was significantly larger than the proportion of fathers ($p < 0.001$).

Marital Status

The Family Assessment Form did not distinguish between being married or cohabiting. A total of 105 parents (43.4%,) reported to be married or living together with the other parent, 25 parents (10.3%) were married or living together with the child's step-parent, and 112 parents (46.3%) were single. Information about marital status was provided for 242 parents (57.3%), while for 180 parents (42.7%) information was missing.

Number of Children

Parents ($N = 402$) had between one and seven children ($M = 2.24$; $SD = 1.02$), whereas eight (1.9%) replied that they were expecting children.

The Parents' Diagnoses

The majority of parents had one diagnosis ($n = 311$, 73.7%), and some had two diagnoses ($n = 70$, 16.6%). A small proportion of parents had three diagnoses ($n = 21$; 5.0%), while few had no diagnosis ($n = 20$, 4.7%). The most commonly occurring diagnosis type were mood disorders ($n = 190$, 45%). Individually, the three most common diagnoses were F32.1 Moderate depressive episode ($n = 60$, 10.3%), F33.1 Recurrent depressive disorder, current episode moderate ($n = 53$, 9.1%) and F43.1 Post-traumatic stress disorder ($n = 31$, 5.3%).

Severity of Mental Illness

The largest proportion of parents in the sample had serious mental disorders ($n = 185$; 46.0%), followed by parents with disorders of moderate severity ($n = 156$; 38.8%). Mild mental disorders were the least common ($n = 61$; 15.2%).

Characteristics of COPMI

Gender

The sample consisted of 290 (52.4%) boys and 263 (47.6%) girls. Information on gender was missing for 28 of the children (4.8%).

Age

The children's ages ($N = 543$) were evenly distributed from 0 to 17 years (**Table 1**). The average age was 8.6 years (SD = 4.97). A total of 76 children (14%) were below the age of 2 years old.

TABLE 1 | Child age.

Child age	N	%
1–12 months	24	4.4
1 year	30	5.5
2 years	22	4.1
3 years	30	4.4
4 years	33	6.1
5 years	32	5.9
6 years	22	4.1
7 years	43	7.9
8 years	34	6.3
9 years	20	3.7
10 years	33	6.1
11 years	37	6.8
12 years	39	7.2
13 years	34	6.3
14 years	32	5.9
15 years	25	4.6
16 years	31	5.7
17 years	22	4.1
Total	543	100
Age missing	39	6.7

Siblings

A total of 432 children (74.6%) had one or more siblings ($M = 1.25$; $SD = 1.04$), and 382 children (66.0%) had one or more siblings under the age of 18, whereas 93 children (16.1%) had one or more siblings above the age of 18. A total of 147 children (25.4%) had no siblings.

Custody For the Child

A total of 186 children (38.9%) lived with both parents, whereas 83 children lived with each parent in a 50–50% manner. A total of 146 children (30.5%) lived with their mother, and 29 (6.1%) with their father.

Access to Adult Resource Persons

Most parents stated that their children had access to adult resource persons other than the mentally ill parent ($n = 424$, 94%). A total of 6% of the children ($n = 27$) did not have this. A chi-squared test was used to see if there was a relationship between being a single parent with a mental illness and whether or not the children had other adult resource persons. The results showed that there was no significant difference; children who lived with a single parent with a mental illness were just as likely to have other close adults as children in homes with two parents, step-parents or other adults.

Care For the Child When the Mother/Father Is in Treatment/Hospitalized

In sum, one or both the parents cared for the child ($n = 308$; 71.7%) when one of them received treatment/was hospitalized (**Table 2**). The majority of the remaining children were taken care of by other family members. If the father was in treatment/hospitalized, the child's mother cared for the child in 88.8% of the cases, whereas if the mother was hospitalized, the child's father cared for the child in 62.7% of the cases, which was a significant difference ($\chi^2(2, N = 430) = 31.348, p < 0.01$). Children with mentally ill mothers were more likely to be cared for by grandparents, stepparents and other family members (17.1%) than children with mentally ill fathers were (4.2%).

TABLE 2 | Caregivers when mother/father is receiving treatment/hospitalized.

Caregiver	N	%
Mother	128	29.8
Father	180	41.9
Grandparent/s	24	5.6
Other family member/s	22	5.1
Fosterparent/s	20	4.7
Emergency placement	9	2.1
Stepparent	9	2.1
Friends/others	4	0.9
Child lives alone	3	0.7
Not applicable	31	7.2
Missing	149	25.6

Children Living With Mentally Ill Parent

About three quarters of the children in the sample (76.2%, $n = 526$) were living with the mentally ill parent. If the patient was the father, a bigger proportion of the children did not live with him (47.3%) than if the patient was the mother (12.6%).

Children Living With a Single Parent With a Mental Illness

A total of 170 of the children (32.5%) lived with a single parent with a mental illness and any siblings, either all the time ($n = 91$, 17.3%) or part of the time ($n = 80$, 15.2%). A greater percentage of the children with a mentally ill mother lived alone with her and any siblings, as compared to the children with a mentally ill father, and this difference was significant ($\chi^2(2, N = 526) = 44.547, p < 0.001$).

Children Living Outside the Home

A total of 45 children (7.8%) lived with neither their mother nor their father. The majority of these children either lived in a foster home ($n = 21$; 46.7%) or in an emergency placement home ($n = 9$; 20%). The other children ($n = 15$; 33.3%) lived with grandparents or siblings of the parents.

School and Day Care

Most of the children were at school ($n = 304$, 67.6%) or in kindergarten ($n = 119$, 26.4%) during the day. The others were at home ($n = 23$, 5.1%), in an emergency placement home ($n = 3$, 0.5%) or in a youth home ($n = 1$, 0.2%). Among the children who were at home during the day, 14 were under the age of 2 years.

Parent's Diagnosis and Disorder Severity, and Its Relation to Where the Child Lives

Parent's Diagnosis

Logistic regression showed that F10–F19 *Mental and behavioral disorders due to psychoactive substance use* was the only significant diagnostic category that could predict children's living conditions, and the odds that the child *did not* live with the parent was higher for parents with diagnoses in this category ($n = 44$; $OR = 8.7$; $p < 0.001$).

Diagnosis Severity

Logistic regression also showed that the odds that the child did *not* live with the parent was 3.3 times higher for the most severe mental disorders ($p < 0.001$) compared to mild disorders.

Factors That Affect Whether Children or Not Children Are Informed About the Parent's Treatment/Hospitalization and Condition

The parents of a total of 67.8% of children ($N = 395$) were asked "*Does your child know that you receive treatment/does your child know that you are hospitalized*?" Of these, a total of 54.9% ($n = 217$) responded "yes," 6.8% ($n = 27$) answered "partially," and 38.2% ($n = 151$) replied "no."

The question "*Has your child received information about your condition?*" was answered by the parents of a total of 61.6% of the children ($N = 359$). Of these, a total of 44% ($n = 158$) responded "yes," 14.5% ($n = 52$) answered "partially," and 41.5% ($n = 149$) replied "no."

Family Composition

Children who lived with a single parent with a mental illness were more likely to receive information than the children living with both parents. The results from a hierarchical logistic regression showed that if the child lived with a single parent with a mental illness, the odds that the child had been given information about both treatment/hospitalization (**Table 3**) and the condition (**Table 4**) were higher. An analysis of whether there were interaction effects between the parent's gender and marital status in how much information the child received, showed no such interaction.

Logistic regression was used to examine the relationship between the information provided and the children's age. The odds that parents had informed their children about the treatment/hospitalization (**Table 5**) and condition (**Table 6**) was higher for each age group ($p < 0.001$); The odds of 15–17-years-olds having received information about the treatment/hospitalization was 43.77 times higher than for 0–2-years-olds, and the odds that they had received information about the condition was 47.78 times higher.

TABLE 3 | Relation between information provided for the child about parent's treatment/hospitalization and whether the child lives with a single mentally ill parent or not.

	$X^2(2) = 12{,}225$, $p = < 0.002$		
	OR[a] [95% CI[b]]	Wald	*p*
Part time	1.93 [1.05, 3.56]	4.43	0.035**
Yes	2.55 [1.36, 4.78]	8.60	0.003*

[a] Odds ratio.
[b] Confidence interval.
*Significant at 0.01 level.
**Significant at 0.05 level.

TABLE 4 | Relation between information provided for the child about parent's condition and whether the child lives with a single mentally ill parent or not.

	$X^2(2) = 12{,}663$, $p = < 0.002$		
	OR[a] [95% CI[b]]	Wald	*p*
Part time	1.73 [0.92, 3.29]	2.88	0.090
Yes	2.87 [1.50, 5.50]	10.13	0.001*

[a] Odds ratio.
[b] Confidence interval.
*Significant at 0.01 level.

TABLE 5 | Relation between information provided for the child about parent's treatment/hospitalization and child age.

	$X^2(5) = 96.128$, $p = < 0.001$		
Child age	OR[a] [95% CI[b]]	Wald	*p*
3–5	5.06 [1.97, 12.98]	11.37	0.001*
6–8	7.93 [3.14, 20.02]	19.19	0.000*
9–11	13.51 [5.19, 35.22]	28.39	0.000*
12–14	29.77 [10.98, 80.70]	44.48	0.000*
15–17	43.77 [14.73, 130.03]	46.27	0.000*

[a] Odds ratio.
[b] Confidence interval.
*Significant at 0.01 level.

TABLE 6 | Relation between information provided for the child about parent's condition and child age.

	$X^2(5) = 95.446$, $p = < 0.001$		
Child age	OR[a] [95% CI[b]]	Wald	*p*
3–5	4.46 [1.51, 13.14]	7.34	0.007**
6–8	9.62 [3.37, 27.44]	17.92	0.000*
9–11	13.37 [4.56, 39.19]	22.34	0.000*
12–14	39.00 [12.75, 119.35]	41.22	0.000*
15–17	47.78 [14.48, 157.65]	40.29	0.000*

[a] Odds ratio.
[b] Confidence interval.
*Significant at 0.01 level.
**Significant at 0.05 level.

TABLE 7 | Relation between information provided for the child about parent's health condition and parent's diagnosis.

		$X^2(5) = 18{,}665, p = 0.002$*		
	N	**OR[a] [95% CI[b]]**	**Wald**	**P**
F10–F19 Mental and behavioral disorders due to psychoactive substance use	44	2.56 [0.82, 7.97]	2.62	0.106
F20–F29 Schizophrenia, schizotypal and delusional disorders	59	8.31 [2.75, 25.09]	14.09	0.000
F30–F39 Mood [affective] disorders	268	3.70 [1.62, 8.44]	9.63	0.002
F40–F48 Neurotic, stress-related and somatoform disorders	121	4.62 [1.86, 11.45]	10.93	0.001
F60–F69 Disorders of adult personality and behavior	25	3.65 [1.06, 12.56]	4.22	0.040

[a] *Odds ratio.*
[b] *Confidence interval.*
**Significant at 0.005 level.*

Children's Gender

There were no significant differences between boys and girls in how much information they had received about the parent's treatment/hospitalization and condition.

Parent's Gender

Results from a chi-squared test showed differences between children with mentally ill mothers and children with mentally ill fathers in terms of how much information they had received about the parental illness. There were significant differences based on the parent's gender in how much information the child had received about the treatment/hospitalization ($\chi^2(2, N = 316) = 8.606, p < 0.05$). There were also significant differences in whether or not they had received information about the parent's condition ($\chi^2(2, N = 349) = 10.015, p < 0.01$). For both variables, the children of mentally ill mothers were more likely to have received information compared to children of mentally ill fathers.

Parent's Diagnosis

The odds ratio that the child received information about the parent *treatment/hospitalization* were highest if the parents had diagnoses from the categories F20–F29 Schizophrenia, schizotypal and delusional disorders (OR = 2.8; $p > 0.05$) (**Table 7**), and F60–F69 Disorders of adult personality and behavior (OR = 3.2; $p = 0.08$). For the diagnostic category F10–F19 Mental and behavioral disorders due to psychoactive substance use, the odds of information being given were nearly halved (OR = 0.5; $p = 0.22$).

The odds ratio that the child received information about the parent's *condition* were highest when the diagnosis fell within F20–F29 Schizophrenia, schizotypal and delusional disorders (OR = 8.3; $p = < 0.0005$) (**Table 8**).

Diagnosis Severity

A multivariate hierarchical logistic regression was used to examine the relationship between the severity of the parent's

TABLE 8 | Relation between information provided for the child about severity of parent's diagnose, child age, child gender and parent's treatment/hospitalization.

	Model 1			Model 2			Model 3			Model 4		
	$X^2(2) = 3.510, p = 0.173$			$X^2(4) = 83.096, p < 0.001$			$X^2(6) = 85.323, p < 0.001$			$X^2(8) = 87.086, p < 0.001$		
	OR[a] [95% CI[b]]	**Wald**	**p**	**OR [95% CI]**	**Wald**	**p**	**OR [95% CI]**	**Wald**	**p**	**OR [95% CI]**	**Wald**	**p**
Severity: Mild	Reference group											
Severity: Moderate	1.40 [0.74, 2.65]	1.06	0.303	1.51 [0.740, 3.10]	1.28	0.257	0.48 [0.08, 2.72]	0.70	0.404	0.75 [0.10, 5.65]	0.08	0.781
Severity: Severe	1.81 [0.95, 3.45]	3.27	0.071	1.56 [0.85, 3.59]	2.29	0.130	0.99 [0.1, 5.50]	0.00	0.992	1.83 [0.25, 13.51]	0.35	0.553
Child age				1.92 [1.64, 2.25]	63.75	0.000	1.56 [1.05, 2.30]	4.94	0.026	1.60 [1.07, 2.39]	5.29	0.022
Child gender (1 = boy)				1.00 [0.62, 1.61]	0.00	0.998		0.00	0.967	1.93 [0.57, 6.52]	1.12	0.290
Mild*age								2.23	0.327		1.92	0.384
Moderate*age							1.41 [0.88, 2.26]	2.04	0.153	1.37 [0.85, 2.22]	1.66	0.198
Severe*age							1.18 [0.75, 1.85]	0.49	0.486	1.15 [0.72, 1.82]	0.33	0.567
Mild*gender											1.73	0.420
Moderate*gender										0.53 [0.13, 2.21]	0.76	0.385
Severe*gender										0.39 [0.09, 1.60]	1.72	0.190

[a] *Odds ratio.*
[b] *Confidence interval.*

diagnosis (mild, moderate, severe), the child's age and gender, and whether the child had been given information about the treatment/hospitalization (**Table 9**) and the parent's condition (**Table 10**). In the first model, the severity of the diagnosis was added alone, while in model 2 the child's gender and age were added. In the last model, the interaction effects were also examined. The interaction effects were examined in individual analyses. The odds that someone had informed their children about their treatment/hospitalization was greater the more serious the diagnosis was, but this difference was not significant. The odds that someone had informed their children about the parent's condition were also greater the more serious the diagnosis was, but this difference was not significant.

Model 2 shows that the child's age could predict whether the child received information or not, which was also shown in the results of the *t*-test, but no interaction effect was found between diagnosis severity and age of the child (Model 3). The child's gender did not affect the information the child received, and there was no interaction effect between diagnosis severity and the child's gender when it came to whether or not the child had been given information about the parent's treatment/hospitalization or condition.

DISCUSSION

Characteristics of the Patient/Parents

Parenting is challenging in itself, and a large number of studies indicate that parents with mental illnesses struggle more with parenting than parents who does not experience mental illness (16, 47). Nearly half of the patients in this study were single, and this may result in both a greater scope of problems in the parent (28), lower SES (48), and possibly an increased risk of the children developing mental disorders (49). In addition to their parent's mental illness, children who also have been through a divorce between their parents may have experienced a stressful and risk-enhancing transition in their family life, because of conflicts, moving and breaks in family relationships (50).

The largest proportion of the parents with mental illnesses were mothers, which is consistent with findings showing that more women than men seek help for mental health problems (51). Mood disorders, such as different forms of depression were most common in the sample, and the diagnosis F33 Major depressive disorder (recurrent), recurrent, was listed as the second most common illness. Recurrent and chronic disorders have been shown to lead to higher risks for COPMI (52, 53).

Most parents in our study had serious psychiatric disorders, which add to the high risk in these patients' children, as several studies have found a significant relationship between severity and chronicity of the parent's mental illness and the risk of negative outcomes in their children (3). In addition, mental disorders of moderate severity may lead to significant functional decline (2) and develop into more serious disorders over time.

The implications of the findings regarding the patients in this study is that the majority of patients are characterized by factors that predict high risk for their offspring. Therefore, health professionals in the clinic need to implement family support to reduce these risks and prevent developmental problems in

TABLE 9 | Relation between information provided for the child about severity of parent's diagnose, child age, child gender and parent's health condition.

	Model 1			Model 2			Model 3			Model 4		
	$X^2(2) = 5.246, p = 0.073$			$X^2(4) = 88.778, p < 0.001$			$X^2(6) = 92.552, p < 0.001$			$X^2(8) = 95.957, p < 0.001$		
	OR[a] [95% CI[b]]	Wald	*p*	OR [95% CI]	Wald	*p*	OR [95% CI]	Wald	*p*	OR [95% CI]	Wald	*p*
Severity: Mild	Reference group											
Severity: Moderate	1.50 [0.77, 2.91]	1.41	0.235	1.50 [0.70, 3.19]	1.08	0.300	0.29 [0.05, 1.75]	1.83	0.176	0.27 [0.04, 2.01]	1.63	0.202
Severity: Severe	2.12 [1.08, 4.15]	4.75	0.029	1.94 [0.90, 4.19]	2.88	0.090	0.47 [0.08, 2.83]	0.68	0.410	0.69 [0.10, 4.93]	0.14	0.712
Child age				2.05 [1.72, 2.45]	64.36	0.000	1.42 [0.96, 2.10]	3.01	0.083	1.42 [0.96, 2.11]	3.03	0.082
Child gender (1 = boy)				0.97 [0.59, 1.60]	0.02	0.904	0.96 [0.58, 1.60]	0.02	0.880	1.25 [0.37, 4.22]	0.13	0.714
Mild*age								4.06	0.131		4.18	0.123
Moderate*age							1.62 [1.00, 2.63]	3.79	0.052	1.62 [1.00, 2.64]	3.78	0.052
Severe*age							1.51 [0.93, 2.45]	2.82	0.093	1.55 [0.95, 2.52]	3.08	0.079
Mild*gender											0.05	0.188
Moderate*gender										1.19 [0.28, 5.02]	1.31	0.817
Severe*gender										0.43 [0.10, 1.84]	2.81	0.253

[a] *Odds ratio.*
[b] *Confidence interval.*

TABLE 10 | Relation between information provided for the child about severity of parent's condition, child age, child gender.

	Model 1			Model 2			Model 3			Model 4		
	$X^2(2) = 5.246, p = 0.073$			$X^2(4) = 88.778, p < 0.001$			$X^2(6) = 92.552, p < 0.001$			$X^2(8) = 95.957, p < 0.001$		
	OR[a] [95% CI[b]]	Wald	p	OR [95% CI]	Wald	p	OR [95% CI]	Wald	p	OR [95% CI]	Wald	p
Severity: Mild	Reference group											
Severity: Moderate	1.50 [0.77, 2.91]	1.41	0.235	1.50 [0.70, 3.19]	1.08	0.300	0.29 [0.05, 1.75]	1.83	0.176	0.27 [0.04, 2.01]	1.63	0.202
Severity: Severe	2.12 [1.08, 4.15]	4.75	0.029	1.94 [0.90, 4.19]	2.88	0.090	0.47 [0.08, 2.83]	0.68	0.410	0.69 [0.10, 4.93]	0.14	0.712
Child age				2.05 [1.72, 2.45]	64.36	0.000	1.42 [0.96, 2.10]	3.01	0.083	1.42 [0.96, 2.11]	3.03	0.082
Child gender (1 = boy)				0.97 [0.59, 1.60]	0.02	0.904	0.96 [0.58, 1.60]	0.02	0.880	1.25 [0.37, 4.22]	0.13	0.714
Mild*age								4.06	0.131		4.18	0.123
Moderate*age							1.62 [1.00, 2.63]	3.79	0.052	1.62 [1.00, 2.64]	3.78	0.052
Severe*age							1.51 [0.93, 2.45]	2.82	0.093	1.55 [0.95, 2.52]	3.08	0.079
Mild*gender											0.05	0.188
Moderate*gender										1.19 [0.28, 5.02]	1.31	0.817
Severe*gender										0.43 [0.10, 1.84]	2.81	0.253

[a] *Odds ratio.*
[b] *Confidence interval.*

the COPMI. Specifically, there is need for interventions which can be tailored to single parents with moderate to severe mood disorders, as well as recurrent disorders. The finding that parents with mental and behavioral disorders due to psychoactive substance use are less likely to have their children at home, also emphasize the need for interventions that suit these families' needs. It seems evident that these parents and their children need support to communicate and engage with each other in safe ways, as well as support if they are moving together again.

The Characteristics of the Children and Their Living Circumstances

One third of the children in this study were 0–5 years old, one third were 7–11 years old and one third were 12–18 years old. Fourteen percent of them were 2 years or younger, and as such within the 1,001 critical days defined by the well-known manifesto "The Importance of the Conception to Age Two Period" from the UK.[2] This manifesto, and others of its kind, highlights the importance of intervening early to enhance the outcomes for children at risk, and this is also a key objective for the development and implementation of interventions in Norway.

It seems fair to assume that most of the children under six in our study probably do not need outpatient help for their own psychological problems yet. However, unless they are identified and can access necessary support and preventive measures by health professionals treating the parent, only some of those who develop social and emotional problems themselves will be reached at a later point in life, and only after the problems have escalated to the point where the child needs to be referred to the specialist health service. This is demonstrated by the national figures for children who receive treatment in Child and adolescent mental health services (CAMHS), where <7% of patients are boys under 7 years, and <5% are girls below the age of seven. Less than 1% of patients in Norwegian CAMHS are under 4 years of age.

At the same time as the youngest children largely do not receive treatment by the mental health services for children and young people, they are especially vulnerable developmentally. Once problems have emerged, they have the worst prognosis and there are relatively few documented effective interventions and measures designed for this age group (4). In this study, no information was collected about how old the child was when the parent's illness was presented. However, recurrent depression was a common disorder in the sample, and for the youngest children, it is therefore reasonable to assume that a large part of their childhood had been affected by the parent's illness. In addition to this, a large majority of COPMI in the present study lived with the ill parent alone, either all the time or part time. As depression is a prevalent disorder in general psychiatric clinics, and has a documented negative effect on parent-child interaction (25), especially for the very youngest (17), feasible interventions for depressed parents of very young children should be a priority in general psychiatric clinics in

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[2] Durkan M, Field F, Lamb N, Loughton T. (2016). Available online at: https://www.1001criticaldays.co.uk/sites/default/files/1001%20days_oct16_1st.pdf

Norway. Collaboration between health personnel in adult mental health services and public nurses in local health clinics, as well as preschool teachers in kindergartens seems especially valuable for this group of children.

Positive social bonds between siblings are an important source of protection for children who undergo stressful life events (54). The majority of our sample had one or more siblings. Sibling bonds can be considered especially beneficial for COPMI with siblings who are older than 18 (16.1% in our sample), where the sister or brother can function as a supporting adult (55). At the same time, dysfunctional sibling bonds, which are not uncommon in families with mental illness, may be a burden and can predict a subsequent psychopathology for COPMI (56). This suggests that sibling relationships between COPMI should also be assessed in mental health services for adults. In our sample, 7.5% of the parents had four or more children, and account must be taken of the negative impact that a larger number of children can have on the family economy, thus contributing to the socio-economic risk (57), as well as the increase in parental burden and need for relevant support for patients with more children. Labor and Welfare Services should be a natural collaboration partner for health personnel in adult mental health services in these regards.

The majority of the children had access to adult resource persons other than the parent with mental illness or addictions. It was usually the other parent or extended family who cared for the child when the parent with a mental illness was hospitalized. In addition, most of the children were at school or in kindergarten during the day. Only a small proportion of the children in the sample were at home during the day (5.1%). These findings emphasize how important the extended family, and personnel in kindergartens and schools are as significant others who may support the child's development. However, the provision of necessary information about the risks and protective factors relevant for each COPMI is crucial to motivate and engage the resources that significant others may contribute with. Child protection services (CPS) may intervene in those families where the children do not have access to resource persons beside the mentally ill parent. However, in a recent study we found that personnel in adult mental health services was reluctant to refer families of concern to the CPS (58). In Norway around 80% of the activity in public child protection services constitutes of preventive and compensating interventions. Examples of such interventions are economic support, practical support for parents and children in the home and related to leisure time activities, visitation homes, as well as parent training interventions offering supervision and guidance for parents in need of professional help to change their parenting practices. Barriers to inter-service collaboration based on lack of knowledge about the CPS in adult mental health services has serious consequences for families in need of support and for dissemination of interventions in the communities they live. Results from our research indicate that there is an unresolved potential for inter-service collaboration involving the children of patients with mental health problems. There is a large potential for improvement in the collaboration between adult mental health services and community services for children and their families in Norway.

COPMI's Family Status and Factors Influencing Their Living Arrangements

Several factors were found that had an impact on the children's living arrangements and factors that led to an increased chance that the child did *not* live with the mentally ill parent were: (1) that the parent had serious mental illness, as compared with mild mental illness, (2) that the parent had an addictive disorder, and (3) that the ill parent was the father.

The severity of the diagnosis was a strong predictor and the odds were three times as high that a child did not live with a parent with a serious mental disorder compared to those with mild mental disorders. This is not a surprising finding, as the more serious mental disorders often lead to higher symptom pressure and malfunction, which to a greater extent leads to a risk of negative outcomes for the child. However, about three quarters of the children in our sample lived with the parent with a mental illness, and one third of them lived alone with this parent all the time or part time. These children will be more vulnerable and at a higher risk for developing mental disorders, as compared with those who also lived with a healthy parent (28).

The COPMI whose parents had addictive disorders were distinct from other COPMI, as their odds of not living with the ill parent were many times higher than for the children of parents whose primary diagnose were from the other categories. This was irrespective of the parent's gender. The reason why families and/or professional helpers choose to move children away from addictive parents is probably associated with substance abuse leading to risk in several different areas, in terms of a problematic social network, crime, physical and mental health problems, impulsive absences from the home, financial problems, on top of challenges with interactions when intoxicated.

About half of the children of mentally ill fathers lived with their fathers, while nearly nine out of ten children of mentally ill mothers lived with their mothers. The majority of children who lived with a single parent with a mental illness lived with their mother. In Norway, most children stay with their mother after a divorce between their parents (59), and it is therefore not possible to say whether it is the break-up or the father's illness that explains this. On the other hand, a previous study showed that mental disorders in fathers may reduce the involvement they have with their children (47). Taken together, our findings indicate that interventions supporting parents with severe mental disorders and their minor children, especially mothers, should be implemented. Service collaboration between community and specialist services is needed to target each family's needs in their everyday lives.

Factors Influencing Whether or Not COPMI Learn About Their Parent's Illness

Half of the children in the clinic had received information about the parent's condition and treatment/hospitalization. Four of ten COPMI had *not* received information about the parent's illness. This indicates that the ill parent and health care providers to a large degree do not speak with the children about mental disorders in parents. The reasons for this are most likely multifaceted, but nevertheless, researchers have postulated that a lack of information may result in an elevated risk of negative developmental outcomes and may run counter to the children's own wishes, needs and rights (7, 36, 38).

The following factors increased the likelihood that the children received information about the parent's condition and treatment/hospitalization: (1) that the child was living with a single parent with a mental illness, (2) that the child was an older child/teenager, (3) that the mother was the parent with a mental illness, (5) that the parent's primary diagnosis was a severe mental illness, and specifically (6) that the parent's disorder was a personality disorder, schizophrenia, or other psychotic disorder.

If the child lived alone with a single parent with a mental illness, the odds that the child had received information were significantly higher. Children who lived with a single parent at all times had almost three times the odds of receiving information about the parent's illness, as compared to children from two-parent homes and children who primarily lived with the other, healthy parent. This may imply that the parent with a mental illness or other adults realized that the child's opportunity to understand and handle their situation would depend on having information about the health status of their primary caregiver.

There were large age differences related to whether the children received information, and the odds that the children received information were higher for each year they grew older. The oldest children were far more likely to receive information about the treatment/hospitalization and about the parent's condition compared to the youngest children. Intuitively, this can be interpreted as a result of older children understanding more about the parent's illness, and thus to a greater extent taking the initiative to talk about it. However, studies have shown that few COPMI seek help from health care providers or their social network (34, 36), which may explain the fact that many of the older children in fact had not received information. It may also be the case that adults feel safer talking to children about a parent's illness when the children are more mature and have a more developed language. Although it could be expected that children under 2 years had generally not received information, the results showed that it was only from age 10 that more than half of the children had received information about the parent's illness. However, school-aged children have both the verbal and cognitive ability to understand phenomena, such as mental illness and substance abuse disorders and they will have perceived that their parents have problems of this type. Therefore, the finding that many children did not receive information about the parent's illness at all could be interpreted as a reflection of that parents and health care providers do not know how to talk to the youngest children about mental illness.(3) For the youngest children this may be due to a culture where adults (both parents and health care providers) have a desire to protect the younger children from "bad things." However, younger children will also be affected by the parent's psychopathology, sometimes even more than older children since they are more depending of predictable daily routines and sensitive care. Therefore, they will also need information adjusted to their age and maturity to help them understand the parent's symptoms, as well as how these affects parenting and the daily routines in the family. In circumstances when someone other than the parents need to take care of the child, an explanation for this should also be offered to the child.

Children of mentally ill mothers were more likely to receive information than the children of mentally ill fathers. This may be partly explained by children often having closer emotional ties to mothers than to fathers, that mother-daughter relationships often is characterized by emotional closeness and increased communication (60), as well as the fact that more of the children lived with their mother than their fathers.

More serious disorders were associated with a greater probability that the child had received information. This may be because these disorders to a greater extent lead to functional impairments in parents, which increases the negative impact on the child and thus the necessity of the child receiving information about the condition.

Children of parents with personality disorders, schizophrenia, schizotypal disorder and other paranoid disorders, also had higher odds of receiving information. A possible explanation for this may be related to the parents having such significant functional impairments and behaving so differently that it would be difficult to keep information about this from the children. For example, psychoses and delusions may present in ways that are very frightening for children and thus necessitate an explanation earlier than many other types of disorders. Again, substance abuse disorders differed from other diagnosis as it led to lower odds that the child received information. Substance abuse disorders are subject to great social stigma (61), and this may be part of the reason that parents are strongly motivated to hide their disorder from their children. It may also be the case that these parents are more worried about losing custody of their children, compared to parents with other mental disorders, and as such openness would seem counterproductive. The stigma of substance abuse disorders may also explain why personnel in mental health care services do not inform the children. Unfortunately, there are few interventions available for children whose parents have severe mental illnesses in Norwegian municipalities. Such interventions should be adopted from other countries, translated, and adapted to the Norwegian context, or developed locally. All new practices should be evaluated to gain knowledge about effects and feasibility.

Limitations

The most important limitation in this study is that the information is based on parents' reports. Other sources of information, such as interviews with the children themselves, as well as supplementary information from teachers, nurses and other relatives, would have given a more accurate, detailed, and comprehensive picture of the children's situation. However, gathering information in this way was not within the scope of the quality assurance framework in this study.

Another limitation is associated with the analyses. To be able to perform a binary logistic regression where two information to the children variables were included, the response "partially" was coded as "yes." These analyses must therefore include the caveat that "partially" could mean anything from almost no information to quite a bit information. Moreover, the extent to which the categorization of diagnoses according to ICD chapters reflect patients' symptom pressures and daily functioning is uncertain. Additionally, many of the diagnostic categories did not have a sufficiently large selection to be included in the analyses, and were therefore excluded.

The categorization of diagnoses by severity was done in accordance with Kessler et al. (22) categorization. They included separate functional goals in their study, which was not included in this study. The severity categorization was therefore only based on the diagnoses, and no controls was made for the fact that psychiatric disorders and their outcomes represent a spectrum of outcomes rather than exact functional measurements. There may therefore be limited correspondence between the assumed and actual degree of severity.

CONCLUSION AND IMPLICATIONS FOR FUTURE CLINICAL PRACTICE AND RESEARCH

The need for new measures seem to be especially significant for COPMI aged 0–5, as this group seems to be the most invisible, and most vulnerable group, while at the same time having few services offered. The finding that this group also receive the least information about their parent's psychopathology, legitimize significant concern. Norwegian law requires health care providers to attend to the children's need for follow-up, but also to ensure that the children receive the necessary information. The youngest group of COPMI is an important target group for early interventions, according to both national and international manifestos, and measures should therefore be developed to support these children and their parents.

The finding that only half of the children receive information about the parent's psychopathology clearly demonstrates that mental health services for adults must be enabled to comply with current legislation. Close to a third of the children live alone with a mentally ill parent, and thus may not have daily access to an adult who can compensate for the functional impairment the parent suffers in their everyday life at home. These children will need significant others to support their development. Enhancing inter-service collaboration seems crucial to reduce the risk that COPMI will develop social problems and mental health disorders themselves.

Based on the findings in this study, there seems to be a strong cause for concern about the continued transmission of psychiatric disorders from one generation to the next. Parents need support and help to inform their children about their mental health problems, and this is especially important for parents of young children. The findings in this study demonstrate how identifying COPMI and their living arrangements, can inform mental health workers about which type of support and interventions their patients need the most.

ETHICS STATEMENT

The data drawn from electronic patient journals in this study is information which health personnel is required by law to obtain. The Data Protection Officer at the University Hospital of Northern Norway approved the quality evaluation project.

AUTHOR CONTRIBUTIONS

All authors agree to be accountable for the content of the work. CR and CL designed the project. CR collected the data. Analyses were conducted by KR, YS, JF, and CR. CR drafted the article. All authors participated in the writing of the manuscript.

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BRIEF RESEARCH REPORT
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Parental Risk for Suicide and Attachment Patterns Among Adolescents With Borderline Personality Disorder. A Clinical-Based Study

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Introduction: Experiencing adverse life events and early disturbed patterns of interaction are crucial determinants for the development of Borderline Personality Disorder (BPD). Parental suicidal attempts can be considered a major adverse life event and a potentially traumatic experience. The aim of this study was to examine differences in parental suicidal attempts among a BPD adolescent population vs. a matched control group. We also aimed to understand if attachment styles and the number of parental suicidal attempts predicted the severity of borderline symptomatology.

Methods: Our study (EURNET BPD) comprised 85 BPD adolescents and 85 matched controls. Axis II disorders were investigated using the French version of SIDP-IV. Parental suicidal behaviors were assessed during a face-to-face interview with the adolescent. Attachment style was assessed with the Relationship Questionnaire (RQ).

Results: Parents of BPD adolescents made more suicidal attempts than controls (34 vs. 11%; chi2 = 13.8, $p < 0.001$). The linear regression showed that the best model explaining the severity of borderline symptomatology (R^2 adjusted = 0.15, $F = 4.65$, $p < 0.001$) was a model including the number of suicidal attempts realized by the parent (standardized beta = 0.28, $p = 0.009$) and the preoccupied attachment style of the adolescents toward their parents (standardized beta = 0.21, $p = 0.043$).

Conclusion: Our results highlight the usefulness of assessing parental suicidal behavior when interviewing patients with BPD traits. This could be useful in clinical practice as an early clue to identify patients with potentially severe clinical profiles and parents needing a specific support. Further research is needed to confirm our findings.

Keywords: borderline personality disorder, BPD, parental suicidal attempts, preoccupied attachment, adolescent

INTRODUCTION

Borderline Personality Disorder (BPD) in adolescence is a severe mental disorder characterized by pervasive and persistent patterns of instability—affective, relational, of the self—and impulsivity. The condition is associated with frequent risk-taking and self-harm behaviors and many psychiatric comorbidities as well as severe psychosocial impairments, and extensive use of mental health services. Cumulative prevalence rates suggest that 1.4% of young people will meet diagnostic criteria for BPD by age 16 years, rising to 3.2% by age 22 years (1), but more importantly BPD is common among adolescents within mental health settings, with an estimated prevalence of 11% in psychiatric outpatients and up to 50% in inpatient settings (2).

Although currently there isn't a unifying model to describe and understand in depth the complex phenomenon of borderline personality, genetics, and environmental factors are both thought to play a role. Concerning environmental factors, experiencing adverse life events and trauma have been largely investigated in the past (3, 4). However, not all individuals who suffer maltreatment will develop a BPD (5). Early disturbed patterns of interaction constitute a risk factor for emotional vulnerability and are crucial determinants for the development of BPD. Maladaptive parenting, including parental hostility, overprotective and rejecting parenting styles and attachment disorganization emerged as strong predictors of BPD symptoms (6, 7). The quality of primary relationships is so important that it might modulate in one way or the other the integration of early traumatic events (5) and therefore the development of personality.

To our knowledge, parental suicide hasn't been studied specifically in adolescents with BPD. Nevertheless, it has been recognized that parental suicide attempts can have a major impact in the offspring and can be considered a major adverse life event and a potentially traumatic experience (8). Parental suicidal attempt can also question the attachment in the young person, through the fear of potential abandonment (9). Besides, behind a suicidal attempt, psychopathology and social adversity is often hidden (10).

The primary aim of this study was to examine differences in parental suicidal attempts among a BPD population vs. a matched control group. We also aimed to test whether attachment styles and the number of parental suicidal attempts predicted the severity of borderline symptomatology.

METHODS

Participants

The study sample was drawn from a European research project investigating the phenomenology of BPD in adolescence [the European Research Network on Borderline Personality Disorder, EURNET BPD; see (11) for a full description of the study methodology]. The research network was composed of five specialist psychiatric centers for adolescents and young adults in France, Belgium, and Switzerland. The final study population comprised 85 BPD adolescents (11 boys, 13%, and 74 girls, 87%). The mean age was 16.5 years (SD = 1.4). 67% ($N = 57$) were inpatients. The control sample included 85 healthy adolescents individually matched for gender, age and socio-economic status. Control subjects were excluded if they had a history of or ongoing psychiatric follow-up, and if they were positive for a DSM-IV diagnosis of personality disorder. The study was approved by our local institutional review board and a written informed consent was obtained from the adolescents and at least one of their parents.

Assessments

All subjects completed a research protocol (consisting of a diagnostic evaluation of Axis I and Axis II disorders) and a self-administered questionnaire to collect socio-demographic and psychopathological data. Axis II disorders were investigated using the French version of SIDP-IV, which is known to have good psychometric properties in adolescents and young adults (12). Borderline severity for each of the 9 criteria was coded as absent (0), subliminal (1), present (2), and severe (3). Borderline severity scores thus varied from 0 to 27. The inter-rater reliability for SIDP-IV was calculated from independent ratings of 10 videotaped interviews. The Kappa coefficient for agreement on the presence or absence of a BPD was very high (0.84) and the values for the presence/absence of other personality disorders ranged from 0.54 to 1.

Parental suicidal behaviors (presence/absence; number of suicidal attempts) were assessed during the face to face interview with the adolescent. The age of the adolescent at the time of the first suicidal attempt of the parent was also recorded.

Attachment was assessed with the Relationship Questionnaire (RQ) (13), a valid and reliable self-report measure of adult attachment [(14, 15) measuring strength of attachment along two dimensions of anxiety and avoidance. The RQ generates a negative to positive score (-12 to $+12$) on the attachment dimensions of anxiety and avoidance, with higher scores indicating less anxiety and avoidance respectively.

Statistical Analysis

Categorical variables (socio-demographics, presence/absence of parental suicidal attempts, insecure/insecure attachment) were compared between groups (borderline and control sample) using chi-square analyses. To compare the mean scores of the borderline and the control sample on the number of parental suicidal attempts and on attachment styles we used a one-way anova. Bivariate associations between attachment styles and the overall severity of borderline symptomatology were examined with Pearson correlations in the borderline sample only. Finally, to test whether attachment styles and the number of parental suicidal attempts predicted the severity of borderline symptomatology we conducted a linear regression analysis with a stepwise procedure. All analyses were conducted using SPSS (25th version).

RESULTS

Parental Suicidal Attempt

34% ($N = 29$) adolescents from the borderline group reported a suicidal attempt made by their parents compared to 11% (N=9) from the control sample. This difference was statistically significant (chi2 = 13.8, $p < 0.001$) (**Table 1**). All the parents of

TABLE 1 | Comparison between the borderline and control groups regarding parental suicidal attempts and attachment patterns.

Variables	Borderline sample ($N = 85$)	Control sample ($N = 84$)	Statistics	
	M ± SD/(%)	M ± SD/(%)	F/Chi2	P
Age	16.5 ± 1.4	16.2 ± 1.3	F = 2.71	0.10
Sex (men) (%)	11 (13%)	20 (24%)	F = 3.3	0.70
Parental suicidal attempt (present/absent) (%)	29 (24%)	9 (11%)	Chi2 = 13.8	**0.001**
N° of parental suicidal attempts	4.2 ± 3.7	1 ± 0	F = 5.7	**0.001**
RQ Secure attachment	2.9 ± 1.6	4.2 ± 1.6	F = 23.4	**0.001**
RQ Fearful attachment	4.0 ± 1.9	2.7 ± 1.5	F = 19.3	**0.001**
RQ Preoccupied attachment	4.2 ± 2.0	3.1 ± 1.7	F = 12.0	**0.001**
RQ Dismissing attachment	2.5 ± 1.8	2.5 ± 1.5	F = 0.01	0.9
Secure/insecure attachment (%)	34%	23%	F = 25.1	**0.001**

Statistically significant differences appear in bold.

TABLE 2 | Multiple regression model predicting the severity of borderline symptomatology.

Predictive variables	Estimate	SE	b	T	P
N° of parental suicidal attempts	3.79	1.4	0.28	2.68	0.009
Preoccupied attachment	0.50	0.2	0.21	2.05	0.043

Linear regression model. Stepwise method. Dependent variable: Severity of borderline symptomatology (SIDP-IV), Independent variables: attachment styles (RQ), n° of parental suicidal attempts, n° of adolescent suicidal attempts. Sample: N = 85. Adjusted $R^2 = 0.21$, $F_{(2,63)} = 4.65$, $p < 0.001$.

the control group had made only one suicidal attempt, while the parents of the borderline group reported several suicidal attempts (4.2, SD 3.7) ($F = 5.7$, $p = 0.022$) with some parents reporting up to 10 suicidal attempts. Mothers accounted for the majority of the suicidal attempts (73%). Borderline adolescents reported that the first suicidal attempt of their parents happened during their childhood and early adolescence (mean 12.7, SD 4.0).

Attachment Styles

Data from the RQ showed that borderline adolescents showed more insecure attachments than controls (34.5 vs. 23%, $F = 25.1$, $p = 0.001$) (**Table 1**). Borderline adolescents had significantly lower scores on secure attachment and significantly higher scores on the fearful and preoccupied styles of attachment.

Correlations to Borderline Severity

Pearson correlations showed that the number of parental suicidal attempts ($r = 0.37$, $p < 0.05$) was significantly correlated to the severity of borderline symptomatology, with fearful and preoccupied attachment styles showing a trend to a significant correlation (**Table 2**).

The linear regression showed that the best model explaining the severity of borderline symptomatology ($R^2_{\text{adjusted}} = 0.15$, $F = 4.65$, $p < 0.001$) was a model including the number of suicidal attempts realized by the parent (standardized beta = 0.28, $p = 0.009$) and the preoccupied attachment style of the adolescents toward their parents (standardized beta = 0.21, $p = 0.043$) (**Table 2**).

DISCUSSION

This study sought to determine whether parental suicidal attempts (and single vs. multiple attempts) could differ in a BPD adolescent population vs. control. First, our study shows that parents of BPD adolescents (specially mothers) made more suicidal attempts than controls. These suicidal attempts happened several years earlier, when patients were still children, or in early adolescence.

Second, the severity of borderline symptomatology seemed to correlate with the number of parental suicidal attempts (the higher the number of suicidal attempts, the increased BPD severity in the offspring) and preoccupied attachment style.

Psychopathology is a frequent underlying cause of suicidal attempts. Severe depression, anxiety and substance abuse are commonly found among suicidal attempters (16). Studies who compared single vs. multiple suicidal attempters in adults showed no differences in axis I disorders but found an increased prevalence of borderline personality disorders and higher impulsivity scores in the latter group (16, 17). Therefore, the likelihood of presenting a personality disorder increases with the number of suicidal attempts. Family members of individuals with BPD may be at higher risk of axis II disorders, including BPD (18, 19) via a potential transgenerational transmission of emotional dysregulation from parent to child (creating insecure attachment patterns). This could explain the impact of parental suicidal attempts on BPD severity in the offspring found in our study as well as the trend to present a preoccupied attachment style.

Our study is somewhat limited by the sampling method, size, and cross-sectional design. Indeed, our data comes from adolescents only, who report their perceived patterns of interaction. Yet number of parental suicidal attempts' report is a rather objective data and less sensible to bias.

CONCLUSION

Our results highlight the usefulness of assessing parental suicidal behavior when interviewing patients with BPD traits. This pattern of behavior is overrepresented in this population and might correlate with psychopathological severity in the offspring. This could be useful in clinical practice as an early clue to identify patients with potentially severe clinical profiles and parents needing a specific support. Further research is needed to confirm our findings.

ETHICS STATEMENT

This study was approved by the ethics committee of the Hôtel Dieu Hospital in Paris (authorization n° 0611259). Results were collected in an anonymous database according to the requirements of the French national committee for private freedoms. All participants, adolescents and parents, signed informed consent after receiving a full description of the study, explanation of its purpose, and information about the confidentiality of the data.

AUTHOR CONTRIBUTIONS

All the authors listed in the manuscript have contributed sufficiently to the project to be included as authors. AM participated in data analysis and interpretation and in writing the manuscript. MS, MC, and AP-S initiated and designed the protocol, collected data, participated in data analysis and interpretation and revising the manuscript. VD participated in data analysis and interpretation and revising the manuscript. All authors read and approved the final manuscript.

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Protective Factors for Early Psychotic Phenomena Among Children of Mothers With Psychosis

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Background: Early identification of sub-clinical psychotic experiences in at-risk individuals is vital to prevent the development of psychosis, even before prodromal symptoms emerge. A widely-replicated risk factor is having a family member with psychosis. The Environmental Risk (E-Risk) Longitudinal Twin Study has shown that better cognitive functioning, a stimulating family environment, and a cohesive community, are protective against psychotic experiences among children; while engaging in physical activity, social support, and a cohesive community are protective for adolescents. In the current study we investigate whether these factors also protect against the development of sub-clinical psychotic phenomena among children and adolescents in this cohort who are at high-risk of psychosis by having a mother with psychosis.

Methods: Data were utilized from the E-Risk Longitudinal Twin Study, a nationally-representative cohort of 2,232 twin children born in England and Wales in 1994–1995 followed to age 18. Psychotic phenomena were assessed in private interviews with children at ages 12 and 18, and mothers were interviewed about their own experiences of psychosis when children were aged 10 and 12. Bivariate and multivariate logistic regression analyses explored associations between individual, family, and community-level putative protective factors and absence of age-12 psychotic symptoms and age-18 psychotic experiences in children whose mothers had a diagnosis of a psychosis-spectrum disorder and/or reported psychotic symptoms.

Results: Higher IQ (OR = 0.97, 95% CI 0.94–1.00, $P = 0.036$) and living in a more socially cohesive neighborhood (OR = 0.88, 95% CI 0.79–0.98, $P = 0.023$) were independently protective against age-12 psychotic symptoms among children of mothers with psychosis. Higher levels of perceived social support were independently protective against age-18 psychotic experiences among children of mothers with psychosis (OR = 0.92, 95% CI 0.87–0.98, $P = 0.006$). However, there were no significant interactions between these protective factors and maternal psychosis in relation to an absence of childhood or adolescent psychotic phenomena in the full sample, indicating that protective effects were not specific to this group of high-risk children.

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Conclusions: These findings provide preliminary evidence that preventive interventions for early psychotic phenomena could focus on improving cognition, social support, and cohesiveness of the local community. Given scarce resources these might usefully be targeted at high-risk children.

Keywords: child and adolescent mental health, early intervention, familial psychosis, maternal psychosis, prevention, protective factors, psychotic experiences, resilience

INTRODUCTION

Psychotic disorders, especially schizophrenia, are responsible for a substantial proportion of disability worldwide (1), are associated with 10–25 years shorter life expectancy (2), and place a huge burden on families, health services, and society (3–5) Current treatments are unable to provide a cure; therefore, early identification of at-risk individuals may help to prevent the development of psychosis. Interventions in the prodromal stage have yielded some success but often fail to prevent poorer functional outcomes in the longer-term (6). It may therefore be necessary to intervene even earlier, before prodromal symptoms have emerged (7).

One potential target for preventative interventions could be sub-clinical psychotic experiences. These experiences, such as hearing voices, having visions, or feeling extremely paranoid, are relatively common in the general population, especially among young people. A systematic review and meta-analysis of population-based studies concluded that the median prevalence of psychotic symptoms was 17% among children aged 9–12 years, and 7.5% among adolescents aged 13–18 years (8). Early-life psychotic experiences are thought to lie on a continuum with psychotic disorders (9) and have been shown to predict elevated rates of schizophrenia by age 38 (10) and share many of the risk factors commonly associated with psychotic disorders (11). Moreover, these sub-clinical psychotic phenomena have also been shown to increase the risk for other psychiatric disorders (12), suicide attempts (10), and poor functional outcomes (13) in adulthood.

Therefore, one approach to informing early preventive interventions for psychotic disorders and potentially other adverse outcomes is to investigate factors that protect at-risk children and adolescents from developing psychotic experiences. A widely replicated risk factor for psychosis is having a family member with the disorder; for instance, having a parent with schizophrenia increases a child's risk of developing schizophrenia themselves by approximately eight-fold (14). A previous analysis of the Environmental Risk (E-Risk) Longitudinal Twin Study, a nationally-representative general population sample of twins, has shown that children were over twice as likely to report psychotic experiences at age 12 if their mother had experienced a psychosis-spectrum disorder (11). This indicates that young people whose mother has experienced psychosis could be considered a high-risk group for the development of early psychotic phenomena. However, not all offspring of affected mothers developed psychotic experiences (11) and thus it is important to understand what protected these high-risk individuals in order to inform preventive interventions.

In the E-Risk sample, it has previously been shown that, in the context of poly-victimization, having better cognitive functioning, growing up in a happier and more stimulating family environment, and living in a cohesive community, were protective against psychotic experiences among children (15), while engaging in physical activity, social support, and living in a cohesive community were protective for adolescents (16). However, it is also important to determine protective factors for other high-risk groups, such as having a parent with psychosis. Therefore, in the current study we use the E-Risk cohort to investigate whether these factors can also protect against the development of sub-clinical psychotic phenomena among children and adolescents who are at particularly high-risk of psychosis, by virtue of having a mother with a psychotic disorder or psychotic symptoms. We explored whether these factors were protective over and above potential confounding factors, including family socioeconomic status (SES) and other childhood mental health problems, which have previously been shown to be associated with the development of psychotic phenomena (11).

MATERIALS AND METHODS

Participants

Participants were members of the E-Risk Longitudinal Twin Study, which tracks the development of a nationally-representative birth cohort of 2,232 British twin children. The sample was drawn from a larger cohort of twins born in England and Wales in 1994–1995 (17). Full details about the sample are recorded elsewhere (18). Briefly, the E-Risk sample was constructed in 1999–2000, when 1,116 families with same-sex 5-year-old twins (93% of those eligible) participated in home-visit assessments. Families were recruited to represent the UK population of families with new-borns in the 1990s, based on residential location throughout England and Wales and mothers' age. Teenaged mothers with twins were over-selected to replace high-risk families who were selectively lost to the register through non-response. Older mothers having twins via assisted reproduction were under-selected to avoid an excess of well-educated older mothers. E-Risk families are representative of UK households across the spectrum of neighborhood-level deprivation: 25.6% of E-Risk families live in "wealthy achiever" neighborhoods compared to 25.3% of households nation-wide; 5.3 vs. 11.6% live in "urban prosperity" neighborhoods; 29.6 vs. 26.9% live in "comfortably off" neighborhoods; 13.4 vs. 13.9% live in "moderate means" neighborhoods; and 26.1 vs. 20.7%

live in "hard-pressed" neighborhoods (19, 20). E-Risk families under-represent "urban prosperity" neighborhoods because such households are likely to be childless. The sample comprised 56% monozygotic and 44% dizygotic twin pairs, and sex was evenly distributed within zygosity (49% male). All families were English speaking, and the majority (93.7%) were White.

Follow-up home-visits were conducted when children were aged 7, 10, 12, and 18 years (participation rates were 98, 96, 96, and 93%, respectively). Home visits at ages 5, 7, 10, and 12 years included assessments with participants as well as their mother (or primary caretaker); the home visit at age 18 included interviews only with the participants. Each twin participant was assessed by a different interviewer. The average age of the twins at the time of the age 18 assessment was 18.4 years ($SD = 0.36$); all interviews were conducted after the 18th birthday. There were no differences between those who did and did not take part at age 18 in terms of socioeconomic status (SES) assessed when the cohort was initially defined ($\chi^2 = 0.86$, $P = 0.65$), age-5 IQ scores ($t = 0.98$, $P = 0.33$), or age-5 internalizing or externalizing behavior problems ($t = 0.40$, $P = 0.69$ and $t = 0.41$, $P = 0.68$, respectively).

The Joint South London and Maudsley and the Institute of Psychiatry Research Ethics Committee approved each phase of the study. Parents gave informed consent and twins gave assent between 5 and 12 years and then informed consent at age 18.

Measures

Maternal Psychosis-Spectrum Disorder

When children were aged 10, mothers were interviewed using the Diagnostic Interview Schedule (DIS) for DSM-IV (21), which enquires about characteristic symptoms of psychosis: hallucinations, delusions, disorganized speech, grossly disorganized, or catatonic behavior and negative symptoms (avolition, flat affect, alogia). They were asked whether they had experienced these symptoms at any time in their life. The interview ruled out symptoms with plausible explanations and symptoms occurring solely under the influence of alcohol or drugs. Following DSM-IV criteria for schizophrenia, women were classified as having a psychosis-spectrum disorder given the presence of hallucinations plus at least two other symptoms, as well as evidence of social, occupational, or self-care dysfunction (12). The goal was not to diagnose clinical schizophrenia, but to identify women who endorsed impairing psychotic-like experiences and beliefs. Of the 1,060 mothers who completed the DIS, 58 (5.5%) were classified as having a psychosis-spectrum disorder.

Maternal Psychotic Symptoms

When children were aged 12, mothers were interviewed with the Psychosis Screening Questionnaire (PSQ) (22) about psychotic symptoms that they experienced over the past 2 years. The PSQ consists of 6 main items covering symptoms of hypomania, thought insertion, paranoia, strange experiences and hallucinations, and 14 follow-up items. The six main items were all presented first and, if any main items were endorsed, the appropriate follow-up questions were asked. A symptom was considered present if the mother positively endorsed the main item and its follow-up questions. The number of psychotic symptoms was summed and then dichotomized into none vs. 1 or more psychotic symptoms due to the skewed nature of the variable. Of the 1,069 mothers who completed the PSQ, 177 (16.6%) reported one or more psychotic symptoms.

Childhood Psychotic Symptoms

E-Risk families were visited by mental health trainees or professionals when children were aged 12 (11). Each child was privately interviewed about 7 psychotic symptoms pertaining to delusions and hallucinations, with items including "have other people ever read your thoughts?," "have you ever thought you were being followed or spied on?," and "have you ever heard voices that other people cannot hear?." This interview has been described in detail previously (11). The item choice was guided by the Dunedin Study's age-11 interview protocol (12) and an instrument prepared for the Avon Longitudinal Study of Parents and Children (23). Interviewers coded each experience 0, 1, 2 indicating, respectively "not a symptom," "probable symptom," and "definite symptom." A conservative approach was taken in designating a child's report as a symptom. First, the interviewer probed using standard prompts designed to discriminate between experiences that were plausible (e.g., "I was followed by a man after school") and potential symptoms (e.g., "I was followed by an angel who guards my spirit") and wrote down the child's narrative description of the experience. Second, items and interviewer notes were assessed by a psychiatrist expert in schizophrenia, a psychologist expert in interviewing children, and a child and adolescent psychiatrist to verify the validity of the symptoms. Third, because children were twins, experiences limited to the twin experience (e.g., "My twin and I often know what each other are thinking") were coded as "not a symptom." Children were only designated as experiencing psychotic symptoms if they reported at least one definite psychotic symptom. At age 12, 5.9% ($N = 125$) of children reported experiencing psychotic symptoms. This is similar to the prevalence of psychotic symptoms in other community samples of children and adolescents (8, 24–27). Furthermore, our psychotic symptom measure has good construct validity, sharing many of the genetic, social, neurodevelopmental, and behavioral risk factors and correlates as adult schizophrenia (11).

Adolescent Psychotic Phenomena

The present study used two measures of adolescent psychotic phenomena which were both obtained from private interviews when participants were aged 18. The primary outcome was a self-report measure of adolescent psychotic *experiences* which reflects the methodology used by many groups in the psychosis prodromal research field (28). At age 18, each E-Risk participant was privately interviewed by a research worker about 13 psychotic experiences occurring since age 12. Seven items pertained to delusions and hallucinations, which were the same as those used in childhood (see above). This interview has been described in detail previously (11). Six items pertained to unusual experiences, which drew on item pools since formalized in prodromal psychosis instruments including the PRIME-screen and Structured Interview for Prodromal Syndromes (28). These included "I worry that my food may be poisoned" and "My thinking is unusual or frightening." Interviewers coded each item 0, 1, 2 indicating, respectively "not present," "probably present," and "definitely present." All 13 items were summed to create a psychotic experiences scale (range = 0–18, $M = 1.19$, SD = 2.58). Just over 30% of participants had at least one psychotic experience between ages 12 and 18 ($n = 623$, 30.2%). This is similar to the prevalence of self-reported psychotic experiences in other community samples of teenagers and young adults (8, 25).

Clinician-verified adolescent psychotic *symptoms* were also examined as a secondary outcome, using the same methodology as used at age 12 in this cohort (11). Adolescents were only designated as having psychotic *symptoms* if they reported at least one definite and verified symptom. At age 18, 2.9% ($N = 59$) of adolescents reported having one or more psychotic symptoms since age 12. This is somewhat lower than the prevalence of psychotic symptoms in this sample at age 12 (5.9%, $N = 125$), consistent with the attenuation of psychotic symptoms documented from childhood to adulthood (8, 29).

Childhood Protective Factors

Iq

The Wechsler Preschool and Primary Scale of Intelligence Revised (WPSSI; (30) was used to assess IQ at age 5. Children were administered two subtests (Vocabulary and Block Design), and IQ scores were prorated following procedures described previously (31) and then standardized with a mean of 100 and standard deviation of 15.

Atmosphere at home

The creation of the atmosphere at home measure has been previously documented (32). It was derived from the Coder's Impression Inventory, which is based on the Home Observation for Measurement of the Environment (33) and the University of Washington Parenting Clinic Questionnaire (Parent-Child Observations) (34). The Coder's Impression Inventory was rated immediately following the study visit at ages 7 and 10 by interviewers who had undergone 4-day training. This measure comprised items representing the state of the home (e.g., "Are visible rooms of the house clean?"), stimulation (e.g., "Is the children's art displayed in the home?"), happiness (e.g., "Is this a happy home?"), and chaos (e.g., "Is the house chaotic or overly noisy?"). The internal consistency at age 7 was $\alpha = 0.77$ and $\alpha = 0.79$ at age 10. The average of the overall atmosphere at home scores at ages 7 and 10 was used for analysis because they were significantly correlated ($r = 0.64$, $P < 0.001$).

Neighborhood social cohesion

Social cohesion within the neighborhood (35) was assessed when children were aged 5 by asking mothers five questions, including whether their neighborhood was close-knit, whether neighbors shared values, and whether neighbors trusted and got along with each other. A total score was derived by summing the answers to all five questions, with higher scores indicative of greater social cohesion.

Adolescent Protective Factors

Physical activity

At age 18, participants completed the Stanford Brief Activity Survey (SBAS; Stanford University, 2001). The SBAS contains 2 items, the first item relates to the extent of physical activity engaged in at work, school, or college and the second refers to physical activity during leisure time. Both questions were rated on a 5-point scale: inactive, low intensity, moderate intensity, hard intensity, and very hard intensity. The scales were then combined to derive an overall activity measure (36).

Neighborhood social cohesion

Social cohesion in the participants' neighborhoods was estimated via a postal survey sent to residents living alongside E-Risk families when participants were aged 13–14 (37, 38). Survey respondents, who were typically living on the same street or within the same apartment block as the participants in the study, reported on various characteristics of their immediate neighborhood. Five items (each coded 0–4) were assessed by asking residents whether their neighbors shared values and trusted and got along with each other, etc. A total score was derived by summing the answers to all 5 questions with higher scores indicating greater social cohesion.

Social support

Perceived social support was assessed at age 18 using the Multidimensional Scale of Perceived Social Support (MSPSS), which assesses individuals' access to supportive relationships with family, friends, and significant others (39). The 12 items in the MSPSS consist of statements such as "There is a special person who is around when I am in need" and "I can count on my friends when things go wrong." Participants rated these statements as "not true" (0), "somewhat true" (1) or "very true" (2). Scores were summed to produce an overall social support scale with higher scores reflecting greater social support (internal consistency: $\alpha = 0.88$).

Confounding Variables

Family SES was measured via a composite of parental income (total household), education (highest for mother/father), and occupation (highest for mother/father) when children were aged 5 (40). The three SES indicators were highly correlated (r's ranged from 0.57 to 0.68, all p's < 0.05) and loaded significantly onto one latent factor (factor loadings = 0.82, 0.70, and 0.83 for income, education, and occupation, respectively). This latent factor was categorized into tertiles (i.e., low-, medium-, and high-SES). A variable for childhood mental health problems was derived to capture children who met criteria for extreme anxiety, clinically-relevant depression symptoms, attention deficit hyperactivity disorder (ADHD), or conduct disorder by age 12. Anxiety was assessed when children were aged 12, via private interviews using the 10-item version of the Multidimensional Anxiety Scale for Children (MASC) (41). An extreme anxiety group was formed with children who scored at or above the 95th percentile ($N = 129$, 6.1%). Depression symptoms were assessed at age 12 using the Children's Depression Inventory (CDI) (42). Children who scored 20 or more were deemed to have clinically significant depressive symptoms ($N = 74$, 3.5%). ADHD was assessed using the DSM-IV and the requirement of symptom onset prior to age 12 was met if parents or teachers reported more than 2 ADHD symptoms at ages 5, 7, 10, or 12 years. We derived diagnoses of conduct disorder on the basis of mothers' and teachers' reports of children's behavior problems using the Achenbach family of instruments and additional DSM-IV items assessing conduct disorder which have previously been described (32). Conduct disorder was assumed present if it was diagnosed at ages 5, 7, 10, or 12 years. The childhood mental health problems variable was dichotomized to distinguish between the presence of any of the above mental health problems (coded 1) vs. the absence of any age-12 mental health problems (coded as 0), as used previously in this cohort (16).

Statistical Analysis

Analyses were conducted in STATA 15 (Stata-Corp, College Station, TX). Bivariate and multivariate logistic regression analyses were conducted to explore associations between individual, family, and community-level putative protective factors and absence of age-12 and age-18 psychotic phenomena, in the E-Risk sub-sample of children whose mothers had experienced psychosis. Maternal psychosis was defined as either diagnosis of psychosis-spectrum disorder and/or presence of one or more psychotic symptoms to utilize the largest possible group of mothers experiencing psychotic phenomena ($N = 408$).

In phase 1 (age-12 analysis), binary logistic regression analyses were conducted to explore associations between each protective factor (IQ, atmosphere at home, and neighborhood social cohesion), individually and then altogether, and the absence of age-12 psychotic symptoms in the sub-sample of children whose mothers had experienced psychosis. We also tested in the whole sample for interactions between maternal psychosis status and each protective factor to examine whether these factors were specifically protective in relation to having a mother who had experienced psychotic phenomena or were more generally protective.

In phase 2 (age-18 analysis), binary logistic regression analyses were conducted to explore associations between each protective factor (physical activity, neighborhood social cohesion, and perceived social support), individually and then altogether, and the absence of age-18 psychotic experiences in the sub-sample of children whose mothers had experienced psychosis. Interactions between maternal psychosis status and each protective factor were conducted to examine whether these factors were specifically protective in relation to having a mother who had experienced psychotic phenomena. Sensitivity analyses substituting age-18 psychotic experiences with the clinician-verified psychotic symptoms at age 18 were also conducted.

Because each study family contains two children, all statistical analyses were corrected conservatively for the non-independence of twin observations by using tests based on the Huber/White variance estimator (43). All analyses were also adjusted for child's gender and family socio-economic status because these factors have previously been associated with psychosis (44, 45). Age-18 analyses were also adjusted for age-12 psychotic symptoms and childhood mental health problems as these have been shown to predict the occurrence of psychotic phenomena in adolescence (29, 46).

RESULTS

Phase 1: Age-12

Sample

The sample ($N = 2{,}182$) was 51.1% ($N = 1{,}114$) female. Family SES was categorized into low- (33.2%, $N = 724$), medium- (32.8%, $N = 716$), and high-SES (34.0%, $N = 742$). Having a mother with either a diagnosis of psychosis-spectrum disorder or at least one psychotic symptom was associated with an increased likelihood of age-12 psychotic symptoms among their children (OR = 2.14, 95% CI 1.38–3.31, $P = 0.001$).

Associations Between Putative Protective Factors and Absence of Age-12 Psychotic Symptoms Among Mothers With Psychosis

Having a higher IQ, a more positive atmosphere in the home, and residing in a more socially cohesive neighborhood were all found to be significantly protective against age-12 psychotic symptoms among children of mothers who had experienced either psychotic symptoms or had a psychosis-spectrum diagnosis after adjusting for gender and family SES (**Table 1**). When additionally controlling for the other protective factors, higher IQ (OR = 0.97, 95% CI 0.94–1.00, $P = 0.036$) and neighborhood social cohesion (OR = 0.88, 95% CI 0.79–0.98, $P = 0.023$) remained significantly associated with a reduced likelihood of the child having psychotic symptoms, but the association for atmosphere at home became non-significant (OR = 0.96, 95% CI 0.92–1.01, $P = 0.133$).

Interactions Between Maternal Psychosis Status and Protective Factors in the Whole Sample

Mean IQ ($M = 101.1$, SD = 14.8), atmosphere at home ($M = 26.6$, SD = 4.8) and neighborhood social cohesion ($M = 7.8$, SD = 2.6) were highest when neither the child nor mother had symptoms or diagnosis of psychosis. Mean scores were lowest when both mother and child had psychotic symptoms or diagnosis: IQ ($M = 88.9$, SD = 14.6); atmosphere at home ($M = 19.5$, SD = 7.0); neighborhood social cohesion ($M = 5.3$, SD = 3.3). However, none of the interactions between maternal psychosis status and the protective factors were statistically significant (**Table 2**). Indeed, higher average IQ (OR = 0.98, 95% CI 0.96–0.99, $P = 0.002$) and a more positive atmosphere at home (OR = 0.93 95% CI 0.89–0.97, $P = 0.002$) were significantly associated with a reduced likelihood of age-12 psychotic symptoms in children of mothers *without* psychosis, with a non-significant trend for greater neighborhood social cohesion (OR = 0.93, 95% CI 0.85–1.01, $P = 0.071$).

TABLE 1 | Associations between putative protective factors and absence of age-12 psychotic symptoms among children of mothers with diagnosis or symptoms of psychosis.

Putative protective factor	**Psychotic symptoms absent** *N* = **359**	**Psychotic symptoms present** *N* = **40**	**Adjusted OR**^{a}	**95% CI**	***P***
	M (SD)	***M (SD)***			
IQ	97.84 (14.77)	88.88 (14.58)	0.96	0.94–0.99	0.007
Atmosphere at home	23.32 (6.92)	19.51 (6.96)	0.93	0.89–0.98	0.003
Neighborhood social cohesion	7.14 (3.02)	5.30 (3.28)	0.85	0.76–0.96	0.007

CI, confidence interval; IQ, intelligent quotient; OR, odds ratio; SD, standard deviation.

[a] *Adjusted for gender and family socioeconomic status. All analyses account for the non-independence of twin observations.*

TABLE 2 | Interactions between maternal psychosis and putative protective factors in relation to the absence of age-12 psychotic symptoms and age-18 psychotic experiences in the whole sample.

Interaction with maternal psychosis	**Adjusted OR**	**95% CI**	***P***
AGE-12			
IQ	0.98	0.95–1.01	0.300
Atmosphere at home	1.01	0.96–1.07	0.739
Neighborhood social cohesion	0.92	0.81–1.06	0.247
AGE-18			
Physical activity	1.08	0.87–1.34	0.494
Neighborhood social cohesion	0.86	0.51–1.43	0.553
Social support	1.01	0.94–1.07	0.818

CI, confidence interval; IQ, intelligent quotient; OR, odds ratio. All analyses are adjusted for gender and family socioeconomic status; age-18 analyses are additionally adjusted for age-12 psychotic symptoms and other age-12 mental health problems. All analyses account for the non-independence of twin observations.

Phase 2: Age-18

Sample

The sample ($N = 2{,}041$) was 52.3% ($N = 1{,}067$) female. Family SES was categorized into low- (33.4%, $N = 681$), medium- (33.1%, $N = 675$), and high-SES (33.5%, $N = 685$). Having a mother with either a diagnosis of psychosis-spectrum disorder or at least one psychotic symptom was associated with an increased likelihood of age-18 psychotic experiences among their children (OR = 1.56, 95% CI 1.21–2.01, $P = 0.001$).

Are Protective Factors Associated With an Absence of Age-18 Psychotic Phenomena Among Adolescents Whose Mother has Psychosis?

Greater levels of perceived social support were significantly associated with a reduced likelihood of adolescent psychotic experiences when controlling for all confounders among children of mothers with a psychosis-spectrum disorder or psychotic symptoms (**Table 3**). No associations were evident for physical activity, but there was a non-significant trend for greater neighborhood social cohesion to be associated with a reduced likelihood of adolescent psychotic experiences (OR = 0.67, 95% CI 0.42–1.08, $P = 0.101$). When additionally controlling for the other protective factors, higher levels of perceived social support remained significantly associated with a reduced likelihood of adolescents having psychotic experiences (OR = 0.92, 95% CI 0.87–0.98, $P = 0.006$).

Sensitivity analyses were then conducted substituting the psychotic experiences outcome with the rarer clinician-verified psychotic symptoms at age 18. None of the protective factors were found to be significantly associated with a reduced likelihood of psychotic symptoms being reported at age 18 when controlling for potential confounders among the adolescents whose mothers had psychosis (**Table 4**). However, the effect sizes were similar to those for psychotic experiences, and there was a non-significant trend for greater neighborhood social cohesion to be associated with a reduced likelihood of adolescent psychotic symptoms (OR = 0.62, 95% CI 0.22–1.69, $P = 0.346$).

Are Protective Factors Only Associated With an Absence of Age-18 Psychotic Experiences Among Adolescents Whose Mother has Psychosis?

There were no significant interactions between each of the protective factors and maternal psychosis status in relation to an absence of age-18 psychotic experiences in the full sample (**Table 2**). Indeed, higher levels of perceived social support were also associated with a reduced likelihood of age-18 psychotic experiences in children of mothers *without* psychosis (OR = 0.91, 95% CI 0.89–0.94, $P < 0.001$).

DISCUSSION

Summary of Findings

Higher IQ and greater neighborhood social cohesion were found to be independently protective against the development of age-12 psychotic symptoms among children of mothers with a psychosis-spectrum disorder or psychotic symptoms. At age 18, higher levels of perceived social support were found to be independently protective of psychotic experiences among adolescents of mothers with a psychosis-spectrum disorder or psychotic symptoms. There was a non-significant trend for neighborhood social cohesion to be protective for adolescent psychotic experiences. As none of the interactions between protective factors and maternal psychosis status investigated were found to be significant, this indicates that factors were not specifically protective against psychotic phenomena amongst these high-risk children. Indeed, all of the factors

TABLE 3 | Associations between putative protective factors and absence of age-18 psychotic experiences among children of mothers with a diagnosis or symptoms of psychosis.

Putative protective factor	Psychotic experiences absent $N = 238$	Psychotic experiences present $N = 144$	Adjusted OR[a]	95% CI	*P*
	M (SD)	*M (SD)*			
Physical activity	2.71 (1.09)	2.72 (1.09)	1.02	0.83–1.25	0.871
Neighborhood social cohesion	2.24 (0.52)	2.09 (0.50)	0.67	0.42–1.08	0.101
Social support	21.02 (4.55)	18.63 (4.99)	0.92	0.87–0.98	0.006

CI, confidence interval; OR, odds ratio; SD, standard deviation.

[a] Adjusted for gender, family socioeconomic status, age-12 psychotic symptoms, and other age-12 mental health problems. All analyses account for the non-independence of twin observations.

TABLE 4 | Associations between putative protective factors and an absence of age-18 psychotic symptoms among adolescents whose mothers had a psychosis-spectrum disorder or psychotic symptoms.

Putative protective factor	Psychotic symptoms absent $N = 360$	Psychotic symptoms present $N = 22$	Adjusted OR[a]	95% CI	*P*
	M (SD)	*M (SD)*			
Physical activity	2.71 (1.08)	2.68 (1.25)	1.00	0.64–1.58	0.991
Neighborhood social cohesion	2.19 (0.50)	2.00 (0.68)	0.62	0.22–1.69	0.346
Social support	20.16 (4.88)	19.36 (4.38)	0.99	0.93–1.07	0.882

CI, confidence interval; M, mean; OR, odds ratio; SD, standard deviation.

[a] Adjusted for gender, family socioeconomic status, age-12 psychotic symptoms, and other age-12 mental health problems. All analyses account for the non-independence of twin observations.

were also found to be protective amongst children and adolescents whose mothers had not experienced psychotic phenomena.

Comparison to Previous Research

Our results are consistent with previous research on the E-Risk study which reported that higher IQ, a more positive atmosphere at home, and greater neighborhood social cohesion were associated with a reduced likelihood of childhood psychotic symptoms in the whole sample and not just in high-risk children exposed to multiple forms of victimization (15). Similarly, previous studies show that possessing a higher IQ is associated with a reduced likelihood of developing psychotic symptoms (47, 48); while lower childhood IQ is associated with a range of mental health problems, including an increased risk of developing schizophrenia, depression and anxiety as an adult (49). Our findings also suggest that high levels of neighborhood social cohesion are independently protective in children with mothers who have either symptoms or a diagnosis of psychosis, and in the whole population. This result is consistent with previous research showing that low levels of social cohesion are associated with greater odds of developing psychotic symptoms (50–52). Additionally, our findings highlight the protective effect of perceived social support in adolescence which is consistent with previous research in E-Risk showing that social support was associated with a reduced likelihood of psychotic experiences among high-risk adolescents who had been poly-victimized (16). A study of clinical-level psychosis also demonstrated the protective nature of social support among high-risk individuals (53).

Strengths and Limitations

Strengths of the study include our novel analysis that examines protective factors for children and adolescents who are at high risk of psychosis by having a mother with a psychotic disorder or psychotic symptoms. Furthermore, our participants were taken from a large, nationally-representative longitudinal cohort study that measured psychotic phenomena at two time-points and enabled us to control for a range of potentially confounding factors. Several limitations warrant consideration. The sample size was relatively small when focusing on children of mothers with psychosis and so we may have lacked statistical power to detect significant effects, especially for the interaction analysis. Moreover, it was not possible to explore protective effects in children who mothers had a diagnosis of psychosis-spectrum disorder separately to those whose mothers had psychotic symptoms due to the small number of children in the former group ($N = 58$). The sample was composed of twins and so we cannot be certain if the findings can be generalized to single children, although the prevalence of childhood and adolescent psychotic phenomena in this study are similar to rates found in singleton samples (8, 25, 27). We limited our analyses to factors that have previously been found to be protective for psychotic phenomena in this cohort and it is possible that a range of other multi-level factors may also be protective. For instance, self-esteem (54), attachment style (55), and positive behavioral support (56). These require further investigation ideally in even larger population-based cohorts. In adolescence several of the measures were assessed at the same time-point and therefore it is not possible to ascertain the direction of the associations found. We did however control for earlier psychopathology, but it would be useful in future to utilize prospective measures, ideally obtained from different informants. Additionally, psychotic phenomena in childhood and adolescence have been associated not only with later development of schizophrenia but also with other mental health problems (10, 12), and thus the findings cannot specifically be generalized to indicate protection against clinically-relevant psychosis. Finally, our analyses focused on psychotic phenomena assessed only at the ages of 12 and 18 so we do not know if the factors analyzed would be protective if symptoms were to develop at other ages in childhood or adolescence, or in adulthood.

Clinical Applications and Future Research

The findings of this study have the potential to inform the focus of interventions to prevent the emergence of early psychotic phenomena and thus ultimately improve the outcomes of high-risk children and adolescents. Intellectual ability, neighborhood social cohesion, and perceptions of social support appear to be key target areas for early intervention. It is possible that possessing a higher IQ may facilitate problem-solving skills, coping strategies in adverse situations, and better self-regulation of emotions, particularly in relation to having a parent with psychosis. Cognitive behavioral therapies are being developed for young people that aim to target and develop skills such as reasoning and emotional coping, as a way of increasing resilience (57). Cognitive remediation therapy is another approach that may offer a way to alleviate cognitive difficulties in young people (58). Additionally, neighborhood support interventions to improve parenting have been demonstrated in a study which found high levels of neighborhood social cohesion were associated with reduced instances of child neglect (59). Such an outcome could occur through alleviating the burden that comes with childcare for mothers with psychosis as neighbors meet the physical and emotional needs of both child and parent. Trust between neighbors may also mean that children approach them for help when they are feeling distressed, providing further adaptive ways of coping (60). Research suggests that when people with early psychosis perceive greater social support, they appear to be more likely to cope with day-to-day stressors (61). Higher levels of social support have been found to correlate with lower levels of positive symptoms and fewer hospitalizations in people with first episode psychosis (62). Therefore, if the current findings are replicated in other cohorts, it would be helpful to investigate whether such interventions might prevent the development of psychotic phenomena in children and adolescents.

CONCLUSIONS

Higher IQ and neighborhood social cohesion were found to be independently associated with a reduced likelihood of age-12 psychotic symptoms among children of mothers with symptoms of psychosis or a psychosis-spectrum diagnosis. Similarly, higher levels of perceived social support were found to be independently protective against adolescent psychotic experiences in this high-risk group. In terms of clinical implications, if replicated, the findings suggest that interventions should be aimed at cultivating these factors for children and adolescents who are considered at-risk by virtue of having a mother with psychosis. All of these factors were also found to be associated with a reduced likelihood of having psychotic phenomena among children whose mother had not experienced psychosis, which suggests that preventative interventions could also be targeted at the general population regardless of risk. These preventative interventions should improve the cohesiveness of a child's community environment, increase their perceptions of social support, and bolster their cognitive functioning. Replication of these findings is required in larger populations, amongst single children, in order to ascertain generalizability and subsequently inform the development and testing of interventions to prevent the emergence of early psychotic phenomena. Ultimately, such preventive strategies may reduce the incidence of psychosis and other mental health problems in adulthood.

AUTHOR CONTRIBUTIONS

HF designed the project and oversaw the analyses and write-up of the manuscript. RB-N, MA, and SR conducted the analyses and wrote the first draft of the manuscript. LA oversaw data collection for the E-Risk Study. All authors critically reviewed and amended the content of the manuscript and approved the final version.

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REVIEW
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Fetal and Infant Outcomes in the Offspring of Parents With Perinatal Mental Disorders: Earliest Influences

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Mental illness is highly prevalent and runs in families. Mental disorders are considered to enhance the risk for the development of psychopathology in the offspring. This heightened risk is related to the separate and joint effects of inherited genetic vulnerabilities for psychopathology and environmental influences. The early years of life are suggested to be a key developmental phase in the intergenerational psychopathology transmission. Available evidence supports the idea that early exposure to parental psychopathology, during the pregnancy and first postpartum year, may be related to child psychological functioning beyond the postpartum period, up to adulthood years. This not only highlights the importance of intervening early to break the chain of intergenerational transmission of psychopathology but also raises the question of whether early interventions targeting parental mental disorders in this period may alleviate these prolonged adverse effects in the infant offspring. The current article focuses on the specific risk of psychopathology conveyed from mentally ill parents to the offspring during the pregnancy and first postpartum year. We first present a summary of the available evidence on the associations of parental perinatal mental illness with infant psychological outcomes at the behavioral, biological, and neurophysiological levels. Next, we address the effects of early interventions and discuss whether these may mitigate the early intergenerational transmission of risk for psychopathology. The summarized evidence supports the idea that psychopathology-related changes in parents' behavior and physiology in the perinatal period are related to behavioral, biological, and neurophysiological correlates of infant psychological functioning in this period. These alterations may constitute risk for later development of child and/or adult forms of psychopathology and thus for intergenerational transmission. Targeting psychopathology or mother-infant interactions in isolation in the postnatal period may not be sufficient to improve outcomes, whereas interventions targeting both maternal psychopathology and mother-infant interactions seem promising in alleviating the risk of early transmission.

Keywords: parents, parental mental illness, prevention, intervention, infancy, pregnancy

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INTRODUCTION

The transition to parenthood is a major life event that brings profound and lasting changes in new parents' relationships and personal identities as well as in the structure and organization of daily life. Becoming parents can be experienced as a highly rewarding but also a highly demanding task (1). The responsibilities of parenthood during the first year where infants fully depend on the caregivers can be stressful especially for parents with (predispositions for) psychopathology. This is why early parenthood is considered to be a period of vulnerability for the new onset and/or relapse of psychopathology in parents.

Among different types of psychopathology that manifest perinatally, the highest incidence rates have been reported for depression. The prevalences of pregnancy and postpartum depression range between 13% (2, 3) and 25% for mothers (4) and between 8.4% (5) and 10% for fathers (4). Anxiety disorders are also highly prevalent and commonly manifest comorbid with depression (6, 7), with incidence rates between 10% and 18% for mothers (8–10) and 5% to 10% for fathers (11, 12) during the perinatal period. Although relatively less prevalent, psychosis (13) and birth-related posttraumatic stress disorder (14) may specifically manifest following birth. Earlier research on perinatal psychopathology has almost exclusively focused on the most prevalent (i.e., depression) and the most severe (i.e., psychosis) forms of psychopathology in mothers (15, 16), whereas the presence of other mental disorders in this period have only recently been acknowledged (4, 17–19). Moreover, fathers have only recently been incorporated into the studies of perinatal mental illness. Psychopathology often co-occurs in new mothers and fathers, reflecting the influences of assortive mating (20) and the effects of living with a partner with a mental illness. The presence of psychopathology in both parents may multiply the risk of transmitting mental illness to offspring (4, 12, 21, 22). Hence, a better understanding of paternal influences, alongside and in interaction with maternal influences is of paramount importance.

The variability in the prevalence estimates across studies of perinatal mental illness in parents is partly explained by other risk factors, for example, socioeconomic disadvantages, unplanned pregnancies, low empathy, and social support from the partner and/or environment (23, 24). Furthermore, the link between parental mental illness and offspring psychopathology may mediate the effect of other disadvantages that are known to be intergenerationally transmitted, such as childhood emotional abuse and neglect in parents (25). Childhood maltreatment constitutes a lifelong risk for depression (26, 27) that may specifically manifest during transition to parenthood (28–30). Depression in parents with these adverse childhood experiences increases the risk of child maltreatment, and infants' postnatal exposure to maternal depression and maltreatment, in turn, multiplies the risk of psychopathology in the offspring.

There is substantial continuity in perinatal psychopathology (31–33), the strongest risk factor for psychopathology during the postnatal period is prenatal psychopathology. Estimates are that over 50% of the mental disorders reported in the postnatal period are relapses of prenatal psychopathology (2, 19). Despite a clear accumulation of risk on parents with earlier mental disorder, psychopathology in new parents goes undetected almost in half of the cases (34, 35). Undetected and untreated psychopathology in this period can take a chronic form, especially in case of a previous history of mental illness. The impact on the child of chronic and recurrent psychopathology in parents, extending beyond the prepartum and postpartum period, would be more profound and present a more pronounced risk for intergenerational transmission of psychopathology (36, 37).

Along with the studies focusing on the prevalence of mental illness during the pregnancy and postnatal period in community samples, a related line of research focuses on the needs and experiences of individuals with chronic and severe mental disorders (such as psychotic disorders) in the reproductive age (38, 39). A meta-synthesis of the qualitative evidence on the early experiences of mothers with severe mental illness reveals several challenges on the way to parenthood (40). At the core, these seem to result from the inherent conflict between the desire to be a good mother as defined by society and the limitations coming from living with a severe mental illness. Mothers experience guilt over their maternal abilities and over the risk of transmitting mental illness to their child. Moreover, the stigma of mental illness seems to be enhanced in the case of motherhood, making mothers less likely to seek help for the challenges they encounter and more likely to end up feeling isolated in this period (40). Early experiences of parenthood in men with chronic or severe mental disorders still remain to be incorporated into this line of research.

Taken together, available evidence on perinatal psychopathology and on the experiences of motherhood in women with severe mental disorders clearly illustrates that the transition to parenthood is a vulnerable phase on the side of parents. The vulnerability on the side of infants, in turn, is related to the tremendous changes and fast-paced development that takes place in the infant brain in this period (41, 42). These changes are highly dependent on infants' environmental experiences. Early experiences have the power to impact on the ongoing brain development either by altering or by moderating the developing function or structure of the infant brain (43). This sensitivity to environmental input by newborns and new parents explains why early environmental adversity including parental psychopathology may have an especially pronounced impact on infants' development in the early years of life (44, 45). For example, prenatal exposure to parental stress in the context of depression and anxiety is linked with changes in the development of infant hypothalamic–pituitary–adrenal (HPA) axis (46, 47), and postnatal exposure to psychopathology is suggested to influence the development of the key emotional brain systems for adult emotion processing, which become functional at around the first year of life (48–50).

Studies on the relationship between mental illness in parents and psychological functioning in the offspring have been categorized broadly into a micro versus macro perspective (44). Within the context of the perinatal period, the micro perspective focuses on the immediate associations of parental prenatal and/or postnatal mental illness with infant development, with a specific focus on aspects of early psychological functioning that may play a role in later psychopathology. The macro perspective, in turn, focuses on the longitudinal measurement of psychopathology in the offspring of parents with perinatal mental disorder over time intervals that extend from infancy up till adulthood.

Available evidence from the macro perspective reveals that parental psychopathology in the perinatal period may be related to child functioning beyond early years. At least in some cases, this link holds after taking into account later psychopathology in parents. This would reflect the specific influence of both genetically inherited dispositions for psychopathology and early environmental influences related to being exposed to a parent with mental illness in utero and in early life. To illustrate with the most studied mental disorder, i.e., maternal depression, studies reveal a significant link between exposure to maternal depression during pregnancy and the first postpartum year, and psychological functioning in the offspring from infancy to adulthood years. For example, infants of mothers with prenatal depression show more internalizing and externalizing problems at 1 year of age (51). Children of mothers with postnatal depression show more behavioral problems at the age of 2 (52), and of 5 and beyond (53, 54), along with a higher (up to fourfold to fivefold) risk of mental disorders such as depression and anxiety at 11 (55), 13 (56), and 16 years of age (57). There is also some evidence revealing similar effects of fathers' depression (58, 59), and parents' anxiety disorders in this period on child outcomes (60–62). Other studies have revealed more modest estimates of this link and have highlighted the importance of incorporating the chronicity of parental mental illness and other risk factors into this line of research (55, 63–67). Thus, further research is needed before we can reach firm conclusions about distinct associations of parental mental disorder at the perinatal period with later psychopathology in the offspring, whereas the evidence accrued so far from the macro perspective points to a link between offsprings' early exposure to parental psychopathology and later development of psychopathology. This highlights the importance of intervening early to break the chain of intergenerational transmission of psychopathology. As suggested by the antenatal investment hypothesis, the earlier the interventions are, the higher the returns would be in terms of economic and social benefits (68).

The findings from the macro perspective illustrate the need to observe early processes that are potential precursors to psychopathology in the offspring of mentally ill parents over the course of development from a micro perspective. The aims of this current review focusing on the immediate infant psychological outcomes from the micro perspective are twofold. The first is to gain insight in the effects of parental perinatal mental illness on early functioning by providing an overview of the associations of parental mental illness with infant psychological outcomes at the behavioral (see the section Behavioral Pathways: The Relationship Between Parental Perinatal Mental Disorder and Early Indices of Infant Psychobehavioral Functioning), biological (see the section Biological Pathways: The Links Between Parental Prenatal Mental Disorder and Early Indices of Infant Psychobiological Functioning), and neurophysiological levels (see the section Neurophysiological Pathways: The Links Between Parental Perinatal Mental Disorder and Early Neurophysiological Indices of Infant Psychological Functioning). The second aim is to answer the question of whether early interventions may mitigate the early intergenerational transmission of risk for psychopathology (see the section Effect of Early Interventions on Parent and Infant Outcomes).

PARENTAL PERINATAL MENTAL DISORDER AND INFANT OUTCOMES

Behavioral Pathways: The Relationship Between Parental Perinatal Mental Disorder and Early Indices of Infant Psychobehavioral Functioning

Infants' socio-emotional development is dynamically shaped throughout the first year as a result of their exposure to emotional expressions in everyday interactions. Indices of psycho-behavioral functioning at this period therefore focus on infants' interactive behavior with their caregiver. Mental illness in parents in the first postnatal year seems to alter parents' behavior in terms of affect expressions, attention, and sensitivity during these early interactions.

Parental Mental Illness and Parents' Behavior and Affect in Early Interactions

Psychopathology in parents may interfere with parents' experience and perceptions of their infant and alter parents' behavior in everyday interactions with their child. Depressed and anxious mothers were observed to be less responsive and/or less sensitive to child signals than mothers without depression or anxiety during early interactions (69–72). Depressed mothers also display more neutral and negative, and less positive affect during their interactions with their infant (73). Moreover, evidence suggests that depression in parents is related to suboptimal amounts of stimulation in everyday activities; for example, depressed parents less often read, sing to, or play with their infants (72). Differently from depressed parents, anxious parents do not differ from reference parents in their positive or negative facial expressions during early interactions (74). Anxious parents, in turn, were reported to display "exaggerated behavior" which is defined by high intensity and frequency of gaze, facial expressions, and vocalizations that are inappropriate with regard to timing and content (75). Moreover, parents with diagnoses of social anxiety were found to show more anxious behavior during their interactions with a stranger in the presence of their infants (76, 77), while parents with panic disorder reported expressing more anger to their infants in disciplinary contexts (78).

The differences in parents' emotional expressions and sensitivity are at least partly explained by psychopathology-related changes in parents' perceptions of their child: For example, parents with depression were found to perceive their child as more negative (79) and to be less likely to detect happy facial expressions of their infants than parents without depression (80). Concerning parenting, depressed mothers' behavior to their infant was classified as intrusive and overcontrolling on one end and withdrawn and understimulating on the other end of the continuum (81, 82). Withdrawn-depressed parents with depression were described to be less engaged and less tuned-in to their child during everyday interactions. Intrusive-depressed parents, in turn, seem to exert more control during play and intervene more frequently with their child's exploration of novel stimuli (82). The withdrawn-depressed parenting style has been linked with an underresponsive physiological profile that is characterized by lower dopamine levels and higher right-frontal EEG activity than the intrusive-depressed style (83–85). These differences were proposed to reflect the behavioral inhibition (BI) and activation systems (83). On a parallel vein, the history of maltreatment in parents seems to indirectly contribute to nonoptimal patterns of parenting, which manifests as more negative and intrusive, as well as harsher parenting practices and less parental emotional availability (86–90). Thus, parents' earlier negative experiences may at least partially explain the observed relationship between parents' depression and parents' negative perceptions of their child, and parenting practices (91).

Earlier evidence has also revealed a relationship between generalized anxiety symptoms and a more intrusive parenting style in parents with infants, along with less challenging parenting (92). Decreased levels of challenging parenting in anxious parents were proposed to be related to anxious parents' reduced ability to encourage their child's approach/exploration of potentially unsafe situations and to the development of child anxiety (93, 94). Findings from few studies that investigated parental behavior in early parent-infant interactions in parents with more severe mental disorders such as schizophrenia revealed that psychopathology-related alterations in mothers' early interactive behavior are especially pervasive in the case of severe mental illness. For example, mothers with schizophrenia were found to be less sensitive, less responsive, and more withdrawn to their infant as compared to parents with affective disorders (95, 96). The effect of these psychopathology-related alterations in parents' experience, perception, and responses to their child is suggested to be especially pronounced in the first postnatal year (48, 97).

Parental Mental Illness and Infant Expression and Regulation of Emotions in Early Face-to-Face Interactions

Psychopathology-related changes in their behavior and affect in early interactions may hamper parents' ability to provide the optimal affective environment for infants' emotional development. Theories of early socio-emotional development assign an important role to parents' emotional expressions and regulation of emotions, as well as to affective synchrony (98, 99). Infants were shown to be highly sensitive to parental affective input at the first postnatal year: Studies in community samples reveal that they tune in to the subtle differences between their mothers' and fathers' expressions of affect in these interactions (100). Although infants have some primitive abilities to regulate negative arousal such as looking away or thumb sucking, these are highly reflexive and limited in effectiveness (101, 102). For the rest, infants highly rely on the assistance of their parents for regulating emotional experiences in negatively arousing situations. Co-regulation of infants' emotional states in early dyadic experiences was suggested to lay the ground for the development of more voluntary emotion regulation strategies that emerge later in the first year (103).

Just like their parents, infants of depressed parents were shown to display more neutral and negative, and less positive affect than infants of reference parents during their interactions (73, 74, 104, 105) and to implement less mature emotion regulation strategies than infants of reference parents (106). Moreover, negative interactive style of depressed parents was suggested to trigger avoidance as an emotion regulation strategy: Children seem to use turning and gazing away from the mother as a strategy to regulate negative arousal possibly resulting from depressed parents' limited sensitivity and responsivity (107). In line with this, it was found that infants of depressed parents use gaze aversion more often during their face-to-face interactions with their parents (108). Although avoidance can be seen as an adaptive strategy in response to parental depression as it would reduce infants' exposure to parents' negative affect, it may be less adaptive in other situations where it may restrict child's exploration and new learning opportunities. On a parallel vein, it was suggested that due to their flat affect, limited responsibility and availability in everyday interactions, infants are less likely to actively seek input from depressed parents in ambiguous situations (109, 110).

Infants of anxious parents, in turn, more often display positive or negative expressions as compared to infants of reference parents in their face-to-face interactions with the parent (73, 111). The evidence also reveals that infants of anxious parents may express less negative affect as compared to infants of reference parents in challenging situations like meeting a stranger (75) but that they become anxious if they are first exposed to parental anxious displays before confronting the strangers (76, 77). In contrast, emotion regulation strategies of the infants of anxious parents do not seem to differ from infants of reference parents (106–108). In an earlier review on the links between exposure to parental depression and anxiety in the first postnatal year and child expressions of affect, it was suggested that infants' displays of affect in everyday interactions in the case of parental depression and anxiety may be mirroring their parents (105): Infants who are repeatedly exposed to parents' flat and negative affect in early face-to-face interactions may show a depressed interaction style characterized by more flat and more negative expressions. Similarly, infants exposed to parents' anxious behavior in specific anxiety-provoking situations seem to show an anxious response characterized by avoidant tendencies in these situations as a result of modeling (76). Likewise, impairments in the parent-child early dyadic regulation of affect and the resulting difficulties in emotion regulation may constitute vulnerability for the development of psychopathology in children, especially in the presence of other vulnerabilities such as insecure attachment and difficult temperament.

Parental Mental Illness and Infant Attachment

According to attachment theory, neonates are biologically programmed to form a strong bond to their primary caregivers to ensure their survival (111). Parents' ability to provide a timely and appropriate response to the infants' dynamically changing attention and affective signals in everyday interactions at this period is of paramount importance for establishing a secure parent-child attachment in the early years of life (112, 113). Along with responsivity and sensitivity, parents' mutuality and synchrony and their positive and supportive attitude during early interactions seem to be factors supporting the establishment of a secure attachment (111). It was suggested that early attachment in infants' first relationships with the caregivers shapes one's internal representations of relating to others. Attachment patterns show moderate stability from infancy to early adulthood years (114). Thus, although there is some room for change, infants' attachment security in their early relationships with the parent provides the ground for later attachment behavior in personal relationships.

Infant attachment is commonly measured using the experimental paradigm the Strange Situation, which is a stressful situation involving parental separation and reunion, as well as stranger anxiety (115). The Strange Situation consists of a series of phases during which the parent leaves the child (alone or with a stranger) for a few minutes (parental separation) before she comes back and reunites with the infant (parental reunion). Several dimensions of infants' behavior are observed during the reunion phase for measuring the attachment to caregiver, including infants' proximity/comfort seeking versus avoidance, resistance against mothers' attempt to contact and comfort them, and their emotional expressions. Securely attached infants express distress in response to maternal separation and positively embrace the reunion, while infants with resistant attachment experience stronger levels of stress in response to separation and show conflictual reactions to parental reunion, characterized by an approach to the parent for comfort, along with a resistance against it. In turn, infants with an avoidant attachment style do not seem to be distressed by maternal separation and/or interested to engage with the mother during the reunion.

A third pattern of insecure attachment, so-called disorganized/disoriented attachment, was later defined by Main and Solomon (116). Children with disorganized attachment overtly show disoriented/disorganized reactions to maternal separation and reunion episodes in the Strange Situation. These children show not only contradictory behavior (such as approaching the parent while averting gaze) and apprehension to the caregiver but also uncommon and out-of-context behavior such as freezing, sudden change in affect, fearful reactions to caregiver, and/or incomplete movements or atypical postures (117). Infants with disorganized attachment were suggested to seek contact with the primary caregiver, without a consistent or coherent strategy to establish that contact (116). It was suggested that at the core of the disorganized attachment style is a difficulty to trust and rely on parents for comfort and soothing. This may potentially be a result of repeated exposure to insensitive or disruptive parenting behavior (including frightening or frightened parental reactions) that is ineffective at meeting infants' needs for proximity and comfort in stressful situations (118).

Earlier evidence has revealed that these insensitive and disruptive parenting behaviors may occur as a result of unresolved traumatic experiences including parents' history of childhood maltreatment. In fact, more than half of the parents of infants with disorganized attachment were shown to have such unresolved trauma (119). In the case of childhood maltreatment, the links between earlier maternal trauma and security of parent-child attachment seem to be mediated by postnatal maternal depression (120). Infants' exposure to parents' postnatal depression and stress during early interactions seems to be linked to a lower likelihood of a secure attachment, along with a higher risk for insecure attachment (121–123). Moreover, higher rates of disorganized attachment were reported in the infants of mothers with borderline personality disorder (124). It is important to note that the association between parental mental illness and child attachment is rather modest in size and was not replicated in some of the more recent studies [for example, the link between parental psychopathology and disorganized attachment was not significant in the case of depression (125, 126), and in the case of anxiety (127, 128)]. Note, however, that most of the presented findings from these earlier studies are from community samples, whereas the association between parental mental illness and disorganized attachment would be especially pronounced in clinical samples of parents [for a more elaborate discussion, see Ref. (129)]. Although limited by similar methodological issues, a significant relationship between early insecure attachment and the development of internalizing and externalizing psychopathology from early childhood to adulthood years was reported in earlier studies (130, 131). To summarize, there is preliminary support for the idea that psychopathology-related alterations in parents' behavior may be related to higher levels of insecure attachment in the offspring, which constitutes a vulnerability for intergenerational transmission of psychopathology. Further evidence from clinical samples of parents with infants is needed to reach firm conclusions about this link between parental psychopathology and insecure attachment.

Section Summary and Conclusions

Taken together, the evidence summarized in this section reveals a significant link between parental mental illness and parents' parenting behaviors, and their expression and regulation of affect during early interactions. These psychopathology-related alterations may limit parents' emotional availability and their ability to respond to their infant in a sensitive manner, rendering the early socio-emotional environment suboptimal for the establishment of a secure attachment bond, as well as for infants' emotional development. Available evidence from infants of parents with anxiety and depression reveal that infants' behavior during these early interactions, defined by high levels of affective negativity and avoidance, along with less mature emotion regulation skills, is reminiscent of the interaction and responses characterizing parents' psychopathology. On the behavioral level, it seems that parents may already pass on negative interaction patterns characterizing affective psychopathology during these early interactions.

Long-term implications of the early suboptimal environment linked to perinatal parental mental health problems include a negative-insecure relational pattern that may be internalized and generalized to the offspring's new relationships with teachers, peers, and romantic partners. The offspring may additionally face the risk of repeating early suboptimal relational experiences by choosing mentors, friends, and partners who behave in similar ways as the parent with psychopathology. Finally, the offspring of parents with perinatal mental disorders may adopt less functional emotional regulation strategies such as self-destructive behaviors, aggression, depression, or avoidance and may experience more difficulty regulating their negative emotions.

Biological Pathways: The Links Between Parental Prenatal Mental Disorder and Early Indices of Infant Psychobiological Functioning

The first environment that a human being experiences is inside the mother's womb. Research in the last decades has shown that this environment can have a great impact on the development of the embryo and fetus (132–135). The fetal programming hypothesis (136, 137) postulates that the environment of the developing fetus affects its development to enhance survival and prepares the infant for the environment to expect after birth. In the context of parental mental health, the mental state of the mother during pregnancy may influence the prenatal as well as the postnatal environment of the unborn child, thereby affecting its development. In this section, we discuss some of the possible mechanisms by which prenatal parental mental health may influence the development of the unborn child, with a focus on infant psychobiological development. We will mostly focus on maternal mental health during pregnancy with the womb as the first (biological) environment, even though fathers may directly and indirectly influence the environment of mother, and thereby her offspring. Furthermore, as mental illnesses co-occur with high levels of stress, and most research in this field is conducted on prenatal depression and anxiety, this section will focus on consequences of (traumatic) stress, depression, and anxiety during the prenatal period.

Human studies have shown that stress during pregnancy has widespread associations with offspring cognitive, emotional, and health outcomes (132–135). Studies in this area differentiate between different types of stress. That is, some studies investigate the impact of traumatic stressors that have happened during the prenatal period and that can be relatively objectively identified, such as having been exposed to the holocaust, the 9/11 attacks (138, 139), and natural disasters (140). Alternatively, some studies investigate the levels of stress that are subjectively experienced during pregnancy, either due to impactful events as mentioned above (141), due to daily life hassles, or due to the pregnancy itself (142, 143). Yet other studies examine more trait- or disorder-related experiences of stress, anxiety and depression (144). In this regard, studies in women that have developed or suffered from posttraumatic stress disorder or depression during the prenatal period often also focus on changes in stress physiology that are associated with these disorders in mothers (138, 145). Irrespective of the type of stress, most of the studies on prenatal stress indicate worse developmental outcomes with problems in the cognitive domain, emotional reactivity, and worse physical health outcomes. In this section we will discuss possible routes *via* which this psychobiological functioning of the infant can be affected by prenatal stress.

As human studies lack the possibility of randomly assigning stress during pregnancy to assess its impact, it is bound by the constraints of observational designs, and views differ on the origins of prenatal stress effects (137). However, studies that examine traumatic events that happened to a large group of people, such as a natural disaster, have the opportunity to more objectively compare women that have and have not suffered from these stressors. Animal studies on the other hand use experimental procedures, ranging from physical constraint to overcrowding, to induce prenatal stress (146). These studies are able to more directly examine causal effects of prenatal stress, independent of predisposing heritable characteristics or postnatal care, and give the opportunity to more precisely examine the potential underlying mechanisms by which prenatal stress may affect the prenatal environment of the fetus. Both human and animal studies comparing pregnancies with high levels of stress versus those with low levels of stress have given us insights in the psychobiological effects of prenatal stress and anxiety, some of which will be discussed next.

The Links of Parental Mental Illness to Infant Psychobiological Development

Recent studies show that prenatal stress and mental health problems in mothers are associated with differential brain development in children (147), although studies in young infants are still rare (148). Some first studies in infants show associations between maternal prenatal depression and amygdala microstructure and functional connectivity in early infancy (149–151) and between maternal prenatal stress and amygdala functional connectivity in preterm neonates (152). Maternal prenatal anxiety has also been found to associate with infant brain microstructures and hippocampal growth (150, 151). Studies in rats complement these studies by showing that these effects can have a causative origin. Indeed, using restraint stress procedures or corticosterone administration in rats has been shown to affect brain morphology and behavior (146, 152).

One line of reasoning is that many of the effects of prenatal stress, anxiety, and depression on infant functioning and brain development are related to changes in the development of the infant HPA axis (153). The HPA axis plays a role in biological stress regulation, where brain areas like the hippocampus and prefrontal cortex are key brain areas regulating these stress responses, and is implicated in cognitive and emotional functioning (154). Quite a few human and animal studies show dysregulations in the HPA axis in relation to prenatal stress (46, 47). Both hyporeactivity and hyperreactivity of the HPA axis have been found in response to prenatal stress, and the effects seem to depend on timing and the type of the stress during pregnancy, time and type of HPA axis measurements, and child sex. For example, we showed that maternal prenatal anxiety was associated with heightened cortisol reactivity to a bathing session at 2 weeks of age but decreased cortisol reactivity to a vaccination at 2 months of age (142), showing moderation by time and type of stress induction. Brennan et al. (155) revealed that maternal prenatal depression was associated with increased baseline infant cortisol levels, while comorbidity with anxiety disorder was related to higher infant cortisol reactivity, showing differential effects on infant outcomes dependent on maternal disorder-specific symptoms. There are furthermore indications that females may be more susceptible to the impact of prenatal stress on HPA axis regulation (46).

Overall, the literature suggests that the HPA axis may be a key player in the association between prenatal stress and developmental outcomes, but longitudinal human studies showing proof for this pathway are still limited (156). From an evolutionary perspective, and according to the fetal programming hypotheses, prenatal stress would prepare the offspring for a stressful, dangerous, or hostile environment to grow up in. Changes in infant HPA axis regulation would thereby prepare for this environment. However, in case the postnatal environment is different than may be expected based on the first experiences, this can lead to a so-called mismatch in environments (157), in which the prenatal developmental changes do not lead to higher changes of survival but may induce susceptibility to pathology (47). While fetal programming has become an important area of research (136), the underlying mechanisms implicated in fetal programming still remain to be fully elucidated, and at different stages during pregnancy different mechanisms may play a role.

A Potential Mechanism: Prenatal Stress Hormones

One area that has been studied extensively in the context of prenatal stress, anxiety, and depression is the influence of maternal stress hormones, most notably cortisol, on the developing fetus. Maternal cortisol levels can directly influence fetal cortisol levels *via* the placenta or *via* stimulation of the infant HPA axis by placental corticotropin-releasing hormones (158, 159). While the fetus is in principle protected from high maternal cortisol concentrations by the placental enzyme 11β-hydroxysteroid dehydrogenase-type 2 (11β-HSD2), this enzyme is found to be inhibited by prenatal anxiety (160), reducing its protection against maternal cortisol. Heightened levels of cortisol during fetal development may in turn affect infant HPA axis regulation and brain development (161, 162). Besides changes in stress hormones, maternal prenatal stress or mental health problems may affect the unborn child in several other ways, including changes in inflammatory and metabolic conditions of the intrauterine environment (163). These endocrinological changes may be dependent on lifestyle factors (e.g., exercise, sleep, and nutrition) that could be direct consequences of heightened levels of stress, anxiety, or depression in the mother (132).

While the prenatal environment may be affected in many ways by changes in maternal hormones, and immune and/or metabolic status, in recent years the focus has shifted to underlying epigenetic mechanisms that may ultimately explain changes in the development of the fetus (135, 163, 164). Epigenetics refers to modifications to the genome that have functional consequences for gene functionality, without changing nucleotide sequences (165). The most common studied epigenetic factor in human research is DNA methylation, which is sensitive to glucocorticoid signaling (166). Epigenetic changes due to cortisol provide a route by which the prenatal environment can impact fetal development, as epigenetic changes due to prenatal stress hormones can directly impact gene activity and functionality during development of the fetal brain and HPA axis (167, 168). Interestingly, not only maternal stress but also paternal prenatal stress has been studied in this context. While paternal stress may impact maternal stress levels *via* behavioral and social routes, it has been suggested that stress in males can also lead to epigenetic changes in the sperm that can be directly transmitted to the offspring (169).

As discussed above, prenatal stress, anxiety, and depression affect the intrauterine environment and thereby the development of the fetus. However, these factors do not act alone and may interact with, or even represent, underlying genetic characteristics. First of all, the effects of maternal stress and mood can interact with genetic susceptibility of the unborn child (170). For example, child brain-derived neurotrophic factor (BDNF) genotype was found to moderate effects of maternal prenatal anxiety on later child internalizing problem behavior (171), as well as on the child's epigenome and structures of the amygdala and the hippocampus (172). Secondly, an infant's genetic susceptibility to emotional or developmental problems will depend on the genes of the parents. In that regard, associations between maternal and/or paternal stress, anxiety, and depression and infant development may partly be due to inherited characteristics (173). As such, dysregulations in the HPA axis of children may very well be directly inherited from the mother, possibly confounding previously discussed associations with prenatal stress. Similarly, the emotional development of children may depend on parental mental health *via* genetic routes. An interesting study by Rice at al. (173) has tried to disentangle some of these effects by comparing children that were born *via in vitro* fertilization (IVF), who were genetically either related or unrelated to the mother. They showed that prenatal stress affected birth outcomes and antisocial behavior independent of mother-child genetic relatedness, indicating prenatal stress as an environmental factor. Likewise, maternal anxiety and depression related to offspring anxiety levels held independent of relatedness. However, associations with symptoms of attention deficit hyperactivity disorder were only present in related pairs and hence implies underlying heritable factors (173). Such clever designs can give a more clear understanding of cause and effect when examining associations between prenatal or postnatal stress and infant outcomes.

So far, we have focused on mechanisms during the pregnancy. Obviously, prenatal stress may also be associated with changes in postnatal care, e.g., with regard to sensitive behavior or emotional availability, and hence affect infant development as well (132, 174); see the section Behavioral Pathways: The Relationship Between Parental Perinatal Mental Disorder and Early Indices of Infant Psychobehavioral Functioning. Furthermore, prenatal and postnatal mood disruptions in mothers can interact or have additive effects on child outcomes (137, 175, 176). In human studies, it is again hard to disentangle effects of the prenatal and postnatal environment, as each may have a different or continuous impact or reflect more underlying characteristics. Here as well, animal studies can guide in disentangling these environments by experimentally manipulating either prenatal or postnatal environment, and by cross-fostering studies (177).

Section Summary and Conclusions

In this section we show the importance of the first biological environment that the offspring experiences, i.e., the womb. Mothers' prenatal stress and mental health status will influence the amount and diversity of hormones and metabolites that permeate the placenta and can thereby directly impact the development of the infant brain and physiology. These changes may be long lasting due to epigenetic changes that can permanently alter the phenotypic expressions of the infant, including heightened stress sensitivity and changes in HPA axis regulation. The long-term implications of these early alterations in infant psychophysiologic and biological functioning may go beyond heightened stress sensitivity and subsequent risk for mental disorders (e.g., anxiety, depression) as it also alters immunity and the brain-gut axis underpinning risk for somatic disorders (e.g., autoimmune diseases) later in development. However, it is important to note that these underlying mechanistic explanations need translational research in animals, as observational designs in humans limit our abilities to draw conclusions regarding the causality of observed associations between changes in parental and offspring psychobiology.

Neurophysiological Pathways: The Links Between Parental Perinatal Mental Disorder and Early Neurophysiological Indices of Infant Psychological Functioning

An accumulating body of evidence illustrates that infants of mothers with mental illness are more likely to develop dysregulated behavior, lower levels of positive affect/behavior, and higher levels of externalizing and internalizing behavior (178, 179). From a developmental psychopathology perspective, child externalizing and internalizing behavior can be partly explained by individuals' inability to regulate their emotions appropriately (180). Two physiological and neural indices play an important role in individuals' emotion functioning. One is vagal tone, indexed by the respiratory sinus arrhythmia (RSA). Vagal activity is related to individuals' facial expressions and to the process of physiological regulation during social engagement (181, 182). The second neural index is related to the amygdala: An enlarged amygdala or heighted connectivity between amygdala and other brain structures is related to heightened negative emotionality and affective disorders (151, 183, 184). In this section of the review, the focus is on the links between maternal mental illness and child's physiological functioning as indexed by RSA and amygdala structure or amygdala connectivity.

Parental Mental Illness and Infant RSA

One of the underlying mechanisms explaining parent-to-offspring transmission of maternal depression and anxiety may be related to the activity in the parasympathetic system (178, 179). Recent evidence from experimental and correlational studies supports this idea (185–187). Activities in the parasympathetic system are usually indexed by vagal tone. The vagus nerve is part of the motor pathway that is connected to striated facial muscles that are responsible for social gaze, facial expression, and vocalization, supporting successful social engagement (182). RSA has been used to measure the functional output of the vagal pathway on the heart (190). It refers to the variability in heart rate that occurs at the frequency of spontaneous respiration. Higher baseline RSA is an index of flexible responding (191) and is linked to better self-regulation (192) and better sustained and focused attention (188, 189). However, higher baseline RSA is also found to be related to greater behavioral reactivity (193) and heightened frustration (192).

The prenatal period and the first year of life are critical periods for the maturation of the vagal system (182, 194), which is indexed by the number of myelinated vagal fibers. Without a working myelinated vagus, more rudimentary defensive strategies such as fight-flight mobilization, tantrum, and shutdown behavior will dominate rather than regulate social behaviors (182). The myelinated vagal fibers keep burgeoning in number, and the myelin thickness continues to increase from 24 weeks through adolescence; however, the greatest increase is observed from 30–32 weeks of gestational age to approximately 6–9 months postpartum (195, 196). Thus, maternal psychopathology [for example, maternal depression reflected in flat affect, unresponsiveness, and low sensitivity (197)] may exert a stronger effect during this stage than later in development.

Infants of mothers who experience prenatal or postnatal depression were shown to be more likely to exhibit lower baseline RSA as early as neonates (84, 198). Infants of mothers with postnatal depression also do not show the usual increase in RSA that is observed from 3 to 6 months in typical development (198). Similar findings were reported in infants of mothers with anxiety disorders (either during lifetime or during pregnancy (199, 200). Low baseline RSA poses several disadvantages for infants (181). Given its connection to the striated facial muscles, the nonoptimal vagal development may impede infants' ability to signal or express their emotions, which in turn may increase infants' risk of developing affective disorders (181, 201). Observational studies support this view such that newborns of depressed (versus nondepressed) mothers showed fewer facial expressions in response to happy and surprised facial expressions (202) (also see the section Behavioral Pathways: The Relationship Between Parental Perinatal Mental Disorder and Early Indices of Infant Psychobehavioral Functioning). Moreover, lower baseline RSA levels limit infants' ability to engage in physiological regulation (203). Taken together, evidence generally supports the idea that infants who have depressed and/or anxious mothers may have difficulty expressing emotions resulting from their nonoptimal development of RSA, and this may in turn impede their social engagement, enhancing the risk for later development of depression and anxiety.

Opposite to lower baseline RSA in infants that is generally seen as maladaptive (181), high baseline RSA is defined as a "biological sensitivity to context" factor (204, 205) such that infants with higher RSA are more susceptible to the environmental influences for better and for worse. This idea is supported by recent evidence that revealed that maternal depression and anxiety are linked to maladaptive infant outcomes (e.g., infant negativity, sleep problems, or disorganized attachment) only for infants who showed higher baseline RSA but not for infants who showed lower baseline RSA (206–208). Thus, in the context of parental mental illness, the finding that infants with higher baseline RSA demonstrate more maladaptive outcomes possibly indicates a misfit between infants' physiology and the level of stress in the environment. Further studies are needed to elucidate the effect of baseline RSA servicing as a "biological sensitivity to context" factor (205).

Maternal Mental Illness and Infant RSA Withdrawal

Differently from the Baseline RSA that is usually seen as an index of a stable resting "physiological state" (181, 203), a decrease in RSA or RSA withdrawal reflects individuals mobilizing resources in response to immediate environmental challenges, such as dealing with a frustrating or stressful situation. This process facilitates an increase in heart rate and allows individuals to shift from maintaining internal homeostasis to coping with external demands (201). After the stressor is over, individuals usually experience a recovery that manifests an increase in RSA (201). Consistent with the theory, the process of infants' RSA withdrawal is associated with concurrent behavioral regulation and recovery from distress (190, 209). A meta-analysis has revealed that children who were able to engage in RSA withdrawal during stressful situations had fewer externalizing, internalizing, and cognitive/academic problems; moreover, lower levels of RSA withdrawal were found in children who displayed clinically elevated behavior problems (210).

Young children have limited ability regulating their negative arousal, and the caregiver serves as an important external regulator for infants *via* physical contact and verbal confirmation (211). Parents who engage in sensitive and responsive parenting usually have infants engaging in optimal levels of RSA withdrawal and normative RSA recovery (212, 213). However, for parents who experience mood disorders, the dyadic coregulation process is likely to be disrupted considering that the mothers' fatigue and depressed mood may result in inability to respond to the infants' need in a timely and sensitive manner (104, 197, 214). Thus, infants lose the opportunities of learning to down-regulate their negative arousal, and they are more likely to develop physiological dysregulation in the long run (211). Empirical studies that considered multiple risk factors in mothers showed that infants in the high-risk group (characterized by mothers' current mental disorder, substance use, or two or more psychosocial risk factors) showed no recovery during the reunion episode of the Still-Face Paradigm suggesting a dysregulated physiological response in infants (187). In another study, no difference was reported in RSA changes between infants of mothers with depression and the control group (215). In contrast, infants whose mothers had bipolar disorder were shown to exhibit an increase in RSA during the stressor task compared to the control group in this study, indicating nonoptimal physiological regulation during a stressful task. To sum up, there is some indirect evidence (i.e., the effect of mood disorder is not teased out) that infants of mothers with mental illness, especially mood disorders, are more likely to develop physiological dysregulation (187, 215). However, more research is needed to uncover the direct association between parental mental illness and infant physiological regulation. Finally, note that no evidence is yet available on the links between paternal mental disorders and infants' vagal functioning. Considering that fathers' mental illness exerts its influence on the children either directly through parenting behaviors or indirectly through negatively affecting mothers' parenting behaviors (216–218), resulting in nonoptimal development in infants' physiological functioning, it is important to incorporate fathers into future studies on this line of research.

Maternal Mental Illness and Amygdala Activity in Infants

The amygdala, a critical brain region in the processing of threat, is susceptible to environmental adversity in early development (219). Mothers with prenatal depression are likely to experience multiple changes physiologically that may affect fetal development such as an increased cortisol production (220, 221). The amygdala is one of the areas rich in glucocorticoid receptors in the fetus' brain, which seems to be especially negatively affected by maternal cortisol levels (222). Increased amygdala activation in response to novelty or threat in children has been linked to higher negative emotionality (223). Furthermore, a larger amygdala in volume, strengthened amygdala connectivity, and greater right amygdala activation are all associated with an increased risk of developing affective disorders such as depression in children and adolescents (183, 184, 224, 225).

Evidence reveals prenatal depression may have a significant effect on the differences in the microstructure of the right amygdala in neonates after controlling for postnatal depression (151). More specifically, significantly lower anisotropy and axial diffusivity, which contribute to increased negative emotionality, were observed in neonates of prenatally depressed mothers (151). Furthermore, evidence supports the idea that maternal depression may also alter the amygdala connectivity in infants. Prenatal depression was shown to be linked to greater functional connectivity in the amygdala with the left temporal cortex and insula, as well as the bilateral anterior cingulated, medial orbitofrontal, and ventromedial prefrontal cortices in 6-month-old infants; these patterns are correlates of major depressive disorder in adolescents and adults (150). Therefore, the changes in the amygdala structure and amygdala connectivity may increase infants' vulnerability of developing affective disorders and may serve as another important mechanism through which prenatal mental illness, specifically depression, is transmitted to infants (151, 226).

Section Summary and Conclusions

Physiological and neural indices serve as underlying mechanisms that may be involved in the transmission from prenatal mental illness to infants' maladaptive functioning. Evidence from literature examining RSA and amygdala activity illustrates that infants of parents with mental illness are more likely to carry physiological risk factors such as lower baseline RSA, reduced RSA withdrawal, and heightened amygdala connectivity. In the long term, these early alterations in RSA and amygdala connectivity may, through mechanisms such as difficulties in emotional expressions, emotion regulation and threat sensitivity, may increase infants' vulnerability of developing mental disorders such as depression and anxiety disorders. Further research on moderating influences (e.g., children's resilience factors and parenting behavior) of the link between parental mental illness and infant physiological and neural functioning is needed before drawing conclusions on responsible mechanisms.

EFFECT OF EARLY INTERVENTIONS ON PARENT AND INFANT OUTCOMES

The findings summarized in earlier sections illustrate the potential value of early interventions targeting parents' psychopathology and related alterations in early parent-infant interactions in the prevention of intergenerational transmission. In light of the short-term and longer-term risks associated with parental perinatal psychopathology [e.g., Refs. (52, 56, 106, 227)] interventions for parents experiencing perinatal psychopathology have focused on infant as well as parent treatment outcomes.

Here, we provide an overview of the interventions for parents with a diagnosed psychiatric disorder [so not, for example, the interventions such as (228–231), where mothers were not diagnosed with psychiatric disorders and where the intervention began before 12 months [so not, for example, Ref. (232) or (233)].

Research into interventions for parents experiencing perinatal psychiatric disorders has predominantly focused on depression, with very few exceptions [for example, a trial for mothers with bulimic eating disorders (234), a trial for mothers with postpartum OCD (235), and a trial registered, but not yet reported, for mothers with anxiety disorders during pregnancy (236); for systematic reviews and meta-analyses, see for example Refs. (237, 238)]. We focus primarily on interventions examined in randomized controlled trials (RCTs), and then only briefly address the interventions examined using less robust designs.

We must emphasize that, to our knowledge, no intervention study has focused on paternal mental disorders and infant outcomes. For over a decade, research has addressed the risks posed by paternal psychopathology (59). It appears that risk pathways from paternal postnatal depression overlap with, but are not identical to, those of depressed mothers (239). Paternal anxiety disorder has received less attention, but, in infancy and toddlerhood, fathers' social anxiety appears to be as important as mothers' in predicting offspring anxiety (76, 240). So, while paternal psychopathology is important, evidence from trials addressing the effect of paternal interventions has yet to be reported.

Interventions for Maternal Mental Illness

Postnatal depression has been the most frequently studied postnatal psychiatric disorder with respect to interventions to address infant outcomes. This section provides an overview of progress in the field, moving from trials examining infant outcomes where maternal postnatal depression alone was the focus of treatment, to trials where mother-infant interactions have been the treatment targets, to having *both* maternal postnatal depression *and* mother-infant interaction as the treatment targets [for systematic reviews for broader considerations (237, 238, 241)].

Maternal Postnatal Depression as the Intervention Target

Two RCTs have examined infant outcomes following treatment of maternal postnatal depression alone (242, 243). The first trial (242, 244) examined the effect of three treatments (psychodynamic psychotherapy, cognitive behavior therapy, and nondirective counseling) versus routine primary care on maternal and offspring outcomes up to 5 years. Although all three treatments were associated with improved depression symptoms compared to routine primary care at the end of treatment (18 weeks postpartum), prevalence of maternal depression diagnosis was reduced only in mothers who received brief psychodynamic psychotherapy. At 5-year follow-up, compared to routine primary care, the treatments had led to no reduction in episodes of depression (244). Regarding offspring outcomes at the end of treatment, mothers in all treatment groups reported lower levels of problems in their relationships with their offspring compared to mothers in routine primary care. Mothers facing high social adversity and receiving nondirective counseling also reported more maternal sensitivity. However, none of the interventions was associated with effects on child attachment or cognitive development compared to the control group, and no effects were found at 5 years on measures of child emotional, behavioral, and cognitive development.

The second RCT (243) tested whether improved maternal mood led to improved child outcomes. Depressed mothers were randomly allocated to either interpersonal psychotherapy (IPT, n = 60) or to a waitlist control group (n = 60), and 56 nondepressed mothers served as control group for comparison. At the end of treatment (mean average, 9 months postpartum), compared to the waitlist control, IPT was superior only in the domain of parenting stress (although this remained higher than in the nondepressed group). At 18 months postpartum, compared to the offspring of nondepressed control mothers, offspring of mothers who received treatment had more behavior problems, lower attachment security, and more negative temperament. In summary, these early RCTs suggested that treatment of maternal postnatal depression alone was inadequate to ameliorate the risk posed to offspring by maternal postnatal depression.

Mother–Infant Relationship as the Intervention Target

In light of results from interventions focused on maternal postnatal depression alone, two RCTs (245, 246) examined the effects of interventions in the context of maternal postnatal depression where the intervention target was the mother-infant relationship, not maternal postnatal depression. First, Van Doesum and colleagues (245) examined the effects of 8 to 10 sessions of home-based video feedback treatment (VFT) (n = 35) and a control treatment of three 15-min telephone sessions offering practical parenting advice (n = 36) on infant attachment and maternal sensitivity. The study did not include treatment for depression. Regarding effects on mothers' behaviors, at the end of treatment and at 6 months follow-up, mothers in the VFT group were observed to be more sensitive and to provide more structure in their interactions with their infants compared to mothers in the control group. Regarding children's development, at the end of treatment, children of mothers who received VFT were observed to be more responsive to their mothers and more involved in interactions when compared to offspring of mothers in the control group. At the 6 month follow-up, prevalence of secure attachment status were higher for offspring of mothers who received VFT. These results must be considered in light of possible attention effects of the intervention (8 to 10 home visits) compared to the control group (three 15-min telephone calls). At 5-year follow-up (247), no main effects of treatment were found for mothers or offspring. However, where families experienced stressful life events, children in the VFT group had fewer mother-reported child externalizing problems than children in the control group. Thus, these results suggested that early, intensive intervention focused on the mother-infant relationship could alter infant development in key domains. Moreover, for those facing further risk in light of subsequent stressful life events, possible protective effects were reported against child externalizing problems.

Second, Horowitz and colleagues (246) reported an RCT with 136 mother-infant dyads, where mothers received an intervention called Communicating and Relating Effectively (CARE) designed to teach mothers to identify, and respond sensitively to, their infant's behavioral cues, or no treatment. All mothers were visited at home at 6 weeks, 3, 6, and 9 months postpartum for observational assessments, with the CARE group receiving additional visits at 2 and 4 months to receive the CARE intervention. Both groups improved on measures of maternal depression, mothers' behaviors, and mother-infant interactions, but there were no significant differences between groups. It is possible that any effects of the two sessions of the CARE intervention were confounded by the attention given to the control group (that is, four home-based observational visits). Further, the mean baseline score on the Edinburgh Postnatal Depression Scale (EPDS) was under 13 for both groups, suggesting that the depression was insufficiently severe to lead to adverse child outcomes. To summarize, the VFT treatment examined by Van Doesum and colleagues (245, 247) reported promising effects for infants and, at 5-year follow-up, protective effects for children who had experienced more stressful life events. Horowitz and colleagues (246) in contrast found no effect of their CARE program. While the interventions in these two trials both focused on helping depressed mothers identify and respond sensitively to their infants' cues, the different "doses" in the two studies, 10 sessions of VFT and two sessions of CARE, might account for the inconsistent results.

In summary, studies examining interventions with their target as *either* maternal depression (see the section Maternal Postnatal Depression as the Intervention Target) *or* the mother-infant relationship (see the section Mother–Infant Relationship as the Intervention Target) have yielded little evidence of short-term benefit to offspring development and almost no benefit at longer-term follow-up. Recent evidence points to the importance of the severity and the persistence of postnatal depression as moderators of risk for adverse childhood and adolescent development (227). In the intervention studies summarized above, the severity of maternal depression (for example, a mean score on the EPDS in the mild to moderate depression range) and the timing of interventions (being completed between 4.5 and 9 months postpartum) possibly limited these studies' ability to clarify the effects of intervention on infant development.

Maternal Postnatal Depression and Mother–Infant Relationship as the Intervention Targets

The first study to examine children's outcomes in the context of severe and persistent maternal postnatal depression, where the mother–infant relationship was a target while mothers also received an evidence-based treatment for depression, was reported by Stein and colleagues (248). In this RCT, 144 mothers were randomly allocated to receive, at home, either video feedback therapy (VFT, with the mother–infant relationship as its target; N = 72) or Progressive Muscle Relaxation (PMR, with stress management as its target; N = 72). Concurrently, all mothers received cognitive-behavioural therapy (CBT) for depression at home (10 sessions between 6 and 12 months postpartum, with two booster sessions in the second postnatal year). In particular, the study examined putative mediators of children's development in the context of postnatal depression, by attempting to use VFT to modify key maternal behaviors (sensitivity, warmth, and contingent responsiveness) which have been shown to be a) impaired in the context of postnatal depression and b) associated with adverse child outcomes (in attachment, behavioral, and cognitive domains). Regarding mothers' parenting behaviors, groups did not differ at the end of treatment or when children were 2 years old. Regarding children's outcomes at 2 years, development was examined in the domains of attachment, behavior, and cognitive development. In all these domains, children's development did not differ between the two groups but was found to be comparable with normative development in nonclinical samples. Stein and colleagues proposed that, given maternal depression had remitted in over 80% of mothers by the end of the first year, and over 85% by the end of the second year, children's developmental outcomes could be understood in the context of no exposure to maternal depression from late in the first year through to the end of their second year. Thus, intensive treatment of maternal depression up to the end of the first year together with the interventions on mother-infant interactions could be adequate to mitigate the impact of maternal postnatal depression on children's development at 2 years.

The trials reviewed above all addressed postnatal depression. The impact on infants of interventions for prenatal depression has received relatively little attention to date. Results are promising, with significant benefits for infants from two pilot RCTs. In their pilot RCT comparing individual, home-based CBT with treatment as usual (TAU) for ante-natal depression, Netsi and colleagues (249) found no significant differences in infant outcomes by treatment. Improved prenatal depression symptoms, however, were associated with easier infant temperament and shorter infant sleep duration 2 months postnatally. Milgrom and colleagues (250) found that group CBT for prenatal depression, compared to usual care, had medium to large effects on infant self-regulation, stress reactivity, and problem solving at 9 months old. These infant outcomes were obtained even when controlling for postnatal depression symptoms. While both pilot studies provide encouraging results, as pilot studies, neither was designed to examine hypotheses regarding fetal programming effects (173). Larger trials will be required to examine the mechanisms of *how* treatment of prenatal depression has its impact on infant development.

So far, we have only reviewed studies reporting RCTs that specifically focused on perinatal depression. However, there are other promising early intervention studies that depressed mothers may profit from and that are worth mentioning briefly. For example, in mindfulness-based programs, parents learn to relate differently to their own psychopathology and to their child (fostering more attentive and less overreactive parenting) through meditation practices. For example, Mindfulness-based Child birthing and Parenting (251, 252), an intervention for pregnant women and their partners, is found to reduce anxiety and depression in both the pregnant women and their partners (250) who play a role in buffering or increasing stress, anxiety, and depression of the future mother during pregnancy. Another intervention for mothers with psychopathology, Mindful with your baby, targets early parenting, babies with (regulation) problems, and mother-baby interaction problems (254, 255). Mindful with your baby was shown to lead to improvements in mothers' psychopathology, babies' or infants' behavior problems, and mothers' observed parenting and the mother-child interaction.

As the literature stands, in the context of maternal perinatal depression, short-term benefits in infant development have followed successful modification of maternal parenting behaviors, with benefits for children's development evident at 5 years of age where children had experienced stressful life events. Conversely, the impact of persistent postnatal depression on children's development can be mitigated, but *via* effective treatment of depression in the first postnatal year, sustained over the second year, without modification of the maternal parenting behaviors impaired by postnatal depression (PND).

Regarding mental illnesses other than depression, literature is less well developed. For example, for mothers with a range of mental illnesses, Fonagy and colleagues (231) conducted an RCT of Parent–Infant Psychotherapy (PIP), compared to TAU, for effects on infant cognitive, language, and motor development. When compared to TAU at 12 months, PIP had no effect on infant cognitive, language, or motor development. To enhance maternal parenting and infant outcomes in the context of maternal substance abuse disorders, Pajulo and colleagues (256, 257) have developed an intervention to promote maternal reflective functioning (RF). In a case series with 34 mother-infant pairs, they reported a significant increase in maternal RF from pretreatment to posttreatment, and that better RF was negatively associated with later relapse to substance use and children being placed in foster care (257). More robust research designs are required to establish the possible effects of enhancing maternal RF in the high-risk context of substance abuse disorders for infant outcomes.

Section Summary and Conclusions

Presently, it appears that treatment of depression prenatally may have beneficial effects on infants' self-regulation, stress reactivity, and temperament. However, postnatal interventions addressing either parental psychopathology or parent-infant relationship in isolation do not seem to significantly improve child outcomes. On the other hand, the combination of interventions targeting parental depression together with interventions on parent-infant relationship or with parental stress management show some promise in adequately limiting infants' exposure to the disorder's impact. It remains to be shown whether these positive effects extend beyond the end of the second postnatal year. Finally, the mechanisms *via* which positive infant outcomes can be achieved remain unclear. Research might fruitfully elucidate how interventions have their effects on enhancing children's outcomes by targeting those who face risks in addition to parental perinatal psychiatric disorder. For example, in addition to parent anxiety disorders infant BI is a risk factor for Social Anxiety Disorder (258). Thus, examining whether the effects of intervention for postnatal parental anxiety differ according to infant temperament (BI or not BI) could show how an intervention impacts infants' development [for example, *via* modifying one or both of postnatal anxiety disorder and BI (259)]. Effective early interventions targeting parental mental disorders and the parent-infant relationship may have a profound beneficial impact on the development of the child up to adulthood in many ways. Potentially such effects may even impact the next generation, as parenting experiences will affect future parenting behavior. As reflected in the focus of this intervention section, we require interventions for other psychiatric disorders and for fathers experiencing perinatal psychiatric disorders.

DISCUSSION

The current review provided a snapshot of the period between pregnancy and the first postnatal year among parents with mental disorders and their children by focusing first on the links between parental mental illness and behavioral, biological, and neurophysiological correlates of infant psychological functioning in this period. Next, to provide insight to the question of whether interventions may help to reduce or reverse this link, we focused on the effects of early interventions targeting parental mental illness (and/or) parenting on infants' psychological outcomes. The summarized evidence provides preliminary support for the idea that parental psychopathology may limit parents' ability to provide an optimal environment for the offspring's emotional and physiological development in this sensitive period where parents' synchrony, responsivity, affect expression, and regulation lays the necessary ground for healthy development in infants. The evidence further suggests that these psychopathology-related changes in parents' behavior and biology in the perinatal period may be related to significant alterations in brain development and to behavioral, biological, physiological, and neural correlates of infant psychological functioning in this period. The accompanying changes in infants' behavioral, biological, neural, and physiological profile seem to be reminiscent of the responses characterizing parents' psychopathology. For example, infants of depressed parents express less emotion and engage less in positive interactions, show lower vagal tone, stronger right frontal EEG activation, and elevated cortisol levels. These altered profiles in themselves may constitute risk for later development of child and/or adult forms of psychopathology and thus for intergenerational transmission.

These findings highlight the essential value of early interventions to alleviate the transmission of psychopathology risk from mentally ill parents to their infant. Although targeting depression or mother-infant interactions in isolation may not be sufficient in the postnatal period, intensive interventions targeting depression earlier, i.e., prenatally, and or more intensively—along with mother-infant interactions—may be promising in alleviating the risk of early transmission. It is important to underline that these early infant psychological profiles that are related to parental mental illness summarized in this article are only probabilistically related to later development of psychopathology and may not fully account for the intergenerational transmission of psychopathology. In fact, not all children of mentally ill parents develop psychopathology or maladaptive outcomes. From a developmental psychopathology perspective, psychopathology in the offspring of mentally ill parents at a given point in development emerges as a result of complex and dynamic interactions between risk and resilience factors operating at the psychological, biological, and social levels of influence up to that point (260). Later adaptation/maladaptation of the offspring certainly depends on further adversity or opportunities that may either aggravate or alleviate the transmitted risk in early development (97, 260, 261). Finally, as child characteristics such as BI start to play an increasingly pronounced role from infancy onwards (262), the bidirectional nature of the associations between parent and child outcome is important to consider in familial transmission.

Although our focus was exclusively on parental mental illness as a risk factor for psychopathology in this review, the inherent complexity of multiple risk/resilience factors and mechanisms that dynamically operate in the development of psychopathology in the offspring makes it necessary to consider the influence of other factors along with parental mental illness and the interventions. These factors include more proximal influences related to the characteristics of the parent [such as history of childhood abuse (90, 91)], the child [such as temperament or BI (258, 262) and gender (140)], the couple [such as coparenting (263) and marital satisfaction (264)], and the more distal influences regarding the family and culture and broader socio-economic determinants. Future studies that incorporate these factors in longitudinal designs in mentally ill parents from pregnancy up to the point where child psychopathology develops will be essential for a more complete understanding of intergenerational transmission.

Moreover, it is important to evaluate the conclusions in view of the limitations coming from the scope of the parental mental disorders addressed by the evidence, as well as by the methodological limitations inherent to the study designs. The summarized evidence predominantly comes from depression, followed by anxiety and traumatic stress, whereas this is likely to change, now that there is an increased recognition of the fact that all disorders along the diagnostic spectrum may manifest during pregnancy and the postnatal period in mothers and fathers (4, 17–19). Methodologically speaking, the reported associations between parental mental illness and infant outcomes are from semi-experimental designs, which preclude any causal inferences. The longitudinal designs therefore provide a unique advantage in establishing a timeline between infants' exposure to parental mental illness and the corresponding alterations in infant outcomes. Finally, methodological limitations are related to the chronic nature and continuity of parental psychopathology from the prenatal period onwards, which make it difficult to delineate the prenatal influence from postnatal and postnatal influence from later effects of psychopathology.

Finally, we note that, despite substantial psychopathology among (future) fathers, and taking into account that most children are raised by two parents, a mother and a father, most studies on the role of parental psychopathology and interventions focused on mothers, disregarding the various roles that parents play directly (for example, through exposure to paternal mental illness) and indirectly (for example, *via* buffering or increasing the psychopathology-related stress in the mother or in the triad). Future studies will need to elucidate these influences by including fathers or co-parents in their future research designs.

FINAL CONCLUSION AND IMPLICATIONS

The available evidence reviewed in the current study leaves no doubt about the importance of reaching men and women with a mental health problem who become parents or who are planning or expecting to become parents as early as possible. A recent meta-synthesis on the factors that prevent women with mental illness to reach out to healthcare services for support during the pregnancy and postnatal year provides insight to the potential ways of enhancing the use of healthcare services and reducing the isolation that mothers experience on the way to and/or in the early phases of parenthood (265). First, the stigma and fears about the loss of custody can be reduced *via* informing the general public on the broader scale and this specific group on a smaller scale about the high prevalence of mental illness in this period and about the possibilities of alleviating the effect of parental mental illness on the parent and the child. Second, it seems that providing some stability on who delivers the care and integrating the services such that the different components can be delivered by the same professionals who are open and accessible to share psychological needs may largely improve the experience of healthcare among individuals with mental illness. Third, a nonjudgmental and compassionate approach and a readiness to provide the needed information by health professionals have been highlighted as important qualities that may facilitate the help-seeking of men and women with mental illness for healthcare services in the perinatal period. Finally, putting an equal weight on the parents' and the baby's needs and involving the parents with mental health problems in the decision-making process related to medical and psychological treatment are of golden value in providing an optimal healthcare environment that parents with mental health problems may turn to whenever needed.

AUTHOR CONTRIBUTIONS

EA wrote the first drafts of the sections Introduction and Discussion, Final Conclusion and Implications and authored the section Behavioral Pathways: The Relationship between Prental Perinatal Mental Disorder and Early Indices of Infant Psychobehavioral Functioning. MT, JQ, and PL authored the sections: Biological Pathways: The Links Between Parental Prenatal Mental Disorder and Early Indices of Infant Psychobiological Functioning, Neurophysiological Pathways: The links Between Parental Perinatal Mental Disorder and Early Indices of Infant Neurophysiological Functioning, and Effect of Early Interventions on Parent and Infant Outcomes, respectively. BE and SB provided advice on the scope, structure, and content of the manuscript and contributed to the writing and revisions of the sections Introduction, Discussion and Final Conclusion and Implications. All authors contributed to manuscript revision and read and approved the submitted version.

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Prenatal Alcohol Exposure and Child Psychosocial Behavior: A Sibling Fixed-Effects Analysis

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Background: The association between low levels of alcohol consumption during pregnancy and children's health remains unclear because of the difficulty in ruling out residual genetic and environmental confounding factors. In this study, using a within-family sibling fixed effects design, we sought to examine the association between low prenatal alcohol exposures (PAE) and children's overall psychosocial behavior in a Japanese cohort.

Methods: We used maternal and sibling data from the Japanese Study of Stratification, Health, Income and Neighborhood 2012-2013. Households were recruited from the Tokyo metropolitan area through clustered random sampling. Children under 18 years old who have siblings ($n = 1,600$) and their mothers were selected. PAE status was retrospectively measured, and classified by binominal and continuous measurements. Outcome measures of children's psychosocial behavior were assessed with the Child Behavior Checklist T-score.

Results: Low PAE was significantly associated with the offspring's anxiety problems ($\beta = 1.54$, 95%CI = 0.26, 2.82) and internalizing problems ($\beta = 2.73$, 95%CI = 0.87, 4.60), and marginally significant with the offspring's total problem scores ($\beta = 2.34$, 95%CI = −0.24, 4.92). There was no significant difference in PAE between boys and girls when it comes to behavioral problems.

Conclusions: Low PAE was associated with children's anxiety, internalizing problems and overall problems, taking into account possible unobserved genetic and environmental confounding influences.

Keywords: alcohol-related disorders, developmental disabilities, maternal-fetal relations, pregnancy, prenatal alcohol exposure

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INTRODUCTION

The U.S. Centers for Disease Control, the U.S. Surgeon General, the American College of Obstetricians and Gynecologists, and the American Academy of Pediatrics uniformly advise women not to consume alcohol during pregnancy (1–5). However, the debate on whether "moderate consumption" of alcohol can be safely practiced during pregnancy continues (6–9). For example, a large-scale study in Denmark recently reported that antenatal exposure to binge drinking was associated with internalizing and externalizing behavioral problems among boys (5). However, the study found no association with lower doses of alcohol exposure. In fact, children of expectant mothers who consumed moderate amounts of alcohol were found to have better mental health than those whose mother abstained from alcohol–a controversial result that was widely reported in the media.

Although it is widely accepted that binge drinking during pregnancy increases the risk of fetal alcohol syndrome (10, 11), whether low-to-moderate levels of alcohol consumption during pregnancy can affect children's health remains controversial (12–14). A recent meta-analysis which assessed both normal and abnormal development as continuous variables concluded that mild-to-moderate prenatal alcohol exposure (PAE) during all trimesters was not associated with child psychosocial outcomes such as cognition and mental development (12). However, it suggested that further studies were needed to rule out residual confounding factors.

There are several reasons why it is challenging to investigate the effects of PAE on children's behavioral problems. First, mothers tend to under report their drinking levels during pregnancy because of prevailing social norms (14). Second, any correlation between maternal drinking during pregnancy and child outcomes is likely to be confounded by a range of unobserved factors including family environment and shared genetic influences. Third, it is ethically infeasible to design a randomized controlled trial to investigate the association between PAE and child behavioral outcomes. As an alternative to directly manipulating the exposure of interest, some sort of quasi-experimental approach is needed, such as the within-family sibling fixed-effects design. To our knowledge, there is a limited number of studies that have used quasi-experimental design to infer causality of deferential low PAE on behavior problems among twins, siblings, or cousins (15, 16). Hence, there has been a call for the implementation of more quasi-experimental study using natural experiment (17).

Notably, studies that considered observed and unobserved confounding factors are rare (16, 18, 19). The basic idea of a sibling fixed-effects design is to leverage the within-family, between-sibling differences in exposure to maternal drinking during pregnancy (e.g., mother drank during first pregnancy, but not during subsequent pregnancies), which effectively differentiates sibling-invariants observed and unobserved confounding variables, such as genetic influences or maternal temperament/personality, or family environment (19, 20).

The findings from the few existing studies using the sibling fixed-effects approach (16, 18, 19) suggest that even low-to-moderate PAE is a risk factor for children's externalizing problems such as attention deficit hyperactivity disorder, infant difficulties and children's behavioral problems. However, the literature remains sparse, especially in the Asian context where alcohol consumption among women is low compared to Western societies (21).

In this study, we examined the association between low PAE and children's overall psychosocial behavior in a Japanese cohort using the sibling fixed-effects design.

METHODS

Participants

The J-SHINE (Japanese study of Stratification, Health, Income and Neighborhood) is an ongoing cohort study established since 2010. Details of the study have been previously described (22). Briefly, the baseline survey was carried out between 2010 and 2011, when a clustered random sample of individuals aged 25–50 years residing in four municipalities in urban or suburban settings of the Tokyo metropolitan area were invited to participate. The household survey asked about the health of all children under the age of 18 years co-residing with the subjects. A follow-up survey was conducted between 2012 and 2013. In the baseline survey, 13,920 individuals were randomly selected from the "*koseki*" registration system, a compulsory domiciliary registration system in Japan that included all residents in target area ($N = 594{,}249$). Among the individuals invited to participate, 4,385 men and women responded (31.6% response rate), including 2,184 households with children under 18 years. By wave 2, the number of households with children increased to 2,244, and of these, 1,520 households (67.7%) agreed to participate in the follow-up survey (including 2,470 children under 18 years). Written informed consent was received from all the participants in the study. We excluded children who did not have the outcome data and the prenatal mothers who drink alcohol more than 2 times per week ($N = 33$, 1.7% of all sample) because we focused on low-level drinkers. Therefore, the number of children in the study was 1,933. In addition, we used only sibling data for the fixed-effects models, the total number of children who had siblings and who had outcome data was 1,600 (mean age 114.1 months ± 52.5 *SD*). Among them, 1,046 had two siblings, 518 had three siblings, and 36 had four siblings. If the number of siblings was more than two in one family, we compared each sibling pair separately. For example, if the number of the children were three (A, B, C) in one household, we compared A-B, A-C, and B-C.

The J-SHINE was conducted using computer-assisted personal interviewing (CAPI), unless the participants requested a face-to-face interview. This study was carried out in accordance with the recommendations of ethical guidelines for medical and health research involving human subjects. The study protocol was approved by the ethics committee of the Graduate School of Medicine of the University of Tokyo. All subjects provided written informed consent in accordance with the Declaration of Helsinki.

MEASUREMENTS

Prenatal Alcohol Drinking

The J-SHINE study asked mothers to report their drinking behavior during each pregnancy retrospectively. Response categories included: (1) 2 times or more per week, (2) 1–4 times

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Abbreviations: PAE, prenatal alcohol exposure; OLS, ordinary least squares.

TABLE 1 | Demographic characteristics of children and families for individual level.

Variables	All sample (*n* = 1933) *n* (%)	Siblings sample (*n* = 1600) *n* (%)
AREA CHARACTERISTICS		
Tokyo urban area (Adachi)	393 (20.3)	332 (20.8)
Tokyo urban area (Mitaka)	389 (20.1)	306 (19.1)
Tokyo suburban area (Kashiwa)	646 (33.4)	546 (34.1)
Tokyo suburban area (Tokorozawa)	505 (26.1)	416 (26.0)
FAMILY CHARACTERISTICS		
Mother’s age (years old) (mean, SD)	37.5 (5.5)	37.2 (5.3)
Father’s age (years old) (mean, SD)	39.4 (6.3)	39.2 (6.2)
MOTHER’S EDUCATION		
< = 12 years	946 (49.0)	388 (24.3)
>12 years	875 (45.3)	1186 (74.1)
unknown	112 (5.8)	26 (1.6)
FATHER’S EDUCATION		
< = 12 years	427 (22.1)	371 (23.2)
>12 years	1441 (74.6)	1181 (77.8)
unknown	30 (1.6)	48 (3.0)
Working mother	875 (45.3)	707 (44.2)
Working father	1631 (84.4)	1363 (85.2)
NUMBERS OF FAMILY		
< = 4	1351 (60.0)	1054 (65.9)
> = 5	574 (27.0)	541 (33.8)
Unknown	8 (0.4)	5 (0.3)
FAMILY INCOME		
<JPY 5 million[a]	438 (22.7)	362 (22.6)
JPY 5–7.5 million	572 (30.0)	484 (52.9)
JPY 7.5–10 million	391 (20.2)	329 (20.6)
>JPY 10 million	369 (19.1)	288 (18.0)
unknown	163 (8.4)	137 (8.6)
Domestic violence	631 (32.6)	538 (33.6)
Prenatal smoking	67 (3.5)	59 (3.7)
CHILDREN’S CHARACTERISTICS		
Number of siblings		
1	333 (17.3)	–
2	1046 (54.1)	1046 (65.4)
3	518 (26.8)	518 (32.4)
4	36 (1.9)	36 (2.3)
Sex (male)	971 (50.2)	800 (50.0)
Age (months) (mean, SD)	113.9 (53.7)	114.1 (52.5)
PRENATAL ALCOHOL CONSUMPTION		
1-4 times/month	110 (5.7)	98 (6.1)
rare	401 (20.7)	347 (21.7)
never	1083 (56.0)	886 (55.4)
unknown	339 (17.5)	269 (16.8)
CBCL *T* SCORE, *N*, MEAN (*SD*)		
4-18 Years old		
Physical problem	52.3 (4.8)	52.1 (4.7)
Social problem	53.3 (5.3)	53.0 (5.1)
Thought problem	51.0 (3.8)	51.0 (3.8)
Delinquency	52.9 (5.2)	52.9 (5.2)
2-18 YEARS OLD		
Withdrawal	53.4 (5.4)	53.2 (5.4)
Anxiety problem	52.7 (4.9)	52.6 (4.9)
Attention problem	53.0 (5.7)	52.8 (5.6)
Aggressiveness	53.4 (5.5)	53.4 (5.5)
Internalizing problem	49.6 (7.8)	49.3 (7.8)
Externalizing problem	49.6 (8.4)	49.5 (8.4)
Total problem	48.4 (9.7)	48.0 (9.7)

[a] *JPY, Japanese Yen; JPY 120 is approximately equal to US 1 dollar.*

per month, (3) rarely (but not zero), and (4) never. We excluded those who responded with “2 times or more per week” to focus on low prenatal alcohol drinking during pregnancy, and combined “1–4 times per month” and “rarely,” so that PAE was binarized into “never” vs. “ever.”

Outcome Variable: The CBCL4-18/2-3 (Child Behavior Check List 4-18/2-3)

The outcome variable, that is, children’s psychosocial developmental problems, was assessed with the CBCL4-18 checklist which targets children aged 4 to 18 years, and the CBCL2-3 which targets children aged 2 to 3 years (23, 24). The CBCL is a scale that assesses internalizing, externalizing, and total behavior problems using 113 items, with good psychometric properties (24). A higher score denotes more problematic behavior. Ratings were completed by caregivers (mother, $n = 1316$, 83.7%). The T score of each CBCL score was calculated using the standardized distribution among Japanese children and mean score represents the 50th percentile, which has been shown to have good reliability and validity (23–25).

Covariates

We considered a wide range of potential correlates of PAE as control variables in adjusted models. There are two types of covariates: sibling variables that are less likely to be variant and sibling-varying variables. With respect to our sibling fixed-effects models, sibling variables that are less likely to be variant include factors that did not vary between siblings, that is, parent’s age, educational level, working status, family income and domestic violence and others that are less likely to be changeable variables. In contrast, sibling-varying variables include children’s age, sex, and prenatal smoking during each pregnancy.

Statistical Analysis

To estimate the relationship between maternal drinking during pregnancy and children’s behavioral outcomes, we conducted ordinary least squares (OLS) regressions controlling for all observed sibling-varying and sibling-invariant variables for the full sample, as well as for the sibling sub-sample. Next, we conducted sibling fixed-effects analysis only among the siblings (i.e., singletons do not contribute to this analysis), with further adjustment of correlation of 3 or more siblings comparisons from the same family. In the fixed-effects model, the effects of PAE on children's outcomes were calculated by differentiating the unobserved, sibling-invariant shared factors, such as genetic or family environmental influences. Finally, we conducted fixed-effects models stratified by child sex (i.e., male-male, female-female, and male-female pair of siblings) as a sensitivity analysis. All analyses were performed with STATA 13.0 (StataCorp LP, College Station, TX, USA).

RESULTS

Table 1 shows the demographic characteristics of families and children comparing the full sample ($n = 1,933$) and the sample restricted to the siblings ($n = 1,600$). In the full sample, the frequencies of prenatal alcohol "1-4 times/month," "rarely," and "never" were 6.1, 21.7, and 55.4%, respectively. The vast majority of pregnant women in this Japanese cohort therefore fitted into the abstention or light drinking categories.

There was a total of 491 sibling pairs whose mother never drank during pregnancies (**Appendix 1**). Further, 37 (4.6%) mothers reported drinking during their first pregnancy but not during subsequent pregnancies. Another 31 mothers (3.9%) reported abstaining from alcohol during their first pregnancy, but consumed alcohol during subsequent pregnancies. Effects of PAE, binarized according to "ever" vs. "never," are shown in **Table 2**. In the OLS model, the effect of PAE was not significantly associated with children's psychosocial problems in both the overall sample and the sibling sample. In the unadjusted sibling fixed-effects model, prenatal drinking was significantly associated with children's delinquency ($\beta = 1.63$, 95%CI = 0.02, 3.25), anxiety problems ($\beta = 1.50$, 95%CI = 0.15, 2.85), internalizing problems ($\beta = 2.62$, 95%CI = 0.61, 4.62), and total problem scores ($\beta = 2.39$, 95%CI = 0.11, 4.66). After adjusting for sibling-varying variables, maternal drinking during pregnancy was significantly associated with offspring's anxiety problems ($\beta = 1.54$, 95%CI = 0.26, 2.82), internalizing problems ($\beta = 2.73$, 95%CI = 0.87, 4.60) and marginally significant with offspring's overall problem scores ($\beta = 2.34$, 95%CI = −0.24, 4.92). Additionally, adjusting for sibling-varying and sibling-invariant variables, such as parent's working status and maternal drinking during pregnancy, remained significantly associated with children's anxiety problems, internalizing problems and overall problem scores. In sensitivity analysis, maternal drinking during pregnancy measured with continuously (i.e., frequencies of alcohol drinking per month) was not statistically significant (**Appendix 2**).

Table 3 shows the sibling fixed-effects results of maternal drinking and children's psychosocial problems, stratified by children's sex. Drinking during pregnancy was not associated with girl's thought problems ($\beta = -2.56$, 95%CI = −7.59, 2.47) attention problems ($\beta = -0.16$, 95%CI = −2.29, 1.96), anxiety problems ($\beta = -2.56$, 95%CI = −0.80, 3.07) and aggression ($\beta = 0.27$, 95%CI = −3.17, 3.70). When it comes to the effect of drinking during pregnancy on internalizing, externalizing and total problems, there was no different between boys and girls.

TABLE 2 | OLS models and fixed-effects models of the association between PAE[a] and children's psychosocial behaviors.

	All sample, OLS, all measured variables adjusted[b]		Siblings sample, OLS, all measured variables adjusted[b]		Fixed effects model,unadjusted		Fixed effects model, adjusted[c]		Fixed effects model, adjusted[d]	
CBCL T score	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**
4-18 YEARS OLD										
Physical problem	0.29	−0.28, 0.86	0.51	−0.10, 1.11	0.94	−0.68, 2.56	1.19	−1.74, 4.11	0.90	−1.85, 3.65
Social problem	0.12	−0.53, 0.77	0.47	−0.20, 1.14	0.74	−0.95, 2.44	1.30	−0.63, 3.23	1.22	−0.76, 3.20
Thought problem	−0.35	−0.77, 0.07	−0.24	−0.67, 0.20	−0.83	−2.11, 0.44	−0.80	−3.34, 1.73	−0.86	−3.35, 1.63
Delinquency	0.48	−0.14, 1.10	0.51	−0.17, 1.19	**1.63**	**0.02, 3.25**	1.73	−0.33, 3.79	1.57	−0.50, 3.64
2–18 YEARS OLD										
Withdrawal	−0.01	−0.65, 0.62	0.19	−0.50, 0.88	−0.51	−1.95, 0.93	−0.40	−1.59, 0.79	−0.41	−1.66, 0.84
Anxiety problem	0.14	−0.43, 0.70	0.43	−0.18, 1.05	**1.50**	**0.15, 2.85**	**1.54**	**0.26, 2.82**	**1.40**	**0.09, 2.72**
Attention problem	0.27	−0.37, 0.91	0.43	−0.23, 1.10	−0.53	−2.20, 1.13	−0.44	−2.57, 1.69	−0.58	−2.78, 1.63
Aggressiveness	0.32	−0.33, 0.96	0.49	−0.21, 1.20	1.18	−0.45, 2.80	1.15	−0.70, 2.99	1.23	−0.69, 3.16
Internalizing problem	0.41	−0.49, 1.31	0.91	−0.07, 1.88	**2.62**	**0.61, 4.62**	**2.73**	**0.87, 4.60**	**2.68**	**0.73, 4.63**
Externalizing problem	0.49	−0.48, 1.45	0.72	−0.35, 1.78	1.77	−0.45, 3.99	1.50	−1.04, 4.04	1.64	−0.94, 4.22
Total problem	0.42	−0.71, 1.55	0.95	−0.30, 2.19	**2.39**	**0.11, 4.66**	2.34[e]	−0.24, 4.92	2.32	−0.34, 4.98

[a] *Measurements of alcohol drinking is binary outcomes (never = 0, more than one time = 1).*

[b] *Adjusted by children's age and sex, parent's age, education and working status, family income, prenatal smoking, domestic violence, clustered by family ID.*

[c] *Adjusted by children's age and sex and difference levels of prenatal smoking baseline prenatal drinking status among siblings, clustered by family ID.*

[d] *Adjusted by children's age and sex, parent's education level, family income, family number, domestic violence and baseline prenatal drinking status among siblings, clustered by family ID.*

[e] *Marginally significant: p = 0.08.*

Bold values means the results which is statistically significant.

TABLE 3 | Fixed-effects model stratified by sibling's sex pairs.

	Male-male pair		Female-female pair		Male-female pair	
CBCL T score	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**
4-18 YEARS OLD						
Physical problem	0.01	−3.88, 3.90	1.05	−4.41, 6.51	2.11	−1.95, 6.18
Social problem	1.34	−2.28, 4.97	1.16	−1.64, 3.96	1.27	−1.71, 4.24
Thought problem	1.39	−1.35, 4.12	−2.56	−7.59, 2.47	−1.43	−4.13, 1.27
Delinquency	0.76	−3.22, 4.75	0.16	−3.06, 3.39	**3.15**	**0.52, 5.77**
2–18 YEARS OLD						
Withdrawal	−0.81	−3.41, 1.79	−1.22	−2.47, 0.03	0.33	−1.28, 1.95
Anxiety problem	2.32	−0.56, 5.20	1.13	−0.80, 3.07	1.37	−0.07, 2.82
Attention problem	1.04	−1.98, 4.06	−0.16	−2.29, 1.96	−1.60	−5.49, 2.30
Aggressiveness	1.27	−2.77, 5.30	0.27	−3.17, 3.70	1.68	−0.86, 4.22
Internalizing problem	2.35	−1.76, 6.46	2.32	−1.18, 5.81	**3.34**	**1.05, 5.63**
Externalizing problem	0.86	−3.96, 5.67	−0.10	−4.37, 4.18	2.76	−1.18, 6.69
Total problem	1.24	−3.24, 5.72	0.64	−3.26, 4.54	4.03	−0.05, 8.11

Bold values means the results which is statistically significant.

DISCUSSION

We found that low PAE during pregnancy is associated with children's anxiety, internalizing and overall problems. Our findings serve as additional evidence for the deleterious impact of maternal drinking during pregnancy on children's psychosocial behavior, and are consistent with previous animal models suggesting that even a relatively small amount of alcohol during fetal development can result in an increase in synaptic connectivity specific to the basolateral amygdala and induce a subtle anxiety-like behavior in rats (26).

Previous studies have reported inconsistent results regarding which sex is more affected by exposure to alcohol *in utero* (5, 27). Sayal et al. suggested that girls whose mother drank alcohol less than once per week during pregnancy have increased risk of behavioral problems; however, these results should be interpreted with caution because of a lack of evidence on dose-response effects (27). In contrast, another study indicated that boys are more vulnerable to PAE than girls (5), because of the brain development trajectory (28, 29). Our findings suggest that PAE is particularly deleterious for certain developmental problems such as attention problems, anxiety problems and aggression in male offspring. However, when it comes to internalizing, externalizing and overall developmental problems, there is no difference between sexes. The mechanism of how PAE affects children's behavior is still unknown and we cannot eliminate social environment completely in this study.

Our study has several limitations. First, the assessment of drinking during pregnancy was retrospectively self-reported by the mothers. Furthermore, we assessed only the frequency of drinking (as opposed to the total amount of ethanol consumed) or the timing of drinking during different trimesters of pregnancy. However, there is evidence that retrospective reports of prenatal substance use can be reliable (30, 31). Nonetheless, further study on the effects of both the timing of alcohol use and the total amount consumed is warranted. Second, the assessment of child psychosocial problems was reported by the parents, which may have given rise to information bias. However, in the sibling fixed-effects model, reports on behavior problems among siblings were made by the same parent, thereby reducing the possibility of differential misclassification. It has also been found by independent researchers that assessment by mothers is often more reliable than laboratory assessment because the latter is only a snapshot of children's behaviors (32). Finally, the sibling fixed-effects models cannot completely control for unknown sibling-varying confounders such as changes in family circumstances and social situations that differed between siblings. Although this study used sibling-invariant variables as parent's age, educational level, working status, family income and domestic violence, these factors could vary between siblings and we did not measure these variables at the time when the mothers became pregnant. Nonetheless, fixed-effects models are known to provide a more credible causal identification strategy compared with traditional OLS regression models (20, 33).

In conclusion, our study provides additional evidence that even low PAE during pregnancy may adversely affect children's psychosocial behaviors, especially anxiety problems. Our findings provide further support for the current recommendation of abstinence during pregnancy, based on the notion that there is no known safe threshold of alcohol consumption during pregnancy.

AUTHOR CONTRIBUTIONS

KI analyzed the data and drafted the paper and designed the study. IK was responsible for conception and design of the study and coauthored the paper, TF was involved in the study design, supervised the data analysis, and coauthored the paper. All authors read and approved the final manuscript.

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Conflict of Interest Statement: 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

TABLE A1 | Distribution of prenatal alcohol consumption among siblings pair (pair =799) N (%).

Both siblings were not exposed to alcohol	491 (61.5)
Senior child exposed, junior child not exposed	37 (4.6)
Senior child not exposed, junior child exposed	31 (3.9)
Both siblings were exposed to alcohol	240 (30.0)

TABLE A2 | OLS models and fixed-effects models of the association between PAE[a] and children's psychosocial behaviors.

	All sample, OLS, all measured variables adjusted[b]		**Siblings sample, OLS, all measured variables adjusted[b]**		**Fixed effects model,unadjusted**		**Fixed effects model, adjusted[c]**		**Fixed effects model, adjusted[d]**	
CBCL T score	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**	**β**	**95%CI**
4-18 YEARS OLD										
Physical problem	0.25	−0.26, 0.76	0.53	−0.02, 1.07	1.25	−0.09, 2.60	1.47	−0.34, 3.27	1.10	−0.47, 2.67
Social problem	0.34	−0.24, 0.92	0.54	−0.06, 1.13	0.76	−0.64, 2.16	0.94	−1.01, 2.89	0.82	−1.22, 2.86
Thought problem	−0.20	−0.58, 0.18	−0.11	−0.50, 0.28	−0.49	−1.55, 0.56	−0.37	−2.20, 1.47	−0.52	−2.41, 1.36
Delinquency	0.47	−0.09, 1.03	0.47	−0.14, 1.08	1.16	−0.18, 2.50	1.12	−0.16, 2.41	0.85	−0.49, 2.20
2–18 YEARS OLD										
Withdrawal	0.17	−0.39, 0.73	0.45	−0.16, 1.07	−0.72	−1.92, 0.49	−0.50	−1.62, 0.61	−0.62	−1.78, 0.53
Anxiety problem	0.17	−0.33, 0.67	0.47	−0.08, 1.01	0.49	−0.65, 1.62	0.57	−0.87, 2.01	0.41	−1.00, 1.82
Attention problem	0.50	−0.07, 1.06	**0.68**	**0.09, 1.27**	0.37	−1.03, 1.77	0.51	−1.36, 2.37	0.29	−1.70, 2.29
Aggressiveness	**0.69**	**0.12, 1.25**	**0.93**	**0.31, 1.54**	0.74	−0.61, 2.08	0.74	−1.02, 2.51	0.64	−1.16, 2.44
Internalizing problem	0.58	−0.22, 1.38	**1.08**	**0.21, 1.95**	1.02	−0.67, 2.71	1.35	−0.18, 2.87	1.25	−0.30, 2.80
Externalizing problem	**1.14**	**0.28, 1.99**	**1.35**	**0.41, 2.29**	1.22	−0.65, 3.08	1.39	−0.83, 3.62	1.36	−0.89, 3.60
Total problem	**1.01**	**0.01, 2.01**	**1.42**	**0.31, 2.52**	1.12	−0.79, 3.04	1.31	−0.76, 3.38	1.14	−1.01, 3.30

[a] *Measurements of alcohol drinking was frequencies of alcohol drinking per month (continuous variables).*

[b] *Adjusted by children's age and sex, parent's age, education and working status, family income, prenatal smoking, domestic violence, clustered by family ID.*

[c] *Adjusted by children's age and sex and difference levels of prenatal smoking baseline prenatal drinking status among siblings, clustered by family ID.*

[d] *Adjusted by children's age and sex, all measured variables about family and baseline prenatal drinking status among siblings, clustered by family ID.*

Bold values means the results which is statistically significant.

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Prenatal Depression and Its Associated Risk Factors Among Pregnant Women in Bangalore: A Hospital Based Prevalence Study

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Background: Depression is the commonest psychological problem that affects a woman during her perinatal period worldwide. The risk of prenatal depression increases as the pregnancy progresses and clinically significant depressive symptoms are common in the mid and late trimester. There is a paucity of research on depression during the prenatal period in India. Given this background, the present study aimed to assess the prevalence of prenatal depression and its associated risk factors among pregnant women in Bangalore, Southern India.

Methods: The study was nested within an on-going cohort study. The study participants included 280 pregnant women who were attending the antenatal clinic at Jaya Nagar General Hospital (Sanjay Gandhi Hospital) in Bangalore. The data was collected by using a structured questionnaire which included. Edinburgh Postnatal Depression Scale (EPDS) to screen for prenatal depression.

Results: The proportion of respondents who screened positive for prenatal depression was 35.7%. Presence of domestic violence was found to impose a five times higher and highly significant risk of developing prenatal depression among the respondents. Pregnancy related anxiety and a recent history of catastrophic events were also found to be a positive predictors of prenatal depression.

Conclusion: The high prevalence of prenatal depression in the present study is suggestive of its significance as a public health problem. Health care plans therefore can include screening and diagnosis of prenatal depression in the antenatal care along with other health care facilities provided.

Keywords: prenatal depression, pregnant women, domestic violence, marital discord, social support, pregnancy related anxiety, Banglaore

INTRODUCTION

The relationship between a pregnant woman and her developing fetus is possibly the most earnest and overwhelming but perplexing of all human relationships. Pregnancy entails physiological, hormonal and psychological changes which could increase the probability of mental and emotional changes resulting in depression, anxiety or psychological distress in the pregnant mother (1).

Maternal and Child Health Programmes in developing countries are commonly focused upon improving the nutritional status and less importance is given toward a woman's emotional and mental health during and after pregnancy (2, 3). Poor mental health of the woman during pregnancy could have profound consequences for the mother and her child in terms of adverse pregnancy outcomes and offspring development (4–7). Most of the existing data, research, and practice policies with regard to perinatal mental disorders center on the postnatal period and there is less research related depression during pregnancy (8, 9).

Depression is the most common psychological problem that affects a woman worldwide during the perinatal period (3, 10). About 15 % of women are known to be depressed at some point during their lifetime and more predominantly during pregnancy and after childbirth (11). The risk of prenatal depression increases significantly as the pregnancy progresses and clinically significant depressive symptoms are common in the mid and late trimester (12). The prevalence rates of prenatal depression differ between high, middle and low—income countries. Studies from various countries around the world show a prevalence rate ranging from as low as 4% to as high as 81% (13–16). The prevalence rate is reported to be lower in high income countries like Australia 7% (17), Hong Kong 4.4% (18), Finland 7.7% (19), and higher in many of the low-income countries like Pakistan 64.6% (20), Bangladesh 18% (13), Nigeria 24.5% (14), and Ethiopia 24.94% (15). The prevalence of depression in India is varies from 9.18% in one study to 36.7 % reported in another study (21, 22).

Even though prenatal depression is an important public health problem, most studies related to maternal depression are focused on post-natal depression and its outcomes; hence there is paucity of research on depression during the prenatal period, especially from India (8). The importance of screening for depression during pregnancy is that prenatal depression, if not treated and diagnosed early, may continue as postnatal depression (23–25) later on and could also result in an adverse influence on birth outcomes and offspring development. Given this background, the present study aimed to assess the prevalence of prenatal depression and its associated risk factors among pregnant women in Bangalore, Southern India.

MATERIALS AND METHODS

Study Setting and Participants

The study sample included of pregnant women who were attending the antenatal clinic at Jaya Nagar General Hospital (also known as Sanjay Gandhi Hospital), which is a public sector hospital in Bangalore. The study was nested within an ongoing cohort study, the study protocol of which was published earlier (26). The eligibility criteria included women above or equal to 18 years of age, with confirmed pregnancy of <6 months (<24 weeks) and having no obstetric or medical complication in the present pregnancy. The study analyzed the data of 280 pregnant women who had enrolled and completed the baseline visit for the study between August 2017 and April 2018.

Data Collection

Data was obtained from the pregnant women by means of an interview, after obtaining written informed consent. A participant information sheet that explained the purpose and nature of the study was issued to those who were willing to participate in the study. The respondents were ensured about privacy and confidentiality of data. The interview process employed the use of a structured questionnaire installed in an Android tablet App. The App included questions about socio-demographic data, obstetric history, medical history and measures for depression, social support, marital discord, domestic violence, and pregnancy related anxiety described below. Data related history of any mental illness and recent catastrophic event was also recorded. Calibrated instruments were used to measure height and weight and calculate the Body Mass Index (BMI). Data on hemoglobin estimation was obtained from hospital records. Depression, being the outcome variable was measured using Edinburg Postnatal Depression Scale (EPDS).

Study Measures

Depression

EPDS is a widely used 10-item self-reporting instrument, specifically designed for assessing both prenatal as well as postnatal depression. It has a sensitivity of 86%, specificity of 78% and positive predictive value of 73% (27). EPDS has been validated for detecting depression in both antepartum and postpartum mothers in many countries. This scale consists of 10 short questions with a choice of four answers that closely reflects about how she was feeling over the past 7 days. Scores are recorded as 0, 1, 2, and 3 according symptom severity. Certain question items (i.e., 3, 2, 1, and 0) are scored in a reverse manner. Respondents who score 13 and above are likely to be suffering from depression and should seek medical attention.

Social Support

The Multidimensional Scale of Perceived Social Support Scale (MSPSS) used to measure social support includes 12 questions, and is validated for use in the South Asian population (28, 29). These questions directly address the adequacy of social support and have a 7-point rating scale ranging from "very strongly disagree" to "very strongly agree." The scale assesses the perceptions of social support adequacy from three specific sources: family, friends, and "significant other." A score of <2 is considered as low support, a score of 3–5 as moderate support while score of more than five indicates high support.

Marital Discord

The Revised Dyadic Adjustment Scale (30, 31) measures seven dimensions of relationship among partners within three categories: decision making, values and affection. It consists of 14 items in which the respondents can rate their relationship on a 6-point scale. Scores range from 0 to 69; higher the score greater, is the relationship and *vice versa*. The cut- off score was taken as 48.

Spouse Physical and Sexual Violence

Spouse physical and sexual violence was measured using the Modified Conflict Tactics Scale (32). It is effective and useful in measuring domestic violence in diverse cultural settings. The scale has 9 questions wherein the respondents affirm whether domestic violence was present or absent.

Socio Economic Scale

The socio-economic class of the respondents was measured by the Modified Kuppuswamy Socio Economic Scale (33). The scale uses education, occupation of the head of the family and monthly family income to calculate socio-economic status. The scores awarded to education and occupation of the head of the family remains unchanged. Revised Consumer Price Index–IW (industrial workers) is used to calculate the monthly income range. The socio-economic status is classified as upper class, upper middle class, lower middle class, upper lower class and lower class.

Pregnancy Related Anxiety

The 10-item Pregnancy Related Anxiety Questionnaire (PRAQ) was used to screen for pregnancy anxiety (34). It appears to have good psychometric and predictive validity for child-birth and childhood outcomes. Each item is scored on a 4-point scale with cut-off scores of 28 and 24 for nulliparous and multiparous women and the internal consistency (Cronbach's alpha) of PRAQ was seen to be 0.79. A score of more than 28 was considered as anxious.

Statistical Analysis

Data were retrieved from the data server. This was followed by data cleaning and analysis using SPSS version 22. Descriptive statistics such as percentage, means and standard deviation were used to summarize the socio demographic data. An EPDS score of 13 and above pointed toward the likelihood of presence of depression. The independent variables were categorized to analyze the association between each independent and outcome variable using a bivariate analysis to calculate the Crude Odd's Ratio with 95% Confidence Interval. Those variables that were associated at a P-value of < 0.2 in the bivariate analysis were entered into a multivariate logistic regression model to calculate the Adjusted Odd's Ratio and to eliminate the effects of confounding. Variables with a P-value of <0.05 in the multivariate analysis were considered to be significant.

Ethical Considerations

The study was approved by the Ethical Committee of Indian Institute of Public Health Bangalore campus (IIPHHB/TRCIEC/118/2017). Written informed consent was obtained from the pregnant mothers and they were assured of confidentiality and privacy of records.

RESULTS

Socio Demographic Characteristics of the Respondents

Table 1 shows the frequency distribution of socio demographic characteristics of the respondents. Of the 280 pregnant mothers, majority (72.9%) of them belonged to the age group of more than 20 years, the mean age of the respondents being 23.02 ± 3.40 years. Over two-thirds among them (72.1%) were Muslim and 40.4% had completed High school. While 92.1% were housewives, the spouses of over half of the respondents (51.8%) were semi- skilled workers. According to the Kuppuswamy Socio economic status scale, more than half of the respondents (57.5%) belonged to Upper Lower class. Nearly seventy percent of the

TABLE 1 | Socio demographic characteristics of the study participants (N =280).

Socio demographic characteristics	Frequency (n = 280)	Percentage (%)
AGE GROUP (IN YEARS)		
≤20	76	27.1
>20	204	72.9
RELIGION		
Hinduism	73	26.1
Christianity	05	1.8
Islam	202	72.1
EDUCATIONAL QUALIFICATION OF THE RESPONDENTS		
Illiterate	07	2.5
Primary school	08	2.9
Middle school	77	27.5
High school	113	40.4
PUC or diploma	52	18.6
Graduate	23	8.2
EDUCATIONAL QUALIFICATION OF THE HUSBANDS		
Illiterate	36	12.9
Primary school	20	7.1
Middle school	71	25.4
High school	95	33.9
PUC or diploma	36	12.9
Graduate and post-graduate	22	7.8
OCCUPATION OF THE RESPONDENTS		
Unskilled worker	11	3.9
Semi-skilled worker	10	3.6
Clerical or farmer	01	0.4
Housewife	258	92.1
OCCUPATION OF THE HUSBANDS		
Unemployed	01	0.4
Unskilled worker	96	34.3
Semi-skilled worker	145	51.8
Skilled worker	35	12.5
Clerical or farmer	01	0.4
Semi professional	02	0.7
SOCIO ECONOMIC STATUS		
Upper middle class	36	12.9
Lower middle class	83	29.6
Upper Lower class	161	57.5

pregnant mothers had no blood relationship with their husbands, where as a notable 13.2% said that their husbands were a first cousin from their mother's side.

Prevalence and Magnitude of Prenatal Depression Among the Pregnant Women

Of the 280 pregnant mothers, the proportion of those who screened positive for prenatal depression was 35.7% (100) suggesting a high probability of clinical depression (**Figure 1**). The mean EPDS score among the respondents was 10.61± 7.48.

Association of Prenatal Depression With Socio-Demographic Characteristics

The association of socio-demographic factors like age group, educational qualification, occupation, and socio-economic status of the respondents with depression was non-significant on bivariate analysis (p-value >0.05). This is seen from **Table 2**.

Association of Prenatal Depression With Obstetric History (Table 3)

On bivariate analysis, the number of pregnancies (gravida) and unplanned pregnancy showed an association with depression at a P-value of < 0.2. However, there was no significant association observed from multivariate logistic regression analysis.

Association Between Social Support, Marital Discord, Domestic Violence, Prenatal Anxiety, Consanguinity, and Catastrophic Events With Prenatal Depression (Table 4)

Association with low social support and presence of marital discord was significant on bivariate analysis but not in multivariate logistic regression. Presence of domestic violence was found to impose a five times higher and highly significant risk of developing prenatal depression among the respondents (COR = 5.438; 95% CI: 1.6–17.5, AOR = 5.916; 95% CI: 1.7–20.5). Pregnancy related anxiety was also found to be a positive predictor of prenatal depression (COR = 1.731; 95%

TABLE 2 | Association of socio demographic characteristics with depression among the pregnant women ($N = 280$).

Socio demographic Characteristics	Depressed mothers (>13) ($n = 100$)	Non-depressed mothers (<13) ($n = 180$)	Bivariate analysis-crude odd's ratio (95% CI)	P-value
AGE GROUP (IN YEARS)				
≤20	29 (29%)	47 (26.1%)	1.156 (0.670–1.994)	0.603
>20	71 (71%)	133 (73.9%)	1	
EDUCATIONAL QUALIFICATION OF THE RESPONDENTS				
<High school	77 (77.0%)	126 (70.0%)	0.710 (0.403–1.250)	0.236
≥High school	23 (23.0%)	54 (30.0%)	1	
EDUCATIONAL QUALIFICATION OF THE HUSBANDS				
<High school	49 (49%)	78 (43.3%)	1.256 (0.769–2.052)	0.362
≥High school	51 (51%)	102 (56.7%)	1	
OCCUPATION OF THE RESPONDENTS				
Working	08 (8.0%)	15 (8.3%)	1.045 (0.427–2.559)	0.922
Housewife	92 (92.0%)	165 (91.7%)	1	
OCCUPATION OF THE HUSBANDS				
Skilled worker	12 (12%)	26 (14.4%)	1	
Semi/unskilled worker	88 (88%)	154 (85.6%)	1.238 (0.595–2.575)	0.568
SOCIO ECONOMIC STATUS				
Upper middle class	12 (12.0%)	24 (13.3%)	1	
Lower middle class	26 (26.0%)	57 (31.7%)	0.912 (0.396–2.100)	0.829
Upper lower class	62 (62.0%)	99 (55.0%)	1.253 (0.584–2.686)	0.563

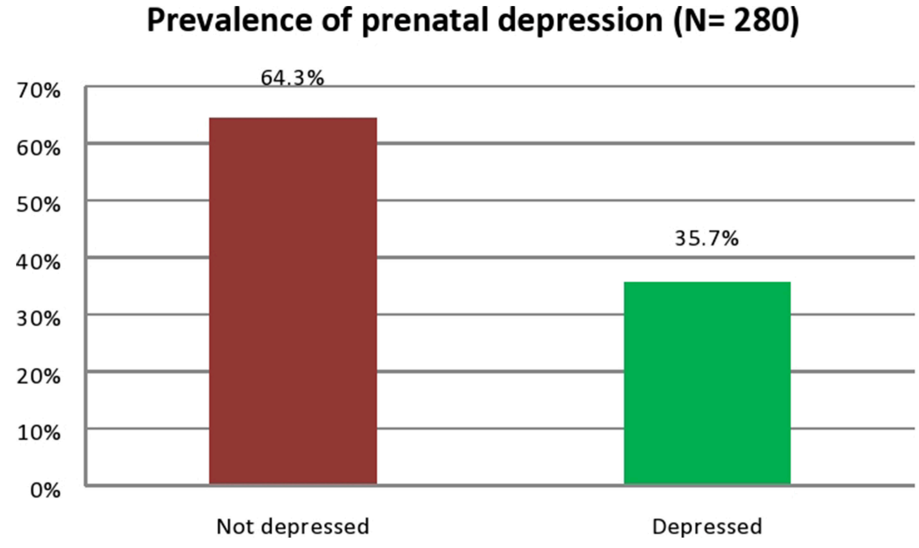

FIGURE 1 | Prevalence of prenatal depression among the pregnant women ($N = 280$).

TABLE 3 | Association of depression with obstetric history of the pregnant women ($N = 280$).

Obstetric history	**Depressed mothers (>13) (*n* = 100)**	**Non-depressed mothers (<13) (*n* = 180)**	**Bivariate analysis crude OR (95% CI)**	***P*-value**	**Multivariate analysis adjusted OR (95% CI)**	***P*-value**
GRAVIDITY						
Primigravida	37 (37.0%)	81 (45.0%)	1		1	
Multigravida	63 (63.0%)	99 (55.0%)	1.393 (0.844–2.299)	0.195	1.386 (0.793–2.424)	0.252
PARITY						
Primipara	43 (43.0%)	86 (47.8%)	1			
Multipara	57 (57.0%)	94 (52.2%)	1.213 (0.741–1.984)	0.442		
HISTORY OF ABORTION						
Yes	25 (25.0%)	37 (20.6%)	0.776 (0.435–1.385)	0.391		
No	75 (75.0%)	143 (79.4%)	1			
PREGNANCY UNPLANNED						
Yes	52 (52.0%)	74 (41.1%)	1.552 (0.949–2.538)		1.604 (0.925–2.782)	
No	48 (48.0%)	106 (58.9%)	1	0.080	1	0.092

TABLE 4 | Association of maternal depression with social support, marital discord, and domestic violence among the pregnant mothers ($N = 280$).

Maternal characteristics	**Depressed mothers (>13) (*n* = 100)**	**Non-depressed mothers (<13) (*n* = 180)**	**Bivariate analysis crude OR (95% CI)**	***P*-value**	**Multivariate analysis adjusted OR (95% CI)**	***P*-value**
SOCIAL SUPPORT						
High support (1–2.9)	44 (44.0%)	94 (52.2%)	1		1	
Moderate support (3–5)	24 (24.0%)	54 (30.0%)	0.949 (0.521–1.729)	0.865	0.839 (0.436–1.616)	0.600
Low support (5.1–7)	32 (32.0%)	32 (17.8%)	2.136 (1.164–3.919)	0.014	1.785 (0.915–3.481)	0.089
MARITAL DISCORD						
No	32 (32.0%)	79 (43.9%)	1		1	
Yes	68 (68.0%)	101 (56.1%)	1.662 (0.995–2.776)	0.052	1.517 (0.862–2.671)	0.149
SPOUSE PHYSICAL AND SEXUAL VIOLENCE						
Yes	11 (11.0%)	04 (2.2%)	5.438 (1.684–17.564)	0.005	5.916 (1.703–20.558)	0.005*
No	89 (89.0%)	176 (97.8%)	1		1	
PREGNANCY RELATED ANXIETY						
Absent	47 (47.0%)	109 (60.6%)	1		1	
Present	53 (53.0%)	71 (39.4%)	1.731 (1.057–2.836)	0.029	2.016 (1.134–3.587)	0.017*
CONSANGUINITY						
No	77 (77.0%)	121 (67.2%)	1			
Yes	23 (23.0%)	59 (32.8%)	0.613 (0.350–1.073)	0.086	0.728 (0.397–1.334)	0.304
CATASTROPHIC EVENTS						
No	64 (31.4%)	140 (68.6%)	1		1	
Yes	36 (47.4%)	40 (68.6%)	1.969 (1.149–3.374)	0.014	2.148 (1.203–3.837)	0.010*

**Positively significant according to multivariable analysis.*

CI: 1.05–2.8, AOR = 2.016; 95% CI: 1.13–3.5). The blood relationship with the husband did not show any significant association with prenatal depression on bivariate analysis and multivariable analysis. Presence of catastrophic events over the past 1 year imposed a two times higher and significant risk of developing prenatal depression among the respondents (COR = 1.969; 95% CI: 1.14–3.37, AOR = 2.148; 95% CI: 1.20–3.83, *p*-value = 0.010). History of mental illness was not included in the analysis because only one respondent had history of this kind and was undergoing treatment with medications.

Association of Prenatal Depression With Physiologic Parameters (Table 5)

Table 5 shows the association between physiologic parameters with prenatal depression. Presence of anemia (COR = 1.621; 95% CI: 0.9–2.7, AOR = 1.586; 95% CI: 0.91–2.75) showed some strong association with prenatal depression although this was

TABLE 5 | Association of prenatal depression with physiological parameters (*N* = 280).

Physiological parameters	Depressed mothers (>13) (*n* = 100)	Non-depressed mothers (<13) (*n* = 180)	Bivariate analysis crude OR (95% CI)	*P*-value	Multivariate analysis adjusted OR (95% CI)	*P*-value
BODY MASS INDEX						
Normal	56 (56.0%)	96 (53.3%)	1			
Underweight	12 (12.0%)	21 (11.7%)	0.980 (0.448–2.141)	0.959		
Obese	32 (32.0%)	63 (35.0%)	0.871 (0.508–1.491)	0.614		
ANEMIA						
Present	41 (41.0%)	54 (30.0%)	1.621 (0.973–2.701)	0.063	1.586 (0.912–2.756)	0.102
Absent	59 (59.0%)	126 (70.0%)	1		1	

not statistically significant. No association found between BMI and depression.

DISCUSSION

In this study we have measured the prevalence of prenatal depression among pregnant women and its association with certain risk factors such as socio-demographic characteristics, obstetric history, social support, marital discord, spouse physical and sexual violence, and physiologic measurements which included body mass index and hemoglobin level.

The mean age of respondents was 23.02 ± 3.40 years, which reflects upon the Indian cultural tradition of early marriage and parenthood. The prevalence of depression during pregnancy was 37.8% which is suggestive of a high probability of depression (using an EPDS cutoff score ≥13) among the respondents. The prevalence of prenatal depression makes it a significant public health issue in the study region. EPDS has been validated for use in India and Karnataka (26). Our study used a cut off score of more than or equal to 13 to identify women with depression; this yields a sensitivity of 100% and specificity of 84.9% in Indian settings (27). Another study from Karnataka showed an almost similar prevalence of 36.8% (21) whereas George et al., Ajinkya et al., and Bavle et al. observed much lower prevalence rates of 16.3% in coastal south India (35), 9.18% in Navi Mumbai (22), and 12.3% in Bangalore (36), respectively. This difference could be attributed to diversity in the socio-economic status, socio-cultural and psychosocial factors such as social support which might vary across different regions in the country. Moreover, this study was conducted in a public sector hospital setting, which in itself could pose as a risk factor and predictor for prenatal depression (28) due to inadequate quality of care in such settings.

In our study, majority of the study participants belonged to the low income group. Although we could not document a significant association with socio-economic status, the risk of depression during, and after pregnancy is higher among the socially disadvantaged group (10, 37, 38). It is hypothesized that low income increases the likelihood of poor living conditions, financial struggle and influences interpersonal relationships which could lead to psychosocial stress. Over a third of the study participants were high school graduates though over 90% were not working; however there was no association of education and occupation with depression. Bavle et al. (36) in their study among pregnant women in Bangalore observed that being educated but not employed outside the house could predispose to depression during pregnancy. Study findings from other low income settings point toward a significant association of a woman's occupation with depression: women who were housewives or employed in the private sector or as a laborer or merchant business were prone to get depressed during pregnancy (39, 40). Other socio-demographic factors such as age, husband's education, and occupation did not predict the occurrence of depression in the present study even though some studies have identified young age as a risk factor (41, 42). In Asian settings, having an unemployed or uneducated husband increases the probability of depression (43, 44).

Among the obstetric history variables, unplanned pregnancy increased the odds of depression on bivariate analysis. However, no significant association was observed on multivariate logistic regression analysis. Other studies show that the chance of getting depressed is higher in case of an unplanned pregnancy (15, 37, 45). Similarly multigravidity appeared to be risk factor for depression on bivariate analysis but not on logistic regression analysis although some studies do report a significant relationship (36, 46).

In this study, among the psychosocial factors, presence of spouse physical and sexual violence and pregnancy related anxiety were significant risk factors for prenatal depression in the multivariable analysis. Earlier research has also reported a strong relationship between domestic violence and the risk of depression in pregnancy in high as well as middle to low—income settings (47, 48). Moderate and low social support were significantly related on bivariate but not on multivariate analysis. The linkage between poor social support and prenatal depression has been well-documented (49, 50). Low social support may increase mental stress by inducing feelings of insecurity, predispose toward substance abuse (51), and promote interpersonal conflict (52). The findings from the present study are concurrent with the study results reported by Nongrum et al. India (53), George et al. in Southern India (35), Silva et al. in Brazil (42), and Bernard et al. in Jamaica (54). Depression and anxiety show frequent co-existence and anxiety may emerge as a strong predictor for depression (24, 37). Mohamad et al. (55) and Edward et al. (56) also demonstrated that anxiety strongly increased the risk of suffering from depression during pregnancy. Even a history of mental illness can pose as a risk factor for depression (57, 58); however in our study only one respondent appeared to have such a history. Marital discord appeared predict presence of depression on bivariate but not on multivariate analysis; other studies report that this is e a well-established risk factor due to its influence on social support (59, 60). Likewise consanguinity seemed to be associated with depression only on bivariate testing. Consanguineous marriages are fairly prevalent in South India and clinical observations have reported a high prevalence of depression in such communities (61) which could be genetically driven (62). A major catastrophic event in the past 1 year was an important risk factor which was significantly associated with prenatal depression in this study; this is consistent with the study results reported by Leigh et al. (63) and Shakeel et al. (58). Another study reported that negative life events may lead to persistent higher levels of depressive symptoms since positive life events can decrease the severity of depression over time (64).

Among the physiologic measurements, anemia was significantly associated with depression on bivariate although not so on multivariate analysis. This is in agreement with the study findings reported by Lukose et al. (65); however Yilmaz et al. depicted the existence of such an association between depressive symptoms and anemia in the third trimester of pregnancy (66). Body mass index was not linked with the risk of prenatal depression in the present study. Research done in other countries reportedly point toward an interconnection between obesity and depression (67, 68). The causal pathway could include inflammation (62), hormonal imbalance (69), or sleep disturbance (70).

STUDY STRENGTHS AND LIMITATIONS

This study focuses on prenatal depression which has received less attention than postnatal depression. All the instruments/scales used to measure the study variables had good psychometric properties. Our study had few a limitations. Antenatal care at such hospitals is mostly availed by pregnant women from the lower and middle—income groups in a community. Hence the findings from this study cannot be extrapolated to pregnant women belonging to the high income group as there could be variations in the psychosocial factors and standard of living. As a part of the cohort study protocol, we excluded women with high risk pregnancies and those with a history of intake of steroidal medication over the past 1 year; this could limit the generalizability of the study findings. Adverse obstetric complications during pregnancy can modulate the mental health of a woman during pregnancy. In the south-east Asian context, conflict with in-laws is also a significant risk factor, although this item was not recorded in the present study but will be included in future data collection. We used the EPDS scale which is a self –reporting screening measure for identifying women at risk for depression. Even though EPDS has a high sensitivity and specificity and can be easily administered by a trained health worker, it is important to confirm the presence of depression by using a structured clinical interview to confirm diagnosis.

CONCLUSION

The present study showed a high prevalence of prenatal depression which is suggestive of its public health importance in the study region. Spouse physical and sexual violence, pregnancy related anxiety and a history of catastrophic events were important predictors of prenatal depression. Obstetric practice should include screening and diagnosis of prenatal depression as a part of routine antenatal care in low and middle—income countries.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of the Ethical Committee of Indian Institute of Public Health Bangalore campus (IIPHHB/TRCIEC/118/2017) with written informed consent from all subjects. All subjects gave written informed consent in accordance with the Declaration of Helsinki. The protocol was approved by the Ethical committee of Indian Institute of Public Health Bangalore campus.

AUTHOR CONTRIBUTIONS

AN and MK: conceptualization. BS, SV, and JV: formal analysis. AN and GM: funding acquisition. CM: methodology. AN and SB: writing-original draft preparation.

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The Impact of the "Semente" Program on the Family-Focused Practice of Mental Health Professionals in Portugal

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Background: Children of parents with mental illness (COPMI) are a high-risk group. There is a strong association between parents' psychiatric disorders and the incidence of psychopathology in their children. These children need to be identified and supported by mental health workers early, and hence, clinical practice in adult mental health services needs to change from a focus on individual patients to a more systemic family focus. "Semente," a mental health promotion program developed by the Psychiatry Service of Fernando Fonseca hospital (Lisbon, Portugal), had been established to identify these children and families and promote their mental health, by decreasing the impact of risk factors and promoting protective factors. The program included preventive COPMI interventions and implementation of activities offered to families with children in the mental health care. The aim of the present study was to evaluate changes in mental health care after the training in "Child Talks" intervention (two to three psycho-educational meetings with parents and children) and implementation of the "Semente" program.

Methods: Participants ($N = 51$) were all professionals from Psychiatric Service of Fernando Fonseca Hospital who received *Child Talks* training. The Family-Focused Mental Health Practice Questionnaire (FFMPQ) was used to measure change in professionals' attitudes, knowledge, confidence, and organizational structure in working with these families. All participants filled in the questionnaire before training and 10 months later.

Results: The results showed that, from pre- to post-measurement, the professionals changed clinical practice significantly. The largest changes were visible in the improved provision of support at the workplace for family-focused practice and the clarity and availability of the policies and procedures. Furthermore, the skill and knowledge of the mental health workers showed significant improvement at posttest.

Conclusion: The positive results of this study were not unexpected; the training, implementation of routines, and procedures as well as workplace support were aims of the "Semente" program. Interpretation of the results should be taken with caution because of the small sample and the lower reliability of some of scales of the FFMPQ. The results indicate that professionals moved from patient focus to family focus during the implementation of the "Semente" program.

Keywords: family focused, mental illness, parenting, implementation, children of parents with a mental illness (COPMI)

INTRODUCTION

Children of parents with mental illness (COPMI) are a high-risk group in society. About 15–23% of children live with a parent with a mental illness worldwide (1, 2). Recent research has demonstrated a strong association between parental psychiatric disease and a higher psychiatric risk in their children (3, 4). About one-third of these children experience serious mental health problems when they grow up, and another one-third experience more serious and long-term adjustment issues (2, 5). To illustrate, a prospective cohort study of 256 children who have a parent with depression or anxiety showed that 38% had developed an affective or anxiety disorder by age 20. At 35 years old, about 65% had depression or an anxiety disorder (6).

In the last decades, many studies showed adverse outcomes in children of parents suffering from mental illness, including negative effects on children's social, behavioral, emotional, and cognitive development (4, 7–9). Moreover, school failure and deficits of social competence at a young age are very common (10–12), sometimes followed by poor functioning at work and in intimate relationships later in life (13) and are at higher risk for suicidal behavior (14).

Mental health stigma and social isolation of these families have contributed to the lack of attention to these children. It is also a population with high genetic and social risk, namely, transmission of mental disorders from parents to children, inheritance of social environment framework of the family, neglect, child abuse, and dysfunctional family interactions associated with the parental psychopathology (3, 8, 15, 16). These children and adolescents frequently suffer in silence from the problems of their parents' mental illness. Several studies have demonstrated that they often experience social isolation. Their social contacts could be limited because of their parents' poor functioning. Frequently, the child takes over a parental role in the family. On the other hand, stigma that involves their parents' mental illness can create negative feelings, such as shame and guilt. Because of that, these children tend to not communicate about their emotional experiences at home (17).

Given this scenario, it becomes evident that preventive intervention is clearly needed to minimize the adverse effects of having parents with mental illness. In their review on interventions to prevent emotional problems in COPMI in the Netherlands, Van Doesum and Hosman (18) postulated that many of the identified risk and protective factors may be sensitive to modulation following exposure to preventive interventions. They pointed to a range of interventions that focus specifically on parents and entire families: making them aware of the impact of the home situation for the children, supporting and informing them, improving the quality of parent–child interaction, and facilitating external social support for children and parents. In Australia and several countries in Europe, prevention programs for children of parents with a mental illness have been developed and are considered a high priority in public mental health policy. Internationally, there is increasing evidence demonstrating the effectiveness of a number of interventions to prevent COPMI from developing problems themselves [e.g., Refs. (7, 19–21)]. However, implementing such interventions requires a systematic approach to changing the behavior of adult mental health service practitioners (22). Children of mentally ill parents need to be identified and supported in adult mental health care in an early phase, and hence, clinical practice in adult mental health services needs to change from a patient focus to a family focus.

However, implementing changes in the practice of adult mental health care to facilitate identification and support of children of mentally ill patients can be challenging, and this large population of high-risk children remains overlooked. There is a lack of natural or systematic communication between psychiatrists treating ill patients and child psychiatry services. Professionals who are treating the parents do not often inquire about the development and mental health of their children. Similarly, pediatric psychiatric teams rarely work with parents' clinicians, even though both groups of mental health professionals are aware that chronic mental disorders burden families (23).

Nevertheless, according to a systematic literature review by Siegenthaler and colleagues (24), interventions to prevent mental disorders or psychological symptoms in COPMI appear to be effective and decrease the risk of developing the same mental illness as the parent by 40%. A current meta-analysis of Thanhauser and colleagues (25) confirmed significant but small effects on preventive interventions for COPMI. They found small significant effect sizes enhancing mother–infant interaction and small effects on global child's psychopathology as well as internalizing problems in adolescents. Interventions addressing parents and children together produced overall larger effects. Research on family-focused practice showed that recognition of the benefits of including families in mental health care is increasingly growing (26–28). It is being reflected in governments' policies and standards, and these interventions are an effective response to these families when collaboration with a range of service sectors and professions is included (26, 29, 30). A continuum of family-focused activities for mental health professionals is recommended when working with parents with mental illness, ranging from assessing their parenting competence to providing appropriate information and resources to family members to providing a liaison with other services to give the family support as needed (31). In a recent meta-analysis, Foster and colleagues (26) identified six core, interrelated family-focused practices: assessment; psychoeducation; instrumental, emotional, and social support; family care planning and goal setting; as well as liaison between families and services. They found that the most commonly described feature of family-focused practices in mental health services was psychoeducation. According to Reupert and Maybery (32), it is essential to develop the most appropriate interventions. They argue that COPMI may be educated about their parent's mental illness through bibliotherapy, peer support groups, and individual consultations with mental health professionals and family consultations with mental health professionals.

It is clear that receiving psycho-education about the parent's mental disorder, understanding the reasons for changes in behavior, and knowing and talking about what is happening within the family is important for children as well as for the parents (7, 27, 33). Communication in the family not only encourages family problem solving but also has been proven to build both child and family resilience. One of the interventions focused at the family is *Child Talks*, developed in the Netherlands (18, 34). It is a brief intervention that was developed so professionals can offer support when their patient has children. Adult mental health workers are trained in talking to parents and children, and they offer this intervention as part of the treatment (34). *Child Talks* consists of three conversations with the parents and the children together and focuses on children in the family, parental concerns, and how parental mental health problems may have an impact on children. It also includes a talk with the children and parents together. It is designed as a basic offer for all parents with mental illness and/or substance abuse and can be used as a standard procedure to talk with parents and children in the adult mental health care. When a parent is registered in the adult mental health care, *Child Talks* is offered during the intake meeting by the therapist of the parent or by a child responsible person specially appointed to talk to the family. The family sessions are offered at the clinic or during home visits. *Child Talks* is implemented and adopted in northern Norway (16). Regarding this intervention, Reedtz and Lauritzen (35) found in a retrospective study of electronic patient journals in the University hospital in north Norway that implementing *Child Talks* in adult mental healthcare may lead to clear identification of patients who are parents and more referrals of children in need of more extensive interventions (N = 5,268 in 2010, N = 5,599 in 2011, and N = 5,705 in 2012).

Nonetheless, the implementation of these interventions requires a great deal of involvement of professionals. Lauritzen and her colleagues (36) evaluated the factors that may facilitate or hinder a family focus in the treatment of parents with mental illness in Norway in a sample of 219 professionals, and concluded that professionals with additional training in family-focused approaches to treatment, such as *Child Talks*, are found to be more open to and active in pursuing a child-focused clinical practice. To evaluate the professionals' perceptions about their own practice, Maybery et al. (37) designed the *Family Focused Mental Health Practice Questionnaire (FFMHPQ)*. The authors compared different groups of professionals in terms of their use of family-focused practices. They concluded that "where more intensive intervention is required for these families, the professionals involved will need training in relation to the evidence-based interventions that are known to be effective with this target group." Given this, it is recommended that all adult mental health professionals should receive workplace training in basic family-focused practice (30).

The Semente Program

"Semente" is a mental health promotion program developed by the Psychiatry Service of Fernando Fonseca Hospital, a general hospital in the suburbs of Lisbon (Portugal). It was established with the financial support of EEA grants,[1] with the objective of identifying COPMI and supporting them with preventive interventions. The program is intended to implement new practices focused on prevention, and targets children and families with parental psychopathology. Being responsible for the treatment of patients with mental disorders, the Adult Psychiatry professionals have easy access to these children through their parents who are also patients.

More specifically, the "Semente" program aims to promote children's mental health and prevent the development of later mental health problems in this vulnerable group, by early identification of these children and improving parental competencies in parents with psychiatric disorders, as well as providing treatment of psychiatric symptoms in the children by the Child Adolescent Psychiatry service, when necessary.

The development of "Semente" was based on the organizational model of the Psychiatry Service, which has a main focus on community interventions, and was made possible through the establishment of partnerships with primary health care, public health, the municipalities, child protection services, schools, and several NGOs. This Service is organized in a community-based model to improve the access of patients with mental disorders, reduce stigma, and reinforce support networks for the patients and their families. The service integrates four community multidisciplinary mental health teams, responsible for the treatment of adult patients—mainly those with severe psychiatric disorders—and a Child and Adolescent Psychiatric Unit. There is a close relation and communication between the child and the adult psychiatry teams, which coordinate their work with complex families, through monthly meetings and regular case discussion and supervision.

"Semente" has focused on making mental health workers aware of the vulnerability of COPMI and has trained professionals in a new approach involving the children of patients with a mental illness as an issue in their daily practice. Training on COPMI preventive interventions focused on their needs is of great value to guarantee further implementation of this program, as well as the accuracy of structured interventions. The chosen approach includes the family-focused preventive intervention *Child Talks*. The choice of this intervention was based on reflections on how to reconcile preventive interventions with the great demands of clinical practice in order not to overload professionals. All the

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[1] The EEA (European Economic Area) Grants represent the contribution of Iceland, Liechtenstein, and Norway to reducing economic and social disparities and to strengthening bilateral relations with 15 EU countries in Central and Southern Europe and the Baltics.

professionals from the Adult Mental Health Community teams and from Child and Adolescent Psychiatry team were trained in the *Child Talks* intervention according to the training model developed in the Norwegian project by Reedtz and colleagues (16). The training consists of 1.5 days of lectures and role playing concerning how to talk to parents and children, as well as discussions about barriers and possible solutions.

The Current Study

The overall aim of the present study was to evaluate changes in the self-perception that clinicians have about their clinical practice after the training in basic family-focused practice *Child Talks*. The following research questions were posed: 1) Will training in *Child Talks* change a patient-centered focus into a family focus in adult mental health practice? 2) Will training in *Child Talks* change professional attitudes towards working with patients who are parents? 3) Will training in *Child Talks* improve organizational policy and support? 4) Will training in *Child Talks* improve workers skills and knowledge in working with the whole family?

METHOD

Participants

The participants in the study are the professionals in the adult and child psychiatric facility at Fernando Fonseca Hospital, since these two groups of professionals were involved in the program in order to better evaluate and respond to the needs of all patients' family members.

At pre-measures, 51 mental health workers responded, corresponding to 90% of the total staff (12% were child providers). As described in **Table 1**, the respondents were mostly women (82% of the cases), and their age ranged between 24 and 56 years old. At post-measures, after the training of *Child Talks*, 46 professionals responded, with essentially the same gender ratio (85% women) and age range (between 25 and 57 years old) as the full sample. The retention rate was 90%. Professionals from both adult and child psychiatry participated at both measurement points. The study was approved by the Ethical Health Commission of the Hospital Prof. Doutor Fernando Fonseca, ensuring a good clinical practice.

TABLE 1 | Sociodemographic characteristics of participants at pre- and post-measurement.

	Pre (*N* = 51)	Post (*N* = 46)
Gender		
Women	42	39
Men	9	7
Mean age		
Women	42	43
Men	34	35
Adult psychiatry	45	40
Child psychiatry	6	6
Profession		
Social worker	5	5
Nurse	10	9
Psychiatry resident	12	12
Psychologist	7	6
Psychomotor therapist	3	3
Psychiatrist	10	8
Child psychiatrist	1	–
Occupational therapist	3	3

Materials

Sociodemographic characteristics. A short questionnaire was developed to collect information on gender, age, and profession.

Family Focused Mental Health Practice Questionnaire (FFMHPQ). This questionnaire was used to measure changes in training needs, interests, and attitudes regarding family-focused practice over a period of time. The scale is designed and tested by Maybery and colleagues (37). Family-focused mental health practice is defined by how professionals involve the whole family in treatment and examines the parenting role of the patient. The questionnaire includes 49 items and 16 subscales with statements relating to professionals' knowledge and skills about family issues; their interest in working with children, parents, and families; their perception of organizational policy and support for family-sensitive practice; and their level of family-focused practice undertaken in their work. **Table 2** shows the questionnaire's subscales, with definitions and examples of items. Each item is rated on a seven-point Likert scale (ranging from *strongly disagree* to *strongly agree*). A low score on the subscales suggests a low degree of family focus, and a high score shows high degree of family focus. The measure has a good content and construct validity and generally fair internal subscale reliability (38). The questionnaire was used in several samples with variety of professional disciplines in the mental health care in Australia (e.g., 30) and in a sample of Irish mental health nurses (39). The questionnaire was translated into Portuguese by the project team and back into English to ensure good reliability. Cronbach alphas were computed for all subscales and range from .42 to .81 (see **Table 2**). The subscale “local issues” had an unsatisfactory reliability of Cronbach's α = .26 and was hence excluded in the results.

Procedure

Training. In July 2015, the whole staff was trained in the *Child Talks* intervention by a trainer from the Netherlands; a supervision meeting with the trainer followed in November. After the training, everyone was able to use *Child Talks* in their practice. The evaluation of the training was carried out through the administration of a qualitative questionnaire, where questions were asked about the global satisfaction with the training, the quality of the content, the structure and the organization of the training, the clarity of the objectives, theoretical background and aspects of application of the intervention, some aspects of motivation to use *Child Talks*, perceived confidence in working with children and families, and overall opinion of this intervention. The participants answered on a scale from 1 (Very dissatisfied) to 5 (Very Satisfied), and responses ranged from 4 to 5—participants considered the training overall very well organized, useful, a way of increasing competencies in relation to COPMI, and very good initiative to improve communication

TABLE 2 | Subscales of the FFMHPQ, definition and item examples (retrieved from 37).

Cronbach’s alpha	Subscale	Definition	Example item
.66	Workplace support (2 items)	*The workplace provides support for family-focused practice*	My workplace provides supervision and/or mentoring to support workers undertaking child-related work in regard to their consumer-parents
.78	Time and workload (3 items)	*Time or workload issues regarding family-focused practice*	There is no time to work with families or children
.50	Policy and procedure (2 items)	*Family focused policy and practices are clear at the workplace*	Government policy regarding family-focused practice is very clear
.53	Professional development (2 items)	*There are opportunities for professional development regarding working with families*	My workplace provides little support for further training in family-focused practices
.42	Coworker support (2 items)	*The support from other workers regarding family-focused work*	I often receive support from co-workers in regard to family-focused practice
.47	Family and parenting support (5 items)	*Providing resources and referral information to consumers and their families*	I regularly provide information (including written materials) about mental health issues to the children of consumer-parents
.53	Work confidence (3 items)	*The level of confidence the worker has in working with families, parents and children*	I am not confident working with consumer-parents about their parenting skills
.71	Support to carers and children (2 items)	*The level of information, advocacy and referral provided to carers and children*	Rarely do I advocate for the carers and/or family when communicating with other professionals regarding the consumer-parent’s mental illnesses
.54	Engagement issues (3 items)	*The opportunity for engagement with family members*	Discussing issues for the consumer parent with others (including family) would breach their confidentiality
.79	Assessing impact on child (2 items)	*How well the worker assesses the impact of the parent illness on the children*	I am able to determine the developmental progress of the children of my consumer-parents
.81	Training (4 items)	*Worker willing to undertake further training*	I would like to undertake future training to increase my skills and knowledge for working with the children of consumer-parents
76	Skill and knowledge (5 items)	*Worker skill and knowledge regarding impact of parental mental illness on children*	I am knowledgeable about how parental mental illness impacts on children and families
.44	Service availability (2 items)	*There are programs to refer families to*	There are no parent-related programs (e.g., parenting skills) to refer consumer-parents to
.69	Connectedness (3 items)	*Workers assessment of parent awareness of child connectedness*	I am able to determine the level of importance that consumer-parents place on their children maintaining strong relationships with other family members
.58	Referrals (2 items)	*Referring family members to other programs*	I refer consumer-parents to parent-related programs (e.g., parenting skills)
.52	Interdisciplinary cooperation (4 items)	*Workers possibility to cooperate with other professionals*	Children and families ultimately benefit if health professionals work together to solve the family’s problems

FFMHPQ, Family Focused Mental Health Practice Questionnaire.

between parents and children, and also indicated that the management supported implementing *Child Talks* in the future.

Practicing *Child Talks*. The patients with children under 18 years old were gradually identified during regular consultation at community mental health teams. An administration form was developed so that mental health workers could identify these families, and refer them to *Child Talks* intervention. During interventions, the professionals filled a logbook of the family meetings. Although *Child Talks* was not initially designed to be delivered by two mental health workers, the project team decided that their meetings with parents and children should be done by two persons, one of them the professional being the professional who regularly provides treatment to the patient. Between two and three sessions were offered to the families who agreed to participate. The fidelity was checked by a supervision meeting with the trainer, and all cases were discussed as part of the monthly staff meetings in the psychiatry service of hospital Fernando Fonseca.

Measurements. The FFMHPQ was distributed by the coordinating team of “Semente” before the training on *Child Talks* (pre-test) to all the mental health professionals of the Psychiatry Service. The second measurement was performed 10 months later when *Child Talks* was already implemented in the mental health service (Posttest).

Data Analysis

All statistical analyses were performed with SPSS (version 25). Descriptive analyses were used to explore the demographic details of the groups. A dependent sample t-test was used to test the difference between the pre- and post-implementation phase. Cohen’s d was calculated to express effect sizes. A Cohen’s $d = .80$ is a large effect, $d = .50$ is a medium effect, and $d = .30$ is a small effect.

RESULTS

Change in Family Focus

In general, the results indicated that, from pre to post, the professionals of the mental health care in the Fonseca Hospital changed their perception about clinical practice significantly by increasing their competencies and moving their focus to the children of their patients (see **Table 3**). Overall, medium to large changes were seen in 11 of the 16 scales of FFMHQ.

Professional Attitudes Working With Patients Who Are Parents

Results showed moderate positive changes (Cohen's *d* between .49 and .59) in the professionals' perception of their clinical practice regarding the way they address family and parenting, and in their self-confidence in working with the whole family. No changes were visible in the engagement with family members, neither in support to caregivers, children, nor other engagement issues.

Organizational Policy and Support

The organizational policy and support improved significantly, especially the provision of support in the workplace related to family-focused practice (like supervision), as well as the clarity in and availability of policy and procedures (Cohen's *d* between .79 and .81). A smaller change was found in the opportunity for professional development regarding working with families (Cohen's *d* = .50). No changes were found in time available, workload, or coworker support.

Worker Skill and Knowledge

Skills and knowledge of the mental health workers related to the impact of parental mental illness on children improved significantly at posttest (Cohen's *d* = .90). Moderate positive changes were found in the improvement of the mental health workers' ability to assess the impact of the parental illness on the children; the professionals believe in making services more available and that they increased the number of referrals Cohen's *d* between.49 and.59. For interdisciplinary cooperation and training, both subscales declined significantly at posttest. The means of these subscales were already high both in pre- and post-training—both areas most valued by professionals, with the highest scores for all the subscales in post-test. No effect was found in the importance of parents' awareness of child connectedness, e.g., relationships with family members and friends.

DISCUSSION

The findings of this study showed that the self-perception about clinical practice in the mental health care of the participating hospital changed from pre- to post-measurement. During this time, professional received training in *Child Talks* and implemented new routines. Consecutively, we found evidence of change in each of the four research questions. 1) Based on the overall improvement in family-focused practice after 10 months, we interpret the findings of the study such that the patient focus of the professionals changed into an approach where they focused more on the whole family. 2) Furthermore, the improvement of professionals' self-confidence in working with the whole family is evident in the professional attitudes in working with patients who are parents, and these attitudes changed in a way where they address family and parenting to a higher degree after training and implementation. 3) The organizational policy and supports showed improvement as the provision of support for family-focused practice in the workplace increased. Also, the clarity

TABLE 3 | Paired t-tests of difference in outcome of subscales FFMHPQ between pre- and post-training.

	Pre-test		Post-test		*t*	Cohen's *d*
	N = 51		*N* = 46			
	M	*SD*	*M*	*SD*		
1. Organizational policy and supports						
- Workplace support	4.37	1.30	5.32	1.04	−4.07***	0.81
- Time and workload	3.94	1.42	3.63	1.22	0.60	0.31
- Policy and procedure	2.91	1.05	3.76	1.10	−4.63***	0.79
- Professional development	5.08	1.23	5.64	0.99	−2.36*	0.50
- Coworker support	5.04	1.01	5.11	0.88	−0.30	0.07
2. Working with patients who are parents						
- Family and parenting support	3.29	0.79	3.69	0.86	−2.66**	0.49
- Work confidence	4.31	1.04	4.77	0.93	−2.49*	0.46
- Support to carers and children	5.29	0.85	5.44	0.79	−0.91	0.18
- Engagement issues	4.41	0.80	4.40	0.70	0.28	0.01
3. Worker skill and knowledge						
- Assessing impact on child	4.11	1.37	4.75	1.20	−2.46*	0.50
- Training	6.07	0.68	5.64	0.85	2.54*	0.55
- Skill and knowledge	4.00	1.06	4.90	0.92	−4.86***	0.90
- Service availability	3.52	1.20	4.16	1.27	−2.10*	0.52
- Connectedness	4.82	0.97	5.12	0.91	−1.88	0.32
- Referrals	3.26	1.33	4.00	1.19	−2.63*	0.59
- Interdisciplinary cooperation	6.26	0.57	5.98	0.58	2.09*	0.49

$p < .05$; $^{**}p < .01$; $^{***}p < .001$ (two-tailed test).

and availability of policy and procedures, and the opportunity for professional development regarding working with families were strengthened. 4) Worker skill and knowledge improved as the awareness about impact of parental mental illness on their children and the way to assess this impact on the children increased. Furthermore, the professionals started believing that they could make services more available and that they could increase the number of referrals. According to interdisciplinary cooperation and training, both subscales significantly declined at posttest, showing that there was less need for training and less need for interdisciplinary cooperation.

Although the results of the study are mainly positive, one should be careful with the interpretation of the results. First, the results of this study were not unexpected, as the training and implementation of new routines and procedures, as well as provision of workplace support, were the aims of the "Semente" project. As extra efforts (time, training, and finance) were available during the 10-month period, this was not surprising. Even though some large changes were found in organizational policy and professionals' skills and knowledge, the changes in clinical practice and the actual work with children and parenting perspectives in patients were smaller. The question is whether the changes in clinicians are sufficient and sustainable over time. The short-term changes in the project can be seen as the result of initial implementation stage, but long-term changes in full operational stages in the innovative project will also need to be evaluated (40). Therefore, the sustainability of the implementation needs to be monitored in the subsequent years. The stage where new interventions are installed and put into use by all staff as a regular service is the most challenging stage of implementation, and according to implementation theories and experiences, it requires 2 to 4 years' time (40). The reasons for this are multifaceted, but for example, skilled mental health workers and other well-trained staff will leave and must be replaced, funding and leadership might change, external systems can change, and new or other social problems arise. Research on the implementation of the same intervention in adult mental health services in Northern Norway showed a significant increase in workers' identification of COPMI between pre- and post-measurement (22), but only a minor, nonsignificant increase at 5-year follow-up measurement (41). In terms of family conversations, the results indicated that there had not been any significant increase in the employees' experience with family conversations during the implementation process (2010–2015). These studies demonstrate that even though interventions can be utilized to prevent and reduce the risk that children of patients develop mental illnesses themselves, implementation of new practice is time and resource consuming. In order to succeed in implementing permanent practice changes, sufficient resources such as time, personnel, and training/supervision must be available to the workforce continually (41).

The second reason to interpret our positive result with caution was that changes were not found in all subscales of the questionnaire. One explanation is that the means of the subscales "Support to caregivers and children" and "Coworker support" were already high at pre-test. The scores on these variables were comparable to a sample of Australian Psychiatric nurses ($M = 5.39$, $SD = 0.84$; 39), and hence, the professionals might already give support to caregivers and children and were possibly working together with colleagues on these matters. Another explanation could be that they gave desirable answers before training, as they already knew the importance of including the children of their patients in their regular clinical practice. The whole staff had been informed about the "Semente" program 2 months before training.

The third reason to interpret the results from this study with caution is that the results are based on one measure, a self-report of how the professionals view their family-focused practice. This might not reflect their actual practice, as have been found in other studies (42). Other data should also be collected to find out how many patients with children are identified and in how many cases *Child Talks* is offered (22).

The fourth reason important to acknowledge to be cautious is the lower reliability (α between .44 and .58; see **Table 2**) in 5 of the 10 scales that showed improvement at posttest. We have decided to leave them in because the original scales had satisfactory internal consistency (37). It might be that due to the small sample size of the study, the lower reliability rates were also found in an evaluation study using the questionnaire in a smaller sample (43).

Nevertheless, positive changes took place in the level of knowledge and awareness in the professionals at the hospital, and these changes may provide a solid platform for sustained changes in the focus of clinicians, as well as necessary changes in clinical practice.

Limitations

This study did not include a control group; this was outside of the scope of the project. Adding a comparison group in a future research might show a clear relation between providing training in COPMI interventions and improvement in family focus practice in the workforce.

Although the FFMHPQ had been proven to be valid and has a reasonable reliability (38), the reliability was poorer in all the subscales in this study. One of the scales (Location issues) had very a low reliability score, Cronbach's α .26, and was excluded. In addition, the reliability scores of all scales were lower compared to the results of Maybery and colleagues (37). This might be due to the smaller group of subjects in the present study and that the translated version of the scale was not tested for reliability in larger groups in Portugal. We recommend to test the Portuguese questionnaire in a larger sample to make it more reliable in detecting changes in family-focused practice. In the further psychometric development of the measure, Maybery and colleagues (38) report also poor Cronbach alphas in the three scales: Location and engagement issues, and support to carers and children. Future research using the FFMHPQ is necessary to improve the reliability of the weaker items.

Furthermore, the time period between pre and post was only 10 months. This is probably too short to see sustainable practice change. The reason for 10 months post-measurement was due to the lack of project funding after the initial training and implementation. We recommend follow-up measurements to evaluate changes after posttest measures.

Also, as mentioned above, the results are based on self-report; this is a limitation because it does not reflect the actual family-focused practice. More aspects need to be monitored like organizational structure, procedures, and support of the management and also experiences of the families who have received the *Child Talks*.

Implications and Recommendations

The results from this project indicate that specific training in COPMI interventions may be a way to enable the workforce to move from treatment to prevention and from a patient focus to a family and children focus. Further research with a comparison group is recommended to show that the changes in the professional attitudes are related to training in COPMI interventions. Goodyear and colleagues (27) showed in their study on the workforce in adult mental health care in Australia that practice-based training in family focus was the strongest predictor of family-focused practice. The fact that this program has been developed in a Psychiatric Service is of great relevance to its acceptance and feasibility for the patients, as it is delivered by the community psychiatric team with whom they have a close relationship. Although this is a prevention program for the whole family, it also contributes to reinforce the patient alliance with their mental health professional. The collaboration and the experience of working together in Child and Adult Psychiatry, as well as the regular communication with partners in the community, is interpreted as facilitating factors in the ongoing change process. These factors were prerequisites to the successful provision of support network for families dealing with psychiatry disorders and their children.

Besides introducing *Child Talks* in mental health service supports in the implementation of family focus in the adult health care, further research should evaluate the effectiveness of this brief intervention.

The ethical questions related to the relevance of intervening, as opposed to doing nothing for these children, prompted the professionals who were planning the project to implement a sufficient strategy by including preventive interventions in the mental health care. Given this, we strongly recommend further training of mental health professionals in order to strengthen this new approach from treatment to prevention.

ETHICS STATEMENT

The Ethical Health Commission of Prof. Doutor Fernando Fonseca Hospital approved the project. They determined that written informed consent was not required for this study.

AUTHOR CONTRIBUTIONS

All authors contributed to the paper in having read and commented on the manuscript text. KvD, TM, CP, ML, CL, and CR put the paper into writing. TM, CP, ML, JM, and LT collected the data. All authors approved the final version of the paper.

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Overcoming Clinician and Parent Ambivalence: General Practitioners' Support of Children of Parents With Physical or Mental Illness and/or Substance Abuse

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Children who are next of kin to parents with physical or mental illness and/or substance abuse need access to mental health support and several cost-effective interventions are available. Because most parents in the target group often consult general practitioners (GPs), GPs may have a crucial role in identifying burdened children and ensuring their follow-up. However, this important topic has received little attention in clinical discussions and research. In response to the knowledge gap, we conducted the research project Burdened Children as Next of Kin and the General Practitioner. Four sub-studies have been completed and published: a sub-study with qualitative analysis of focus group interviews with GPs (paper 1), a qualitative analysis of focus group interviews with adolescents as next of kin (paper 2), and a qualitative analysis of individual interviews with parents with illness and/or substance abuse (paper 3). The results from these sub-studies were incorporated in a survey sent to members of a nationwide GP organization (paper 4). The aim of the present sub-study was to gain further knowledge about conditions for the encounters between GPs and parents with impairments to be supportive for the children as next of kin. The material of the present sub-study derived from the project's four previous sub-studies and comprised a secondary analysis of the four prior sub-studies. We conducted an overarching thematic analysis of these sub-studies' results sections. We searched for statements from the GPs, the adolescents, and the parents on their experiences and evaluations of the needs of the children and their families, and the possible ways of accommodating these needs in general practice. The analysis shows that both GPs and parents were ambivalent about addressing the topic of the patients' children during consultations. This was the case although the GPs were in a good position to identify these vulnerable children, and the parents were worried about their children's situations. Possible strategies for GPs to overcome this ambivalence can be to (1) strengthen their competence in the topic, (2) gradually build trusting relationships with parents, and (3) gradually gain contextual knowledge about the families' situations. GPs can do this by performing ordinary GP tasks and acknowledging the parents' efforts to give their children good daily lives.

Keywords: children as next of kin, parents with impairments, general practitioners, health prevention, health promotion, qualitative research

INTRODUCTION

Approximately 20–30% of children 0–18 years at some point in time experience a parent with physical or mental illness and/or substance abuse (1–4). While the prevalence of children living with parents with illness and/or substance abuse varies, as the definitions of these parental problems are often not the same, the negative effects that this situation can have on the children are clear. These children are at risk of poor psychosocial outcomes and health problems (5–7). In addition, the children describe difficulties in their daily lives, health problems, school problems, loneliness, and instability in the family situation (8–13).

During the last few decades, children's legal rights have been strengthened (14). In Norway, a law was enacted in 2010 concerning care for children as next of kin to parents with severe physical illness, mental illness, and substance abuse. Health personnel who have parents in treatment are obligated to inform their children (with their parents' informed consent) about their parents' situations and ensure follow-up if necessary (15). The same year, Sweden enacted an identical law (16).

Families affected by illness and substance abuse face many challenges that can be exacerbated by stigmatization, parental prognosis, exposure to violence and trauma, and the impairment's impact on the family economy. Children as next of kin to parents with the abovementioned problems experience varying risk and protective factors, and the impairments have different impacts on their lives and developments. Despite these differences, the children have in common, at least in periods, that their parents often struggle to fulfill their parental tasks and give their children the developmental support that they need (17, 18). They are children at risk but are often described as "invisible" in public life and to support services, and thus are difficult to reach for health promoting and prevention (17, 19). Identifying these children and giving the family necessary support is an important preventive task. Evidence-based interventions are available (7, 20–22), including home visits, individual and group sessions for parents, support groups for children, and family intervention programs. However, there is a need for more high-quality studies on the effectiveness of these programs and how they adapt to different children and family situations (23, 24).

A study from the United Kingdom (UK) reported that 23% of children between 9 and 17 years of age with mothers with depression met the criteria for a DSM IV diagnosis. However, only 33% of the children with a diagnosis had been identified and were in treatment (25). The mothers were mainly recruited from general practices. The authors called this a situation of missed opportunities because the professionals in contact with the mothers could have identified these children and offered them appropriate follow-up. It was our belief that general practitioners (GPs) might be in a good position to change this situation. Most patients with parental responsibilities who suffer from mental illness, severe somatic illness, and/or substance abuse will repeatedly consult a GP, often the same GP, several times over the years (26). Although this topic has been raised in clinical discussions and research (27–29), there is still a knowledge gap concerning how the GPs could support patients' children as next of kin. Therefore, we conducted the research project Burdened Children as Next of Kin and the General Practitioner. The results of four sub-studies have been published (10, 30–32).

In these four studies, we found that the GPs were in a good position to identify the children, but they experienced substantial obstacles to ensure them follow-up care. Parents and adolescents, however, wanted the GPs to address their family situations in the encounters. This had to be enacted within a trusting relationship where parents and adolescents felt that their struggles to manage their challenges were recognized.

This article presents the results from the fifth sub-study of the project. This is a thematic analysis of the four sub-studies' result sections. The aim is to gain further knowledge about how GPs can take on a child-focus and support children as next of kin when their parents with illnesses and/or substance abuse seek the GP for their own health problems.

Context and Setting

In Norway, where this project took place, general practice provides an open access for people with all types of health-related problems, and the GPs are often the patients' first medical contact within the healthcare system. GPs cooperate with others in the primary healthcare setting, giving the patients coordinated care. They also act as gatekeepers and coordinate entrance into secondary care by referrals. A patient list system is operational, and almost all citizens are enlisted with a personal GP, which facilitates continuity of the doctor-patient relationship. The children are usually enlisted with their mothers' GP. GPs primarily work at their office in which they receive patients for consultations.

Theoretical Assumptions

The prevailing consultation model among Norwegian GPs is the patient-centered consultation model (33) which encourages the GPs to explore and understand the patient's expectations, background, and feelings. Then, in dialogue with the patient, the GPs combine these insights with the examination-results and his or her medical knowledge. According to this model, the GPs must explore the context, including family matters and the children's situations. However, if the patients for some reason do not want the GPs to address the children's situations, the subject might be omitted even though the GPs ethically and legally have obligations toward the children.

Patient trust is often a precondition for the patient to allow for and engage in difficult conversations about vulnerable themes with a medical professional (34, 35). In a study from Skirbekk et al. on patient-GP consultations (36), trust is conceptualized as the patient's implicit willingness to accept the physician's judgment in matters of concern to the patient. Several studies on general practice concluded that an attitude of recognition from the GP can encourage the patient to share his or her story (37, 38). Here, an attitude of recognition is described as relational, mutual, and based on respect for the person as a subject, as an authority of one's own experiences. This attitude may make it easier for the GP and the patient to tolerate their different viewpoints during the encounter. The concepts of patient-centeredness, patient trust, and patient recognition provide theoretical support for our analysis (39).

As this project's sub-studies were completed, the researchers discussed the implications of the results. GPs mostly meet their patients in a doctor's office. These consultations between the GPs and the parents are the central arena that can allow the GPs to learn about the children's situations and, in collaboration with the parents, ensure follow-up care of the children. Our research suggests that while the GP may be a crucial figure in helping the children of parents with impairments, there are many missed opportunities in practice (31). Therefore, this secondary analysis of our results might deepen the understanding of the possibilities and limitations of these consultations.

MATERIALS AND METHODS

There are four sub-studies in our project considered in the present analysis (10, 30–32) (**Table 1**): a qualitative analysis of focus group interviews with GPs (paper 1), a qualitative analysis of focus group interviews with youth as next of kin (paper 2), a qualitative analysis of individual interviews with parents with illnesses and substance abuse (paper 3), and a survey sent to members of a nationwide GP organization that incorporated the results of the previous sub-studies (paper 4). The current sub-study comprised a secondary analysis of the four prior sub-studies. The results sections of these four sub-studies provide the data analyzed in the present study. The empirical material of this study allows us to combine the perspectives of GPs, youths and parents on how to support burdened children. To reach a deeper understanding of this topic, a thematic analysis (40) is appropriate as it allows for a systematic treatment of the material. This method involves searching the dataset to find repeated patterns of meaning. The analysis is structured and consists of different steps, as described below. During the analysis the researcher continuously compared the codes and themes they developed with the text as a whole to ensure that the results were grounded in the original data set.

After defining the aim, two of the authors (MH) and (FG) read the four articles several times to *familiarize* themselves with the material seen as a whole and recorded their preliminary ideas. Then we searched the results sections specifically for elements that described the informants' experiences and evaluations of the needs of the children and their families, and the possible ways of accommodating these needs in general practice. We marked these elements with *codes*. We found that the text dealt with 37 different codes relevant to our aim. We gathered the textual elements with the corresponding codes. We then determined how the codes from the different sub-studies were interconnected by comparing and integrating them, and then developed preliminary subthemes and overarching themes. Each *subtheme* should cohere together meaningfully, and there should be clear distinctions between them. This process ended with 11 subthemes. We grouped the subthemes under three *overarching themes* that organize the results section of this analysis. The 11 subthemes are used as subheadings (**Figure 1**). To assure the quality of this analytic process, a third author (KvD) compared the four sub-studies with the list of codes and development of subthemes. At this stage, we searched the text for citations that would best illustrate the subthemes. During this analysis, we evaluated the developed codes, subthemes, and overarching themes against the four sub-studies to ensure that they were based on the empirical material.

RESULTS

The adolescents described a lack of knowledge about their parents' situations and that they wanted to be offered follow-up on their often-stressful life situations. During the analysis, we found that their needs were acknowledged by the GPs and the parents, but both groups expressed ambivalent feelings about addressing the children's situations in the consultations.

The major overarching themes and sub-themes derived from analysis are (1) The parents had mixed feelings about receiving support on parenting (sub-themes "Parents acknowledged their children's need for information and to talk about their experiences," "Parents lacked knowledge about support services for their children," and "Parents gave double messages"), (2) GPs often missed their opportunities to support parents and children ("GPs faced obstacles in the general practice framework," "GPs feared jeopardizing the doctor-patient relationship," and "GPs lacked knowledge about talking to the children"), and (3) How can the GP lay the ground for reduced ambivalence of talking about the children? (sub-themes "Recognizing the parents' struggle for an ordinary family life," "Taking the initiative," "Awareness of the therapeutic alliance," "Gaining contextual knowledge and building multidisciplinary networks," and "Building competence").

Overarching Theme 1: The Parents had Mixed Feelings About Receiving Support on Parenting

Parents Acknowledged Their Children's Need for Information and to Talk About Their Experiences

During the focus group interviews, the adolescents indicated that they had incomplete knowledge about their parents' conditions and expected outcomes. This caused worries and uncertainty and made their daily lives unpredictable. They described a

TABLE 1 | Overview of the four articles in the project Burdened Children as Next of Kin and the General Practitioner.

	Aim	Design/data collection	Participants	Parental problems	Analysis
Paper 1	Explore GPs thoughts and experiences with handling the special needs of children as next of kin in general practice	Qualitative interview study/4 focus group interviews	27 GPs, 9 women, 38–65 years of age, 6–33 years in GP		Thematic analysis (40)
Paper 2	Explore significant experiences of adolescents as next of kin that the GP should identify and recognize	Qualitative interview study/4 focus group interviews	15 adolescents, 12 women, 16–25 years of age	5 physical illness 5 substance abuse 5 mental illness	Systematic text condensation (41)
Paper 3	Identify important factors for the GP to bear in mind during encounters with ill and substance-abusing parents to enable the GP to provide appropriate support to the children	Qualitative interview study/Individual semi-structured interviews	12 parents with a total of 28 children, 9 women	4 physical illness 2 substance abuse 8 mental illness	Systematic text condensation (41)
Paper 4	Investigate the experiences of GPs concerning their involvement with their ill patients' children and their evaluation of the opportunity to help these children	Web-based survey with some open questions	499 GPs 244 men		Numeric data analyzed by cross tables, t-tests, chi-square testing and multiple regression. Text material from the open questions analyzed by thematic analysis (40)

daily struggle to balance their own needs for an ordinary adolescence—participating in social activities and focusing on school performances—with the boundaries and burdens caused by their parents' problems. For these adolescents, it was important to have someone to talk to about their family situation and who acknowledged their challenges. This individual could be the healthy parent, a teacher, a friend, a family member, a support group, and someone from the health care system, including the GP. A girl with a mother with mental illness, now living in a foster home, said the following (paper 2):

> It is so nice to talk to some adults who can tell you that this is NOT how you should live. You should not wash the dishes after a huge dinner that you didn't eat. That is not how life should be for a kid. You should be out playing, because it is sunny outside. That kind of information is incredibly important.

The majority of parents recognized their children's lack of information and emotional burdens. However, giving such information to the children was perceived as difficult. Often, due to the parents' own medical condition, they did not sense their children's worries. For example, consider the statement from one mother with severe chronic back pain (paper 3):

> …….because all kids get worried when the mother stays in bed all day, and when they peep into the bedroom, she is laying there crying with pain. Of course, my kids got worried. They were terrified. They thought that I would die. They did not see the difference whether I laid there not being able to move because of back pain, or if I had cancer. For them, there was no difference. I did not manage to sense these worries.

Some parents expressed that their children avoided talking about their parents' problems at home. Often, choosing the best time to inform the children of their problem was difficult. Moreover, the parents were unsure what information was relevant to share. Many parents wanted concrete and individualized advice on how to talk to their children about their situations. For this purpose, they said that a helper close by would be the best person to seek counseling from. Some parents wanted their children to be offered help from professionals.

Parents Lacked Knowledge About Support Services for Their Children

The adolescents described constraints in their lives caused by their parents' impairment. It was important for them to relax and take part in social activities such as going out with friends and joining sports activities. They needed "time-outs." One 16-year-old girl living alone with a father with schizophrenia echoed this sentiment (paper 2):

> I just wanted to have time out with them, my friends, where nobody knew about my dad. I found that relaxing. I didn't want to be pitied for living with him, I just wanted to be seen as an ordinary girl.

In addition, parents missed having relieving activities for themselves and their children, help for daily household concerns, and financial support. They often did not have the energy, social network, or competence to search for support services. Parents wanted their GP to ask questions about their family's needs and to help them to find relevant support. A mother with bipolar disorder who lived alone with two children said the following (paper 3):

> It is important that the GPs have knowledge about where they can recommend us to get help when it comes to the children. Once the doctor knows that we have children, there should be

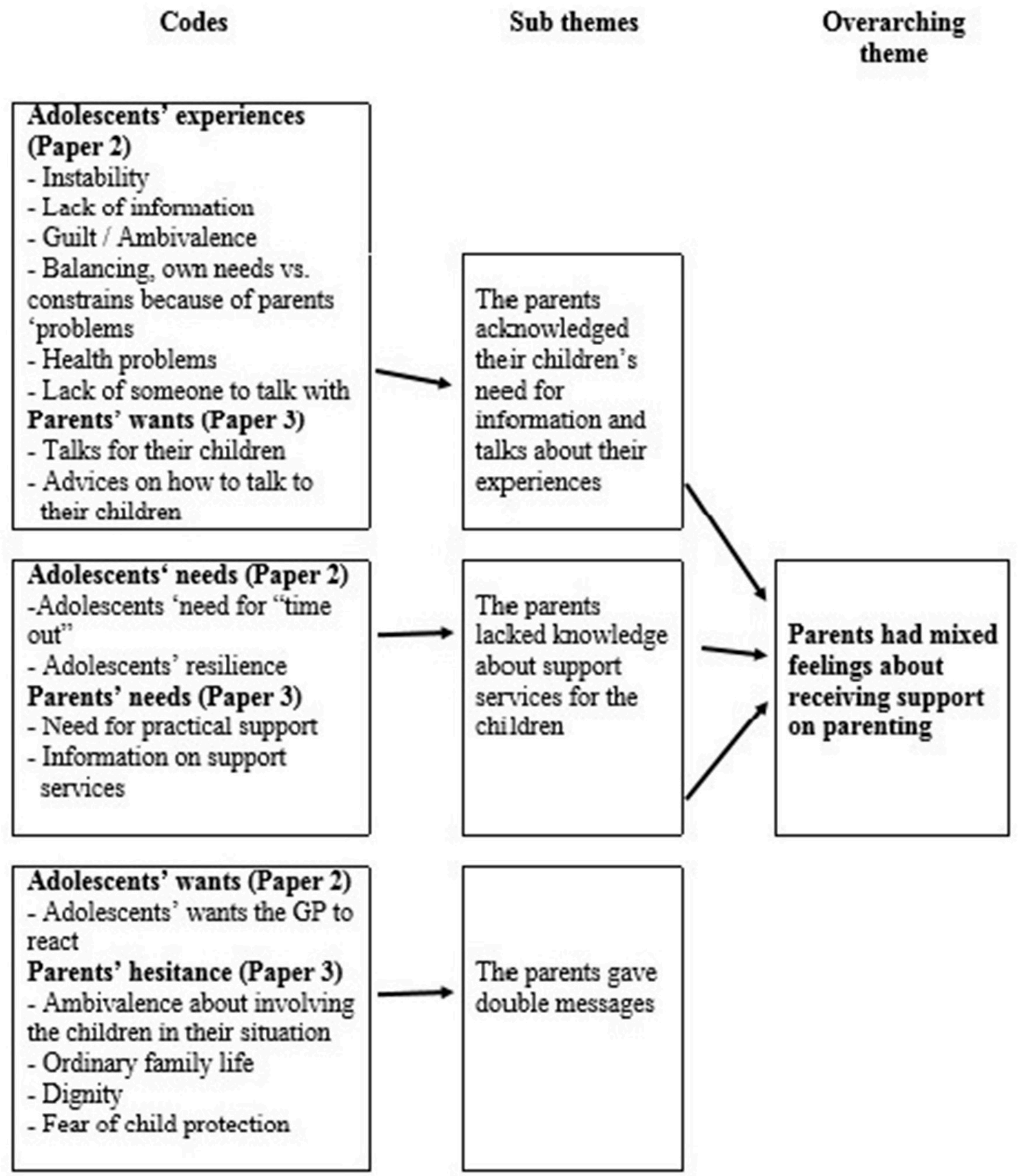

FIGURE 1 | Analysis process from codes over sub-themes to overarching themes. Result section "Patients had mixed feelings about receiving support on parenting" as an example.

> an alarm ringing telling them: "Okay, now these kids need to be protected." The doctor should tell the parents: "I have some advice for you, and some helpers you can contact, and here are the phone numbers," …. a brochure to hand out or stuff—I think that can be very helpful.

Parents Gave Double Messages

The youths often wished for more information and to have talks about their life experiences. In particular, they wanted to know more about their parents' conditions, how to understand their sometimes-deviant behaviors, their disease progressions and help to understand their own life situation and the rationale for their struggles. They wanted someone to talk to about their experiences. The parents acknowledged their children's needs to be informed and supported. In addition, they realized that advice on parenting could be beneficial. Despite this, they revealed ambivalent stances on the topic and gave what could be termed a double message; they both wanted and did not want their children's situation to be a topic in the consultations. Some openly stated that they were afraid of being considered bad parents. Likewise, some feared their children would be taken from them if they revealed the problems at home. Some GPs in the survey noticed this resistance and attributed it to the parents' fear of losing custody. The adolescents, however, wanted the GPs to intervene. For instance, one girl blamed the GP for not contacting the child protection services after a consultation she had attended with her mother who was heavily drunk. Related to this, many parents wanted to preserve their dignity and social acceptance. They described their struggle to keep up the appearance of an ordinary family life. Last, the parents were unsure about how much information should be provided to the children, as knowing too much about their parents' problems could cause their children unnecessary concerns. A single mother with a personality disorder living with an 8-year-old son maintained, as did many parents, that keeping up the impression of a normal family was in a child's best interest (paper 3):

> I have simply avoided talking about it. I have been afraid about making our situation abnormal, that he might think we are living differently. That he would be ashamed. For this is what is normal for him.

Overarching Theme 2: GPs Often Missed Their Opportunities to Support Parents and Children

GPs Faced Obstacles in the General Practice Framework

Some GPs knew their patients' children from local communities and from collaboration with the health visitors in the geographic area. Others pointed out that they only had a "peephole" into their patients' everyday lives, and the patients regulated the GPs' knowledge of their families. As one GP said: "It is easy to hide from a GP." Due to the continuity of care, this "peephole" sometimes gradually broadened through knowledge gained over time during ordinary GP tasks.

In particular, the GPs emphasized three factors within the GP framework that hampered their opportunities to address the children's situations during consultations: busy practices with heavy workloads, short consultations, and the registry system where family members could be enrolled with different GPs. Most of the respondents in the survey (paper 4) answered that they felt responsible for their patients' children when they were enrolled on their own list. However, only half of the GPs felt the same responsibility when the child was enrolled with another GP. The following glimpse from an interview between the interviewer (FG) and a GP illustrates this point (paper 1):

> FG: "Does the general practitioner already have so many tasks that this becomes difficult to handle during the workday?"
>
> GP: "I think that's a good point, especially in a situation where the rest of the family is not on your list. Then you think there are other people involved who will take care of them. The rest of the family can be people you don't know and whom you have never seen."

However, these children might have GPs who do not know about their parent's problems. If a child's GP does not routinely ask about his or her parents' condition, this GP may not recognize if the child is at risk. This is often the situation for children of ill or substance-abusing fathers because at birth Norwegian children are automatically enrolled with the same GP as their mother.

GPs Feared Jeopardizing the Doctor-Patient Relationship

Relational constraints—such as focusing on the parents during the consultations and thus, forgetting to address the children—caused problems for the GPs. In addition, the GPs were also concerned about the possibility of hurting or losing their vulnerable patients if they brought up the children's situations. Some GPs said that they avoided the topic because they did not want to add guilt and place burdens on parents who were already struggling. One female GP expressed this notion (paper 1):

> It is difficult, because then it's as though I am also saying that her problem is her children's problem. Then I am putting the blame on her, and here she has come to get help for herself. I am just placing one more burden on her shoulders, I should think.

Similarly, some GPs avoided the topic because they thought they could offend their patients. These GPs viewed that ending a long-term doctor-patient relationship would be a disadvantage for the patient and the family.

GPs Lacked Knowledge About Talking to the Children

The GPs often engaged in discussions about parenting, giving advice to both ill and healthy parents. A female GP told a relevant story about a family with an 8-year-old boy (paper 1):

> The mother had asked me for advice on how to inform her son about the father's drug problem. (....) Then, the father died in an overdose. Afterwards I gave advice on how to tell this son about death and why it happened. Naturally, I also visited their home a few times after he died and talked with the little 8-year-old boy. That was not easy!

The GPs who addressed the children's situations were confident in informing and advising their parents. However, GPs felt more uncomfortable about talking directly to the children.

Overarching Theme 3: How Can the GP Lay the Ground for Reduced Ambivalence of Talking About the Children?

The parents gave a double message on whether to thematize the children's situations in the encounters with the GPs. The GPs said that they often avoided the topic of their patients' children out of fear of placing more burdens on their struggling patients or of losing the therapeutic alliances. However, the data-set contained thoughts and experiences from both the parents and the GPs on how to overcome this mutual ambivalence who might hamper identification of and support to the children.

Recognizing the Parents' Struggle for an Ordinary Family Life

Many parents tried to make ordinary daily lives for their children. Overall, they wanted their impairment to have as little negative impact as possible. Parenthood gave them social belonging and self-respect. Crucially, it sent a message to them and those around them that they managed parenting despite their problems. One mother with substance abuse framed it this way (paper 3):

> For the last 6 months, a woman from child protection has been coming to my home twice a week to take urine tests. In addition, she does an inspection in our home. I wanted it that way. I wanted these people to come home to me, to let them see that we manage just as well as our neighbors.

For these parents, admitting that they needed support threatened their self-image of being a competent parent who managed daily life in an ordinary family. Therefore, before they could admit their shortcomings and collaborate with the GP, many parents needed recognition for their efforts and love of their children.

Taking the Initiative

Despite their expressed ambivalent feelings, parents interviewed in Paper 3 wanted their GPs to address their children's situations. However, the parents did not want to put forward the topic themselves. Rather, they had to be prompted by the GP. The adolescents expressed the same sentiment: They wanted the GP to ask about their family and situation at home during ordinary consultations. The adolescents' expectations of the GP could be negatively formed by their parents' previous medical experiences such as delayed cancer diagnosis or psychiatric diagnoses. The GP might misunderstand this hesitant attitude or ambivalence from parents and children as avoidance or that the topic was not relevant.

Awareness of the Therapeutic Alliance

Sometimes, the GPs worried that their rapport with patients would be affected negatively if they addressed the children's situations (see above "GPs feared jeopardizing the alliance"). The parents, however, told stories of how they had previously tolerated direct speech from a trusted helper, someone whose alternative viewpoints and corrections they could accept. The parents often had a trusting relationship with one professional, a person who had provided continuity of care and demonstrated strong personal involvement. One father who had recently lost his wife to cancer expressed this view (paper 3):

> Support from the GP, a cancer nurse, or health visitor is really important. To have helpers genuinely interested in helping you and not just doing a job because it is their duty to do so. You tell more to a person you know and trust than to a person you see only once. These helpers have been there during the illness. It started with the GP, the GP has been there all the time, and it is there you go if new troubles come up.

Gaining Contextual Knowledge and Building Multidisciplinary Networks

It was important for the parents to have helpers who knew their situation well, including the social and family settings. In addition, the parents appreciated when the GP had adequate knowledge about support services and participated in multidisciplinary meetings. In these meetings, the GP could contribute with valuable information. For the parents, it was important that the different helpers collaborated, as stated by a single mother of two children (paper 3):

> My GP is very active participating in collaborative meetings. Then she gets more information about my situation—more than if she just sees me at her office. In those meetings, we talk about almost everything. It is of great importance that the GP participate. Otherwise, she would have had no insight. I am not that often at the GP's office.

Some GPs emphasized that collaborating with health visitors in preventive child health networks provided them with knowledge about local societies and the families' daily lives. House calls could also give important information and generally made it easier to become aware of the children's situation. Most importantly, however, continuity of care gave the GP gradual insights into the families and their social situations. Some GPs did not participate in multidisciplinary meetings, but those who did experienced that they could also support the parents this way.

Building Competence

In the survey (paper 4), the participant GPs were asked about what would help them to ensure the support of these vulnerable families. In particular, younger GPs and GPs who were not specialists in general practice reported "more competence about children as next of kin." Some of the GPs wanted more training in talking to children about sensitive matters. Notably, many of them did not know about the 2010 law (15) that requires them to, given the parents' informed consent, ensure that children receive information and follow-up. The GPs called for, among other things, net-based courses, booklets, and overviews of the services for families in primary care and social services.

DISCUSSION

The parents and the GPs who participated in the four sub-studies generally accepted that the parents' health problems might have a negative impact on their children's current and future wellbeing, health, and psychosocial adaptation. Despite this, both groups were ambivalent about addressing the children's situations during encounters in doctors' offices, and the topic was often omitted. This analysis suggests recommendations for how GPs can overcome these barriers: namely (1) recognize the parents' good intentions, (2) ask directly about their children, (3) learn more about children as next of kin, (4) learn more about how to talk to children, (5) build a trusting alliance with the parents and rely on it, and (6) participate in multidisciplinary networks concerning these families. Some of these elements are previously described from general practice and specialist health services (10, 29, 30, 42). Our research, however, points out opportunities to incorporate these elements into a clinical context in general practice.

Many authors recommend using a family-focused approach in general practice to reach children in need (26, 29). Based on a qualitative study of GPs in Denmark, Holge-Hazelton and Tulinius (19) defined "cases with a child in need" in general practice as "a case that directly or indirectly involves problems with a specific child, an as-yet unborn child, or one or both parents of a family, currently or potentially threatening the wellbeing of the family and the child." They found that in general practice, most cases with a child in need are found during indirect consultations; indirect in the sense that the child is not present, or the primary cause of the consultation itself may not be the child. This is in accordance with the aim of our study: to indirectly support children as next of kin during consultations with their parents. However, the results from the present analyses suggest that a family focus is a necessary, but not sufficient prerequisite. In addition, the GP usually has to prompt parents to bring up the children's situations and overcome the ambivalent feelings both in themselves and the parents.

Our results showed that the GPs were often unsure whether the parent tolerated their inquiry into family matters. Their concern was that some parents might think this topic was not a GP's business, be offended, leave the office and—in the worst case—leave their patient registry. Broholm-Jorgensen et al. (43) recently published a study on GPs' strategies for retaining patients during preventive health checks. Opportunistic health checks during consultations for other topics might be provocative for patients. The researchers found that respect was a core element for the GP to succeed in a professional urge to promote better health practices in smokers, overweight persons, etc. They identified two complementary fields of respect: the GP's respect for the patient's autonomy and the patient's respect for the GP's professional authority. If the GP balanced the emphasis on his or her authority with their respect for the patient's autonomy, there was an increased chance that the patient would come back for another consultation. During such consultations, there would ideally be an exchange of mutual respect. Because GPs have ethical and legal obligations to address children of parents with illnesses and substance abuse during consultations with these parents, it is crucial—given the result from the aforementioned study—that GPs treat the topic of children with respect. Trust and recognition might be useful concepts to apply here as well.

The parents wanted information and advice from a trusted helper from whom they could accept direct speech and alternative viewpoints. This lead into the question of how the GP can evaluate the strength and quality of the patient's trust. Skirbekk et al. (36), having studied GPs' consultations, concluded that doctor-patient trust is mostly indirectly, and rarely openly, addressed by doctor and patient. The patient gives the doctor a "mandate of trust" in which the patient sets the conditions for what is an accepted topic during the consultation. This mandate can be limited or broad, and it may change during the consultation. The GP might negotiate the mandate of trust by taking the initiative to talk openly about the patient–doctor relationship. To what extent does the patient trust the GP? What are his or her doubts and mistrust if the topic of the children's situation is addressed? An open talk might disclose that the patient already indirectly has given the GP an open mandate, or that the patient does not have enough trust in the doctor to reveal problems at home that might affect the children. Then these concerns might become a topic in the consultation.

As the study of Broholm-Jorgensen et al. demonstrated (43), recognition may be a crucial aspect of the exchange of mutual respect. When asked about daily family lives, most parents in the present study first emphasized how well they managed their ordinary routines. Later, some revealed difficulties, such as worries about their children and needing support for themselves and their children. As described in the section "Theoretical assumptions," an attitude of recognition allows for the acceptance of another person's experiences and opinions. This attitude has been shown to be beneficial to a doctor–patient relationship in which the patient may gradually accept the doctor's viewpoints. For example, after the parent has told the GP about the efforts that he or she puts into maintaining as ordinary a life as possible for the children and the GP has verbally recognized this efforts, the parent may respond and give the GP trust in return and respect the GPs authority and advice about the children's need for support. However, the GP is responsible for laying the groundwork for this process to develop.

If the GPs manage to overcome the mutual ambivalence to address the family situation during consultations, this might benefit not only the children but also the parents with impairments. In a Dutch interview study of parents with mental health and substance abuse disorders (44), the parents stated that parenthood and the demands of parenting gave meaning and structure to their lives, and thereby provided them with strength. Therefore, if the GPs come in a position to support and coach parents by taking the parents' experiences and goals as a starting point, this will be of help for both children and parents. In the parental interviews (Paper 3), we learned how important their identity as responsible parents was, but also their need for emotional and practical support.

The GPs wanted to build more competence in the topic of children as next of kin, and stated that this could raise their awareness and ensure adequate support for the children. However, the topic is inconsistently referred to in the medical education curriculum and continuing education for specialists in general practice. Therefore, medical schools, governmental health authorities, and the Norwegian Medical Association must take measures to prevent transgenerational transference of psychosocial problems. For example, child-focused and family-focused content should be strengthened in the training of medical students as well as in the continuing education for GPs, including ways to overcome barriers in raising potentially confrontational issues about children's welfare during consultations.

Strengths and Limitations

To our knowledge, the research project Burdened Children as Next of Kin and the General Practitioner is one of very few focusing on the care of these children in general practice. Because GPs are central to the primary health care of the parents in the target groups and their children, our project may act as a basis for clinical work and further research. It is a strength of the project that it combines experiences from all three relevant informant groups: children, parents, and GPs.

The empirical material for this analysis was a text composed of the result sections of the four published articles and not the original raw data from the four projects. This may constitute a limitation regarding richness. However, the aim of the present secondary analysis was to gain further knowledge about the consultation situation, where the main experiences of the actors were juxtaposed and abstracted into salient themes. This bird's eye view of the situation was possible only by looking at the main experiences after they had been identified in the previous sub-studies. That said, there were some limitations in the data collection methods used in the sub-studies. First, the parents were recruited through their GPs, and because of this it is possible that they had more positive experiences with their doctor than informants on average. We have little information from those who could reveal shortcomings in GPs' services, did not have a rapport with their GP, or had their health services covered by specialist services. Second, the material contains no direct information from "invisible" children because the adolescents were recruited from support groups. Some of them, however, told stories in retrospect about their daily lives before they were identified and helped. Third, the results section of paper 4 refers to 499 respondents to a survey sent to ∼6,000 GPs. We can assume that the results then are based on answers from GPs who were more interested in the topic that the average GP (45).

Finally, some interviewer and interpreter biases may have occurred because the interviewers were GPs (FG) and child and adolescent psychiatrists (MH). How the participants presented themselves and their experiences might have been influenced by this fact (46). The GPs may have avoided telling stories about encounters where they felt that they failed and the adolescents and the parents generally avoided sharing bad experiences from encounters with GPs.

Overall, the material possibly contains less information about bad experiences of encounters with GPs where the children's life situations were addressed. We assume that there are more aspects concerning these encounters than our analysis has brought forward. Nevertheless, the suggestions we have provided for GPs to overcome mutual ambivalences during these encounters are valid and can be transferred to general practices in Norway and most likely to comparable GP and health care systems.

Implications and Advice for the GP (Memory box)

- Assume that parents are striving for and want the best for their children. Acknowledge parents' efforts to maintain ordinary daily lives for their children.
- Keep in mind that most of these parents want their children's situations to be addressed, but the parents must be prompted by a GP that they trust.
- Conduct critical evaluation of the doctor–patient relationships with parents. Undertake necessary efforts to obtain a working mandate of trust that ensures openness about the children's situations.
- Expand personal knowledge and skills on the topic, including the local psychosocial networks of families with problems, relevant support resources, and everyday challenges children as next of kin are facing.
- Give priority to and actively participate into collaborations with the other helpers for a specific family.
- Establish contact with support services. Provide children and parents with a list of possible support services for families with children as next of kin.

FURTHER RESEARCH

It is important to continue the work to develop evidence-based guidelines for GPs during encounters with parents with illnesses and/or substance abuse to ensure their children adequate information about their parent's impairments and follow-up if necessary. The following specific recommendations for research are:

1) A web-based survey with a representative sample of Norwegian GPs. The aim of the survey would be to assess (1) the distribution of views and attitudes and current practices related to what most Norwegian GPs consider to be good service to burdened children and their families in general practice, (2) how the expectations of the children and their parents (Paper 2 and 3) can be met, and (3) GPs evaluation of how the preliminary guidelines from the present sub-study can be applied in their practices.
2) A multicenter randomized controlled trial with GPs in Scandinavia. The intervention could be education on the topic of children as next of kin and clinical training in applying the preliminary guidelines from the present sub-study. The outcome measure would be if children of the GPs' patients are identified and offered follow-up. Are there differences between the intervention group and the control group? We plan to include centers in Sweden and Denmark to achieve a sample that is large enough to give statistically significant results. The context for general practice in Scandinavia is fairly equal.

ETHICAL APPROVAL

This study was carried out in accordance with recommendations of the Western Norway Regional Authority and theResearch Council of Norway. The empirical material in the present article stem from our previous four articles in the project. Two of the articles (10, 30) received approval from Regional Committee for Medical and Health Research Ethics, Western Norway (2012/2336-3). According to the Regional Committee for Medical and Health Research Ethics, the Act does not apply to the sub-studies published in the two other articles (31, 32) and the present article. All subjects gave a written informed consent in accordance with the Declaration of Helsinki.

AVAILABILITY OF DATA AND MATERIAL

All audiotapes and transcripts are stored in secure, password-protected storage at the University of Bergen. De-identified transcripts from the interviews may be made available to interested persons or organizations on request to the corresponding author at Marit.Hafting@uni.no.

AUTHOR CONTRIBUTIONS

MH headed the research group Children as Next of Kin and the General Practitioner and had formal responsibility in all stages of the present research process. FG and MH coded the material in the present article. FG and KvD followed the research process closely and commented during the work process. MH, FG, NA, TS-S, and GR worked together on the foundation of the project and had substantial contributions to the four published articles (10, 30–32). NA, TS-S, and GR commented on the last versions of the present article, and all the authors approved it.

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frontiers in Psychiatry

ORIGINAL RESEARCH
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Identification of and Support for Children of Mentally Ill Parents: A 5 Year Follow-Up Study of Adult Mental Health Services

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Background: Children of parents with mental disorders are more likely to develop mental difficulties during their childhood and adulthood. Based on this knowledge, the Norwegian health legislation has been amended to better identify and protect children of parents with a mental illness. In this project, two interventions were implemented in a regional clinic for adult mental health services. These interventions were (i) Assessment Form and (ii) Child Talks. Both interventions aimed to support healthcare professionals in identifying and providing support for children of patients within adult mental health services. The process of changing relevant practice to become more family-focused was evaluated in 2010 and 2013, and the results showed some changes slowly materializing in the adult mental health services. The purpose of the current study was to investigate long-term effects of the interventions at 5 year follow-up (2015). The main aim was to investigate whether the workforce perceived that their clinical practice had changed as a result of the legislative change and the implemented interventions.

Method: This longitudinal study consists of a pre-test, post-test and follow-up test. The sample ($N = 219$ at pre-test, $N = 185$ by post-test and $N = 108$ on follow-up test) included healthcare staff from a participating hospital, responding to an online survey about their routines for identifying children of patients, their attitudes, as well as concerns and expectations related to having a child perspective in their clinical work. Employee experiences with family conversations were also investigated, as well as their knowledge about the consequences parents' mental disorders may have for children.

Results: Our findings showed a significant increase in participants identifying children of patients between pre- and post-measurement but a minor, non-significant increase at follow-up measurement. There was no significant increase of participants who reported that they had a lot of experience with family conversations. From post-test to follow-up, there was no increase in the workforce' reported positive attitudes, knowledge or expectations about the effects of the interventions.

Conclusion: There have been some changes in clinical practice, but it seems that the changes required by law are a very time consuming process. It is necessary to increase the pace of the implementation process.

Keywords: implementation, changed clinical practice, children of mentally ill, mental health care for adults, children's perspective

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INTRODUCTION

Mental illness among parents is a risk factor that can have serious consequences for their children. In the course of the last two decades, findings from a number of studies have indicated that children of mentally ill parents have an increased risk of developing mental illness themselves (1–3). The risk for this outcome has been shown to vary between 41 and 77% (2, 4–7). Children of mentally ill parents are more vulnerable to depression, and compared with other children they are also more likely to develop reduced social skills, behavioral problems, hyperactivity, attention deficit disorders and lower self-confidence in their childhood and youth (8–10). In addition, children of mothers with depression have been shown to score lower on cognitive ability tests and perform worse in school (11). Furthermore, studies have shown that children of mentally ill parents have an increased risk of developing a variety of psychiatric disorders as adults, including depression, anxiety and substance abuse (9, 12).

A large number of children and youth live with parents who are suffering from mental illness, which is linked to the high rates of mental disorders in the population (13). Calculations carried out by the Norwegian Institute of Public Health in 2011 estimated that 410,000 children in Norway live with either one or two parents who have a mental illness, and 115,000 of these children had parents with serious mental illnesses (13). Another Norwegian study has shown that 13% of patients at 24 clinics and 33% of patients at psychiatric outpatient clinics were responsible for caring for children (14). These figures correspond to studies from other countries (15, 16).

It is assumed that the mechanisms that cause mental illness to be transmitted from parents to their children may be due to a number of different factors (17, 18). It is not the parents' diagnosis in and of itself that transmits this risk, but rather an interaction between factors such as disease severity and duration, genetics, coping style and social conditions (19). However, it is well documented that general parenting skills and the quality of the interaction between the parents and the child plays an important role in this transmission (20, 21). Among other things, it has been shown that this group may be less sensitive to their child's signals and needs (22, 23). Several studies can document a correlation between the quality of parenting skills and dysfunctional emotional regulation, insecure attachment and lower self-esteem (9, 22, 23).

In addition to interventions that are geared toward improving parenting skills, a growing literature has shown that the situation for children of mentally ill parents improves when they have the opportunity to talk about their parents' illness (24–26). In a large Norwegian multicentre study, researchers found that children of mentally ill parents have an unmet need for understanding their parent's condition, as well as its consequences (27), and it is not uncommon for the children to feel shame or guilt related to their parents' struggles (24). Interventions that aim to provide age-appropriate information to the children of mentally ill patients have been shown to help increase the children's comprehension of their parents' illness, improve communication within the family and enable the children to better master their situation when the parent is ill (24). Combined, these findings provide grounds for optimism about the results that can be expected if measures are implemented to provide children and parents with information about mental illness, and the opportunity to talk about the parents' mental illness.

In addition to the above benefits of preventing transmission of mental illness from one generation to the next, it has been argued that incorporating the child's perspective in psychiatric clinics also may provide additional health benefits for patients who are parents. An American study showed that mothers with mental illness identified their role as mothers as a primary factor in their treatment. They reported that not being able to raise and care for their children affected their mental health negatively, and prevented their progression toward improvement (28). Interventions that aimed to inform families about how depression affected the parents' functional level, and to provide strategies to detect and deal with stress, reduced symptoms of anxiety and depression in both parents with depression and their children (26, 29). A meta-analysis conducted by Pitschel-Walz et al. (30) reports that treatment that includes the children reduces relapses for patients with schizophrenia by up to 20%. Similarly, (31) found that family-focused interventions resulted in fewer relapses for those with bipolar disorder than in patients who only received medication and maintenance measures. Thus, it is evident that treatment approaches that include the patient's children and family can lead to positive outcomes and improve the situation for both the children and the patient.

Mental health care providers have a unique opportunity to identify children at risk through direct contact with the parents of the children in question. Incorporating a child's focus in mental health services for adults involves having an attitude that children and family are central parts of the patient's life that therefore must be taken into account in the treatment. This requires that health care providers incorporate the family in the treatment process, address the needs of the children and engage in the patient's parenting role (32). Clinics in mental health services for adults can be major contributors in the effort to provide the necessary help for children of the mentally ill. Central tasks include assessing the children, examining how they experience their situation, contributing information and psychoeducation, and taking steps to ensure further action if necessary (33). Norwegian health legislation has since 2010 obligated health personnel to identify whether the patient has children, and to help fulfill the children's need for information and follow-up if necessary.

Challenges in Clinical Practice Related to Children of Parents With a Mental Illness (COPMI)

Despite political guidance and the fact that the children of patients now have a statutory right to be identified and cared for, a number of studies have revealed challenges in the efforts to incorporate the child's perspective in mental health services for adults (27, 34, 35). These studies suggest that there is a gap between what health services *should* offer and what is *actually* offered in services for adults.

In studies conducted by Lauritzen et al. (34, 35), the findings indicated that some of these barriers were at the organizational level. The challenges were related to the high workload staff were under, a lack of ownership of the issue, as well as insufficient allocation of time and resources (35). Other barriers relate to the characteristics of the staff in the clinic. Research has shown that some of these challenges may have two sides. On the one hand, health care providers say that they believe it is important to support the children of patients, but that they at the same time do not consider this to be one of their tasks (35, 36). On the other hand, health care providers express concern about including the patient's children in the course of treatment (37). Furthermore, Lauritzen et al. (34, 35) results indicated that there are barriers related to health care providers' knowledge, lack of training and uncertainty. In this context, other studies have shown that experience and positive expectations related to working with children and the family have an important bearing on the implementation of a focus on children in clinical practice (32).

The Norwegian COPMI Project

The COPMI project is a longitudinal research project 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. The project started in 2010, and involves a long-term strategy for changing clinical practice. The current study is a five-year follow-up study.

The aims of the current study were to investigate:

(a) Has the proportion of health care providers identifying patient's children increased, 5 years after the legal changes?
(b) Has the proportion of health care providers reporting that they support the patient's role in parenting and safeguarding the patient's minor children changed over time?
(c) Has the implementation of the new routines led to higher levels of employee's expectations of positive outcomes, attitudes and knowledge about children as next of kin?
(d) Has the implementation of the new routines led to lower levels of health care providers' concerns related to assessments of whether the patient/therapist relationship will be disturbed by the focus on the patient's children?
(e) Are there differences in expectations, knowledge, concerns and attitudes between employees who have little experience with family conversations and those who have a great deal of experience?

METHODS

Participants

In the pre-study, 219 employees responded (50% response rate), whereas in the post-study, 185 employees responded to the email, corresponding to a response rate of 40.5%. In this follow-up study, 108 employees participated (24.5% response rate). Demographic information is presented in **Table 1**.

TABLE 1 | Study sample: descriptive statistics of the study sample and result of analysis of variance of sample characteristics according to measurement point.

	Pre-test ($N^{\text{total}} = 219$)	**Post-test ($N^{\text{total}} = 185$)**	**Follow-up ($N^{\text{total}} = 108$)**	
	***N* (%)**	***N* (%)**	***N* (%)**	***p***
Sex				0.171
Women	166 (76.1)	123 (71.1)	76 (74.5)	
Men	52 (23.9)	50 (28.9)	26 (25.5)	
Age				0.525
<30	25 (11.5)	11 (6.0)	6 (5.6)	
31–40	55 (25.2)	38 (20.7)	22 (20.6)	
41–50	57 (26.1)	65 (35.3)	36 (33.6)	
51–60	60 (27.5)	51 (27.7)	31 (29.0)	
>60	21 (9.6)	19 (10.3)	12 (11.2)	

The participants were health care providers employed in the adult psychiatric clinic at the University Hospital of North Norway. The clinic consisted of a total of 16 outpatient clinics, and included a geographical area consisting of 31 municipalities. Participants were recruited through a written invitation sent to their email address, in which they were asked to answer online questionnaires (Quest-Back). It was the clinic leadership that made lists of the employees' email addresses available to the research group. The participants were informed that participation was voluntary, and that they would remain anonymous. As a result of the participants' anonymity, there were independent selections in the pre, post, and follow-up studies. The study was approved by the data protection supervisor at the University Hospital of North Norway, and was carried out in accordance with the Declaration of Helsinki on ethical guidelines for medical research involving people (38).

Procedure

The clinic initiated new procedures to identify and offer the necessary follow-up for children of psychiatric patients and this work included the implementation of two interventions: *Assessment Form* and *Child Talks*. The *Assessment Form* is an intervention for treatment providers to increase the identification of patients' minor children. The Assessment Form consists of a general section where information that the providers were required to collect as a result of legislative changes is requested (for example, the child's name, age, siblings, living situation, etc.) and a section that maps the parents' concerns and skills related to parenting. The *Child Talks* is an intervention designed to provide support for patients with mental illnesses who are parents and for their children. The purpose is for the talks to promote health and prevention by having the treatment provider speak with the family about the children's situation and their needs, as well as to provide support to the family and assist them in seeking help from other health services if necessary (39).

Measures

Routine for Identification

The employees' routine for identifying children was measured through the test item: "Have you completed the Assessment conversation?" Participants were asked to answer *Yes* or *No*. Due to miscoding, some responses were recoded into missing values (Pre-test N recoded = 3, Post-test *N* recoded = 2).

Family Conversations

Experiences with the family conversations were measured through the test item "To what extent do you have experience with family conversations?" Participants were asked to rank their response on a five-point Likert scale, from "To a very small extent" (1) to "To a great extent" (5). Experience with family conversations was also used as a predictor variable in one set of analyses. When used as a predictor, the variable was categorized into "low experience," "neither low nor high experience," and "high experience" with family conversations.

Expectations About Positive Outcomes

Participants' expectations about the effect the new routines may have for the children of patients was measured using a scale consisting of four test items. The scale was calculated as the average score of the four test items. Cronbach's alpha for this scale was 0.91 (calculated with re-coded variables without 0 responses). Test items included questions about expected outcomes for patients and their children, and one example of the questions is "I think conversations about and with children can help improve the life situation of children of parents with a mental illness." Participants were asked to rank their response on a five-point Likert scale, from "To a very small extent" (1) to "To a great extent" (5).

Knowledge

Participants' knowledge of the children of mentally ill parents was measured using questions retrieved from the *Family focused mental health practice questionnaire* (40). The questionnaire was adapted to Norwegian conditions/contexts with the permission of the authors. The nine test items included both questions that dealt with knowledge about the children and questions related to knowledge about the new legislation. One example question is "To what extent do you have knowledge about mental illnesses and the consequences such disorders may have for the parenting role." Participants were asked to rank their response on a five-point Likert scale, from "To a very small extent" (1) to "To a great extent" (5).

A factor analysis was conducted to reduce the number of test items. Data was considered to be suitable for factor analysis, with a KMO value of 0.86 and Bartlett's test of sphericity was significant. A Principal Component Analysis (PCA) with Varimax rotation indicated that all test items, with the exception of one, were high loading on one factor. This test item was "To what extent have you received training in the new legislation?" This test item was thus excluded, and a scale was calculated based on the average score of the eight remaining test items. The scale was named Knowledge, and Cronbachs alpha for this scale was 0.86.

Attitudes

The ten test items included questions related to the willingness to change within the organization, attitudes to the new practice and attitudes to the importance of having a focus on the children of mentally ill parents. In addition, the test items related to attitudes about whether or not a child perspective in the practice interferes in the therapeutic alliance with the patient. Test items included questions such as "We should offer the children of patients information and support as next of kin," and "The relationship between the treatment provider and patient may be negatively impacted if the patient's parenting role is brought up." Participants were asked to rank their response on a five-point Likert scale, from "To a very small extent" (1) to "To a great extent" (5).

A factor analysis was conducted to reduce the number of test items (re-coded data without 0 responses). The estimated KMO value was 0.88 and Bartlett's test of sphericity was significant, which indicated that the data was well suited for factor analysis. A Principal Component Analysis with Varimax rotation revealed two components. Positive attitudes to a focus on patients' children was high loading on component 1, while negative attitudes associated with concern about disruption in the treatment and interference in the parenting role was high loading on component 2. The interpretation of the two components is consistent with previous research for the scale associated with Attitudes (34). Two scales were calculated based on the average scores for the test items in the two components. The two scales were named Positive Attitudes and Concerns, and the reliability analysis calculated Cronbachs alpha to be 0.94 for Positive Attitudes and 0.79 for Concerns.

Demography

Age and sex were also reported in the questionnaire.

Procedure

The treatment provider was to conduct a family assessment conversation and fill out a form together with the patient, and should then enter it into the electronic patient record. The family assessment consists of questions such as "Do you have children?," "How many children do you have?," and "Do you have the daily care of the child?." At the end of the conversation, the treatment provider should inform the patient about the Child Talks, and offer this to the patient (41). If he or she agreed to participate in Child Talks, the talk would normally be held in the course of the next 4 weeks.

Data collection was carried out electronically using email through Quest Back on three different measuring points in pre (2010), post (2013), and follow-up (2015) studies. Participants received an email in which they were asked to answer an online questionnaire. If they did not respond, Quest Back generated reminders about the study after 2 weeks. This was done for a maximum of three intervals. This article reports the results from the follow-up study, where we wanted to examine whether the implementation of the two interventions (Family assessment and Child Talks) had led to a change in clinical practice 5 years after health legislation was altered to make assessment and support mandatory for families affected by parental mental illness.

Data Analyses

The data was exported from Quest Back to SPSS. All statistical analyses were performed in IBM SPSS (version 24). Descriptive analyses were used to examine the demographic characteristics of the group. Hierarchical logistic regression analysis was used to test the probability of having identified children of patients, in a two step manner. First the crude odds ratios were estimated, and next the estimates were adjusted for sex and age. Multivariate analyses of variance were used to test the degree to which familiarity with family conversations, knowledge, expectations, positive attitudes, and concern varied according to measurement point. The estimates were adjusted by sex and age. Similar multivariate analyses of variance were conducted to test the degree to which knowledge, expectations, positive attitudes and concern varied according to experience with family conversations. Once again, sex and age were adjusted for.

RESULTS

Assessment and Identification

There was a significant increase in the proportion of participants who identified the patients' children between the three points at which measurements were taken. In the pretest measurement, 44% of participants reported that they had used the Assessment Form, while at the posttest stage, 66% reported that they had used it. At the follow-up test, the proportion had again increased to 72% of the participants. Results are displayed in **Table 1**.

The results indicated that the different measurement points did have an impact on the probability that the employees used the Assessment Form, and thus identified the patients' children. Both the adjusted and unadjusted estimates showed a significantly greater probability of having used the Assessment Form at the post-test (adjusted OR = 2.29) and at follow-up (adjusted OR = 3.37) compared to the pre-test. The confidence intervals between the posttest and follow-up test were overlapping, indicating that there was no significant difference between these two measurement points. See **Table 2** for an overview.

Experience With Family Conversations

In order to examine whether more health care providers had experience with family conversations at the post-test and follow-up test compared to the pre-test, a multivariate analysis of variance was run. The estimates were adjusted for sex and age. The results revealed no significant difference [$F_{(2, 467)} = 1.085$, $p = 0.339$]. The average value and standard deviation for the scores are presented in **Table 3**.

Knowledge, Positive Attitudes, Concerns and Expectations

Multivariate analyses of variance were conducted in order to estimate the degree to which the participants in the pre, post and follow-up groups scored differently on the outcome variables Knowledge, Positive Attitudes, Concerns, and Expectations. The estimates were adjusted for sex and age. The results of the multivariate analyses are presented in **Table 3**. There was a significant difference in the average score for Positive Attitudes between the participants in the three groups ($p = 0.002$). The mean score on positive attitudes were lower both at the post-test ($B = -0.25$, $p < 0.001$) and at follow-up ($B = -0.016$, $p = 0.044$) than at the prtest.

There was also a significant difference in Knowledge between participants at the three measurement points ($p = 0.003$). Knowledge was significantly higher both at the post-test ($B = 0.18$, $p = 0.001$) and at follow-up ($B = 0.16$, $p = 0.022$) compared to at pre-test. Overlapping confidence intervals indicated that there was no significant difference between the post-group and the follow-up group. There were no significant differences in reported Expectations or Concern between the participants in the pre, post and follow-up groups.

High and Low Experience With the Family Conversations

Furthermore, we wanted to investigate whether there were differences between health care providers who had a high and a low degree of experience with family conversations, in regards to Knowledge, Positive Attitudes, Concerns and Expectations of positive outcomes. The multivariate anova, with adjustments for sex and age, showed a significant difference only in Knowledge [$F_{(2, 460)} = 154.253$, $p < 0.001$]. The group with low experience with family conversations had on average -0.93 ($p < 0.001$) lower scores on Knowledge, whereas the group with neither high nor low experience had on average -0.54 ($p < 0.001$) lower scores on Knowledge, compared to the reference group with high experience with Family conversations. Level of experience with family conversations was unrelated to Positive attitude, Concerns and Expectations.

DISCUSSION

The objective of this follow-up study was to examine whether there have been permanent changes in clinical practice related to assessing and supporting children of patients in units for mental health services for adults at UNN. The results indicated that some changes have been integrated since the project's start-up, however health care providers had not changed their practices in such a way that all children of patients are identified and offered support. The results further indicated that after the project withdrew from the participating hospital in 2013, there has not been sufficient systematic work around assessing and safeguarding the need for information that children of mentally ill patients have.

Our findings indicated that the biggest changes happened in the first few years after the legal changes came into effect and at the time the implementation process started up at UNN. There was a significant increase in the number of employees who reported that they identified the children of patients after the Assessment Form was implemented in 2010: the proportion rose from 44% in the pre-study to 66% in the post-study in 2013. Subsequently, there was a smaller, non-significant increase until 2015, when 72% of employees reported that they implemented the required form and identified the patients' children. The results showed that 28% of the participants still do not assess if patients have children. The registration of patients' children is a key part of the 2010 legislation, and after the project withdrew in 2013 there has not been any statistically significant increase in the number of participants who identify if patients are parents.

TABLE 2 | Logistic regression analysis of the probability of identification of children of patients according to measurement time (pre-test, post-test and follow-up).

		Model 1[a]			Model 1[b]		
	N^{yes} (%)	*OR*	*CI*	*p*	*OR*	*CI*	*p*
Pre-test	95 (44)	Ref.[c]			Ref.[c]		
Post-test	120 (65.6)	2.32	1.53–3.51	<0.001	2.29	1.51–3.74	<0.001
Follow-up	73 (71.6)	3.37	2.00–5.70	<0.001	3.37	1.99–5.71	<0.001

[a] *Unadjusted estimates.*
[b] *Estimates adjusted by sex and age.*
[c] *Reference group.*

TABLE 3 | Multivariate analyses of variance of differences in having performed Family conversations, Knowledge, Expectations, Positive attitudes, and Concerns according to measurement time (pre-test, post-test, and follow-up), adjusted by sex and age.

	Pre-test (*N* = 203–215)[a]	Post-test (*N* = 176–182)[a]			Follow-up (*N* = 99–105)[a]			Total
	B	*B (CI)*	*SE*	*p*	*B (CI)*	*SE*	*p*	*p*
Family conversations	Ref[b]	0.15 (−0.03 to 0.36)	0.10	0.143	0.56 (−0.19 to 0.30)	0.12	654	0.339
Knowledge	Ref[b]	0.18 (0.07 to 0.30)	0.06	0.001	0.16 (0.02 to 0.29)	0.07	0.022	0.003
Concerns	Ref[b]	0.10 (−0.05 to 0.25)	0.08	0.188	0.17 (−0.01 to 0.35)	0.09	0.063	0.143
Positive attitudes	Ref[b]	−0.25 (−0.38 to 0.11)	0.07	<0.001	−16 (−0.32 to 0.01)	0.08	0.044	0.001
Expectations	Ref[b]	−0.14 (−0.26 to 0.01)	0.06	0.031	−0.12 (−0.27 to 0.03)	0.07	0.104	0.067

[a] *The variability in N is caused by missing values for some of the variables.*
[b] *Reference group.*

A successful implementation of the Assessment Form may in this context be considered a quality indicator (27), as it accords with the health authority's guidelines and provides a thorough review of the circumstances such as the child's name and age, as well as clarifications about the patient's care responsibilities. Unfortunately, our results indicated that the implementation had not been successful, as a high proportion of the participants report they are not using the Assessment Form. One possible explanation could be that they register patients' children in other documents in the patient journal (42). If so, this is a less systematic documentation of patients' children than the Assessment Form, and the degree of information registered in the record may vary between treatment providers.

An even greater challenge has related to implementing the Child Talks in the clinical practices of health care providers at the hospital. The results indicated that there has not been any significant increase in the employees' experience with family conversations since the implementation process started in 2010. At that time, 20% of the employees reported that they had high experience with family conversations, while in 2013, 23% of the participants said the same. In 2015, 26% reported having a high experience of these conversations. It is possible that "family conversations" meant something different to the employees at the last two measurement points than it had at the pre-measurements. In 2010, the implementation of the Child Talks had not yet started up, and it is therefore possible that the reported experience of family conversations had a more general meaning and was not necessarily linked to this specific intervention. One would still expect that if more employees had begun to implement the Child Talks after the implementation started in 2010, participants would also have reported higher levels of experience with family conversations in 2015, which does not seem to be the case. We therefore do not think that different perceptions of what the family conversations are is a plausible explanation for the fact that 75% of the employees still report low levels of experience related to talking with the patients' families, even five years after the new law required health care providers to support and safeguard the information needs of children of mentally ill patients.

However, our findings still support the claim that the employees value a focus on children in the clinical practice. Participants generally had a high degree of positive attitudes to the introduction of the Child Talks in the clinical practice, which is reflected in the high average scores at all three measuring points. However, participants were the most positive to the introduction of a child's perspective in 2010, the same year that the new law came into force. The participants reported a significantly lower degree of Positive Attitudes in 2013, while there was no significant change between the measurements in 2013 and 2015. The participants reported that they largely agreed that health care providers should identify the children of patients, offering them information and support, and initiate a dialogue about the parenting role with patients who have children. The results indicated that there is no significant change between the three measurement points in the employees' concerns related to having a focus on patients' children in the treatment. The participants were generally not very concerned that talking with patients about their children and parenting role may damage the patient/therapist relationship, which is reflected in the low scores for concern. The employees' positive attitude as well as the low level of concern, may indicate that there is a certain degree of willingness to change among the employees (43). Despite this, more than 25% of the participants still report that they do not seek to clarify whether the patients have children, and more than 75% have little experience with talking to the patient's children at the follow-up point in 2015. The results thus indicate that a significant proportion of the staff has not integrated the new procedures in their clinical practices, despite the fact that they show a high degree of positive attitudes to including a focus on the patient's children. This is in line with the findings from the pre and post study in the project (34, 35), as well as past research (36, 44), which indicates that the positive attitude of the staff is not sufficient for them to incorporate a focus on patients' children in the treatment.

We also wanted to investigate whether there has been any change in the employees' level of knowledge since the implementation of the new interventions started in 2010. The results show a significant increase in participants' self-reported knowledge from the implementation process started in 2010 and to it reached its highest level of activity in 2013. There was no change in participants' knowledge from 2013 to 2015, which indicates that the employees' level of knowledge has remained stable after the project withdrew from the hospital. However, it is possible that the reported increase in knowledge between the measuring points in the pre and post study is not very significant in the clinical practice. Although there was a statistically significant change between the measuring points in 2010 and 2013, the calculation of the effect size showed that this was virtually negligible.

The results from this survey indicate that there also was no difference between these two groups related to concerns or positive attitudes. Both those with high and low degrees of experience with the family conversation had a very high degree of positive attitudes to focusing on the patient's children during the treatment. However, our results did indicate a difference in the reported level of knowledge between the two groups. Employees who had more knowledge about children of mentally ill parents also reported more experience with family conversations than those with lower levels of knowledge. This can be seen in the context of discoveries made in the measurements carried out in 2010 by Lauritzen et al. (34). This finding showed that employees who reported that they identified the children of patients also had a higher level of knowledge about children as next of kin and of the legislation than those who did not identify the children at this measurement point. These findings are also consistent with results from studies that have shown that the ability to support patients in the parenting role increased through the health care provider's personal experience and their professional experience and education in this area (45, 46). On the other hand, studies have revealed that a lack of knowledge and skills can be obstacles to the implementation of the child's perspective in clinical practice (44).

There are reasons to assume that there is potential to strengthen the training of health care providers, so as to increase their level of knowledge and skills related to working with children as next of kin. The results from a Norwegian multicentre study showed that 40% of the employees in mental health services for adults had not participated in training related to practizing the new legal provisions (27). In this context, Maybery and Reupert (44) propose that the training should have a two-part focus. One part should be directed toward the patient's parenting role, and should provide knowledge of parental guidance and increase the ability to discuss the consequences that mental illness can have for the child in an empathetic way. The second part should deal with the direct work with children, where the ability to talk with children and provide age-appropriate information should be key (44). Another suggestion might be to include knowledge about work with children and families in the education programmes for the various health professions (27, 35), as this can give future health care providers an expanded and solid knowledge that increases their sensitivity to the children of patients once they enter the workforce.

Child Perspective Team

It is not uncommon for innovations to have to be changed in order to adapt to the practices they are to be implemented in Meyers et al. (47), and it is possible that it is necessary to rethink the implementation of the family conversations in the participating hospital. A strengthened partnership between the adult and child services can help promote the safeguarding of patients' children and families (27). In addition, the interdisciplinary cooperation between different health services is important in the efforts to offer support to children as next of kin (27, 48). By involving key personnel, who are often referred to as "champions" in the literature, conditions can be created that promote the implementation process (47). The champions have a high level of knowledge and motivation, as well as the ability to engage the rest of the staff (47). Champions can uncover barriers that delay the implementation process in the clinic, and help develop solutions to address these obstacles (48). It has previously been proposed that an employee with responsibility for children can fulfill such a role in the clinic (48), but this requires that more time and resources be set aside for this work (33). Furthermore, the employee with responsibility for children must be anchored at a sufficiently high level in the organization, and must be given influence in the clinical practice (27).

With regard to the identification and assessment of the patients' children, this will still be the responsibility of the patient's treatment providers, as is done in the existing routines. Possible measures to increase the identification and registration of patients' children may be to provide adequate training in keeping patient records for health personnel (27), and/or to introduce the registration of patients' children as a quality indicator for the hospital's service (42).

CONCLUSION

The children of mentally ill parents is a group with a known risk of developing mental illness, and since 2010 they have had a statutory right to be identified and cared for by health care providers in Norway. The interventions "Assessment Form" and "Child Talks" comply with the statutory requirements, and can prevent and reduce the risk that children of patients themselves develop mental illnesses. Our results show that the hospital has not managed to implement changes in its practices to ensure that all minor children of mentally ill patients are assessed and cared for in a satisfactory manner. On this basis, we have proposed specific measures to establish dedicated teams at psychiatric centers within the hospital, which will be responsible for conducting conversations with the patient's children. Furthermore, we believe that the key personnel in such teams can help to overcome important obstacles at several levels in the organization, and speed up the implementation of the new procedures. However, this will be difficult to implement without adequate allocation of time, resources, and personnel.

LIMITATIONS OF THE STUDY

The most central limitation in this study is the relatively low response rate in the study, particularly at the follow-up point (50% for the pretest, 40% for the posttest and 24% for the follow-up measurement). This can affect the results if the decision to participate in the survey relate to or are affected by the respondent's attitudes. It is also possible that those who choose to answer the survey already have more positive attitudes attached to having a family perspective in the treatment they practice. One consequence of this may be that this article represents more positive attitudes in health care providers about the topic than what actually is the case. However, a response rate of 24% is quite common for online surveys. The cohorts of respondents did not differ significantly in terms of age and gender at the different times of measurement. Confront **Table 1** for information about the studied population in terms of age and gender.

Another limitation of this study is the way some of the questions are worded in the questionnaire. Participants are asked whether they to a high or low degree have "experience with family conversations." In our study, we are interested in examining whether the employees have begun to make use of the "Child Talks" intervention. It is possible that the participant interprets experience of family conversations in a larger context and is not responding specifically to whether they have experience with the "Child Talks." Subsequent studies should therefore examine whether health care providers have started to make use of the "Child Talks."

This study is based solely on measurements from self-reporting about attitudes, knowledge, expectations and current work practices. Future studies should also include objective measurements such as patient record data to investigate how many patients' records report on assessments of family conversations.

The samples at pre, post and follow-up tests are not independent, due to the anonymity of the respondents. The respondents may therefore have been influenced by being nested within the same department, which may have biased the results. In future research personnel should be evaluated individually to track changes in levels of competence and clinical practice.

In terms of the existing evidence for these interventions, there are few studies available. The interventions are being tested in Norway and Portugal, and we expect results about their effects to be published eventually. However, there is generally a lack of intervention studies that document the positive effects of incorporating a focus on the patient's children in clinical practice and treatment within the mental health services. Hence, future studies should be conducted to study effects of interventions that aim to promote well-being among children of parents with a mental illness. This may motivate hospital management and health care providers to make use of such interventions to a greater extent.

ETHICS STATEMENT

The Data Protection Officer at the University hospital of Northern Norway approved the project.

AUTHOR CONTRIBUTIONS

All authors agree to be accountable for the content of the work. CL and CR designed the project. CL collected the data. Analyses were conducted by KR, MN, AW, and CL. CL drafted the article. All authors participated in the writing of the manuscript.

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Key Informants Specify Core Elements of Peer Supports for Parents With Serious Mental Illness

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Background: Researchers have documented the lack of evidence-based interventions for parents with serious mental illness (SMI). Given the prevalence of parenthood among individuals with SMI, the value placed on parenthood, and their diverse vulnerabilities, a robust, theoretically sound and empirically tested model of peer supports would likely provide a valuable complement to psychiatric services. In this paper, we lay the groundwork for a model of peer supports using a program theory development process and guided by stages of evidence-informed innovation outlined by the National Implementation Research Network.

Methods: This study employed a developmental design in the initial stage of a larger study of the development, implementation, and testing of peer supports to address three questions: (1) What needs of parents with SMI are particularly well-suited to peer supports? (2) What do peers have to offer parents? and (3) What is unique about peer supports for parents? A purposeful sampling strategy was used to recruit key informants ($n = 22$) familiar with peer supports, family-focused care and the experiences of families living with parental mental illness. Individual interviews were conducted face-to-face or on the telephone and a full-day group interview was conducted using a workshop format. Interview data were analyzed qualitatively to identify themes reflecting potential core program elements.

Results: Consistent themes drawn from data comprise four core program elements: engage, explore, plan, and access and advocate. These core activities are likely founded on practice principles that include a focus on families and their strengths, cultural sensitivity, and acknowledgment of the trauma experienced by many parents. The findings raised a number of challenges in contemplating peer supports for parents with SMI, including the need for ongoing support for peers.

Discussion: In developing this model, aspects of organizational context must be considered along with specification of the characteristics of parents with SMI suited to this approach and the attributes of peers providing support. A fully-articulated model must include parallel theories of change for the workforce, as well as for participating parents, to support well-being in the context of peer relationships and the success of parents with SMI in family life.

Keywords: parents with serious mental illness, peer supports, family-focused care, psychiatric services, program theory, evidence-based practice

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INTRODUCTION

Numerous researchers have documented the lack of targeted evidence-based interventions for parents with serious mental illnesses (e.g., schizophrenia, bipolar disorder, major depressive disorder) (1, 2). Effective psychiatric treatment is recommended, of course, dependent on a person's history, diagnosis, preferences and response. However, inattention to issues of importance to individuals with serious mental illnesses (SMI) who are parents, and the probable impact of unaddressed issues on a person's participation and progress in treatment, suggest the potential benefit of attending to a parent's circumstances and priorities, particularly if they are contributing to stress. Given the prevalence of parenthood among individuals with SMI (3, 4); the value placed on parenthood by these individuals (5, 6); and their diverse vulnerabilities (e.g., limited resources and supports) (7), a robust, theoretically sound, empirically tested model of peer supports for parents with SMI would likely provide a valuable complement to traditional psychiatric treatments. In this paper, we lay the groundwork for a model of peer supports for parents using a program theory development process (8) and guided by stages of evidence-informed innovation outlined by the National Implementation Research Network (NIRN) (9).

Peer-delivered service models have increased markedly over the past two decades and are now an integral component of the behavioral health care system in the USA (10, 11). Peer support is based on the idea that individuals with lived experience of mental health and/or substance use disorders who have made progress in recovery may be uniquely positioned to offer support, validation, and hope to others (12, 13). Within the field, peer support specialists are persons who identify as having mental health conditions who receive formal, skills-based training to deliver services in the behavioral health care system (14). Peer support specialists occupy a variety of roles in behavioral health care that vary by setting, credentialing requirements, and service model (6, 15). In the USA, states are authorized to bill Medicaid, the public insurer, for peer support services to assist individuals in recovery from mental illness and substance use disorders.

A growing body of literature supports the benefits of peer-delivered services in behavioral health care (16). Much of this literature suggests positive outcomes as demonstrated by increasing consumer engagement and retention in care, linking individuals to community-based support services, fostering hope, addressing social isolation, increasing activation and self-care, and reducing inpatient utilization of services (11, 13, 16–21). However, recent meta-analyses of peer-delivered services, particularly for individuals with SMI, provide a more tentative assessment of the empirical evidence and raise questions about the methodological rigor of published studies (18, 22, 23). Adequate categorization and clarity surrounding the role, objectives and performance of peers is often lacking (24–26). Theory-based program models of peer support with well-defined change mechanisms are needed (22, 25, 27), as is increased attention to implementation processes and barriers that might affect participation and anticipated outcomes (22, 25).

The task of specifying the core program elements and underlying processes of a model of peer supports for parents with SMI, along with clear specification of the role of parent peer specialists, is informed by review of the literature regarding peer supports in mental health in general (13, 20, 28, 29). Authors describe and link peer support processes to conceptual underpinnings. Chief among these, by definition, is the importance of lived experience with mental illness and within the system of services (13, 20, 29). The peer specialist's use of positive self-disclosure and the perception of commonalities shared with and by the patient or client contribute to relationship engagement and foster hope for change (13, 20, 29). The peer relationship is enhanced by the emotional engagement that develops in this work through trust, acceptance, understanding and empathy (13, 20, 29). Peer specialists have the ability to role model progress in recovery and coping. They may be more likely than clinical practitioners to take a strengths-based approach, rather than focusing on psychopathology, and facilitate access to social and practical support, both informal and formal (13, 20). The peer specialist role has been described as having benefit to the peer specialist as well as to the patient or client, via the assumption of the helper role (13, 29).

However, the possible emotional closeness between peer specialist and patient or client, in combination with the fact that the peer specialist role definition often lies somewhere between that of traditional mental health clinician and patient or client, may contribute to difficulties (29). The peer specialist may be vulnerable to over involvement, unsustainable boundaries and burnout (29), suggesting the need for ongoing training and support. The organizational or agency context (e.g., hospital in-patient setting, community mental health agency or health center) in which peer support is offered must be considered as well (29). Authors recommend that the process of creating peer positions should include the active involvement of non-peer staff, organizational leaders and people in recovery, especially those who may serve as champions in the effort (20).

This paper reports on the initial, preparatory and foundational stage of a multi-stage study to develop, implement and assess the effectiveness of peer supports for parents with SMI. It reports on work done together with diverse stakeholders in laying the groundwork for model development of peer supports targeting parents with SMI. The initial tasks include developing program theory linked with core elements and processes, and identifying essential qualities and characteristics of parent peer specialists. Research questions include: (1) What needs of parents with SMI are particularly well-suited to peer supports? (2) What do peers have to offer parents? and (3) What is unique about peer supports for parents (i.e., what distinguishes them from peer supports in general)? Findings from this initial exploration will inform the next stages of the project: first, further elaboration of the peer supports model for parents with SMI and, second, the installation, implementation and testing of the resultant innovative, empirically informed model.

METHODS

Design

The study reported here employed a developmental design as the initial stage of the larger study of the development, installation, implementation and testing of parent peer supports. A developmental design has the purpose of helping develop an innovation, intervention or program (30). Feedback loops inform ongoing program, staff and organizational development. This approach is particularly relevant in situations in which programs or models are being adapted to new conditions, contexts or target populations (30), that is, the exploration and installation stages outlined in the NIRN framework for the implementation of evidence-based programs and other innovation (9). The larger study methods combine three iterative approaches to program theory building (8): (a) a review of the academic and gray literature (deductive approach); (b) interviews with key informants (articulating stakeholder mental models); and (c) Implementation Team meetings as the potential program model is considered for installation at the community-based agency (inductive approach). In this paper, we report findings from interviews with key informants. The extensive literature review, reported above only briefly, focused on the experiences and needs of parents with SMI and the development, implementation and testing of peer supports in general. Implementation Team meetings are underway as we prepare to install peer supports for parents with SMI in the community agency context.

Key Informant Interviews: Articulating Stakeholder Mental Models

Mental models for programs reflect the ways in which stakeholders describe what a program would look like, how it would work, and how success would be determined, particularly as stakeholders consider the characteristics, needs and experiences of the target population (8). The selection of key informants is informed by the notion that engaging stakeholders provides access to expertise, facilitates the development of a shared understanding of the potential program and promotes empowerment, communication and uptake of the final result (8). Diverse perspectives are included to develop a more comprehensive program theory.

Sample Recruitment

A purposeful sampling strategy was used to recruit initial key informants familiar with peer supports, family-focused care and the specific needs and challenges of families living with parental mental illnesses. A preliminary list of potential study participants was constructed by the investigators, chosen to reflect diverse, relevant perspectives including state agency policymakers and funders, provider agency managers and staff, experienced peer specialists, parents with SMI, program developers and researchers. Additional informants were added to the sample as they were identified by study participants. Potential participants were sent a recruitment email informing them of the study and inviting their participation in an interview to discuss peer supports for parents with SMI, including relevant factors to consider in developing the proposed model and ideas about how to implement, sustain and assess the effectiveness of the model. All invitees agreed to participate ($n = 22$). The investigators were included as participants given the co-production nature of the project and their partnership role in facilitating key informant engagement and contributing to the design of the ultimate parent peer supports model (8, 30).

Interview Procedures

Interviews were conducted by one of the investigators, experienced clinical researchers, either face-to-face or on the telephone, at the convenience of the participants, between March 2016 and July 2017. A study overview including the interview items was emailed to participants prior to the scheduled session. The individual interviews lasted 1–2 h. One full-day group interview was conducted using a workshop format with expert stakeholders to draw out rich perspectives on key elements of program theory in an iterative, face-to-face manner (8). At the beginning of each session, informed consent was obtained and participants completed a brief background survey (e.g., age, gender, race/ethnicity, education, training/discipline, organizational affiliation, and years in their current role). For those individuals interviewed by telephone, informed consent forms were emailed in advance, signed and returned by the participant. Study procedures were reviewed and approved by the state agency research review committee and the institutional review boards of the investigators' universities.

The investigators employed a semi-structured interview protocol informed by a series of open-ended questions as suggested in the NIRN Exploration Stage of innovation development (9). The questions were used to orient conversation. By way of introducing the topic of peer supports for parents, the interview began by asking participants for their perspectives on the needs of parents with SMI. Follow-up questions elicited information on current programs and practices, potential resources for the proposed parent peer supports, anticipated implementation and sustainability challenges, and additional stakeholders to contact. The interview concluded with an open-ended question asking whether the participant had anything else to add. The investigators began reviewing data as they became available, so that earlier interviews informed later interviews as knowledge was obtained. Verbatim notes were taken during all interviews, reviewed and transcribed by the study investigators using standard word processing software, and de-identified to maintain the participants' privacy. In addition, for the workshop session, extensive notes were made on easel sheets along with verbatim notes to facilitate group discussion. These were transcribed as well.

Analysis

A framework approach to the interviews was employed, given that a starting set of issues to investigate was identified prior to data collection. These included recommended considerations in the NIRN exploration stage of innovation related to implementation context, such as the identification of essential resources, potential barriers, and lessons learned from prior initiatives (9). Open-ended interview questions were developed to provide for unstructured responses to these implementation topics as well as to allow for rich responses from study participants regarding the specific needs and experiences of parents with SMI (31, 32). While the overall framework of the interviews as focusing on developing and implementing peer supports was identified in advance, themes related to essential elements and considerations for the development and implementation of peer supports for parents with SMI emerged from the data, in a grounded theory type of approach, as thematic coding progressed (33). The investigators worked independently initially, recording notes of themes that emerged from the data, as well as informed by the research objectives. They reviewed and updated themes based on the frequency of themes within a response and the emphasis granted certain themes by participants. Together, investigators revisited and updated themes and came to consensus regarding the codes and the language used to define them. Coding was facilitated through the use of Dedoose software (34). The investigators generated memos regarding impressions and emerging themes and relationships, and reviewed findings as they supported, diverged from or augmented prior knowledge.

Trustworthiness of the qualitative analysis process and findings was established in multiple ways (35). Transcripts were reviewed and coded independently, impressions compared, and differences reconciled to achieve complete agreement in all cases. Trustworthiness was further established through member checking (33). Preliminary findings were reviewed by independent stakeholders on the study's Implementation Team and actively working in the field.

RESULTS

Participants

Massachusetts, the seventh smallest state in the US, is located in the Northeast region of the country. The state population is estimated at 6.93 million. The state's capital, Boston, is the most populous city; more than 80% of Massachusetts residents live within the greater Boston metropolitan area. Prevalence estimates suggest that more than a half a million adults in Massachusetts experience SMI; unmet need for care is high. While Massachusetts is lauded as among the best states in the nation regarding access to behavioral health care, many individuals with SMI and substance use disorders struggle to find services.

Twenty-two informants participated in the study. The majority of informants were female (77%), over 45 years of age (77%), and Caucasian (86%), reflecting the characteristics of the Massachusetts human services workforce. Most had a four-year university degree or more in education (81%). The education of the remaining participants ranged from some high school to some college, with two completing a high-school equivalency diploma or trade school. Just over one-third of the participants (36%) were in their current positions for a dozen years or more; two were unemployed outside the home. Just over half (59%) described themselves as being in practitioner roles (e.g., social work, rehabilitation counseling, clinical psychology); 23% were in policy and program administration positions; and 18% were in other roles (e.g., mental health advocate; parent at home). Informants represented community mental health and state human service agencies, and included community leaders from varied practice settings and academic researchers, as well as advocates and parents with SMI. Their familiarity with peer supports and as service providers and recipients was extensive. Their experiences varied from working as peer support specialists and educators, to directing and supervising peer and recovery services to receiving peer services as patients or clients. Several informants were funders and administrators of peer services. A significant number identified as individuals with lived experience of mental illness or as having family members with lived experience. All were parents. The notion of peer support work in the US implies that those who direct or supervise these programs are often peers themselves, with lived experience of mental illness and as service recipients. It would be incorrect to assume that only those parents unemployed outside the home had ever received services. Participants were asked to designate their primary role as they identified it at the time of the study, but were encouraged to draw from their range of experiences.

Research Question #1: What Needs of Parents With SMI Are Well-Suited to Peer Supports?

The gaps in supports and resources for parents with SMI were frequently noted by key informant participants. In addition, parents with SMI may have questions or concerns that are specific to their situations or circumstances. Needs that are common among all parents may be exacerbated in situations in which parents live with mental illnesses.

Gaps in Supports and Resources

Many participants touched on the lack of personal or professional support that parents with SMI experience, the pronounced sense of not knowing with whom to talk, and the implications of this isolation. As one key informant noted, "Often parents say they have no one who they rely on." Participants observed that because so many parents with SMI do not have a "non-threatening person with whom they can confide" they may compartmentalize their needs and fear disclosure about salient information including the "use of psychiatric medication" and "childcare challenges." Participants suggested that among parents with SMI, fear of the child welfare system and child protective services is a significant source of stress that may exacerbate parental isolation. Such parents might avoid seeking services lest disclosure of their psychiatric diagnoses or challenges in childrearing result in their children being taken from them. As one participant suggested, "the child welfare system exists to protect children, but there has to be a way for parents who are struggling to reach out—without the fear of having their children removed [from their care]." Participants also pointed out that parental isolation and the lack of support give rise to doubts and fears about a common concern of parents with serious mental illness, that is, if and when to disclose their illness to their children and others.

Concerns Specific to Parents With SMI

Peers, given their own experiences, may be best suited to discussing a number of issues that specifically relate to family life and serious mental illness. Participants strongly endorsed the use of peer supports for parents who may be "contemplating having another child" as peers may have experience with balancing the demands of parenting multiple children and managing serious mental illness. Peers may have insight into complex or stressful family relationships, for example, and suggestions for parents who have complicated relationships with their own parents, yet must "rely on [them] for child support."

Many participants noted the crucial support that a peer may provide in helping parents reframe their experiences, to "deconstruct stigma" as they make negative assumptions about the relationship between their own illness and their children's behavior, and to "normalize the day-to-day challenges of parenting" that exist independent of a psychiatric diagnosis. Parents may blame themselves and their illnesses as contributing to behavior in children that actually may be developmentally appropriate (e.g., temper tantrums in toddlers). According to one informant, "It's common to ascribe any difficulties in parenting to depression, but there are normal experiences and challenges of parenting that exist apart from major depression." Further, parents with serious mental illness may need extra support in "the pragmatic steps necessary to get through the day," for example, figuring out "how to get their kids to the bus on time in morning" or "how to get them enrolled in a good afterschool program." In helping parents with serious mental illness navigate these day-to-day challenges, peers may suggest alternative strategies that help to change parents' perspectives from that of feeling overwhelmed by demands and "failing" to one of coping and succeeding in managing everyday family life.

Parents may benefit from peer supports for situations unique to them. As participants pointed out, parents who do not have custody of their children may have supervised visitation as required by child protective services or may hope for reunification with their children. Peers may have experience with child welfare and advice for navigating the system successfully. They may be preferred, willing and able to supervise parents' visits with their children. A key informant described, "This is complicated, but it's worth taking on. How can a parent interact with a child, particularly if they have limited contact?" Another informant suggested, "Parents may need support for grieving the loss of parenthood—for those who opt not to have children" and for those who have lost custody or contact. Parents with SMI may need help repairing and renewing bonds with children. For older parents, peers may be helpful in assisting them to reconnect with their adult children.

Generic Parenting Issues Magnified

Participants also remarked on the needs of parents with SMI that are generic to all parents. Concerns about housing, healthcare, and education were frequently cited. However, many suggested that even the very basic needs of vulnerable individuals take on a different quality and urgency for parents with SMI who are primary caretakers of minor children. As one informant noted, "Do I have enough money to pay my electric bill this month?" may mean something very different for individuals who are parents than for those individuals responsible only for themselves. For parents with SMI caring for children, failing to provide for the essentials of daily living may exacerbate their doubts about their parenting abilities, mentioned above, and draw the attention and concern of others.

Research Question #2: What Do Peers Have to Offer Parents?

Peers may be sensitive to the importance of empowerment and the contribution of successful planning and problem-solving to feelings of self-efficacy and recovery. They may be well-suited to helping parents access and advocate for supports and resources for themselves and their families.

Plan

Peers can draw from their own experiences to help parents solve problems, envision change and make concrete plans to achieve their goals. "Peers workers facilitate peer-driven goal planning." Because of their unique perspective as peers, they may be better able to partner with the parent to "go to a school meeting; go to a doctor's appointment; go to get a prom dress…help plan a [parent-child] visit, plan birthday parties; talk about court and steps to get children back." They may have tested strategies, not only for managing day-to-day life but, specifically, household, money and time management. According to an informant, the peer can focus on "whatever it takes for families to be successful." Peers may see "that there is more than one way forward" and appreciate that recovery is not a straightforward path; there will likely be set-backs and relapses along the way. Peers may be more inclined than non-peers to recognize that "It's about helping the client with self-determination, autonomy and self-advocacy" and that by succeeding in planning and problem-solving, parents begin to see and believe that change is possible and that they are capable of making it happen.

Access and Advocate

One informant indicated, "The role of peer support is to provide information and support as needed." Parents may not know how to access resources, that is, "how to get adequate support" from friends or family or how to "navigate communication" across a network of professional providers. Peer specialists who are parents may be better acquainted with community resources useful to other parents and especially to those living with mental illness. Peer specialists may facilitate "referral to grassroots, community-based programs; in-home recovery services; and community-based supports" and may also be helpful in "talking with family members at the request of the client [parent]" to help the parent build natural supports. Advocacy is a key element of the process of accessing and building supports and resources. An informant pointed out, "A significant element of advocacy is integrated into the peer-based model of care. The [parent peer specialist] must understand this context, and the role of advocacy and networking with other providers serving families."

Research Question #3: What Is Unique About Peer Supports for Parents?

Informants suggested that peers could offer a unique and specialized approach to supporting parents with SMI in terms of their posture or approach to engaging with parents as well as the specific skills and expertise they employ to explore parents' strengths, resources and supports, and elicit motivation to make desired changes. Specific challenges were suggested that reflect both the benefits as well as the vulnerabilities inherent in the peer-parent relationship.

Engage

Participants indicated that peers may readily be able to engage with parents with SMI with greater authenticity than can traditional mental health clinicians. As one pointed out, peers create "relationships that feel normal, not clinical." They can "get real with parents in a way traditional mental health workers can't." The assumption is that peer relationships are based on the concept of mutuality, which "tends to make people more open." Mutuality is sparked by the perception of similarities and sharing of lived experiences. Peer specialists are trained in the use of self-disclosure to enhance relationship-building. A parent who has lost contact with children, for example, may feel better supported by someone who can talk openly about feelings attached to similar experiences and provide examples of ways in which they coped. Informants recommended a life span approach to parenting, that is, providing peer supports to parents at all stages of family life.

Participants suggested the importance of cultural sensitivity. In addition to cultural variations in the framing of mental illness and treatment, for example, the ways in which parents talk about mental illness with their children or their approach to child behavior management may differ from culture to culture. The notions of authenticity, mutuality and cultural sensitivity led to discussion of the match and fit between parents and peers. Participants emphasized the importance of choice. As one informant who is a parent with SMI pointed out, "some pairs fit better than others.…Either side should be able to decline the match or ask for a re-match.… [You] need to consider the parent's needs and the peer specialist's needs in making the match." According to another informant, "Not everyone wants peer support. For some, it's not the right time." The question of whether peer supports for parents must be provided by persons who are parents emerged and, if so, what aspects of parenting should be considered as criteria for the match (e.g., age of children, custodial or non-custodial status, etc.). And in weighing the criteria for fit, the felt connection between peer and parent may matter more than particular aspects of lived experience.

The notion of choice was echoed by participants who emphasized the importance of peers taking a non-judgmental approach, given that parents with SMI so often feel judged. The value of a non-judgmental approach plays out in working together with parents to explore their families' strengths and vulnerabilities, plan for themselves and their children, and deal with the consequences of their actions. According to one informant, "People have to be given choices, including potentially risky or harmful choices," suggesting that parents may make choices that peers consider ill-advised. Another suggested, "Peer workers endorse the dignity of risk." Parents with SMI are often viewed negatively, with little regard paid to their efforts or capacity to care for their children. A peer specialist may take a more "normalizing" approach focusing less on "clinical appraisal" to make "more room for exploration of feelings that are stigmatized in clinical settings." Allowing thoughts and feelings to be expressed without fear of censure may reduce the level of blame or shame parents feel about their choices.

Explore

Peers may be especially helpful in assisting parents to identify strengths and reframe their capacity to make change in their lives. In the words of an informant, "You may be combatting a family's story or narrative. Parent peer specialists may be able to support the client around this better than others…can change the story and uplift the children." By helping parents to "figure out the life they want to live…in contrast to the treatment world, which she describes as 'so clinical,'" the parent peer specialist instills hope for change. A parent informant described a particular peer specialist as bringing "creativity, fun and positivity to her work" that allowed the parent to explore and envision a more positive future. This helps parents "to become unstuck," to participate in planning and to muster the energy and resources to achieve goals.

Peers may provide examples of success to parents with SMI, according to key informants. Peer specialists are often selected into their positions by virtue of the progress they have made in their own treatment and recovery. Parent peer specialists may offer examples and role model successful efforts to cope with the demands of parenting and illness management or negotiate more positive relationships with children and family members. They may draw from their own life experiences, not only to build relationships through perceived mutuality, but to provide examples of ways in which they themselves have coped or made changes that might work for the parent.

Lived Experience of Family Life May Convey Challenges

Peer specialist training may involve exposure to practice skills and approaches that are generally relevant to parents with SMI (e.g., shared decision-making, problem-solving, skills-building). The work of peers is further informed by their own family values and lived experience of family life, as well as their experiences with mental illnesses. Consequently, in drawing from and disclosing their family experiences, peers may become vulnerable in ways that differ from or are experienced more keenly than when providing more generic supports. Peer specialists may understand that "boundaries between peer and client are more permeable [than those between a traditional clinician and client]" and "boundaries and use of self-disclosure are [therefore] emphasized in…training." The focus on family life in working with parents has the positive potential to provide rich opportunities for sharing, but carries the risk of reminding the peer of family experiences or memories, possibly traumatic, that were painful or unpleasant. Adding family experiences to the shared lived experience of mental illness and recovery not only multiplies the opportunities for mutuality, but increases the pool of potentially painful or unresolved issues for the peer as well as for the parent.

The peer specialist may also be inclined to extend themselves for parents in ways they might not for patients or clients who are not parents, particularly because the well-being of children may be involved. As one informant pointed out, "Peers, for example, shouldn't be driving people to appointments, unless it's in the service of their work…talking to the client, introducing them to a support group." And, "Peers are not taxi drivers or junior counselors." Another informant suggested, "Peers cannot be held responsible for client behaviors, nor should they be asked to do favors for clients, including holding money or assisting with medications." These issues could become particularly salient and potentially confusing to the parent peer specialist when children are involved in the home, for example, when a parent is unable to drive a child to a doctor's appointment or has no food in the kitchen. However, as another informant concluded, "Sometimes people need to be taken care of; that's what staff do. Sometimes people need someone to be with them; that's what peers do."

DISCUSSION

Parents With SMI May be Helped by Peer Supports

Diverse informants, including parents themselves, agree that parents with SMI are often isolated, dealing with gaps in available supports and resources. They may be challenged by situations or circumstances related to parenting while managing mental illness, including very practical, day-to-day challenges of raising children, relationships with children across the life span, and issues other parents face but that are exacerbated by the challenges of navigating treatment and recovery. Peers offer special knowledge, drawn from personal feelings and their own experiences, that may serve as a resource to parents. What is unique about peer supports for parents is the potential for relationship building and the capacity for open discussion based on the sharing of the lived experience of managing mental illness, family relationships and parenthood, and the authenticity and mutuality all this inspires. Parent peer specialists potentially offer examples of success, serve as role models, reframe deficits and set-backs, normalize parenting experiences, and disclose information about themselves with purpose and intent. The lived experience that peers share also may include expertise in navigating the health care and social service systems as a parent with SMI. For example, they may be aware of community resources or problem-solving strategies that have worked for them.

Potential Challenges

The findings raise a number of challenges in contemplating peer supports for parents with SMI. Some are broadly relevant to peer support and others are particular to the role of parent peers (e.g., boundaries within the parent-peer relationship, the judicial use of self-disclosure, the potential for raising the peer specialist's own concern for painful family issues, and the peer's values associated with families, parenting and children). To some extent, these challenges exist across the range of peer supports (i.e., substance abuse recovery coaches, peer supports in elder care), but because young children may be involved—either living in the home or not—a more cautious approach may be warranted.

Working with parents, especially parents of young children, underscores the need for explicit, ongoing training and perhaps a willingness to re-examine norms or standards of practice, particularly given the assumptions, values and range of feelings that often surround parenting and family life. For example, the idea of a peer's role in honoring parent choice was noted by several informants, the underlying notion being that peer specialists are there to honor and support the autonomy of their peers. In reality, this aspect of peer support is not terribly distinct from the approach of traditional mental health clinicians, in the sense that clinicians may not weigh-in directly and/or advise patients or clients specifically about everyday decision-making. However, honoring patient or client choice is essential to peer support, in part, because in supporting autonomy, peers support clients' wellness, as opposed to their illness, and their potential for growth and recovery (21). This may be the mechanism behind a peer's authentic use of self. It is certainly one of the most salient and important aspects of peer support. For parents whose lives may be heavily intertwined with people telling them what to do (e.g., partners, extended family members, child protective service workers), the dignity of choice, of having a support person who does not mandate or require a particular response seems nothing short of essential.

However, there may be instances when the question is not really about the autonomy or wellness of the parent but, rather, the implications of the parent's behavior or actions for a young child. This may present new challenges for peer support specialists; indeed, supporting a parent's autonomy to make less than ideal decisions may convey far-reaching implications. With respect to peer supports for parents, this suggests two related ideas. First, peer specialists working with parents in the home may experience more moments of dissonance or distress than they might otherwise in working with adults with no children or in settings outside the home. The peer specialist working in the home may have greater exposure to the consequences of a parent's decision-making (e.g., an empty refrigerator). Peer support specialists may benefit from targeted training, coaching and support from supervisors and peers, and from knowledge about specific supports and resources for the parents and families with whom they work, to deal with this increased exposure. The very opportunity for enhanced relationship-building conveyed by shared lived experience also may result in peers being aware of issues about which more traditional office-based clinicians have no knowledge.

Second, peer supports may not be right for everyone—the parent or the peer. Even well-trained peer specialists may not feel comfortable "intruding" in the parent's family life. Peer support specialists may be asked to interact with family members, school personnel and other professionals. This may be distinct from work undertaken by other peer support specialists. It may not be lived experienced of parenting, *per se*, that makes the work successful but, rather, the ability to tolerate ambivalence and utilize personal disclosure in a way the fosters a sense of possibility, regardless of family circumstance. This suggests that peers who work with parents may not necessarily have to be parents themselves. Similar issues have been raised in discussions of "peerness," the fit and match of peer specialists and patients or clients (28, 36). Even individuals who are not parents themselves have lived experience of being parented and family life.

Study Limitations

This was an exploratory study with a developmental design. While the number of participants was relatively small, they drew from considerable breadth and depth of experience—as professionals and as parents. The needs and experiences of parents with SMI identified by informants were consistent with those identified in previous research and in other countries. In subsequent stages of the study we propose to specify fully, implement and test a model of peer supports for parents with SMI that will be in many ways determined by the context in which it sits. Peer support models have developed in the USA to fit not only the regional or local service system context and need, but to fit the fiscal structure of the American health care system and reimbursement for services. Therefore, the study findings generally reflect parent/family and system characteristics and conditions known to Massachusetts, though the participants included a number of national experts from other locations. While a peer support model for parents with SMI may well have international application, given the similarities in the needs and experiences of parents across countries, aspects of the implementation context must be considered if the model is to be adapted and replicated in other sites.

CONCLUSIONS AND IMPLICATIONS FOR FUTURE PRACTICE AND RESEARCH

Moving forward to inform a model of peer supports for parents, consistent themes drawn from data obtained to address the original research questions can be grouped into four core program elements: engage, explore, plan, and access and advocate. These core elements or categories of activity are likely founded on practice principles that include a focus on families and their strengths, cultural sensitivity, and acknowledgment of the trauma experienced by many parents living with mental illness, at home (e.g., when they lose custody of children) and in the system of services (e.g., if they are approached by others through the lens of negative attitudes and expectations). Further translation of core elements and practice principles into peer support activities, and the implementation and testing of the peer supports model for parents with SMI remain the focus of next steps in the multi-stage study. We anticipate that parent peer specialists will require additional training and coaching to supplement content covered in the state's mandatory training of peer support workers. The training and coaching materials will be targeted and relevant to the population of parents served. The content of these materials will be determined in consultation with key informants and other engaged stakeholders.

As noted in the introduction, a number of questions have emerged about the efficacy and impact of peer supports in behavioral health services. However, in thinking about peer supports for parents, the question isn't about whether peer support makes a difference, but under what circumstances and with whom (21). This question needs to be asked not only of the potential parent patient or client, but of the potential peer support specialist as well. Not everyone has an affinity for this work. This is a simple statement, yet one that needs to be explicitly addressed. Clearly, those who choose to provide peer supports to parents must be supported themselves (24), if their goals and the goals of the parents with whom they work are to be met.

Aspects of the organizational context must be considered along with specification of the characteristics of parents suited to this approach and the attributes of peer specialists providing support. The job description of peers working with parents with SMI must be well-specified, with adequate training, coaching and supports spelled-out and provided, to ensure that peer specialists are activating core program elements and processes, without putting their own well-being at risk. The role of peer specialists vis a vis the roles of others in the service provision setting must be clearly articulated, with organizational players prepared for and supported to promote productive collaboration and respect for the contributions of each. It is fair to assume that, amongst clinicians, there are individuals with experiences of mental illness and family life, though they may not disclose nor explicitly draw from their lived experience as do peer specialists. All staff members may require organizational leadership and support in taking a family-focused approach. A fully-articulated model of peer supports for parents with SMI, therefore, must include parallel theories of change for the workforce as well as for participating parents, to support well-being in the context of peer relationships and the success of parents in family life.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of the Dartmouth College Committee for the Protection of Human Subjects (CPHS#00030199) and the Massachusetts Department of Mental Health Institutional Review Board (Protocol#2017-04) and approved by these committees. Written informed consent was obtained from all participants.

AUTHOR CONTRIBUTIONS

JN: designed the study; JN and AV: collected the data, performed analyses, interpreted results, prepared sections of the manuscript, and contributed to manuscript revision, read, and approved the submitted version.

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Developing an Intervention to Improve the Health Related Quality of Life in Children and Young People With Serious Parental Mental Illness

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Introduction: Children and adolescents living with parental mental illness (CAPRI) are at increased risk of behavioral, social and educational difficulties, mental and physical health problems and have poorer quality of life (QoL). Adverse outcomes can extend into adulthood but are not inevitable. Recent policy and stakeholder consultation recognize the urgent need for interventions that extend beyond objective, service-led measures of health. Systematic evidence synthesis has demonstrated a lack of evidence-based interventions for enhancing holistic, child-centered outcomes. We aimed to co-develop a manualised, community-based intervention to improve QoL in CAPRI. Precedence was given to the QoL domains that were prioritized by stakeholders and deemed feasible to modify within a health and social care context. We describe here the modeling phase of developing the intervention emphasizing co-production activities with CAPRI, their families and professionals who support them.

Methods: Semi-structured interviews and focus groups with CAPRI ($n = 14$), parents ($n = 7$), and professionals from health, social and educational sectors ($n = 31$) in the UK. Topic guides qualitatively explored participants prior experiences, unmet needs, perceived barriers and facilitators to receiving/delivering support, and their ideals for a new intervention. Findings were synthesized with existing research evidence and presented to a mixed panel of clinical academics and health and social care professionals. A consensus exercise was used to identify the preferred structure, format and content of the manualised intervention.

Results: An 8-week group intervention for 6–16 year olds and their parents, called Young SMILES, has been co-developed along with associated training materials for facilitators. Each session addresses an identified need, but is underpinned by cross-cutting themes pertaining to mental health literacy, parent-child communication, and problem solving skills. Sessions are delivered by two trained facilitators and held in accessible and acceptable community locations weekly for 2 h.

Conclusion: Young SMILES captures a broad age range and level of need for CAPRI and can be evaluated with quantifiable child-centered outcomes. In line with current policy directives, this is the first UK-based, multi-context intervention to improve QoL in this population. Implementation and referral mechanisms are currently being evaluated in a multi-site feasibility trial.

Keywords: co-production, children, young people, serious mental illness, parents, intervention, health-related quality of life

INTRODUCTION

Children and adolescents living with parental mental illness (CAPRI) are poorly provided for in current social care and educational settings (1, 2). Children and young people (CYP) growing up in families affected by parental mental health disorders have an elevated risk of physical health (3–5) and mental ill health (6, 7), attention or educational difficulties (1, 8, 9) and emotional and behavioral problems (10–12). They may be more socially isolated (13), and some will experience neglect (14), family separation (15), or social care involvement (16). Longer term outcomes for these children extend into adulthood and include chronic psychological difficulties, social and occupational dysfunction and substance misuse (17).

Risk of adverse outcomes is of increasing public health concern. Population estimates from the United States, Sweden and Australia suggest that between 38 and 50% of women with serious mental illness (SMI) will be mothers and approximately 25% of men with SMI will be fathers (18). In the UK (and Sweden) 1 in 4 children at any time are living with parental mental illness and, by 16 years of age, with over 50% of children will have experienced parental mental disorder of a severity sufficient to present to services.

Adverse outcomes are not inevitable and the impact of parental mental illness on children's outcomes is modifiable. Data suggests that at least half of all children with a parent with mental illness may not experience any psychiatric symptoms (19), and only a small proportion will access mental health services (10). Individual and family resilience can be heavily determined by an ability to find positive meaning in challenging events, to recognize a need to change social interactions or environmental conditions, and the increased availability of health-sustaining resources. Thus, how a parent or child makes sense of their experiences of mental illness may be as, if not more, important than the actual experience itself.

There is a growing literature exploring community-based interventions for CAPRI which has been comprehensively reviewed (20). Traditionally, parents have been considered the primary change agent for their children and parent-centered interventions have been evaluated for secondary effects on child outcomes. Quantified outcomes have primarily been behavioral or psychological in origin, and mediated by improved parenting or enhanced parental health (11, 21). Meta-analysis suggests that such parent-centered interventions are of variable quality and reliability (20).

Increasingly child and family-based approaches are permeating research arenas and these interventions offer an alternative and promising avenue for change. Child-centered interventions seek to establish the child as the major change agent. Research and consultation has suggested that children often have a different view of their situation, and a different idea of what would help compared to parents or mental health workers (19, 22). A philosophical shift away from "children as patients" has led to clinical, therapeutic interventions being replaced by more strength-based approaches. Peer support interventions have been advocated as a possible means by which to provide respite and reduce the isolation for CAPRI but are often not standardized or time bounded to a degree that facilities their roll-out or implementation across resource-constrained services. Additional work is needed to agree intervention priorities, identify the most likely active ingredients, and deliver these in the most cost and time efficient format.

In evaluating adult and services, focus has moved from simply the absence of disease to a more holistic approach, recognizing that improving the QoL, and specifically health-related QoL (HRQoL) as has great significance (23). QoL concerns how an individual perceives their own well-being and life experiences with respect to their personal beliefs, goals and expectations (24). In the context of severe parental mental illness, it can thus be conceptualized as both outcome and a modifier of children's short- and longer-term resiliency.

Multi-dimensional models of QoL reflect the scope and complexity of an individual's QoL judgments and offer a coherent, empirical based framework through which to potentially maximize intervention impact by simultaneously addressing multiple QoL domains. Targeted approaches to modifiable dimensions of QoL have the potential to improve the shorter and longer term availability of internal assets as well as external resources. These combined resources can strengthen protective factors, counteract or moderate risk factors and help to achieve positive adaptation in the face of adverse life experience (25–30). Effective adaptation can in turn, precipitate improved QoL appraisals, creating a positive feedback loop and maximizing return on investment.

Systematic evidence synthesis has demonstrated a striking lack of evidence-based interventions for enhancing holistic, child-centered outcomes. Bee et al. (20) identified only three trials focused on CYP with severe parental mental illness; and none of these explored QoL outcomes of CAPRI. This knowledge provides a compelling argument for a focused exploration of CAPRI need and the theoretical development, delivery and evaluation of novel, effective interventions (2, 20, 31). This paper describes development of a new intervention called Young SMILES [Simplifying Mental Illness and Life Enhancing Skills; (32)]. Development of the intervention was conducted in line with the Medical Research Council (MRC) complex interventions framework. Following identification of existing evidence and theory development, the MRC framework stipulates modeling an intervention by identifying key components of the intervention and its evaluation prior to conducting a feasibility trial (33).

MATERIALS AND METHODS

Co-development of the intervention involved three phases: Phase 1—Needs analysis—review of existing literature and primary qualitative research with stakeholders to identify needs and preferred delivery models; Phase 2—stakeholder consensus—professional synthesis exercise to identify emerging themes and agree provisional content and delivery preferences and; Phase 3—manual development and refinement—locating stakeholder preferences in existing knowledge literature (20) and theory of change involving team work.

A planning group of research team members, including those with clinical and service delivery experience, met face-to-face monthly throughout the project to discuss existing intervention literature and materials, emerging research data and the development of the Young SMILES intervention. Additional remote meetings were held as required for manual development.

The activities and findings from each development activity follow.

Phase 1 Needs Analysis

Existing Literature

The work presented here reports on a commissioned piece of work that aims to shift the spotlight away from the medicalization of CAPRI to focus on the development of a child-centered, community-based intervention to improve quality of life for all children affected by severe parental mental illness.

Literature exploring community-based interventions for CAPRI has been recently and comprehensively reviewed (20). This review, and the intervention resources identified with it, was used as a starting point for intervention development. An existing intervention, The Family SMILES intervention (34), provided a starting template for intervention development. Family SMILES is based on the SMILES programme, a 3-day intervention for Australian CYP aged 8–16 with a parent experiencing mental ill health (35). SMILES has been evaluated positively with respect to improving CYPs knowledge of mental health and coping skills (36).

Family SMILES has been piloted in the UK but not yet rigourously evaluated. FAMILY SMILES takes a deliberately narrow approach; focusing only on CYP at-risk of maltreatment or neglect. The intervention comprised 8 weekly groups: 6–8 CYP sessions; 6 one-to-one weekly sessions with parents; and a final CYP-parent joint session with each family. Its aim was to enhance children's resilience and self-esteem and parents' protective function; and to improve parent-child communication and family relationships. Preliminary evaluation of Family SMILES has highlighted potential benefits for CAPRI in increased social functioning and confidence, reduced social isolation and reduced blame associated with parental illness. For parents, benefits included less distress and unhappiness, a shift of thinking from own needs to those of their children; and for families overall a more relaxed atmosphere, openness about parental mental illness, empathy between CAPRI and parents and shared responsibilities (34).

Agreeing the Working Aim

The working aim, agreed between the research team and the project steering group, was to broaden the scope and content of Family SMILES to make it specific to families whose parents have SMI; to make it applicable to a wider age-range of CAPRI, to align it with NHS priorities and service structures and to make it deliverable in different practice settings by a varied staff skill mix, including NHS and voluntary sector providers. In the context of the UK NHS, our intention was to co-create a child-centered approach with far broader reach (geographical and across ages and needs), with a specific focus on enhancing children's QoL. QoL was defined according to a published, empirically-led model derived specifically for the target population (37). This definition upheld QoL as a multi-dimensional construct comprising of 5 domains spanning emotional, physical and social well-being, family context and experience and children's self-esteem and self-actualization. Precedence was given to three domains identified as priority by stakeholders and potentially capable of being modified via a time bounded health service intervention. These domains comprised emotional and social wellbeing, family experience and self-esteem and actualization. A fourth domain, physical wellbeing was represented as a secondary goal of the intervention and targeted through in-session education.

Within these priorities the importance of improving problem-based coping skills, increasing mental health literacy, and alleviation of parental mental health symptoms were evident.

Recognizing the importance of stakeholders' voices, our explicit intention was to place childrens' needs at the center of the process to create a best evidence, feasible and acceptable intervention to CAPRI improve health-related QoL (2). We used this approach to ensure that a priori beliefs and existing Family SMILES components did not drive our assumptions about what would and would not work for children. To achieve this, we interviewed participants blind to the Family SMILES intervention and explored ways to support CAPRI directly and separately from the experiences and needs of their parents. Despite the incidence rates of mental illness being increased for CAPRI, as a large percentage will not experience mental illness, we anticipated an intervention relevant to all CAPRI.

Primary Research

Stakeholders, including children and adolescents, parents and practitioners from NHS and voluntary settings (including managers), were invited to participate in discussion groups and individual interviews if they preferred. Individual discussion groups were held for each stakeholder group to maximize opportunities for participation. Consent was taken from

TABLE 1 | Stakeholder consultation methods and participants.

	Young People ($n = 14$)	**Parents ($n = 7$)**	**Practitioners ($n = 31$)**
Method:			3
Focus Group (number conducted)	2	1	
Interview (number conducted)	2 face-to-face	2 telephone	
			0
Family SMILES experience (n, %)	Previous experience 6, 43% No previous experience 8, 57%	No previous experience 7, 100%	Previous experience: Voluntary organization A practitioners (10, 32%) No previous experience: Voluntary organization B practitioners (15) and; NHS practitioners (6) (68%)
Gender	9 girls (64%), 5 boys (36%)	7 women (100%)	28 women, 1 man
Age (mean, range)	11, 10–16	41.14, 33–47	
Siblings (*n*, %)	11, 79%		
Live with parent experiencing SMI (*n*, %)	13, 93%		
Awareness of parental mental health type (n, %)	10, 71%		
Number of children under 17 years (mean)		2.43	
Time experienced SMI (mean, range)		14, 2–25	
Professional qualifications			Social work, teaching, counseling, clinical psychology, OT, mental health nursing, family therapy
Numbers in managerial position (*n*, %)			7, 23%

all participants prior to the discussion group/interview commencing. Parents were asked to consent for their child or adolescent taking part. For CYP, parental/guardian consent was required in addition to their assent. All parents/guardians approached agreed for their child to take part. Participation of both CYP and their parent was not necessary, but all were invited. Practitioners in the recruiting sites initiated contact with families and assisted in obtaining consent. Participants were asked to complete a short demographic questionnaire at the beginning of the discussion (separate questionnaires were developed for each stakeholder group). Discussion groups and individual interviews were held at a community location convenient to the participants. Travel expenses were reimbursed and refreshments provided.

Attendees were informed about the aim of the study and the value of their involvement in developing a new intervention to improve the QoL of CAPRI. The terminology used was altered for each stakeholder group to ensure understanding. Semi-structured topic guides explored experiences of previous support, unmet needs, barriers and facilitators to receiving/delivering support, gaps in current care and what an ideal intervention would look like. Interviews and focus groups were audio recorded or notes were taken. For CYP, methods to enhance engagement, such as using post-it notes to provide views anonymously for discussion, emojiis to express feelings about aspects of their ideal intervention and pens to draw were implemented. Data were analyzed using thematic analysis (38).

Interviews and focus groups took place between June and October 2016. Participants were recruited via two different voluntary organization regional branches (A and B) and the NHS (practitioners only). Some had direct experience of Family SMILES. **Table 1** provides an overview of the data collection methods and participant demographics.

Key themes were identified in relation to the purpose and composition of the Young SMILES intervention with comparisons between participant groups recognized. Core content topics or themes identified across all stakeholder groups highlighted that Young SMILES should educate (improve mental health literacy); reduce isolation and support. A theme about delivery preferences was also documented.

The following provides a summary of these findings and evidence of differing viewpoints.

Educative—Improving MH Literacy

Improving mental health literacy was regarded as important by young people and parents; and considered a key element of Young SMILES. This related to one's own understanding but also understanding within the wider society. For parents, lack of understanding of their own problems was acknowledged as having an impact on their views and beliefs about their ability successfully to undertake their role within the family. One parent highlighted the effect that depression can have and how improving their understanding is needed for help seeking.

> *"I understand what depression is now I can see it coming and see it in other people but the very first time you're not sure if it's … you question yourself a lot. You question whether you're a good mum. Is it really you that's annoying everyone and people pick up on the signs. So at the beginning of the depression it's really understanding that you have got a mental illness yourself and recognizing it and then going to the doctors to do something about it without feeling embarrassed or stupid. That's a really big part of mental health."* (Parent)

There was a general sense, particularly among CYP, that society lacked awareness of what the experience of having a parent with an SMI is like. The negative consequences of this had an effect on their daily lives.

> *"no-one knows what I'm dealing with, they bully me because of my mum, they know I'm a young carer."* (CYP)

Attending school was challenging for many; lack of understanding and awareness of the effects of having a parent with an SMI extended beyond their peer group to their teachers. CYP were eager for changes to occur:

> *"I want them [school] to understand more, what I am as a young carer going through... understanding of why I come in late."* (CYP)

Whilst it was acknowledged that the intervention could not necessarily change society's awareness and perceptions, professional stakeholders recognized the importance of educating children and teachers about their parent's mental illness and saw this as a key element of providing support and overcoming the challenges those children can face:

> *"And that's often leaving children with a lot of space to create their own ideas of what's going on which is often much more frightening in the reality isn't it?"* (NHS practitioner)
> *"it was quite a normal reaction for the children to be able to want to understand their mother's difficulties which had been quite pronounced and defined really."* (NHS practitioner).

Reducing Isolation

As a result of living with a parent experiencing SMI, children reported that they often felt emotionally isolated. These experiences extended to their social lives, where some sacrificed activities for friends to look after their parent(s).

> *"I just can't go out with friends – need to make sure mum is OK first."* (CYP)

Parents acknowledged this was often the experience of their children and that there was often a reversal of roles, with their children taking on responsibilities they felt they shouldn't. This was distressing for parents, who felt a sense of guilt because of their inability to parent:

> *"you're having this problem you cannot get yourself out of, when you look at your children, you cannot help them."* (Parent)
> *"mental health becomes who you are."* (Parent)

Consistent with these experiences, professionals acknowledged the difficulties that CAPRI experience emotionally, highlighting their inability to understand the root cause.

> *"Yeah, they don't understand their own emotions, they don't understand the parents' emotions. They think that their parents' emotions are a reaction to their behavior, which sometimes that is what's going on, and the child takes all the responsibility for that."* (Voluntary organization A practitioner)

It was acknowledged that an intervention focused on children's needs has to negotiate the sensitivity of helping parents understand that their illness and its behaviors may adversely influence their children.

> *"It's that introductory process really, around, not just introducing what [the intervention] is about, but you're introducing the concept that there's some idea that this issue, that this parent has probably been living with for 20 years or more, can potentially impact on their children, and impact on how they are parenting their children."* (Voluntary organization A practitioner)

Support

Practitioners recognized the need to provide support to CYP and parents. For all stakeholders, the value of support on a predominantly emotional, but at times practical level was considered important. The value of providing a group intervention including peers was recognized as a key element. Many saw it as a way of reducing isolation among CYP and parents. Reflecting upon feedback from families they had worked with previously, one practitioner stated:

> *"It's ultimately about I thought it was only us who was struggling. I thought it was only me that at times hates my mum. And from a parent's perspective, I thought it was only me that is really struggling with my teenager, and all those issues. So I think there's also that experience really, about coming together and that mutual support and that I'm not on my own. And that there are similar experiences shared really."* (Voluntary organization B practitioner)

The additional value that involving their parent in an element of the intervention would provide was thought of positively across both sectors, with additional benefits for the family identified:

> "*And I think permission as well, I think permission to have conversations, that families... that they might be thinking of in their heads but not actually have the courage or feel they've got the permissions to have those conversations with each other."* (Voluntary organization B practitioner)

The importance of helping parents to overcome their unwillingness to approach difficulties via a more family or child-centered approach was acknowledged. Practitioners identified that they could play a supportive role to promote parent engagement:

> *"... my experience is often parents don't want you to, or they're maybe a little bit more reluctant, to have children involved say with the family meeting. And children will often be at school when people are calling... I sense there would be a reluctance and so some of the skill is about connecting to parents I think initially and maybe doing some work there about what maybe could be talked about and things and whether they can do some of that and we can support them in doing that."* (NHS Practitioner)

Parents recognized that talking to their children about mental health was challenging; particularly when they were "*really bad*" and that providing their child with the opportunity to speak to other CYP in similar circumstances helped their child to address some of the issues they were facing via different supportive avenues.

> *"It's like when you're talking to your own friends isn't it, you can open to your best friends and your partner, and whatever. But sometimes it's like, well, I can tell her anything but not about the mental health because you don't want them panicking... But when they're together and they've been through the same experiences... they can open up and they can say, my mum's done that. And then your [other parent's child] little one might say, well, my mum does that as well, or, you know, stuff like that, so they're not alone."* (Parent)

CYP and parents valued having the opportunity to have some "respite." Within the discussions it was evident that there were similarities between the views of parents and CYP such as the recognition of the importance of retaining their family unit but subtle differences were identified. CYP expressed a desire to receive support in an environment separate to their parents, for the most part, to discuss the difficulties they were experiencing and they felt being away from parents would make it easier for them to do so and reduce any impact this may have upon their parent:

> *"Or like anything's happening, any bullying or anything like that, if it did happen to me, I'd rather speak to [voluntary organization] about it than my mum because I don't want to put that pressure on my mum and everything... if it's something that's gone on at school or something that's happened, I have to keep it to myself. If I did keep it to myself, I would be okay with it, but then I'd just get a bit worried."* (CYP)
> *"My mum makes me nervous". (CYP)*

Parents mirrored these views, identifying the importance of '*children having a separate opportunity*' (Parent), but also recognized that, at times, their children's sense of responsibility for them was overwhelming and became a barrier to engaging in activities without them:

> *"I think it would be better if the children were separate and they had, like, a little group together and then all together. I know when our [child's name] had a first referral to [the voluntary organization] and she [facilitator] was, like, come with me. I said, she's dead nice. Because I'd spoken to her before and we'd gone through it all. I said, she's dead nice, she's lovely. And our [child's name] usually... if you meet her, she's, like, hello. And she was, like, no mum. And she got hold of my hand, you're coming with me. And I'm, like, are you sure you want me with you? ... We're both turning up, she doesn't want to leave me by myself."* (Parent)

Delivery Preferences

For CYP, it was important that they had "*fun*" and that despite Young SMILES being an opportunity to learn more about their parents' mental health, it was vital that did not mirror the school environment, offering an opportunity to learn in non-traditional formats.

> *"Instead of just sitting down and talking... doing an activity... engaging in a different way so it doesn't feel like you are in school." (CYP)*

CYP focused their discussions on activities that could be included to ensure it was "fun." They wanted an opportunity for relevant team building and physical activities and somewhere to share their own experiences. Older children additionally expressed the value of "*anonymous self-expression*." Whilst they recognized the potential use of technology in the activities included in the Young SMILES sessions, they were reluctant for it to be incorporated because of its potential effect on their ability to engage with others in the group highlighting it was *"anti-social" and reflecting on the impact that involving technology may have upon the intervention:*

> *"It's [technology] wasting our time together." (CYP)*

While parents acknowledged the need for "*fun*" and allowing their children to experience "*childhood*" they focused less on the type of activities they thought Young SMILES should contain; predominantly exploring the potential outcomes that could be achieved for their children such as encouraging independence/increase confidence, educating and normalizing:

> *"Because they kind of have that attachment to us [our children] and that worry, that anxiety. And that's true really, because they don't want it, they're constantly seeing you on the couch, you know. So that time when they are go off [to Young Carers activity] not to worry, because that's what they're there for. So they kind of get used to that, oh mum can be okay when I'm not around."* (Parent)

Practitioners identified that "selling" the intervention was important and, in doing so, they needed to understand what would be attractive to CYP from an individual perspective:

> *"But they would all have their different reasons for coming, won't they? I mean some may come back because they found it was fun, some may come back because they liked the food, some might come back... So everyone's coming from a different angle, young people and even parents. So it's about trying to attract them in some way, and we do that quite well with our activities and stuff, to try and sell it to young people. You think well, I know they like coming because their friend comes, or they like coming because they like that. So they all have different reasons, motivations for coming."* (Voluntary organization B practitioner)

Phase 2: Stakeholder Consensus

As part of our co-development methodology, a stakeholder synthesis day was held. The aim of the synthesis day was to review existing and new research knowledge in order to agree provisional content and delivery preferences for intervention. The synthesis day was open to all individuals currently working with or potentially working with children of parents with severe mental illness in the future. This involved practitioners, academics and managers representing voluntary sector and NHS services attended. During the day, key findings were presented to professional stakeholders alongside existing research. A consensus exercise was used to identify the preferred structure and key components of the finalized intervention, focusing predominantly on implementation and delivery. Tensions between findings and views were explored and acknowledged.

Two qualitative research team members facilitated the day. The aim was to use group consultation to identify agreement and to resolve areas where the evidence was ambiguous or less established. There were three consecutive activities:

1. Listening
2. Identifying gaps and ambiguities
3. Reaching consensus

Nineteen people (9 research team members and 10 non-team individuals) attended the synthesis day. A variety of different professional organizations and roles were represented including those working in academia e.g., health service researchers, psychiatrists, psychologists and a PhD student and voluntary sector organizations e.g., social workers, team managers, business managers and NHS organizations e.g., family therapists, mental health nurses). Attendees were experienced in at least one of the following—working with vulnerable CYP (who may or may not have parents experiencing SMI); working with vulnerable families (where there may or may not be a parent experiencing SMI); working with adults or children with mental health difficulties; or conducting research in the field of adult or CYP mental health.

1. Listening

   Key messages that emerged from existing literature and Phase 1 stakeholder consultation were presented. Attendees were asked to think about what the data implied about what Young SMILES should look like and to make notes that would be useful for activity two.
2. Identifying gaps and ambiguities

   Attendees were asked to reflect on the information from activity one in order to populate a *synthesis matrix*. The matrix aimed to ascertain three areas: intervention format (e.g., content, facilitation, delivery); intervention resources (e.g., training manuals, service resources), and any additional relevant information.

   The two workshop facilitators reviewed the matrix, identifying any contradictory or missing areas. **Table 2** presents a summary of aspects that participants were in agreement about and those where some inconsistencies or evidence gaps were acknowledged.
3. Reaching consensus

   In response to the findings from activity two, attendees in four small multidisciplinary groups were asked to discuss one of the four areas where gaps in the evidence or ambiguities had been identified (detailed in **Table 2**). Participants were told that the aim was to draw upon their own experiences, express their views to the group and listen to those of others to reduce uncertainties and to come to a consensus.

   Summaries of the discussions, including a clear rationale, were fed back verbally to the wider group.

   Following the synthesis day, the planning group consulted with the steering committee and parent representatives prior to finalizing the Young SMILES intervention. The steering committee and parent representatives reviewed and agreed the proposed intervention format, guidance and training materials. Innovation of Theory of Change was used as a guiding methodology for finalizing the change model underpinning the intervention. The intervention was intended to be child-centered and the primary outcome of the intervention, determined a priori at study commissioning stage was enhanced QoL.

TABLE 2 | Summary of the outcome of activity two discussions.

Agreement	Inconsistencies/lacking evidence
Aim: Respite, social networking, accessing help, integrated services/wider engagement [Mental Health Education] **Content:** Safety plan, mental health education, signposting, communication skills, practical support for parents **Who:** Discrete age groups **Where:** Transport **Who:** Support worker for parent **Engagement:** Build trust, consolidate peer support, develop between session resources **Resources:** Assessment is crucial, food **Facilitator training:** Group management skills	1. Intervention resources 2. Delivery format • Group composition • How long and over what time period/legacy 3. School/multi-agency liaison and engagement 4. Measures/legislative frameworks • Assessment • Progress/success

RESULTS

Figure 1 presents the Theory of Change diagram developed as a result of consultation and consensus activities.

Theory of change conceptualizes Young SMILES with respect to the problems faced by CAPRI, Young SMILES inputs and change mechanisms, primary outcome(s) for children and impact upon associated risk of negative outcomes. It additionally takes into account the QoL domains and priorities as determined by CAPRI within previous literature [e.g., (20, 37)]. The change model built on Phase 1 data synthesis and Phase 2 stakeholder consultation and as such was inclusive of multiple perspectives and participants in its design. It required stakeholders to make a distinction between desired and actual outcomes and to identify their desired outcomes before deciding on possible intervention content and processes to achieve those outcomes.

Processes that normalize children's experiences, improve their social and peer support networks and/or enhance their mental health literacy and problem-based coping skills are upheld as important change mechanisms and influenced both the format and content of the Young SMILES intervention. Children's psychological resistance is dependent on these elements to ensure enhanced wellbeing or protection from the impact of potential risk factors by enabling them to gain a better understanding of parental SMI and interact with their family context. Similar priorities have been reported in the literature (39, 40), and empirical work (41). Studies have identified that many CAPRI

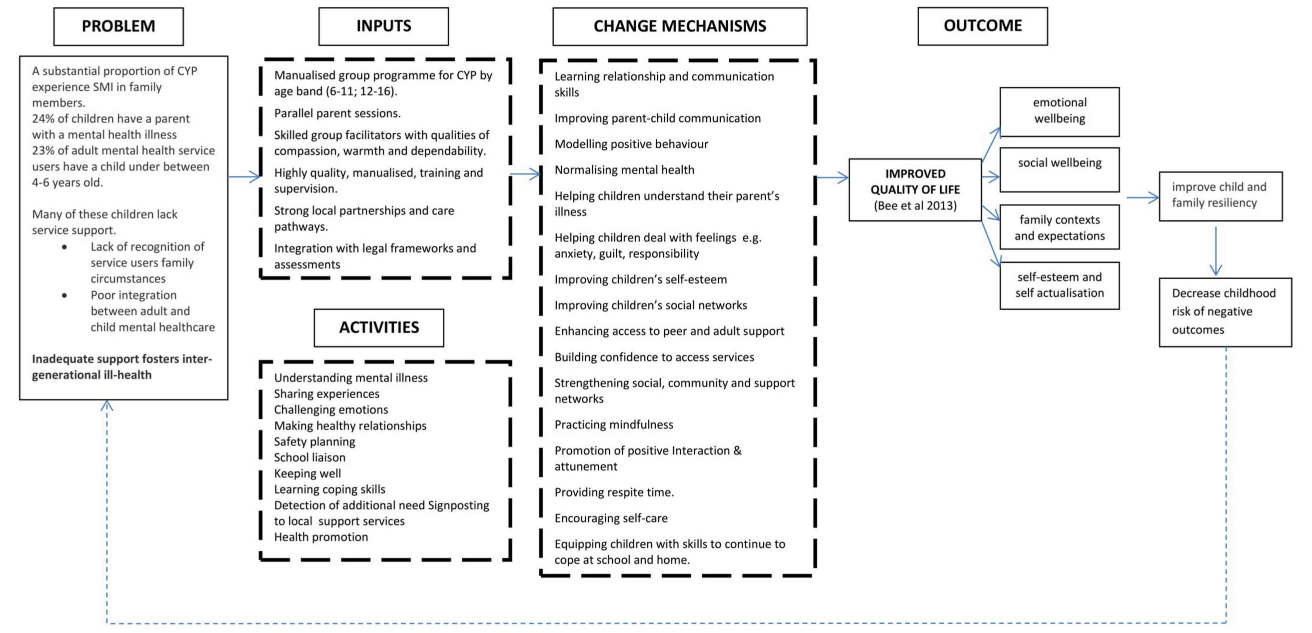

FIGURE 1 | Young SMILES theory of change.

adopt a caring role for this parent(s), a responsibility that can extend to looking after or supporting other family members (42). The importance of Young SMILES including problem-focused approaches to enhance coping strategies was identified as an important element among stakeholders to empower children to maintain their long-term emotional health. The adoption of a group format, and the inclusion of parent sessions, was hypothesized to strengthen children's and young people's support and encourage child-centered developmental opportunities. Establishing and strengthening social networks is recognized as one potentially effective way of enhancing self-management capacity and well-being (43), and positive family and peer interactions are recognized as important contributors to children's QoL (41, 44).

Programme Outline

On establishing the theoretical framework, the planning group finalized the intervention outline and facilitators' manual. Resulting data showed some overlap with Family SMILES such as the involvement of parents, the opportunity to meet other children in similar circumstances and increase knowledge, as well as the need for new components. Integration of these two led to Young SMILES being recognized as separate from, but a derivative of, Family SMILES. It was called Young SMILES in recognition of common components.

Young SMILES is a manualised 8-week group programme for CAPRI designed to work with small groups of children/adolescents: it is recommended that a minimum of 4 to a maximum of 6 children/adolescents are involved per group. Its explicit focus is to improve children's health related QoL. The wide age range of Young SMILES groups is split according to the school the young person attends, either primary (6–11) or secondary (12–16). Each group work session is allocated a 2-h time slot, which includes time for a short break and refreshments (with parents/carers) during and after the group. The structure of each session is presented in **Table 3**.

TABLE 3 | Session structure for CYP and parent sessions.

CPY sessions (week 1–8)	Parent sessions (week 4–8)
• **Welcome:** 10 min	- **Welcome:** 10 min
• **Warm-up game:** 10 min	- **Warm-up activity:** 10 min
• **Activity 1:** 10 min	- **Weekly Reflection:** 20 min
• **Made-up family:** 20 min	- **Snack Break:** 10 min
• **Snack break:** 10 min	- **Feedback from children's sessions:** 20 min
• **Activity 2:** 10 min	
• **Weekly home activity:** 10 min	- **Wrap-up discussion:** 20 min
• **Wind-down game:** 10 min	- **Groups join for end snack:** 30 min
• **Snack and closure:** 30 min	

Within each session the following is always covered:

- "Ice-breaker" warm-up activities, including links to previous sessions, to enable the group to recap the main learning points and raise and discuss any issues or questions.
- "Checking in" to identify how things have been since the last session and identifying if any of the CYP need individual time to talk over any particular issues that may have happened since the last session.
- Setting the agenda and objectives for the session—Facilitators set out the session's aims e.g., "today we aim to learn about

managing a crisis: who we can contact in a crisis; how to manage our feelings of fear in times of uncertainty; what to do when we think our parent is going into crisis etc." CYP are given the opportunity to tell the facilitators what they would like to learn or achieve and if they had any anxieties about the session.
- Education and interaction included presentation and discussion of information conveyed via flip charts or drawings, videos, play, creative writing, case studies, and scenarios, all relevant to the learning objectives of the session.
- "Wrapping-up" at the end of the session to elicit feedback on the session, recap on the main learning points, answer questions and agree on activities to be done between sessions and a brief taster of what the next session will cover.
- Getting together as a group for something to eat with in communal space before going home.

At week 4 of the CYP's work group, parallel sessions are offered to the parent/carer who is unwell; and to an additional significant adult in the child's life (identified by the child and their carer) who might attend with the unwell adult, or attend in their place. From week 4 onwards, the CYP and parent groups get together for a snack at the end of the sessions.

Based on stakeholder consultation, we assigned different objectives to each session, but all CYP and parent sessions were underpinned by three emergent themes: mental health literacy, communication and problem-solving skills. Outlined in the facilitator's manual are activities that can be used to achieve the aims of the session. Whilst the overall aims and objectives of each session should not be altered the ways that they can be achieved is flexible to ensure responsiveness to CYP and parent needs. **Figure 2** provides an overview of the structure and aims of each proposed CYP (8) and parent (5) sessions. CYP and parent sessions were delivered by two trained facilitators from the NHS or voluntary organizations and held in accessible and acceptable community locations for 2 h each week.

Intervention Refinement

The final intervention was presented to service users (parents), practitioners and the trial steering committee for feedback and refinement. Positive feedback was received with individuals indicating that it adhered to the aims of the overall feasibility study and was an accurate reflection of what has been identified via exploration of the research evidence and the views of CYP, parents and professionals. Despite this, some practitioners assigned to delivering the intervention requested that additional step-by-step guidance to conduct the in-session activities be provided to improve their understanding and, in some cases, confidence. A more detailed structure for the in-session activities could also support fidelity to the intervention's objectives and consistency of delivery across sites.

DISCUSSION

CAPRI are a growing and vulnerable group of multiply deprived young people whose QoL is compromised significantly (2). Little specific provision is available to meet their needs in current services where the focus has been on the parents. In a wholly novel approach, we undertook a series of consultations, focus groups, and a synthesis workshop with CAPRI themselves, with their parents and a broad range of professional stakeholders involved in the support of CAPRI to co-develop a child-centered QoL intervention. Our intervention development aimed to maximize the involvement of all stakeholder groups at the same time as minimizing burden, but further consultation with CYP in the refinement stages of Young SMILES manual could have been beneficial.

There was consistency and overlap between the perceived needs of the CYP, but parents and professional stakeholders did not appreciate their requirements in detail, nor did they appreciate their need for more basic, quotidian support. Although mental health literacy, communication, and problem solving skills emerged as themes for all, CAPRI were clear in wanting peer-focused help and information to understand and manage their parental illness away from their parents in their own ways and in their own space. The young people described feeling isolated socially and in other ways; as well as lacking support and understanding from teachers and schools with a need for greater recognition about their situation by peers, schools and teachers. It was recognized that helping parents understand how and when their illness and behaviors influenced the lives of their children adversely was important but this communication needed some sensitivity. The consistency between perceived needs amongst stakeholders was inconsistent with previous research (19). This could be as the CYP and parents involved were already engaging with services and as a result may have been more informed about the impact that parental mental health was having within the family. The focus of the questions within focus groups and interviews around what an ideal intervention to meet their needs, rather than focusing solely on what their specific needs are, also may have influenced the commonality of responses.

These insights helped us to create a novel, child-centered intervention to deliver away from parents initially in group-based, peer-focused sessions over 8 weeks. It included valued elements for CAPRI such as fun, creative and physical activities and snack times with parents in the latter half of the intervention weeks. Thus, we were able to prioritize CAPRI needs within the wider context of ensuring the feasibility and acceptability of a multidisciplinary team-developed intervention that integrates best practice from mental health and social care.

Young SMILES may provide opportunity for greater collaboration between NHS and voluntary organizations to support CAPRI and mean individuals in different sectors work together and share their knowledge and expertise to meet the needs of this vulnerable and underserved population. Young SMILES, therefore, optimizes the potential value, impact and

FIGURE 2 | Young SMILES CYP and parent session overview.

scalability across statutory services none of whom traditionally are not targeting this group which occupies a space between the health and social care interface.

Our future work lies in piloting the broader acceptability and feasibility of the new programme and testing our ability to deliver it to scale within a randomized controlled trial method for future evaluation of clinical and cost effectiveness.

CONCLUSIONS

There is a clear need for a child-centered, specific and focused approach to supporting vulnerable young people living with severe parental mental illness across the UK and globally (2). We have recognized the need to consult closely with children themselves and have co-developed an intervention shaped by their input and understanding with an emphasis on peer support separate to the support their parents receive. Future formal evaluation of Young SMILES will help to consolidate these close links with CAPRI keeping them at the center of solutions to the difficulties they face on a daily basis.

ETHICS STATEMENT

The study was approved by North West - Greater Manchester East Research Ethics Committee (Ref: 16/NW/0207 13th April 2016). Approval was also obtained from Barnardo's Research Ethics Committee (BREC) 26th July 2016. All participants gave written informed consent in accordance with The Declaration of Helsinki.

AUTHOR CONTRIBUTIONS

JG, PB, DH, LG, CC, and KA participated in the development of Young SMILES. JG, PB, and AK analyzed the qualitative data. JG, PB, KA, LG, and HH prepared the manuscript. All the authors read and approved the final manuscript.

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REVIEW
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An Online Intervention to Promote Mental Health and Wellbeing for Young Adults Whose Parents Have Mental Illness and/or Substance Use Problems: Theoretical Basis and Intervention Description

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The transition to adulthood can be a vulnerable period for certain population groups. In particular, young adults aged 18–25 years who have a parent with mental illness and/or substance use problems face increased risks to their mental health compared to same aged peers. Yet these young adults may not have access to age-appropriate, targeted interventions, nor engage with traditional face-to-face health services. To support this vulnerable group, services need to engage with them in environments where they are likely to seek help, such as the Internet. This paper describes the risk mechanisms for this group of young adults, and the theoretical and empirical basis, aims, features and content of a tailored online group intervention; mi.spot (**m**ental **i**llness: **s**upportive, **p**reventative, **o**nline, **t**argeted). The participatory approach employed to design the intervention is described. This involved working collaboratively with stakeholders (i.e., young adults, clinicians, researchers and website developers). Implementation considerations and future research priorities for an online approach targeting this group of young adults conclude the paper.

Keywords: young adults, parents, substance use, mental illness, online intervention

BACKGROUND

A key risk factor to young peoples' mental health and wellbeing is having a parent with a mental illness and/or substance use problem (1). The mechanisms underlying this association are accumulative and involve interactions between genetic, individual, parent, familial, environmental, and societal factors (1). Parental mental illness or substance use problems have been associated with various adverse outcomes for young people, including the development of their own mental illness or substance use problem, academic failure, high incarceration rates, and stress-related somatic health conditions such as asthma (2–5). These adverse impacts may be maintained into adulthood. The transition period from adolescence to adulthood is a critical time to intervene and attempt to reduce the onset of mental health problems (6). Given that 21–23% of children grow up with at least one parent with a mental illness (7) and 11.9% of children live with at least one parent who was dependent on or abused alcohol or an illicit drug (8), it is imperative that efforts are made to reduce the risk of intergenerational mental illness and substance misuse.

This article presents the theoretical and empirical basis for an online intervention, mi.spot (**m**ental **i**llness: **s**upportive, **p**reventative, **o**nline, **t**argeted) for young adults (aged 18–25 years) whose parents have a mental illness and/or substance use problem. First, the extant literature on the mechanisms by which risk is conferred on these young adults will be reviewed, followed by service gaps and young adults' preferences for support. This literature provides the foundation for a theoretical approach to the development of the online intervention, mi.spot. The approach employed to develop mi.spot is then described, followed by an outline of the intervention. The issues associated with introducing a new intervention such as mi.spot into regular service delivery, and opportunities for future research conclude the paper.

MECHANISMS OF RISK RELATED TO MENTAL HEALTH AMONG YOUNG ADULTS

Mental health disorders account for the highest burden of disease in adolescence and young adulthood. The majority of lifetime prevalence for mental health conditions occurs by 24 years of age, and substance abuse disorders by 25 years (6). According to Arnett (9), emerging adulthood is a distinct transitional period between adolescence and young adulthood (18–25 years) that marks a major transition in terms of identity, work, relationships, and place of residence. The changing roles and increasing levels of responsibility during this period increases stress (10) which may precipitate depressive episodes (11). Additionally, substance use problems and risky sexual practices increase during these ages (10). However, young adults have lower rates of mental illness treatment than adults or adolescents (12) and only 35% of young people experiencing mental health problems seek professional help (13) due to access issues and stigma (14). Although peer relationships are integral in this developmental phase, decreases in social support, which may occur as young people leave school, are linked with increases in depressive symptoms (15). During this transitional period emotional stability is not yet established, specifically dysregulation of anger, which is high compared to other age groups (16).

There are various mechanisms involved in the transmission of mental illness for young adults whose parents have a mental illness and/or substance use problem. The first potential risk mechanism is young adults' relationship with their parent and their need for independence. Marsh and Dickens (17) found that some young adults find it challenging to separate themselves from their families to pursue normative developmental tasks such as attending university. When they do attend university, these young adults describe greater psychosocial adjustment difficulties than their peers (18). Mitchell and Abraham (19) found that amongst university students, those with a parent with a mental illness experienced higher levels of homesickness compared to other young adults. Likewise, Bountress et al. (20) found that young adults whose parents had substance use problems were more likely to have negative experiences during the leaving home transition. In turn, this predicted an increased risk of affective disorders in adulthood. They suggested that parents with substance use problems may attempt to limit their children's independence from the family of origin or fail to appropriately scaffold their leaving home transition. Family dynamics can be strained. Abraham and Stein (21) found that young adults with a mother with a mental illness reported lower levels of affection from their mothers compared to their same aged peers who did not have a mother with a mental illness. Relatedly, parents with substance use problems have been found to be more emotionally withdrawn from their young adult children and to show less sensitivity and more hostility toward them, compared to those whose parents do not have substance use problems (20, 22).

Similarly, many young adults find it difficult to be emotionally independent from their parent who has a mental illness or substance use problem. Drawn from interviews with 12 young people aged 13–26 whose parents had substance use problems, Wangensteen et al. (23) described their struggle in balancing emotional closeness and distance with their parent. In particular, those aged 18–26 felt sorry for their parent and described a sense of obligation toward them and the subsequent struggle to separate themselves emotionally when they moved out of home. As some young adults whose parents have a mental illness find it difficult to be emotionally independent, they may find it challenging to regulate their emotions (24). Such research may explain, at least partially, why some young adults experience difficulty forming close personal or romantic relationships (3, 25). Kumar and Mattanah (26) found that early attachments provide a template for romantic and other relationship developments in early adulthood, and this appears to be particular pertinent for young adults whose parents have a mental illness or substance use problem.

Role reversal, parentification or caring responsibilities are other mechanisms of risk for this group of young adults (27). They may be given or assume the responsibility of caring for their parent and siblings and maintaining the household (25). These responsibilities may have adverse impacts on employment, schooling, and friendship groups (28, 29). Developmentally, stigma and a sense of shame associated with mental illness may increase as adolescents and young adults appreciate the manner in which others in the community perceive those with a mental illness, and so impede help-seeking for themselves and their family (30).

Whether children of parents with different mental health concerns have the same mechanisms of risk and subsequent intervention needs are questions that have been previously debated in the literature (31). In two systematic reviews of the impact of different parental mental illness on children's own diagnosis, van Santvoort et al. (32, 33) found that children of parents across the spectrum present with a broad-range of adverse outcomes, not limited to their parents' diagnosis. On this basis, the authors suggest that all children can be offered the same intervention, though simultaneously suggest that additional, tailored interventions might be required, such as might arise through the exposure to violence or abuse (33). Others have also argued that across diagnostic groups, young people may be exposed to similar familial and contextual stressors such as marital discord, housing instability, isolation, and poverty (27). Other common risks across parental diagnoses might include stigma, caring responsibilities, and a lack of accurate knowledge about their parent's illness (1, 27).

Young adults have highlighted a lack of knowledge about their genetic vulnerability to addiction and mental illness (34). While younger children are often left out of explanations about their parent's illness, Seurer (35) suggests that, for parental depression at least, some young adults may be viewed by their parents as confidantes even though these discussions do not always provide the full picture of what is happening for the parent. Knowing more about mental illness/substance use and in particular their parent's specific illness can help young adults understand and distance themselves from their parent (36, 37).

During the transition to adulthood, parental control declines while the influence of peers gain in importance (38). Similarly, young adults seek support and information from their peers, rather than parents (39). Giving opportunities to share experiences with peers is important when services want to connect with difficult to engage populations (40) and is also important for young people whose parents have a mental illness or substance use problem (41). Klodnick et al. (42) found that for young adults, associating with peers who have similar mental health experiences can provide strong affectional bonds, meaningful connections and useful exchanges about services. Emerging adulthood is perhaps the first time when individuals can formulate a "new and ideally integrative understanding of one's life story" and integrate "different personifications of the self within a single self-defining life story" (43). Thus, young adults may be actively involved in the communicative sense-making processes in coming to terms with their parent's illness or substance use and can articulate and share their life experiences with others (35), and if given the opportunity, with their peers (44).

In summary, the transition to adulthood can be a vulnerable period especially for those whose parents have mental illness and/or substance use problems. At the same time, emerging adulthood represents an ideal time to develop new strategies and foci in readiness for adulthood, in relation to key developmental tasks, such as identity formation, role transition, the formation of new social connections and intimate romantic attachments, and independence from parents. Moreover, promoting access to treatment services is necessary to improve mental health outcomes and reduce the burden of mental illness, especially those aimed at increasing young adults' willingness to seek help (45).

AVAILABLE INTERVENTIONS AND SERVICE GAPS

There are various evidence-based interventions developed for children and young people aged under 18 years, who have a parent with a mental illness or substance use problem. These include peer support programs, family interventions and psychoeducational resources (46). In a systematic review and meta-analysis, Siegenhaler et al. (47) found that the risk of acquiring a parent's mental illness was reduced by 40% for children participating in targeted interventions. Most interventions for children and young people in these families draw on cognitive, behavioural, and/or psychosocial theories and deliver topics on mental health literacy, adaptive coping, and problem solving (48). However, the majority of interventions for children and young people in these families are limited to those aged under 18 years (48).

When asked about their preferred supports, adolescents aged 13–17 years and living in families where a parent has a mental illness or substance use problem, reported a clear preference for online supports (41). Extending that study, Matar et al. (44) employed a Delphi study with 282 young adults aged 16–21 years and whose parents had a mental illness and/or substance use problem and asked them what they wanted from an online intervention. Online opportunities to share with other young adults living in similar families was a common request as were topics on psycho-education, managing the parent-child relationship, and strategies to build resilience, and improve mental health, wellbeing, and coping. Finally, they wanted assurances that any online bullying would be dealt with appropriately.

There are some online interventions (mostly from the Netherlands) for young people whose parents have mental illness though these are still in the early stages of development and generally target both adolescent and young adults. These interventions include *Survivalkid* for 12–25 year olds (49, 50), *Grubbel* for 15–25 year olds (51) and *Kopstoring* for 16–25 year olds (52). To date, one randomised controlled trial evaluation has been completed on Kopstoring with positive trends found toward a reduction in internalising symptoms (52). No significant differences were found in self-reported depressive symptoms or internalising problems compared to treatment as usual in a 3-month follow-up, though the authors suggested this might be due to problems with the evaluation design (52). These interventions are based in Europe and developed specifically for those sociocultural and mental health service contexts. To date, there have been no reported interventions for this group of young adults from English-speaking countries, including Australia.

To succeed in identifying and supporting young adults who have a parent with a mental illness and/or substance use problem, services need to engage with young adults in environments where they seek help and interact. Wetterlin et al. (53) found that 61.6% of 521 young adults aged between 17 and 24 years had utilised the Internet to access information or seek help for how they were feeling, and 82.9% indicated that they were likely to use a mental health website to find information in challenging times. As young adults often prefer anonymous sources of help to traditional services (54), online interventions provide an ideal opportunity to intervene with this vulnerable group. Online approaches are important as they have the potential to link young adults with others in similar situations, especially those in rural and remote areas (55).

TABLE 1 | How mi.spot was developed.

Sources	References
Reference group using participatory design principles	(56, 57)
Theory:	
• Competence enhancement model	(58–60)
• Theory of health information seeking behaviour	(61)
• Theory of development: emerging adulthood	(9, 10)
Previous research on parental mental illness and substance use problems in particular	
• Common intervention ingredients	(46)
• Adolescents' preferences for support	(39)
• Young adults' preferences for online support	(42)
Elements of effective online interventions for young people	(51, 62, 63)
Four feasibility trials conducted, with feedback elicited from 66 participants and five facilitators	(64)

The present paper is the first to provide a theoretical overview and empirical basis for an online intervention for this particular group of vulnerable young adults from an English-speaking country. Indeed, theoretical frameworks for online interventions are often missing (56) but critical for ongoing intervention monitoring and evaluation (57).

mi.spot: DEVELOPING AN ONLINE APPROACH

The mi.spot intervention was developed iteratively over a 36 month period using participatory design principles and drawing on relevant theory, existing research and feedback from feasibility trials (see **Table 1**).

From the outset, a *participatory design approach* was employed (58, 59) through the creation and facilitation of a reference group consisting of researchers, clinicians, website designers, and young adults who have lived experience of parents with mental illness and/or substance use problems. The group was collaborative and collegiate with all stakeholders recognised as having different but equally important skills to offer. In particular, young adults were included to ensure that the intervention was user friendly and acceptable and clinicians were involved as the intervention needed to be clinically feasible. It was acknowledged that what young people want can be different from what parents and clinicians believe they need (60), which meant privileging the views of the young people on the reference group, especially in regard to site aesthetics and content and in terms of how clinicians might respond to young people's questions and concerns. Regular face-to-face meetings were facilitated with the aim of raising a diversity of views and learning from each other. Different members of the group had worked together previously (on other research projects, or on face-to-face peer support programs) which provided opportunities for capacity building. The reference group met regularly over 36 months to discuss website developments and the implications of research (see below) to the website. Notes were taken at each meeting to document decisions and follow-up meetings with individuals were conducted as required to ensure that all perspectives were considered. Overall, the participatory design approach covered several phases; the identification of problems for young people living these families (drawing on existing research, see below) the generation of solutions, the development of and feedback on an online intervention, and the development of an implementation and evaluation strategy, including dissemination procedures.

Theoretically, mi.spot is based on the competence enhancement model, discussed in detail by Barry (65). The model takes a lifespan approach to the promotion of mental health, and focuses on enhancing strengths and promoting resilience (61, 65). According to Eccles and Appleton (62), the model is most beneficial when there are explicit efforts to promote connections to others and enhance participants' competence in developmentally appropriate domains. Cognitive and behavioural approaches are commonly incorporated within the competence enhancement model (62), given their effectiveness and efficacy in improving dysfunctional cognition, regulating emotions and the promotion of adaptive problem solving (63). The theory of development for young adults was used for the current intervention, with a focus on identify formation, managing relationships and independence (9, 10). Additionally, the theory of health information seeking behaviour (64) was applied to promote optimal use of the intervention. This approach acknowledges the role of passive and active retrieval of information, and the role of peers and clinicians in helping to process and interpret information (64).

In terms of *research,* the intervention drew on previous work that identified the ingredients commonly associated with effective interventions for this target group (48). Previous research that sought the views of young adults whose parents have mental illness and/or substance use problems regarding support in general (41), and online interventions in particular (44), were incorporated. Evidence on online behaviour change interventions was used to inform online functionality, especially those designed for adolescents and young adults (53). A systematic review found that effective online interventions provides opportunities for interaction, personalized and normative feedback, and self-monitoring (66). Likewise, Short et al., (67) drew on user engagement research across multiple disciplines to identify those factors that influence how users engage with online interventions. In their model, engagement is influenced by (i) the environment including the length of time available to the user and the user's access to the Internet (ii) individual factors related to perceived personal relevance and usefulness of the intervention and their expectations and (iii) intervention design including the interactivity, aesthetics, credibility and opportunities to interact with a counsellor. These features were considered in the development of mi.spot.

Seven feasibility trials of mi.spot have been conducted since its inception, with 66 participants. According to Eldridge et al. (68) feasibility trials ascertain whether an intervention can be done, whether it should be done and if so, how. Related issues that feasibility studies may address include participants' willingness to be recruited, the time required to collect and analyze data, and the acceptability and suitability of any given intervention for both clinicians and participants. Various methods were used to recruit participants for mi.spot with Facebook advertisements found to be the most effective and efficient. Feedback from previous participants and online facilitators in semi-structured interviews were used to modify the initial iteration of the intervention. For example, many previous participants wanted to see a greater emphasis on managing friendship, work, and significant relationships and not only managing relationships with their parent and these additions were subsequently incorporated; facilitators requested additional functions such as notification of someone typing (i.e., "…") in the online group chats to allow for more effective facilitation.

mi.spot: THE INTERVENTION

mi.spot is an online, 6 week voluntary intervention for groups of up to 20 young adults (aged 18–25), who have a parent with a mental illness and/or substance use problem. mi.spot aims to (i) improve knowledge about mental health and wellbeing, (ii) promote adaptive coping, (iii) build, expand, and sustain healthy relationships, (iv) increase resilience, (v) encourage help seeking behaviour, (vi) facilitate a sense of peer connection and finally (vii) foster mental health and wellbeing. mi.spot is a manualised approach that offers real time (synchronous) and anonymous peer online networking opportunities and an individually tailored, interactive and professionally led intervention, as evident by the following functions:

- Six, one hour, professionally facilitated psychoeducational modules delivered online in synchronous mode (see **Table 2**).
- A private, online diary (called mi.thoughts.spot) to prompt and encourage participants to apply a cognitive behavioural approach to current stressful situations, with the support of a facilitator (provided asynchronously).
- Opportunities for participants to chat informally with each other on threads (i.e., topics) initiated by a participant or facilitator.
- Opportunities for one-to-one private online counselling sessions between a participant and a facilitator (synchronous).
- Video, audio, print resources, and self-monitoring questionnaires offered as weekly activities for participants in order to consolidate and extend learning from weekly sessions.

Participants may elect to join all, some or none of the offerings. The password protected site is moderated twice daily including weekends. Procedures exist for rule violations (e.g., online bullying) and risk procedures (e.g., if a participant expresses suicidal thoughts).

TABLE 2 | mi.spot weekly modules.

Module	Module description
Week 1: What's mi.spot all about?	This week provides an orientation to the intervention and various cognitive-based strategies to help participants identify unhelpful thinking patterns, reframe these in a more constructive manner, and regulate emotions. Time is spent practising these strategies.
Week 2: Learning about mental health and mental illness	Participants are offered the opportunity to ask questions about mental health and illness, with a particular focus on their parent's illness, or substance use problem. They are encouraged to discuss how their parents' illness or substance use problem may impact their own health and wellbeing. Information about generic vulnerabilities is provided with an emphasis on ways to promote mental health and wellbeing.
Week 3: Me, my parent and other relationships	Participants are encouraged to reflect on their current relationship with their parent, and how the dynamics of the parent-child relationship might influence other relationships. Strategies for healthy boundary setting are discussed.
Week 4: Managing stress	Participants are invited to identify current stressor/s and as a group, discuss adaptive coping strategies.
Week 5: Caring—who, me?	Participants are asked to identify ways they might look after themselves (i.e., physically and emotionally), the barriers to self-care, and how these might be overcome. Their caring responsibilities are discussed and integrated with the prior module on boundary setting in relationships.
Week 6: Taking control of my life	The final module consists of a general overview of key aspects of previous modules. It emphasizes strengths, provides recommendations for self-care, and encourages help seeking and the continued use of adaptive appraisal strategies.

ONLINE FACILITATOR ROLE AND TRAINING

Online facilitators for mi.spot assume an active role in encouraging ongoing, supportive conversations between participants, facilitating the weekly sessions, and providing one to one online counselling sessions as required. They also fulfil a monitoring role and ensure that the site is a safe, respectful space for everyone by identifying and addressing any instances of bullying, harassment, and racism and/or descriptions of harm to self or others.

Currently, facilitators are master's level psychology students working in a university clinic but any qualified mental health clinician would be able to deliver the intervention after appropriate training had been attended. Two days of facilitator training using manualised content are provided. The first day focuses on generic online counselling skills (in both group and individual counselling mode) and the second specifically examines the mi.spot intervention and how it should be delivered. The second day starts with a presentation from two young adults with lived experience who discuss their experiences and preferences for online interactions. The remaining part of the day is spent on simulated training scenarios to practice responses on the site. Fidelity checks for the weekly sessions are built into the intervention at the facilitator level, to ensure all topics are covered. Future considerations are to deliver the training online [as per (69)].

FUTURE PRIORITIES

Building on the current evidence base in this area (52), further randomised controlled trials are needed to establish the effectiveness and efficacy of mi.spot, including cost effectiveness. Comparing the online intervention, mi.spot, with face-to-face peer support programs will be important to evaluate comparative effectiveness. Uptake, engagement and drop out numbers as well as website analytics need to be documented to establish which participants engage with the different components of the intervention and the impact that this may have on participant outcomes. Monitoring of the intervention is required to ascertain to what extent the intervention been implemented as designed (as per the manual) and what modifications might be required. Safety is another evaluation component which may be measured through clinical deterioration related to intervention use and inappropriate use (e.g., online bullying). Developing online interventions for young people aged 13 to 18 years living in these families is another priority.

Further information is needed on how mi.spot may be embedded into services. Batterham et al. (70) identified various barriers for delivering online interventions including policy, safety, and political restrictions, incompatible reimbursement systems, limited availability of trained clinicians, and clinicians' negative attitudes toward Internet interventions. Nonetheless, a recent study found that Australian clinicians were highly supportive of online interventions for this particular group of young people as they acknowledged that many miss out on face-to-face peer support programs (71). Most research on online interventions has focused on efficacy and effectiveness trials, with little investigation on implementation processes (72), making this a priority for future research. Within these deliberations, the applicability and effectiveness of a stepped care model needs to be investigated, where mi.spot may be provided as a first step in intervention, or alternatively delivered in conjunction with individual face-to-face support. Relatedly, a strategy for targeting young people is needed. Building on the successful method of recruiting participants in the feasibility trials using Facebook advertising, other potential approaches include Twitter and YouTube advertisements and by promoting the intervention via various university and community mental health agencies. Young people's input into these approaches will be critical.

At present mi.spot is delivered from a university clinic but our vision is to expand the service into mainstream settings. Which service or services might assume this responsibility is being considered. Child and adolescent mental health services might appear to be an appropriate service but traditionally focus on those aged under 18 years. Others have highlighted the problems transitioning adolescents into the adult mental health system as well as the inappropriateness of adult mental health services for young adults (73). This is concerning, as service inadequacies and gaps during the transitional period from adolescence to adulthood have the potential for long lasting functional difficulties (74). Conversely, compared to standard adult services, age-specific interventions may increase young adults' use of mental health services (75). Given the vulnerability, prevalence, and the unique needs of young adults whose parents have a mental illness or substance use problem, it is crucial that developmentally informed and relevant services and interventions are developed and, arguably more importantly, delivered on a broad scale.

AUTHOR CONTRIBUTIONS

AR, CB, RC, KF, JM, DM, and LP contributed to the conception of the paper. AR wrote the first draft of the manuscript. RC, KF, JM, DM, and LP wrote sections of the manuscript. All authors contributed to manuscript revision, read and approved the submitted version.

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"…and How Are the Kids?" Psychoeducation for Adult Patients With Depressive and/or Anxiety Disorders: A Pilot Study

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Depressive and anxiety disorders are highly prevalent and form a substantial burden for individuals and their family members. A recent study showed that approximately two-thirds of the children of patients with severe depressive and/or anxiety disorders develop one of these disorders themselves before 35 years of age. In the Netherlands, various preventive interventions are available for children of parents with mental illnesses. However, the actual reach of interventions is small (<1% of all children participate). A major barrier appeared to be parents' hesitancy to let children participate in preventive programs. In order to address this barrier, we designed a pilot study to implement a psychoeducation program on parenting and mental illnesses. The main aim of this study was to investigate how a preventive family-focused approach can be embedded in routine adult psychiatric care. The pilot started in April 2017 and has ended in September 2018. It was conducted in the University Center Psychiatry (UCP) in Groningen, The Netherlands. This article describes the implementation process so far. The main intervention was a monthly organized group-psychoeducation called "parenting and a mental illness," which could be attended by parents currently treated in the UCP. In 18 months, implementation activities were divided in four phases; (1) Creating awareness, (2) Adoption of the intervention, (3) Implementation and evaluation, and (4) Continuation phase. The program development process was evaluated using both qualitative and quantitative data. Based on the pilot study we were able to make recommendations for the further implementation of this approach so that more parents can be reached in future. Further study with follow-up activities is needed to measure the effects of the psychoeducation, for instance on parenting functioning and the quality of the parent-child interaction.

Keywords: anxiety, depression, implementation, parental support, child wellbeing, prevention

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INTRODUCTION

Up to 45% of patients who receive adult psychiatric care are parents (1). Having a parent with a mental illness greatly affects children's wellbeing. Approximately two-thirds of the children of patients with severe depressive and/or anxiety disorders develop such a disorder themselves before their 35th year, and half of them even before their 20th year (2). These children form an important target group for prevention (3).

In the Netherlands, various interventions for prevention of mental health problems in offspring of depressive and anxious adults have been developed. Current interventions are aiming at the parents (psycho-education and parenting skills), the children (psycho-education, coping skills, mutual support), or the family as a whole (group sessions, online support) (4, 5).

However, the vast majority of children and parents does not receive the services they require (6). Regarding the Dutch preventive interventions aiming at the offspring of parents with a mental illness, no more than 1% of children take part (4, 7). The primary challenge, therefore, is to increase the reach of the preventive interventions for parents and children.

A first and crucial step in increasing the range of participants in preventive interventions is to get the offspring in the picture. A substantial group of these high-risk children can easily be detected; namely through the parent being treated. van Doesum et al. (8) found that mental health agencies are the most common source of referrals to preventive programs for offspring of parents with a mental illness. Moreover, screening on the presence of children in adult psychiatric care has already been improved in several countries. In Norway health legislation was altered in 2010, making it mandatory to assess whether or not psychiatric patients have children (9). In the Netherlands, it has been obliged to register the number of minor children of adult psychiatric patients, since 2017.

However, Skogøy et al. (10) underline that legal changes are helpful, but not sufficient to secure equal chances of protection and support for children of parents with mental illnesses. Research has indicated that both parents and professionals experience several barriers to be involved in prevention. Barriers of parents include: perceived lack of necessity for intervention, overburden, reluctance to bother their offspring, concern for shame and stigma, and practical problems such as lack of transport facilities (8, 11–13)

Professionals may perceive talking about parenthood with their patients as a sensitive topic (8, 14–16). Furthermore, between disciplines differences may exist in the ease with which professionals talk about parenting and wellbeing of the children. Findings of Maybery et al. (17) suggest that social workers feel more comfortable with talking about these family issues than nurses working in adult psychiatric care. Foster et al. (18) however, argue that nurses in mental health care are in prime positions to support parents who are patients. These authors proposed a framework for family-focused actions that psychiatric nurses can carry out at different phases of client care, such as providing age-appropriate information on mental illness to children and facilitating parental attachment with offspring. Furthermore, based on consultation with a group of senior clinical leaders in adult mental health services in Australia, Goodyear et al. (19) introduced essential and explicit practice standards for mental health nurses working with families. These efforts illustrate a growing knowledge base of what should be implemented into adult health care to serve patients who are parents and their children. However, papers on program development that investigate how knowledge can be successfully translated into clinical practice are scarce.

AIM OF THE CURRENT STUDY

The main aim of this pilot study was to investigate how a preventive family-focused approach can be embedded in routine adult psychiatric care. This aim resulted in the following research questions:

1) How can a preventive family-focused approach be made feasible and acceptable, for parents and professionals?
2) Which remedies can be identified in order to increase feasibility and acceptability?
3) Which recommendations can be given to further implement this approach?

METHODS

Design

We designed a pilot study to implement a preventive, family-focused approach for adult patients treated for depressive and/or anxiety disorders. We tried to build on existing routines so that changes were kept as small as possible. In the University Center Psychiatry (UCP) a psychoeducation program for parents already existed for a few years, but the program was not offered on a structured base, i.e., at maximum three to four times a year, at different times and dependent on the number of parents staying in the inpatient ward. A social worker of the inpatient ward coordinated these sessions and any other actions for patients regarding their children. Also, she led a special working group of professionals. Nurses were used to refer to this social worker, who invited parents to talk about the impact of parents' illness and to help explain the situation to children, on an age appropriate level.

In a four-phase implementation plan, we worked toward a situation in which all professionals mention patient's role as a parent at least once during routine treatment, and motivate parents to attend the group-psychoeducation "parenting and a mental illness" which was now offered monthly. Since it is known that parents may perceive different kinds of barriers to participate in preventive interventions, we listed common barriers and made a plan to address them in this pilot study (**Table 1**).

The medical ethical committee of the University Medical Center Groningen evaluated this study as not falling under the Medical Research Involving Human Subjects Act (WMO). Nevertheless, we followed the required procedures and obtained written informed consent for the qualitative components of the project. No written informed consent was requested for participation in a psychoeducation meeting because this option was offered as part of the treatment. Patients knew that their own therapist would be informed of their participation. Collection of participant information was kept to a minimum and reported at group level, such as the number of participants who joined the psychoeducation. In (online) registration the current privacy legislation was followed and only the necessary information was requested, such as an e-mail address confirming the registration, and the age category of the children to adjust the psycho-education on that particular evening.

TABLE 1 | Overview of the implementation process.

Barriers	Initial approach	Feasibility	Acceptability	Adjustment	Recommendations
Perceived lack of necessity to talk about parenthood and mental health	Creating awareness by providing information about the project e.g., flyers, newsletter, survey of professionals' attitudes	Overload of professionals	Professionals' view that talking about parenthood is important but does not belong to their core tasks Patients do not mention the subject spontaneously when the professional skips it	Involve management to make talking about parenthood part of the department policy Informative, normalizing online video (infographic) to increase awareness and acceptability Website with possibility for parents to sign up for a psycho-educative session themselves	Consideration of appointing an attention officer for patients' offspring Involvement of management needed to ensure that every patient who is a parent receives information about mental health and parenting Improvement of ICT facilities in electronic patient files, for e.g., a button "parenthood" and a treatment plan on parenting and child wellbeing
Reluctance to bother offspring; unfamiliarity with possibilities for prevention	Professionals should make the parent understand the importance of information for children.	Parents need support how to talk about their illness with their children	Parents' anxiety to loose custody	Relation building with parents, empathy for being reluctant while emphasizing the importance of information Peer exchange of parents' experiences; Information about further referral possibilities	Collaboration with other disciplines (public health, gp's) to continue and improve support for families
Feelings of shame and guilt ("being a bad parent")	Normalization of the attention for patients' children and their parenting roles	It appeared difficult for professionals to apply a family focused approach (which is broader than child safety procedures)	Different opinions among professionals regarding their task; who can best deal with family matters of patients and when?	Project leader attends the team meetings of all participating wards to discuss how this "child-aware" approach could best be implemented into work processes in their ward	Training of professionals how to introduce and talk about parenting with patients
Overload of the parent	Make professionals mention the topic at least once during therapy Refer to existing 2-h session of psycho-education	Acute phases of decompensation may ask for alternative ways and/or moments to raise the topic Group sessions can only take place with sufficient participation	Patients' feedback: timing is important when to talk about parenting	If there are reservations regarding the timing of mentioning parenthood, the timing will be discussed in the team meeting Psycho-education sessions are offered on a structural base to improve feasibility	Create guidelines how and when to talk about parenting and when to refer parents to the psychoeducation meeting Discussing parenthood should be adjusted to patient's condition (but will not be skipped) Automatic notifications in electronic patient file to reconsider action of practitioners at a later moment
It appeared difficult for professionals not to focus mainly on child safety procedures	Training for professionals	Due to an overcrowded training schedule, the training is postponed	Professionals differed in opinion about who could best deal with family matters of patients	Project leader attends the team meetings of all participating wards to discuss how this "child-aware" approach could best be implemented into the work processes in their ward	Postponed training will be scheduled Recommendation to document the working procedures for this point in each department

Participants

The target group consisted of adult patients who are parents of children in the age range of zero till 24 and who are currently being treated for depressive and/or anxiety disorders in an outpatient or inpatient ward. Patients will be informed by their own practitioner (psychologist, psychiatrist, psychology/psychiatry resident, or nurse practitioner), social workers, and/or nurses of the inpatient wards. The participating wards are one outpatient ward and two inpatient wards for patients with depressive disorders, 2 day-clinics for depressive disorders, one outpatient ward for bipolar disorders, an outpatient ward for anxiety disorders, and a day-clinic for anxiety disorders. Since the UCP is a tertiary care center, symptom severity of most patients is moderate to very severe.

In 1 year about 1,200 adult patients with depressive and/or anxiety disorders are treated in the UCP. It has been estimated that 30–50% of these patients have children. Systematically, information on the parent status of a patient is gathered when patients enter the UCP to receive care, via a general questionnaire. In this way practitioners know by forehand (a) whether the new patient is a parent, (b) how many children he or she has, and (c) what the age of the children is.

Primary Intervention: A Preventive, Family-Focused Approach

We performed this pilot to investigate the possibilities to permanently embed a preventive, family-focused approach in routine treatment protocols. In concrete terms, this meant that:

1. Practitioners and/or nurses mention patient's role as a parent at least once during routine treatment, and motivate parents to attend the group-psychoeducation "parenting and a mental illness."
2. Psychoeducation meetings for patient-parents are offered on a regular base.
3. We examined possibilities to permanently embed the preventive, family-focused approach in the organization and management structures.

Psychoeducation "Parenting and a Mental Illness"

We organized a monthly group-psychoeducation, on every first Thursday evening of the month, called "parenting and a mental illness." An experienced social worker of the inpatient ward for depressive disorders of the UCP and a child psychologist gave the psychoeducation, using a power point presentation. Project leader MP (resident psychiatry) coordinated applications for the evenings and was available to stand in when one of the group leaders was hindered to give the psychoeducation. Patients attended one meeting and were encouraged to bring their partners. The psychoeducation could only be attended by parents currently treated in the UCP. The content of the psychoeducation was theory-based and consisted of the following main items:

(a) Patients introduce themselves shortly (name, family situation, the current treatment status), followed by an introduction to the theme parenting and a mental illness. In particular, attention is paid to illness perception and the relation with the patient's role as a parent, before and during treatment. Group leaders pay attention to feelings of guilt or shame that may be present regarding shortcomings in the parenting role. Furthermore, the role of the other parent is a topic of conversation.

(b) How do parents tell their children about mental illness? This starts as an open conversation in which parents share their experiences. Later, group leaders give a summary and provide examples of how to talk with children about their mental illness, using age-appropriate information. Group leaders may notice barriers in talking with their children among parents, for example not wanting to bother their children and bring up these subjects.

(c) How do parents know when it is (not) going well with their children? Do they have worries about their children's wellbeing? For each age category, the group leaders provide information on signals of (ab)normal development. Sharing of experiences among parents is encouraged.

Additionally, at the end of the psychoeducation meeting further supportive options were offered to the parents, such as continued conversations about parenting with their current practitioner, or other professionals (e.g., the general practitioner, a social worker). If necessary, referral of the child for child psychiatric consultation was offered.

We estimated that 68 parents would have attended the group-psychoeducation after 1 year, based on data that about 1,200 patients with a depressive and/or anxiety disorder are treated in the UCP in a year. About 480 of the patients have children (40%) and we estimated that 336 parents could be motivated by practitioners to join the psychoeducation, with an eventual reach of 20% of the motivated parents.

Implementation Procedure

The pilot study started in April 2017 and was planned to end at the end of September 2018. In this time span of 18 months, implementation activities were divided in four phases; (1) Creating awareness, (2) Adoption of the intervention, (3) Implementation and evaluation (following an iterative process), and (4) Continuation phase. We described intended activities in these four phases in our implementation plan that we wrote prior to the start of the project. The implementation plan, including the four phases, was based on a workbook for guideline implementation in youth help and protection services (20).

Phase 1: Creating Awareness Among Parents and Professionals (April–October 2017)

During the first months of the project, we spread information about the project via various ways. In April we announced the project in the general newsletter of the UCP. During the next months, the project leader (MP) attended the regular team meetings of all participating wards, to discuss with colleagues how the family-focused approach could be embedded in their ward. In addition, these meetings served to spread flyers with practical information about the project and to present a flow chart of general procedures regarding children of patients and child safety. At the launch of the actual implementation (September 2017), more information on the project was given in a presentation for all professionals and researchers in the UCP. We stressed that the primary aim of the project was to pay more attention to patients' normal role as a parent, for example by asking how parents manage to perform this role now they are being treated for a mental illness. Existing child safety procedures were mentioned, but were not the main focus of the project.

Finally, we created awareness for the importance of implementing a preventive, family-focused approach. For both parents and professionals, we developed an infographic film of 3 min illustrating the impact on the family when one of the parents has a mental illness. This infographic was placed on the project website https://kopp.umcg.nl that we had developed in this first phase of the implementation. The launch of the project (including the infographic) was also spread as a local news item by the press agency of the UMCG, among others via Facebook. On the project website, more information was provided regarding the monthly group-psychoeducation on parenting and a mental illness. Recently, the infographic was translated to an English version which can be found via the following link https://www.youtube.com/watch?v=mZbsXjh2hYM.

Phase 2: Adoption of the Intervention (September–December 2017)

This phase predominantly focused on activities that promoted the adoption of the intervention, such as creating awareness and support of various groups, including the management.

First, via a survey among professionals of the department mood and anxiety disorders, we inventoried general practices regarding the attention for patients' role as a parent in routine treatment protocols. This raised awareness among participants and gave us an impression of the current state of preventive, family-focused care at the department. Second, in order to gain broad support for the project, we organized participation meetings with the client counsel, social workers, residents, the secretaries, and managers of the department. About once in 3 months, we wrote an update of the implementation process to publish in the UCP newsletter.

Phase 3: Implementation and Evaluation (September 2017–August 2018)

From September 2017 on it was possible to sign up for the group-psychoeducation sessions. Both parents themselves and practitioners were able to sign up, online via the project website, via an email or via a registration form on paper. The first psychoeducation session took place on the first Thursday in October 2017. After the start of the psychoeducation sessions, we constantly adjusted the original implementation plan to the actual situation in clinical practice, using an iterative approach. Feedback of parents and practitioners was essential during this phase. For example, after introducing the pilot project, the importance of the topic "parenting and a mental illness" was endorsed by many professionals. However, some of them indicated that they had not enough time to discuss parenting in the treatment sessions within the scheduled time. To solve this problem, we suggested that they could use the infographic film of 3 min to introduce the topic and refer parents to the group-psychoeducation. Another example of a change in the initial plan was prompted by the observed difference in working methods at the various wards. By attending team meetings of the wards, we explored together with the professionals how the family-focused approach could best fit the current working methods in their opinion. Furthermore, parents were asked to fill out a feedback form after attending a meeting (**Supplement 1**) and some of them were asked for a telephone interview. This information was used to help to define barriers and remedies (**Table 1**).

Parallel to the implementation of the family-focused approach, the experiences of nurses with talking about parenting were investigated by students of the School of Nursing (University of Applied sciences). They performed two qualitative research projects, in which nurses were interviewed both individually and in focus groups (which included other professionals). For the interviews, topic lists on the theme parenting were used. With respect to the aim of this study, we do not extensively report on the nursing projects in this article. However, these projects may have contributed to increased awareness of parenting and continued positive reinforcement. In general, nurses were highly motivated to participate in the interviews and brought up some interesting topics, such as task ambivalence. On the one hand they felt family-focused care as one of their core responsibilities, but on the other hand, many nurses found it unclear when and how specific tasks should be performed and by which professional.

Phase 4: Continuation (September–December 2018)

The main goals of this final phase of the implementation were (a) to create support and guidance for the intervention in the organization and management structures, (b) to realize professional guidance on the work floor and/or training, and (c) to make arrangements with management and stakeholders for continuation of the project. To date, we have taken the following steps. First, we have developed the parental questionnaire "parenting and a mental illness," involving questions on experiences of patients' role as a parent and wellbeing of the children (**Supplement 2**). The reason for developing this questionnaire was to ease conversations about parenting between parents and practitioners or nurses. It is now available for use as part of the routine outcome measurements (ROM) of the department. Patients who have started a treatment automatically receive an email with an invitation to fill out the parental questionnaire (online). When the questionnaire is completed, the practitioner of the patient can see the answers in the electronic patient file. In this way, we expect that practitioners will have a better view of the family situation, e.g., how patients perceive their parenting role, which may ease family-focused conversations with the patient. Evaluation of the use and effectiveness of the questionnaire will take place at a later stage. Second, to meet the wish of professionals who indicated they needed more practical support, we set up a targeted training. However, eventually, we decided not to schedule the training since it was not considered realistic in a year in which two new electronic patient registration systems were introduced.

Therefore, training programs have been designated as a future priority. Lastly, we asked the management and stakeholders to express the intention to continue the family-focused approach as part of routine treatment protocols. This has already resulted in initiatives such as including information on mental illness and parenthood in the training program of psychiatry residents.

Analysis

The implementation was evaluated using both quantitative and qualitative data. The quantitative part consisted of a survey among professionals of the department of mood and anxiety disorders and registration of the number of participants at the monthly group-psychoeducation meetings. The online survey (**Supplement 3**) was created using the program Survey Monkey and was accessible for the professionals via an email that was sent by the secretaries. We asked ten multiple choice questions which were determined through discussions in the project group (MP, LD, PH, CH, RS). MP analyzed the outcomes of the survey, which are documented in a word file (anonymous). Registration of the actual number of participants was done by social worker HH, who was present at all but one psychoeducation meetings, and further processed in an SPSS file by MP. All data were checked and verified by at least two researchers (mostly by MP and LD).

The qualitative part consisted of written evaluation forms of the psychoeducation meetings (filled out by parents) and semi-structured telephone interviews with some of the parents who participated in a psychoeducation meeting. Questions asked were for example: "What was the most positive of the evening and what are you least satisfied with?" and "Do you have any suggestions for us to improve this psycho-education?"

Lastly, we reflected on the barriers that we had listed, whether and how we addressed them and which remedies were found (**Table 1**).

RESULTS

Survey

In September 2017, at the launch of the implementation, we performed a survey among professionals of the department of mood and anxiety disorders. All professionals received an email with a link to the online survey which consisted of ten multiple choice questions about the general way of practice regarding family-focused care. Thirty-two professionals filled out the survey in a time span of 6 weeks. Most of them were nurses (13/32), followed by psychologists (7/32) and other professionals, such as creative therapists (5/21). Furthermore, three psychiatrists and four residents in psychiatry filled out the survey. Respondents were about equally spread among the different wards of the department of mood and anxiety disorders; professionals from the inpatient wards (12), the day-clinic wards (11), and the outpatient wards (13) were represented. Some professionals worked at more than one ward. Nineteen of the 32 respondents (59.4%) answered that they mostly or always mention parenthood when they talk to patients who have children in the age of 0–24 years. The other 13 respondents answered they do not mention the topic regularly. Regarding the timing for bringing up the topic, most respondents thought the beginning of treatment (until the third meeting with the parent) is most suitable. Although most professionals said to talk about parenting and/or child wellbeing, they indicate a need for further education or skills training in their current work. Eleven professionals want to have more general information on mental illness and parenthood; for example on the actual risk that children will develop problems if their parents have a mental illness. Furthermore, 19 professionals indicate that they miss practical information, such as to which organization they can refer parents and/or children if they signal any problems. Finally, skills training in conducting conversations about parenting and child wellbeing is wanted, as indicated by 11 professionals.

Psychoeducation "Parenting and a Mental Illness"

In **Table 2** we show the number of parents that participated in the psychoeducation meetings. The number of participants varied per meeting. In the meeting of June, the highest number of parents participated, i.e., 12 parents (**Table 2**). In July only one person applied, probably that was due to the summer holidays. Most patients came with their partner or, in exception, with another family member. Twelve patients came alone. After December 2017, when we had launched the online possibility for registration, the majority of the participants applied via this way. In total 64 parents participated in the psychoeducation meetings, which was lower than the estimated number of 68 at the start of the study. Regarding the age of the children that were reached via psychoeducation, we are only able to provide a rough estimate in age groups. Most parents that applied for a psychoeducation meeting had one or more children in the age ranges of 4–11 (at least 18 children) and 12–17 (at least 14 children). Fewer parents had one or more children in the age ranges of 0–3 (at least 9 children) and 18+ (at least 4 children). For 6 participants data were missing regarding the age of the children.

At the end of the psychoeducation meetings, we asked participants to fill out an evaluation form in which we asked whether parents regarded the meeting as useful and which needs they had regarding parenting and wellbeing of the children. Twenty-five participants filled out the evaluation form. All regarded the meeting as useful. Most of them further commented on this question, as shown in **Table 3**. Of the 25 participants, 12 regarded the information of the meeting as sufficient, 2 expressed the need for further information, 3 preferred to talk about parenting more frequently with a nurse of the ward, 6 would like to go to a social worker or therapist, and 2 had other preferences for further support. One patient who preferred to talk about parenting with a nurse said: "It keeps me bothering whether the situation at home is good, whether it is safe enough, comfortable, and also a bit "normal." A parent expressing other preferences wished for: "The possibility for our children to talk with someone."

In addition, we interviewed four participants by telephone after they had attended a group-psychoeducation session. The participants who were interviewed concerned one woman who had been treated for depression and her husband and two

TABLE 2 | Number of parents* participating in the psychoeducation meetings.

Date	Number of participants	Way of application			
	(Actual/ applied)	Via practitioner	Written form	Online form	e-mail or other
2017/10/05	8/10	2	2	NA	4
2017/11/17	4/4	.	.	NA	3
2017/12/17	4/6	0	0	4	2
2018/01/04	2/3#	0	0	3	0
2018/02/01	4/6	0	4	2	0
2018/03/01	8/8	.	.	5	1
2018/04/05	6/6	.	.	5	.
2018/05/03	4/4	0	0	4	0
2018/06/07	12/12	4	0	8	0
2018/07/05	1/1#	0	0	1	0
2018/08/02	9/9	4	1	4	0
2018/09/06	2/4	0	0	4	0
Total	**64/73**	**10**	**7**	**38**	**10**

**in most cases the other parent came along with the patient; in a few cases a grant parent or other family member was present. # individual appointment via social worker instead of the regular group psychoeducation.*

TABLE 3 | Reactions of parents who attended a psychoeducation meeting.

Question: what did you find useful of this meeting?
"It was nice to share our experiences with other parents"
"It was a relief to know that many things are going well in the way we approach the children regarding my illness"
"It gave me insights in the possible impact of my illness on the children, and that early signaling is important to prevent that they will have problems"
"The importance to keep in touch with our children"
"Some things I recognized, other things are not applicable to our family"
"It was confronting (we recognized the impact of my illness on our son), and we now have more knowledge than before this meeting"
"Now we know that we need to talk about it with the children"

women who were partners of patients in care. Beforehand they all consented to be interviewed about their experiences. All four were positive about the organization and contents of the evening. Exchange of experiences was mostly appreciated, although sometimes confrontational, for example when it appeared that other participants had received more familial support than the respondent. The timing (to offer this intervention during hospitalization) was seen as very important. Recommendations on which words to use to explain about the mental illness to their children were appreciated. One of the respondents suggested that this meeting should be mandatory for all patients with children.

Barriers and Remedies

Table 1 shows how the implementation process changed during the course of the project. The initial implementation plan was based on the barriers for parents to talk about their parenting we knew from the literature. With the help of the logbook, it was recorded which parts of the plan were feasible and acceptable and which were not (found barriers). Solutions were sought and found in consultation with those involved. Successful adjustments to the implementation procedure (remedies) were, for instance, the introduction of the online infographic, the discussions in the team meetings, involving the management, and the website with possibility for parents to sign in for a psycho-education session themselves. At the end of the project an overview could be made of the recommendations we deemed necessary for a successful continuation of the implementation, such as improvement of ICT facilities and the scheduling of the training that was postponed.

DISCUSSION

In this pilot study, we have described the development of a program to implement a preventive family-focused approach into routine care for adult patients who are parents and treated for moderate to severe depressive and/or anxiety disorders. The focus was on feasibility and acceptability. Most professionals expressed their sympathy to the project. Our survey showed that professionals consider it as important to inform patients who are parents about the possible impact of mental illness on parenting (nearly 60% of professionals already talked about parenthood and child wellbeing with their patients at the start of the implementation). The other 40% did not mention the topic frequently, and the need for additional training (education and/or skills) was indicated by a majority of professionals.

Team discussions revealed that opinions differed between disciplines whether talking about parenthood was part of their job. At wards where a social worker was part of the team, patients were preferably referred to the social worker for further consultation. However, the option of referral to a social worker is not present at all wards of the UCP. It gradually became clear that the family-focused approach fitted very well with the core business of nurses. First of all, the survey was filled out by nurses for 41% (13/32). This fits with literature on this topic, which shows that nurses are in a unique position to initiate conversations on parenthood and child wellbeing because of their direct, frequent, and sustained contact with patients and families (18). Nevertheless, family-focused care should not be reserved for specialized professionals. Small talk about normal life (events), including parenting, can, for example, be initiated by a secretary who can also hand over a leaflet with information about targeted psychoeducation. Furthermore, therapists of all kinds can stress that mental illness may have impact on parenting and that they, therefore, want to pay attention to it during therapy sessions. Management support and professional education in a positive organizational culture (13) are conditions to realize a therapeutic climate where support of patients who are parents becomes a normal part of treatment. This can benefit both parents and children (21).

As Foster and Isobel (14) have pointed out, talking about parenting involves special capacities. In our survey, 11 out of 32 professionals expressed their interest in training opportunities to address parenting and child wellbeing in conversations with patients. Other respondents mentioned (and perhaps rightly so) that they had enough conversation techniques to be capable to discuss parenthood. Interviews and case discussions during the team meetings provided more insight into the emerging dilemmas of professionals, such as the idea of having to choose between the good of the parent and that of the child (22). Reupert et al. (13) warn that professionals, including social workers and nurses, seem to avoid such dilemmas by arguing that talking about parenthood can harm the confidentiality of the relation between professional and patient. They also hold a plea to be creative in finding a solution when there is the conviction that these conversations are necessary.

The monthly organized psychoeducation meetings for parents were visited by 64 parents in 1 year. By far most of them (38) applied via the online registration form on the project website. Parents' reactions after the psychoeducational meetings were mainly positive, and they provided useful feedback to further improve the meetings. A good timing of the psychoeducation, for instance, was deemed indispensable. Parents' reactions were discussed in the team meetings by the project leader and once more colleagues were asked to draw their patients' attention (on time) to the educational evenings. This illustrates that the implementation of a preventive approach is not achieved after one or two actions, but requires continuous positive reinforcement (23).

The number of parents participating in the monthly psychoeducation meetings was variable. More professionals started to refer parents, but the most visible increase in registrations was in the number of parents who applied directly via the website. This may indicate that the psychoeducation meets the needs of, at least a subset of parents treated in the UCP, which also corresponds with the positive feedback of parents who had joined a psychoeducation meeting: all regarded the meeting as useful, and about half of the parents would like further support. Furthermore, feedback and interviews indicated that awareness among parents about the impact of the mental illness on child wellbeing has grown. We think that for e.g., better provision of information about the preventive offer may have facilitated parents to be more aware and to participate in the psychoeducation. Furthermore, the easy way of registration may also explain the higher number of online applications, or the fact that the involvement of a (busy) professional can be avoided.

A limitation of our research is that it was carried out in a specific setting under specific circumstances. It is unlikely that the same circumstances are present in any other organization for adult care, which could result in different outcomes. On the other hand, a strength of this pilot is that we were able to demonstrate how an iterative implementation method can be used to realize a translation from current knowledge to clinical practice. Further prospective studies with a control group are needed to assess whether preventive psychoeducation, as part of a family-focused approach, is effective. Findings of this pilot study provide useful information for setting up larger studies with a higher impact. Our aim is to continue the psychoeducation program in the UCP and to further develop it, using the outcomes of this pilot study. Regarding policy, we strive to offer more parents the possibility to attend the psychoeducation meeting on parenting and a mental illness at the UCP, for example by sharing our knowledge with other health care providers and governmental organizations.

In conclusion, our findings indicate that our preventive family-focused intervention met the needs of parents that participated in the psychoeducation meetings. Feedback of participants showed that they regarded the evening as useful, and more importantly, they seemed to be more aware of the potential impact of their illness on their children. However, a large number of parents were still not reached by our intervention and further study with follow-up activities are needed to measure the effects of psychoeducation on, for example, parenting, the quality of the parent-child interaction and child wellbeing. Lastly, we regard the structural embedment of interventions and continued positive reinforcement as essential elements for long-lasting attention for the prevention of mental health problems among children of parents that are treated for mental health disorders.

AUTHOR CONTRIBUTIONS

MP, LD, and PH set up the project, conducted the pilot study, and wrote the manuscript. CH supervised methodology, RS supervised it all.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2019.00004/full#supplementary-material

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Identity in Personal Recovery for Mothers With a Mental Illness

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Developing a "positive identity" is considered a core component of personal recovery, and mothering offers meaning in life and a valued identity. Few studies have highlighted the factors influencing identity within a personal recovery paradigm for mothers with mental illness. This study explores how mothers describe their identity in relation to recovery, including the factors that influence identity. Using constructivist grounded theory methodology, in-depth interviews were conducted with 17 women who were mothers and experienced mental illness. Women defined their self-concept broadly, accentuating motherhood, but also including vocational, community and social roles. Analysis revealed six categories: defining self, becoming a mother, being a "good" mum, feeling different, doing it my way and speaking out. Valuing identity in parenting was found to be linked to recovery. Services may facilitate personal recovery by supporting mothers to enhance a self-concept associated with mothering, as well as other diverse attributes and roles.

Keywords: gender, identity, mental illness, motherhood, personal recovery

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INTRODUCTION

Personal recovery is a unique journey for those experiencing a mental illness, however common underlying characteristics and processes are now becoming better understood (1). Distinct from clinical recovery, which emphasizes a remission of psychiatric symptoms, personal recovery is grounded in the subjectivity of people who have live experience of mental illness (2). Personal recovery is concerned with holistic life functioning and social participation and agency, regardless of the presence of ongoing symptoms (3). Recovery experiences are unique and individual, and also differ for men and women due to the influence of gender (4).

Identity, and particularly the development of a positive sense of self that is less illness dominated and more strength based, is one of the key processes of the CHIME personal recovery framework developed by Leamy et al. (1). Resulting from a systematic review of personal recovery literature, the CHIME framework highlights Connectedness, Hope, Identity, Meaning and Empowerment as core categories that transform in a recovery journey. The significance of redefining and reclaiming a valued identity may comprise "determining the direction of one's life, grieving for lost opportunities, and yearning for belonging and acceptance" (5).

The ways in which mothers with mental illness describe their identity and the influences upon it within the process of personal recovery, has remained peripheral within recovery discourse. The aim of this study is to explore the ways in which mothers with mental illness describe their identity, and the factors that support or hinder development of a positive identity. Critical research aims to reveal and question social inequalities that are embedded within stereotypes (6). This current study adopts a gendered lens to examine the identity experiences of mothers with mental illness, and to investigate multiple identity possibilities including and beyond motherhood.

Identity Theory

Identity is broadly defined as one's self concept and is constructed through self-awareness of what and who one is "like" and "not like" (7). Identity is constructed within one's social and cultural context (8) and is therefore related to the construct of connectedness. Identity is a highly contested concept within feminist theory. An overarching theme is that gender based identity is predominantly oppressive. Butler (9) has problematized the concept of gender identity as the reproduction of the subordination of women, and explored the ways in which it is reproduced through linguistics. She also questions whether identity is a stable and continuous entity, and if it is static, the social regulatory structures that produce this outcome:

> 'To what extent is 'identity' a normative ideal rather than a descriptive feature of experience? And how do the regulatory practices that govern gender also govern culturally intelligible notions of identity? In other words, the 'coherence' and 'continuity' of 'the person' are not logical or analytic features of personhood, but rather socially instituted and maintained norms of intelligibility' (p.23).

Côté, defines identity as multifaceted and states that "manifestations of identity exist at three levels of analysis; the subjectivity of the individual, behavior patterns specific to the person and the individual's membership in societal groups" [(10), p. 8]. Within psychology, these differentiations have more broadly been separated into two categories: personal and social identity.

Personal identity relates to the internal characteristics including attributes and values that one recognizes as inherent to one's self concept. Social identity meanwhile, reflects how individuals view themselves as affiliating with and belonging to particular societal groups. Vocational, family, community, political, or gender-based categorizations are realms in which social identity may exist.

A fundamental debate in identity theory is whether the self is essentially a stable and enduring entity, or whether it is constantly changing and evolving (7). According to self-schema theory, the core identity constructs of an individual are thought to remain stable over time, although there may be some change in the more peripheral aspects of one's identity (11). Rosenfield (12) used schemas relating to self-salience, to explain disparities in mental health outcomes across gender, race and class, where a social determinants framework was ineffective in accounting for results that were inconsistent with structural inequality. Self-salience is associated with the relative importance individuals place on self, vs. the collective (13, 14). Rosenfield (12) cites the gendered socialization processes that contribute to the social construction of femininity and masculinity, and recognizes how these differ in relation to race and culture (15). Historical and enduring social conditioning underlies the gendered internal self-salience tendencies that predispose white women to experiencing more internalizing problems (e.g., depression) and white men to have a greater prevalence of externalizing problems (e.g., aggression) (12).

In contrast, self-categorization theory is based on the assumption that individuals' identity can and does change and evolve in response to the social and environmental context in which people live, and in response to external and internal processes associated with major life events. Ontorato and Turner (7) compared the two theories and found evidence to support self-categorization theory, with two studies demonstrating "the dynamic nature of self" (p. 276) that is context dependent. Self-categorization is a useful theory to inform exploration of the influences on identity development occurring through recovery from mental illness. Underlying assumptions imply that recovery emerges through social and psychological processes of personal change and development (1). The depth and breadth of self-reflection and change embedded in many of the subjective narratives of those with lived experience in mental illness and recovery, suggest that recovery entails a transformation at a deep psychological level, not merely some pruning or enhancement on the margins. Thus, self-categorization theory relates to this study.

Although gender identity can be a source of solidarity amongst women, it is more frequently problematized within feminist debate as a site for the reproduction of existing power differentials and maintenance of patriarchy. Within feminist theory:

> "…the reproduction of normative identities cannot be understood simply as a question of positioning within language but as a lived social relation that necessarily involves the negotiation of conflict and tension" [(16), p. 185].

For women who are mothers with mental illness, those conflicts and tensions may be the precipitant as well as the result of the psychiatric diagnosis and subsequent treatment and recovery. One method of developing deeper understanding of lived social relations is through amplification of the voices of those women who are marginalized in dominant discourse. This study aims to contribute to that endeavor.

Mental Illness and Identity

Mental illness has been characterized as a loss of self (5), as psychiatric symptoms may conceal or distort an individual's skills, knowledge, values and attributes. Social identity can also be disrupted as the illness often manifests in ways that prevent people from continuing in social roles that they had previously occupied and enjoyed (17). Individuals have spoken of enduring confusion, grief and regret as they seek to reconstruct a sense of self and reconcile the differences in their identities before, during and after the mental illness experience (18).

Researchers have explored the relationship between psychiatric symptoms and disruptions or incongruence in self-concept (19, 20) including models of conceptualizing and mitigating the impact of complex trauma on ones' identity and healing (21). Wisdom et al. (5) found loss of self to be the most prominent focus of narratives, with the illness "often described as taking away…their previously held identity" (p. 491).

Feminist writers have observed that historically, society's response to mental illness in women has resulted in sanctions for deviance and non-conformity to the prevailing cultural expectations [e.g., (23)]. A focus on social "integration" and adaptation of the individual, rather than advocacy for social change to create more equal and accessible social environments that foster diversity and inclusion, are evident in critiques of the recovery paradigm (4, 22).

Mothering With a Mental Illness

Mothering with mental illness is increasingly common (24, 25). Studies conducted with mothers with mental illness have emphasized the importance of a mothering identity to women in providing meaning and purpose (26), love and connection (26), and fulfillment (27).

Researchers have highlighted that parenting stress can compromise mental health (28–30), and discovered that mothering confidence and competence can be undermined by the scrutiny and prejudice imposed by over-zealous service providers (26, 31) and family members (32), who are often operating within a risk aversion framework.

Shor and Moreh-Kremer (33) emphasized the strengths for women with mental illness in being able to claim a normative maternal identity, thereby reducing their vulnerability to stigma and alienation. Within that study mothering identity was compared to mental illness identity, without consideration of other potential sources of identity in women's lives. While there may be individual benefits to conforming to gendered norms, if this is through developing a public persona that is incongruent with one's internal value system, and acceptance of oppressed social status, there will also be psychological costs (23).

AIM

The aim of this study was to explore how a cohort of Australian women who were mothers with mental illness described their identity, and how this related to personal recovery from mental illness. The research furthermore aimed to explore the factors and processes that mothers with mental illness describe as influencing their identity.

METHOD

Constructivist grounded theory (CGT) methods were employed to gather and analyse rich descriptive qualitative data (34). Grounded in feminist understandings of gender inequality that can result in and exacerbate the effects of mental illness, the research aimed to amplify the voices of women from a marginalized population group (35). Congruent with feminist approaches to research, CGT challenges the objectivity claims of positivist methods, instead advocating for transparency in acknowledgment of the values, perspectives, experiences and biases of the researcher which all influence the research decisions, processes, and outcomes (34). CGT emerged in the 1990's [e.g., (36, 37)] in response to the epistemological assumption that if "social reality is multiple, processual, and constructed, then we must take the researcher's position, privilege, perspective and interactions into account as an inherent part of the research reality" [(34), p. 13]. While traditional grounded theory methods emphasize that researchers arrive at their own data fresh, without prejudice or influence from previous studies (38) this sequencing is not required for undertaking constructivist grounded theory as the background research and theoretical context investigated, contributes to the unique subjectivity of the researcher. As all experiences in the social world influence the perspectives, language, assumptions and biases a researcher brings to each project (34), it would be exceedingly ineffectual to refrain from reviewing literature as just one of infinite potential influences. Hence in this study, literature on identity and gender were explored prior to the data collection with participants, with a more focused search ensuing after the categories were formulated.

Procedures

Following ethics approval from two health service and one university Human Research and Ethics Committees, mental health clinicians from a regional clinical mental health service supported recruitment processes by promoting the study amongst eligible women on their caseloads. Steps were taken to ensure no harm or distress was caused, and that women felt comfortable to end or pause the interview at any time. All interviews were conducted by a researcher who was also an experienced social worker employed as a senior mental health clinician and informed consent was obtained in writing. Local services guides were developed and distributed to participants in the event that the interviews triggered psychological response requiring follow up.

Nine women were recruited via clinical mental health services, and the remaining 8 from the general community via social and print media (promoted on Twitter and in local newspaper articles). Interviews were conducted by one researcher (RH), and consistent with CTG procedures, four of these 17 women were interviewed on more than one occasion as part of theoretical sampling. These participants were invited to participate in a second interview due to specific characteristics they possessed, that meant they were able to provide data that could increase understanding of phenomena relating to emerging codes. A total of 21 interviews were conducted, the mean duration being 42 min. The participants were at different stages of recovery. None of the women were in an acute crisis or experiencing active symptoms at the time of the data collection, although one woman had been hospitalized within the last month, and 6 were engaged with a clinical mental health service at the time of the interview. Three women were supported by a community mental health service and the remaining eight reported not receiving any specific mental health service at that time.

Interviews were conducted in person within a confidential space at various community health venues that were accessible to participants. Interviews were audio recorded and transcribed verbatim. The interview schedule was open ended and flexible, containing questions pertaining to women's experiences of personal and social identity. The interview schedule is available as a **Supplementary Material**. To set the context, women were asked about their mental illness and recovery journeys. Women were then asked broad questions regarding how they describe themselves, their social roles, personal characteristics, if and how the way they see themselves has changed over time, about their mothering and parenting, how they envisage others see them, and what or who influences how they view themselves in various settings (e.g., work, mothering/family, community). The total recruitment and interview time frame was from July 2015 to February 2016.

Data Analysis

Data analysis occurred concurrently with data collection, coding and reflection beginning immediately after transcription of the first interview and continuing after each interview. Analysis consisted of initial coding following close reading of all data on multiple occasions. Memo writing, purposive sampling, focused coding, mind-mapping were subsequently undertaken to enable development of categories (34), hence the categories emerged directly from the data. Regular dynamic discussion amongst the three member research team stimulated analysis and questions relating to the emerging categories. This led to second interviews with some participants to provide additional details to define the properties of emerging categories, as consistent with purposive sampling.

Participants

Participant characteristics are summarized in **Table 1**. A total of 21 interviews were conducted with 17 women who were all mothers with a psychiatric diagnosis. The women were a heterogeneous group and varied in relation to their living arrangements, socio-economic status and cultural background. Ages ranged from 23 to 53 year, with an average age of 36.29.

RESULTS

The study resulted in identification of six categories surrounding the concept of identity. The categories name psychological or social processes that were predominant within the data collectively. "Defining self" explores how women describe their self-concept and highlights the important elements of personal and social identity. As mothering was a core component of participants' identity, the categories "becoming a Mother" and "being a 'good Mum'" illuminate the relationship between women's mothering role, their mental illness and their recovery journey. The relationship between an illness identity and a mothering identity is explored in these sections. The category "feeling different" focuses on the women's experience of lacking a social identity and the implications of this on their personal identity. The final two categories reflect women's recovery progress, and highlight the importance of developing a positive identity, for attaining other personal recovery outcomes such as empowerment and meaning in life.

Defining Self

In describing their sense of self, all of the participants sought to contextualize their current temporal personal identity within their life histories, and inter-relational experiences, beginning in childhood. Ten women shared accounts of trauma from interpersonal violence occurring within childhood and/or adult relationships, and it is not known whether the remaining women may have also experienced violence but not disclosed. Through articulating key formative events, the women sought to construct identity narratives that provided meaning to explain their current circumstances, including their mental illness diagnosis.

Personal and social identities were described by participants as changing and evolving, while also containing stable components that persisted over time as illustrated by the assertion "I've always been this way" (P7). Participants described their identity in relation to their personal attributes, social roles and key relationships. Not all women were able to eloquently describe themselves, however there were exceptions:

> "(I'm) a woman, a mother you know, I'm a feminist, I'm an atheist, I'm left wing, very left wing in a lot of ways. I'm very politically minded, I'm very also socially conscious I guess. I do a lot of social activism. I'm bisexual so I've done a lot of campaigning for marriage equality and stuff like that. I'm very creative… I do a lot of crafty stuff. I love having kids because it gives you an excuse to colour in… I do a lot of puzzles, I watch probably way too much television and movies, I'm a huge film buff. I love politics as well" (P11).

Moral characteristics such as honesty, independence, generosity and creativity were viewed positively in the ways in which women viewed themselves, and they validated this through reflecting on how others might see them "I think people would describe me as nice and caring" (P13). Recognition of their own resilience was also evident ($n = 9$) in comments such as "…sometimes I look back on what I've coped with and I think, wow" (P5).

For other women, there was recognition that attributes that had been characterized as strengths prior to the mental illness experience could also become barriers to the help seeking that may be required to address mental health challenges:

> "I would describe myself as very independent. Probably too independent… I like individual sports like running and tennis. I've never really played team sports. I like to achieve, like at work I became fairly obsessed with achieving at work. But that was another thing after having the baby. It was very different…" (P2).

The social roles that the women identified embodying were relational, vocational, and community orientated. They included mother, sister, daughter, partner, friend, worker, health professional, student, mental illness advocate or educator, committee member and volunteer. Relationships with others in these spheres contributed to how they viewed themselves, and their personal identity could be particularly susceptible to messages they received from significant others regarding their performance within those roles.

Four women referred to their religious affiliation as being important in defining their moral and ethical framework. Although three women spoke of attending church related activities, they did not view themselves as sharing many characteristics with other members of their church except for their religious beliefs, therefore this was salient for personal rather than social identity.

Cultural identity was discussed by five women, two of whom were of Aboriginal descent (P7 and P8). Cultural disconnection was apparent with one of the women who indicated she knew

TABLE 1 | Participant characteristics.

Age; cultural background	No. of children and age	Housing type and lives with	Self-reported diagnosis and did diagnosis occur prior to motherhood?	Income source	Current mental health service engagement
38; Irish and French	3:10 years, 8 years, 6 years	Private rental Lives alone	Bipolar, OCD, BPD Yes	Government payment	None
35; Australian	2:2 years, 10 months, pregnant at time of interview	Own home on farm Lives with husband and children	Anxiety and depression No	Farm income	None
44; Australian	2:13 years, 11 years	Own home Lives with husband and children	Anxiety and depression No	Husband's full time wage	None
39; English and Australian	1:15 years	Own home Lives with partner and child	PND, anxiety and depression No	Own full time wages	None
39; Finnish and Italian, adoptive family Australian	1:14 years	Private rental Lives with child	BPD and bipolar disorder. No	Government payment and part time wages	Community MHS
40; Australian	1:2 years	Public housing Lives with child	Drug induced psychosis Yes	Government payment	Clinical MHS
29, Aboriginal	8:15 11 10 8 4 3 plus 2 younger in foster care	Public housing Lives with eldest 4 children	Anxiety and depression, paranoia, bi-polar disorder No	Government payment	Clinical MHS
31; Aboriginal	6:14 years, 12 years, 9 years, 4 years 2 years, 8 months	Transitional housing Lives with youngest 3 children	Anxiety, PTSD No	Government payment	Clinical MHS
43; Australian	1:15 years	Own home Lives with husband and child	Bipolar Disorder Yes	Government payment	Community MHS
26; Australian	1:6 months	Private rental Lives with partner and child	Bipolar Disorder Yes	Partner's full time wage	Clinical MHS
34; Australian	3:9 years, 8 years, 5 years	Private rental Lives with children	PTSD and depression, PND No	Government payment	None
53; Australian	3:17 years, 16 years, 13 years	House on the family owned farm Lives with husband and children	PND and depression No	Farm income, and wages from 3 part time jobs	None
28; Australian	2:5 years, 2 years	House on the family owned farm Lives with husband, children and boarder	Depression Yes	Part time wages and farm income	None
44; Australian and English	1:9 years	Private rental Lives with mother and child	Depression and anxiety No	Government payment	Community MHS
23; Australian	1:10 months	Own home Lives with partner, step child and son	Depression and anxiety Yes	Full time wages and partners' wages	Clinical MHS
45; Australian and German	2:14 years, 12 years	Own home Lives with husband and children	Anxiety, OCD and depression Yes	Part time wages and husband's full time wage	None
26; Caucasian	1:14 months	Parents' home Living with parents and child	Depression, Anxiety and PND No	Husband's full time wage	Clinical MHS

BPD, Borderline personality disorder; MHS, Mental Health Service; OCD, Obsessive compulsive disorder; PND, Postnatal depression; PTSD, Post traumatic stress disorder.

little of her heritage as her Aboriginal father had died when she was young. Another Aboriginal woman had experienced significant trauma during childhood and in adult life and had lacked opportunities to develop cultural knowledge or connection, stating "I want to know my culture, but I don't want to *do* my culture" (P8).

Participants articulating devalued personal identities were more likely to be in roles or relationships where they derived little pleasure or fulfillment:

> "I have issues with my work and because my boss is bully… when they cut my hours I took it really personally and I couldn't go to work the next day–I just cried" (P16).

Some women had difficulty describing themselves, identifying their strengths and imagining themselves in the future: "at the moment I can't see past tomorrow… I'd really like to enjoy things in my life a little bit more" (P15).

Identity across different domains could manifest in either positive, socially valued ways, or as negative and socially devalued. For example, a mothering identity could be positive if one perceived oneself to be a "good" mother, and felt a sense of belonging to a mothers' group. Alternatively, a mothering identity could be experienced as negative if one considered herself to be deficient and incompetent, thereby judging herself as a "bad" mother. Similarly, if a woman held a position of esteem within the community as a committee member in a sporting club, this would foster positive personal and social identity across other domains, whereas a lack of community recognition or a sense of social distance could contribute to a devalued identity reinforcing isolation and social exclusion.

Becoming a Mother

> "When I gave birth, I felt connected to something bigger and stronger than myself. I'm not religious, but I'm spiritual. So I felt connected to the bigger scheme of things like connected to other women and the feminine force of the universe" (P11).

Becoming a mother was a particularly significant component of women's self-concept. It could transform a woman's personal and social identity, fostering a deep sense of connection and meaning. However, it could also negatively impact a woman's view of herself if she struggled to feel competent in the role. Diversity was apparent in the responses the women had to embodying a mothering role and identity. For some it was an identity they immediately embraced and had always expected. The majority of participants ($n = 12$) had always had aspirations of parenting, and one woman with her partner had been planning the pregnancy for some time, including changing psychiatric medication and exploring fertility options. Having a baby brought a sense of empowerment and fulfillment for 8 of the participants. For one woman it was an opportunity to exercise her own autonomy and make choices that would not necessarily be endorsed by her own mother;

> "I breastfed until 21 months and I loved it. And my mum said 'don't you think it's time you gave it up?' at six months. And I'm like, hey, I felt something was right" (P5).

However, the experience did not always live up to expectations, especially in the early days and weeks.

> "I hid it from the maternal and child health nurse that I wasn't coping and then on the Friday I'd had enough and so I rang (family services worker) and said 'I need your help'" (P10).

For four women, all of whom had relished the idea of motherhood since childhood, childbirth was accompanied by debilitating postpartum depression when they had thought they were emotionally prepared. This left them with guilt and regret for the aspects of early parenting that they missed.

Experiencing breastfeeding difficulties was a source of significant stress and eroded self-confidence for four women. One woman delayed disclosing her decision to bottle feed to her new parent's group, fearing social rejection:

> "It took me probably two weeks to tell them that I wasn't breastfeeding, because I was just so anxious about it because everyone else was breastfeeding and I was like, oh they're going to want to kick me out of the group, yeah they won't want to talk to me ever again. But once I told them they were really supportive" (P17).

Having responsibility for a child prompted two women to re-assess their social behavior and temper their anger. They explained how accepting the responsibility associated with caring for an infant had led them to cease drug use, necessitating the severing of social relationships that would undermine this new healthy lifestyle choice. This assisted in them feeling greater competence in managing emotions, which led to a more positive identity.

Becoming a mother could have positive or negative implications for the women's personal identity. This was partially influenced by the availability of supportive relationships and assistance to adjust to the early parenting phase. How women perceived themselves to be performing in the mothering role profoundly shaped their identity, as did the quality of their relationships with family members and health professionals and the ways in which their inherent value was reflected through interactions within these relationships.

Another factor was whether the women's mental illnesses emerged before or after they became a mother. While 6 women had been given a psychiatric diagnosis prior to becoming a mother, an additional 7 participants expressed the belief that the mental health issues that eventually led to the subsequent diagnosis (after motherhood) had existed for many years, in some cases from childhood or adolescence. In these situations women described the mental illness label as providing an explanation for psychological, cognitive and behavioral events, along with the opportunity for enhanced social identity through developing peer relationships.

Being a "Good Mum"

A "Good Mum" was defined as accepting responsibility for one's children, prioritizing her children's needs over her own, being present and responsive and "making it fun" (P7); using humor and actively engaging in play. Attending to children's emotional needs and being available was a part of this: "I want to be the person that my kids will come and talk to me when something's going on, you know. Instead of hiding that away" (P7). All of the women interviewed expressed a desire to be identified by others as a "good mother." Furthermore, they wanted to embrace this label for themselves, although for three participants reconciling their thoughts and emotions regarding the mothering role with an ideal self-as-mother, was problematic. One woman rationalized that she wasn't a good mother because

> "…most of the time I feel like I'm just getting through… it's the extras that play with the mind and question how well you are doing. Am I stimulating them enough? Am I doing the right things for learning at this age?" (P2).

Characteristics of being a "good" mother ranged from meeting children's basic needs for nutrition, sleep and intellectual stimulation to being physically and emotionally available to children; "a good mother is showing love to their child, their little one, talking to them, validating their feelings… understanding his point of view" (P5). It involved persisting through hard days. Persisting entailed getting up in the mornings despite exhaustion or symptoms of depression, putting "a smile on a lot in front of them and for them" (P7) and for two mothers, remaining in undesirable employment to provide financial stability. In speaking about striving to be a good mother, two participants referred overtly to the sexism embedded in their own interpersonal relationships that saw them taking primary responsibility for child rearing while their husbands enjoyed more leisure time.

Eight women expressed sadness and regret regarding their own upbringing. Attributing their subsequent mental illness to the cumulative impact of trauma, abuse, neglect and disadvantage, participants felt that if they had been raised in a family with a responsive adult figure attentive to their needs, their lives may have taken a different path. This fuelled a desire to be present and considerate and sensitive to their children's traits, strengths and needs, even within challenging socio-economic circumstances.

> "Making sure I do a good job and (my child) gets a good education and just the little things, speaking to him nicely. I've never ever yelled at him, I don't believe in that and I don't like people who smack their children" (P6).

Despite best intentions, there were times when women's stress levels were high and their mental health was compromised. They became aware of how difficult this high expectation of parenting was in such times. For five women, serious physical health conditions such as pneumonia also took a toll. At this time children.

> "…learnt to become very independent and it was hard to parent them at that time. I didn't have the energy to discipline them and I noticed a lot of things went out the window—just their manners and the way they behaved and stuff—like I just couldn't be a good parent" (P16).

Breaking the pattern of cycles of intergenerational poverty, trauma and substance use were important to women in demonstrating their parenting attributes. They hoped for easier life circumstances for their children and hoped to guide them toward healthy choices:

> "It's being able to see beyond their pain and just walk with them and be their friend and guide them and show them this is what Mum and Dad have been through and this is why we don't want you to go down this path" (P1).

Viewing oneself as a good mother therefore had a positive impact on personal identity, while feeling incompetent or guilty had the opposite effect. Women used different measures to assess their parenting capabilities however an increased capacity for self-reflection and self-compassion was associated with greater progress toward recovery.

Feeling "Different"

A sense of disconnection and alienation from peers and family members pervaded development of a valued and intact self-concept for a number of women and they related experiences stemming from childhood to illustrate this. For some this was characterized by additional sensitivity, "I was a very clingy needy child" (P16) or having different needs and abilities to siblings "when we were younger I would clean (my sister's) room or do stuff for her just so I could spend time with her. Because we're very different" (P10). These examples suggest an unmet need for nurturing and connection in childhood. Resulting from this was a fragmented sense of self that was dominated by rejection.

For others it was about possessing a unique skill set or perspective on life, "the way my mind works I've never known anything different…and because of that I got treated different" (P7). Being able to deflect childhood labels of deviance and learning to embrace their own uniqueness fostered a sense of wellbeing, but was difficult to achieve outside the context of a supportive relationship. Women who felt validated by supportive intimate partners were better able to reflect of their childhood experiences of exclusion and externalize the cause of this experience.

Doing It My Way

This category was associated with increased confidence in one's experience and competence in mothering and signified a recovery milestone. Over time, women's self-awareness grew, and they became more insightful as to their own strengths, values and needs. This informed the resources and strategies they accessed to support their mental health and wellbeing. They gradually became proficient at seeking the support they required, whether that involved psychiatric medication, talking therapy, social connection, creating art, returning to study or if it was a viable economic option, taking respite from employment.

A key recovery milestone was reached when women grew in confidence and self-belief, enabling them to recognize and confront people in their lives that they saw as exerting disproportionate influence. In relation to parenting choices and styles, four women spoke of rejecting the preferred methods of others and asserting themselves. A sense of empowerment emerged when they were able to exert control over their baby's wellbeing. For one woman, ensuring that her own mother was not verbally or physically violent toward her in her infant son's presence was of paramount importance, and she asserted herself around her expectations through threatening to withdraw access to her son.

Another participant reflected on receiving what she defined as a "one size fits all" (P5) approach to parenting and attributes much of her mental anguish to the circumstances of her adoption. Being able to successfully breast feed her baby for 18 months was an empowering experience for this mother, and perhaps the most powerful example of her defying her adoptive mother with great success for the health and wellbeing of herself and her son.

Living on her in-law's rural property, another woman (P2) explained how she withdrew from her husband's parents as a strategy to maintain her independence and to reduce the feelings of inadequacy her mother-in-law instilled. In declining offers of child care from the children's grandmother, this woman sacrificed the potential for respite from her two young children, in the interests of sustaining her need for independence and autonomy.

Self-expression through choosing unusual clothing was how another participant asserted her own style. Creativity was employed to physical represent her mood, and she shared how her curious outfits at times draw smiles from community members, which then made her feel "that little bit happier." On one occasion she related dressing in a pink ball gown with a purple top hat to go to the supermarket:

> "I just got up feeling… I'm in a mood today, what can I wear? And I will spend hours because I need to find (the right outfit) and I won't wear something that won't match my personality, if I don't I think that matches my personality for today l won't wear it" (P7).

Speaking Out

The category of speaking out included disclosing, becoming a mental health advocate/educator, addressing stigma and challenging stereotypes. Although in speaking out, mental illness became dominant in women's personal identity, it was viewed positively and enabled social identity through peer networks to flourish. Enduring the ups and downs of mental illness was seen as a valuable asset that enabled women to take on an educator role, to connect with others through shared experience and to be knowledgeable in ways that others were not.

> "My mental health journey has meant that I've got experience in that to be able to connect with people on that level and that's what I want to be able to do" (P13).

Sometimes the desire to engage in community education stemmed from experiences of discrimination that woman believed arose from ignorance.

> "I was angry with the way society treated us. And just cast us aside like we were nothing… you can't necessarily see the pain that we carry with us. Its soldiers. And that's what I call it. We're all soldiers. We're all in this together…" (P1).

When women began speaking out, they embraced their illness as a core component of self. The illness symptoms, treatments, and ramifications were integrated into their lives and were a part of how they viewed themselves and presented themselves to others:

> "…doing the mental illness education was a really big part of my recovery so to go back and give out to the community, our point of view of how we feel, that's recovery too" (P1).

Participants expressed being selective in how and to whom they disclosed. Generally there was a correlation between the relative perceived safety of the participant's social environment and the extent of their disclosing. Women who felt supported and who had a multi-faceted, secure and positive social identity within their community were more likely to fully disclose. Past involvement with child protection was a deterrent that led women to conceal or minimize the impact of their mental illness.

DISCUSSION

The majority of women who participated in this study described multifaceted identities that represented their mothering and familial roles, their employment or vocational occupations, community relationships, hobbies and interests, religious and cultural status as well as the ways in which they defined their political or social values and attitudes. These descriptions encompass broader dimensions of identity than have previously been reported in studies with this cohort (39). Each of these could be considered positive and socially valued, or negative and socially devalued, with implications for social and personal identity, depending on the perceived competence and autonomy in the role. For instance, self-identifying as either a "good" or "bad" mother. Mothering was found to be a particularly powerful influence on women's self-concept, and participants identified the characteristics of being a "good Mum" as self-sacrifice, being present and enjoying the role. These concepts are congruent with the prevailing Western ideals of mothering that are grounded in traditional gendered roles that are oppressive to women (40). The findings emphasized the importance of identity to recovery, consistent with the CHIME framework (1).

Also significant, were women's expectations of the future. Impending opportunities and obstacles were related to women's ideas about their own competence, social status, agency and aspirations. The findings also highlight that identity is inseparable from social connection with participants described the ways in which their sense of self was heavily shaped by interactions with others (41). **Figure 1** represents conceptually the ways in which the women described the factors influencing

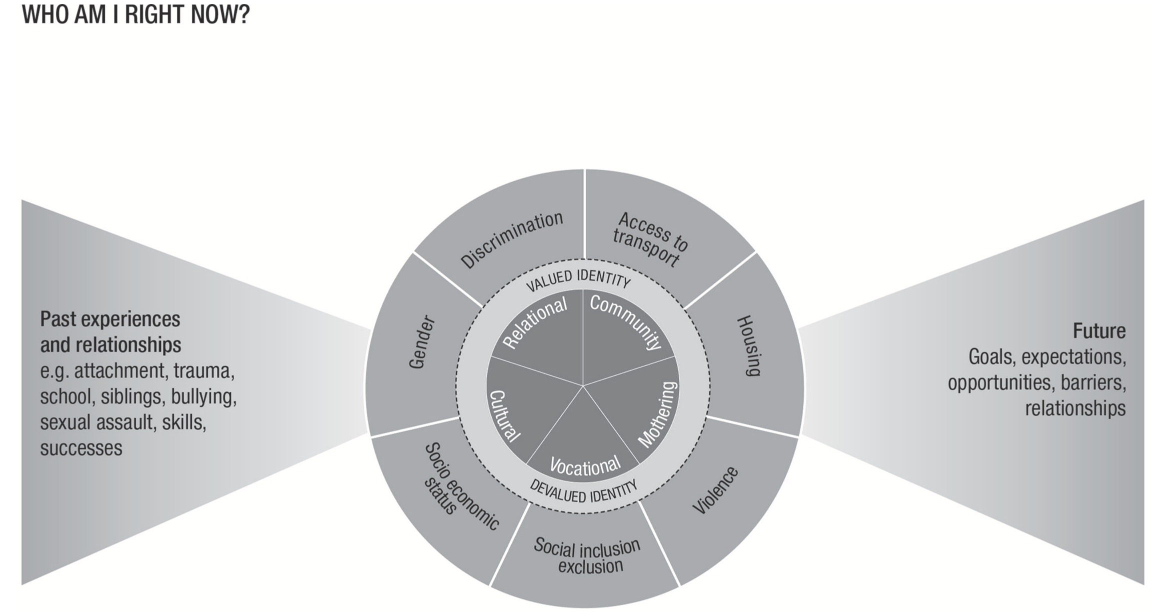

FIGURE 1 | Influences on personal and social identity for mothers with mental illness.

their personal identity. This figure was developed from the theoretical concepts that emerged from the data. Women described their identity being dependent on how they conceived their self-concept across a range of social domains. Each of these domains could be viewed as socially valued and positive or devalued and negative. Women spoke of being influenced not only by events and relationships from their past, but also their beliefs and aspirations regarding the future, thereby hope was a critical ingredient for a positive identity.

The categories of "doing it my way" and "speaking out" were indicators of significant integration of the illness experience into a woman's personal and social identity. Although the illness identity was at the forefront in "speaking out," it was constructed positively, intrinsically linked to meaning in life and empowerment, which are other recovery processes identified by Leamy et al. (1). Women emphasized recovery outcomes attained through being supported to identify their strengths, validate and normalize their challenges and self-reflect on the development of their identity across their life span. Often, however, prejudiced attitudes of others, especially associated with their mothering capacity, undermined women's self-concept.

Mothering Identity

Consistent with previous parenting studies [e.g., (26)], participants spoke of disconnection from other mothers. This was associated with socio-economics, parenting styles and difficulty infiltrating closed (well-established) social groups. Feeling disconnected, socially isolated and "different" is a common experience for mothers with mental illness [e.g., (42)] and is considered a barrier to both sustaining a positive social identity and to recovery (43) and wellbeing more generally (41).

Identifying as a "good" mother was symbolic of women's reflective capacity and internalization of social and cultural expectations as they related to the morality of parenting and the quality of relationships to children. Narratives reflected how women compared their mothering experiences to archetypes represented in their social environment. This finding echoed Venkataraman and Ackerson's (44) study on sources of parenting norms in popular culture, the media and parenting literature. Importantly, the signals women perceived from service providers, portrayed a deep understanding of how society assesses "good" mothering, and an intense desire to be viewed as competent. This facet of personal identity was susceptible to women's own internalized self-criticisms, in addition to the censorious messages conveyed by health professions or family members.

Having a psychiatric diagnosis was a core component of identity for some women, however this was not always experienced negatively, as disclosure could also bring meaning, purpose and connection, when used to educate, support or advocate around mental illness. For other women, having experienced mental health difficulties was just one relatively insignificant facet of a rich and varied life history. In this study a distinction between positive and negative identity was apparent, however this was not necessarily associated with the extent to which women embraced the illness as part of their personal identity.

Identity, Mental Illness and Trauma

The data also parallels Agnew and colleagues' study that "highlighted the complex and intertwined nature of traumatic experience, personality organization, and self/identity" [(19), p. 8]. Crucially important in defining a self-concept within the current study, were women's experiences from the past, including trauma resulting from physical, psychological or sexual abuse, the quality of early life attachments, transience in housing and schooling, relationships with siblings and parents, experiences at school and access to physical and economic resources. These factors, along with the ways in which women conceptualize them have also been previously highlighted (32, 42, 45, 46). In this study, these factors shaped the identity journey and the way the women saw themselves in the present.

For some women, having a diagnosis provided meaning and understanding that made sense of their symptomatic experiences. Additionally, this enabled them to connect with a peer network of others who shared similar thoughts, feelings and behavior and this offered validation. Mental health education and activism constituted a positive interpretation of lived experience that could simultaneously maintain the mental illness part of identity at the forefront.

Identity and Change

Identity is assumed to be fluid and dynamic within self-categorization theory (7), and while the participant interviews in this study are a snapshot in time, identity was described by participants as flexible, changing and evolving. This was evident in women's descriptions of themselves over time, and the ways in which they connected with others including disconnecting from unhealthy relationships and becoming more discerning or alternatively, learning to trust. However, there was also reference to consistent and enduring components of identity associated with preferences and strategies for managing stress and mental health difficulties. Crossley (47) adopted a narrative approach in investigating the disruptive impact of trauma, identifying the capacity for trauma to unseat previously coherent conceptualizations associated with self. Crossley (47) found that assumptions regarding one's usual patterns of thoughts, behavior and emotions are undermined along with one's temporal awareness that ordinarily provides meaning and context. This is congruent with women's descriptions of managing distress, in the early period of the illness. For the participants in Crossley's study, narratives become prominent in creating meaning, when customary psychological processes fail under the vast strain of traumatic occurrence.

Not only was it observed that events from women's past shaped their current identity, but their perceptions of their future lives including hopes, plans and aspirations were also influential in defining how they perceived themselves in the present. Identity development appeared to be a non-linear process that was highly permeable to social influence.

Recommendations for Research, Policy and Practice

The findings demonstrate mothering identity to be important for mental illness treatment and recovery. Women primarily related as mothers in articulating their self-concept and strove to be recognized as proficient in this role, highlighting their skills, strengths and underpinning parenting values as critical components of their identity. Therapeutic interventions need to explore mothering relationship to self and others, as "therapeutic understanding that takes into account the deficiencies within diagnostic criteria and acknowledges the diverse nature of self and identity of an individual may improve the therapeutic relationship" (19). Within such interventions, women need to feel safe and secure to explore their identity "journey" including the interactions and events that have led to their current self-concept.

A strong therapeutic alliance is critical to cultivating a safe space within which women can begin to address the issues that underpin their healing and recovery (48). Women spoke of the importance of this particularly in the early parenting phase, while adjusting to a new mothering role, and not yet feeling confident in their parenting knowledge and skills. The women who participated in this study indicated that validation of normalcy of parenting challenges can be useful at this time, as well as a more conversational approach that moves beyond the assessment checklists, to the development of an authentic relationship that offered individualized support, rather than reinforcing a sense of being "monitored" and judged.

Elevating the significance of identity and self may reveal areas of intervention that can support more flexible, nuanced and realistic expectations surrounding women's multiple roles and activities. Identity work, integrating past experiences that may challenge individual's assumptions regarding their identity (43) and reflecting on emotional and behavioral responses, is acknowledged as a core component of the recovery process (1, 48).

Beyond this, the current research suggests that investigation of identity and self in the context of social and environmental conditions must incorporate critical reflection on the dominant norms that may be oppressive and result in devalued status within various identity domains. Challenging and contextualizing these dominant assumptions may be particularly significant for marginalized population groups (e.g., Indigenous, people identifying as GLBTI), who are demonstrated to be at increased risk of developing mental health difficulties (49).

Discussions at this deeper conceptual level may be perceived as challenging to establish in the midst of psychological distress, and practitioner judgment in ascertaining readiness for such discussions is essential. However, avoiding these topics can pathologise individual responses to issues that are associated with layers of structural inequality. Future research should also focus upon overcoming workforce barriers within mental health and family services to engaging in identity work with women who have a mental illness.

Mental health policy needs to recognize and reflect the importance of identity work as a crucial part of practice. A disproportionate focus on medication and risk management within clinical mental health continues to stifle recovery oriented practice that encompasses a holistic view of people including consideration of their diverse and multi-faceted roles and relationships.

CONCLUSION

The importance of developing and sustaining an identity that is multifaceted and socially valued has been under-acknowledged within mental health services, despite positive identity development being repeatedly identified as a key characteristic of the recovery process. Fostering a positive self-concept, particularly associated with one's parenting role, can assist in facilitating personal recovery in mental illness.

For women who are mothers with mental illness, gendered norms around the mothering role can result in the imposition of unrealistic expectations of women's functioning, particularly if they are living in impoverished social and economic circumstances. Identity work needs to incorporate consideration of the personal level including past experiences, relationships, thoughts, emotions and behavior, as well as the broader environmental context.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of the Australian National Statement on Ethical Conduct in Human Research. The protocol was approved by South West Healthcare Multi-disciplinary HREC, Monash University HREC and Ballarat Health Services and St John of God Hospital HREC. All subjects gave written informed consent in accordance with the Declaration of Helsinki.

AUTHOR CONTRIBUTIONS

RH, DM, and MG developed the research design and methodology together. RH undertook the literature review and the data collection and led data analysis. DM and MG contributed to data analysis and all researchers were involved in initial coding and conceptual analysis as well as developing the findings. RH wrote the manuscript with editing/contributions from DM and MG.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2019.00089/full#supplementary-material

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A Narrative Inquiry Into Global Systems Change to Support Families When a Parent Has a Mental Illness

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The issues that confront families when a parent experiences mental illness are complex. This often means that multiple service systems must be engaged to meet families' needs, including those related to intergenerational experiences of mental health and illness. A multisystem approach to public mental health care is widely recommended as a form of preventative intervention to address the effects of mental illness and its social, psychological, and economic impact upon parents, children, and families. Globally, a multisystemic approach to care requires a change in the way systems are currently organized to support families, as well as the way systems are interacting with families, and with each other. This qualitative secondary analysis emerged from a primary study examining global systems change efforts to support families, including components of change that were common and considered successful in different countries. A narrative inquiry method was used to re-analyze the data by compiling the stories of change described by individuals from participant countries. The data were interrogated to ask questions about story content, and to identify who was telling the story and how they described important changes across different geographical and cultural contexts. The individual stories of 89 systems change experts from 16 countries were then compiled into a shared global narrative to demonstrate international progress that has occurred over time, toward multisystemic change to support families where parents experience mental illness. While the global narrative demonstrates considerable overlap between pathways toward change, it is also important to document individual stories, as change pertains differently in different contexts. The individual stories and the global narrative illustrate how countries begin a journey toward change at different time points and may have various outcomes in mind when they commence. Study findings raise questions about the extent to which systems change can be standardized across countries that have unique social, cultural, political, and economic features. This study provides several potential points of reference for countries considering, or currently undertaking systems change to support families where a parent has a mental illness. It also provides an important story about international efforts undertaken to improve outcomes for families.

Keywords: system change, global mental health, parental mental illness, intergenerational mental health, family mental health, children of parents with a mental illness, qualitative secondary analysis, narrative inquiry

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INTRODUCTION

Mental illness represents a substantial proportion of the world's health problems, with lifetime prevalence estimates between 18% and 36% (1), accounting for 13% of the global burden of illness (2). Globally, approximately 15–23% of children may live in families with a parent who has a mental illness (3–5). Mental illness affects families. Children who have a parent with a mental illness are at increased risk of developing their own mental health problems with approximately one third at risk of serious mental illness and another third at risk of any mental illness (6, 7). Mechanisms of risk include disrupted parent–child interactions, exposure to social disadvantage, genetic and epigenetic processes, and a lack of mediation of other risk factors (8). Risk is most likely to be transmitted through a complex interplay of neurobiological, genetic, and psychosocial factors (9). Families affected by parental mental illness can experience vulnerabilities including sustained stress and cumulative adversity (9) and are at significant social disadvantage due to factors such as poverty and social isolation (8). The effects of parental mental illness are likely comprised of bidirectional interactions between risk and protective factors at the individual, relational, community, and societal levels (10), requiring multilevel social and collective action (11). Families are affected by their environments, including a wide range of ecological factors (10), such as poverty, homelessness, interrupted education, incarceration, and political or environmental disadvantage (11). These factors can have long-standing impacts on families across generations (12).

Recovery from mental illness is also increasingly being understood to be a social and relational process occurring often within family contexts, particularly when parenting roles and children are involved (13, 14). A multisystem "whole of family" approach to mental health care has been recommended as a form of preventative intervention to address the impact of intergenerational mental illness on parents and children (15, 16). Understanding the needs, experience, and context of the whole family is of central importance to addressing parental mental illness, but is not identified to be universally well embedded in practice within any one system (4, 17). A systems approach requires partnerships beyond mental health, within education, welfare, primary health, social care, public health, and social policy development (18). Many challenges exist though at the level of the practitioner, organization, service system, and cross-service systems to provide a "whole of family" approach that meets the needs of these families. Gaps between what is known in research as best practice to promote family mental health and actual practice delivered and sustained on the ground is an issue for many countries across the world (18).

The help families do receive is most often from services that are themselves fragmented, particularly between the adult and child service systems (19). Multiple barriers prevent easy integration across service systems to meet the many needs of families. Funding for services is limited, and system barriers include a focus on the individual with the presenting issue as the point of service, rather than the family (20). A significant factor influencing systems level change is the ability for health and mental health funding arrangements to permit a broader focus on the needs of the individual with mental illness to, for example, incorporate key relationships with family members and the social context of a person's life (e.g., parenting) (18).

Mental health promotion strategies require an intersectoral approach to implementation, as well as the use of multiple methods (21). These forms of cross sector engagement that require an integration of approaches are also known to be slow processes that unfold as a series of cumulative developments over time (21). For families where parents have a mental illness, the indication for system change comes from compounding vulnerabilities and the health, social, and economic factors that contribute to them (22). Health promotion frameworks suggest that change should be situated across all the systems that connect with families and their individual members (21).

In the context of risk to children and their families where a parent has a mental illness, Foster (23) identified that intergenerational mental illness may indeed be a "wicked problem." This is because it is a significant social problem and complex public health issue, with multiple causal pathways and policy implications that are resistant to simple resolutions. Stories of change that have taken place or may still be unfolding in differing contexts may provide insight into global progress on the issues affecting families where a parent has a mental illness and on outcomes for these families. Such stories may also provide important guidance for service settings, including those of individual countries, who see themselves at the beginning of systems change on this topic. Rather than each country embarking on a separate or isolated journey toward change to address similar issues, a shared narrative of global change may be of benefit to those who can utilize the knowledge gained.

STUDY DESIGN

Aim

This study aimed to construct a narrative of global systems change to support families where a parent has a mental illness through an examination of individual stories of change from countries across the world.

Research Questions

How do individual experts in different countries describe stories of system change to support families where a parent has a mental illness?

What do individual narratives of change tell us about systems change processes globally?

Study Data

The analysis reported in this article is a qualitative secondary analysis (QSA) of data produced in an international Delphi study examining the concept of systems change to support families where a parent has a mental illness. QSA reuses existing data, collected for prior purposes, to investigate new questions or apply a new perspective to an "old" question. An immediate conundrum in speaking and writing about QSA is the difficulty posed by language. QSA is a study in its own right; to avoid confusion, researchers generally refer to the primary study (the Delphi), as a way of distinguishing between the previous work and that to be performed in the secondary analysis (24).

The Primary Study: An International Delphi Study Into Systems Change

The primary study was based on the experience of an international research group working in the area of families where a parent has a mental illness. For the primary study, key system change experts on the topic were identified through the networks of the research team and their professional affiliates in each of their respective countries. Individuals in each country were invited to participate in the study *via* email, followed by snowball sampling thereafter (25). Ethics approval was provided by the lead university and the affiliated recruiting service in each country where required.

Participants in the primary study included service development workers, policy makers, managers, practitioners, and health or welfare workers who had expertise at working at the system level as well as an understanding of change over time in their respective countries of work. This included psychologists, social workers, occupational therapists, mental health nurses, psychiatrists, researchers, consumer advocates, government officials, and administrators.

The first round of the Delphi study asked open-ended questions about experiences of system change within the participants' geographical and cultural contexts. Specific systems were not specified but systems change was defined as any workforce, policy, legislation, or other mental health promotion strategy or development that aimed to identify and support parents with mental illness and their families, including their children. Participants were asked questions about: steps and approaches to systems change initiatives undertaken within their countries, locally or nationally; factors that facilitate change or remain as barriers to change; the most significant change that had occurred up to the point of the study; and considerations about future work needed to bring about systems change. The direct role of the participants in systems change was also explored as a way to understand their experience and efforts undertaken over time.

The QSA: Developing a New Research Question About the Experience of System Change Using Narrative Inquiry

In preparation for subsequent rounds of the primary Delphi study, a thematic analysis was used to identify key categories based on data from the preliminary round of questioning addressing individual experience with systems change in each country (26). As the thematic analysis was reviewed, questions arose about how to retain the stories of change described by participants and how, or if, individual stories might fit together in order to construct a global narrative of change. A common problem identified in qualitative data analysis is that categorization can result in data fragmentation such that the "meaning of the whole" is obscured or lost altogether (27). The thematic analysis, while useful for developing further rounds of the primary Delphi study, was not designed to capture narratives of systems change because the analytic emphasis was on determining common elements related to each country's experience as it unfolded across time and place. Consequently, narrative inquiry was chosen as a methodology for carrying out secondary data analysis, to answer a different question about "systems change," as storied.

Narrative Inquiry Methods

Following a modified version of Labov's (28) narrative framework, specific questions were used to analyze and interpret the data from each participant in the primary study. Analytic questions included a focus on how the participant orients the audience to a story about systems change, asking, "What happened first?" (the orientation). This was followed by questioning "What happened next?" or "What happened once the initial change had taken place?" (the complication). "What still needs to happen?" reflected an analytic focus on understanding how stories of change might be unfolding currently, considered unfinished, and seeking resolution of some kind (the resolution). The next step in the analysis and interpretation involved compiling the individual country-by-country stories into a collective narrative (29), or broader story of global systems change.

The narratives produced in this study are based on an analysis and interpretation of data produced by participants in the primary study. Participants varied according to their country of origin, as well as the disciplinary and professional contexts (or systems) in which they worked as experts in systems change. This variation was also the case for the research team that performed the QSA. It is important to acknowledge that these contexts influence the stories that are told—those that are both proximal, local, or nearby, as well as more distant contexts, which lie outside of but influence the immediate encounter in which the data are produced, and while these stories are embedded in a wider story of change, they are not all of that story.

Reflexivity is a key analytic strategy in qualitative research because critical self-reflection (including collective team-based reflexivity) is used to question and document assumptions individuals bring to a study topic. This knowledge is accounted for and used to produce additional insight into the data during analysis and interpretation, and to acknowledge that all analysis is always partial, tentative, and provisional, and open to re-interpretation (30). In our case, understanding the significance of contexts, as part of a reflexive research strategy, provided additional insight into the complexity and subtleness of the situation under study, enriching our ability to interpret what the data might mean, and acknowledging that this is not the entire story of systems change that might be told. Reflexivity was maintained as a research practice to ensure the trustworthiness of the QSA among the research team throughout the study (31). For example, regular fortnightly data analysis meetings were used to reflexively consider and document researcher assumptions and responses to the data, consider the ongoing analysis, and interrogate and challenge emerging interpretations, including how stories were represented in writing up the analysis.

RESULTS

The 89 participants who responded to the Delphi study self-identified as systems change experts. They came from 16 countries, primarily from high-income countries. The majority were aged between 40 and 60 and had worked for over 10 years in their fields, suggesting a depth of experience. Most worked in public mental health services in a variety of professional roles, which is important in considering their points of reference when talking about the systems (see **Supplementary Table 1** for participant demographics). Findings are presented using the modified analytic categories of Labov's model described previously (the orientation, the complication, and the resolution).

What Happened First? (The Orientation)

The orientation introduces how the story of change began, according to the study participant. For example, change was described as being inspired by shifts in national policy, the introduction of an intervention model, or the creation of practice guidelines; however, it was not always apparent what motivated initial change. Sometimes change occurred, or became apparent, because it was opportunistically connected to other, related change initiatives, such as social movements associated with consumer advocacy or general mental health reform. In other instances, change could be traced back to a critical incident, such as the death of a parent or child and the system review that followed, or to the explicit efforts of a particular group of leaders who were variously described as, "pioneers," "early adopters," "enthusiasts," "advocates," or "champions" within systems change. Participants did not always situate the beginning of the story in the years that led up to change, but rather they chose a particular point in time to represent when change happened. For example, a participant described securing funding as the point of change rather than the years of lobbying that had led up to this point. Participants who had been involved in years of advocacy often went further back in time to locate the beginning of change among particular disciplinary efforts toward awareness raising, as described by a participant who stated that, "The movement began in the late 90's where some child psychiatrists and adult psychiatrists became interested in the relationship between parental mental illnesses and children's difficulties."

Participants positioned stories of change within multiple types, levels, and dimensions of systems. Stories were based within local initiatives and projects, regional collaborations, or national campaigns and policies. The need for change was directed toward the individual practitioner or the wider service system and at the local, national, or international level. Participants reflected on the efforts of small motivated groups of individuals working to advocate for and drive local practice change within systems, as well as efforts to foster the government and political buy-in required to provide resources and sustain future systems change.

Although at times participants used the concept of time to orient their stories, which included notions about progress or the movement from one point of change to another, this was not consistent within or across stories. Participants varied in how they located their country's progress toward change. For example, they described their country as being at the "beginning" of a journey, having come only recently to an awareness of what some of the issues for families in these circumstances might be. This is in contrast to others who described the journey as more of a refinement toward systems change, with already-established and multilevel buy-in from key players. How they perceived their country's position in terms of systems change, as a temporal ordering of events, was relative and dependent on how they interpreted what progress might look like. This was the case where struggles to provide basic services within the mental health system were still considered progress. While others, who had advocated for many years in this field, and had made significant legislative changes specific to the inclusion of families in care provision, described their story as, "still having far to go." Participants from different countries often compared themselves to other countries in taking a position and providing a rationale about their (lack of) progress on the topic, as illustrated by the following participant, who said, "We are back years with respect to other countries that have developed preventive programs. The stigma towards mental illness is still high and there is no culture of prevention."

As individuals, participants positioned themselves in different ways within their own stories, and in the process of change that they wanted to see happen. For example, systems change was sometimes described as something external to the individual, which had to change, whether or not the participant actually worked in these systems. Participants depicted other players as more responsible for change in stories where *they* (not *I*) were the main protagonists, or the system (*itself*) was expected to change, as if it could do so devoid of human agency. This was in contrast to other stories where the pronoun "we" suggested that the participant expected to play a significant role in changing power structures for systems change to occur. The specific use of pronouns in these stories indicated how participants thought about their role and responsibility for systems change, to what extent they expected this to be an active rather than a passive role, and whether or not they saw themselves as an integral part of the change process. This is nicely illustrated in the following statement in which the participant uses the inclusive pronoun, "we," to suggest that there is a collective responsibility to hold others to account for change that may have taken place, but has not been effectively implemented, "We need to be more hands on and make them accountable for adhering to the policies that are already in place."

BOX 1 | Orientation to the global narrative.

Globally, systems change to improve outcomes for families where a parent has a mental illness has been initiated within differing systems and at multiple system levels.
The identified starting point of change can occur after years of action, awareness raising, and advocacy by individuals within and outside systems examining ways to improve internal and collaborative processes. Change can also begin suddenly or opportunistically in response to other pressures or actions within systems. Actions to initiate local change to practices or processes require strategic buy-in to progress toward sustainable change within or across systems.

What Happened Next in the Stories? (The Complication)

The complication explores what happens after change is initiated in these stories. Initial change was often followed by other actions within and between organizations to drive overall systems change. For example, further change might occur through changing current organizational policies and procedures, or by establishing a network of champions to support practitioner development. Collaborative models, particularly between mental health and social care/family welfare systems were frequently sought. Some countries sought government funding to scale up the integration of evidence-based family interventions across all services working with parents. Not all countries included stories of systems collaboration as the necessary next step. Some were yet to decide on which system should take primary responsibility for supporting families, or how to collaborate with each other, as illustrated in the following quote, "There is no consensus about whether the aid [for families] should be in the health or social sector and how cooperation should take place."

Regardless of where change first began, efforts to move in a concerted direction were questioned with respect to sustainability, with progress seemingly tenuous, comprised of fragmented examples of good work, requiring ever more resources to sustain change. "There are pockets of good practice but this is very patchy so I feel we are a long way from being able to say that there has been any significant shift in the right direction." Initiatives were described as scattered, carried out by a small group of enthusiasts or driven by a motivated individual, and even if initially successful, were compromised by loss of funding. Aspects of change were described as not being large or cumulative enough to be sustainable.

Whether participants from different countries believed they had achieved systems change was not always clear. In some instances this lack of clarity was illustrated by the limited descriptions about what kind of "shift" was actually hoped for and how change would be recognized once it had taken place. In some cases there was a desire to move beyond simple awareness raising (about the needs of families), and the enthusiasm of advocates, toward ensuring individuals would act to achieve actual change: "It is not enough to raise awareness or gain people's enthusiasm. They must know that it is within their grasp to make a difference."

Even where some significant change had occurred, the need for change was characterized as unremitting because as this participant points out, once change has occurred in one area it must be followed up by a change in another area related to "the problem," "There has been a shift from having to argue the need to work with families … to one about having to drive how to work [with families]." In another example, even in countries where systematic processes for identifying and taking responsibility for families was mandated by policy, or by law, there was difficulty bringing about the requisite change to ensure these requirements were met. Participants suggested that even when there was significant policy change in place this did not guarantee that individuals would change their practice to ensure policy was adhered to. Nearly all stories described change as a process that was ongoing, and that significant multisystemic shifts had not yet been sustained, but were desired.

Stories included specific and more general ways that change had occurred, including: seeking government funding and endorsing new legislation; implementing strategic approaches to identify families and their needs, as well as thinking about holistic approaches to care; increasing awareness of familial mental illness; and piloting new interventions aimed at family members. Desirable aspects of systems change were noted in the adjectives attributed to systems and services that were considered "trustworthy" and "honest" and had an "understanding" of families embedded within their organizational cultures. How services might work together or systems might be coordinated to address families' needs was not detailed in the data. Stories of change over time might be best described using the metaphor of "a journey" to draw attention to change as a process, rather than suggesting it necessarily has a specific or predictable end point. This idea is illustrated in the following quote: "Change is a journey rather than a destination … the reality is that it isn't a linear process but one that takes up and down journeys."

BOX 2 | Complicating Action in the global narrative.

Any initial change to support families within systems is always followed by a need for more action to drive and maintain overall systems change. Difficulties identifying and responding to families continue broadly across current systems. Limited sustainable resources and a lack of cohesion about consistent approaches to support families between and within systems contribute to these difficulties. There are multiple differing pathways toward change that are effective and no identified "best way." Fragments of change exist globally, but clarity around a shared vision to achieve sustainable systems change to support families is a desired next step.

What Still Needs to Happen in the Stories? (The Resolution)

The resolution explores what still needs to happen to advance systems change and to sustain progress achieved to date. The resolution brings the story into the present, but also looks to the future. All stories suggested that there was much work still to be done, as summarized by the statement: "…there is still a long way to go." This suggested an implicit idea that stories are meant to achieve resolution, but this was portrayed as far off. Often, an end point seemed to move further away the more progress the country made, as the complexities of a systems change process became more apparent. Countries with established intervention programs, models, and guidelines identified as having a long way to go, whereas those who situated themselves early in the journey described a clearer path forward.

Time frames used to describe ways forward were somewhat arbitrary and difficult to compare. Unsurprisingly, perhaps, stories were framed by the concept of progress. Progress was valued as a positive aspect of change, because it implied that journeys were moving toward some type of goal even if this was not yet defined. The end point was not necessarily explicit or shared openly. However, while stories were not always told as a linear narrative, most described "moving forward" in some way to achieve a "whole of family" or "whole of system" approach, where services worked collaboratively to provide support to all family members. Progress toward change was considered slow and ongoing but implicitly finite. "I have been working already for 25 years in this topic and it is still not finished."

Participants were clear on what steps were required to make progress; the need for resources was frequently emphasized. A common story thread described the frustration of funding given, but later retracted. As one participant indicated, "implementing without money gets to be tiresome." Stories proposed future steps that included; continued lobbying, awareness raising, the participation and inclusion of the voices of families, and continued funding for development of systems change initiatives. Monitoring systems were thought to also help keep the work on the agendas of local organizations and governments. University education and academic research are important for a sustained focus on the topic. Shifting toward prevention was identified as an aspiration for many, although how this is defined or might be undertaken was not described.

Legislative change was perceived to be the most ideal form of change, both by those who had experienced it, and those who wanted to achieve this, "Now we have these national recommendations but it is only recommendations … on what should or could be done and not a judicial document on what needs to be done as they have in other … countries." Legislation was seen as necessary in order to bring other measures to enact change, such as practice guidelines and recommendations. Legislative change on its own was not enough because other changes were required to ensure the intentions of the laws were systematically implemented and the individuals responsible for carrying out these laws were suitably skilled.

The change people were working toward was broad and cross-sectorial, requiring complex solutions that go beyond changes to individual systems. Rather, change required broad cultural or paradigmatic shifts in thinking. Participants identified that cultural and social understandings, and beliefs about mental illness, influenced systems responses to families. As a result, many stories included community awareness campaigns to reduce structural barriers to support families as noted by a participant who said, "To continue to break down the stigma of mental illness so families are open to access supports." Within and across services, participants expressed shared ideas for shifting the culture of systems to focus holistically on families, rather than individuals, and for moving the focus of funding and practice toward prevention and early intervention. As illustrated in the data, even in countries where the change process was seemingly well underway, there still remained a dominant biomedical and individual model of illness and health which determined funding and service system structures. Participant's suggestions for creating such a shift included, piggybacking change initiatives onto other movements toward collaborative care work, trauma informed care, recovery, and social determinants of health.

BOX 3 | Resolution to the global narrative.

Current efforts toward systems change are occurring at multiple and different levels, resulting in potential reconfiguration of systems as well as change within individual systems. Sustained change will be limited without significant shifts within broader political, cultural, and economic structures toward prevention and health promotion; as well as recognizing the importance of family within models of health and illness. Globally, there is significant and broad commitment to improving outcomes for children, parents, and families and numerous strategies in place to ensure awareness and response to families within systems.

DISCUSSION

It is known that improving mental health outcomes cannot be achieved by changing one system alone but by engaging in collaborative practices between health, education, child protection, and social welfare systems to help shift the sociopolitical and economic determinants of mental health (2, 32). The current study illustrates the complexity of systems change to support families where a parent has a mental illness and highlights the need for coordinated action in multiple spheres for long-term sustainable change (18, 23, 33, 34). While there is a growing knowledge base of programs and strategies that can support family focused practice within systems, an approach aimed at changing the actual systems is required to ensure integrated, consistent, and intergenerational support for families (16, 18).

Underlying any systems change work are assumptions about which systems are relevant to families and that outcomes for families can indeed be improved by cumulative and progressive shifts in systems and the practices that they employ. The current study highlights diverse understandings of systems change and indicates the significance of considering what drives perceptions and recognition of the "need for change" in this field. This includes for example, a question about how "systems" are defined by different individuals (in different cultural contexts) and how or why this may be implicated in decisions about which kinds of changes are necessary to support families.

An important question raised by our narrative analysis is what is considered necessary to achieve change, and why systems change to support families is indicated. The stories in our data described legislative development as a desirable outcome and an important marker of systems change. However, despite significant and strategic but singular changes like this, a prevailing individual model of illness dominates mental health services globally and will likely continue to impede the integration of families into social and mental healthcare (2). Broad paradigmatic change in the ways we think about and practice global mental health to support families is necessary, to address the complexities of systems change described in these stories. As a result, the change process needs to be adaptable, possibly to work outside "systems," and in ways that acknowledge that there is no one right way of doing things (35). A dynamic process to explore systems change includes challenging currently held assumptions about the topic and an iterative approach to theory and practice to systems change that occurs over time (36).

As a method of analysis narrative inquiry imposed a useful linearity on the complex storied characteristics of our study data concerning systems change. This analytical lens helped us to compile stories from individuals in different countries, and to structure these stories into a plausible narrative of global change. We do not intend to imply through this methodological choice that there is a set point of entry into stories of change, or to suggest that we know when stories about systems change are finished or complete. This caveat is reminiscent of the implementation science literature in which innovation takes place as a series of stages or phases (37). While change may be depicted as linear, in practice it often follows a variety of nonlinear, recursive, or re-iterative pathways (38), which are characterized by shocks, setbacks, and unanticipated events, similar to those described in our data (33).

Narrative analysis allowed us to see that it is difficult to determine when systems change is achieved, precisely because broader social and political contexts influence what is considered a successful outcome. These stories did suggest that simply aiming to change systems to improve outcomes for families might not be an end in itself, or enough on its own. This also has implications for thinking about how to define and measure systems change, whether or not the focus is on successful change. While systems change explored in this global narrative had no specified shared outcome related to an understanding of work to be completed or finalized in this field, it is clear from the responses of our participants that there is a global and multisystemic commitment to improving outcomes for children and families. There are large amounts of standalone and shared efforts to make progress on local, regional, or national systems change, however they may be improved upon with the resources available in different social, cultural, and geographical contexts. Further examination would be needed to track sustained benefits, to be able to understand outcomes achieved within global systems change (34) within its specific context, over time (33, 39, 40)

A recognized and well-defined "field" was necessary to identify systems change experts for the first stage of the primary (Delphi) study. However, the countries whose contributions are absent from this narrative also form an important part of the global picture of systems change. There are likely countries who have yet to begin work in this field, or who might conceptualize the problem and the solution, differently. It is hoped that providing a narrative analysis of systems change may help to guide countries who are just beginning to recognize this as a topic of concern, although their stories will likely be different. The systems change experts in this study came from a broad range of cultural and professional contexts and were shaped by the countries, cultures, and systems within which they lived and worked. They were influenced by local understandings and cultural discourses about mental health, as well as wider discourses about health and illness and what it means to be a family. These contexts form part of a global narrative of change, with each story contributing to the narrative, and many aspects not yet explored or understood. The stories responded to study questions about change over time. Consequently time was depicted as relative in these narratives, something happened and then more events occurred, but there was always something more to be accomplished. As people may experience and organize how they talk about their lives through time (29), it was not surprising that participants began their stories at point in time that made sense within their contexts. The resulting global narrative reflects the current moment in time during which this study was conducted.

To change systems there has to be a belief that systems are structural entities that do exist and are amenable to change. This belief may be a function of living in a highly resourced and privileged context that can support "systems" in a particular way. Across the globe there is scarcity of resources to meet the health and mental health needs of populations, resulting in inequitable systems to support people with mental illness and their families (2). While the capacity to include families in approaches to mental health care will certainly be impacted upon by the financial, cultural, and local processes of countries, global mental health directives maintain a strong focus on mental health promotion and prevention (41). Such a focus requires shared actions of governments, civil societies, international development agencies, and academia/researchers (41). Going forward there is a need to utilize systems change approaches that enable mutual knowledge exchange and enhance integrated understandings of global systems change. This includes overt recognition of differing notions of systems, families, and their needs across countries, cultural groups, and populations, as well as partnerships with emerging systems for mental health support in low- and middle-income countries.

While many societies may be replete with change agents, "a restless mix of individuals and organizations set on transforming the world," their achievements have been described as "islands of success in a sea of failure" (35, p. 12). Stories in this study suggested that the longer countries undertake change, the further away success may appear. It is possible that those in the process of change for longer periods of time may identify the power dynamics embedded in policies and structures that continue to disadvantage families, and this may shed new light on further work to be done to bring about systems change. Considering why things don't change can be an important step toward understanding global barriers to change (35). In the meantime as Green (35) argues, "We must become comfortable with ambiguity and uncertainty, while maintaining the energy and determination to succeed" (p. 28).

CONCLUSION

Systems change experts around the world describe stories of change across systems that are diverse, pathways that are convoluted, and have only an incremental awareness of the need for family integration in individual service systems. The stories analyzed in this study can inform other stakeholders and countries as they embark on journeys toward sustainable change to support families where a parent has a mental illness. Systems change requires motivated and passionate individuals, opportunistic endorsement of the need for change, and sustained financial backing. Change can occur in different ways and with differing intentions about desired outcomes. The compilation of individual stories, coupled with a global narrative, allowed us to show how the complexity of this field might be addressed by focusing on family-focused practices within individual systems, as well as collaborative care practices across systems. The findings tell a story of complex change that highlights the different ways in which countries begin their journey; how their pathways overlap, and how the successes as well as the setbacks are experienced in cumulative attempts to improve health and social outcomes for families. The global narrative reminds us that there are many pathways to change and that it is important to recognize achievements along the way, including the potential to develop a shared outcome to support families. On this last point it is important to acknowledge that shared outcomes must be developed by including the voices of children, parents, and families who experience and use services within systems. They are missing from the current narrative but are integral to any evidence for developing systems change that might work better to support families.

LIMITATIONS

The findings and the global narrative are a compilation of the perspectives of those who contributed to the study data at a particular point in time. The participating individuals from select countries were involved in the QSA because they were associated with the network of international researchers in the primary Delphi study, and because they self-identified as experts in system change in this field.

ETHICS STATEMENT

The primary study was carried out in accordance with the recommendations of The National Statement on Ethical Conduct in Human Research, 2007, National Health and Medical Research Council Act with implied informed consent from all subjects. All subjects gave informed consent in accordance with the Declaration of Helsinki. The protocol was approved by the "Monash University" Human Research Ethics Committee.

AUTHOR CONTRIBUTIONS

MG leads the primary Delphi study. BG guided the QSA method. SI, BA, and MG led the data collation and early analysis. SI drafted the paper. All authors analyzed the data and contributed and edited the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2019.00310/full#supplementary-material

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BRIEF RESEARCH REPORT
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Collective Impact on Prevention: Let's Talk About Children Service Model and Decrease in Referrals to Child Protection Services

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Background: Families with parental mental health issues often have numerous problems needing multilevel measures to address them. The "Let's Talk about Children Service Model (LT-SM)" is a community-based service approach aiming at collectively impacting population needs regarding child protection services. Three municipalities in the Raahe District (RD) of Finland requested implementation of the LT Service Model. This paper describes the model and first results.

Methods: The LT Service Model connects relevant stakeholders with families and their social networks aiming at the shared goal of supporting children's everyday life at home, kindergarten, school, and leisure environments. Parents, teachers, and other caretakers are supported by LT interventions. An infrastructure for collaboration, decision making, monitoring, training, and feedback is established, embracing health, social and educational services, and other stakeholders. Referrals to child protection services were compared with national data before (2009–2013) and after implementation of the LT Service Model (2013–2016). Analyses were conducted using the joinpoint regression method.

Results: There was a significant decrease in the underage population referred to child protection services in RD (AAPC $= -6.9$; $p = 0.013$) between 2013 and 2016, in contrast with an increased rate nationwide (AAPC $= 1.9$; $p = 0.020$).

Conclusion: In the LT Service Model, prevention starts in children's everyday life as the uniting, common goal for multiple stakeholders and an integrated service structure is developed to support this effort. The first results are promising, showing an appreciable decrease in referrals to child protection services, although further research with longer follow-up and across other municipalities is needed.

Keywords: collective impact, child protection services, family adversity, Let's Talk about Children intervention, Let's Talk about Children Service Model, parental mental health disorders, prevention

INTRODUCTION

Intergenerational transfer of parental problems and family adversity is an individual, family, and societal concern. In Finland, parental mental health and substance abuse problems, often associated with other adversities, are also major reasons for child protection measures (1). Family adversity, as well as family resilience, is multi-determined and embedded in the social ecology of the family and society (2). Multilevel prevention is necessary (3) and comprehensive approaches are needed to achieve a collective impact on population-level outcomes (4).

The "Let's Talk about Children Service Model (LT-SM)" is a community-based model for promoting child and family well-being and resilience and preventing family and child dysfunction. It is aimed to overcome the fragmentation of services and lack of common goals, which have been major obstacles for integrated approaches in health, social, and educational services (4). The core of LT-SM is supporting children in their everyday life at home, kindergarten, school, and leisure environments (children's developmental contexts). Parental and family problems interfere with everyday routines and interactions with a risk of compromising the child's well-being and development (3). In LT-SM, health and social services, as well as kindergartens, schools, and available non-governmental organizations, commit to a shared goal: to support, in collaboration with others, children's everyday life in all developmental contexts and to build the corresponding service structure.

Within this model, LT interventions (5–8)[1] are used with families; LT is a relational, cognitive, and behavioral intervention approach aiming to support parents, teachers, and other caretakers in their everyday interactions and activities with children. It is used within mental health and substance use services as well as in cases of severe somatic conditions. In kindergartens and schools, LT for Schools and Early Education (mielenterveysseura.fi) is used universally. The focus is to enhance sensitive and supportive interactions with the child as well as mutually supportive parent-teacher relationships based on a shared understanding of the child at home and school. Children participate in this program depending on their age.

LT Intervention 2018 (www.mielenterveysseura.fi) includes two steps. LT-Discussion depicts the child's ordinary day in all developmental contexts and an action plan is made with parents to enhance the identified strengths and to give support in vulnerabilities; this step is performed in one or two semi-structured meetings. When needed, the second step LT-Network meeting is planned with parents and convened to carry out the necessary support with the family's social network, services, and possible others. LT-Network meeting provides a forum for case-based cross-sectoral collaboration, including also the families and their social network.

RD decided to implement LT-SM in 2011 with the purpose to increase family and child well-being indicated by a reduction in referrals to child protection services (9, 10). It was hypothesized that there would be a decrease in the referrals after an initial increase due to revealing latent needs and that this process would take several years to yield results (11).

The present research paper describes LT-SM and findings related to referrals to child protection services. A detailed description of the implementation of this model is beyond the scope of this paper.

RD includes an industrial town and two smaller rural communities consisting of a population of 35,000 inhabitants with 8,300 under-aged children. The unemployment rate was 10% in 2012. Finland has national health and social services that also cover RD.

MATERIALS AND METHODS

Let's Talk About Children Service Model and Its Implementation

Strategy Level

Both administrative and political leaders in the health, social services, and educational sectors committed to implementing LT-SM at the turn of 2011-2012. LT-SM was incorporated into the Plan for Children's Well-being.

Organizational Level

Multiagent management group (MMG)

MMG is needed to run the implementation and develop cross-sectoral collaboration. This group includes leaders from all relevant sectors. One of its main functions is to report the progress and challenges concerning the implementation of LT-SM. Issues that need to be solved are related to subjects such as decision-making, budgeting and allocating resources across sectoral divides, ethical and practical guidelines relating to reporting and monitoring, documenting patient/client/student records, and networking with families.

MMG was founded in RD in early 2013. MMG meets four times a year and also based on ongoing needs.

One contact service (OCS)

OCS coordinates case-based collaboration. It arranges LT-Network meetings with the necessary participants. Therefore, it also encounters challenges in collaboration and reports them to MMG. Functional feedback system allows the organization to learn from experience.

OCS was created in RD in August 2013 and is being run by two social workers. One phone call or email is enough to activate the Service. As the work of this service concerns prevention and resilience, even a timid and isolated child needing a hobby in a poor family is a valid reason for action.

Workforce Capacity

Different levels of the workforce, from administrative leaders to practitioners working with service users, are educated concerning the main principles, and the ethos and praxis of LT interventions and LT-SM. Method training in LT interventions (in a span of 3 days) is organized for those practitioners working with family members. Training trainers (5 days) is organized to ensure the sustainability of the model. A long-term plan for training is prepared to overcome disruptions due to changes in staff.

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[1] https://www.copmi.net.au

Method training in LT interventions started in RD in August 2012. In the first round, 30 practitioners coming from health, social services, and educational sectors were trained. Twenty-five of them also participated in the trainer's training. Dr. Niemelä was responsible for this training program.

Between 2012 and 2013, in RD method training included all 220 workers in health and social services and 270 kindergarten teachers and assistants. Within the schools, practically all employees were trained in this industrial town, including 350 teachers, assistants, and student welfare professionals, while in the two rural communities 30 special education teachers and student welfare professionals were trained.

Population-Level Communication

The population was informed about LT-SM and LT interventions through local media and during evening meetings with parents at the schools and kindergartens. Service users received face-to-face information and leaflets.

Implementation of the Model by 2013

By the end of 2013, LT-SM infrastructure was functional and LT intervention was offered to parents at the mental health and substance use services; attention was also provided to parents with severe somatic conditions. In kindergartens, LT interventions with parents were carried out yearly, and biannually in schools.

Data and Statistical Analyses

Data regarding referrals to child welfare services from 2009 to 2016 were obtained from Sotkanet, an Indicator Bank produced by the National Institute for Health and Welfare (THL) (12). The size of the population between zero and 17 years in RD and the whole country was obtained from the PX-Web population database provided by Statistics Finland. No ethical approval was required for using combined data from open-access data sources.

With regards to time trend analyses, a joinpoint regression program and the average annual percentage change (AAPC) method (13, 14) were employed to estimate the percentage rates of the 0- to 17-year-old population referred to child protection services. The AAPC method used the underlying joinpoint regression model to calculate a summary measure above the fixed pre-specified interval. In the current study, the year 2013 was used as a cut-off year regarding the change in trends. Due to the relatively small number of referrals causing a high variability in the rates of the 0 to 17-year-old population referred to child protection services in RD compared to those of the whole country, a 3-year moving average (3-year moving average at the end of time periods) was used for the statistical modeling.

RESULTS

Figure 1 shows the trend in percentage rates of the 0 to 17-year-old population referred to child protection services in RD and in the whole country. The overall test for parallelism revealed a statistically significant difference in trend rates between RD and the whole country ($p = 0.009$). From year 2013 onwards, the rate of the 0 to 17-year-old population referred to child protection services showed a significant decrease in RD (AAPC = −6.9; 95% CI: −10.8,−2.8; $p = 0.013$) compared to an increasing trend in the whole country (AAPC = 1.9; 95% CI: 0.6, 3.3; $p = 0.020$) (AAPC difference = 8.9; 95% CI: 6.2, 11.5; $p < 0.001$). In absolute numbers, during the year 2016, a total of 359 children in RD were referred to child protection services compared to 451 children in 2012, showing a decrease of 25%.

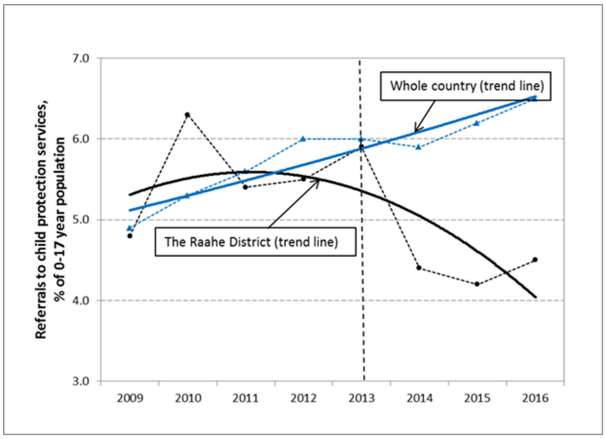

FIGURE 1 | The time trends (solid lines) and the observed percentage rates (dotted lines) of referrals to child protection services among the 0 to 17-year-old population in Raahe District and the whole country. The vertical dotted line indicates the year 2013 from which the Let's Talk about Children Service Model was in the routine use in the services of RD.

Before the year 2013, the rates had significantly increased in the whole country (AAPC = 4.1; 95% CI: 3.1, 5.2; $p < 0.001$), while no noticeable change was observed in RD (AAPC = −1.5; 95% CI: −4.6, 1.8; $p = 0.373$) (AAPC difference = 5.6; 95% CI: 2.2, 8.9; $p = 0.001$).

DISCUSSION

Let's Talk about Children Model was implemented in RD from 2012 to 2013. We analyzed referrals to child protection services based on the municipal register data and compared them with the national register data from 2009 to 2016. Referrals in RD decreased significantly between 2013 and 2016, while the national data showed a significant increase. We will first discuss the results and then the LT-SM.

The significant decrease in referrals in RD with the launch of LT-SM and the clear departure from the national trend suggest an association between the two. However, possible favorable contextual changes affecting child and family population might also have an impact on this outcome. RD has been an area with high unemployment. However, rather than a decrease, between 2012 and 2016 in RD, there was an increase in unemployment in the general population from 10 to 13%, and among individuals between 18 and 24 years from 14 to 20% (15). Unemployment is known to be a family stressor and related to use of child protection services (16). This was not reflected in the data, which may support the possible preventive impact of LT-SM. Furthermore, there were no other major concurrent projects targeted on families; however, as this was not a randomized controlled study, we cannot make definite causal conclusions. RD will continue to be evaluated to gain a more comprehensive picture in the future.

The decrease in referrals to child protection services suggests that families were getting the needed support before the family situation reached a breaking point. One Contact Service is likely to be crucial for convening LT Network meetings without delay. This might also explain why there was no early increase in the referrals as we expected.

We further hypothesized that the process would take years to yield results, but this was not the case. There are several possible reasons for this, including the expeditious implementation. The leaders of RD were deeply motivated to implement LT-SM and decisions could be made without delay. Training trainers also contributed to the rapid implementation and access to practical work with families.

LT Service Model

LT-SM and its implementation present a novel approach regarding prevention, promotion, and service development. Rather than focusing on individual impact, it aims at the collective impact on population-level outcomes (4). Key aspects of the model are discussed as follows.

First, a commitment from both administrative and political leaders of the model is crucial. The implementation of LT-SM touches all corners of the service system and, through schools and kindergartens, the whole child population and their parents. The role of the Multiagent Management Group is important, which is a decision-making body including leaders from different service sectors. Problems are discussed and successes achieved around the same table.

The core of the Model is the agreement to promote children's well-being in everyday life in all developmental contexts. It is a goal that makes sense and can be scientifically argued and expressed in common language. It is the smallest common denominator for the fragmented services.

It is noteworthy to mention that the starting point for the process is the provision of practical support for children and families. Rather than waiting for the collaborative infrastructure to be developed before the corresponding work with families begins, LT-SM is built up through interaction with the experiences and needs identified in LT interventions with families. The work with families informs the change process and is its generator.

In conclusion, LT-SM recognizes that child and family well-being involves the whole ecological context. Our results are promising, but they are just a starting point from one district. Nevertheless, they encourage the implementation of the model in different types of municipalities and demand research on implementation strategies and impact mechanisms with long-term evaluation.

AUTHOR CONTRIBUTIONS

MN and TS developed the LT-Model and the implementation strategy. MN was responsible for taking the Model into practice and consulting and guiding the implementation process. TS was responsible for negotiating with the Ministries of Health and Social Affairs and Education concerning the needed approvals for the Model and writing up the manuscript with contributions from MN and SR. BA-A and JJ contributed to implementing the Model especially in the child protection and family social services. HK, was the motivational force for the implementation of the Model in Raahe District and instrumental for getting the political support. SR consulted implementation process especially relating to mental health services and made valuable contributions to the manuscript. HH had responsibility of statistical analyses and writing up the data analysis and results section of the manuscript. TR, on the impact of parental illness on children, provided important guidelines for implementing the Model in different services. All authors have approved the final version of the manuscript 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.

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Mothers With Postpartum Psychiatric Disorders: Proposal for an Adapted Method to Assess Maternal Sensitivity in Interaction With the Child

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About 15% of mothers suffer from postpartum psychiatric disorders, such as depression, anxiety, or psychosis. Numerous studies have shown maternal caregiving behavior to be negatively affected under these circumstances. The current study sets out to shed light on specific caregiving behaviors of affected mothers in the context of parental mental illness at an early stage. There are several methods to assess maternal caregiving behavior in terms of sensitivity. However, all of them have limitations regarding the peculiarities of mothers with postpartum disorders, that is, changes in affect regulation, and the early onset of the disorder postpartum. With the current study, we provide an adapted method to assess maternal sensitivity based on methods recently approved in attachment research. Two groups of mothers, who were either healthy or had different postpartum disorders, were recorded on video during interactions with their infants. Behaviors were rated regarding responsiveness, promptness, appropriateness, intrusiveness, and positive and negative affect. A first analysis revealed an increased number of deficits on all subscales in mothers with postpartum psychiatric disorders as compared to healthy mothers. Depressive mothers with a single diagnosis had lower scores in responsiveness, promptness, and appropriateness and higher scores on intrusiveness as compared to those in healthy mothers. Here, maternal behavior appears more parent-centered, whereas affect seemed to be relatively unharmed. Moreover, as compared to healthy mothers, mothers with comorbid depression and anxiety symptomatology achieved lower scores on responsiveness, appropriateness, and positive affect and higher scores on intrusiveness and negative affect. It is suggested, that increased deficits are related to the severity of illness in mothers with comorbidities. Results on promptness indicate that these mothers are still capable of maintaining higher vigilance to infant cues. Variance in maternal behavior was relatively high in clinical mothers, showing that some of them are well capable of behaving in a sensitive manner toward their child. One strength of our adapted method is that particular aspects of sensitive parent–child interactions are assessed separately. This may shed light on specific behavior patterns of different postpartum psychiatric disorders, which may in turn relate to specific child outcomes. The manual is open for usage, while reliability testing is required.

Keywords: maternal sensitivity, postpartum depression (PPD), postpartum anxiety, mother–child interaction, AINSWORTH

THEORETICAL BACKGROUND

Postpartum Mood Swings and Psychiatric Disorders

The birth of a child is typically represented as a fascinating and happy event. In reality though, a few days after child birth, about 40–80% of mothers experience symptoms of depression and emotional lability (1–3). This phenomenon, commonly known as "baby blues," usually declines after a couple of days without psychiatric treatment. Tremendous changes in hormonal levels and psychological difficulties in adapting to the new situation after childbirth are supposed to be responsible for this emotional status (4–7). However, when these symptoms manifest in a mental disorder during pregnancy or postpartum, as it does in 10–15%, treatment is mandatory (8, 9). Maternal risk factors are a history of (familial) psychiatric disorders, sociodemographic variables, such as early motherhood, low income, low educational level, and low social support, as well as experiences of maltreatment during childhood or later life (10–13).

The most common and significant diagnoses of postpartum psychiatric disorders are postpartum depression (14), followed by postpartum anxiety (15, 16), postpartum obsessive–compulsive disorder (OCD) (17), and postpartum psychosis (18). Additional compulsive infant-focused thoughts are common in perinatal mental illness (19). Postpartum diagnoses often appear comorbid [e.g., Refs. (20, 21). In sum, they are comparable to disorders outside the postpartum period that refer to alterations in affect and cognition. However, additional peculiarities are that fears, obsessive thoughts, and sense of guilt focus on the child [e.g., Refs. (22–24)].

Maternal Sensitivity in Mentally Ill Mothers

Various symptoms with postpartum onset interfere with the demands of caring for a child and handling motherhood. Therefore, mothers with postpartum disorders often exhibit specific behavioral restraints during interaction with their infant. More specifically, postnatally depressed mothers, as compared to healthy mothers, show fewer positive caregiving behaviors, less emotional involvement, and less responsiveness when interacting with their children (25). In contrast, anxious mothers show more exaggerated behaviors and increased arousal when interacting with their children (26). Some mothers suffering from compulsive thoughts in terms of harming their child tend to withdraw from interactions as they are afraid to hurt their child (27, 28). In postpartum psychosis, Hornstein and colleagues (29) describe affected mothers as quickly overstrained, emotionally tense, uncoordinated, and sometimes awkwardly moving their infant.

In sum, postpartum psychiatric disorders compromise a mother's ability to interact with her child in a sensitive way, which decreases the quality of the mother–child interaction [e.g., Refs. (25, 26, 30)]. Sensitive parenting behavior means to consider the child's perspective, adequately infer and respond to emotional needs, and provoke positive reciprocity during interaction. Sensitivity itself is defined by the *perception*, *correct interpretation*, and a *prompt* and *adequate* reaction to a child's signals and communication attempts (31). There is evidence that sensitivity is affected in mothers with postpartum disorders. As prior work suggests, there seem to be symptom-specific patterns regarding alterations on different subscales of sensitivity, which are described below.

Responsiveness. The perception and reaction toward the child's signal was found to be lowered and less contingent in mothers with postpartum depression (25, 32–35). Also, maternal trait anxiety reduces responsiveness in healthy mothers (36, 37), whereas, in contrast, clinically anxious mothers show some overactivity (38).

Appropriateness of maternal behavior can be observed if an interaction sequence is "well-rounded" by being supportive and child-centered in an adequate way. Such supportive behavior is reduced in mothers with depressive symptoms (33). Anxious mothers behave less appropriately (39) and are more open to misinterpretations of children's cries, which may then lead to inappropriate reactions (40). Also, repeating sorrowful thoughts can lead to lower structuring during mother–child interactions (26, 41–43). For postpartum psychosis, results regarding the appropriateness of maternal behavior are ambiguous. A recent review postulates that mothers with postpartum psychosis are as likely as postnatally depressed mothers to harm their children (44), whereas Riordan et al. (45) reported observations of greater interaction deficits than those with affective disorders, namely being more remote, insensitive, intrusive and self-absorbed.

Another important component closely related to the concept of sensitivity is *intrusiveness*. Higher scores on this scale are used when the caregiver denies the child's autonomy and acts in a self-centered controlling way. On the other hand, a nonintrusive caregiver considers the child's perspective an important part of its individuality and respects it. Mothers with postpartum depression were found to exhibit behaviors of intrusiveness and withdrawal inconsistently [e.g., Refs. (16, 25)]. Highly anxious mothers show more intrusive behaviors compared to mothers with low anxiety levels [e.g., Refs. (36, 37, 43)] as less support of the infants' autonomy (46). Similarly, at an older age, teenagers with parents suffering from OCD report to be confronted with more controlling behavior and boundaries from their parents (47). Finally, reactions of psychotic mothers toward their children are assumed to be more intense in their intrusive behavior (48).

Several methods used to assess parental interactional behavior emphasize the importance of warmth and positive climate, although these aspects are not included in the original definition of sensitivity. Key aspects of depression are depressed mood and loss of interest. Thus, mothers with this diagnosis often experience blunted emotionality or an increase in negative affect. These emotional characteristics compromise the affective climate during mother–child interactions. More precisely, depression lowers the mothers' capability to flexibly switch between affective states and adjust to their child's altering affective states and needs sensitively (49, 50). Also, increases in hostility and negativity could even be observed in mothers who only exhibit subclinical levels of depression (34, 51). Finally, depressed mothers are described as less warm, more irritable, and lack joy in interacting with their child (35, 52).

Negative affect in mothers with postpartum anxiety disorder reveals itself in stressful and sorrowful behavior (53), increased criticism, and less satisfaction regarding the child's behavior (54, 55). Some studies also describe mothers with postpartum anxiety disorders as less warm and positive in contact with their children, indicated by fewer smiles or positive gestures (46, 54, 56, 57). However, other studies suggest that maternal anxiety may also yield more positive interactions (26) or that maternal affect is at least comparable to healthy mothers (58–60).

The relevance of maternal sensitivity is evidenced by numerous studies that identify this domain of caregiving behavior as an important predictor of the child's attachment (61–66). Indeed, the quality of attachment is highly sensitive to the appropriateness of early experiences (67, 68). In this line, insecure attachment strategies can be viewed as adaptions to less emotionally available or reliable caregivers, which means that they are functional in a given environment (69). Secure attachment relationships, however, provide the child with a range of competencies and have a positive impact on its social and language development (66, 70–72). With regard to maternal depression, a lack of sensitivity can yield insecure attachment, whereas maternal sensitive behavior can buffer negative effects of depression and increase the likelihood of establishing a secure attachment relationship (73, 74). Interventions focusing on improving maternal sensitivity have the potential to increase the offspring's attachment security (75). However, specific associations between certain aspects of sensitivity and child outcome variables are still unknown, which is why more fine-grained analyses on a subscale level are needed. Finally, this may be especially important with regard to behavioral impairments exhibited by mentally ill mothers.

Previous Measures of Maternal Sensitivity

Observational tools to assess maternal behavior in mother–child interactions are numerous (76–79). Most combine scales measuring sensitivity with other observable behaviors like affect or even play and generate an overall score. In the following, we describe the four most influential methods that a) assess parenting behavior in terms of maternal sensitivity and b) have successfully been used to assess predictors of children's attachment security.

First, the Ainsworth Maternal Sensitivity Scale is one of four scales assessing parental behavior (31): Sensitivity versus Insensitivity to the baby's signals, Cooperation versus Interference with baby's ongoing behavior, Physical and Psychological availability versus Ignoring and Neglecting, and Acceptance versus Rejection of the baby's needs. Here, the scale sensitivity turned out to be a key variable because it was found to be correlated to acceptance, cooperation, and availability and to be a reliable predictor of attachment security (61). Affect is considered in the Acceptance versus Rejection Scale because it deals with the balance between the mother's positive and negative feelings about the baby. The Ainsworth Maternal Sensitivity Scale requires the observer to make one global assessment of sensitivity. In further analyses, it has been related to attachment quality and a range of child behavior. It is the origin of other developing measurement tools like the NICHD–Study of Early Child Care Mother–Child Interaction (ECCN) Scale.

Second, the NICHD-ECCN Scale for mothers of children from 6 up to 24 months quantifies different aspects of maternal behavior rated from videotaped dyadic semistructured play interactions. It assesses the following aspects of parenting behavior: *Sensitivity/Responsivity to distress* (adapted from the original Ainsworth Maternal Sensitivity Scale), *Sensitivity/Responsivity to nondistress*, *Intrusiveness, Detachment*, *Stimulation of development, Positive regard for the child, Negative regard for the child,* and *Flatness of affect* (80). Even though there are two separate global rating scales measuring maternal sensitivity/responsivity, most studies use a composite measure of overall maternal sensitivity as an indicator of mothers' sensitive behavior [e.g., Ref. (81)]. The NICHD-ECCN sensitivity scales are one of eight widely used observational measures; however, there are indeed a total of 50 measures related to the construct of maternal sensitivity (79). A number of studies have shown that maternal sensitivity measured by the NICHD-ECCN Scale is predictive of different child outcomes, for example, attachment security (82, 83) or cognitive competence (84).

Third, Biringen and colleagues (85–87) developed the emotional availability (EA) Scales. The EA Scales describe the caregiver's ability to perceive and react to both positive and negative emotions, as well as the child's reaction to the caregiver's behavior. It includes a multidimensional set of features (e.g., caregiver's sensitivity, nonhostility, structuring, nonintrusiveness). Moreover, the child's behavior is observed, too. This leads to the evaluation of the affective quality of parent–child relationships from the first weeks of life to adolescence. There is no given structure for the observational situation, and it has been applied in separation–reunion situations (62), still face procedure (88), and structured and semistructured play situations (89, 90).

Fourth, the CARE-Index is a screening tool intended to enable trained professionals to make judgments about the necessity of a family intervention (91). Similar to the EA Scales, the method evaluates the quality of caregiver–infant interaction.

Observational situations are 3 min of play interaction recorded on video under nonthreatening conditions. It is applicable from birth to 15 months and up to 2.5 years when using the toddler form. The coding procedure focuses the observer's attention on seven aspects of the caregiver's and the infant's behaviors, including affect and cognition. For caregivers, these are sensitivity, control, and unresponsiveness. These codes define four patterns of caregivers' interactional behavior. The "sensitive pattern" involves how the caregiver accommodates to the infant's behavior and shares the most commonalities to the concept of sensitivity. The "controlling pattern" identifies behaviors that are either overtly hostile or covertly hostile comparable to intrusiveness and hostility in other scales, whereas the "unresponsive pattern" consists of items describing forms of withdrawal.

Limitations of Existing Methods for Mentally Ill Mothers

With many other methods, but the four named as influential in attachment research, it should be just a matter of choice to evaluate maternal sensitivity in postpartum psychiatric disorders. Indeed, all of them have been in use for clinical research [e.g., Refs. (76, 79] with their strengths and weaknesses. However, to integrate sensitivity research and specific parenting characteristics with postpartum psychiatric disorders, each of the named methods has its own limitations.

First, the onset of postpartum depression starts either already during pregnancy with signs for mental health problems or manifestations of depression and anxiety or immediately after birth or at least very few weeks or months after delivery. In practice, the diagnosis is given with diagnostic criteria until children reach the age of 1, in some hospitals even 2. Here, it is important to note that sensitivity measures vary regarding recommendations on the child's age range: Whereas the scales of Ainsworth, EA, and CARE-Index allow observation from birth on, the NICHD Scale starts from 6 months of age and was not specifically intentionally developed for newborns or younger infants. Moreover, the CARE-Index and the EA Scales measure dyadic interaction, which increases in older infants. This might account for the number of null findings in studies using the EA Scales in depressed and nondepressed mothers (86).

Second, the development from and application with regard to attachment theory and research are different. The CARE-Index and the EA Scales can be regarded as further developments of the Ainsworth sensitivity scales and are both very promising in evaluating the dyadic interaction within the developing attachment relationship because sensitive maternal behavior is related to secure attachment. According to Bowlby (92), attachment to the primary caregiver develops not earlier than after 8 weeks, with increasing differentiation in the second half of the infant's first year and attachment measures are applied with 12–18 months. Because maternal postpartum psychiatric disorders occur within the infant's first year of life, with an onset immediately until 4 weeks after birth, observational measures of attachment security need to be assessed at follow-up. The Ainsworth sensitivity and the NICHD-ECCN scales have been shown to be good predictors for measures of attachment security. Moreover, the latter two are freely available and open to use for everyone. For research, reliability testing is mandatory. Training to learn the manual of the CARE-Index takes about 8 days plus practice and reliability testing. Also, the EA Scales require 1 week with additional training by Biringen, too. Whereas the Ainsworth sensitivity and the NICHD scales are free for usage, the other two are subject to charges.

Another limitation is that the NICHD ECCN sample mostly stems from the middle class. For samples who are at specific risk to show highly dysfunctional parenting behaviors, however, it is necessary to use scales that particularly differentiate behavioral nuances at low levels of parenting competence and that allow to measure distinct impairments of specific aspects of parenting behaviors.

Furthermore, all methods vary with regard to the observational situation. Whereas the Ainsworth Maternal Sensitivity Scale and the EA Scales can be applied in different settings, the CARE-Index favors free-play situations, and the NICHD-ECCN Scale requires semistructured situations like diaper change or feeding situation or in the toddler form a manualized procedure. Also, the required recording length ranges from 3 min (CARE-Index) to 30 min (EA Scales). Regarding the special group of mentally ill mothers, the observational situation must be carefully chosen. It should be easy to implement during daily routine of several treatment institutions. Because those mothers are limited in their capacity to play freely with their children, it must not enforce the feeling of insufficiency.

Next, a crucial diagnostic criterion of postpartum depression is depressed mood and loss of interest, whereas arousal is increased in mothers with postpartum anxiety. Affect measures have been considered in all scales but only to a limited degree: Hostility or warmth is measured in the Ainsworth Maternal Sensitivity and EA scales. The NICHD-ECCN sensitivity composite score includes positive and negative regard of the child for youngest children, with changes in the negative affect scale in the toddler form.

Further considerations refer to how differentiated maternal behavior is measured, which is especially important with regard to mothers' symptom comorbidity. Recent research finds that depression and anxiety during the first 8 weeks postpartum occur simultaneously in 13% of mothers (20). Symptoms of anxiety often come along with symptoms of compulsive behavior (93). Triple comorbidity of depression, anxiety, and posttraumatic stress disorder (PTSD) is relatively rare with a prevalence of 2% to 3% (94). Other comorbidities are not well studied, although they appear in clinical practice. Next to comorbid symptoms, stresses and strains as parenting or childhood stress, immigration history, and maternal vulnerable personality predict higher rates or symptoms (20). A main concern with the recent methods is that behavioral variance caused by comorbidity may not be represented sufficiently because all ratings use sum scores or general ratings. For instance, a mother with depression can partly be responsive and adequate but not prompt, whereas a psychotic mother might act responsive, prompt but not adequate. By using a sum scale of sensitivity, both mothers would receive low scores, but how can one differentiate behavior between these mothers regarding sensitivity? And how can such differences then be related to later child outcome? With the future goal, that is to describe the mother–child interaction in postpartum disorders with the full spectrum of comorbidities and risk factors, a separation of the concept of sensitivity into its original components described by Ainsworth is valuable.

To sum up, we propose an adaptation of the described methods to measure sensitivity to fully represent the spectrum of altered behavior in postpartum psychiatric disorders with all its comorbidities. Our intention was to develop a method that is applicable in a clinical routine setting (e.g., mother–baby unit) with reasonable effort for patients and the therapeutic team. At the same time, it should be usable for scientific purposes. Future scientific questions can then focus on how maternal caregiving behavior differs with regard to different diagnoses while taking into account the high number of comorbidities. It is also of special interest if other risk factors associated with postpartum psychiatric disorders are related to specific deficits, for example, parenting stress, traumatic experiences, or lack of social support. And finally, we aim to relate our findings to specific child outcomes to increase our understanding on symptom-specific effects of alterations in parenting behavior that are related to maternal mental illness.

The present paper describes the development of an adapted measure of maternal caregiving behavior for scientific and clinical use in mothers with postpartum psychiatric disorder. The adapted measure specifically considers the clinical setting, peculiarities of postpartum disorders, and a fine-grained description of maternal behavior. We then present a preliminary pilot implementing the adapted measure on 38 mothers admitted to a mother–baby unit and 35 healthy mothers.

METHOD

Ethics

This study was carried out in accordance with the Declaration of Helsinki and the permission and recommendations of the Ethics Committee of the Friedrich-Alexander University Erlangen Nuremberg and the Ethics Committee of Technical University Dresden (Ethics committee of the FA: 320_15 B, Ethics committee of the TU: EK450 22013). All participants gave written and informed consent. For all data concerning children, written informed consent was obtained from the parents.

The Pilot Project: Video Screening for the Development of the Adapted Sensitivity Scale

Sample

Mentally ill mothers currently attending a video-based interaction therapy in a mother–baby day unit were asked to participate in the study. Video recordings were made on a voluntary basis. Treatment institutions were the Mother-Baby-Day-Unit of the Clinic of Psychiatry and Psychotherapy of the Paracelsus University Clinic Nuremberg Psychiatry in Nuremberg and the Mother-Baby-Day-Unit of the Clinic for Psychotherapy and Psychosomatics of the University Clinic Carl Gustav Carus at the Technical University in Dresden. In these units, mothers spent time from 8 to 16 o'clock each day for an average of 8 weeks, during which they receive a variety of interventions (e.g., group therapy, psychoeducation, personal counseling, and art therapy). A special focus lies in the improvement of mother–child interaction by providing sensitivity training, baby massage, and mother–baby bonding therapy. Video interaction therapy starts soon after the initial assessment with numerous sessions during the whole stay in treatment. In these sessions, mothers watch videotapes of themselves interacting with their infant and get feedback about positive sequences from their therapist. They are then supported by their therapist to enhance such behaviors and integrate them into daily life situations.

The total sample of mothers who agreed to participate in the current study consisted of 102 mothers with postpartum disorders (67 stemming from the Dresden Unit and 35 from the Nuremberg Unit) and of 38 healthy/nonclinical mothers. For mentally ill mothers, the diagnoses were depression, anxiety, compulsive disorder, PTSD, substance abuse {of medium extent [alcohol, tetrahydrocannabinol (THC), or nicotine during pregnancy and continuing]}, bipolar disorder, personality pathology, and/or psychosis (order according to frequency). More than half of the patients had more than one diagnosis.

Clincial diagnoses were based on clinical assessment according to International Statistical Classification of Diseases and Related Health Problems (ICD-10) criteria, video recordings were made by an interaction therapist, and questionnaires were assessed as self-ratings.

Observation of Mother–Child Interaction

Observation and analysis of mother–child interaction were used for the development of the adapted sensitivity scales. Videos were recorded in clinical routine for therapeutic use at the beginning of their therapy. Mothers were instructed to act as normal as during daily routine.

Description of the Adapted Sensitivity Scales for Observation

The method is based on the Ainsworth Maternal Sensitivity Scale (31) and the NICHD-ECCN Scale (80, 84, 95) and was adapted to the survey situation, children's age, and peculiarities of interaction behavior in mothers with postpartum psychiatric disorders. Analysis of video-recorded interactions was done according to the manual attached (see **Supplementary Material**).

With respect to behavioral deficits in mentally ill mothers (as described above), the following scales were defined: caregiver's ***responsiveness, promptness*** of reaction, ***appropriateness*** of the caregiver's reaction, ***intrusiveness*** of the caregiver, and ***negative affect*** and ***positive affect*** toward the child.

The scale ***responsiveness*** refers to whether the mother reacts to the child's signals. Here, given that the child sends a signal, overtly or subtle, the mother needs to signal back that she recognized it. For example, during diaper change, this can be the exchange of a look or comments on what is happening, when the child displays discomfort. ***Promptness*** is defined whether the response occurs immediately, not exceeding 3 s. Otherwise, the child may not relate its own action to the maternal reaction and experience himself as agent of this interaction.

The scale ***appropriateness*** measures whether the mother's reaction fulfills or at least adequately attempts to fulfill the baby's needs presented in the sequence. Generally spoken, this refers to security, warmth, and comfort. An appropriate reaction is well-rounded and children experiencing appropriate behavior are usually less stressed.

These three scales were chosen based on Ainsworth's definition of sensitivity and the NICHD descriptions, whereas both of them use an overall score for all three types of behaviors referred to as "sensitivity."

Intrusiveness measures to what extent the caregiver respects the child's autonomy and accepts its individuality. High intrusiveness indicates low respect of autonomy. Practically, this can often be observed when mothers unclothe their children in an uncomfortable but quick manner, instead of doing so with respect to the child's reaction and by providing guidance through, for example, verbalization. Mothers with postpartum psychiatric disorders have difficulties here because they often seem to follow an adult-centered script and do not seem to be able to take over the child's perspective.

Negative affect is defined as the presence of active expressions of negative affect either verbally, facial, or by hostile actions. Because infants cannot differentiate whether they are the cause for maternal bad mood or whether there are other causes, all kinds of negative affect and hostility are rated. ***Positive affect***, in contrast, is mirrored in the caregiver's joy within the interaction. These scales already existed in the NICHD-ECCN Scale and were modified with special regard to the altered affectivity in psychiatric disorders.

The method and the rating scheme are described at length in the manual at the appendix (compare **Supplementary Material**). We added examples that were found in the videos to the descriptions.

Scales and blinded videos were repeatedly discussed on lab meetings and with trained sensitivity observers. Other scales, as measuring maternal speech or body contact, were rejected during adaptation because they were not directly related to the concept of sensitivity, although many mothers also show deficits here.

In total, six scales were rated on a 9-point scale ranging from one to five with semisteps in between. Defined anchor points are 1, 2, 3, 4, and 5. Low values indicated low characteristics of the regarding scale, whereas high values go along with frequent observation of the behavior in the scale.

Coding of Mother–Child Interaction

Analyses were conducted by five postgraduate psychology students who participated in a seminar on "observational tools to assess baby's signals" and underwent intense reliability training by experienced researchers. Reliability training included reading literature on sensitivity, discussing videos with mother–child interaction and doing pre-ratings to find misperceptions or positive or negative observation bias on a weekly basis for 6 months. They were trained with the help of the manual and an assessment sheet (see the Manual) analyzing the categories: Responsivity, promptness, appropriateness, intrusiveness, and negative and positive affect. Note: Observers who are familiar and reliable with other sensitivity scales should be able to use the method with less intense training.

Raters were blind to diagnoses. All five students were required to become highly reliable, with all $\kappa > 0.85$ before they were allowed to score the videos included in the study. According to McHugh (96), this indicates that interrater reliability is strong to perfect. *Post hoc* calculated interrater reliability (weighted kappa) was based on 17 double-coded video tapes after 6 months. Here, mean reliability was moderate, with $\kappa = 0.73$. All raters showed strong reliability on the scale intrusiveness, $\kappa = 0.85$, whereas on the scale, promptness reliability was lowest and just weak, with $\kappa = 0.59$. Reliability for the other scales were moderate with responsiveness, $\kappa = 0.66$; appropriateness, $\kappa = 0.73$; negative affect; $\kappa = 0.76$; and positive affect, $\kappa = 0.80$.

First Statistical Analyses Comparing Data on Depressed and Anxious Mothers to a Healthy Group

Sample

For the present study, we examined a subset of the clinical sample including 31 mothers from the Nuremberg Unit and seven mothers from the Dresden Unit with postpartum depression and/or anxiety disorder. The clinical sample was recruited consecutively from 2016 to 2018. Infants were healthy as seen by Pediatrics from the hospital. There was a massive reduction of usable videos because during the pilot phase, criteria for the recordings of the videos for scientific usage have been developed.

Videos were excluded if they were <3 min in length (according to the standard CARE-Index procedure), recorded after the third week of admission, the patient presented with three clinical diagnoses on the basis that the underlying disorder was unclear (e.g., depression and anxiety and personality disorder and substance abuse), invisibility of important parts of the interaction, strong sound problems, or lack of questionnaire data.

In contrast, inclusion criteria were a good quality of the video, recording at the beginning of the therapy, and at least one of the diagnoses from the spectrum of depression, anxiety, or compulsive disorder.

The control group was acquired in the area of Erlangen Nuremberg, Germany, and videos were recorded during home visits. The control mothers were recruited *via* flyer or word of mouth and agreed to participate in a home visit, including the recording of a video and assessment of questionnaires. They were paid 20€ and received a small toy for the child.

All mothers were instructed to act as usual during daily routine. All videos included a semistandardized situation, where they changed the diapers of their infant or fed him or her. In addition, parents were asked to play with their infant without instruction.

Questionnaires

Brief Symptom Inventory

The Brief Symptom Inventory (BSI) (97) is a short form of the Symptom Checklist SCL-90-R (98) and aims to assess mental stress of participants within the last 7 days. Participants respond on a 5-step Likert Scale, ranging from 0 ("not at all") to 4 (very heavy) how much they agree to a total of 53 items. The BSI includes nine scales assessing, for example, depression, anxiety, or psychotic experiences. Finally, the global score [Global Severity Index (GSI)] is calculated to assess general mental stress.

In addition, there are three global scores for general mental stress: GSI, intensity [(Positive Symptom Distress Index (PSDI)], and number of reported symptoms [positive symptom total (PST)].

Scores can be transformed into T scores with different norms for academic students or adults in general, as well as men and women. Here, we used T scores for women (nonstudent) to assess the global severity of mental illness. Clinically relevant GSIT scores are above ≥63 (99).

Edinburgh Postnatal Depression Scale

To assess postpartum symptoms of depression, we used the Edinburgh Postnatal Depression Scale (EPDS) (22). The screening instrument includes 10 items asking for the mood of the last 7 days of young mothers. Mothers had to answer on a 4-point Likert (0–3) Scale how much they agreed with each item. Scores were summed up, with 30 as the highest possible value. High values indicate strong symptoms. Mothers with scores of 13 or higher are regarded as likely to suffer from depression. Validation studies (100) reported good sensitivity (79%) and specificity (85%).

Statistics

We used SPSS 25.0 for analysis. To examine differences between group variables, we used *t*-test. Intercorrelation analysis was done by Pearson correlation. Potentially influencing factors, as age of the child or mother, were also controlled by using Pearson correlation. Group comparisons regarding sensitivity were done with multivariate analysis of variance (ANOVA). All *post hoc* pairwise comparisons were performed using LSD. Significance level followed $p \leq 05^{*}$, $p \leq 0.01^{**}$, $p \leq 0.001^{***}$. **Tables** show means and standard deviations.

RESULTS

Sample Descriptives

Mothers of both groups were at the same age in the beginning of the thirties and had the same, relatively high, educational background (**Table 1**). The sex of the children had a similar ratio in both groups. Children of mentally ill mothers were significantly younger than children of the control group. In the clinical group, the number of participants with at least one close relative (parents or grandparents) who suffered from mental disorders was twice as high as in the control group.

Psychopathology was significantly higher in the clinical group as indicated by measures of the BSI (GSI) and EPDS. In the control group most participants (27 of 35) had scores below 10, while 5 participants had an EPDS score between 10 and 12, indicating a medium degree of postpartum depressive symptoms, and 3 of them had scores above 12 (clinical relevance). In contrast, in the clinical group, almost all mothers (36 of 38) had scores above 12, whereas 2 of them scored between 10 and 12.

Group Comparisons

We used multivariate ANOVA to test if the group of healthy mothers differs in maternal sensitivity significantly from the clinical group. Although not significant ($F_{(6,66)} = 1.690$; $p = 0.137$), results indicate that the clinical group performs poorer in all subscales compared to the healthy control group (**Table 2**). Nonsignificance is typical when the dependent variables are highly correlated (**Table 4**). Further analysis underlines it: extracting promptness as a dependent factor from analysis leads to approaches of significance ($F_{(5,67)} = 2.042$; $p = 0.084$). However, univariate comparisons result in significant effects for each of the dependent measures. Healthy mothers are more responsive, more prompt, and more appropriate than mentally ill mothers. They respect more of the child's autonomy, whereas clinical mothers are more intrusive. Negative affect is lower and positive affect is higher in the healthy group than in the clinical group. Whereas the group of healthy mothers perform in the upper range of the scale (range 3.87–4.33), clinical mothers perform still in a mediocre to upper range (range 3.12–3.88).

TABLE 1 | Demographic data for healthy and clinical mothers.

	Healthy mothers (n = 35) Mean SD	Clinical mothers (n = 38)	
		Mean SD	Significant differences
Maternal age (years)	31.49 ± 4.27	30.02 ± 5.38	n.s.
Education with A Level (%)	62.85	55.26	n.s.
Age of child (weeks)	49.60 ± 25.22	27.75 ± 21.39	$t(71) = 4.002$, $p = 0.000$
Sex of child (boy:girl)	16:19	19:19	
PTs having relatives with psychiatric disorder	11	22	$X^2 (1, n = 73) = 5.152$, $n = 0.023$
EPDS (sum)	5.77 ± 3.98	17.32 ± 5.55	$t(70) = -10.097$, $p = 0.000$
BSI (GSI)	46.97 ± 12.03	74.83 ± 7.72	$t(71) = -11.763$, $p = 0.000$

Values indicate means and standard deviations. PTs, participants; EPDS [sum], sum score of the Edinburgh Postnatal Depression Scale; BSI [GSI], Brief Symptom Inventory [Global Severity Index]; n.s., not significant.

TABLE 2 | Comparison of the sensitivity subscales for healthy and clinical mothers.

	Healthy mothers Mean SD	Clinical mothers	
		Mean SD	Significant differences
Responsivity	4.16 ± 0.73	3.67 ± 0.89	$F_{(1,71)=}$ 6.431; p = 0.013
Promptness	4.01 ± 0.66	3.54 ± 0.93	$F_{(1,71)=}$ 6.214; p = 0.015
Appropriateness	3.97 ± 0.86	3.32 ± 1.00	$F_{(1,71)=}$ 8.942; p = 0.004
Intrusiveness	2.13 ± 0.83	2.88 ± 1.15	$F_{(1,71)=}$ 10.203; p = 0.002
Negative affect	1.67 ± 0.69	2.12 ± 0.86	$F_{(1,71)=}$ 5.984; p = 0.017
Positive affect	4.33 ± 0.86	3.85 ± 0.94	$F_{(1,71)=}$ 5.003; p = 0.041

Values indicate means and standard deviation. P values indicate significance from multivariate testing.

TABLE 3 | No correlation of maternal behavior with maternal or child age.

		Maternal age	Child age
	Healthy mothers		
1	Responsivity	−0.078	0.221
2	Promptness	−0.185	0.122
3	Appropriateness	−0.157	0.297
4	Intrusiveness	0.048	−0.154
5	Negative affect	−0.039	0.042
6	Positive affect	0.212	−0.045
	Mothers with PPD		
1	Responsivity	−0.074	0.137
2	Promptness	−0.043	0.084
3	Appropriateness	0.033	0.066
4	Intrusiveness	−0.100	−0.164
5	Hostility	−0.276	−0.320
6	Positive affect	0.230	0.017

Factors Influencing Maternal Sensitivity

We tested for maternal and child age as possible parameters influencing sensitivity. Spearman correlation revealed no significance with these factors for none of the scales neither in the group of healthy nor clinical mothers (compare **Table 3**). For further testing if the difference of child age between the group still affected the results, we included it as a covariate into the analysis. Here, significance was reduced to a trend level in three scales, namely, responsivity ($F_{(1,70)}$ **=** 2.820, p = 0.098), promptness ($F_{(1,70)}$ = 3.594, p = 0.062), and negative affect ($F_{(1,70)}$ = 3.816, p = 0.055).

Intercorrelations Between the Scales of Sensitivity

As visible in **Table 4**, all scales correlate significantly with each other in the healthy control group (range from r = 0.934 to r = 0.446) and the clinical group (range from r = 0.955 to r = 0.620).

Influence of Severity of Symptoms in Mental Illness on Sensitivity

One goal of the development of the adapted method is to analyze sensitivity with respect to specific symptoms of depression to better differentiate between groups of different mental disorders. In the group of healthy mothers, we found mothers who scored subclinical to moderate on depression or EPDS and/or BSI and were clinically conspicuous. We decided not to exclude them from earlier analysis because they were "healthy enough" not to be in treatment. However, in an additional analysis, we put them in a separate group, named "healthy with mild depressive symptoms" (n = 8). Moreover, we separated the group of clinical mothers. Whereas 16 mothers were diagnosed with depression only, 19 mothers had depression and anxiety disorder or compulsive thoughts comorbid. We excluded three mothers because their comorbid diagnosis appeared to raise a personality disorder. The healthy group without depressive symptoms consists of 27 mothers. It is obvious that, with increasing symptoms, EPDS and BSI scores rise (**Table 5**).

We used multivariate analysis to test whether the sensitivity scales differ significantly between the four groups (healthy vs. mild symptoms vs. clinical with depression vs. clinical with depression and comorbidities). Not significantly, but descriptively, with increasing severity, sensitivity gets worse. Healthy but mildly depressed mothers have lower values in sensitivity but not in their affect. Clinically burdened mothers differ significantly in their sensitivity pattern from that of healthy mothers. Whereas mothers with depression respond less, are less prompt, less appropriate, and more intrusive, their affect does not differ significantly from that of healthy mothers. Mothers with additional diagnoses of anxiety are less responsive, less appropriate, and more intrusive and show more negative and less positive affect but are not significantly less prompt.

TABLE 4 | Intercorrelations of maternal sensitivity in the control group (n = 35) (upper values, gray background) and the clinical group (n = 38) (lower values).

Clinical group		Healthy control group					
		1	2	3	4	5	6
1	Responsivity	1	0.934***	0.883***	−0.661***	−0.529***	0.506**
2	Promptness	0.955***	1	0.835***	−0.585***	−0.478**	0.460**
3	Appropriateness	0.826***	0.846***	1	−0.753***	−0.592***	0.594***
4	Intrusiveness	−0.664***	−0.620***	−0.824***	1	0.493**	−0.446**
5	Negative affect	−0.758***	−0.707***	−0.760***	0.742***	1	−0.837***
6	Positive affect	0.694***	0.659***	0.756***	−0.715***	−0.913***	1

*Asterisks label significance (**p ≤ 0.005, ***p ≤ 0.001).*

TABLE 5 | Comparisons of the sensitivity subscales for groups with different degrees of depressive symptoms, ranging from no symptoms to comorbidities.

	Healthy mothers			**Clinical mothers**			
	No depression *n* **= 27**	**Mild depression**		**Depression**		**Depression + anxiety**	
		***n* = 8**	***p***	***N* = 16**	***p***	***N* = 19**	***p***
EPDS	3.96 ± 2.31	11.87 ± 1.35	0.000	14.87 ± 5.43	0.000	18.61 ± 5.03	0.000
BSI GSI	43.03 ± 7.83	61.50 ± 12.75	0.013	72.81 ± 7.47	0.000	76.89 ± 3.98	0.000
Responsivity	4.24 ± 0.69	3.87 ± 0.79	n.s.	3.65 ± 0.97	0.029	3.73 ± 0.88	0.047
Promptness	4.05 ± 0.62	3.87 ± 0.79	n.s.	3.53 ± 1.04	0.050	3.57 ± 0.91	n.s.
Appropriateness	4.07 ± 0.80	3.62 ± 0.99	n.s.	3.21 ± 1.04	0.006	3.42 ± 1.03	0.025
Intrusiveness	2.02 ± 0.82	2.50 ± 0.75	n.s.	2.88 ± 1.27	0.008	2.79 ± 1.04	0.012
Negative affect	1.67 ± 0.65	1.69 ± 0.84	n.s.	2.00 ± 0.98	n.s.	2.16 ± 0.74	0.048
Positive affect	4.33 ± 0.82	4.31 ± 1.03	n.s.	4.00 ± 1.01	n.s.	3.78 ± 0.85	0.040

Mean and standard deviation. p values belong always to comparisons from the group left to them with healthy mothers without depression.
n.s., not significant. See ref. (24).
EPDS, Edinburgh Postnatal Depression Scale; BSI GSI, brief symptom inventory, global severity index.

DISCUSSION

General Discussion

The present study aimed at adapting observational methods assessing sensitivity in caregivers to a group of mothers with postpartum mental disorders. Therefore, we screened 102 videos of mentally ill mothers interacting with their infants to identify specific parenting behaviors associated with different postpartum mental disorders. These specific behaviors are described in the manual, which can be found in the supplements. With respect to the main deficits of maternal caregiving behavior, we combined different measurement scales for sensitivity. More precisely, with special regard to attachment research, the Ainsworth's Maternal Sensitivity Scale (31) and the NICHD Scale were chosen as a basis for the present method while integrating aspects taken from the EA Scales, the CARE-Index, and from a range of other observational tools focusing on parental behavior (76, 79). This procedure resulted in the examination of responsiveness, promptness, appropriateness, intrusiveness, negative affect, and positive affect. The compilation of these final scales is adapted to the special group of mothers with postpartum disorders because it considers peculiarities regarding the onset of illness, child age, and disorder-related behavior.

In a second step, we used the adapted scales for a preliminary analysis aiming to investigate whether the method is applicable in a clinical group. Therefore, we compared caregiving behavior in a group of mothers at the beginning of treatment in a psychiatric mother–baby unit to that of a group of healthy mothers. Results revealed that mentally ill mothers have difficulties in all aspects of sensitivity, intrusiveness, and affect. Mothers with postpartum psychiatric disorders were found to be less responsive, respond slower, less adequate and to be more intrusive and show more negative and less positive affect toward their children. These findings are not surprising because it has been repeatedly described that postpartum depression can result in tremendous problems regarding mother–child interactions [e.g., Refs. (25, 26, 30, 55, 98)].

A second aim was to see whether assessing different aspects of sensitivity separately would help identify typical behavior patterns of mothers with mental disorders dependent on their diagnosis or other risk factors for altered mother–child interaction. Here, we found that the severity of symptoms is an important factor influencing maternal sensitivity in the present sample. Even in the healthy group, we observed that mild depressive symptoms already reduce sensitivity, however, the effect was rather small. This is in line with recent research showing that subclinical depressive symptoms may increase hostility and negative affect (51) and reduce maternal sensitivity (101). Moreover, we separated the clinical group into those with depression only and those who have an additional diagnosis for anxiety disorder because postpartum depression and postpartum anxiety are frequently comorbid (102, 103). Comparing results to the healthy control group revealed deficits in both groups of mothers with mental illness, but we found different behavioral patterns: In contrast to healthy mothers, those with depression only scored significantly lower on all scales of sensitivity (responsiveness, promptness, appropriateness) and intrusiveness; they did not differ significantly on the affect scales, although values are lower than those in the healthy group. In comparison, mothers with comorbidities of depression and anxiety scored, apart from promptness, significantly lower on all subscales, indicating general deficits in mother–child interaction including own emotion regulation.

The difficulties with sensitivity and intrusiveness in depressive-only mothers have repeatedly been found in earlier studies. Responsiveness, the perception and reaction to a child's signal, has repeatedly been shown to be lower in postpartum depression (25, 32–35). Moreover, maternal responses are less contingent (25) and less appropriate (33). Intrusiveness and withdrawal are reported to appear inconsistently (16, 25), whereas in our study, depressed mothers generally tend to show less respect for child autonomy. Without the investigation of a behavioral pattern, the lack of responsiveness alone may lead to the assumption that signals are not perceived, likely as a result from a cognitive deficit. In combination with measures of lower appropriateness and higher intrusiveness, it can be assumed that mothers ignore their children purposely, followed by a parent-centered response inappropriate to the child's needs. The underlying cognitive process behind this behavior remains unmeasurable, but it seems as if intuitive behavior is disturbed in depressed mothers (97) and that they rather follow a script or external recommendations during interaction. Also, depression worsens the ability to manage changes between affective states, and it is more difficult to adjust to their child's affective states and needs sensitively (49, 50). Interestingly, in our study, maternal depression did not significantly alter affect during mother–child interactions in mothers with depression only, although depressive mood is central in depression. In contrast, Cohn and colleagues (104) argue that negative affect is increased four times in depressed mothers. Also, prior work found mothers with former or postpartum depression to show more negative behaviors as criticizing or destructive reactions and fewer positive behaviors, such as praise or constructive guidance during interaction sequences (105, 106). One reason why we did not find this could be that, in the present sample, the variance in the affect scale is relatively increased in clinical mothers as compared to healthy mothers. This indicates that some of them are well capable of positive interaction with their infants, whereas others are not. Laucht et al. (107) suggest, that, because of some well-functioning, self-control mechanisms, many depressive mothers are capable of acting as nondepressed in contact with their children, warranting a positive child development (107).

As general explanation, the disturbances in altered mother–child interaction are caused or accompanied by altered brain activation. Mothers with postpartum depression have shown weaker activation of reward and motivation areas, such as the thalamus, nucleus accumbens, caudate, and key emotion regulation areas, such as the lateral orbitofrontal cortex (108). Lower responsiveness may be the result when motivation is lowered to increase contentment and infant stimuli do not feel as rewarding for some of them as for healthy mothers.

In our subsample, mothers with depression and anxiety comorbid presented another behavioral pattern. This group showed deficits in all scales, except promptness, which may represent the increase of impairment caused by the double diagnosis.

Earlier findings on maternal anxiety describe mothers as less responsive and withdrawing from interaction, also in trait anxiety of healthy mothers after birth [e.g., Refs. (36, 37)]. More precisely, anxious mothers behave less appropriate (39) and are more exaggerating or overprotective (26, 38). This exaggeration or overprotection, probably caused by the typical higher arousal in anxiety, is mirrored in the "promptness" scale. In our study, we found mothers with comorbid anxiety to have somewhat lowered scores on this scale; however, they did not significantly differ from those of the healthy group. In the face of comorbid anxiety, promptness may be a sign of higher alertness to threatening keys (such as unfamiliar baby signals combined with low maternal self-efficacy). In our, as well as in recent, studies, highly anxious mothers present more intrusive behaviors compared to mothers with low anxiety levels [e.g., Refs. (36, 37, 43)] and support less autonomy (46). They interfere with and try to control the child's behavior (37, 38, 41), often with the objective to be overprotective (53, 109).

The finding that responsiveness, appropriateness, and intrusiveness are worsened in comorbidly ill mothers may reflect a commonality with depressed mothers' behavior, namely, that they act more parent-centered and less intuitive. The constant alertness reflected in the promptness scale could give a hint to their inner conflict between "waiting for signals" and "not being able to react flexible." This conflict is also visible on the affect scale because they show less cheerful play and more negative affect mimically and verbally, more distressed behavior, or emotional flattening. Negative affect in maternal anxiety has been observed through stress and sorrowful behavior (53), increased criticism, and less satisfaction of the child's behavior (54, 55). In our study, we observed lowering of positive affect in depressed and anxious mothers in contrast to some other studies (26, 58–60). Our finding is in line with those studies describing depressed and anxious mothers as generally less warm and positive in contact with their children, indicated by less smiling and positive gestures (46, 54, 56, 57). The lack of joy in the postpartum phase might also have its cause in altered brain processes, namely, that higher levels of anxiety in relation to parenting correlated with reduced activation in the substantia nigra, a region implicated in reward prediction and processing (110). When a child's smile feels less rewarding, experience of joy is probably reduced, too. Consequently, intentions to make the child laugh may be lacking.

In sum, the presented scales successfully identify specific deficits in mother–child interaction of mothers with postpartum psychiatric disorders. It further shows that these deficits differ depending on the diagnosis or comorbidity of mothers and thus provide simplifications for tailored therapeutic intervention. Although for depressed-only mothers, a training on improving appropriate reactions and on understanding of infant's signals and needs might be sufficient, mothers with depression and anxiety comorbidity may need further support in their own emotion regulation. Our method is very detailed, but it is still closely related to the concept of sensitivity. Therefore, it is suited for the investigation of maternal behavior as a predictor of later attachment security.

Limitations

Results of our study are based on a group of healthy mothers and a subgroup of mothers admitted in a psychiatric/psychosomatic Mother-Baby-Day-Unit Nuremberg or Dresden. Healthy and nonhealthy mothers did not differ in their educational degree or age. One limitation was that the children of clinical mothers were younger than those of the control group because of a change of admission requirements in the hospital while the study was still ongoing. However, there was no correlation of the scales with child's age.

Behavioral assessment was located differently for clinical and healthy mothers that may have caused different levels of stress or comfort during observation.

Another limitation of the study is the rather small sample size in the group with comorbidities with respect to the fact that they are diagnosed with different and/or several anxiety disorders. Anxiety disorders manifest in a wide range: generalized anxiety disorder, OCD, panic disorder, and birth-related PTSD. In the postpartum period, sometimes the severity and symptoms do not rise to the level of an anxiety disorder diagnosis (e.g., hypervigilant concerns and attention for the baby, extreme lability, constant worry) but nevertheless can cause significant distress and disturb mother–child interactions (93, 111). Creating further, more clearly defined subgroups would have yielded in too small sample sizes. So, conclusions for different diagnoses of anxiety on sensitivity must be postponed to future studies. Still, the investigation of subgroups and comorbidities remains an important goal for the present method.

Further Thoughts on the Use of the Method in Postpartum Psychiatric Disorders

Intercorrelations

During development of the method, it was noted repeatedly that "responsiveness" and "promptness" correlate highly significantly and were difficult to separate. We thought about combining these two scales again as the NICHD-ECCN Scale does but decided to keep them separate. First, we found "promptness" to differentiate between depressed-only and depressed-anxious mothers, as presented above. Second, during scale adaptation, we often found single videos (psychosis or very severe depression), where the difference was more obvious as responsiveness was very delayed. Third, all scales correlate with each other what is typical in this research sector. All EA Scales (86) correlate with each other, and in the Ainsworth Maternal Sensitivity Scale, the author describes the scale "sensitivity" as a key variable correlating with the other scales. The positive and negative affect scales correlated very high, too, especially in the group of mothers with mental illness. This is likely to represent the emotional flattening of mothers with depression.

Scaling

A related discussion concerns the scaling of the subunits. A rank of 3 (the mean) relates to a mother who behaves a bit more often with good caregiving skills than bad. Healthy mothers rank relatively high (around 4 from 5). Our present clinical subsample ranges from 5 to 2; only in training videos did we find ratings of 1. Why is this so and should we have adapted the scaling? The scale is not conceptualized to separate very good from perfect maternal behavior. With a rank of 4 or 5, we can assume that the behavior in the observed sequence was ideal. It must be noted that the situation was short had only little instruction. A healthy mother often knows how to perform ideally and can act so for a certain amount of time in stress-free situations. Mentally ill mothers are not necessarily "disturbed" in their sensitivity but limited in their capacities (107) and can partly behave normal. Moreover, although the group presented here is severely impaired, the therapeutic staff decided that the child's well-being is not critically endangered. Otherwise, they must have separated mother and child and decided to treat the mother as inpatient first. We believe that, with more demanding observational settings or other types of mental disorder as psychosis, scores will be lower. Methods of stress reduction or increase can change maternal performance, leading to higher variance between mothers.

Suggested Video Length and Observational Situation

In hospital routine, we implemented video observations with a length of 10 min displaying diaper-changing situations and free play. Especially in the group of mentally ill mothers, videos from the beginning of our studies were strikingly shorter. Whereas the CARE-index requires 3 min of observation of mother–child interaction only, the EA Scales (86) recommend analyzing much longer observations, up to 2 h. Both observations pursue different goals, namely, to decide about intervention with the CARE-Index (91) while extracting information about attachment measures with the EA Scales. Therefore, different recording times are reasonable. In our study, we recommend recording times around 10 min and include a semistructured situation and free play. Because attention span is reduced in depression, video observations should reveal deficits in maternal behavior quickly (112, 113), although many are also capable of sustaining normal behavior for a while. To increase motivation for participation and compliance for therapy and science, stress should be as low as possible during recording.

According to our clinical observations, mentally ill mothers seem to lack ideas for interactive play that may evoke feelings of insufficiency in the face of a given play task. If in future studies, higher differences between the groups are favored, stress in observation situation can be increased by using more difficult tasks or observe mothers with siblings (114). Not only attention itself but also divided attention is reduced in depression (112), what should also affect mother–child interaction in more demanding situations. In this sense, it has been shown that contact to a stranger reduces responsivity to child's signals in mothers with social phobia (115).

Future Directions

The present adapted method is useful to measure sensitivity in postpartum psychiatric disorders. Moreover, it should allow testing for relationships to attachment qualities with later strange situation procedures. Still, the next phase of research includes validation work with other observational tools and measures of attachment. Moreover, an extension of the sample with other diagnoses as psychosis and OCDs is planned. Analyses for further understanding of the disorder and its relation to maternal behavior and then child outcome are prospects. Of further importance in the group of mothers with postpartum psychiatric disorders is the interplay of genetic and caregiving influences in the prediction of attachment and further development of the child. It could be shown that higher maternal sensitivity can be a buffer for disorganized attachment in cases of genetic risk (116).

To describe the full spectrum of deficits in mother–child interaction, risk and protection factors for altered sensitivity postpartum psychiatric disorders should be assessed. Own childhood experiences are a crucial factor for the quality of sensitivity, with adverse experiences leading to less sensitivity, whereas positive experiences make warm and sensitive parenting more likely (117, 118). Moreover, experiences of emotional or physical abuse during childhood are predictors for hostile behavior toward her own child (119), as well as for postpartum maternal depression (120). Lack of social support is related to lower sensitivity and responsiveness toward her own children (121, 122) and a key factor for the development of depression (12). Socioeconomic pressure, as low income or unemployment, leads to lower quality in interaction (123) and increases depressive symptoms (12). In contrast, higher education and maternal employment are related to more sensitivity and less control of children (124). For future studies it may be interesting to study those risk and protection factors that play a role in the development of mental health for both parent and child.

ETHICS STATEMENT

This study was carried out in accordance with the Declaration of Helsinki and the permission and recommendations of the Ethics Committee of the Friedrich-Alexander University Erlangen Nuremberg and the Ethics Committee of Technical University Dresden (Ethics committee of the FA: 320_15 B, Ethics committee of the TU: EK450 22013). All participants gave written and informed consent. For all data concerning children, written informed consent was obtained from the parents.

AUTHOR CONTRIBUTIONS

CH and MG designed and conducted the study, created the manual, and wrote the manuscript. SG was involved in study design, setup of the manual, assessment of control group, and editing the manuscript. JJ-H and SS were involved in study design and assessment of the clinical group and editing the manuscript. JF worked on the manual and the introduction. GS was involved in study design, edits on the manual, statistics and edits on the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2019.00471/full#supplementary-material

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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

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The challenges involved in engaging young people who have a parent with a mental illness in research and in programs are well documented. Social networking sites provide a potentially useful medium to include at-risk youths and their families by removing some accessibility barriers that may prevent engagement and connection with individuals whose parents or family members have a mental illness. This paper examines how social networking sites can be used to recruit youths and/or their families and engage them in research. Applying a case study analysis, the implications of using social media as a tool for recruitment and data collection and the ethical considerations and limitations will be discussed. Results tentatively indicate that social networking sites may be an effective method to engage young people of parents with a mental illness. The study argues for more informed use of social platforms for the translation and dissemination of research and intervention prevention. Overall, this paper will contribute to public mental health practice through guidelines and policy about social media research with at-risk young people and their families.

Keywords: young people with parents with mental illness, social networking sites, mental health practices, recruitment, research with young people, online research methods, youth at risk, research ethics

INTRODUCTION

Young people who have a parent with a mental illness can experience difficulties throughout their lives, sometimes beginning in their early years. These difficulties can impact on the young person's well-being and can lead to their own mental health challenges (1). An epidemiological study suggests that around 21–23% of young people have been found to have, or have had, a parent with a mental illness (2). Research consistently reports higher rates of emotional, behavioral, and developmental difficulties in these youths compared with those in the general community (3), with long-term effects including social and occupational issues (1). They may experience increased rates of psychiatric disorders through an interplay of both genetic and environmental factors (4). Self-harm and suicide rates tend to be higher in these youths (5, 6). The young person may harbor resentment towards the parent (7) as well as experience negative emotions including shame, depressed mood, fear of conflicts, loneliness, feelings of abandonment, anger, or envy of peers (8). It could then be said that young people who have parents with mental illness and/or substance abuse disorders are an at-risk group within society. Despite the risks that the young person with a parent with a mental illness may experience, many demonstrate resilience. Researchers agree that some children with a parent with a mental illness can cope well (9), acquire and practice effective problem-solving skills (10), and access functional social supports external to the family (9). Young people need protective factors, for example, adequate community support and intervention (11) and accurate mental health information (12), to reduce the impact of negative risk factors. In this paper, young people or youths are considered to be aged between 14 and 18 years old.

Children of parents with a mental illness are aware that there are some predictable and unpredictable elements of their parents' behavior, and it is the unpredictable elements that often impact the young person the most (13). Their understanding of parental mental illness changes with age as well as with the amount and type of information about mental health they acquire (14). Slominski (15) measured adult children's perceptions of growing up in a family where a parent was diagnosed with a mental illness. He, too, proposed that it was not only the experiences themselves that shaped the individual but also the way the individual made sense of those experiences. When young people do not receive complete, accurate mental health information, they may come to their own—often inaccurate—conclusions that can exacerbate their fears. A general lack of awareness of their parent's condition may negatively influence the youth's ability to access systems of support. Some young people make attempts to obtain information from the mental health professionals who care for their parents, but many report that they do not get the recognition they feel they deserve, for example, a lack of acknowledgment of the young person's role in caregiving and their need to understand what is happening for their parents and themselves (7, 16).

While extensive research has identified young people's risk of adverse outcomes and the potential for resilience (2, 17), it is challenging to recruit young people and families where a parent has a mental illness in research and into prevention programs (18). The difficulties of engaging with youths from high-risk communities in research may be due to barriers in recruiting vulnerable youths (19, 20). These barriers can include i) insufficient understanding of research (21, 22), ii) mistrust of research (21), iii) stigma associated with participation in clinical research (23), and iv) issues surrounding parental consent (23). There are few papers that outline "adolescent friendly" recruitment strategies that can be employed to overcome barriers to recruitment and effectively engage with at-risk young people in research initiatives (24). A potential way to address these barriers can be to recruit at-risk youths through the mediums in which they connect with most—that is on the Internet *via* social networking sites (25). How social networking sites are used to recruit youths and/or their families and engage them in research should be explored, as well as the potential to use social networking sites for translation and dissemination of accurate public health information. This paper is the first attempt to examine social networking platforms as a potentially useful medium to include at-risk youths and their families by attempting to remove barriers that could prevent access to and involvement of individuals in research.

A TECHNOLOGY-BASED RESPONSE: SOCIAL NETWORKING PLATFORMS AS A POTENTIALLY USEFUL MEDIUM TO INCLUDE AT-RISK YOUTHS AND FAMILIES

Social networking platforms are online communication systems that facilitate community-based contributions, interactions, content sharing, and collaboration in virtual networks *via* social media websites Facebook, Twitter, blogs (i.e., Tumblr), and/or YouTube (26). Social networking sites have potential when recruiting a younger population as well as targeted groups of individuals (24), such as youth with a parent who has a mental illness. For some young people, the Internet is their primary source of information and their main means of communication with the world (25). There has been a large growth of online social connections: Facebook has around 1.59 billion monthly users (Facebook 2018), while on Twitter around 500 million tweets are sent per day (Twitter 2018). Social media is increasingly an information source for youths, where they use websites to learn about the world and themselves and connect with others (27). Young people are more inclined to use social networking sites than older adults (28). As a result of the rise in social media usage, researchers and policymakers have begun to consider how such platforms can be harnessed to support research, share accurate public health messages, and inform robust evidence-based policymaking.

A systematic review of 25 studies that employed Facebook to recruit young people (10–18 years old) found that paying Facebook for advertising about specific research projects was a successful tool for recruiting adolescent participants into mental health research (24). However, the review noted that there have been few studies in which underrepresented or at-risk adolescents have been recruited *via* social media platforms (24). There have been no studies that examine social media use in recruiting children and families where a parent has a mental illness. There is relatively little guidance on using online platforms in social and mental health research. Further research is clearly needed particularly given that social networking sites are considered to be an emerging data collection, recruitment, and research dissemination strategy that is gaining traction in research (25, 26, 29).

Social networking sites can be a useful tool for gathering data, which has the potential to address barriers in research. For example, they can be a platform to deliver health interventions and recruit participants for studies in a cost-effective manner (25, 26, 29). Some of the barriers to recruiting and engaging young people who have a parent with a mental illness in research may be addressed through social platforms. Ongoing discussion about the purpose of social media methods is necessary to identify how social networking sites are applied across individual research studies. This will contribute to the field's understanding of the ways in which these platforms can be used to access at-risk groups in the community (19). There are, however, several factors to consider, such as whether to use social media research as the sole method or in conjunction with other methods, processes of data collection and analysis, ethical implications of consent and privacy, and the presentation of social media research findings. There is a lack of policy and guidelines for using social networking sites in research (19). This paper will apply a case study analysis of a research project that used social media as a strategy to recruit young people who have parents with a mental illness and to translate and disseminate research knowledge.

METHOD AND RESULTS OF CASE STUDY

Stage 1 of the research project was to explore whether a social networking site could be used to recruit and engage with youths whose parents have a mental illness (the focus of this paper). Stage 2 gathered youth people's experiences of help seeking using a mixed methods approach (16). The objective of stage 2 was to identify their expressed preferences for supports (for example, informal supports such as talking to peers or parents) and more formal interventions (such as developed programs). In order to answer this research focus, a self-administered questionnaire followed by individual semi-structured interviews was employed. In total, 173 young people (13–17 years) who have a parent with a mental illness completed a questionnaire and 6 of these participants took part in interviews. Initially, 175 youths were recruited; however, 2 in the sample were male and subsequently removed, leaving the sample of females only. During stage 1 of the study, the social networking site Facebook was used in two ways: i) as a recruitment strategy in response to the research objectives and questions, and ii) as a technology-based mechanism for knowledge translation and dissemination with the aim to provide accurate and helpful information about mental health and support for young people and their families.

Online Research Presence

A Facebook page was created to provide participants with information about the research project (see **Figure 1**). It was titled "Children of Parents with a Mental Illness: Views of Intervention." The main objective of the Facebook research page was to disseminate the online questionnaire, promote awareness of the study, and disseminate public mental health information. Posts made on the Facebook page included information about the research team and project, support services, the latest research about mental health and well-being, and study announcements that included a link to the study questionnaire. On average, the lead researcher made two Facebook posts a week. During the recruitment period, the researcher made 100 Facebook posts, received 75 Facebook followers, and liked 78 organizations relating to young people and mental health. The lead researcher reviewed and approved all wall posts in a timely manner and took them down if inappropriate due to swearing or cursing. The author interacted with other Facebook pages by "liking" the pages of community

You are posting, commenting and liking as Children of Parents with a Mental Illness: Views of Intervention — Change to Christine Jeanette

Ready To See Your Updated Page?
Your Page will automatically be updated on Thursday.

Does your Mum or Dad have a Mental Illness? Are you between 8-17yrs? If yes you can fill in a survey & get a voucher http://tinyurl.com/ViewsIntervention

Does your Mum or Dad have a Mental Illness? Are you between 8-17yrs? If yes you can fill in a survey...

Like · Christine Jeanette likes this.

FIGURE 1 | Facebook research page.

organizations. The organizations were found using keyword search terms on the Facebook search engine, such as "children of parents with a mental illness," "young carers," "youth mental health," and "youth well-being." The "About" section on the Facebook research page included detailed information about the study, the researchers' names and affiliations, and links to the researchers' official affiliation website.

Advertising Research on Facebook

The researchers paid for targeted advertising to meet specific criteria. The criteria included the age group of the targeted youths (13–18 years), a specific population (children who have parents with a mental illness), and geographical location (Australia only). The study advertisements were only presented to Facebook users who met the study criteria set by the researchers *via* the Facebook administration page. It was shown to potential participants on their personal Facebook pages *via* their desktop computer and/or mobile device. Advertisements on Facebook must adhere to language and image guidelines. The headline had a limit of 25 characters, and the body text of the advertisement had a limit of 90 characters. Images were required for all advertisements. The study advertisement (see **Figure 2**) was placed on Facebook from August 2013 to February 2014, inviting youths to participate. Those who clicked on the poster were directed to the research explanatory statement at the beginning of the survey on a survey website called Qualtrics. Consent was obtained from the young person when they agreed to complete the questionnaire through an online form at the beginning of the questionnaire. Participants were requested to obtain parental consent before completing the survey; this request was on each page in the survey (e.g., "Ask your parent if you can take part in this study").

A total of 1,382 people clicked on the link to the questionnaire on the advertisement on Facebook. Of those who accessed the website link, 192 provided consent. Of these, 175 met the study inclusion criteria and subsequently completed the questionnaire. No other methods of data collection (such as mail-out surveys) were used. Of the 175 samples, 6 participants agreed to take part in an individual semi-structured telephone interview and were offered a $10 gift voucher upon completion. The study found that the use of social media, specifically Facebook, was a more effective recruitment tool than using traditional methods (such as flyers or mail-out surveys) to recruit young people of parents with a mental illness (12, 16).

Online Snowball Sampling

Snowball sampling occurred online during the recruitment phase of this study. Snowballing transpired when community organizations shared the Facebook research page and the survey on

Children of Parents with a Mental Illness: Views of Intervention

Just now

Does Mum or Dad have a mental illness? If yes, please consider filling out in this survey!

Tell us what you think

tinyurl.com

FIGURE 2 | Example of advertisement used.

their websites and social media pages. From there, a snowballing effect occurred: the Facebook research page and posts made by the author were distributed *via* social media through community organization followers (individuals who followed the page of the organization). These were then subsequently shared by individuals on their individual social network site, that is, on blogs, in personal newsletters, and/or on their Twitter/Facebook pages. In addition, online snowball sampling occurred through group and friendship connections between Facebook users, where the study details were shared further.

DISCUSSION

There are benefits and challenges of using social media as a recruitment tool when engaging with vulnerable youths in research. Some benefits include using a platform that is widely interacted with by young people, whereby the research can be shared across many social sites within a young person's network, enabling a questionnaire to be disseminated quickly among many potential participants (30, 31). Amon and colleagues (24) suggest that advertising on social networking sites alongside traditional means of recruitment (i.e., flyers, mail-outs, and telephone calling) could be helpful in accessing larger and more specific groups. Social media presents a fast, affordable method of recruiting study participants for questionnaire research. In the current study, using social media was found to be a successful strategy to recruit a large sample of young people who have parents with a mental illness in a short period of time (16). Clearly, online recruitment for structured questionnaires *via* social media is a feasible and efficient tool that can be used to access at-risk individuals.

The Australian Psychology Society (32) released a national survey examining the impact of technology and social media on the well-being of young people (aged 14–17 years old). The survey found that young people are self-reporting that they use social media for an average of 3.3 h each day, on five or more days of the week. The vast majority of teenagers reported that their screens and social media accounts were a positive part of their lives. Many use social media channels to connect with family and friends and to entertain themselves. Despite social media playing a positive role for most, the high use of social media and technology can have a negative impact on youth self-esteem. Two in three young people feel pressure to look good, and nearly a third of youths have been bullied online (32). Clearly, there are ethical and potential limitations when using social networking platforms in practice or research that should to be taken into account.

Young people and their families from rural or lower socioeconomic status have been found to be more difficult to recruit and engage than those from higher socioeconomic status or from urban areas (33). This may be due, in part, to disadvantages associated with living in these areas that include reduced quality of services (34), but can also be attributed to the challenges that coincide with living in poverty or far from central towns including lack of a reliable telephone service and limited or nonexistent access to the Internet (33). Thus, a possible limitation is that participants recruited through social media may not be representative of the population as not all young people have access to computers or the Internet. Some research suggests that the digital divide is reducing as more individuals are online; however, age is a crucial factor. Nine in 10 people aged between 15 and 54 use the Internet; however, this statistic reduces to 8 in 10 of those aged 55–64 years, and to under 6 in 10 of those over 65 years (35). The age of young people who can be recruited *via* social platforms is limited to those over the age of 13. Due to age restrictions, individuals 13 years and below cannot create a Facebook, YouTube, or Tumblr account.

A challenge of social networking sites is that the participants are not representative of the population. The findings here thus might not be generalizable to the wider population. It could be that the population is not representative because children of parents with a mental illness may search for information elsewhere than on social media platforms. A predominant limitation of social media for collecting data is that it may introduce self-selection bias. Self-selection bias can lead to a sample bias. Self-selected and self-reported data may affect the reliability and validity of results (36). The validity of the completion of the online survey should be considered, as members of social media sites may not be truthful in their responses and/or complete the survey as a "fake" participant. The online identity of a person may not be the same as their real-life identity. Often, social media identities are to a certain extent more "positive." This limitation could be helpful, though, if the research is focused on a particular group that is active on a social media platform, and there is the potential to gather data from participants who may not tend to respond to other types of research methods (37). The anonymous nature of online recruitment can be a challenge in that researchers may not be aware if a young person is upset or triggered by the content in the survey. Researchers are not able to check or provide more information if the youth does not understand the project. The Facebook research page seems to have multiple purposes, which makes it difficult to disentangle to what extent this medium is useful for research participation versus general information versus referral or information to other prevention services. For example, the current case study recruited mainly females; thus, the findings or suggestions here may not be appropriate for the recruitment of males. While Facebook is still one of the most used networks in general, currently among some young persons' Facebook is not their only or main social site they access, when compared to other platforms such as Snapchat, Instagram, or YouTube (38).

Rarely are there papers that include strategies for recruiting at-risk youths (18, 24). Effective recruitment strategies are needed as it is widely recognized that underrepresented adolescents can be challenging to recruit due to barriers that impact their participation. A barrier to youth participation is the way in which at-risk adolescents are recruited. Consent for youths to participate in a study is usually attained from an adult in the young person's life, such as a parent or guardian, known as a "gatekeeper." The parent/guardian is not typically the intended audience for a study directed at youths, yet recruitment strategies are often focused at connecting with the "adult gatekeeper" in order to attain consent for the young person's participation. The challenge in accessing youths *via* a "gatekeeper" is that two sets of individuals are to be recruited for the study and the researchers usually need the parent/guardian to be interested in the research and then the youth. Depending on the study considerations required in recruiting vulnerable youths, such as whether the young person themselves can self-select into a study by providing their own consent, young people can then have "buy in" into a project in that they make the decision to participate in the study. The young person then becomes the "gatekeeper" who chooses to share study information with their parent/guardian, potentially building trust and respect of the young person. This approach respects autonomy and privacy of the young person, who, as noted by the United Nations Rights of the Child under the Convention on the Rights of the Child (39), can decide and choose to take part in matters that directly impact them, and that includes their inclusivity in firstly opting into developmentally appropriate research. A "youth-centered approach" has ethical justification. Some researchers advocate, however, that "adult gatekeepers" are a necessity in that they protect the adolescent from exposure to information that may not be appropriate (40). However, first and foremost, researching alongside at-risk youths requires procedures for the researcher to be aware of, or sensitive about, the "risk status." An understanding and sensitivity is required of researchers regarding the information they are gathering and to be done so in ways that convey respect for the young person and their circumstance, for example, using a non-judgmental attitude, respect of opinions, and collaborative approaches to consult with young people. A paradigm shift is needed for some whereby youths are seen as experts on their lives, where they have the right to share their perspectives and to initially choose to engage in research.

An ethical consideration when removing "adult gatekeepers" is regarding the ability of minors to provide consent. In the study by Grové et al. (16), formal consent was required from youths only and formal consent was not required from the parents. However, youths were invited throughout the online questionnaire to inform their parents of their participation in the study. The young persons were reminded that they did not have to be involved in the research if they chose not to. The gatekeepers were not in the role of insisting or convincing a student to participate in the research project.

Some youths express concern about identifying or sharing their at-risk status, such as being a child of a parent with a mental illness. If youths are recruited in community or public places, such as at school or through an organization, they know, and are fearful others know, they are being sought for a problem or issue and may not indicate their interest. A fear of exposure of their at-risk status and a lack of privacy are a barrier to youths participating. Social networking sites attend to their concern in that they can take part in the research anonymously and privately. Respect for youth privacy and autonomy, again, are key to recruiting and retention (41) whereby youths view the research as "something interesting and not because of a problem." However, anonymity can be a challenge in that researchers may not be aware if a young person is upset or potentially triggered by the content in the survey or if the youth has the language to understand the research. Young people who participate in preventive interventions may not necessarily want to participate in research or vice versa. Social media could help with intervention recruitment and delivery—an area that future research could explore.

Research using social network sites is a relatively new phenomenon. Social networking sites appear to be suited to recruiting a younger population who regularly use these platforms and also may be hard to reach, for example, young people of parents with a mental illness and/or substance abuse disorder (18). More generally, the emerging role of social media in research raises ethical concerns:

- privacy of participants (Are their IP addresses identifying)?,
- ownership of the data gathered (Do the data belong to the third-party website as well as the research team? What is done with these data and who are they shared with)?,
- participants providing informed consent (Is informed consent needed when conducting research studies on public websites and social media platforms and, if so, how should the researchers attain this consent)?, and
- access to participants (Who has access to which participant groups in the community and which participants are being excluded)?.

Research that uses social media platforms may consider guidelines for social networking platforms in research that are specific to their study and its context. The guidelines may outline the benefits and limitations of interacting with social media, including the relevant ethical, legal, and governance issues. The practice guidelines will depend on the sensitivity of the research area and if privacy and confidentiality (or lack of) can be provided to participants. For example, depending on the social media user's privacy settings, posts that they follow, like, or comment on can be seen by others online, creating a digital footprint or trail of interaction. Researchers may also choose to monitor and record the frequency of occurrence of the online snowball sampling strategy to determine its impact.

Researchers may consider whether the method of social media recruitment and dissemination is the appropriate tool to use, with attention to ongoing and regular interactions by the research team of content and public communication within the platform. Given the infancy of social media research, researchers need to make methodological decisions based on previous case study illustrations of research using social media rather than prior personal experience. Social media methods often make use of existing, publicly available data, so the need for participants to complete demographic questions can be reduced. However, existing data may not be accurate and ethical discussions of whether consent should be provided for its use are warranted. Researchers should provide information about their project and the research team publicly, including the research purpose and how the data gathered will be used, stored, and accessed. Ways to protect the interests of vulnerable populations in the community, such as young people who have a parent with a mental illness and/or substance abuse disorder, need to be included in project-specific guidelines. The guidelines may choose to provide suggestions about support, promote help seeking, and disseminate summary information gathered during the study. More research is vital to determine how scholars can use social media to explore participants' understanding, engagement, and active participation in public mental health practice guidelines and policy.

A challenge with some of the research with children of parents with a mental illness could be that many participants are already connected to support networks. Often, they are recruited *via* this connection. Unless the research aims are to examine outside of this network specifically (recruiting external to support networks, in rural areas, or those not accessing support), it can otherwise be challenging to recruit. Using a social networking site (i.e., Facebook) to aid in recruiting participants outside associated, connected networks did help in addressing this bias. However, if researchers are recruiting vulnerable families who may not be connected to support, educational information about supports (i.e., online or telephone counselling services or how to connect with local doctors) should be provided.

CONCLUSION

There are challenges in connecting with and engaging with young people in research, particularly if the young person is considered underrepresented in the community. An untapped approach in attaining youth involvement in research can be through social networking sites. Social networking sites are platforms that young people use directly and regularly, and could be used as a possible recruitment tool depending on the research study objectives. Further research will do well to illustrate the use of social media in research, the risks and benefits, and how efficacious this strategy is in recruiting different at-risk groups of youths.

ETHICS STATEMENT

Ethics for this study was approved through the relevant Monash University committee. Ethical approval number is MUHREC CF13/1766-2013000918. Paid advertising on Facebook was used to recruit participants. Potential participants were shown the information that is on the recruitment poster as an advertisement. The Facebook recruitment advertisement that was shown to potential participants comprised of 1) short title (e.g., "Tell us what you think"), 2) an image (e.g., stock image of young people), and 3) main text, up to 74 characters in length: "Does Mum or Dad have a mental illness? If yes, please consider filling out in this survey!" (See **Figures 1** and **2** for an example of the advertisement used for mobile device and desktop computer). If participants chose to take part in the project, they then clicked the advertisement. This took them directly to the explanatory statement and participant consent forms for participants to take part in the study.

AUTHOR CONTRIBUTIONS

The author developed the conception and design of the research work, conducted the data collection, the data analysis and interpretation, drafted the article, provided critical revision of the article, and examined the final approval of the version to be submitted.

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Mental Health Literacy of Youth That Have a Family Member With a Mental Illness: Outcomes From a New Program and Scale

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A program evaluation examined mental health literacy levels and coping outcomes for youth (ages 10–16), before and at the end of their participation in a manualized, school-based mental health literacy program called Youth Education and Support (YES). Most of the youth reportedly had a parent or other family member with a mental health disorder such as depression, anxiety, and/or substance abuse. The mental health literacy levels of program participants from pre to post were evaluated with the developing Knowledge of Mental Illness and Recovery (K-MIR) scale. This scale was validated using item-response theory, demonstrating good psychometric properties. Youth answered two coping questions about their use of positive coping during the program and coping skills compared from pre to post intervention. Findings revealed that youth levels of mental health literacy increased significantly from pre to post program participation. Over 90% of the youth reported an improved use of positive coping strategies from pre to post intervention. The program appeared to deliver enhanced levels of literacy and coping for this sample of youth. The scale appeared to be appropriate to measure youth mental health literacy. Recommendations for practice, policy, and research are offered.

Keywords: mental health, substance abuse, adolescents, children, families, psychoeducation

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INTRODUCTION

Mental health disorders are one of the most common sources of disability in the world (1). The social, emotional, cultural, and economic costs of mental illnesses affect people across community, business, school, health care, family, and individual sectors (2, 3). Despite the magnitude of the impact, mental illness is an all-too-often a stigmatized topic all over the world. There is a need for mental health literacy programs that provide accurate, non-stigmatized information about mental health disorders and recovery (4). The programs provide practical application of how to seek help for mental health concerns and how to help others who may have mental health symptoms (5).

Children of a parent, or other family member, with a mental illness face particular risks for acquiring a mental health disorder such as depression, anxiety, schizophrenia, bipolar disorder, and/or substance (6). The children, including youth ages 10–16 included in this study, have for too long been "invisible" to services providers of mental health services (7, p. 86). Within individual medical model systems, mental health consumers are rarely if they are parenting minor children. In most of the world, there are little to no mental health literacy programs for youth with a parent or other family member with a mental health disorder (7).

This study is an evaluation of a new youth-focused mental health literacy program. Most of the youth have a parent or other family member with a mental health disorder. The current study includes the psychometrics for a developing scale to measure youth levels of mental health literacy.

BACKGROUND

Youth with a parent with a mental illness comprise a large population. One of five people have a mental illness and parenting rates are similar for adults with a mental illness, as compared to adults without a mental illness. England and Sim (8) explored government and private health care databases to find that over 22 million children in the U.S. have a parent with a diagnosis of major depression. Maybery et al. (9) used Australian national health care data to estimate that around 21–23% of young people have a parent with a mental illness.

Risk and Resiliency

Risk and resilience theorists have provided a theoretical framework to understand how to promote healthy psychological development in individuals that face increased levels of psychosocial adversity (10–15). Youth that have a parent with a mental illness are at a higher risk of developing a mental health illness when compared to their same age peers (16, 17). These youth can be at a higher risk of developing behavioral, developmental, and emotional difficulties (18). Young people of parents with a mental illness may experience school problems and difficulties with attention or self-regulation. For example, they experience higher dropout rates at school (19), an increased likelihood of being taken into foster care (20) and an increased risk of developing a substance abuse disorder (21). Self-harm and suicide rates are higher in young people who have a parent with mental illness (22). Some young people may need to be re-located if their parent is hospitalized and/or very unwell (23). The youth may face increased risk to acquire intergenerational mental illness (24).

Childhood development can be affected adversely when they take on caregiving for parents in a way that is long-term and disproportionate to the child's developmental level of emotional maturity and understanding (25). They can experience negative emotions including shame, fear of conflicts, loneliness feelings of abandonment, sadness, anger, or envy of peers (26). Sometimes, they may harbor resentment toward their parent (27). Often these youth do not receive developmentally appropriate information about mental health (28) and, similarly, do not have the opportunity to develop an adequate and accurate understanding of their parents' mental illness (29).

Not all children are affected adversely by parental mental illness, nor will all children in the same family be affected in the same ways (18). Young people also differ in their beliefs and understanding about the nature of mental illness, its causes and the ways to treat mental health difficulties (30). Darlington et al. (31) suggest that mental illness in a parent does not automatically result in negative outcomes in children. Many parents with mental illness can form healthy attachments and provide nurturing care to their children. Even some situations where children take care of their parents can lead to positive outcomes (25). Caregiving may help the youth build a sense of purpose, potentially supporting resiliency, and reinforcing the parent-child bond (32).

Despite the risks and outcomes that the young people of parents with mental illness may experience, many of these youth demonstrate resilience. Resilience appears when youth engage in positive adjustments to a situation that includes conditions of challenge, risk, and/or adversity (33–35). In general, the more that a young person are seen as resilient, the better their mental health (36). Youth can achieve resilience when there is a balance between adverse events and protective factors. The presence of protective factors can safeguard young people from the impacts of parental mental illness by potentially "buffering" the impact of the risk factors and reduce the vulnerability of a young person acquiring a mental illness. Protective factors include nurturing care by a parent, a close relationship with parents, the child's own problem-solving skills in response to stressors, psycho-education, and accessible social support from peers and other adults (such as teachers) or family members, such as aunts, uncles, grandparents, or siblings (37, 38).

Stress, Coping, and Adaptation

Another model for considering youth with a parent with a mental illness is stress and coping theory (39, 40). Youth that have a parent with a mental illness may share some of the secondary effects of living in a home where a parent has a mental illness such as underemployment, unemployment, poverty, and/or parental divorce and/or separation (41). Some youth report that it can be stressful living with a parent, sibling, grandparent, aunt/uncle, and other family member with a mental illness as family interactions may change as the family member's illness symptoms can increase or decrease from day to day (41, 42). Further, youth may worry the family may not be able to pay the bills if a parent or other breadwinner is unable to work due to mental illness symptom exacerbation; they may fear others will judge the family member harshly and make fun of the person and, sometimes, the family as a whole (41). They may worry they will inherit a relative's mental illness (43, 44). They may not know how to talk about the relative's mental illness (45).

Coping is part of health and wellbeing, that includes healthy environments, responsive parenting, sense of belonging, healthy activities, resilience, and if mental illness symptoms arise, treatment of the illness (46). Youth can learn to manage stress by engaging in healthy coping activities such as coping self-talk and positive self-talk (47, 48). For example, positive coping behaviors can include exercising, talking to friends, writing, making a craft, and/or listening to music. It can include seeking help for family mental health crises. Stallman (46) notes that youth can make a coping plan to recognize personal and family stress and then engage in stress-reducing coping behaviors. As youth increase their coping behaviors, they can learn to better manage stress and, over time, adapt to stressful situations. They can even move beyond mere adaptation as their crisis plan offers new buffers or protective factors toward youth developmental resiliency (49).

Mental Health Literacy Interventions and Measures

Mental health literacy interventions, sometimes called psychoeducation, aim to provide accurate, non-stigmatized information about mental illness and recovery to mental health consumers and/or family members. Jorm et al. (50) describe mental health literacy as consisting of several components such as the:

> ability to recognize specific disorders, knowledge of how to seek mental health information, knowledge of risk factors and causes, knowledge of self-treatments and of professional help available, and attitudes that promote recognition and appropriate help-seeking (p. 469).

Mental health literacy interventions can teach young people coping skill and strategies to help them reduce stress. They can also respond to specific requests of young people who often ask for more mental health information to help with their understanding about their parents' illness (28, 29). Many young people report that they do not receive enough information about their parent's mental health (28) and are left "guessing" or "figuring out" what is happening to their parent. Some youth may develop misconceptions about mental illness such as blaming themselves for their parent's illness, or believing the mental illness can be caught like a cold or be "passed" onto friends (51). A lack of understanding about parental mental illness hinders the recognition and promotion of appropriate help seeking and reduces the likelihood of young people pursuing help for their difficulties (52). Mental health literacy programs often offer social support; for example, youth may use the mental health information and supportive relationships acquired in mental health literacy programs to manage practical issues such as finding support when their parent is hospitalized and when they need a break from caring for their parent (23, 51).

Clearly, youth would like to be "kept in the loop" and informed about their parents' mental illness (53) and want to talk about their experience (29). However, they may be unsure of the implications of accessing support, such as discussing the impact of stigma for having a parent with mental illness (54). Each young person will process information about his or her parent's illness differently and so will have varying information needs. Young people's understanding of their parent's illness is constantly evolving and changing including the amount and type of information they acquire (55).

Gladstone et al. (29) suggest that psycho-education should attempt to include young people's views of their parent's illness and should recognize the youth's role within the family context. However, it is important to note that the kind of mental health knowledge, and how it is shared with youth, is mostly developed and implemented by adults who decide what youth need (56). Mental health information for youth should be examined from a child-centered approach; there is a need to investigate how mental health information is used by youth and whether this is helpful for the young person, if at all (29). The voices and experiences of young people and their families when developing mental health literacy programs also need to be heard (53, 57).

There are many emerging mental health literacy programs designed to promote resilience in children who are with parents or family members with mental illnesses (7, 18). For example, in Finland (58) and Sweden (59), there are national implementations of a family intervention program to encourage child and parent communication about the parent's mental health condition, recovery strategies, and social service coordination (58). However, in most areas of the world, only a small percentage of youth with a parent or other family members with a mental illness have access to mental health literacy programs. Even in Australia, where there are a number of family-, youth-, and parent- centered programs with mental health literacy content, there is limited access to the programs across many regions of the country. Many of the programs are emerging; they are not yet considered evidence-based (18). A lack of evidence-based programs that provide mental health information and support for youth, parents, and other family members could be a potential barrier to funding. Many private and government organizations prefer to fund evidence-based programs (60).

A lack of mental health literacy scales with sound psychometric properties are a barrier to building evidence-based practices, especially in evaluating interventions for young people with a parent with a mental illness. O'Connor et al. (61) conducted an extensive literature review of mental health literature scales. They found that of the measures they reviewed none covered all of the constructs of mental health literacy, such as disorder recognition, help seeking, risk factor identification, information seeking, causes of mental illness, self-treatment/coping, and risk factors. They also reported that most measures used had methodological gaps in psychometric validation and norming.

Since the publication of that article, new scales with increased rigor have been published (62, 63). The first measure was normed primarily on college students (62). The second measure was normed on a combination of health care practitioners and those in the general population ages 15 and up (63). Additionally, the second measure focused on three specific disorders (schizophrenia, anxiety, and depression). The most rigorous mental health literacy measures available appear to be normed only on adults (62). The work of Kutcher et al. (64) included a scale designed to measure mental health literacy as an indicator of increased mental health resiliency among high school students. However, the reading level appears to exceed that needed for middle school students.

Measures have been used to assess mental health literacy programs delivered to young people with a parent with a mental illness (5, 43, 65) or with young people without a parent with mental illness (66). However, these measures do not appear to capture all of the mental health literacy constructs. They also contain limited information about scale psychometric validation or norming. Therefore, building on these current works in the field comprehensive and robust mental health literacy scales are needed for youth of varying ages (4). Specifically, YES program services providers indicate that their evaluation of the program is need of a measure for youth aged 10–16 that have a parent or other family member with a mental illness. More evidence is needed for youth mental health literacy programs and scales to move the field forward toward building evidence-based practices.

METHODS

This study evaluated the outcomes of the YES mental health literacy program. The guiding research question asked, "What are the pre-post mental health literacy and coping outcomes reported by youth attending the Youth Education and Support program?" Given the lack of psychometrically validated measures of youth mental health literacy (4), the first phase of the evaluation study included development and analysis of an instrument to measure youth mental health literacy. The second part assessed youth participant self-reported levels of pre to post program mental health literacy and coping behaviors.

The Knowledge of Mental Illness and Recovery (KMIR) Scale

The KMIR scale items includes 36 questions. Eighteen are set in a true and false format. Eighteen use a four-item multiple choice response set. The primary aim of the scale is to assess the mental health literacy of children ages 10–16. The Flesch-Kincaid scale assigned the KMIR a 4.8 reading level. This means the scale is an appropriate reading level for youth 8 months into the fourth grade. In the US, most of these youth would be about 9 years old. Completion time is 11–14 min. The primary investigator used or modified questions found on mental health websites (67, 68), and as found within a nonfiction book written for teens with a parent with a mental illness (69). In addition, the scale author wrote 16 new questions, as the first version of the KMIR scale was pretested initially with six middle school children and who took the test and then offered suggestions for improvement. Their comments led to the second version of the scale administered to 39 YES program youth. Version two was then administered to 216 middle school students in three schools in the Midwest, which was used in the psychometric validation presented below. The validation sample included 55.10% female participants, with a mean age of 13.54 years (standard deviation 0.71 years, ranging from 11 to 16 years). The participants identified as Caucasian (80.10%), Black/African American (6.50%), Latino/Hispanic (6.00%), American Indian (2.30%), Asian American (0.50%), and other or did not answer (2.8%). Following the validation, the scale was administered to the 46 YES participants in the current study.

To determine the validity of the Knowledge of Mental Ill00ness and Recovery (KMIR) were evaluated for goodness of fit to a graded response model using Microsoft R Open version 3.4.2 (70) and R Studio version 1.1.383 (71). The M_2 limited information goodness of fit statistic (72) was calculated for each subtest, with significant results ($p < 0.01$) indicating less than adequate fit. However, when sample sizes are relatively small, a corresponding small Root Mean Square Error of Approximation value (RMSEA < 0.09) suggests that a lack of fit may be due to a limited amount of "model error" (73); common in strong parametric models (72). The Standardized Root Mean Square Residual (SRMR) is the square root of the difference between the residuals of the sample covariance matrix and the 0covariance model (74). Values range from 0 to 1.0, with well-fitting models obtaining values <0.06 (75). Marginal reliability coefficients (analogous to Cronbach's alpha coefficients) are calculated using Lord and Novak's true score model (76).

TABLE 1 | Rasch model fit statistics for KMIR scale.

	M_2	*df*	*p*	RMSEA	SRMR	Reliability
KMIR total score	1199.88	629	< 0.001	0.06	0.11	0.87
Mental illness	83.79	65	0.06	0.04	0.08	0.69
Recovery	225.37	65	0.11	0.09	0.13	0.68
Stigma	147.34	65	< 0.001	0.06	0.11	0.68

At the item level, fit was investigated further with $S–X^2$ item level diagnostics (77, 78); infit and outfit (79, 80); local dependency (81); and differential item functioning by gender (82). Non-significant $S - X^2$ item values are indicative of good item level fit, while infit and outfit scores between the range of 0.5 and 1.5 are considered productive for measurement. Local dependency indicates a relationship between items where the ability to answer one item is predicated on the ability to answer another item. For example, to demonstrate knowledge of bipolar disorders, one would likely need to be able to demonstrate knowledge of depressive disorders. Local dependency is identified using positive correlation coefficients of 0.20 or greater in a correlation matrix of residuals. Differential item functioning indicates a gender difference in likelihood of correctly endorsing an item for participants of the same underlying ability. One example of differential item functioning would be a question regarding the role of social support in recovery, which is generally answered correctly by females and not by males—reflecting differences in gendered experience and norms rather than knowledge.

The results of the Rasch analyses suggested that aside from a small number of locally dependent items (one item pair in *Mental Illness*; two pair in *Recovery*; and one item pair in *Stigma*), the subscales and total KMIR scales demonstrated reasonable model fit. There were no redundant items, no items demonstrated significant misfit, and no differential item functioning was observed. Infit and outfit statistics were all within the recommended range of 0.5–1.5. The fit statistics are presented in **Table 1**.

The Youth Education and Support (YES) Program

Forty-six youth participants were drawn from an urban, Midwest American middle school. Social workers employed by the school visited grade 5–8 classrooms to talk about students' opportunities to participate in the Youth Education and Support (YES) program to learn about mental health and recovery. If considering YES program participation, youth submitted a note of interest later to the mailbox of the social worker. This ensured that interested youth could not be identified as those turning in program inquiries at the time of the social workers visits to the classrooms. School social workers then met with inquiring youth and once their interests were confirmed, communicated with parents to explain the program, and if parents allowed, obtained signed informed consent for participation. The primary investigator trained the school social workers' in obtaining informed assent from youth and consent from parents in accordance with the overseeing university's Institutional Review Board. Parents also completed youth and family demographic information. Demographic information included parents' checking a general category of relative that they identified as having a mental health disorder. At no time were specific individuals identified by name.

Participation in the study did not require that youth have a parent or other family with a mental illness but it was a preferred criteria for inclusion in the program. Prior to the first group session of the YES program, school social workers obtained a pre-intervention youth completion of the Knowledge of Mental Illness and Recovery scale (KMIR) to assess youth beginning knowledge of mental health literacy information. The KMIR scale included knowledge about common mental illnesses, recovery, and stigma.

The Youth Education and Support (YES) program is a manualized, ten-session mental health literacy program for youth in grades 5–8. It was developed by the first author over a 12 year span of time with recommendations from children, youth, and adults that reportedly have a parent or other family member with a mental illness. Recommendations for the content were also drawn from mental health professionals, family members with a mental illness—especially parents, the professional literature, and reviews of existing mental health literacy program curricula from the United States, Australia, England, and Canada. The program continued to be modified over time in response to program evaluation data drawn from the comments of youth participants, their parents, other family members, middle school teachers, fidelity observers, and program facilitators.

Each session is about 50 min so there is around 8 h of total program time. Across the program, the content covers mental health literacy content including knowledge of mental illness in general, including risks/causes; specific mental health disorders, including major depression, types of anxiety, schizophrenia, bipolar disorder, and substance abuse, e.g., alcohol, marijuana, meth amphetamines, inhalants, and opioid addiction. It covers risks for mental illness such as stress and genetics, while emphasizing that it is difficult to know what causes any one individual to develop, or not develop, a mental illness. A holistic recovery model anchors the recovery-focused learning content. Youth discuss how to seek help for mental health concerns and how to help others with these concerns. Mental illness "truths" (facts) and "myths" (stigma) are included regularly within the program.

Words of the day guide session topics that include YES (introduction to the YES program), mental illness, coping, depression, recovery, substance abuse, co-occurring (mental illness and substance abuse), family (impacts of mental illness on family members), planning (for mental health crises and the future of the youth), and graduation (end of group celebration). Each session begins with attendance, word, of the day, and food and drink, i.e., cookies, fruit, cheese, and water. Every session includes a check-in on stress and coping, as well as discussion of ways to manage stress. Some kind of hands-on active learning activity takes place in each session. For example, youth construct a crisis mobile with drawings and names of behaviors that they believe will help them reduce stress and "stay in balance." They develop a customized individual coping plan with illustrations. They color pictures of a depressed vs. a non-depressed brain based on an MRI example. They engage in a hopscotch-like team competition to plan for ways to work toward their future goals. They watch a movie clip about a family with a parent with a mental illness and discuss what each family member may be feeling and/or thinking. They walk rapidly about the room in a circle demonstrating how substance abuse and depression can have a "circular" effect, sometime leading to increased levels of mental health symptoms.

All of these activities seemed to work well with all of youth, but those ages about 13–16 spent more time on discussion. For youth ages 10–12, facilitators talked at a slightly slower pace and used lower language levels. At each session end, each youth chooses a small item from a "prize box" to reward their participation. Each session ends with a take home educational document to share with parents. For example, some of the parent handouts are how to talk to a child/youth about mental illness and when and how to seek help should a parent have concerns about their son or daughter's mental health. These are general information handouts; no specific youth, parent, or family application is included.

The program was co-led by a school social worker and university faculty member with expertise in mental health practice and research. Sessions took place during the school day at varying days of the week and times of the day to reduce youth participants from missing too much of one class. Almost every session included a trained observer who completed a YES program fidelity scale that measures the extent the session met stated learning objectives. The observer sat in the rear of the classroom and did not participate in programming.

At the last session, facilitators asked youth participants to think about their coping at the beginning of the program. A pause followed for youth reflection. Then they were asked to think about their level of coping at the end of the program. The facilitator emphasized that honest responses were most helpful; youth were encouraged to choose the answer that seemed to fit their experience. Youth reported the extent they thought their pre to post coping met one of the following responses, i.e., a lot worse, a little bit worse, about the same, a little bit better, or a lot better. Facilitators also asked youth to report how frequently they were able to use positive coping behaviors during the time they were participating in the program. Positive coping behaviors are activities that youth choose to deal with their stress that are likely to make the situation better. For example, thinking positive thoughts or playing sports may help some youth reduce their stress. Negative coping behaviors such as hitting a window or yelling at someone are likely to make the situation worse. Youth circled a positive coping frequency estimate of not very often, sometimes or often.

These simple questions were included in the midst of developing the program. This was primarily because the

A-COPE, used in early YES program measurement, seemed to rely on youth implementing a wide array of coping measures to score higher in coping (83). Since the YES program focused on implementing one to three youth-selected coping behaviors regularly, combined with relatively short programming time and the fact that the A-COPE scale did not include computer and online coping behavior choices, this simplified coping measurement appeared to capture youth ideas about their coping.

RESULTS

Forty-six of fifty-two youth, ages 10–16, completed the YES program after development of the 2012, or third version, of the K-MIR scale. Reasons for non-completion were schedule conflict with team sports ($n = 1$), second parent withdrew consent ($n = 1$), not interested in continuing ($n = 2$), moved out of area ($n = 1$), and not known (n=1). **Table 2** notes the demographic composition of the YES participant youth. More than half of the sample were non-Caucasian. Most participants were in grades 6–8. About 59% of the sample reportedly had a relative with a mental illness, most frequently a parent with reported diagnoses of depression, bipolar disorder, or borderline personality disorder. Over 10 percent of the youth reported had a parent with a dual diagnosis of mental illness plus substance abuse (primarily alcohol abuse). Notably, parents reported that over 28% of the youth also had a mental health diagnosis, most commonly ADHD, anxiety, and depression.

TABLE 2 | Demographic and household details of the YES program sample ($N = 46$).

Demographic	% or mean
Female	63.00%
Male	32.60%
Age	13.07 years (SD 0.82)
RACE	
Black	21.30%
White	48.90%
Latino/Hispanic	12.80%
Mixed Race/Other	17.10 %
GRADE	
6th	19.60%
7th	8.70%
8th	69.60%
Missing	2.20%
REPORTED FAMILY MEMBER WITH DIAGNOSIS	
Parent	34.80%
Sibling	15.20%
Grand parent	4.30%
Aunt/Uncle	2.20%
Other family member	30.40%
None reported	30.40%
Missing	10.90%
Child reported to have a diagnosis	28.26%

Table 3 details the distributions of correct answers by item, subscale, and total scale; and statistical comparisons from pre to post intervention.

Youth reported how often they were able to use positive coping strategies across the program often (60.9%), sometimes (32.6%), and not very often (6.5%). Ninety three and a half percent of the youth said they were able to use positive coping strategies often or sometimes. Participant youth also rated their coping skills at the end of the program as compared to their skills at the beginning of the program; they said their coping skills were way better (51.1%), a little better (40.4%), and about the same (8.5%). Therefore, 91.5% of youth estimated their coping skills were way better or a little better from pre to post intervention.

The fidelity assessment revealed that fidelity objectives were met 94% of the time within the YES program delivery. For the 6% not met, it appeared that reduced session time available was a main barrier. Especially in the second semester and near holiday breaks, the schools sometimes shortened class hours to accommodate special activities such as pep rallies, standardized testing, and award ceremonies.

DISCUSSION

Data responded to the guiding research question: "What are the pre-post mental health literacy and coping outcomes reported by youth attending the Youth Education and Support program?" It appears that this sample of YES participant youth reported significantly increased levels of mental health literacy from pre to post program participation. Over 90% of the sample youth described the use of positive coping during the time they participated in the Youth Education and Support program. Over 90% of youth also reported improved coping skills from pre to post.

The YES mental health literacy outcomes align with the work of Kutcher et al. (64) who found participating in a mental health literacy program led to increased knowledge of mental health among Canadian high school students. The YES program youth participants showed less stigmatized assumptions from pre to post intervention. This result is similar to the work of Wahl et al. (66) in their implementation of a middle school mental health education curriculum. Since most mental health literacy programs are delivered to adults, this study may contribute to building knowledge about *youth outcomes* of mental health literacy programs. This is an important knowledge gap to address among mental health literacy program evaluations.

The Knowledge of Mental Illness and Recovery scale measuring levels of youth mental health literacy demonstrated promising psychometric properties including, reasonable model fit. Rasch analysis offers an approach to instrument development and validation that differs both conceptually and mathematically to the more commonly used classical test theory, where the aim is to measure across the range of the latent construct. Overall, the three subscales and the total scale score demonstrated adequate or good fit to the model. Reliability coefficients were generally fair for the subscales, and considered good for the total scale.

TABLE 3 | Repeated-measure *t*-test of KMIR by item, subscale, and total scale scores.

Items and scales	No. correct pre-program		No. correct post-program		*t*	*p*	Effect size	
	Mean	**SD**	**Mean**	**SD**			**Cohen's *d***	**95% CI**
1. Mental illness is more common than…	0.63	0.49	0.78	0.42	1.86	0.07	0.27	0.16 to 0.66
2. The most common mental illness is…	0.22	0.42	0.61	0.49	4.32	<0.001	0.63	0.27 to 1.11
5. Mental illness most often begins when a person is…	0.72	0.47	0.84	0.4	1.30	0.20	0.21	−0.21 to 0.61
6. About one of _______ people develop a mental illness…	0.44	0.49	0.87	0.34	5.93	<0.001	0.80	0.26 to 1.11
10. What percentage of people with mental illness gets help?	0.26	0.43	0.54	0.5	3.49	<0.001	0.48	0.10 to 0.93
16. The mental illness of a person is likely caused by…	0.38	0.48	0.41	0.5	0.83	0.41	0.06	−0.35 to 0.47
20. In the U.S.A., more people go to the hospital because…	0.4	0.5	0.44	0.5	0.00	1.00	0.06	−0.35 to 0.47
21. People with mental illness often do fairly well in life…	0.46	0.51	0.85	0.36	4.12	<0.001	0.64	0.14 to 0.97
22. Parents can develop a mental illness when their kids…	0.34	0.47	0.52	0.51	2.14	0.04	0.29	−0.11 to 0.71
23. If someone in your family has a mental illness…	0.62	0.49	0.83	0.38	2.66	0.01	0.38	−0.07 to 0.76
30. Most people with mental illness are likely to get…	0.42	0.5	0.63	0.49	1.94	0.06	0.35	0.07 to 0.76
36. Epilepsy, or having seizures, is a mental illness…	0.5	0.51	0.67	0.48	1.64	0.11	0.27	−0.15 to 0.67
Mental Illness subscale	**5.35**	**1.9**	**7.96**	**2.04**	**7.4**	**<0.001**	**1.09**	**0.69 to 1.58**
3. One of the first steps to recovering from serious mental…	0.5	0.51	0.67	0.47	2.07	0.04	0.30	−0.12 to 0.70
7. Recovery from mental illness means…	0.35	0.48	0.39	0.49	0.47	0.64	0.06	−0.34 to 0.47
9. A lot of things can help a person recover from mental illness…	0.57	0.5	0.7	0.47	1.35	0.18	0.20	−0.22 to 0.60
13. One of the best combinations of things that can help people…	0.35	0.48	0.63	0.49	2.91	<0.01	0.43	0.02 to 0.85
15. Most people with mental illness…	0.44	0.5	0.44	0.5	0.00	1.00	0.00	−0.41 to 0.41
17. Family members can help a person with mental illness by…	0.28	0.46	0.59	0.5	3.18	<0.01	0.47	0.07 to 0.90
19. Getting help for mental illness works as well as getting help…	0.46	0.5	0.57	0.5	1.09	0.28	0.16	−0.24 to 0.57
28. If a person is trying to get attention by saying he or she will…	0.85	0.36	0.94	0.25	1.43	0.66	0.22	−0.22 to 0.60
31. Mental illness keeps a person from being able to develop…	0.53	0.5	0.8	0.4	2.60	0.01	0.40	−0.05 to 0.77
32. It is very important to know the cause of a person's mental…	0.2	0.4	0.24	0.43	0.63	0.53	0.09	−0.32 to 0.50
33. People with mental illness are usually able to make…	0.52	0.51	0.8	0.4	3.28	<0.01	0.48	0.02 to 0.84
35. Most people with a serious mental illness get well quickly…	0.85	0.36	0.89	0.31	0.63	0.53	0.09	−0.33 to 0.49
Recovery subscale	**5.89**	**2.06**	**7.65**	**1.84**	**5.87**	**<0.001**	**0.87**	**0.40 to 1.25**
4. The main reason people do not get help for their mental…	0.4	0.49	0.65	0.48	3.52	<0.001	0.46	0.04 to 0.87
8. People with mental illness…	0.52	0.51	0.74	0.44	3.16	<0.01	0.47	0.03 to 0.85
11. Some people think that people with mental illness are…	0.53	0.5	0.91	0.29	4.38	<0.001	0.67	0.13 to 0.96
12. Stigma toward people with mental illness happens when…	0.5	0.51	0.72	0.46	2.22	0.03	0.33	−0.10 to 0.72
14. People with mental illness…	0.35	0.48	0.59	0.5	2.69	0.01	0.40	−0.01 to 0.82
18. Most people with mental illness…	0.44	0.50	0.70	0.47	3.60	<0.001	0.53	0.10 to 0.93
24. Mental illness may be caused by being an emotionally…	0.33	0.47	0.61	0.49	2.93	<0.01	0.43	0.03 to 0.85
25. People with a mental illness are usually more dangerous…	0.67	0.47	0.89	0.32	2.66	0.01	0.40	−0.07 to 0.75
26. It is easy to tell if someone has a mental illness…	0.67	0.47	0.87	0.34	2.45	0.02	0.37	−0.09 to 0.74
27. People with a mental illness usually can't think well…	0.54	0.5	0.87	0.43	2.49	0.02	0.56	0.11 to 0.94
29. People with mental illness are more likely to be the victim…	0.48	0.51	0.37	0.49	−1.09	0.28	−0.16	−0.57 to 0.25
34. A person recovering from a mental illness is usually better…	0.7	0.47	0.87	0.34	2.43	0.02	0.35	−0.10 to 0.72
Stigma subscale	**6.12**	**2.23**	**8.67**	**2.1**	**7.14**	**<0.001**	**1.05**	**0.58 to 1.45**
Total KMIR scale	17.35	4.65	24.28	4.8	9.94	<0.001	1.47	1.03 to 1.95

Full items can be made available by the first author. For completeness, changes in score by item have been included, however interpretation is recommended at the subscale and total scale level. Cohen's d effect sizes calculated after correcting for the correlation between pre and post-program scores, as per (84).

Item redundancy were not found to be an issue for any item pair across all KMIR subscales. Given the relatively small number of items in each scale, this is somewhat expected, as the probability of redundant items increases with the number of items given that there are a finite number of ways in which a single construct can be represented in written language. However, even small numbers of items are not immune from the effects of redundancy, with items that are too similarly worded or of very similar difficulty likely to result in correlated residuals after accounting for the latent constructs. As noted earlier, differential item functioning was not explored given the sample size. Local independence was observed for several item pairs; however, given that there were no significant gains to be had from the removal of dependent items, all were retained.

There remain a number of areas for improvement, and it is important to note that the KMIR scale is still under development. Data collection is underway to measure test-retest and test convergence analysis of the KMIR compared to the Wahl et al. (66) knowledge scale. The development of scales with adequate psychometric properties is a key need for the development of evidence-based mental health literacy programs for youth.

The YES program does provide an early response to the requests for more mental health information echoed among youth that have a parent or other family member with a mental health disorder (57). The active learning format of the YES program and the readability of the KMIR scale seems aligns with the recommendation of parent mental health consumers for delivering mental health literacy programs that are fun and developmentally appropriate (85).

The YES evaluation includes youth-estimated coping skills is useful in beginning to collect behavioral change data. This is important since resiliency and coping theories include a need for knowledge and behavioral foci. The YES small group format may have provided an additional support network for the youth. Support networks are key components within resiliency and coping theories (11, 49).

However, it is not possible, even for this sample of youth, to know the extent that participation in a mental health literacy program may serve as a developmental factor in the prevention of mental health disorders or their reduction in symptoms severity. The YES program participant sample is relatively small and the KMIR scale is still undergoing psychometric testing. Family mental health disorders reported by parents were not subject to professional confirmation. Most critically, the study did not use a control group. It is likely that having the YES qualitative data analyzed by outside reviewers may have enhanced findings validity; an audit of the data using the trails of a codebook and raw data would also have contributed an external review of analyses (86). The findings cannot be generalized to all children of a parent or other family member with a mental illness.

There is a need for more funding, including long-term evaluation of mental health programs for youth. Specialized curricula may be needed for children that have a parent or other family member with a mental health disorder (4). The voices of youth, parents, and other family members should be included in the program development and evaluation. Intergenerational dialogue among family members may be helpful (87). Future research should include studies with matched comparison or control groups. The use of a randomized wait list controls may be an option. The inclusion of additional standardized scales to examine youth, parent, and family functioning would provide a stronger picture for assessing pre, post, and follow up intervention.

Mental health disorders one of the most common sources of disability in the world and mental health literacy may be able to build human developmental resiliency. Therefore, federal and state governments, as well as private funders of mental health research, should fund mental health literacy programs. Seeking this funding will require policy advocacy on the part of community members (30). It makes sense to build and test mental health literacy programs for youth, parents, and intergenerational family members (24, 88) to explore what works for whom, when, how, and for how long.

Despite many limitations, this study offers hope in findings that youth participating in a mental health literacy program reported increased levels of mental health literacy and coping skills. The target group was largely composed of youth that reportedly had a parent with a mental health disorder. The work is likely a small step of progress toward reducing the knowledge gaps for building evidenced-based mental health literacy programs for youth at risk of acquiring a mental health disorder. To do so, offers even more hope for a future world where mental illness is discussed respectfully, professional help is forthcoming, and accurate information about mental health is widely available.

ETHICS STATEMENT

The study was carried out in accordance with the recommendations of the human subject guidelines of the Michigan State University Institutional Review Board. The protocol was approved by the committee. All subjects gave assent (if minors) and their parents gave written consent.

AUTHOR CONTRIBUTIONS

JR lead the project and worked on all sections of the paper. SC author led the norming of the scale and wrote **Tables 1–3**. DC wrote the background section on scales and the information on resiliency and also coded all the data from which the paper was developed. CG wrote the background on children at risk and added to the discussion section. All authors helped with reviewing drafts.

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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

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Introduction: Offspring of parents with severe mental illness have an increased risk of developing mental illnesses themselves. Familial high risk cohorts give a unique opportunity for studying the development over time, both the illness that the individual is predisposed for and any other diagnoses. These studies can also increase our knowledge of etiology of severe mental illness and provide knowledge about the underlying mechanisms before illness develops. Interventions targeting this group are often proposed due to the potential possibility of prevention, but evidence about timing and content is lacking.

Method: A large, representative cohort of 522 7-year old children born to parents with schizophrenia, bipolar disorder or controls was established based on Danish registers. A comprehensive baseline assessment including neurocognition, motor functioning, psychopathology, home environment, sociodemographic data, and genetic information was conducted from January 1, 2013 to January 31, 2016. This study is the first follow-up of the cohort, carried out when the children turn 11 years of age. By assessing the cohort at this age, we will evaluate the children twice before puberty. All instruments have been selected with a longitudinal perspective and most of them are identical to those used at inclusion into the study at age 7. A diagnostic interview, motor tests, and a large cognitive battery are conducted along with home visits and information from teachers. This time we examine the children's brains by magnetic resonance scans and electroencephalograms. Measures of physical activity and sleep are captured by a chip placed on the body, while we obtain biological assays by collecting blood samples from the children.

Discussion: Findings from the VIA 7 study revealed large variations across domains between children born to parents with schizophrenia, bipolar and controls, respectively. This study will further determine whether the children at familial risk reveal delayed developmental courses, but catch up at age 11, or whether the discrepancies between the groups have grown even larger. We will compare subgroups within each of the familial high risk groups in order to investigate aspects of resilience. Data on brain structure and physical parameters will add a neurobiological dimension to the study.

Keywords: parental schizophrenia, parental bipolar disorder, early signs of mental illness, longitudinal cohort, psychopathology, MR scanning

INTRODUCTION

For decades, familial high-risk studies have shown that individuals who are born to parents with schizophrenia and affective disorders have a higher risk of developing mental disorders and neurocognitive impairments than non-predisposed individuals (1–3). Cohort studies of children with familial risk with assessments carried out over several time points in their lives allow us to study the early processes that precede illness manifestation. Such studies increase our understanding of the etiology of these complex diseases, and they give us an excellent starting point for developing a specialized and focused preventive approach in terms of early intervention for high risk individuals.

A meta-analysis concluded that ∼55% of all familial high risk (FHR) children will experience some kind of mental illness during early adult life (1), and a third will have a severe mental illness (SMI). Offspring of parents with SMI have a higher risk of developing the same disorders as their parents, but also of developing other mental disorders (4). Former FHR studies have reported neuro-integrative problems, social impairments, deficits in attention, information processing and other neurocognitive functions, poorer neuromotor functions and early symptomatology such as anxiety, sleep and mood disorders (2, 3), among offspring of parents with SMI. Moreover, in the premorbid phase the early signs of later schizophrenia (SZ) or bipolar disorder (BD) can present themselves already in childhood as e.g., anxiety, depression, or other non-psychotic disorders (5, 6). The first results from The Danish High Risk and Resilience Study—VIA 7 confirmed this by showing that already by age seven, children born to parents diagnosed with SZ and BD present much higher rates of a psychiatric diagnosis as well as cognitive deficits (only FHR SZ) across several domains, and motor problems (7–10). However, research is sparse when it comes to the question whether these abnormalities or developmental delays diminish over time, indicating that the individuals only suffer from those deficits and symptoms in a transitory manner and then catch up compared with the typically developing children, or whether the problems become even worse. A comprehensive review (11) synthesizing the data of neurocognition in individuals with FHR of schizophrenia concluded on the basis of 30 studies and nine cognitive domains that mild cognitive deficits (i.e., intermediate/between healthy controls and individuals diagnosed with schizophrenia) are reliably present in young FHR individuals, and that longitudinal studies should aim to elucidate the trajectories of cognitive changes to improve early intervention strategies. Concerning individuals at FHR of bipolar disorder there is evidence that neurocognitive deficits also play a role (12), although studies are smaller and tests are seldom repeated at different ages. It is therefore relevant to carry out careful examinations of early signs or symptoms of mental disorders among high risk children several times before puberty to learn more about these individuals and the trajectories of their emerging symptoms or difficulties.

Abbreviations: ADHD, attention deficit and hyperactivity disorder; BASYS, The Bipolar and Schizophrenia Young Offspring Study; BD, bipolar disorder; DNA analytic, deoxyribonucleic acid; FHR, familial high risk; FHR-BP, familial high risk for bipolar disorder; FHR-SZ, familial high risk for schizophrenia; FORBOW (study), Families overcoming risks and building opportunities for well-being; GWAS, genome wide association analyses; MANOVA/MANCOVA, multivariate analyses of variances/covariance; MR/MR scan, magnetic resonance scanning; PLEs, psychotic like experiences; PTSD, post-traumatic stress disorder; SIPS/SOPS, Structured Interview for Prodromal Syndromes and Scale for Prodromal Symptoms; SZ, schizophrenia.

Puberty is a period characterized by massive changes in brain structures and connectivity as well as changes in physical appearance, hormonal status and psychological and social constitution (13, 14). It is also a period with high incidence rates for mental disorders and for some, it is a period with changes of behavior, including risk taking activities and new relational patterns, for example higher degrees of independency. From a developmental perspective it is a period in life that is of high importance, but also very complex to study since both age and hormonal status as well as social and psychological aspects matter when comparing individuals in e.g., a cohort. Social mechanisms like bullying or other forms of social defeat that take place in childhood and early adolescence are frequently reported and have systematically been shown to be directly related to later emergence of mental illnesses like depression (15) and other negative life outcomes. Attributional style or coping, however, have been less investigated and may be of importance.

Brain changes in schizophrenia are present in drug naïve adult patients, and the strongest risk factors exert their influence already in the pre- or perinatal period (16, 17). Lately, the study of endophenotypes and biomarkers has advanced the field of SZ and BD as well as our understanding of the neurodevelopmental nature of both disorders (18, 19). This approach will also allow for early identification and intervention of serious mental illness (20). Despite the fact that only populations of young individuals before the mean age for the onset of the disorder are genuinely "at high risk" (19), few studies to date have examined adolescents at risk with the same means of brain mapping, and none before the onset of puberty. Taken together, the existing literature suggests the study of familial high-risk individuals as fruitful for the understanding of correlates for vulnerability and resilience (compensatory mechanisms), but findings have been inconsistent. In particular, magnetic resonance scans (MR scans) before puberty will permit the study of brain changes during early disease formation and provide important information on differences between individuals with and without familial risk.

Patients with SZ and BD have a higher risk of *somatic comorbidity* than the background population, and they have higher mortality rates due to medical diseases and untreated physical conditions (21). It is likely that early markers of physical illness can be traced already in childhood, either because of common life style factors in the family and/or because of a shared genetic vulnerability to mental disorders and physical illnesses, but evidence is limited. It is therefore relevant to examine early markers of evolving physical illness in children with FHR for mental illness and to analyze whether variations can be explained solely by differences in life style factors such as nutrition, environment, sleep, and physical activity. Also, very little is known about whether e.g., frequency of absence from school and the children's health service use differ between the groups. A Danish register study demonstrated, however, that children with FHR for SMI are more likely not to complete elementary school or to score lower grades than children without a predisposition for mental illness (22).

Family functioning is very often influenced when one of the parents (or both) are suffering from a severe mental illness, also in cases when the child does not live with the affected parent, but only with the co-parent (23). The daily routines can suddenly be disrupted by behavior or needs due to the mentally ill parents and resources, both materially, socially, culturally, and emotionally may be scarce. Dysfunctional family patterns are often seen in several dimensions of family life such as problem solving, communication, role functioning, affective responsiveness, affective involvement and behavior control in families with parental mental illness (24). These dimensions are thoroughly described by the authors behind Family Assessment Device, a self-administrated and widely used questionnaire based on social and system theoretical principles to measure family functioning [FAD (25)].

Briefly, the first *results from the VIA 7 study* showed considerable associations between parental mental illness and several domains of child functioning. Children born to parents diagnosed with schizophrenia showed markedly impaired neurocognitive functioning on the majority of applied measures; i.e., processing speed, working memory, executive functioning, visuospatial functions, sustained attention, and declarative memory (7). At the same time, we found an increased risk of displaying mental health problems or even mental disorders among both groups of children with familial high risk, but in particular among the children with familial high-risk of schizophrenia. Of children with FHR for SZ 38.7% had a life time diagnosis, while this was found in 35.6% of the children with FHR for BD, and in 15.2% of the controls. Also dimensional measures of psychopathology showed this (8). Especially anxiety, ADHD, and stress and adjustment disorders were more prevalent in both of the FHR-groups. Children with FHR for SMI also showed impairments in motor functioning (9) and in sustained attention and interference control (10). Further, children with FHR for both mental disorders were found to be at a greater risk of growing up under low socioeconomic status and reported more adverse life events than the controls based on interviews with the caregivers (unpublished data). Results further indicate an increased risk for living in a home environment with insufficient levels of stimulation and support for children predisposed for SZ and to a lesser extent also BD, and also social cognition and language development was found to be impaired in the FHR-SZ group (manuscript in preparation).

Increasing our knowledge about both the risk and the protective factors that influence these children's development is important to improve our chances for identifying the most vulnerable groups of children. More knowledge concerning the underlying psychological and neurobiological mechanism will guide future developments of early interventions with a preventive approach.

AIMS

In a cohort of children with familial high risk for schizophrenia or bipolar disorder our aims are:

1) To improve insight into early disease processes in schizophrenia and bipolar affective disorder including symptom formation, impairments or delays of maturation in different domains of cognitive functioning incl. social

cognition, paralleled by difference of brain structure and of patterns of brain activation compared to controls.

2) To investigate the development over time (i.e., from age 7 to age 11) of children with familial high risk compared to controls in the domains of neurocognition, psychopathology, social cognition, motor function and adverse life events and to identify and describe possible associations between familial high risk and factors that increase resilience in children.
3) To identify the influence of genetic and environmental exposures and their interactions.
4) To identify early amendable risk factors such as lack of stimulation and support, traumatic events during childhood, insufficient parenting, low socioeconomic status, neurocognitive and social cognitive deficits, and early, subtle signs of psychopathology.
5) To improve knowledge about physical health status among children with familial high risk for severe mental illness.

DESIGN

The Danish High Risk and Resilience Study is a representative nationwide cohort study consisting of 522 children born to parents with schizophrenia, bipolar disorder or neither of these disorders. The participating families were recruited from Danish registers and investigated thoroughly during 2013–2016 when the children were seven years old. The first assessment is referred to as the VIA 7 study, for details please see study protocol for the VIA 7 study (26).

The cohort consists of 522 individuals, now 11 years old (see **Figure 1**):

a) 202 children with at least one parent diagnosed with schizophrenia spectrum psychosis
b) 120 children with at least one parent diagnosed with bipolar disorder
c) 200 children with neither of the parents treated in mental health services for the above diagnoses.

The children of parents with SZ and controls were matched on municipality, sex and exact age of the child. The representative cohort is based on data from The Danish Civil Registration System (27) linked to the Danish Psychiatric Central Research Register (28). Saliva from the children and blood samples from the parents were used for genome wide association analyses (GWAS, PsychChip). The children and their parents were thoroughly examined with interviews, neurocognitive, social cognitive and motor tests, questionnaires, home visits, and observations. Assessments were supplemented with data from questionnaires sent to school teachers. Outcomes for the children were assessed thoroughly in the domains of neuromotor functioning, neurocognitive, social cognitive functioning, social functioning, and psychopathology. Also, parents were interviewed about their mental health, and data on their neurocognitive as well as social cognitive functioning was collected.

Compared to previous studies this cohort is unique due to the very large representative sample of children, all of the same age at the time of assessment, which allows inference about development in the repeated cross-sectional examinations and ultimately it will allow us to follow developmental pathways in the longitudinal design. By including genetic material in combination with thorough estimates of environmental factors, measures of illness severity, and level of functioning of the parents we have a unique possibility of investigating the gene-environment interplay.

The full assessment battery in the VIA 7 study lasted ∼3 days (5–6 h per day), as does the VIA 11 study-battery and the vast majority of the families completed the whole battery in the VIA 7 study. Parents were offered feedback on their child's performance, all participants received a gift card, and all families were informed that a follow-up at age 11 was planned.

METHODS

In the VIA 11 study we are aiming at re-assessing all children exactly 4 years from the date when they were included in the VIA 7 study. The adult who is taking care of the child on a regular basis and who is registered with the same address as the child is invited to give information (i.e., interviews and questionnaires) about the child's actual well-being and behavior ("primary caregiver"). This is often but not always one of the biological parents. The completion of the entire test battery lasts at least three days, and when organizing the appointments

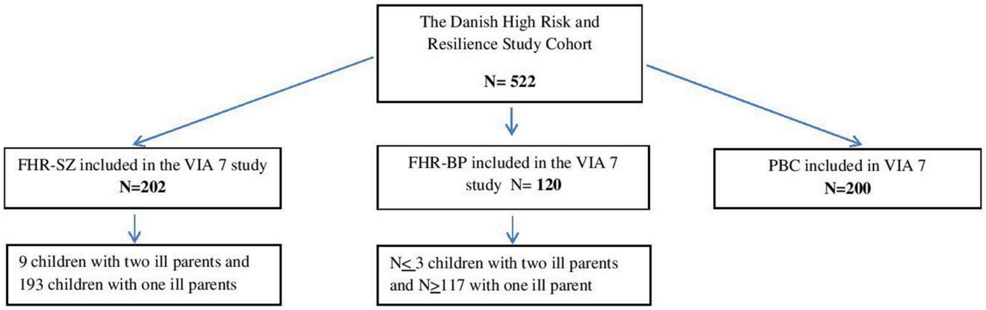

FIGURE 1 | Flowchart of the Danish High Risk and Resilience Study illustrating all children being assessed in the VIA 7 study and who are now being assessed again.

all kinds of flexible arrangements are being made to meet the families' specific needs without compromising the quality of the data collection.

All outcome measures are examined with validated instruments, specifically developed and selected for this age group, sensitive to small changes and suitable for later follow-up (**Tables 1**, **2**). We will take advantage of the fact that most variables will be measured twice, making analyses of developmental predictors and associations possible between level of functioning at age seven and age 11, e.g., by multivariate analysis. Moreover, our analytic strategy will capitalize on the fact that the genetic exposures can be modeled directly with polygenic risk scores. Analyses will include measurement of the current environment to which the child is exposed. For each of the outcome domains listed below, we will analyse

1) the differences between the three groups of children at age 11,
2) the differences in associations of outcomes from age seven to age 11 in the three groups
3) the extent to which direct measures of environmental factors (e.g., home environment) and direct measures of genetic risk (e.g., polygenic risk scores for SZ and BD) are responsible for variations in outcomes and development.

The analyses will thematically be divided into groups and will be carried out by Ph.D. students and postdoc researchers. Researchers are based in two centers, one in Copenhagen (Research Unit at Mental Health Center Copenhagen, Gentofte Hospital) and one in Aarhus (Psychosis Research Unit, Aarhus University Hospital Risskov) and have close collaboration at all levels including regular face-to-face meetings and video conferences. The outcomes are organized in the domains listed in **Tables 1**, **2** and described below.

DOMAINS AND INSTRUMENTS

Neuromotor Function

Fine motor speed is being assessed with Finger Tapping Test (30), while manual dexterity, ball skills and balance are assessed with Movement Assessment Battery for Children-2, Movement ABC-2 (29). Physical activity will be measured by a sensor in an easily wearable adhesive patch [SENS motion® (31)] which can directly measure sleep disturbances and level of physical activity during a 1-week observation period.

Neurocognitive Function

The comprehensive test battery includes Word Selective Reminding and Memory for Stories [TOMAL-2 (33)], Rey Complex Figure Test and Recognition Trial (34), Rapid Visual Information Processing [CANTAB (35)], Conners' CPT-II [Continuous Performance Test II (36)], Verbal Fluency 1–3 and Trail Making Test 2–4 [D-KEFS (38)], Symbol Search and Coding [WISC-IV (39)], Stockings of Cambridge, Intra-Extra Dimensional Shift, Spatial Recognition Memory, Spatial Span, and Spatial Working Memory [CANTAB (35)], Letter-Number Sequencing and Arithmetic [WISC-IV (39)], Cambridge Gambling Task [CANTAB (35)], and Reynolds Intellectual Screening Test [part of the RIAS (43)].

Social cognition is measured by Animated Triangles (41), Beeds Test (44) (tendency to jump to conclusion), Frederic's Stories (42), and the Social Cognition paradigm from the Human Connectome Project (78).

Psychopathology

General psychopathology and psychotic-like experiences (PLEs) will be examined with the diagnostic interview K-SADS-PL (45) including a specialized assessment of sub-threshold psychotic-like experiences (79). Magical Thinking Questionnaire (47) is included to ensure optimal information about tendency to magical ideation that is not covered by the K-SADS-PL-interview.

The aberrant salience-hypothesis (80) suggests that patients with schizophrenia have deviances in attributional style. Results from the VIA 7 study showed that familial high-risk children more often reported being bullied (unpublished results), which could be a sad truth, but it could also be partly explained by deviances in attributional style. We have developed and included a predefined social situation in a virtual reality scenario that will help us identify any liability to social anxiety and paranoid ideas. By using a virtual reality scenario, we will be able to investigate if a subgroup of the children is more inclined than the others to appraise an ambiguous social situation as threatening or bullying using a virtual reality scenario. We will apply a Danish children's version of the State Social Paranoia Scale (55), which asks about positive, neutral and paranoid appraisal of the virtual reality environment.

Executive functioning including affective regulation and flexibility will be assessed with the questionnaire Behavior Rating Inventory of Executive Function [BRIEF (51)] from both caregiver and teacher. Autism spectrum traits are evaluated with Social Responsiveness Scale [SRS (59)] also completed by the child's caregiver and the teacher. Dimensional measures of psychopathology will be covered with Child Behavior Checklist [CBCL (48)], ADHD rating scale [ADHD-RS (49)], and Strengths and Difficulties Questionnaire [SDQ (57); both by parent and teacher] while the ratings of the clinical impression of the child during the testing are registered with the Test Observation Form [TOF (81)].

Social Functioning and Behavior

Self-esteem will be coved by "I think I am"-questionnaire [Sådan er jeg (53)]. Social development is captured by the Vineland-2 Parental interview (58) and with the Strengths and Difficulties Questionnaire [SDQ (57)]. Data from the school will also be included via questionnaires sent to the teacher (see **Table 1** for details). Bullying is included with the questionnaire developed by Olweus for the child itself (54). Resilience is measured by a short version of Child Youth Resilience Measurement [CYRM (56)] as well as by associating the life situation and context of the child with their course of psychopathology.

TABLE 1 | Domains and instruments used for testing the children at age 11 in the Danish High Risk and Resilience Study VIA 11.

Domains	Outcomes	In VIA 7	Child	Primary caregiver	Teacher	Type of instrument
Neuromotor and physical measures	Motor development and milestones	Yes	Movement ABC (29)			Test in clinic
	Anthropometry	Yes	Height, weight, waist			Observations in clinic
	Motor speed and dexterity	Yes	Finger tapping (30)			Test in clinic (or home)
	Physical activity and sleep	No	SENS chip (31)	Anamnestic interview		Chip on tight for one week (32) Interview with primary caregiver
Neurocognition	Verbal Memory and visual memory	Yes	Word Selective Reminding and Memory for Stories from Tomal-2 (33) RCFT (Rey Complex Figure Test and Recognition Trial (34) and Spatial Recognition Memory from CANTAB (35)			Test in clinic or at home
	Attention	Yes	RVP (Rapid Visual Information Processing;3-5-7 mode) from CANTAB (35). Conners CPT (Continuous Performance Test (36)			Computer test
	Communication and pragmatic/social interaction	Yes			CCC-2 (Children's Communication Checklist-II) (37)	Questionnaire
	Speed of Processing	Yes	Verbal Fluency 1-2 and Trail making Test 2-4 from D-KEFS (38) and Symbol Search, and Coding test from WISC-IV (39)			Test in clinic or home
	Executive functions (planning and flexibility)	Yes	SOC (Stockings of Cambridge) and IED (Intra-Extra Dimensional Set Shift) from CANTAB (35) and Verbal Fluency 3 from D- KEFS (38)			Computer test
	Executive functions (visual and verbal working memory)	Yes	SSP (Spatial Span) and SWM (Spatial Working Memory) from CANTAB (35) and Letter-number Sequencing and Arithmetic from WISC-IV (39)			Computer test and test in clinic or home
	Executive functions (error monitoring)	Yes	Flanker Task (40) – before and during fMRI			Computer test
	Social cognition	Yes No	Animated Triangles (41) before and during fMRI (without the goal directed animation) Frederic's stories (42)			Computer test Practical test
	Intelligence	Yes	RIST (Reynolds Intellectual Screening Test) (43)			Test in clinic or home
	Decision making	No Yes	Beads' Test (44) Cambridge Gambling Task from CANTAB (35)			Computer test
Psychopathology	Psychiatric symptoms, incl. depression, anxiety, psychotic symptoms, thought disorders, PLEs, obsessive-compulsive symptoms, eating disorders, sleep disturbances, self harming behavior and traumatic life events	Yes Yes No Yes	Kiddie-SADS-PL interview (45) PLE assessment (46) Magical Ideation Questionnaire (47)	Kiddie-SADS-PL interview (45) Anamnestic interview CBCL (48) (Child Behavior Checklist)	TRF (48) (Teachers Rating Form)	Interview (categorical psychopathology) Questionnaires, (dimensional psychopathology)

(Continued)

TABLE 1 | Continued

Domains	Outcomes	In VIA 7	Child	Primary caregiver	Teacher	Type of instrument
Do	Attention/hyperactivity	Yes		ADHD-Rating Scale (49)	ADHD-Rating Scale (49)	Questionnaire
	Affect regulation/flexibility	Yes	CEMS (50) (Children's Emotion Management Scale)	BRIEF (Behavior Rating Inventory of Executive Function (51)	BRIEF (Behavior Rating Inventory of Executive Function (51)	Questionnaire
Do	Anxiety	Yes	STAIC (State-Trait Anxiety Inventory for Children (52)			Questionnaire
Social functioning and behavior	Self-esteem	Yes	Sådan er jeg' ['I think I am' (53)]			Questionnaire
	Bullying	No	Olweus Bully/Victim Questionnaire (54)			Questionnaire
			SSPS (State Social Paranoia Scale (55)			Questionnaire after Virtual Reality scenario
	Resilience	No	CYRM (Child and Youth Resilience Measure short version (56)			
	Social Functioning	Yes		SDQ (Strengths and Difficulties Questionnaire (57)	SDQ for teachers (Strengths and Difficulties Questionnaire (57)	Questionnaire (emotional symptoms, conduct problems, hyperactivity/ inattention)
		Yes			CCC-2 [Child Communication Checklist (37)]	
	Social development	Yes		Vineland Adaptive Behavior Scales –II (58)		Interview
	Autism spectrum traits	Yes		SRS−2 (Social Responsiveness Scale (59)	SRS-2 (Social Responsive ness Scale (59)	Questionnaire
Environment and emotional climate	Stimulation and support in actual rearing environment	Yes	HOME Inventory, Early Adolescent version (60)	HOME inventory, Early Adolescent version (60)		Interview made in the home with both child and parent
	Life events and trauma	No	CTS [Childhood Trauma Screener (61)]	Anamnestic interview focusing on age 7-11		Questionnaire and semi-structured interview
	Perceived support from social network	Yes		SPS [Social Provision Scale (62)]		Questionnaire
	Attachment style	Yes, but new test	Secure Base Script Test (63)			Test in clinic or home
	Expressed emotions/emotional family climate/familiar relations	Yes No		FMSS [Five Minute Speech Sample (64)] FAD [Family Assessment Device (25)]		Interview Questionnaire
	Stress	Yes	Hair test for cortisol Items from DLSS [Daily Life Stressor Scale (65)]			Hair sample Questionnaire
	Use of social media	No		Anamnestic interview		Questionnaire

(Continued)

TABLE 1 | Continued

Domains	Outcomes	In VIA 7	Child	Primary caregiver	Teacher	Type of instrument
Physical health	Puberty status	No	Tanner stages (66, 67)			Illustrations
			Hormone level			Blood sample
	Physical health	No	HbA1c. leucocytes, CRP			Blood sample
			Exercise on bicycle,			Test (Copenhagen only)
Genetic and epigenetic analyses	Polygenic risk scores	Yes	Dry blood spots from Danish Neonatal Screening Biobank and blood samples			Day 1, hospital
	Inflammatory and infectious markers.	Yes	Dry blood spots from Danish Neonatal Scrbeening Biobank and blood samples			Day 1, hospital
Brain scan	Functional and structural MRI	No	MR Scanning and EEG			Day 2, Scan at hospital
	Electrophysiology	No	The Copenhagen Psychophysiological Test Battery (CPTB) (prepulse inhibition, P50 gating, mismatch negativity)			Day 2, before the scan at hospital

TABLE 2 | Instruments for the assessment of the parents' mental health status, actual level of functioning and parenting issues in the Danish High Risk and Resilience Study - VIA 11.

Domains	In VIA 7	Parent/actual caregiver	Co- parent	Type of test
Mental health status (previous 4 years)	Yes (lifetime)	SCAN [Schedules for Clinical Assessment in Neuropsychiatry (68)] for the person him/herself and the other parent	do	Interview
Daily Functioning	Yes	PSP [Personal and Social Performance Scale (69)]	do	Interview
Actual state of illness	Yes	SANS [Scale for the Assessment of Negative Symptoms (70)]	do	Interview
Do	Yes	SAPS [Scale for the Assessment of Positive Symptoms (71)]	do	Interview
Do	Yes	Hamilton Rating Scale for Depression (72)	do	Interview
Do	Yes	YMRS [Young Mania Rating Scale (73)]	do	Interview
Affective regulation	Yes	ALS [Affective Liability Scale (74)]	do	
Family functioning	No	Family Assessment Device, short version [FAD-12 (25)]		Questionnaire
Perceived support from social network	Yes	Social Provision Scale [SPS (62)]	do	Questionnaire
Social Response, adult	No	Social Responsiveness Scale -Adult, self-report [SRS-A (75)]	do	Questionnaire
Relation to child and relation to other parent	Yes	FMSS [Five Minute Speech Sample (64)]		Interview (recorded and transcribed)
Adverse childhood experiences	No	ACE Study Questionnaire [Adverse Childhood Experiences Questionnaire (76)]	do	Questionnaire
Attachment style	Yes	PAM questionnaire (77)	do	Questionnaire
Knowledge about mental illness and talking to children about mental illness if relevant	No	Included in anamnesis		Interview

Environment

Familial environment in the home is measured by the use of a semi-structured interview, HOME, that must take place at a home visit with the caregiver and the child being present at the same time [Home Observation for Measurement of the Environment (82)]. We chose the version called Early Adolescence (EA-HOME) which is appropriate for children from age 11. An anamnestic interview with the primary caregiver includes sociodemographic data, data on the child's health service use, adverse life events, school performance and leisure activities as well as social network. Childhood trauma is measured directly from the child through the section in the K-SADS-PL (45) about post-traumatic stress disorder (PTSD) combined with the Childhood Trauma Screener [CTS (61)]. Attachment style categorization will be based on Secure Base Script Test [SBST (63)] while level of stress will be captured by Daily Life Stressor Scale [DLS (65)] and from level of hair cortisol in a hair sample.

The parents' level of daily functioning is measured with Personal and Social Performance Scale [PSP (69)] and is used as a measure of the environment together with Five Minute Speech Sample (64), which contains data about the primary caregiver's impression of the relationship with the child.

Physical Health Status

We will describe the children's physical condition including a long range of tests relevant for risk of physical illness at age 11. This will include: VO_2max, anthropometry, Body Mass Index, white blood cells, C-reactive protein, HbA1c, and level of cortisol in hair samples. Puberty status will be determined by analyses of sex-hormones in blood tests and with the Tanner test (66, 67), which is letting the child look at drawings of different stages of puberty, and choose which stage is closest to the child's actual development.

Genetic and Epigenetic Analyses

Saliva and peripheral blood are being sampled from the children and will be analyzed with DNA-analytic results from the dried blood spot from birth and from saliva-sample results collected at age seven with the purpose of studying epigenetic changes.

Structural and Functional Magnetic Resonance Imaging (fMRI) an Electroencephalography (EEG)

We will examine the children with anatomical and functional magnetic resonance imaging (MRI) of the whole brain at 3.0 Tesla at Aarhus University, Center for Functionally Integrative Neuroscience (CFIN) and Hvidovre Hospital's, Danish Research Center for Magnetic Resonance (DRCMR). We will acquire 3D T1-weighted (MP2RAGE) and high-quality diffusion weighted imaging (DWI) data to calculate a set of measures that will allow us to detect regional changes in brain microstructure and structural brain connectivity (DWI based tractography) across groups. The measures include fractional anisotropy, apparent diffusion coefficient, and diffusion kurtosis indices. These measures will be used to compare regional changes in brain microstructure, as indexed by diffusion weighted imaging (DTI) across groups. Functional MR scans will map task related changes in brain activity and connectivity during three experimental tasks: the Eriksen Flanker paradigm, which addresses cognitive control and motivation, the Social Cognition (Animated Triangles) paradigm from the Human Connectome Project (78) which tap into theory of mind, and a Self-Reference paradigm to probe the ability to relate external events to oneself.

It is our intention to use these MR-scans carried out at age 11 as a basis for identifying developmental trajectories of brain changes during disease formation as we plan to repeat the same MR protocol later in adolescence (at age 15 or 16). The trajectory in children developing schizophrenia is expected to include reduced maturation of inhibitory pathways and excessive pruning of excitatory pathways leading to altered excitatory–inhibitory balance in the prefrontal cortex. We also anticipate structural and functional alterations of the brain's connectivity. Although some data support each of these possible neurodevelopmental mechanisms for schizophrenia, no study has previously provided possibilities for identifying measures of abnormal structural brain maturation (such as less or excessive pruning, less maturation of major limbic/cognitive fiber tracts, abnormal global brain connectivity, dysfunctional processing), and differences in integration in key circuits that will be probed with task-based fMRI and EEG in young individuals at risk for those disorders before puberty. We plan to correlate the abundant biological, clinical, and neurocognitive information already collected at age seven to future imaging data to see whether earlier data are predictive of imaging outcomes.

In a subgroup of ∼50% of the participants, EEG will be performed at DRCMR to record functional task-related cortical activity during an auditory 40 Hz stimulation paradigm, a mismatch negativity paradigm, and a Flanker paradigm which is closely matched to the one used during functional MR to enable data integration. We selected these tasks based on consistent evidence for deficits in neural synchrony and event-related potentials (ERPs), evoked by using those paradigms in patients with SZ (83, 84) and, although to a lesser degree, in patients with BD (85). Reduced power of cortical activity and reduced amplitudes of event-related potentials in neural mechanisms are assumed to be related to both automatic sensory processes as to higher-order cognitive functions (86) and provide a window into some of the neurobehavioral symptoms (87). No study before has studied these neural correlates in a large sample of children at familial high risk for schizophrenia and bipolar disorder.

PROCEDURES AND PERMISSIONS

Contact and Procedures

All families have been informed by the completion of the first assessment in the VIA7 study that a follow-up was planned at age 11. To maintain contact with the participants of the cohort, Christmas cards and birthday cards have been sent to the children each year and a short survey consisting of two questionnaires [the "I think I am" (53) and the Child Behavior Check List CBCL (48)] was sent out to each family when the child turned 9 years of age; this survey is called the VIA 9 study.

As in the VIA 7 study, the first contact for the assessment is made by sending out a hard copy letter and an illustrative folder to the child's address (see **Figure 2**), followed by a phone call by the research coordinator, who invites the family to an information meeting. All parents give informed consent before data collection can start and for those who are divorced or separated and have joint custody over the child, informed consent from both parents is mandatory. We meet all families with a very friendly and flexible approach, taking into account any individual concerns or obstacles that they may have with regard to participation. The first results from the

VIA 7 study are available for those participants who ask for them.

The data collection takes place during 3–4 meetings, each lasting up to ∼5 h. The caregiver and the child are assessed simultaneously by two researchers, and whenever possible the assessor testing the child is kept blinded for the familial high-risk status and for the potential diagnosis from the first assessment at age seven to avoid any subjective influences. The first two days take place in the clinic, the third day is at the MR-scan facility, while the fourth day is in the family's home in order to complete the home environment assessment. However, some children may need more time or cannot concentrate sufficiently and thus need shorter but more frequent meetings. The child receives a gift card after each day completed and at the end of the whole assessment, the parents are offered a feedback on their child's performance and results. If any specific concern is raised on the basis of the interviews and testing, the researchers will guide the parents to relevant support or even make the referral or inform the social authorities or hospital service, if necessary. All diagnoses retrieved from the diagnostic interviews K-SADS-PL (45) and the SCAN interviews (68) are discussed and confirmed or declined at weekly clinical conferences, with a child and adolescent psychiatrist (A. Thorup) being present together with the research team members.

Permissions and Data Protection

The study was approved by the local Ethical committee (Protocol number: H 16043682) and the Data Protection Agency (ID-number RHP-2017-003, I-suite no. 05333).

The majority of tests that we used in the VIA 7 study were chosen at the outset (among other reasons) because they would also be suitable at age 11 and this is a considerable advantage when aiming to study relatively small changes over time (**Tables 1**, **2**).

FIGURE 2 | Lay out for the invitation to all families in the VIA 11 study.

Statistical Analyses

Several descriptive and inferential statistical tools will be applied to analyse the data. For example, for descriptive analyses, frequency distribution, mean or median, and graphical representation will be applied. For inferential analysis, linear regression and/or analysis of variance or covariance (ANOVA/ANCOVA) will be the standard method for analyzing the association between exposure variables (i.e., high-risk status) and continuous outcome measures (e.g., psychopathology). If a variable does not follow a normal distribution even after possible transformations, non-parametric methods (e.g., Kruskal–Wallis test) or non-linear statistical models can be an alternative approach. Moreover, to control for the Type I error probability as well as the correlation between multiple outcome variables, multivariate analyses of variances/covariance (MANOVA/MANCOVA) will be applied. Furthermore, dichotomous/count outcome measures will be analyzed with generalized linear models (e.g., logistic or Poisson regression). To account for longitudinal effect from VIA 7 to VIA 11 data, linear mixed effect model for the continuous outcome and the generalized estimating equation for the dichotomous outcome will be applied. Based on our specific research question, we may consider clinically meaningful interaction terms (e.g., high-risk status and other covariates) in the models. Furthermore, as we are collecting genetic data for children and parents, to account for genetic background, polygenic risk scores (e.g., for Schizophrenia, education) would be considered as a covariate in the statistical models. In the case of missing data, multiple imputations will also be considered given that data are missing at random (MAR). However, in case of participating families with lower functioning there might be the chance that missing data are not at random (i.e., non-ignorable missing), but solely due to the inability or unwillingness to complete instruments, test, or questionnaires. Thus, we will apply sensitivity analyses for investigating the possible violations of the missing at random assumption. For families not participating at all, we will have the opportunity to include data from Danish registers in the sensitivity analyses. To handle the latent factors like resilience and vulnerability, we will apply structural equation modeling (SEM) or different kinds of path analyses. In addition, multiple regression will be applied for analyses of predictors and associations from age 7 to age 11 regarding changes in mental and physical symptoms.

RESULTS

Results will be presented within the context of both cross sectional and longitudinal analyses, i.e., comparing the results from the first assessment at age seven with the results found at age 11. Cross sectional results will be reported in all new areas or instruments like MR scans, Virtual Reality setting, SENS motion data, and blood sample data, while developmental trajectories will be investigated for those measures that are repeated, i.e., most of the cognitive measures, psychopathology, motor development, as well as many of the questionnaires, and the home environment evaluation (see **Tables 1**, **2**).

Results concerning the actual psychopathology and the association to the mental health status at age seven will be investigated, including data on attributional style (tendency to report being excluded/bullied) and physical health. Data on social cognition will be linked to data on the developmental trajectories of Psychotic Like Experiences (PLE's; i.e., intensity, frequency, and severity) from age seven to age 11. Neurocognitive data will be analyzed to learn more about the extent to which these children's deficits or advantages measured at age seven remain stable, deteriorate or diminish over time. Brain scan data will focus on potential differences between high risk groups in terms of structural properties, neural networks, and connectivity.

DISCUSSION

This protocol describes the research strategy for the first follow-up of the unique familial high-risk cohort consisting of 522 children all the same age called The Danish High Risk and Resilience Study. This second wave is called the VIA 11 study since all children are now 11 years of age. The advantages of this cohort are especially that all children have the same age and that all families of the cohort have been recruited from national registries. Further, the comprehensive battery and the multidimensional approach will allow us to combine results from many research areas in order to add knowledge to the field of familial high risk, developmental psychopathology, brain development and risk, and resilience assessment.

The field of familial high risk studies is not new, actually the first study with this design was described and conducted by Barbara Fish (88) in New York in the early fifties, and from this study came the "pandysmaturation"-hypothesis, giving inspiration to the neurodevelopmental hypothesis of understanding the etiology of schizophrenia. In the field of schizophrenia research several familial high-risk studies have been conducted since Fish' study and recently reviewed by Hameed (89). However, we believe that the possibility of recruiting the participants from registries and not from clinics (i.e., biased toward including only those in treatment or with severe courses), with all children having the same age is a major advantage. From the perspective of neurodevelopmental psychopathology, age greatly matters when comparing results of cognitive tests, social and behavioral functioning and developmental milestones.

There are other important familial high-risk studies that are ongoing and should be mentioned here. One is the Canadian FORBOW study by Uher et al. (90) which is still recruiting both parents with schizophrenia, major depression and bipolar disorder and their offspring and testing various types of prevention and early interventions in the cohort, while also conducting annual assessments. The Spanish study led by Sanchez-Gistau (91, 92), called The Bipolar and Schizophrenia Young Offspring Study (BASYS), is a multi-center, naturalistic study which aims to evaluate psychopathology and neuropsychological and neuroimaging variables in child and adolescent offspring of patients with schizophrenia or bipolar disorder in two child and adolescent psychiatry departments in Madrid and Barcelona. The sample consists of 41 offspring of parents with schizophrenia, 90 offspring of parents with bipolar disorder and 107 controls. The age span is wide, 6–17 years and all participants are recruited via parents seeking treatment. The Dutch Bipolar Offspring Study led by Manon Hillegers in the Netherlands is a longitudinal fixed cohort study established in 1997 and followed up until early adulthood (93). It consists of 140 offspring from 86 families where one of the parents suffered from bipolar disorder. Many interesting results have been reported from this cohort, e.g., that the impact of life events especially in the early phases of the illness may be mediated by a passive reactive coping style. Further, a study led by Iacono et al. (94) has recent data on a smaller group of offspring of parents with bipolar disorder. However, all studies have much wider age ranges and smaller Ns.

Conducting a Longitudinal Study

Conducting a longitudinal study and aiming for several assessments of both parents and children over time is an ambitious and demanding project. Longitudinal studies must be well-prepared and sufficiently financially supported and the team behind the study must set a long-term plan, have a strong collaboration and develop clear and good ways of communication with regular meetings and other forms of contact. Research questions must be based on a theoretical framework and reflect current state of the art and must map onto the aims of the study. Recruitment procedures of participants and strategies for retaining participants in the study are crucial and must be planned and discussed with potential participants/pilot users. The assessor group must be properly trained, have regular meetings and good collaboration, and high levels of interrater reliability is essential. Data must be handled in accordance with rules of data protection and analyzed on the basis of a statistical, preset strategy. Finally, results must be published for both professionals and the population (95). Communication of results that reveal some aspects that could be interpreted as negative for the participants must be planned and handled with empathy.

Clinical Implications

Developmental trajectories will inform us on how the delays and abnormalities observed at age seven will evolve and influence the individual's risk of mental symptoms or reduced level of daily functioning at age 11. For example, much more knowledge is needed if we are aiming for understanding, treating or giving advice to children and their parents when children report psychotic like experiences (PLEs). Are PLEs at age seven just a normal, transient phenomenon that we as clinicians should not worry about? What is the impact if PLEs are found to correlate with an increased risk of psychopathology at age 11 and should persistent PLEs (i.e., found at both age seven and age 11) be considered as more severe early risk factor? We intend to investigate the nature of all possible PLEs mentioned by the children in the VIA 11 study in a very thorough way to be able to analyze data at a detailed level. Results from the VIA 7 study indicated that PLEs are much more common among FHR-SZ children (Ellersgaard et al, in preparation) and do correlate with poorer functioning, and this fact is important to follow very closely to help us learn more about their value as a risk marker for later mental illness, as proposed by others (96). A large cohort study reported that up to 2/3 of a population of school children aged nine to 11 would report some kind of PLEs on a self-report measurement, and for those individuals who reported the PLEs to be persistent after 2 years, a correlation with an increased risk for internal and external psychopathology later in life was found (96). However, methodological considerations are important, since other studies find much lower rates (97), for example the frequency was 5.9% based on a clinical interview in a large birth cohort of 12-year-old twins. In the VIA 11 study we initially considered using a self-report instrument, but concluded that due to the long test battery, the data collectors will have time to build a good relationship with the child and thus confidentiality will most likely be high, allowing the child to tell the interviewer if she or he has had any PLEs. Still, a well-validated rating scale for PLEs among children and adolescents with a clear and informative manual would be a considerable contribution to the field for the future. An adapted version of the SIPS/SOPS instrument (79) could be a place to start.

In a more general perspective, we hope that the results from this important cohort will shed light on the problems that these families face and struggle with every day. We appeal for authorities, institutions, and organizations, who are responsible for providing help and support to the whole population, to pay extra attention to families where severe mental disorders are involved, sometimes in two generations (see **Box 1**).

Limitations

The comprehensive and thorough test battery is also quite time consuming which may lead to some families not being able to complete all instruments, tests, and questionnaires. Although we try to motivate every family to participate again, some degree of attrition is inevitable, and that this could be expected to be more pronounced in the group of lower functioning families, but it could also be in the busy and well-functioning ones. In the VIA 7 study, however, many families were positive about their participation and for some, the feedback on their child's performance and the results that we provided, meant a lot to them. We also offer all families a feed back after completed testing in the VIA 11, and in those cases, where the situation calls for immediate help or support from authorities, we try to guide the family, or we send referrals and information to the municipalities.

Since a large group of testers are working with the same instruments, we will be ensuring very carefully that all instruments are used, and results interpreted in the same way by having monthly meetings where we discuss any issues or problems and where we score clinical examples together. Testers often work together in pairs, but in varying combinations also with colleagues from the other center.

Most tests are chosen with the follow-up perspective, allowing us to compare results from the VIA 7 study to the VIA 11 study directly. However, this was not possible in some cases and it will thus be necessary to look into the comparability of tests. This is for example the case for the measure of attachment, where the method used in the VIA 7 study was the Story Stem Assessment Profile [SSAP (98)], which is suitable for age four to nine. In the VIA 11-study we use the Secure Base Script Test [SBST (63)], which is recommended from age 9 to 14. However, both tests are based on the same theoretical basis that a narrative setting and an invitation to create a story that is inspired from everyday situations will tap into the child's inner world and working models of adult- and self-presentations. Optimally, we would conduct a validity study with e.g., 30 nine-year-old children performing both tests and then see how the attachment profiles correspond in the two tests.

Box 1 | Case Benjamin

Benjamin is a curious 11-year-old boy,who lives with his mother and his younger brother. Benjamin's mother has been diagnosed with bipolar disorder when Benjamin was four years old and she divorced Benjamin's father a few years ago. Benjamin visits his father every other weekend, sometimes more frequent, if his mother is depressed and in need of hospitalization. For most part of his life,Benjamin was told that his mother was hospitalized due to influenza, but recently Benjamin learned that it was because of her 'thoughts in her mind', that his mother sometime had to go away and letting him and his brother stay with their father and his new wife, and their baby twins.

Benjamin is usually a happy boy who likes to go to school, play football and Minecraft. However, he is also a rather anxious boy; who is not comfortable with sleeping at his friends or going too far away from home. He has also developed a sleep disorder as it takes him a very long time to fall asleep every night. At school and at home Benjamin sometimes gets very angry and has tantrums,although it is now much less frequent than when he was younger. He is doing fine in school and has one good friend. For a while,Benjamin was seriously bullied by some boys from another class. He was afraid to tell his mother about the bullying, because he wanted to protect her from getting sad. Eventually,the teacher found out and contacted Benjamin's mother. Now, the bulling has stopped but Benjamin still thinks a lot about having been bullied. Maybe that is why he sometimes have problems concentrating in school.

Benjamin participated in the VIA 7 study and the VIA 11 study and was fond of it, although the assessment at the hospital lasted three full days and was quite demanding. He talked to a psychologist in the VIA 11study about the sounds and voices that he had been hearing almost monthly for the past 8 months. The sounds and voices always seem to come when he is nervous or on his own, and although he realizes that they are not real, they make him feel uneasy. A few times he had shouted back at them. He had never talked to anybody about this before and found it as a relief to share it with a friendly person.

We decided to use two scanner facilities, one in Aarhus and one in Copenhagen to be able to cover all of Denmark apart from the similar scanner systems across sites, and to introduce the site variable in the data analyses, so that bias due the site will be detected. Finally, we will pay close attention to securing equal distribution of the scans of the cohort between the two centers in terms of HR-status of the children, so the two centers can analyze data independently.

Mean age of onset of puberty has lowered for the last decades (99), and for some children at age 11 the process of puberty has already started. We will be able to control for this difference in our analyses by using data from the blood tests and data regarding Tanner state of puberty as covariates.

Perspectives

It is highly relevant to carry out careful examinations of early signs or symptoms of mental disorders among high-risk children several times before and during puberty to measure longitudinal patterns of symptoms. Especially, it is important to determine constellations and interactions with external influences that determine the event of a more negative progression of symptoms and function vs. constellations that facilitate resilience in an individual with a similar vulnerability. The current study allows us to explore these patterns, including a wide array of measures, reaching from phenotyping, to measures of brain structure and function.

The children at age 11 are facing puberty, which is a period characterized by massive changes in brain structures and connectivity as well as changes in physical appearance, hormonal status and psychological, and social constitution (13). Paradoxically, even though self-regulatory control develops at a fast pace during puberty, it is also a period with changes of behavior, including risk taking activities and new relational patterns, for example higher degrees of independency, and a period with high incidence rates for mental disorders. From a developmental perspective puberty is a period in life that is highly formative but also complex to study because both age, hormonal status and social and psychological aspects matter when comparing individuals in e.g., a cohort. This is why cohorts that focus on a narrow age range may be helpful in determining differences of development and behavior inter-individually, or systematically across specific groups. Social mechanisms, such as bullying or other forms of social defeat that may take place in childhood and adolescence, are frequently reported and are related to later emergence of mental illnesses like depression and psychosis (15) and other negative life outcomes.

It will be of immense value if a better understanding of the early phases of the development of mental disorders, neurodevelopmental changes including brain development and the needs of these children will permit early interventions with potential prevention or preemption of psychosis. Increased knowledge about the impact of the thoroughly described and closely investigated environmental risk factor like home environment, parenting style and relation, parental functioning and mental health, social network, socioeconomic status, and adverse life events will create the basis for developing specific supportive and preventive interventions. These children have for decades been known to be at an increased risk of mental illness. The more we know about the early development of their potential course of illness and resilience factors, the better we can provide early and specific help.

AUTHOR CONTRIBUTIONS

AET wrote the manuscript together with MN. All co-authors NH, AS, MG, ÅP, MK, JB, LC, MM, DE, BB, AG, MU, JO, AN, LJ, ALT, AA, LV, CK, HS, JJ, OM read the manuscript, commented etc. MU was responsible for the statistics section. KP, HRS, LØ, and VB were responsible for all content regarding the MR scans.

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Improving Identification and Child-Focused Collaborative Care for Children of Parents With a Mental Illness in Tyrol, Austria

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Background: Children of parents with a mental illness (COPMI) are more likely to experience negative long-term adversities. However, interventions to support their needs early can significantly enhance adjustment and reduce negative outcomes. Approximately one in four children currently lives with a parent with mental illness worldwide. The lifelong impact for individuals, governments, and broader society is likely to be substantial. There are significant workforce barriers to the early identification of COPMI and addressing their needs, particularly within the adult mental health care system. The current study aims to reduce such barriers and to improve identification of COPMI in the current health care systems.

Objectives: The project "The Village" is a multidisciplinary health and social care policy intervention and seeks to improve child development and well-being outcomes for children of parents with a diagnosed mental illness. This will be achieved through the co-development, implementation, and evaluation of a practice approach to the early identification and collaborative care for COPMI, through establishing child-focused support networks. This will be done with open innovation science (OIS) approaches engaging the public in Tyrol, a geographical region of Austria, throughout 4 years. As part of the co-development process, we will work with stakeholders to co-develop the practice approaches based on evidence-based approaches and determine the most appropriate study design to evaluate those, as well as the implementation processes we will undertake.

Methods: The project is underpinned by theories from different disciplines (i.e., public health, psychology, sociology, linguistics, economic sciences) as well as drawing on different approaches (i.e., co-development, implementation science, symbolic interactionism, and realist evaluation). It is based on the seven content work packages (WPs): 1) management, 2) focusing on children and methods to understand their "voice,"

3) scoping, 4) co-development, 5) implementation, 6) evaluating the practice approaches, and 7) knowledge dissemination. "Scoping" will involve exploring the existing evidence, practice, and current state of identification and collaborative care in Tyrol, Austria. "Co-development" involves the co-design of practice approaches to identify and support children in partnership with key stakeholders and service providers working in Tyrol. The "implementation" of practice approaches will be based on the results of the co-development phase and will involve working with organizations to develop support strategies that draw on known organizational drivers from the field of implementation science to support the rollout of the practice approaches. In "Evaluation" we will follow principles of a realist approach; this includes developing program theories and logic models for the practice approaches. Those will set out the outcomes hypothesized to achieve and the processes that are expected to lead to those changes. This will refer to changes in children, parents, and practitioners. We expect that the main focus will be on measuring child quality of life and mental health outcomes, and outcomes that are on the path to those (such as social support needs, resilience, mental health literacy, stigma, and help-seeking behavior) as well as costs. The "child voice" WP focuses on children's perceptions and needs as the importance of "assent" and support of children to develop their own "voice" in health care is increasingly recognized within child health research. The "dissemination" step focuses on reaching a broad public audience of different stakeholders, researchers, and families involved.

Discussion: The research project aims to directly improve identification and support of vulnerable children across selected regions in Tyrol, Austria, and by doing so, improve the health and well-being of future generations, through breaking the cycle of intergenerational transfer of adverse childhood experiences.

Keywords: children of parents with mental illness, practice approach, implementation, participatory co-design, realist approach, symbolic interactionism, open innovation in science

INTRODUCTION

It is estimated that 50% to 66% of people with serious mental illness are living with one or more children (1), and that approximately 25% of children live with a parent who has a mental illness (2–6). Having a parent with a severe mental illness (SMI) has been associated with adverse child development outcomes, which can have long-lasting effects throughout a child's life including multiple physical and mental health problems (3, 7–10); lower academic achievement (11, 12); and reduced employment opportunities (13). While most studies on COPMI have been carried out in the United States, United Kingdom, and Australia, there is now also evidence from a German national mental health survey showing that parental mental illness is associated with increased risk of mental health problems in children and adolescents (14). A community study found that children of parents with SMI carry a higher risk of developing mental disorders compared to children of parents with mild to moderate mental illness (3). Thus, children of parents with a mental illness (COPMI) are more likely to present as the next generation of people living with SMI and who will use mental health services (10). Further, studies show the overrepresentation of this population in child community mental health services: 48–79% of the children using community mental health services had a parent with a mental illness (15–17). This transgenerational transmission of mental disorders (TTMD) is associated with high costs: In the United Kingdom, the estimated costs of adverse child impact linked to maternal mental illness during the perinatal period alone is £5.8 billion per year (18, 19). The risks associated with the TTMD could result in impaired parent–child interactions, genetic and pregnancy risks, emotion regulation deficits, individual vulnerabilities such as difficult child temperament, stress reactivity, cognitive skills, as well as social environmental factors such as school/work environments and social support. These risk factors are summarized in the Hosman et al. (20) model of the TTMD, though as yet we know currently of only one study testing this model (21).

The treatment of the parental disorder is associated with improved mental health and well-being in COPMI (22–27), though overall there are only a few studies on such effects (25, 28). These studies typically target the same disorder in children and parents, e.g., depressive symptoms in the offspring of parents with depression. However, such a specific transmission of disorders is not typical, as the outcomes often follow a *multifinality* pathway (20) with children of parents with depression developing various kinds of disorders (10, 29). A meta-analysis with nine studies on the effects of psychological treatment of maternal depression on children's psychopathology resulted in an overall moderate effect size, with a Hedge's g = .40 (28). An earlier meta-analysis on preventive interventions for COPMI demonstrated a significant relative risk reduction of 40% in the same disorder as their parent, and overall small effect for children's internalizing (Hedge's g = -.22) and externalizing (Hedge's g = -.16) symptoms (30). The most recent and comprehensive meta-analysis on preventive interventions for COPMI (31) resulted in effect sizes similar to those of Cuijpers et al. (28) for young children, and overall smaller effects for older children that equal those of Siegenthaler et al. (30). Different longitudinal studies on parental anxiety and depressive disorders present heterogeneous effects of parental treatment on children. A 6-year prospective longitudinal study on the effects of parental panic treatment demonstrated that parental treatment is a significant predictor of children's anxiety symptoms (25). The Sequenced Treatment Alternatives to Relieve Depression (STAR*D) Child study is designed to examine the association between maternal remission from depression and children's functioning and psychopathology. The study demonstrates differential effects on child psychopathology in early, late, and nonremitting mothers, with early remission being associated with reduced child externalizing problems (27); similar results have been obtained in another large longitudinal study (22). Thus, overall positive parental treatment effects are associated with reduced psychopathology of the children, though changes in important markers such as well-being, or for children that have not developed psychopathology symptoms yet, are missing.

A major barrier for such positive effects of the parental treatment is the lack of identification of children in families with mental illness. A recent review identified only nine studies systematically assessing whether adult mental health patients admitted to either in- or outpatient care had children or not, and revealed a substantial number of patients that were either not asked about parental status or where this was not documented in patient files (32). The same holds for child and adolescent mental health services (CAMHS) that do not systematically assess the health status of the parents of children and youth in treatment (17). Many systemic barriers exist preventing the early identification of COPMI in adult mental health services including a lack of skill and knowledge of practitioners to talk about parenting, lack of collaboration between the different services involved (i.e., adult and child mental health), individualized funding and treatment focused models of care that limit a family focus in treatment, and a reluctance by parents to talk about their children for fear of child removal into care (33–35).

Hence, according to the review by Maciejewski et al. (36), we are currently lacking opportunities for early identification and thus preventive interventions for COPMI due to adult mental health services neglecting the impact of the TTMD by not asking about parents and children. Developing opportunities or practices that help integrate a focus on intergenerational impacts of mental illness in adult mental health in ways that manage these systemic barriers are considered to be essential (37).

Interestingly, other disciplines have realized the importance of such early identification, especially pediatric primary care (38). As up to 15% of mothers and 7% of fathers suffer from postpartum depression (PPD) (39), they suggest PPD screening in pediatric primary care in order to identify and treat parents and to prevent negative PPD impact on the children, as this has been associated with a high risk of developmental delays and behavior problems (38). This study thus further highlights the necessity of strong collaborative care that, in practice, is rare and challenging between relevant disciplines (40, 41). Currently, this lack of collaboration leads to a lack of identification and thus missed prevention opportunities.

Another significant barrier is the current lack of collaboration itself. Current research suggests a lack of collaboration between different sectors that may impact COPMI—such as adult psychiatry/psychotherapy, child and adolescent psychiatry/psychotherapy, youth welfare agencies, pediatrics, gynecology, as well as family and social services. Instead these sectors offer individualized services that neglect the family as a whole, and the opportunity to capitalize on coordinated services is lost (42, 43). Thus, services that are in touch with families where a parent has a mental illness do not connect and miss chances that might improve overall family and especially child functioning and well-being (33, 40). Even in countries that have taken legislative steps to improve identification of children in families with mental illness, such effective collaboration is still scarce (44). As a result, COPMI often remain invisible, and their needs may be unmet, as their "voice" usually remains unheard (45).

Our project "The Village" has been designed to address these barriers to the care of COPMI through utilizing an open innovation approach (46–48) to design and trial evidence-informed practice approaches in adult mental health and other family support services using principles of symbolic interactionism, realist evaluation, participatory co-design, and knowledge of translation to practice implementation strategies from the field of implementation science (for elaboration on those methods, see below).

The current study has two major objectives: in our model region in Tyrol, Austria, we will 1) facilitate identification of COPMI with a *Sensitive Screening* (SENSE); and 2) establish a *Collaborative Village Approach* (CVA) that will enhance the formal and informal support opportunities for children. The project will be carried out with a focus on understanding children's experiences, challenges, and opportunities to support a child-focused SENSE and CVA, the child's voice. The project aims to ultimately improve child development and well-being outcomes for children and families.

METHODS

The project will be carried through seven work packages to scope, co-design, implement, and evaluate the practice approaches (see **Figure 1**).

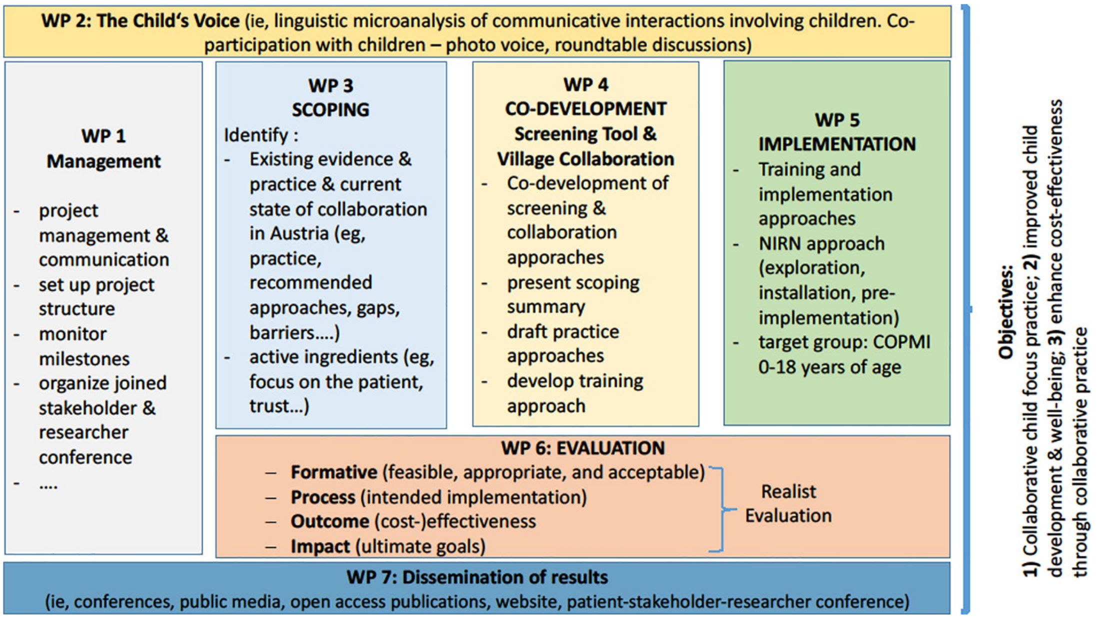

FIGURE 1 | Objectives and work packages (WP) of "The Village."

Design and Theoretical Framework

The project is underpinned by the theories from implementation science (49), a realist evaluation approach (50), and symbolic interactionism (51, 52).

Installing any new practice in routine care is difficult. Many gaps exist, particularly in mental health services, between what is known to be effective evidence-based practice and the translation of these practices into routine care (53). To help move beyond a "train and hope" strategy for workforce development, working with ways to address the systemic barriers within and across services that prevent practice change is a particular focus of implementation science, particularly for practice approaches that aim to address the intergenerational impact of mental illness in families (37). According to the National Implementation Research Network, implementation is defined as a specified set of activities or strategies designed to put into practice an evidence-based program or activity into routine use within specific settings (54). The field of implementation science acknowledges the need to focus on the feasibility and acceptability of new "intervention"/practice change, in order to facilitate practice change. The principles of co-development and co-design approaches that aim to work alongside key stakeholders in applied settings to co-create and adapt the evidence-based practice with local contingencies in mind, particularly in complex health environments, are one such approach drawn from implementation science to address the gap from research to practice (55).

A realist approach toward evaluation is a theory-driven type of evaluation that is particularly suitable for the examination of complex programs in the health and social care field. It advocates the investigation of theoretical assumptions underpinning a program and whether those are responsible for changes that may occur. The aim of each realist evaluation is to provide a detailed description and analysis of contexts, assumed causal origins, and mechanisms that, in turn, affect the outcome of an intervention (56). Realist evaluation is based on the theory of critical realism as proposed by Archer et al. (57). According to this underpinning theory, the potential mechanisms of causation reside both in the individual actors as well as in society and are real and present even when not active and may or may not be observable when actualized. Realist evaluation is thus different from traditional evaluations that focus on overall effectiveness in that it provides a basis to describe how and why a complex intervention works or does not work (56). Both empirical qualitative and quantitative data will be used to examine not only whether the practice approaches were (cost-) effective but also possible processes and contextual factors that influenced outcomes (or costs).

The theoretical framework of symbolic interactionism is based upon the epistemology of constructionism and refers to the belief that meaning and knowledge are constructed and maintained through social interactions; there is no one truth to be discovered, but many depending on the way in which the question is asked, approached, and analyzed (51, 52). As such, it is important to continuously gather data and perspectives of multiple stakeholders throughout the Village project that provide feedback into the project design. Such an approach focuses on the importance of language and interactions in shaping how people make sense of themselves and their social world (58). Given the stigma experienced by people and families impacted by mental illness, along with the inherent sensitivities of trying to find and support children in this situation, examining the social and interactional nature of experiences is particularly relevant for this study. Research using symbolic interactionism values multiple data sources, commonly obtained through qualitative approaches, and perspectives in order to continuously build and reflect the social interaction of interest. Aligned with this theory, we undertake a period of exploration (scoping). The co-developed approach also acknowledges the importance of collecting multiple views in order to produce new practices that will more likely fit the situation. Obtaining multiple data sources, such as interviews or observations from different perspectives or media, known as data triangulation, can enhance interpretive rigor, a valuable quality check used within qualitative research.

With an open innovation science (OIS) approach, i.e., the strategic use of the public to increase innovation (46, 47), we will decide together with the relevant stakeholders on the research steps to be taken and consult monthly with a group of "experts by experience" (young adults with a parent with a mental illness). Our project's use of OIS will be internally assessed by the Open Innovation Centre at the Ludwig Boltzmann Gesellschaft using novel evaluation criteria based on opening up disciplinary boundaries, fostering public engagement in the research process, and establishing new forms of stakeholder interaction and collaboration that lead to interdisciplinary and transdisciplinary research (59).

The processes of this methodological framework as well as their linkage are described in detail in the work packages "scoping," "co-development," "implementation," and "evaluation" (see below).

The other three work packages of the Village describe the overall management, dissemination of research findings, and our central overarching focus of the project on capturing the "child's voice."

Scoping

Drawing on implementation science, in a staged model of organizational change, scoping (known as the exploration stage) is a critical first step needed to understand the contextual factors shaping existing practice and practice challenges that need to be addressed in the implementation of practice approaches for the translation to routine care (60). This initial step will therefore generate the knowledge and conceptual frameworks to be informed and to be tested in the co-development, implementation, and evaluation of the project. This will include summaries of evidence and draft logic models that inform the development of the two practice approaches, SENSE and CVA. Scoping will be supported by the knowledge generated on conceptualizations of the child's voice (described below).

Evidence will be collected through reviews, and OIS principles such as stakeholder views; sharing and exchanging will be applied to understand and describe 1) current practice, recommended practice, and gaps in relation to the identification of COPMI as well as in relation to current and recommended collaborative care that puts the child at the center of attention ("child's voice"); 2) the (unmet) needs for COPMI; 3) what works for whom and when, what is cost-effective, and what are the mechanisms (active ingredients); and 4) contextual factors that can inform the development of logic models and the development of approaches.

As this work is underpinned by the theoretical framework of symbolic interactionism (51, 52), it is essential to gather perspectives of multiple stakeholders to inform co-development and implementation.

Particular pieces of work to address the scoping objectives will be conducted, using varied techniques:

- mapping of existing Tyrolean support structures and epidemiological dimensions, and mental health service use;
- semistructured interviews with Tyrolean stakeholders in the community, from services and families, to analyze the current situation of supporting COPMIs, existing needs, and barriers in Tyrol;
- mapping of key topics in the COPMI research field to identify what topics are studied together and which are not (and what the current research focus is) using co-word analysis, bibliometric coupling, and co-citation analysis;
- systematic review identifying facilitators and barriers in identification and support provision for COPMI;
- interviews with international experts who have attempted to change adult mental health care to be family-focused in order to improve identification of children at risk of mental illness;
- review of collaborative practice within the COPMI setting to identify the "active ingredients" or what has been shown to work.

The results from the Tyrolean mapping and situational analysis are currently summarized in a separate paper (61). Our preliminary evidence suggests that a variety of services are available both for parents who have a mental illness as well as for families and individual family members. However, services do not address the needs of COPMIs specifically and lack coordination across sectors. Findings further indicate that there is awareness of the problems related to COPMIs at all levels, but there is a lack of installed support processes to meet their needs.

Finally, initial logic models will be drafted in line with a realist approach (50). This work will inform the co-development process where the logic model will be developed further and agreed upon with stakeholders, and the co-creation of new practice approaches with stakeholders that draw on the evidence base for SENSE and CVA.

Co-development

One key element of this research project is that it follows OIS principles (46). This includes the co-development of the practice approaches for SENSE and CVA (informed by knowledge generated during the scoping in the form of initial logic models and the research literature) with local stakeholders and the training and implementation support for the application of these co-created practice approaches into routine care. Furthermore, stakeholders will be involved in decisions about key indicators and (outcome) measures and study design for the evaluation of the practice approaches.

A series of design workshops with stakeholders representing key service providers from the Tyrol region will be held monthly over a 6-month period to draw on the evidence and logic models developed in the earlier stages and existing practice wisdom of key stakeholders to co-develop the components of the two practice approaches (SENSE and CVA). Key stakeholders include leadership representation from psychiatric/psychotherapeutic and social care services across the region supporting adults and/or children, with a mix of professional background, and include people with lived experience of parenting with a mental illness or being an adult child of a parent with mental illness. Consultation will also occur with COPMI (the beneficiaries) acting in an advisory capacity. A training concept and training materials as well as feasibility and implementation indicators will also be prepared with the input of key stakeholders to support the implementation of the practice approaches in their respective contexts. To date, four out of six workshops have been held and draft models for SENSE and CVA for the Tyrolean region are already available. A preliminary Theory of Change model was presented to the workshop participants, which was based upon initial logic models, literature, and data collected during the scoping phase (see **Figure 2**). All workshops will be recorded, transcribed, and analyzed. Additional data will be collected *via* participant observation and by holding a focus group after the final workshop. Qualitative methods will be applied to analyze the data collected to measure the impact and process undertaken during this co-development phase. Core themes will be identified (based on a content analysis), clustered, and critically analyzed (62).

Implementation of the Practice Approaches (SENSE and CVA)

The objective of this step of the project is to implement the key practice approaches and position the evaluation of the impact into the applied settings decided on in the co-development stage. This stage of the project will draw on best practice evidence using known facilitative implementation drivers (49) to support organizational change to embed the 1) SENSE and 2) CVA approaches into selected Tyrolian study sites. Implementation drivers (also known as core components) are a key focus of implementation of the new practice approaches to ensure that the project

- develops, improves, and sustains the ability of selected sites to deliver the new practice approaches and
- creates an enabling positive organizational context that can support and sustain the operation of the new practice over time (49).

Implementation drivers are activities and processes for building the capacity and infrastructure of an organization to influence a program's success and are the "engine of change" needed to initiate and support changes to achieve positive outcomes for children, families, and community (49).

Underpinning these drivers is the level of Program Acceptance and Program Buy-in by staff, including managers and leadership, to the new practice approaches. When attention and action are paid to these key drivers, we are much more likely to see the practice approaches implemented as designed, resulting in change to practice and therefore improvements in family outcomes.

The evidence-based implementation framework will inform the project from the scoping through to the evaluation phase. Implementation is concerned with the use of strategies to adopt, integrate, and use evidence-based interventions/practices to change practice patterns within specific settings. This approach will utilize the NIRN implementation framework developed by the National Implementation Research Network (49) (exploration, installation, implementation), and undertake a series of activities across the entire project that will attempt to understand the context of implementation (the scoping of current practice and existing barriers at selected sites), to design a feasible approach that fits to the context (the co-development approach), to help prepare the environment, to train dedicated staff, and to coach the practice of the approach at selected sites. This implementation phase will also monitor the uptake and address issues with the on-going implementation of the approach at sites participating in the evaluation.

Evaluation

The main objective of the evaluation is to generate knowledge as to whether SENSE and CVA were implemented successfully and whether they were (cost-) effective. More specifically, the objective is to understand feasibility and acceptability of the practice changes, with a focus on the co-developed nature of the process, and on mechanisms and contextual factors; changes in outcomes (i.e., child quality of life and psychopathology, or outcomes that are expected to be on the path to child quality of life and mental health improvements, such as social support, mental health literacy, stigma, confidence); and changes in service use for COPMI and their family. We will also seek to capture impact at the wider system level. We will need to agree on outcomes with stakeholders, but at the moment, according to our current theory of change (**Figure 2**), we will measure mental health literacy, stigma, help seeking, quality of life, and psychopathology.

Evaluation of SENSE and CVA will follow principles of the realist approach, which is particularly suitable for complex interventions, and takes into account the mechanisms and conditions for change (50). Generation of the logic models will inform the framework of evaluation. Planning for process evaluation will start during scoping and co-development and inform modifications before full implementation of the practice approaches. Indicators will include a number of professionals trained, parents screened and children identified, children or families referred, collaborative "Village" meetings held, as well as awareness and behavior change in professionals. Knowledge generated about program theories (e.g., in the form of logic models) during scoping and co-development will inform the evaluation of outcomes and costs. Outcomes, resource use, as well as views and experiences will be captured at baseline and 6- and 12-month follow-up (i.e., after baseline) to assess changes for children and parents. Evaluation parameters and measures will be agreed upon during the scoping and co-developing phase. Personnel at study sites will be trained in the recruitment of study participants and practice approaches during the implementation phase. The cost-effectiveness analysis (in the form of cost-consequences analysis) will examine direct and indirect costs from a public sector as well as a societal perspective, and compare them against outcomes. Statistical analysis will be carried out to examine the causality of SENSE or CVA on outcomes and service use (and costs). The choice of statistical method will depend on the final choice of the study design.

Understanding the "Child's Voice"

Each of the steps outlined above will be influenced by an overarching focus on capturing the child's voice in the research undertaken. The project will explore and identify the concept

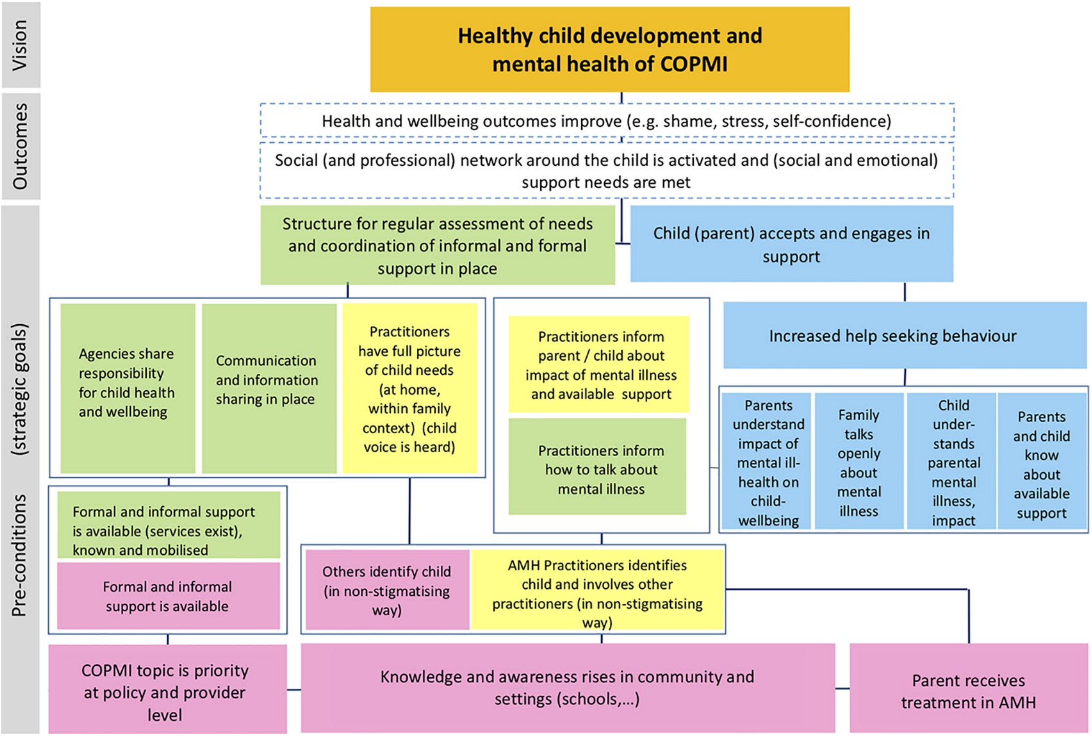

FIGURE 2 | Preliminary theory of change. Green boxes refer to the CVA approach; yellow boxes to SENSE; pink boxes refer to general elements; and blue ones to the family. AMH = Adult Mental Health; COPMI = Children of Parents with a Mental Illness.

of the "child's voice" in COPMI health settings, and knowledge produced will inform the overall research process, particularly in the co-development, implementation, and evaluation. This part of the project will identify how children currently, and wish to, participate in discussions about their needs, from the perspective of the child, service providers, and parents. It will uphold the project's underpinning philosophy to hold the child at the center of their care, and to listen and support them in identifying their own formal and informal support network. It will provide opportunities to educate professionals in the implementation step in appropriate ways to engage and support COPMI to become more communicatively active in conversations with adults about their needs in the evaluation step.

Focus groups and interviews primarily conducted for scoping and co-development will include questions that will address the topic of "child's voice" and communication between children and service providers [c.f. Ref. (63) for an example of methodological approaches]. This work will support the International Charter for Human Values in Healthcare—Compassion, Respect for Persons, Commitment to Integrity and Ethical Practice, Commitment to Excellence, and Justice in Healthcare. It will also draw upon research regarding health care communication with vulnerable people, such as those with disabilities or individuals from migrant populations, in which traditional patient–doctor power balances are amplified, as is the case with doctor–child interactions.

Through close analysis of naturally occurring health care interactions and analysis of stakeholder perspectives, this work aims to understand and improve the nature and possibilities of children's interactive participation in conversations about their needs and concerns. This work spans across the project and makes use of and informs data collected in the other work packages. A series of sociological and linguistic micro-analyses will be performed across collected data sets. Additional data collection will include (audio or video) recordings of health care encounters with children, as determined through consultation with stakeholders in scoping and co-development. This part has already started as we record communications between health care professionals and children. A total of 40 interactions will be recorded. For comparison purposes, those include interactions between health care professionals and children in need of psychotherapeutic/psychiatric treatment as well as interactions between adult health care professionals that treat the parents and their children. We will also investigate current practice (i.e., case meetings) to determine how others do and can speak for the child if they are unable or not present.

The communicative effectiveness of SENSE and CVA to listen to the child's voice and address their needs will be evaluated once this phase of the project starts through recording observations and will also inform and utilize the evaluation. Discourse analytic techniques will be applied, such as Conversation Analysis (CA), pragmatics, and interactional sociolinguistics, to analyze interviews, focus groups, and naturally occurring interactions, which will provide opportunities to educate professionals in this field.

Dissemination

The objectives of the dissemination focus are based on OIS (46–48): (1) to gain ownership and buy-in for the research among stakeholders throughout the different research phases and (2) to achieve impact at an individual, community, regional, and (inter-)national level. This includes impact in regard to research, practice, and policy. All communication and dissemination will be designed sensitively, with the aim to reduce stigma as a relevant barrier to the realization of the project (45).

Communication and dissemination will be an ongoing process from start to end of the research. This includes identifying organizations and representatives of those who will be part of the project; relevant stakeholders will be recruited into advisory and steering groups; a series of open-day forums will be organized throughout the project to which a wide range of stakeholders will be invited. This will include families, service managers, politicians, researchers, commissioners, and practitioners (including clinicians, social workers, school teachers). We inform about upcoming events and provide information and material for all events already having taken place (https://village.lbg.ac.at/news). Events will inform stakeholders about the research and include educational components; for example, keynote speakers might talk about the latest evidence and good practice; policymakers and influencers (including patient and professional associations) will be engaged; partnerships will be built and managed with organizations and projects that have similar aims and work with similar target groups such as early interventions that target families with newborns to support them and promote healthy child development; and formal collaboration agreements will be established. Our first two formal events for stakeholders were held in Innsbruck in June and September 2018, including the project inauguration ("Kick off") and the first of our annual "General Assemblies." These events are an opportunity to present updates to our official and unofficial stakeholders, receive feedback on design and findings, promote the project to policymakers and people working with children in Tyrol, and raise awareness in a sensitive approach that aims to reduce stigma surrounding mental health. Ongoing information is made available *via* our Village research project website (http://village.lbg.ac.at). A range of innovative communication tools (e.g., video, infographics; see: https://www.youtube.com/watch?time_continue=64&v=XHBv7ebFkWU) will be developed, which will be disseminated together with research reports throughout the project to promote the research aims, methods, and findings. Peer-reviewed papers will be produced and published in scientific journals, and a joint researcher–stakeholder conference is planned at the end of the project to communicate findings to the general public.

Analytical Strategy

As outlined in our theoretical framework, as well as in the detailed descriptions of the different work packages, the Village intervention differs substantially from traditional RCTs or other traditional scientific approaches. Although this may bring several risks to the project, it provides a significant opportunity to understand and implement practice approaches that address the local needs in Tyrol. Currently, in Austria, as well as the other German-speaking countries Switzerland and Germany, there are no sustainable structures for COPMI. The Village aims at understanding the currently available structures and how to improve and connect them in order to achieve an improved identification of COPMI with our SENSE approach, as well as improved care with the CVA approach. For this, we will use the tools provided within our theoretical approaches. As those strongly depend on OIS and stakeholder participation, those will be developed and decided on interactively and in collaboration. We thus cannot provide a fixed analytical strategy, as this is work in progress. We have, however, done some preliminary sample size calculations to determine numbers needed to treat to observe change (calculations were performed using the "n4means" function from the R package "CRTSize"; http://r-project.org). As outlined in the Introduction, we can expect small to moderate effects (31). As there are currently no studies on COPMI and the SENSE and CVA approaches, we assume at best small effects. Depending on the number of clusters (i.e., participating sites), the number of participants to be included varies largely. For instance, if we assume a small effect (Cohen's d = 0.1) of our CVA approach as well as clusters participating in Tyrol with an intraclass correlation of 0.01 with $\alpha = 0.05$ (two-sided), $1\text{-}\beta = 0.8$ in a parallel-group cluster-randomized trial, we would need 3,420 participants in 342 clusters (cluster size m = 10). **Tables 1** to **3** provide information on the number of participants to be included for different cluster sizes. However, those numbers depend on whom we will include at what stage with what

TABLE 1 | Required total sample size of clusters and patients to demonstrate an effect of Cohen's d = 0.1/0.2, assuming an **ICC of 0.1** in a parallel-group cluster-randomized trial with respect to cluster size m.

Cluster size m	Cohen's d = 0.1		Cohen's d = 0.2	
	Number of required clusters	Number of required patients	Number of required clusters	Number of required patients
10	596	5,960	150	1,500
20	456	9,120	114	2,280
50	370	18,500	92	4,600
100	342	34,200	86	8,600
500	320	160,000	80	40,000
1,000	316	316,000	80	80,000
2,000	316	632,000	78	156,000
5,000	314	1,570,000	78	390,000

$\alpha = 0.05$ (two-sided), $1\text{-}\beta = 0.8$, ICC = 0.1.

TABLE 2 | Required total sample size of clusters and patients to demonstrate an effect of Cohen's d = 0.1/0.2, assuming an **ICC of 0.05** in a parallel-group cluster-randomized trial with respect to cluster size m.

Cluster size m	Cohen's d = 0.1		Cohen's d = 0.2	
	Number of required clusters	Number of required patients	Number of required clusters	Number of required patients
10	456	4,560	114	1,140
20	306	6,120	76	1,520
50	216	10,800	56	2,800
100	186	18,600	48	4,800
500	162	81,000	42	21,000
1,000	160	160,000	42	42,000
2,000	158	316,000	42	84,000
5,000	158	790,000	42	210,000

α = 0.05 (two-sided), 1-β = 0.8, ICC = 0.05.

TABLE 3 | Required total sample size of clusters and patients to demonstrate an effect of Cohen's d = 0.1/0.2, assuming an **ICC of 0.01** in a parallel-group cluster-randomized trial with respect to cluster size m.

Cluster size m	Cohen's d = 0.1		Cohen's d = 0.2	
	Number of required clusters	Number of required patients	Number of required clusters	Number of required patients
10	342	3,420	86	860
20	186	3,720	48	960
50	94	4,700	26	1,300
100	62	6,200	18	1,800
500	40	20,000	12	6,000
1,000	36	36,000	10	10,000
2,000	34	68,000	10	20,000
5,000	34	170,000	10	50,000

α = 0.05 (two-sided), 1-β = 0.8, ICC = 0.01. Calculations were performed using the "n4means" function from the R package "CRTSize" (http://r-project.org).

expected effect. They are thus currently only an orientation. The same holds true for potential outcome measures. There are measurements often used in the research of COPMI, such as the psychopathology screening instrument, the Strengths and Difficulties Questionnaire (3), or measures on child-well-being (64), but based on our theoretical framework and as laid out above, we decided to actively involve the relevant stakeholders in this process and give them a voice in our research.

Potential primary and secondary outcome measures might include well-being and psychopathology outcomes for children, improvements in family functioning and social support, and process measures of feasibility and acceptability of the new practice approaches in chosen sites. Barriers and facilitators to service change will also be an important indicator of the co-development and implementation processes of this study. We will also have a focus on measurement of the co-development process and satisfaction with outcomes achieved, as well as documenting the implementation drivers and program adaptation needed to translate the co-designed practice approaches into routine care.

For all activities, we ensure performance according to the Declaration of Helsinki and its later amendments and will obtain approval from the relevant Human Research Ethics Committees (see Ethics Statement). Within the scoping project, we have already performed interviews with relevant stakeholders that are currently analyzed. We have obtained ethical approval for the local scoping interviews and development process measures from the Human Research Ethics Committee of Monash University Melbourne, Australia. In addition, internal approval from the London School of Economics, London, UK for the international interviews is undertaken as part of the scoping stage. Once the recruitment of participants starts, we will beforehand obtain ethical approval from Innsbruck Medical University to ensure accordance with national Austrian guidelines.

Implications

COPMI are a highly vulnerable group that are still neglected in the health setting in general and the (adult) mental health setting specifically (15, 17, 32) and thus prone to the TTMD (20).

"The Village" aims to change the invisibility of this vulnerable group due to lack of identification and collaborative care in the model region of Tyrol, Austria. To achieve this, we follow a robust collaborative approach based on child voice and OIS principles involving beneficiaries, relevant stakeholders, and policymakers as outlined in the different phases of the project. In the end, this project will have informed and instigated a low-threshold, nonstigmatizing collaborative care system for COPMI in Tyrol, Austria, which efficiently and effectively integrates services and support from the perspective of the child. Accordingly, practitioners and other front-line staff working with children and adults will know how to sensitively identify families with mental illnesses, and how to carry out sensitive screenings (SENSE) to identify COPMI. They will know when additional services are required. If so, they will be able to put the child's voice at the center and carry out sensitive, low-threshold interventions, based on the child's and family's needs. This is expected to improve child development and quality-of-life outcomes and reduce the risk for the development of mental illness in the children themselves, promoting children's quality of life (65–67). Further, such a best-practice model will be capacity building, providing the evidence for upscaling the practice approaches to other regions of Austria and countries with similar practice challenges. A range of materials about the evidence-informed approaches (SENSE, CVA), and the resource impact of scaling those up, as well as implementation and dissemination strategies will have been made available.

Impact on service users: The project seeks to improve child development and quality of life for a population that is often invisible, i.e., COPMI are affected by multiple disadvantages, and often not known to services until presenting with their own problems later during child- or adulthood (42, 43). The project will improve the child's situation by providing access to a strengthened social support network that supports their emotional, practical, and social needs (45). Although not a specific aim of the project, children will benefit from destigmatized services that put the child at the center of support. This will likely lead to sustainable changes in the child's life and have positive effects on long-term outcomes that reach into adulthood, such as school performance, employment, and mental health.

Impact on policy, practice, and research: The project will test an innovative whole-system approach in an Austrian region. Based on the findings, a training approach, tools, and implementation support package will be developed that (if the project proves cost-effective) would allow scaling up the approach at a national level and may assist practice internationally that seeks to provide holistic care to COPMI. The evaluation can inform a resource impact analysis, which will provide an estimate of the costs to the government (and society) if the approach was to be rolled out more widely. Findings will be disseminated through a number of research outputs. The use of the stakeholders and the media to promote the project will be maximized throughout the project using an OIS design, to increase awareness of the importance of supporting COPMI and ways that this can be done. The message will be that many people in the community can support COPMI. By working with stakeholders, such as patient associations, local community leaders, and health and social care professionals, throughout the entire research project period, knowledge about mental illness in parents and its impact on child health and development will be disseminated. Throughout and toward the end of the project, project stakeholders will be invited to knowledge exchange events, where project progress and results are presented in informal, accessible language.

Through the co-developed research process, the capacity of stakeholders will be built to improve not only their collaborative- and child-centered approaches, but also their evaluation of practice. This includes an increased capacity to collect and analyze data on process, costs, and outcomes to reflect on their practice approaches. This can inform a long-term approach to generating evidence in this area.

Economic impact: Economic analysis will estimate the cost-effectiveness from the perspective of government and society. This will include the costs of informal care that occur when children look after their mentally ill parents, as well as other costs that should be included when planning public services (e.g., out-of-pocket expenditure to families and potential long-term costs if children are removed). In Austria, knowledge of the (cost-) effectiveness is currently not well utilized to inform the planning of services. The research team will work with policy stakeholders and support them in translating the research findings into practice.

Methodological Considerations

"The Village" is a multidimensional project that involves a range of potential risks. Examples of risks include not finding appropriate study sites; too many tasks for the 4-year time period; the unwillingness of stakeholders to be engaged; lack of practice change and fidelity in delivery; and delivering a local project through a multinational team. However, the project includes significant preparatory steps to scope potential study sites, to engage key stakeholders, to co-develop feasible practice approaches, and to use the latest thinking in implementation science to address these potential risks. The project officially started in February 2018 with an initial kick-off event in June 2018. For this kick-off, people with lived experience as well as the range of relevant stakeholders were invited and actively participated in the event. This resulted in positive feedback (https://science.orf.at/stories/2921593/), already suggestive of a high acceptance of and high involvement in "The Village" in Tyrol, Austria.

Finally, the research team will take substantial steps in planning for early identification and mitigation of risks and has indeed started taking some of those steps.

CONCLUSION

The research project aims to directly improve identification and support of vulnerable children across selected regions in Austria, and by doing so, improve the health and well-being of future Austrian generations while breaking the cycle of intergenerational transfer of adverse childhood experiences. The described project underwent a rigorous public review process (https://ois.lbg.ac.at/en/methods-projects/crowdsourcing-research-questions-in-science) with different stakeholders as well as people with lived experience and scientific experts of the field having reviewed our proposal (https://ois.lbg.ac.at/en/methods-projects/ideas-lab). This ensures high acceptance and most likely high probability of implementation of our approach, a highly relevant fact for sustained impact. Further, the research group is genuinely interdisciplinary with researchers from public health, (clinical) psychology, economics, linguistics, and implementation sciences thus combining not only different expertise but also perspectives that are crucial concerning cooperation and collaboration. We believe that the research findings from the described public health intervention will also be relevant for health care providers and policymakers in other countries, and the international research community.

ETHICS STATEMENT

The study is performed according to the Declaration of Helsinki and its later amendments and was approved by the Human Research Ethics Committee of Monash University Melbourne, Australia, in addition to internal approval from the London School of Economics, London, UK. Local human research ethics in Austria will be obtained as appropriate. Written informed consent will be obtained from all study participants. If participants are underage, they will be informed about the study and their written informed consent will be accompanied by the written informed consent of their legal guardians.

AUTHOR CONTRIBUTIONS

HC wrote a first draft of the manuscript. AB, BF, MG, IL, IZ, and JP reviewed and commented the version. HC and JP revised the commented version, which was reviewed again by all coauthors.

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Children of Mentally Ill Parents at Risk Evaluation (COMPARE): Design and Methods of a Randomized Controlled Multicenter Study—Part I

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Objectives: Mental disorders are frequent, associated with disability-adjusted life years, societal, and economic costs. Children of parents with a mental illness (COPMI) are at an increased risk to develop disorders themselves. The transgenerational transmission of mental disorders has been conceptualized in a model that takes parental and family factors, the social environment (i.e., school, work, and social support), parent-child-interaction and possible child outcomes into account. The goal of the "Children of Mentally Ill Parents At Risk Evaluation" (COMPARE) study will thus be twofold: (1) to establish the efficacy and cost-effectiveness of a high-quality randomized controlled trial (RCT) with the aim of interrupting the intergenerational transmission of mental disorders in COPMI, (2) to test the components of the trans-generational transmission model of mental disorders.

Methods: To implement a randomized controlled trial (RCT: comparison of parental cognitive behavioral therapy/CBT with CBT + Positive Parenting Program) that is flanked by four add-on projects that apply behavioral, psychophysiological, and neuro-imaging methods to examine potential moderators and mediators of risk transmission (projects COMPARE-emotion/-interaction/-work/-school). COMPARE-emotion targets emotion processing and regulation and its impact on the transgenerational disorder transmission; COMPARE-interaction focuses especially on the impact of maternal comorbid diagnoses of depression and anxiety disorders and will concentrate on different pathways of the impact of maternal disorders on socio-emotional and cognitive infant development, such as parent-infant interaction and the infant's stress regulation skills. COMPARE-work analyzes the transmission of strains a person experiences in one area of life to another (i.e., from family to work; spill-over), and how stress and strain are transmitted between individuals (i.e., from parent to child; cross-over). COMPARE-school focuses on the psychosocial adjustment, school performance, and subjective well-being in COPMI compared to an adequate control group of healthy children.

Results: This study protocol reports on the interdisciplinary approach of COMPARE testing the model of the transgenerational transmission of mental disorders.

Conclusion: The combination of applied basic with clinical research will facilitate the examination of specific risk transmission mechanisms, promotion, dissemination and implementation of results into a highly important but largely neglected field.

Clinical Trial Registration: DRKS-ID: DRKS00013516 (German Clinical Trials Register, https://www.drks.de/drks_web/navigate.do?navigationId=trial.HTML&TRIAL_ID=DRKS00013516).

Keywords: children of mentally ill parents, transgenerational transmission, mental disorders, intervention, prevention

INTRODUCTION

Children of Mentally Ill Parents

The German social report of the year 2013 indicates a total of 19 million children/adolescents living in 1.6 million single and 8.1 million dual family households (1). Given the estimated lifetime prevalence rate of 27.4% for mental disorders associated with significant disability-adjusted life years (DALYs) for the age group of 18–65 year olds (2), approximately 25% of the children/adolescents in Germany are living with a mentally ill parent, a percentage resembling international rates (3–7). Having a parent with a mental illness has been associated with multiple psychological and developmental risks for children, such as lower academic achievement (8), increased stress-related somatic health conditions (e.g., higher rates of asthma and other atopic diseases (9), internalizing/externalizing symptoms (10, 11), and the development of severe mental illness (SMI) (12). Our own study of $n = 15.904$ adult patients from three different psychiatric/psychosomatic clinics revealed that 65% of the patients had children and that 73.4% of those parents were currently caring for their children (13). The children of the parents of those three different clinics were already exhibiting symptoms of mental disorders themselves (depending on the sample between 15 and 38.4%), thus providing evidence that the trans-generational transmission of mental disorders (TTMD) is a major risk factor for the development of SMI, as demonstrated in numerous other studies (11, 12, 14–16). Long-term studies have shown that children of parents with a mental illness (COPMI) have a higher life-time risk of developing SMI that ranges between 41 and 77%. However, subclinical symptoms often emerge earlier in life (14, 15). The BELLA study revealed a parental mental illness as a powerful risk factor (OR 2.4) for the development of probable mental health problems in children and adolescents (17). Recent studies have added evidence that offspring with two generations previously affected by SMI are at an even greater risk (18, 19). Thus, COPMI are most likely to constitute the next generation of patients with a mental illness (12) associated with significant DALYs and economic costs (20–22). They therefore constitute an essential target high risk group to be addressed by selective prevention programs (23). This is the aim of the COMPARE-family project (see part II of the study protocol published in this research topic by Stracke et al. (24).

Transgenerational Transmission

A recent meta-analysis of the trans-generational transmission of parental mental disorders on children's symptomatology revealed specific effects of parental disorders on children (12). According to this meta-analysis, both transgenerational concordance (specific parental mental disorders increase children's risk for certain disorders) and multifinality (parental mental disorders increase children's risk for mental disorders in general) are used to define the scope of the child's diagnostic outcomes of a particular parental disorder. The concepts of transgenerational specificity and equifinality, respectively, refer to similarities and differences in parental disorders preceding a child's diagnostic outcome. Multi- and equifinality are more common in children of parents with unipolar and bipolar affective disorders, whereas the risk of children of anxious parents is mainly restricted to develop anxiety disorders. For all children, risk transmission is assumed to be partly specific since studies indicate a strong tendency for children to develop the same disorders as their parents. A meta-analysis ($k = 61$ studies included) of the cross-sectional association between paternal and maternal psychopathology on children's internalizing and externalizing behavior problems further revealed parent-specific gender effects (25). There is a general lack of studies on the specific risk transmissions for the broad range of parental disorders other than affective and anxiety disorders (12) and studies assessing a wider range of parental disorders as well as their effects on the children are missing, as are studies on parental comorbidity. This lack is addressed in the current study, as we will consider a broad range of parental disorders as well as comorbid ones.

A comprehensive model (see **Figure 1**) of the TTMD identifies four major domains [(1) parent, (2) family, (3) child, and (4) social environment] that interact with their respective systems and are influenced by five transmission mechanisms [(1) genetics, (2) prenatal factors, (3) parent-child-interaction, (4) family, and (5) social factors]. Child development over its whole span is considered, as well as the concepts of multi- and equifinality, concordance, and specificity. While there is empirical support for the model's different domains and factors, many research has solely addressed single dimensions (i.e., focusing on family or child factors or on genetics), without taking the whole model into account, which is clearly necessary (for a review of the model components see (13). Furthermore, most of those studies were conducted on individual disorders in parents, and comparative studies on the range of parental disorders and on crucial factors such as parental comorbidities are rare (26) or entirely lacking (12). Moreover, the specific impact of the various moderating and mediating processes between the incidence of mental disorders in parents and their children—such as genetics, epigenetics, stress reactivity, emotion regulation, parenting skills, parent-child-interaction, children's cognitive abilities, family, and environmental influences—as well as their interactions have not been clarified so far. Thus, the aim of the COMPARE consortium with its subprojects COMPARE-family/emotion/interaction/school/work is to test the TTMD model and to simultaneously assess the four major domains as well as the transmission mechanisms. This will enable us to establish specific transmission profiles (equi- vs. multifinality vs. concordance vs. specificity) for a range of parental disorders with/without comorbidities. Furthermore, we will be able to identify risk-profiles for children at high vs. low risk, since studies have shown that not all children will develop disorders themselves (25) although their overall risk is significantly increased (14). This will improve the development of targeted interventions, connecting our first aim with the second one, since the bi-directional influences of interventions on the TTMD as well as specifics of the TTMD on interventions have not been investigated so far.

Summing up, we expect that our study will substantially deepen our knowledge of the relevant risk transmission mechanisms and related child outcomes, as well as markedly improve children's health, by simultaneously assessing the domains and mechanisms of the TTMD model as well as the bi-directional influences of a selective intervention in one study. This paper reports on part I of the overall protocol paper describing the consortium structure with the clinical trial and the four add-on research projects. Part II is the protocol paper of the clinical trial, also published in this research topic (24).

Aims of the Research Consortium

The research consortium "Children Of Mentally III Parents At Risk Evaluation" (COMPARE) is a network of researchers that includes disciplines from clinical, work and organizational, developmental, and educational psychology. Using a randomized controlled trial (RCT) and accompanying add-on projects, the COMPARE consortium addresses the following main goals:

1. To establish effects of parental treatment on child outcome [see the protocol on COMPARE-family by Stracke et al. (24) in this Frontiers research topic].
2. To tests the domains of the TTMD model within the RCT.
3. To identify specific transmission mechanisms.

The primary hypotheses of the model testing part of the study are:

COMPARE-family: To test the transgenerational transmission of mental disorders from parents to children. We will establish risk profiles and analyze the transmission pathways of multifinality, equifinality, specificity, and concordance. We further test the effects of parental psychotherapy treatment on the children and whether a training of parenting skills will result in additional effects [for details see (24) in this research topic].

COMPARE-emotion: Emotion processing and emotion regulation demonstrate processes that, on the one hand, are reduced in COPMI compared to children of parents without mental illness. This relationship should be moderated by factors related to the mental diseases of the parents, such as the type of disorder, disease duration, age of the children at disease onset and disease severity. On the other hand, we assume these processes to be influenced by parental treatment and to simultaneously moderate the effectiveness of the intervention. Therefore, the current subproject was designed to (a) compare COPMI with children of parents without mental illness (COPWI) regarding emotion processing (EP) and emotion regulation (ER) skills, (b) to evaluate the impact of EP and ER on treatment outcome, and (c) to assess the effects of different treatments on EP and ER in COPMI.

COMPARE-interaction: Maternal psychopathology, infant stress reactivity, and the quality of the parent-infant interaction at 3–4 months predict infant development at 24 months postpartum, and maternal comorbid depression and anxiety coincide with greater impairment in the infant development at 12, 18, and 24 months than depression alone. Furthermore, it is hypothesized that the parent-infant interaction as well as the infant stress reactivity at 12 months of age mediate the relationship between the maternal psychiatric diagnosis 3–4 months postpartum and infant development (socio-emotional as well as cognitive) at 24 months of age.

COMPARE-work: We expect to observe that mentally ill parents are confronted with more severe working conditions as compared to healthy parents, and that stronger adverse spillover effects of work-based strain toward the family emerge for them. With respect to the transmission of parents' strain to their children, we as well assume that such crossover-effects depend on the parents' health status. Overall, we expect to identify specific patterns with regard to the spillover-crossover effect assuming that the maladaptive cycle leading from the parents' poor working

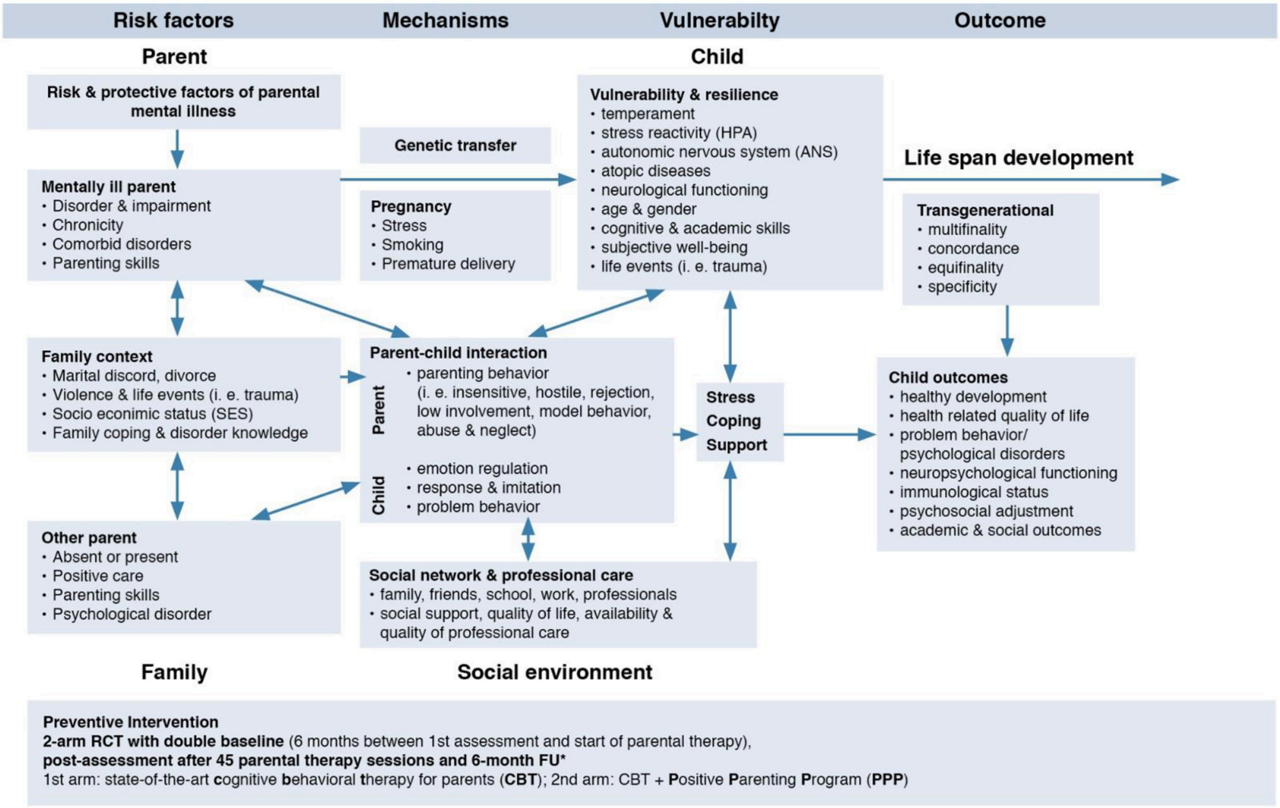

FIGURE 1 | Model of the trans-generational transmission of mental disorders adapted from Hosman et al. (14), p. 253, identifying risk factors, mechanisms, vulnerability, and child outcome. The influence of interventions on the different aspects of the model is an inclusion of the consortium.

conditions (in particular those with a mental illness) via spillover to the family can be interrupted so that no crossover to their children occurs.

COMPARE-school: We expect that COPMI perform poorer on academic achievement scales, display less positive psychosocial adjustment patterns, and suffer from lower general and domain-specific subjective well-being compared to children of mentally healthy parents. Due to the specific parental mental disease and parents' gender, we anticipate identifying different effects in the aforementioned child outcome variables in COPMI.

METHODS

COMPARE-Family: Effects of Parental Psychotherapy and Additional Parent Training on Children

This subproject is central for the entire consortium and planned as a randomized controlled trial establishing the effects of high quality parental therapy (cognitive behavioral therapy/CBT) on the children in comparison to parental CBT and a parent training, the Positive Parenting Program (CBT+PPP). We have chosen CBT for the intervention, as CBT has the soundest evidence base for the psychotherapeutic treatment of mental disorders (72). Studies further demonstrate poorer parenting skills in parents with mental disorders (27–30), and the enhancement of such skills has been a significant mediator in improving child outcomes (31). The Positive Parenting Program (PPP) is a well-established program to enhance parenting skills, and specific effects on child psychopathology could be demonstrated (32, 33). Studies explicitly testing effects of parental treatment in conjunction with parenting skills are lacking so far.

A total of seven university based study sites participates in the RCT and each of them specializes on different mental disorders. We will thus be able not only to address depression and anxiety, but a wider range of disorders such as sleep-wake-, and somatic symptom disorders, obsessive compulsive, trauma and stressor related disorders, eating and personality disorders, bipolar, and schizophrenic disorders. As patients in Germany are required to be abstinent for outpatient psychotherapy, patients with substance use disorders will not be included in the trial.

The structure of the RCT will allow us further (i) to establish effects of the parental disorder(s) with/without comorbidities on children's health; (ii) to establish effects of the different interventions (CBT vs. CBT+PPP) on children's health; (iii) to test assumptions of the TTMD model and bi-directional influences of different treatments on the model; (iv) to analyze specific transmission mechanisms; and (v) to establish risk profiles for children with urgent needs for preventive interventions.

Key inclusion criteria are parents with a mental illness in outpatient treatment caring for a child between 1.5 and 16 years of age. Key exclusion criteria are insufficient German language skills, severe impairment of the children requiring comprehensive treatment.

Description of the Primary Efficacy/Test Accuracy Analysis and Population

The primary analysis is conducted according to the intention-to-treat (ITT) principle and includes all randomized patients. The confirmatory test for treatment group differences between T2 and T3, and T2 and T4 with respect to the primary endpoint each applies linear mixed multi-level model with patients at level 1 and children at level 2, adjusting for center, number of comorbidities, number of children, baseline TRF score at T2, and length of waiting period between T1 and T2 (in weeks). The overall type I error rate is set at 5% (two-sided) and will be controlled by applying the multiple test procedure for hierarchically ordered hypotheses. Effect size assumed for estimation of sample size is set at $d = 0.25$. With a two-sided significance level of $\alpha = 0.05$ and a power of $1–\beta = 0.8$ using a two-sample t-test, 253 patients per group (n = 506 total) are required. Taking a drop-out rate of 20% into account, $n = 634$ patients need to be enrolled into the trial. Since it is assumed that parts of the outcome variance can be explained by the inclusion of covariates, the actual power of the analysis by linear mixed multi-level model is expected to be higher than $1–\beta = 0.8$. Sample size calculation was performed using SAS v9.4. For details on COMPARE-family and the full study protocol please refer to Stracke et al. (24) in this research topic.

The projects COMPARE-emotion/interaction/work/school are closely linked to this trial, as the parents of children of COMPARE-family will also be assessed for those subprojects. We will thus gain knowledge on the interaction of therapy effects with the specified TTMD-domains.

COMPARE-Emotion: Emotion Processing and Regulation in Children of Parents With Mental Illness

The TTMD model identifies child factors as one of four major domains of the transmission of parental mental disorders to the child (see above). Therefore, specific characteristics and behavioral traits in the child are likely to raise or lower the risk for a mental disorder in the child of a parent with a mental illness for him- or herself. Emotion processing and emotion regulation have been identified as such factors. Emotion processing (EP) is a broad concept that includes sub-processes of emotion perception, affective stimulus interpretation, and affective reaction to another's emotions. In the current sub-project, we want to focus on (a) emotion recognition, (b) perspective taking, and (c) affective arousal (sympathy vs. personal distress) as a reaction to another person's emotional situation (34). EP aberrations constitute a central characteristic of both individual and a wide range of mental disorders, reflecting a transdiagnostic approach (35–37). Emotion regulation (ER) comprises processes applied by individuals to influence the incidence, kind, intensity, and duration of their emotions as well as their effects on feelings and behaviors (38, 39). Strategies can be adaptive, for example if they increase positive or decrease negative emotions, or be maladaptive, having the opposed effect. Both, EP and ER constitute important skills that are transdiagnostically related to mental disorders. Both skills are affected by parental behavior throughout child development. The research conducted on this topic so far reveals direct and indirect associations between a parental mental illness and aberrant EP or ER in their children. However, no comprehensive study on the impact of the child's EP and ER on the trans-diagnostic trans-generational transmission of mental disorders has been conducted so far, despite the fact that these processes constitute important targets for preventive interventions. Furthermore, the impact of EP and ER on treatment effects has not been studied to date.

To examine these processes, experimental tasks assessing EP and ER will be administered in 200 children of parents participating in the COMPARE-family study [see study by Stracke et al. (24) in this research topic] at the assessment points T1, T3, and T4 of the clinical trial (100 children of parents receiving cognitive behavioral therapy and 100 children of patients additionally receiving the Positive Parenting Program). Furthermore, a sample of 100 children with parents without a history of mental diseases or psychotherapy and no indication of current mental disorders will be assessed with the same tasks at a single measurement point (see **Figure 2**). A version of the Video Sequences Task will measure various aspects of emotion processing including emotion recognition, perspective taking, personal distress, and sympathy. This task has been chosen because of it's high ecological validity. Children are shown short film clips containing scenes with the protagonist conducting a certain basic emotion. Additionally, the morphing task will be adopted to assess emotion recognition with respect to quality and speed. In this task, film clips with neutral faces morphing into affective faces are shown and participants are supposed to press a button as soon as they recognize the emotion. To measure emotion regulation, an emotional Go/Nogo paradigm will be applied. In this task children are supposed to press a button following certain affective stimuli, e.g., happy faces, and inhibit a button press following other stimuli, e.g., sad faces. These two experimental tasks allow for a precise and fine grained assessment of the relevant constructs. Dependent variables include behavioral task performance measures and peripheral physiological activity during the tasks (heart rate variability, skin conductance, and mimicry). Furthermore, questionaires will measure emotion regulation strategies and cognitive and emotional empathy of the children and their parents.

To investigate differences in emotion processing between COPMI and COPWI not manifest in behavioral or physiological measures, patterns of neuronal activity and neuroanatomical connectivity will be compared between subgroups. Especially the amygdala-prefrontal cortex network is assumed to display a specific coordinating role in this context, and different mental disorders in childhood and adolescence have been found to be associated with aberrant amygdala and/or prefrontal cortex activation during EP. Therefore, out of each subgroup 40 participants will be measured with functional magnetic

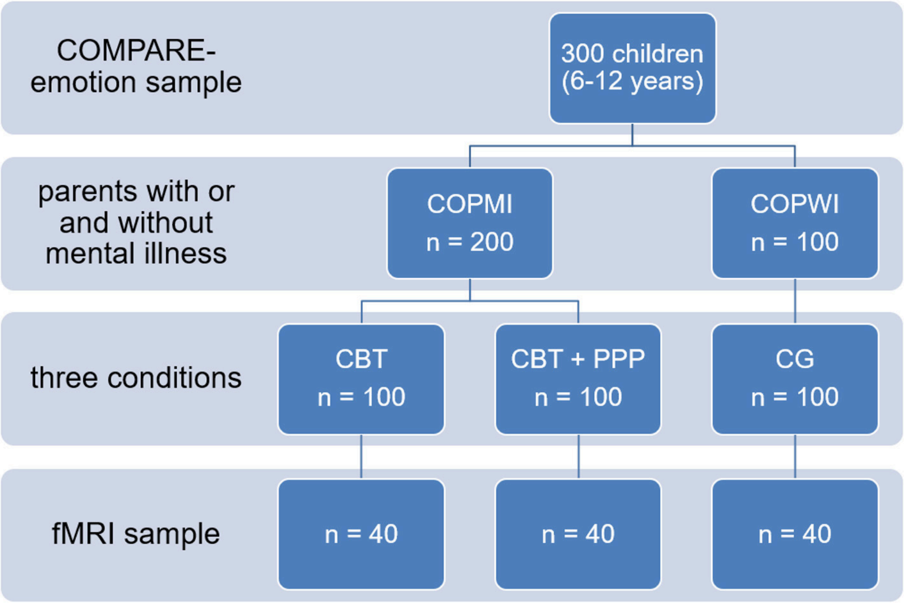

FIGURE 2 | Sample of COMPARE-emotion. fMRI, functional magnetic resonance imaging.

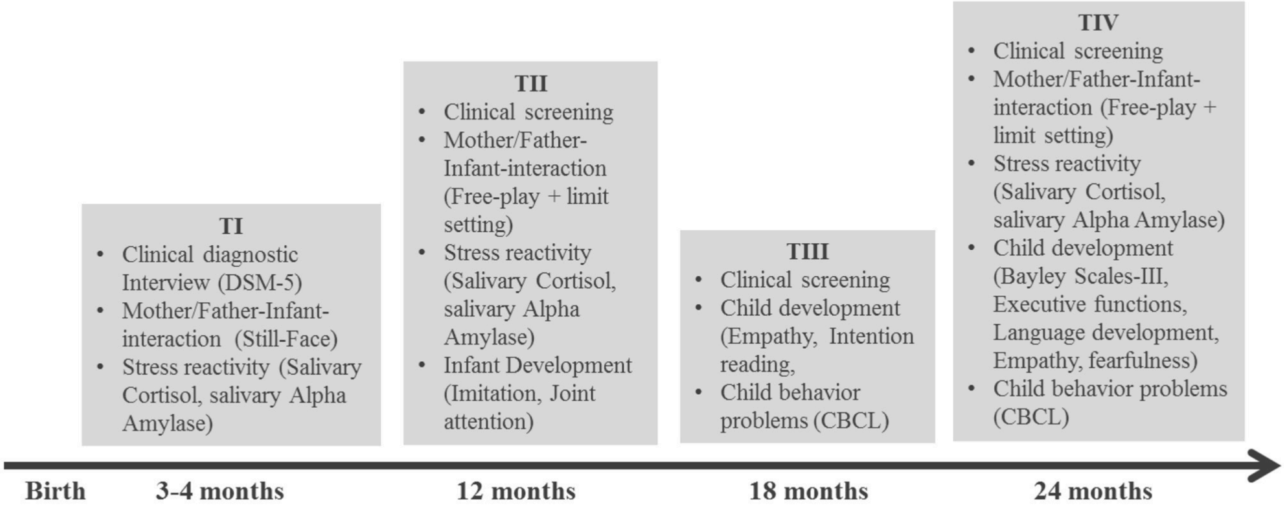

FIGURE 3 | COMPARE-interaction study design.

resonance imaging. A paradigm will be applied that involves processing of emotional faces as well as implicit emotion regulation via emotion labeling. We assume that COPMI will show higher activity in emotion processing brain areas and less habituation of the amygdala than COPWI, in particular if there is no task to perform. When the participants are required to draw attention away from the facial emotional expressions, we additionally expect higher neural costs in the COPMI, especially overactivity in the anterior cingulate cortex. During emotion labeling, COPMI should exhibit less regulatory prefrontal activity than COPWI, i.e., less activity of the ventrolateral prefrontal cortex combined with higher activity in the amygdala and less functional connectivity between both structures. Additional diffusion tensor imaging will be applied to investigate differences in white matter tract integrity in emotion processing networks.

COMPARE-Interaction: Impact of Maternal Comorbid Depression and Anxiety in the Peripartum Period on Infant Development: The Role of Parent-Infant Interaction and Infant Stress Reactivity

The Compare-interaction subproject pursues a developmental approach which aims to identify risk factors for the generational transmission of parental mental health disorders in early childhood as displayed in the TTMD model. This model identifies parent-child interaction as a core mechanism in the transgenerational transmission of parental mental disorders to the child. COMPARE-interaction focuses on the long-term consequences of maternal depressive and comorbid depressive and anxiety disorders during the peripartum period on child development, especially on socio-emotional and cognitive development. Special attention will be paid to the mediational effect of parent-infant interaction and infant stress reactivity on the relationship between the maternal disorder (depressive, comorbid, healthy controls) and infant outcome. Moreover, it will significantly contribute to model testing in the model's following domains: mentally ill parent, family context, other parent, parent-infant interaction, infant outcome. Given the high prevalence of depressive and anxious disorders during the peripartum period and the increased risk for children of depressed and anxious mothers to exhibit adverse developmental problems, further research in this field is urgently needed. The negative impact of depression on infant socio-emotional and cognitive development is well-documented in the literature (40). To date, there are no studies comparing mothers with peripartum depression alone to depressed mothers with comorbid anxiety disorders. As comorbidity is associated with greater impairment and symptom severity related to the primary diagnosis, comorbidity in mothers might raise their offspring's risk of developing internalized disorders even more than has been noted in conjunction with depression alone (41).

N = 168 families (n = 56 per subgroup and n = 84 per study center in Heidelberg and Munich) will be recruited through inpatient and outpatient centers as well as maternity hospitals in Munich and Heidelberg. A drop-out rate of 20% is expected.

This study is designed to assess peripartum depressed mothers with and without comorbid anxiety disorders according to DSM-5, fathers and their infants, as well as a healthy control group at four measurement points over the first 2 years (T1: 3–4 months postpartum, T2: 12 months postpartum, T3: 18 months postpartum, an T4: 24 months postpartum; see **figure 3** for details). Besides the evaluation of parental psychiatric status at all measurement points, parent-infant interaction will be videotaped and coded according to the Coding Infant Behavior Scales (CIB) (42). To determine infant stress-reactivity, cortisol will be extracted from infant saliva, which is collected before (C1), 20 min (C2), and 30 min after the interactional episodes.

At the age of 12 months, declarative and imperative point production and understanding (43), as well as imitation of object-related (44) and intransitive action skills (45) as predictors of later social-cognitive development will be assessed. This will be expanded upon at 18 months by an assessment of empathy (46) and intention reading (47) as milestones of constructive social behavior and mindreading, and at 24 months of empathy, executive functioning (gift delay (48) and reverse categorization (49), language abilities (SET-K (50), and child fearfulness [spider task (51)]. The measurements taken at 24 months will serve as our primary outcome measures for the longitudinal study. Infant socio-emotional development will be rated by mothers, fathers and additional caregivers via the Child Behavior Checklist/Caregiver Teacher Report Form (52, 53) at T3 and T4. Cognitive development will be assessed at T4 using the Bayley Scales of Infant Development-III (54).

This sub-project will yield new insights into the specific effects of maternal depressive and comorbid depressive and anxiety disorders disorders on different aspects of the infant's social and cognitive development, as well as on the interaction mechanisms contributing to this process. We need a deeper understanding of the underlying mediation effects to develop future prevention or intervention approaches, since research on comorbid depressive and anxiety disorders and their impacts on the parent-infant relationship and infant development is still lacking. Therefore, better understanding will enable us to suggest starting points for further research into this area. The multi-perspective approach is novel as we will be analyzing socio-emotional and cognitive parameters together and also include fathers in this study. This is unique as no other research projects have studied these research questions using additional biopsychological measures. Research of this kind is particularly important to help us to better understand adverse developmental pathways and to pave the way for future implementation of prevention and intervention programs.

COMPARE-Work: Parental Work Environment as a Risk Factor: Spillover-Crossover Effects From Parents to Children

Work has a central meaning to our lives: it shapes our identities, offers us social support and appreciation, and helps us to achieve collective goals (55). How important work is and the effects it has on children can be illustrated by the example of being unemployed. Studies indicate that the children of unemployed parents are disadvantaged: their birth weight is lower, they reveal slower growth and have more accidents (56, 57), they exhibit more self-destructive behavior, and that they tend to drop out of school and are more frequently unemployed as adults ["intergenerational unemployment"; (57)]. Similar effects are to be expected if working conditions are detrimental (58); then long-term negative health-effects occur that can spread over to the family and thus be a risk factor for the mental health of children. A negative downward spiral could be activated when parents, because of being emotionally exhausted for example, cannot provide their children any support when doing the homework while their children absorb their parents' work-related worries. Parents' adverse working conditions may have a negative impact on both their children's school performance and mental well-being. To the best of our knowledge, no research so far has attempted to clarify this process.

A first aim of this project is to explore whether mentally ill as compared to healthy parents are confronted with more severe working conditions, namely that they are confronted with more stressors (e.g., time pressure), possess fewer resources (e.g., lack of social support) at their workplaces, and report on a worse career development (e.g., job loss). Across countries findings of the (59), p. 30) revealed that the "average income of people with a moderate mental disorder is around 90% of the total working-age population, and it is 80% or less (...) for those with a severe mental disorder." These statistics suggests that also mentally ill parents face a high unemployment risk or work more frequently in precarious jobs. Moreover, research has identified two different ways in which work demands or strain are carried over: spillover and crossover. Adverse working conditions can strain parents, and such strain is expected to be transferred from the work domain toward the family, rendering it a risk factor for family life (work-family conflicts) and hence for the mental health of children. This "spillover" is the within-person, across-domain transmission of strain from one area of life to another (60). The next step following spillover-processes from work to family is to explore parent-to-child crossover, which has not been studied to date. Crossover represents a phenomenon, where experiences of stress and strain are transmitted between individuals (61). The core assumption is that one's own stress and strain experiences have an impact on others in close surroundings. To clarify the underlying mechanism in the spillover-crossover-process (62), we will try to shed light on the mediators. As proposed in the concept of emotional contagion (63), negative states are transferred easily and automatically. Moreover, crossover can be of cognitive nature through shared social cognition, a factor that can be a source of information to facilitate the interpretation of ambiguous situations. Hence, the way crossover from parents to children works—driven emotionally or cognitively—will also be researched.

In this project, working conditions of parents of children of the RCT [COMPARE-family; see (24) in this research topic] and parents of a representative group of school children not differing in basic demographic variables (assessed together with COMPARE-school) will be compared. We will assess task- and job-related working conditions (e.g., time pressure, autonomy, interruptions at work, and skill utilization) with the Instrument for Stress Oriented Task Analysis [ISTA; (64)] and the Copenhagen Psychosocial Questionnaire [COPSOQ II; (65)]. Further, working conditions on the social level (e.g., social stressors, social support, appreciation) will be explored using well-established measures. To explore spillover, besides working conditions, strain (work-family-conflict, need for recovery) and well-being (life satisfaction, mood, emotional exhaustion) of both groups of parents will be assessed via existing self-report measures. To study crossover, also the children will be investigated regarding their well-being (life satisfaction, mood, emotional exhaustion) and their closeness to their parents. To detect the underyling transmission process, an *ad-hoc* measure that reflects emotional vs. cognitive processes in crossover for children will be developed and pre-tested.

Using the software G-Power and specifying alpha as 5% and statistical power to equal at least 80% and applying MANOVA to test for differences between mentally ill vs. healthy parents' working conditions considering parents' gender, a minimum sample size of 200 parents is suggested. However, to test the more complex models (SEM) detecting the spillover-crossover process, a minimum sample size of 300 parents would be optimal to detect medium-sized effects. As higher attrition rates are not uncommon for longitudinal research in the Occupational Health Psychology field, we plan to contact a total of 887 mothers and/or fathers. Depending on the parents' consent to participate—which we expect to be about 50% (aiming at 450 parents in total)—the exact sample size will be somewhat lower, though.

In sum, this project will deepen our knowledge of the TTMD model. By assessing parental working conditions, an essential element in the TTMD model can be explained. Various stressors and resources in the working context of both mentally ill and healthy parents as well as work-related strain and work-family conflict indicators will be explored, characterizing the aforementioned spillover-crossover process in (all) its complexity.

COMPARE-School: Psychosocial Adjustment, School Performance, and Subjective Well-Being in Children of Mentally Ill Parents

Most studies have focused on the mental health and child psychopathology of COPMI and just a few studies have focused on other relevant child outcomes proposed in the TTMD, such as academic attainment, psychosocial functioning or subjective well-being by investigating an adequate control sample of children with healthy parents [e.g., (66–68)]. One main aim of this subproject is to compare a school control sample to the COMPARE-family intervention groups [see (24) in this research topic]. This control sample will be recruited in elementary and secondary schools during regular school lessons. School children comparable in age to the RCT [COMPARE-family; see (24) in this research topic] will be examined with the same tests as COPMI. All tests will be given during regular school lessons to entire school classes. About $N = 900$ school children will initially be investigated. Within this sample, a propensity score matching [see (73)] will be performed controlling for age, gender, and parental socioeconomic background to obtain a school control cohort comparable to and as large as the COPMI group.

Different indicators of academic achievement will be assessed (standardized achievement tests in reading and mathematics, school grades). Social skills will be measured via self-reports as well as via parental assessments. Subjective well-being will be examined by investigating both cognitive as well as affective variables regarding life as a whole as well as regarding family, school, and peers (69).

Moreover, there is a serious lack of knowledge how parental mental disease affect lower educational attainment. Besides parenting behaviors that are related to child learning (such as homework support) and that in turn affect academic achievement, other mediating child characteristics might also be of influence (e.g., temperament and cognitive abilities). In this context, the child's school values and academic expectations are especially interesting, as research has demonstrated that these variables both differ between children of mentally ill and healthy parents and influence academic achievement (70, 71). Besides other child outcomes such as psychosocial functioning and subjective well-being, especially parenting skills and parenting styles in general might be important process variables.

Research has shown that some COPMI reveal no detrimental outcomes regarding their educational attainment, psychosocial adjustment, or subjective well-being [e.g., (6)]. There is little research to date comparing those children with children suffering from their parent's mental health disorder to identify protective and risk factors. The TTMD model describes various potentially-relevant vulnerability factors such as temperament, cognitive skills or the parenting style and parenting behavior. These vulnerability factors will be additionally examined within COMPARE-school.

Finally, treatment-related changes in educational attainment, psychosocial functioning, and subjective well-being will be examined. The aforementioned outcomes will be assessed at T2 (2nd baseline), T3 (post-test), and T4 (6 months follow-up) in the cohort of COPMI and in the representative control group [for a full description of the RCT-design see (24) in this research topic].

The outcome variables measured in COMPARE-school are closely related to children's health, and provide a holistic perspective onto a child's healthy development. Our project will yield important insights into the specific transmission profiles proposed in the TTMD. In sum, we will contribute to both model testing and the intervention emphasis of the main COMPARE project.

TRANSFER INTO THE ROUTINE PROVIDER SYSTEM

The results of the COMPARE consortium will be made available to the larger scientific community via peer-reviewed publications in scientific journals, presentations at scientific meetings and presentation on the COMPARE website. Partners will be encouraged to publish in "Open Access" journals whenever appropriate, ensuring accessibility to the widest readership worldwide. Throughout the study and beyond, patients, parents, and carers (stakeholder board) will be kept informed about the study through the COMPARE website, flyers, newsletters, and personal contacts. Politicians, public health services, and stakeholders will be informed via teaching seminars and conferences on the background and results of the present study, thus improving public policy and health care decisions whose aim it is to prevent and treat COPMI. Two large health care insurances, the "Allgemeine Ortskrankenkasse/AOK" and "IKK Südwest" have ensured us to disseminate results to their insurance holders. Existing networks (i.e., Kindernetzwerk Deutschland, Diakonisches Werk Baden, unith e. V., BVKJ, DGPS, DGKJP, DGPPN, DGKJ) will be used to disseminate our results to relevant target groups. A joined patient conference on parenting skills is planned. Since all participating Psychology Departments have outpatient departments for children/adolescents and adults and are members of unith e. V. (a collaboration among university-based therapeutic training institutions), the implementation of results in ongoing and future practice is warranted, especially since many of the researchers involved already teach at the respective therapy training institutes and unith e. V. supports this project. The consortium's members also include experts from child and adolescent psychiatry as well as pediatrics, meaning our results will be implemented in those relevant fields as well. As we will be cooperating with a large adult psychiatry institution having many clinics throughout Germany (Schön-Kliniken), our results are certain to be implemented within a major stakeholder. Results will be relevant for and thus included in treatment guidelines as well as in psychology textbooks. Furthermore, as we have included experts from educational psychology will help transfer our findings to educational practice.

DISCUSSION AND LIMITATIONS

COMPARE with its subprojects family, emotion, interaction, work, and school, is the first comprehensive research programme on COMPI in Germany, as to date there is neither standard care nor systematic research with respect to this highly vulnerable group.

The COMPARE-family project is the central RCT testing the effects of high quality parental CBT on the children and whether additional PPP will result in incremental effects above and beyond CBT alone [for details see (24) in this research topic]. Effects of this trial as well as baseline data is informative for the other subprojects, as based on this trial the different domains of the TTMD (influences of emotion processing, parent-child-interaction environmental influences such as work and school) can be tested. So far the TTMD model has not been comprehensively tested.

The COMPARE-emotion subproject focuses on emotion processing and emotion regulation as potential child factors for the transgenerational transmission of mental disorders. Furthermore, these processes will be assessed as predictors and outcome measures for the RCT. Emotion processing and emotion regulation will be assessed with the help of different approaches, comprising behavioral measures, peripheral physiological markers, and neuro-imaging techniques. Thus, we expect to accomplish a comprehensive understanding of relevant child factors that display a significant role in the TTMD model. Moreover, the identification of underlying processes is equivalent to the identification of targets for effective intervention in the future.

The COMPARE-interaction subproject focuses on the long-term consequences of maternal psychopathology during the peripartum period on infant development, especially on socio-emotional and cognitive development. Special attention will be paid to the mediational effect of parent-infant interaction and infant stress reactivity on the relationship between the maternal disorder (depressive, comorbid, healthy controls) and infant outcome. Moreover, it will significantly contribute to the model testing in the model's following domains: mentally ill parent, family context, other parent, parent-infant interaction, infant outcome. This sub-project will yield new insights into the specific effects of maternal mental disorders on different aspects of the infant's social and cognitive development, as well as on the interaction mechanisms contributing to this process. It is of great importance to gain a better understanding of the underlying mediation effects to develop future prevention or intervention approaches, since research on comorbid depressive and anxiety disorders and their impacts on the parent-infant relationship and infant development is still lacking.

COMPARE-work explores the working conditions of mentally ill parents in contrast to those of healthy parents, identifies how these working conditions impact on work-related strain and how this causes strain in family life (work-to-family spillover). The comparison of strain and well-being levels in both groups of parents as well as their children enables us to detect transmission effects (parents-to-children crossover), thereby focusing on the specific transmission mechanisms (mediators), and finally analyzes whether treatment buffers or aggravates the spillover-crossover process (moderators). With respect to the preventive intervention, the consideration of parental work-environments is a significant moderator of therapy outcome. More or less supportive or adverse work-environments can be seen as framing conditions (partly outside the control of a person) shaping the opportunities of a therapeutic intervention at least partly; in particular in such cases where working conditions cause, strengthen or work to maintain mental diseases.

COMPARE-school focuses on different indicators of academic achievement, social functioning, and subjective well-being. A main aim is to compare the children of mentally ill parents with a healthy school control sample in terms of academic and psychosocial outputs and to seek variables that explain those differences. Moreover, we will test whether the specific type of parental mental illness and the mentally ill parent's gender are relevant concerning the aforementioned child outcomes. To create targeted interventions for children suffering from their parent's mental disease, we will additionally focus on those children with a low risk for developing health problems themselves and examine how they differ from those who have mental health problems. This comparison will enable us to explore approaches that can support children suffering from their parent's mental disease. Treatment-related factors in academic and psychosocial outcomes will be examined as well. Taken together, we will examine the theoretical model underlying COMPARE in terms of different outcomes other than psychopathology in COPMI and we will test for intervention effects by taking another adequate control sample into account.

However, there are also some limitations to be considered.

First of all, due to funding decisions, some aspects of the TTMD-model, such as (epi-) genetics and somatic outcomes will not be part of the study. However, if this study proves to be successful in the identification of risk mechanisms, this might provide a strong basis for follow-up (epi-)genetic projects on this topic.

Second, the research programme with a clinical trial at the center [see (24) in this research topic] and the four add-on projects puts high demands on the participating patients and their families that are already under increased stress due to the parental mental illness. On the other hand, the intensive assessment of different areas of life that might be impaired or prove to be associated with resources will substantially increase our knowledge on when to provide which interventions in what areas of life for whom.

ETHICS STATEMENT

The study is carried out according to the Good Clinical Practice (GCP) guidelines, the Declaration of Helsinki and its later supplements and local legal requirements. The lead ethics committee at the department of psychology at Philipps-University Marburg approved the study procedure and all study documents. A positive ethics committee vote is required at a study site, before the inclusion of a first patient at the respective site.

DISSEMINATION

Via peer-reviewed publications in scientific journals, the results of this study will be made available to the scientific community. Using PsychData all primary data will be made available for re- and meta-analyses. Politicians, public health services and stakeholders will be informed throughout the study and beyond, thus, improving public policy and health care decisions concerning preventive interventions and treatments for COPMI.

AUTHOR CONTRIBUTIONS

HC has drafted the main manuscript. CR and A-LZ wrote the section on COMPARE-interaction and commented on the whole manuscript. KO wrote the COMPARE-work section and commented on the whole manuscript. RS and LW wrote the COMPARE-school section and commented on the whole manuscript. SW, RS, and CS wrote the COMPARE-emotion section and commented on the whole manuscript. DE and CB are the study advisories, read the manuscript, and commented. JK, CK, and MK are the trial biometricians, read, and commented on the manuscript.

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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

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Background: Mental health problems are highly frequent, as well as being associated with enormous societal and economic costs and significant disability-adjusted life years. Children of parents with a mental illness (COPMI) are at a tremendously increased risk to develop disorders themselves. According to the literature, parental mental disorders launch a wave of risk factors that in turn predict the emergence of psychological problems in the offspring, and effective treatment of the parental disorder has been associated with reduced child psychopathology (launch and grow assumption). Furthermore, studies focusing on parent-child interaction demonstrate generally poorer parenting skills in parents with mental disorders, and the enhancement of such skills has been a significant mediator in improving child outcomes (parenting assumption).

Objective: To implement a preventive intervention for COPMI with the aim of interrupting the transmission of mental disorders in children of a parent with mental disorders. An RCT will compare state-of-the-art cognitive behavioral therapy (CBT) for a parent with mental disorders to CBT plus the Positive Parenting Program (Triple-P), a well-established and evidence-based program that enhances parenting skills.

Methods: A total of 634 patients seeking treatment in 8 outpatient clinics in Germany and their children will be included between January 2018 and April 2021 in the study. We use (clinical) interviews and self- as well as other-report questionnaires to assess the families at four main measurement points [T1: beginning of waiting period for psychotherapy treatment (duration of waiting period depends on usual waiting period in the study center: multiple baselines), T2: begin of parental psychotherapy, T3: post-assessment, T4: 6 months follow-up]. The total observation period will be 39 months. The patients will be randomly assigned to either the control condition (25 to 45 CBT sessions) or the experimental condition (25 to 45 CBT sessions + 10 Triple-P sessions). For evaluating the treatment process, the patients and clinicians will also be assessed after each treatment session. Furthermore, there will be a continuous assessment and report of adverse events during treatment.

Discussion: This trial will be the first ever to address the launch and grow as well as the parenting assumption in one study and to establish effects of the two different interventions on children's health. Our study will also likely be the first one to provide data on the comparative cost-effectiveness and will therefore provide essential information relevant for the potential implementation of such programs. The structure of the RCT will allow us to establish effects of the parental disorder(s) with/without comorbidities on children's health, to test assumptions of the trans-generational transmission model of mental disorders and bi-directional influences of different treatments on the model and to analyze specific transmission mechanisms. A deeper understanding of risk mechanisms will reveal specific transmission profiles that will result in the early detection of and effective reduction in risk factors and thus improve the health of the children at risk.

Ethics: The study is carried out according to the Good Clinical Practice (GCP) guidelines, the Declaration of Helsinki and its later supplements and local legal requirements. The lead ethics committee at the department of psychology at Philipps-University Marburg approved the study procedure and all study documents. A positive ethics committee vote is required at a study site, before the inclusion of a first patient at the respective site.

Dissemination: Via peer-reviewed publications in scientific journals, the results of this study will be made available to the scientific community. Using PsychData all primary data will be made available for re- and meta-analyses. Politicians, public health services, and stakeholders will be informed throughout the study and beyond, thus, improving public policy and health care decisions concerning preventive interventions and treatments for COPMI.

Trial Registration: DRKS-ID: DRKS00013516 (German Clinical Trials Register, https://www.drks.de/drks_web/navigate.do?navigationId=trial.HTML&TRIAL_ID=DRKS00013516)

Keywords: children of mentally ill parents, transgenerational transmission, randomized controlled trial, prevention, intervention, mental disorders, parenting training

INTRODUCTION

Children of mentally ill parents (COPMI) are at a high risk of developing severe mental illness (SMI) themselves and are likely to be the next generation of mentally ill patients (1). There are studies pointing to the fact, that the parental mental disorder launches a wave of risk factors that in turn predict the emergence of psychological problems in the offspring (2). Numerous studies have shown that a parental mental illness is a powerful risk factor for the development of a SMI in children (OR in the BELLA study of 2.4) (1, 3–8). Long-term studies were also able to show that COPMI have a higher life-time risk of developing SMI themselves (ranging from 41 to 77%) with subclinical symptoms emerging earlier and more often (5, 6). The treatment of the parental disorder has been associated with improved outcomes in COPMI (7, 9–13), although there are few studies on such effects (7, 14), and they typically target the same symptoms in the child as the parent's, while such specific transmission of disorders is not typical for COPMI (1). A recent meta-analysis (k = 9) by Cuijpers et al. (14) on the effects of psychological treatment of maternal depression on children's psychopathology resulted in an overall effect-size of $g = 0.40$. However, the studies included were very heterogeneous ($k = 5$ targeting women with post-partum depression; $k = 4$ targeting pregnant women or mothers of young children or mothers of children with psychological/psychiatric problems/disorders), and only 2 studies explicitly applied Cognitive Behavioral Therapy (CBT) as an intervention that resulted in an overall effect of $g = 0.31$ (14). An earlier meta-analysis by Siegenthaler et al. (15) on preventive interventions for children of mentally ill parents demonstrated a significant relative risk reduction of 40 % for the same disorder as the parents', and overall small effects for children's internalizing ($g = -0.22$) and externalizing ($g = -0.16$) symptoms. This analysis included interventions targeting children though and not specifically those that assessed parental psychotherapy effects on their children (15). Our own meta-analysis on preventive interventions for COPMI (16) resulted in effect sizes similar to those of Cuijpers et al. (14) for young (up to 5 years of age) children ($g = 0.31$), and overall smaller effects for older children ($g = 0.14$) that equal those of Siegenthaler et al. (15). Different longitudinal studies on parental anxiety and depressive disorders present heterogeneous effects of parental treatment on children. A 6 year prospective longitudinal study on the effects of parental panic treatment demonstrated that parental treatment is a significant predictor of children's anxiety symptoms ($d = 0.49–1.09$ for different parental psychopathology predictors) (7). The Sequenced Treatment Alternatives to Relieve Depression (STAR*D) Child study was designed to examine the relation between maternal remission from depression and children's functioning and psychopathology. The study demonstrated differential effects on child psychopathology in early, late, and non-remitting mothers, with early remission being associated with reduced child externalizing problems (∼5% of Child Behavior Checklist (CBCL) externalizing symptoms explained) (13); similar results have been obtained in another large longitudinal study (9).

Studies focusing on the parent-child interaction tended to demonstrate poorer parenting skills in parents with mental disorders (17–20). The enhancement of such skills has been identified as a significant mediator in improving child outcomes (21). The Positive Parenting Program (Triple P) is a well-established program to enhance parenting skills in parents of children aged 0–16 years (Triple P Kids and Triple P Teens). Universal prevention effects have been established for the Triple P Kids program (22), as well as specific effects for child psychopathology ($d = 0.473$) (23, 24) that differ for mothers ($d = 0.61$) and fathers ($d = 0.42$). The effectiveness of the Triple P Teen program was also shown (25). Studies explicitly testing the parenting assumption in conjunction with the launch and grow assumption are lacking so far.

Regarding psychotherapy research, there have been suggestions that moving from an approach comparing an active treatment with a control group ("does it work?") to one that examines putatively active treatments resulting in relative questions ("which works best?" or "how do the treatments differ?") is advisable. If demand artifacts and differentiating non-specific from specific treatment factors are included, we arrive at this formula: Treatment A $= E + D_T + T_{NS} + T_{S(A)}$ and Treatment B $= E + D_T + T_{NS} + T_{S(B)}$ with E = all extraneous factors; D_T = demand characteristics treatment; T_{NS} = non-specific treatment factors; T_S = characteristic-specific treatment factors. Thus, the relative difference between the two active agents in the proposed study $T_{S(A)}$ = CBT and $T_{S(B)}$ = CBT+PPP estimates how the two treatments differ and enables us to determine relative effects (26). Such head-to-head studies on preventive interventions for COPMI assessing differential effects have been extremely rare; the classic format are "does it work?" studies (15).

Thus, we aim to implement a preventive intervention for COPMI with the aim of interrupting the transmission of mental disorders in children of a parent with a mental disorder. The preventive intervention is planned as a two-arm RCT to establish whether strengthening parenting skills results in incremental COPMI effects above and beyond state-of-the-art CBT for parents. Including an economic evaluation alongside the clinical trial, the results of this study will have an effect on the decision making process on resource allocation for this highly vulnerable group of children of mentally ill parents.

The RCT will thus target the following hypothesis: (1) the treatment of the parental disorder will result in improved child outcome (1st arm: CBT) (2, 7, 12), and (2) the parenting skills of parents with mental illness are impaired and enhancing such skills leads to better child outcomes, thus incremental effects will become apparent in the 2nd arm: CBT+Triple P (17, 18, 20, 27, 28).

Further research questions are: Is the clinical outcome associated with reduced direct medical, direct, and indirect non-medical costs? Are CBT and CBT+PPP associated with improved quality of life for parents and children? Are CBT and CBT+PPP associated with increased psychopathology knowledge of parents and children? Is CBT+PPP associated with higher parenting skills than CBT alone? Are the effects independent of type of diagnosis, comorbid disorders, and psychopharmacology?

METHODS

Design

The planned study is a prospective, multicenter, confirmatory, randomized controlled phase III-trial with two parallel arms comparing the effects of state of the art CBT (control intervention), and CBT + Triple P (experimental intervention) for parents with a mental illness on their children. The study is coordinated by the Department of Psychology, Clinical Child-, and Adolescent Psychology at the Philipps University Marburg (UMR).

After 3 months of study preparation (October–December 2017), a 15 months recruiting period has started in January 2018. The assessment period (first patient in until last patient out) will last for a total of 39 months. Six months are scheduled for data freeze, data cleaning and analysis. Thus, the duration of the whole trial is 48 months (see **Figure 1**).

Using the same instruments (self- and other-report questionnaires as well as (clinical) interviews) each time, patients, and their families will be assessed at four main measurement points (see **Figure 2**): pre-assessment 1 (T1, beginning of waiting period for psychotherapy treatment (duration of waiting period depends on usual waiting period in the study center: multiple baselines), pre-assessment 2, (T2, beginning of parental psychotherapy), post-assessment (T3, after parental psychotherapy), follow-up-assessment (T4, 6 months after parental psychotherapy). Altogether, assessment time comprises between 4 and 5 h for each main assessment (T1, T2, T3, T4) for each family. Patients and therapists are also assessed after each treatment session for treatment fidelity and satisfaction. Every fifth session, the remission status of the patient is also assessed.

Participants

A total of 634 parents with a mental illness (patients) shall be included in the study. The following criteria must be met for patients and their families to be included in the study: (1) patient seeks outpatient psychotherapeutic care, (2) patient currently meets diagnostic criteria for DSM-5 disorder (29), and (3) patient is caring for at least one child between the ages of 1.5–16 years. Patients and their families will not be included in the study if (1) patient is already in psychotherapeutic treatment, (2) patient needs acute inpatient treatment (e.g., acute risk of committing suicide or acute psychosis), (3) all children fulfill criteria for a severe mental illness and are in need of prompt treatment, (4) patient uses benzodiazepines continuously (intermittent drug use less than once every 2 weeks is allowed), or (5) family has insufficient German language skills. No further exclusion criteria will be applied. Hence, we seek to establish effects of parental psychotherapy on children in a naturalistic setting and want to specify the effects of the transgenerational transmission to test the launch and growth assumption. Patients in need of acute inpatient treatment are referred to cooperating hospitals. For children in need of prompt care, treatments are initiated at the study sites or the families are referred to other local specialists/psychiatric hospitals.

The study will be conducted at eight university outpatient clinics throughout Germany: Bielefeld, Bochum, Gießen, Landau, Leipzig, Mainz, Marburg, and Munich. All cooperating sites have established outpatient clinics for research and teaching as well as psychotherapy training institutes and are well-known specialized treatment centers for psychotherapy. The patients are primarily recruited from the university outpatient clinics at each study site. The study centers have continuous experience in conducting psychotherapy studies with recruitment of large patients numbers (30, 31). The patients to be recruited per center and fulfilling all inclusion criteria will be attainable over a 15-month acquisition period at all study sites without the need to change the existing infrastructure. If possible, all children between 1.5 and 16 years of age (at T1) and a partner living together with the family shall be included in the study. A legal guardian not living together with a child can upon request be offered to participate in the study and to complete questionnaires about the respective child. Inclusion only takes place if the family participates voluntarily in the study. There is no compensation for study participation.

Interventions

All patients will receive between 25 (short-term) and 45 (long-term) weekly or bi-weekly sessions of individual state of the art Cognitive Behavioral Therapy (CBT). A high frequency therapy with more than one session per week is also allowed. Depending on the amount and the frequency of therapy sessions, the treatment will usually last between 6 and 12 months. The number of sessions and therapy duration will be documented, thus, allowing us to include therapy duration and number of sessions as a moderating factor in the analyses.

CBT can be considered the current "treatment of choice" for mental disorders in general (32). Central elements are psychoeducation that provides a framework for psychotherapy, cognitive components (i.e., debate of dysfunctional cognitions alias Beck), as well as behavioral interventions (i.e., exposure). As all study sites are outpatient clinics for research and teaching located at university psychology departments, they commit to the highest possible state of the art standard for CBT as outlined in "proceedings in psychotherapy/Fortschritte der Psychotherapie" of which Prof. Rief (Co-PI of COMPARE-family) is an editor (33). As the study shall be conducted as naturalistic as possible and a broad range of disorders shall be included, no specific study manual has been created, rather, the respective therapy manuals of the proceedings in psychotherapy will be used in this study.

Patients randomized to the experimental group will receive 8–10 additional group or individual sessions of the Positive Parenting Program (Triple P) parallel to the last third of the psychotherapy sessions to test whether the enhancement of parenting skills will result in improved child outcome, as suggested in different studies (17, 18, 20, 27, 28), above and beyond parental CBT. Triple P is a well-established, widely used and evidence-based program to enhance parenting skills. The Triple P Kids program can be used in groups or individual sessions with parents of children aged 0–12 years (34, 35) and the Triple P Teens program in groups with parents of children aged 12–16 years (36). The Triple P elements are: (1) promotion of a positive parenting style, (2) promotion of child development, (3) dealing with problem behavior, (4) behavior training sessions (34–36).

Outcomes

Primary Outcome Measure

The primary outcome is the children's pathology according to the [(Caregiver-) Teacher Report Form (C-)TRF, caregiver/teacher version of the Child Behavior Checklist] score between baseline 2 (T2) and post treatment (T3) as well as between T2 and the 6-month follow-up (T4) after end of treatment (37, 38) as the literature shows that mentally ill parents can be biased in rating their children's symptoms (39). Further, teacher ratings have been shown to demonstrate greater predictive validity in the long term ratings (40). In comparison to shorter questionnaires [e.g., Strengths and Difficulties Questionnaire (SDQ) (41)], the CBCL also produces more variance. As the CBCL is used in various studies as a primary outcome measure (15), we will be able to compare our data to a broad range of other results. Further outcome measures are mental disorders according to the Diagnostic Interview of Mental Disorders for parents

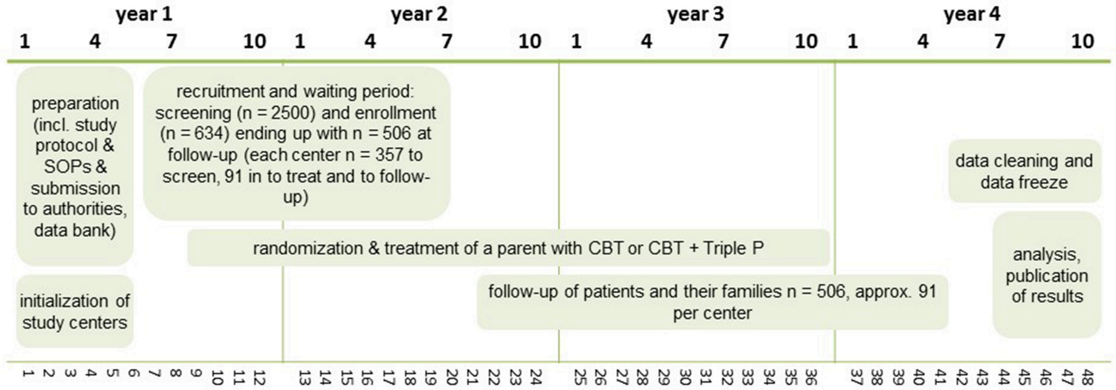

FIGURE 1 | Study design.

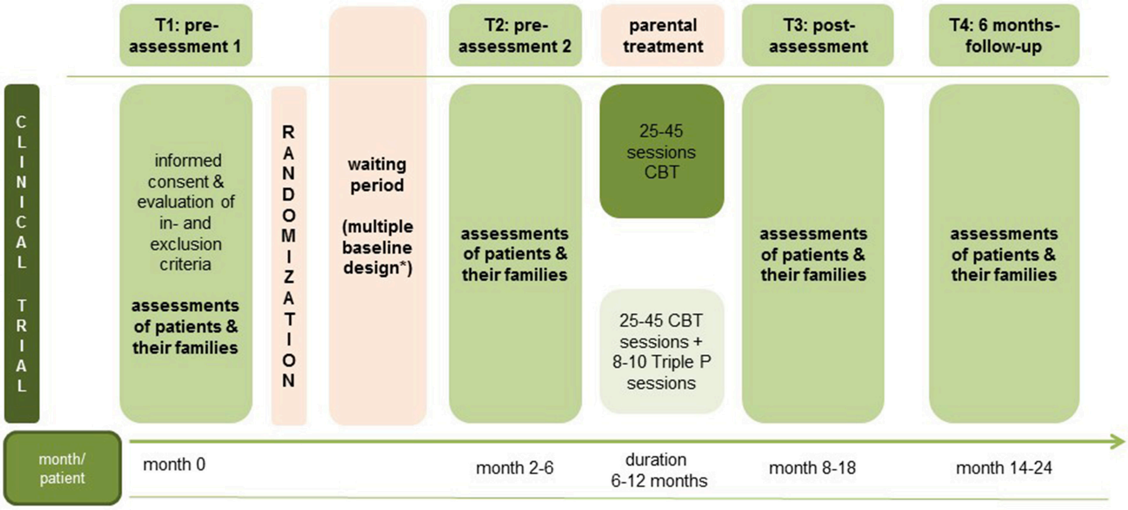

FIGURE 2 | Study assessment points. *Waiting period can vary according to the regular waiting period in the study centers.

and children (DIPS and Kinder-DIPS) (42–44) respectively for children under the age of 6 years the Structured Interview for Preschool Ages (SIVA) (45) and the parent rating of the children's pathology according to the CBCL (for detailed descriptions see below). Thus, we will be able to validate the teacher ratings with the Kinder-DIPS diagnoses and can compare the concordance of the teacher and parent ratings.

Secondary Outcome Measures

Children's and parents dimensional severity index of a broad range of mental disorders according to the DSM-5 based clinical interview (Kinder-)DIPS (42–44) respectively the SIVA (45), parent's psychopathology according to the Brief Symptom Inventory (BSI) (46), personality traits (PID-5-BF) (47), children's psychopathology according to the CBCL-parentversion (37, 38), parenting skills (EFB) (48), parental stress (ESF) (49), children's and parents knowledge about mental disorders [semistructured interview adapted from Beardslee and Röhrle, (50)], direct medical costs and direct and indirect non-medical costs (TiC-P) (51), health-related quality of life in parents [EuroQoL/EQ-5D-5L, AQoL-8D (52, 53)], health-related quality of life in children (KIDSCREEN-10) (54, 55) (for details see **Table 1**).

Instruments

The (Kinder-) DIPS is a diagnostic interview for mental disorders and is available for the diagnoses of mental disorders from age 6 to adulthood according to ICD-10 and DSM-5. The SIVA is a diagnostic interview for mental disorders for preschool ages according to ICD-10 and DC: 0–5. A translation table to DSM-5 diagnoses has been created for this study. For parents we use the DIPS; for their children the Kinder-DIPS or SIVA. Conducting the interviews with the patient takes around 60–90 min as well as the child assessment with the Kinder-DIPS or SIVA for each

TABLE 1 | List of diagnostic domains and measurements.

	Who?	Where?	T1	T2	Intermediate	T3	T4
Eligibility screening	Family	Local	x				
Socio-demographics	Parents	Local	x				
Structured clinical interview with the patient (DIPS)	Patient	Local	x	x		x	x
Structured clinical interview for children (Kinder-DIPS/SIVA)	Parent	Local	x	x		x	x
Brief Symptom Inventory (BSI)	Parents	Online	x	x	x (patient)	x	x
Personality traits (PID-5-BF)	Parents	Online	x	x		x	x
Child Behavior Checklist (CBCL)	Parents	Online	x	x		x	x
(Caregiver-) Teacher Report Form [(C-)TRF, caregiver/teacher version of the CBCL]	Teacher	Online	x	x		x	x
Parenting Skills (EFB)	Parents	Online	x	x		x	x
Parental Stress (ESF)	Parents	Online	x	x		x	x
Knowledge about mental disorders	Parents, children	Local	x	x		x	x
Direct and indirect costs in children and parents (adapted TiC-P)	Parents	Online	x	x		x	x
Health-related quality of life in parents (EuroQoL/EQ-5D-5L, AQoL-8D)	Parents	Online	x	x		x	x
Health-related quality of life in children (KIDSCREEN-10)	Parents, children (>8)	Online	x	x		x	x
(Serious) adverse events	Parents, children	Local		x	x (patient)	x	x
Psychopharmaka and other drugs	Parents, children	Local	x	x	x	x	x
Concomitant interventions	Parents, children	Local	x	x	x	x	x

child. Different studies have shown good quality criteria for these interviews (43, 45).

The Achenbach System of Empirically Based Assessment (ASEBA) (56) consists amongst others of questionnaires for preschool (C-TRF and CBCL 1,5-5, consisting each of 100 problem items) and school-ages (TRF and CBCL, consisting each of 113 problem items). Answers are rated on a three point Likert-scale (0 = Not true to 2 = Very True or Often True) and can be scored on different subscales, the second order scales internalizing problems and externalizing problems as well as on a total problem scale. Studies have reported good to very good internal consistency with $r > 0.86$ for the second order scales and the total problem scale of the preschool age-versions (37) and Cronbach's alpha >0.80 for the second order scales and at least.93 for the total problem scale in the school age-versions (38). In the current study we use the parent (CBCL) and teacher (TRF) ratings of the ASEBA.

The Brief Symptom Inventory (BSI) is a self-report questionnaire consisting of 53 items that are rated on a five point Likert-scale (0 = not at all to 4 = very much). Answers are scored on nine Primary Symptom Dimensions and three Global Indices. Internal consistency with Cronbach's alpha > 0.70 for the subscales and > 0.90 for the Global Indicee GSI is good to very good (46).

The Personality Inventory for DSM-5-Brief Form (PID-5-BF) is a self-report questionnaire assessing 5 personality traits (negative affect, detachment, antagonism, disinhibition, and psychoticism). The 25 items are rated on a 4 point Likert-scale (0 = very false or often false to 3 = very true or often true). An average score for each domain and an overall score can be calculated with higher scores indicating greater dysfunction. The average domain and overall personality dysfunction scores were found to be reliable in the DSM-5 Field Trials (47).

The Elternstressfragebogen (ESF) is a German self-report questionnaire with 38 items assessing parental stress. The answers are rated on the four scales Parental Stress, Role Restriction, Social Support, and Partnership. The internal consistency is good with Cronbach's alpha > 0.76 (49).

The Erziehungsfragebogen (EFB) is the German adaptation of the English Parenting Scale (57). The self-report questionnaire consists of 35 items and can be rated on the scales Overreactivion, Laxness, and Verbosity as well as on a total score. The internal consistencies of the scales Overreaction, Laxness, and the total score is acceptable to good with Cronbach's alpha > 0.74. For Verbosity the internal consistency is lower with Cronbach's alpha > 0.59 (48).

To assess knowledge about mental disorders, a semi structured interview was adapted from Beardslee and Röhrle (50). Domains of the interview are knowledge about mental disorders in general, knowledge about the primary diagnosis of the mentally ill parent, causes of the mental illness, coping with the mental illness and communication. Conducting the interviews separately with all family members over the age of 6 years takes 15 min each.

The questionnaire on healthcare consumption and productivity losses for patients with a Psychiatric disorder (TiC-P) is a comprehensive and widely used self-report questionnaire focusing on establishing direct medical costs and indirect costs (e.g., productivity losses due to absenteeism and presenteeism) (51). The TiC-P is a feasible and reliable instrument for collecting data on medical consumption and productivity losses in patients with common mental health conditions (58). The TiC-P has been previously adapted for use in Germany (59). For this study, we adapted the TiC-P for use in children.

The EQ-5D-5L is a widely used instrument to calculate quality-adjusted life years (QALYs) (52). The EQ-5D-5L comprises 5 items covering 5 domains (mobility, self-care, usual activities, pain/discomfort, and anxiety/depression), each of which is rated as causing "no problems," "slight problems," "moderate problems," "severe problems," and "extreme problems." Theoretically, the EQ-5D-5L generates 3,125 different health states. Preference-based utilities for each of these health states are available for Germany with "full health" and "death" being anchored at 1 and 0, respectively (60).

The Assessment of Quality of Life (AQoL) instruments are health-related multi-attribute utility quality of life instruments. The AQoL is a reliable and valid instrument (53). The AQoL-8D consists of five psycho-social and three physical dimensions. With one exception (dimension "senses"), each of these represents a psychometrically valid sub-scale [e.g., tests indicate they measure a common construct (61)]. Utility scores were obtained by a four-stage methodology (62).

The KIDSCREEN-10 index consists of 10 items each answered on a 5-point Likert scale. The index provides a good discriminatory power along the health-related quality of life trait-continuum. The KIDSCREEN-10 Index shows good psychometric properties (63). Utility scores will be derived by an algorithm for mapping the KIDSCREEN-10 index onto the CHU9D utility scores, a preference-based instrument developed specifically for application in cost-utility analyses (54).

Sample Size

The sample size calculation is based on the primary outcome measure (change in TRF score between T2 and T3) which is hierarchically ranked on top of the multiple testing procedure. Based on the results described above (14, 15, 64), it is assumed that the standardized treatment effect for this outcome expressed by Cohen's d amounts to $d = 0.25$. With a two-sided significance level of $\alpha = 0.05$ and a power of $1–\beta = 0.8$ using a two-sample *t*-test, 253 patients per group ($n = 506$ in total) are required. Since the primary outcome is measured on the child level and it is possible to enroll multiple children per patient, a hierarchical multi-level model with patients at level 1 and children at level 2 will be fitted. Taking a drop-out rate of 20 % into account, $n = 634$ patients need to be enrolled into the trial in the analysis. This attrition rate is conservative and based on results from the longitudinal STAR*D study that also carried out long-term follow-up assessments (13), as well as on other studies in the field that report even lower attrition rates (65, 66). The problem of attrition and missing values will also be addressed in the analysis by applying the intention-to-treat principle. For missing values, imputation techniques will be applied, thus partly resolving this problem. Since it is assumed that parts of the outcome variance can be explained by the inclusion of covariates, the actual power of the analysis by a linear multi-level model is expected to be higher than $1–\beta = 0.8$. Assuming that the number of children enrolled per patient amounts to 1.5, we expect that $n = 950$ children will participate in the trial. The enrollment of more than one child per patient is expected to yield an additionally increased power. Sample size calculation was performed using SAS v9.4 (SAS Institute, Cary, NC).

Randomization

Patients will be assigned in a 1:1 ratio to either the control intervention (CBT) or the experimental intervention (CBT+Triple P) through a centralized web-based tool (www.randomizer.at). Randomization will be performed stratified by center, comorbidity (yes/no), and total number of children (1/more than 1). To achieve equal group sizes per stratum block randomization will be performed. The block length will be defined by the study biometrician and treated confidentially to prevent selection bias.

Blinding

Assessment interviews will be conducted and analyzed by clinician-raters blinded to the treatment condition. Raters at post and follow-up assessments must not be the therapist of the particular patient being assessed and analyzed.

Statistical Methods

Our primary efficacy analysis will be based on the full analysis set (FAS) according to the intention-to-treat (ITT) principle, reflecting the recommendations given in relevant guidelines (67). The FAS is defined to include all patients enrolled who are assigned to the treatment group they were originally randomized to, regardless of whether they actually underwent the assigned treatment or not. This will be the primary population for evaluating all efficacy endpoints and subject characteristics. Additionally, the per-protocol (PP) population, including all FAS patients with no major protocol deviations, will serve as a secondary analysis population and be used for sensitivity analyses. Before lock of the database, each patient's allocation to the FAS or PP population will be defined in the statistical analysis plan.

The two hypotheses to be assessed in the primary efficacy analysis are ordered hierarchically: In the first step, the null hypothesis $\mathrm{H}_0^{\mathrm{I}}\colon \mu_{\mathrm{CBT+PPP}}^{\mathrm{T3-T2}} = \mu_{\mathrm{CBT}}^{\mathrm{T3-T2}}$ for the primary outcome "change in teacher CBCL (Teacher Report Form/TRF) score between T2 and T3" is tested at the two-sided significance level of 5% against the alternative $\mathrm{H}_1^{\mathrm{I}}\colon \mu_{\mathrm{CBT+PPP}}^{\mathrm{T3-T2}} \neq \mu_{\mathrm{CBT}}^{\mathrm{T3-T2}}$. If $\mathrm{H}_0^{\mathrm{I}}$ can be rejected, the null hypothesis $\mathrm{H}_0^{\mathrm{II}}\colon \mu_{\mathrm{CBT+PPP}}^{\mathrm{T4-T2}} = \mu_{\mathrm{CBT}}^{\mathrm{T4-T2}}$ for the second primary endpoint "change in TRF score between T2 and T4" is tested at the two-sided level of 5% against its alternative $\mathrm{H}_1^{\mathrm{II}}\colon \mu_{\mathrm{CBT+PPP}}^{\mathrm{T4-T2}} \neq \mu_{\mathrm{CBT}}^{\mathrm{T4-T2}}$. Application of this multiple test procedure for a priori ordered hypotheses ensures control of the family-wise type I error rate at a level of 5%. The null hypotheses will be assessed using a linear mixed multi-level model with patients at level 1 and children at level 2, adjusting for center, number of comorbidities, number of children, baseline TRF score at T2, and length of waiting period between T1 and T2 (in weeks).

Data missing for the primary outcome variable will be replaced by using multiple imputation (68) which takes the covariates treatment group, center, number of comorbidities, number of children, baseline TRF score at T2, and length of waiting period between T1 and T2 (in weeks) into account by applying the fully conditional specification method (69). This will be realized using the option "FCS" of the SAS "MI" procedure implemented in SAS 9.4. Sensitivity analyses will be performed by applying alternative methods dealing with missing data such as complete case analysis. All secondary outcomes will be evaluated descriptively, and descriptive *p*-values are reported together with 95% confidence intervals for the corresponding effects. Further exploratory analyses will be performed to identify potential prognostic factors (e.g., parental disorder, child psychopathology, socio-economic status) and mediators (e.g., Brief Symptom Inventory/BSI, Parenting Questionnaire/EFB, Parental Stress Inventory/ESF) for an intervention effect. The safety analysis includes calculation of frequencies and rates of adverse and serious adverse events together with 95% confidence intervals. All analyses will be performed with SAS version 9.4 or higher.

The health-economic evaluation will involve a combination of a cost-effectiveness analysis (CEA) and a cost-utility analysis (CUA) (70, 71). The economic evaluation will be done from a societal perspective (all relevant costs) and a public health care perspective (only direct medical costs) within a 6 month time frame. In the CEA, the incremental cost-effectiveness ratio (ICER) will be expressed as the incremental costs per point improvement on the primary clinical outcome (CBCL scores in children; parents' BSI scores). In the CUA, the ICER will be expressed as incremental costs per quality-adjusted life year (QALY) gained as based on the EQ-5D-5L (52, 72) (parents) and KIDSCREEN-10 (63, 73) (children). Sampling uncertainty in the ICER will be handled using non-parametric bootstrapping by resampling patient-level data to generate 2,500 simulations of the ICER. We will bootstrap the SURE model (seemingly unrelated regression equations; sureg command in Stata) to allow for correlated residuals of the cost and effect equations (74). Ninety five percent confidence intervals (CI) will be obtained by the bootstrap acceptability method, since parametric techniques are inappropriate for use on skewed variables and ratios (75).The bootstrapped ICERs will be plotted in a cost-effectiveness plan where the horizontal axis reflects differences in effects and the vertical axis differences in costs. The bootstrapped ICERs will also be shown in a cost-effective acceptability curve disclosing the probability that the intervention is cost-effective for a range of willingness-to-pay ceilings (76). To test the robustness of the base-case findings, a probalistic sensitivity analysis will be done. Several assumptions made in the base-case scenario will be changed to assess their impact on the ICER (e.g., QALY calculation based on AQoL-8D).

DATA MANAGEMENT, MONITORING, AND QUALITY ASSURANCE

Training of Study Personal and Treatment Fidelity

All study personal (assessors, therapists, and supervisors) will be trained before taking part in the study. As all sites are part of associated university training institutes that conform to the highest psychotherapeutic standards. All therapists will receive intense training in CBT for the different disorders. The Triple P institute in Münster, which is the official and certified Triple P institution in Germany, will carry out Triple P training. Trained and certified supervisors will supervise every fourth treatment session at the study sites. At each participating center, a clinical project manager is responsible that the study is conducted in accordance to the procedures outlined in the study protocol. Concerning all information and data collected during the study, all study personal maintain professional secrecy and confidentiality.

After each treatment session, the therapists indicate CBT adherence by rating a checklist extracted from the disorder specific therapy manuals as outlined in the proceedings in psychotherapy (33). Upon termination of a study therapy, a supervisor rates the overall adherence of the therapy based on the adherence checks completed after every session. The adherence rate is documented in the eCRF for further analyzes.

Treatment fidelity/integrity will be analyzed with rating schemes for 5% randomly selected videotaped treatment sessions by the study coordination at the UMR. Before the videotapes are rated; they will be checked for any cues/hints potentially indicating the treatment condition; those will be erased.

Data Collection and Retention

For each included family, a study file will be created at the study site in which all local study documents will be archived. An electronic case report form (eCRF) will be used for the data collection using the secure web based electronic data capture (EDC) tool REDCap (77). The participants complete most of the questionnaires directly in the eCRF. A study staff at each site creates log-in-codes that are handed out to the participants during on site sessions with which the participants (parents, children over the age of 8 years and caregivers/teachers) can log into REDCap to complete the questionnaires at home. The families are asked to fill out the forms independently. The eCRF is programmed to not allow skipping answers to prevent missing data. If questions occur, filling out the eCRF can be paused and continued later on with a trained assessor at a study site. A study staff will enter locally collected data into REDCap preferably on the day of data collection. Upon termination of the study, all local study documents at the study sites will be sent to the UMR where the documents will be digitalized. For supervision and to ensure treatment fidelity, all study sessions will be videotaped. The video data will be stored in encrypted form using VeraCrypt which is useable free of cost under the Apache License 2.0. All videotapes will be send to the study coordination at the UMR and stored there. Upon termination of the study, all video data will be destroyed.

All local study documents are part of the Investigator Site File (ISF) as outlined in section 8 of the ICH Consolidated Guideline which is stored and archived according to the legal retention period. Data quality assessment of the eCRF will be done continuously by the Institute of Medical Biometry and Informatics Heidelberg during the study. Any entry and correction in the EDC system will be documented automatically in an audit file. Completeness, validity, and plausibility of data will be checked in time of data entry (edit-checks) and using validating programs which will generate queries. All assessments and modifications are immediately accessible via web access. The clinical project manager at each study site is also responsible for online transmission of data and site-specific quality assurance. Upon completion of the study, all data will be exported into different data formats for further analyses. The primary data will be made accessible to the public for re- and meta-analyses through PsychData at the Leibniz Institute for Psychology Information (https://www.psychdata.de/). All data will be used in accordance to data protection regulations and the data safety guidelines of the German Psychological Society (DGPs).

Ethical and Legal Aspects

Working with a highly vulnerable group, the procedures set out in this study protocol are designed to ensure that the investigators abide by the principles of the Good Clinical Practice (GCP) guidelines of the International Conference on Harmonization (ICH) and the Declaration of Helsinki and its later supplements. The study will be carried out adhering to local legal requirements. The lead ethics committee at the department of psychology at the UMR has approved the study procedure, study information, and informed consent forms in December 2017 and its subsequent amendments in May and July 2018. A positive ethics committee vote is required at a study site (ethics committees at the departments of psychology at Bielefeld University, Ruhr-University Bochum, Justus Liebig University Giessen, University of Koblenz-Landau, University of Leipzig, Johannes Gutenberg University Mainz, and Ludwig-Maximilians-University Munich), before the inclusion of a first patient at the respective site. At the beginning of the study, patients and their families are informed verbally and written about the aims of the study and the study procedures. All patients and their families (children over 6 years of age) need to provide written informed consent for study participation. For children to participate in the study, written informed consent by all legal guardians is required. Every participant can drop out of the study at any time. All dropouts will be documented in the eCRF.

Pseudonymization

After signing the informed consent, each study participant is assigned a pseudonymized screening-ID. The screening-ID consists of three parts: specific number for the study site, consecutive screening number and individual coding number (1 = patient, 2 = partner, 3, 4, 5 = included children in order of increasing age, X = ex-partner). Members of a family can be recognized by the first two parts of the screening-ID (specific study site number and consecutive screening number).

There is a coding list on paper at each study site on which names and screening-IDs are documented. The coding lists, which are kept locked away, are accessible only to study personal. The lists will be destroyed upon termination of the study but no later than 2021/12/31. After that, all data will be fully anonymized. As long as the coding lists exist, participants can request the deletion, respectively the destruction of all their collected data.

Clinical Monitor

A clinical monitor is responsible for overviewing the implementation of the study in accordance to the ICH-GSP guidelines at the study sites. The clinical monitoring includes pre-study, initiation, intermediate and close-out visits to the study sites. During the site visits, the clinical monitor examines the protocol adherent study implementation, the safety of the patients and the data consistency (e.g., comparison of local study documents and eCRF records).

Data Safety Monitoring Board

An independent Data Safety Monitoring Board (DSMB), consisting of a university professor, which is an expert in the field of the study, and a biometrician, is responsible for monitoring the study as well as assessing the study protocol adherence and the study progress (especially the recruiting plan). The DSMB will be informed about all safety aspects of the study (especially serious adverse events, SAE) and will review them regularly. If necessary, the DSMB will recommend changes to the study protocol or the termination of the study.

DISSEMINATION

The results of this study will be made available to the larger scientific community via peer-reviewed publications in open access scientific journals. Politicians, public health services and stakeholders will be informed through the COMPARE website, conferences, teaching seminars, flyers, newsletters, and personal contacts throughout the study and beyond, thus improving public policy and health care decisions concerning preventive interventions and treatments for COPMI. As all study sites are university based outpatient clinics the implementation of results in ongoing and future practice is ensured.

Upon termination of the study, all primary data will be made available to the scientific community in a completely anonymized manner for re- and meta-analyses using PsychData, a data-sharing platform developed by the Leibniz Institute for Psychology Information (ZPID, https://www.psychdata.de).

DETAILED STUDY PROCEDURE

A detailed flowchart of the study procedure is presented in **Figure 3**.

Pre-assessment T1 and Assessment of Eligibility

After an initial screening of eligibility for an as early and effective as possible selection of patients and their families and a regular intake consultation with the patient in the outpatient clinic, the patients and their families (children over the age of 6 years) are invited to an informational session at the study center. During this session, the families are informed about the study procedure before signing their consent to participate in the study. For pseudonymized data collection, each participant is assigned a Screening-ID. To assess a broad range of DSM-5 based psychological disorders and comorbid ones as well as severity ratings, structured clinical interviews for patients (DIPS), and children (Kinder-DIPS parent version or SIVA) are conducted with the patient respectively a parent (for each child separately). The parent, with whom the Kinder-DIPS/SIVA is conducted, has to be the same for all children of a family at all measurement points. The in- and exclusion criteria are checked on the basis of the information collected during the screening and the diagnoses

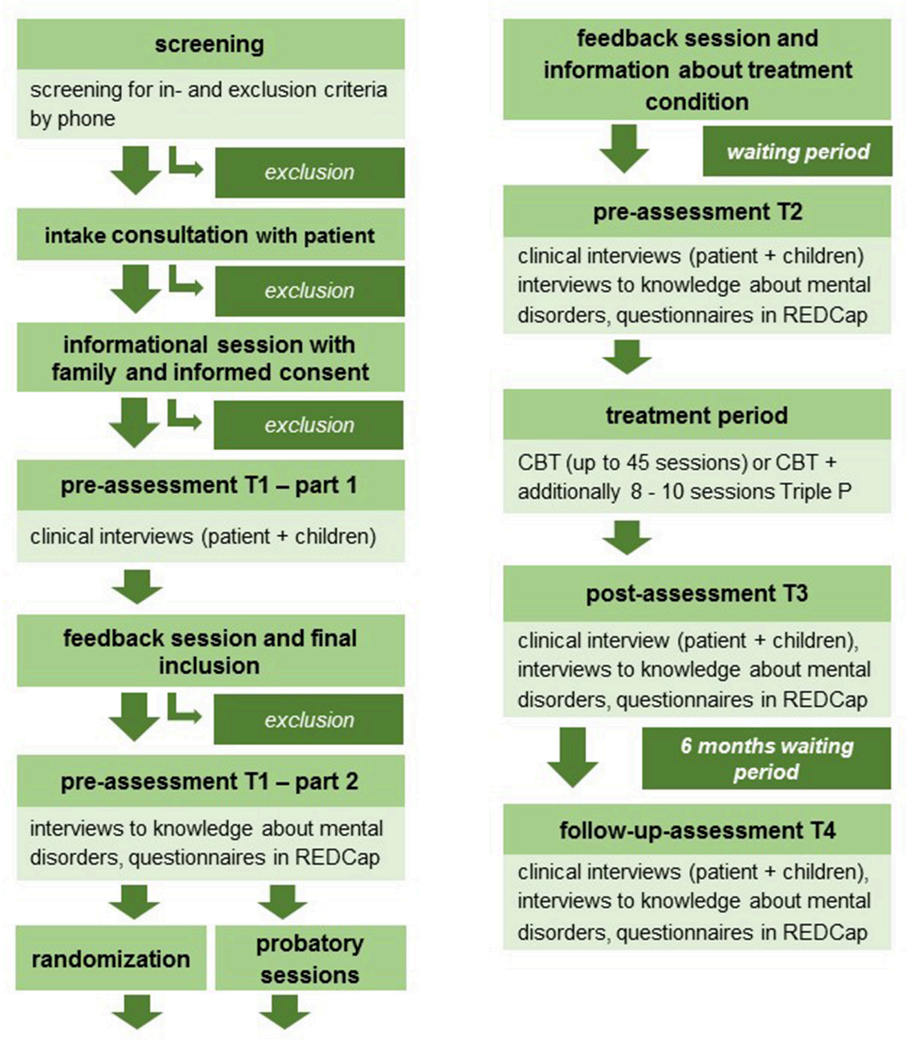

FIGURE 3 | Study flowchart.

based on the clinical interviews. Feedback to diagnoses is given in a separate session. If a patient and its family fulfill all the in- and none of the exclusion criteria, the family is included in the trial and records for the eCRF are created for all the included family members. The family is invited to another session in which interviews about knowledge about mental disorders are conducted with each included family member. After this session codes are handed out with which the family members can log into REDCap at home to complete the questionnaires. Forms with REDCap-codes for the children's caregivers or teachers (one caregiver/teacher for each included child) are also handed out to the family with the request to forward them to the respective caregiver/teachers. There is no reference to the clinical trial of the COMPARE-family project on the caregiver/teacher's form; rather, the questionnaire is generally presented as part of a study at the department of psychology the UMR. After the final inclusion in the study, the randomization to one of the two treatment arms (CBT or CBT+Triple P) takes place. If needed, further probatory sessions can take place at the study centers. In a separate feedback session, the patient is informed about the treatment condition and results of the diagnostic process.

Pre-assessment T2

After the usual waiting period for a psychotherapy treatment in the study centers (waiting period can differ between the study centers depending on their usual waiting periods for a psychotherapy treatment), structured clinical interviews for patients and children are conducted again with the patient respectively the same parent as in T1 (for each child separately). The family is invited to another session in which interviews about knowledge of mental disorders are conducted with each included family member, and codes for logging into REDCap at home to complete the questionnaires are handed out. The forms with REDCap-codes for the children's caregivers/teachers are also handed out to the family with the request to forward them to the respective caregivers/teachers.

Treatment Period

All patients receive between 25 to 45 sessions of individual CBT. Patients allocated to the experimental intervention (CBT+Triple P) receive in addition 8 to 10 group or individual Triple P sessions parallel to the last third of the CBT sessions. After every therapy session, patients, and therapists complete a session-feedback screener including aspects of therapeutic alliance and symptom intensity, as well as on fidelity. (Serious) adverse events [(S)AE] are also assessed after every therapy session with a checklist. If a (S)AE occurs, a report has to be filed and send to the study coordination at the UMR. Every fifth session, the patients are also handed out a code for logging into REDCap at home to complete the BSI to assess remission status (early, late, and non-remitters).

Post-assessment T3

Using the same measurements as in the pre-assessments, the post-assessment is conducted by an assessor who is blind to the treatment condition. Structured clinical interviews for patients and children are conducted again with the patient, respectively the same parent as in T1 (for each child separately). During a separate session, interviews on knowledge about mental disorders are conducted with each included family member separately, (S)AE are assessed for every included family member, and codes for logging into REDCap at home to complete the questionnaires are handed out at the end. The forms with REDCap-codes for the children's caregivers/teachers are also handed out to the family with the request to forward them to the respective caregivers/teachers. If a (S)AE occurs, a report has to be filed and sent to the study coordination at the UMR.

Follow-Up-Assessment T4

After a waiting period of 6 months, the follow-up-assessment is conducted by a blind assessor in the same manner as the post-assessment. The structured clinical interviews for patients and children are conducted again with the patient respectively the same parent as in T1 (for each child separately). In a separate session, knowledge about mental disorders and (S)AE are assessed for each included family member separately. At the end of this session, codes for logging into REDCap at home to complete the questionnaires are handed out. The forms with the code for the children's caregivers/teachers are also handed out to the family with the request to forward them to the respective caregivers/teachers. If a (S)AE occurs, a report has to be filed and sent to the study coordination at the UMR.

DISCUSSION

We expect that in arm I (CBT), the parents will improve through treatment as will their children (2, 7, 12). Since, parenting skills of parents with a mental illness are likely impaired and enhancing such skills leads to better child outcomes (17, 18, 20, 27, 28), we expect that additional incremental effects will become apparent in arm II (CBT+Triple P). We will use the Positive Parenting Program (Triple P) in addition to CBT, as Triple P is an evidence-based, widely used, and well-established program. The positive effects of Triple P have been demonstrated in parents and children (22, 23, 78), although incremental effects above and beyond parental CBT have not been researched in conjunction with COPMI so far.

The established magnitude of this specific incremental effect will result in precise recommendations for this high-risk group that will impact clinical practice (i.e., practice parameters, treatment guidelines). As outlined above, COPMI are most likely to constitute the next generation of patients with a mental illness, and we assume that this intervention will contribute to the prevention of SMI in this specific high-risk group.

As studies so far have focused primarily on specific disorders (e.g., anxiety, depression) and have excluded comorbidities (1, 3), though comorbidities actually occur with the most patients, the COMPARE study will fill this research gap by including a broad range of mental disorders and by not excluding comorbidities. A double baseline measurement with multiple baselines depending on the usual waiting periods in the study centers (T1: 1st assessment of parents and children via clinical interviews and questionnaires; T2: 2nd assessment with the same instruments) will enable us to assess parental disorder effects on the child that can then be related to parental treatment effects (T3: after completion of 25 short-term to 45 long-term sessions of state-of-the-art CBT or CBT+TripleP) for a 6-month follow-up (T4). As there are differential parental psychotherapy effects on children depending on the parental remission status (early, late, and non-remitters), we will also assess such effects (9, 13). By including different therapy durations (short- vs. long-term therapy) we will be able to shed light on treatment duration effects.

Research and Clinical Implications

The RCT is part of the COMPARE consortium [see Christiansen et al. (79) in this research topic] with the subprojects COMPARE-emotion, COMPARE-interaction, COMPARE-work, and COMPARE-school. The findings of the Compare consortium will not only allow for the estimation of parental treatment and parenting skill effects on their children, but also for the novel approach of testing the core assumptions of the transgenerational transmission of mental disorders model (5) comprehensively. The cost-effectiveness and cost-utility analyses from a societal and a health care system perspective will be a basis for negotiations with health care providers, as COPMI at risk can currently not receive professional help, except if they have already developed disorders themselves. The evidence of this trial will hopefully contribute to an understanding of preventing the development of disorders in COMPI to reach this goal. The identification of specific risk profiles will contribute to such an improved understanding and will result in tailored interventions that might either be more preventive or interventive in character, depending on the individual risk.

Limitations

COMPARE-family plans four extensive assessments for all family members as well as up to 55 therapy assessments. Even though the majority of patients has positive attitudes toward extensive diagnostic assessments (80), this might also be a burden on the family, especially as the add-on projects require additional assessments at the same assessment times.

The implementation of a self-wait control design in the RCT was originally planned with a waiting period of 6 months between the first assessment of the patients and their families and the beginning of the therapy. Due to the new psychotherapy guidelines in Germany, that became effective in April 2017, the waiting periods to receive psychotherapy treatment have decreased enormously at the study sites. Although the time span to receive a place in a treatment program can still be up to 9 months, many of the recruiting outpatient clinics have tried to cut those time spans down (e.g., 1–3 months waiting periods). With the original 6 months waiting period, patients wanting to participate in the COMPARE study would consequently have had to wait longer for their psychotherapy treatment than necessary. For the study centers this not only created ethical but also practical issues with interested patients preferring treatments with shorter waiting periods to the participation in COMPARE. With the now implemented multiple baseline design, the waiting period can vary between the study centers depending on the usual waiting periods on-site, therefore not disadvantaging study participants.

The observation period of only 39 months in total is rather short for capturing effects on the prevention of SMI, but due to funding, a longer observation period could not be implemented. For assessing long-term effects, a follow-up study is already planned and all participants are asked permission to be contacted again later on.

The majority of outpatient clinics will most likely treat patients with depressive and affective disorders, a fact possibly hampering the aim of the COMPARE study to cover a broad range of disorders. However, as all outpatient clinics are university-based and all recruiting centers have different research foci, chances are good that the range of disorders will be represented. This will also raise the generalizability of findings.

AUTHOR CONTRIBUTIONS

HC, MS, and KG contributed to conception and design of the study. CB drafted the health economic evaluation study. MK, CK, and JK organized the database and performed the statistical analyses. MS wrote the first draft of the manuscript. All authors contributed to sections of the manuscript, manuscript revision, read, and approved the submitted version.

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CONSORT CRITERIA

CONSORT Criteria were applied in writing this study protocol.

RECRUITING CENTERS

Members of the recruiting centers are:

Bielefeld: Angelika Schlarb

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Gießen: Arleta Luczejko, Christina Schwenck, Rudolf Stark, Anett Werner, Julia Zimmermann

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Psychoeducational Intervention for Perinatal Depression: Study Protocol of a Randomized Controlled Trial

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Perinatal depression (PD) is a severe and disabling condition impacting negatively on children in terms of adverse neonatal outcomes and on the well-being of women and their families. All pregnant women attending the unit of Gynecology and Obstetrics Service of the University of Campania "L. Vanvitelli" will be screened for PD using the Edinburgh Postpartum Depression Scale (EPDS). Women with a score $\geq$10 at the EPDS will be invited to receive a full psychiatric assessment. The required sample size is of 126 women with PD which will be randomly allocated to either an experimental group, receiving a uni-familiar psychoeducational intervention, or to a control group, receiving the Best Treatment Option (BTO). Patients will be evaluated through several assessment instruments: Hamilton Depression Rating Scale (HAM-D), Hamilton Anxiety Rating Scale (HAM-A), Global Assessment of Functioning (GAF), Clinical Global Impression (CGI), Manchester Short Assessment of Quality of Life (MANSA), Family Assessment Device (FAD), Family Coping Questionnaire (FCQ), and Pattern of Care Schedule (PCS). Patients will be evaluated at baseline, 3, 6, 9, and 12 months post-randomization. The severity of depressive symptoms at the HAM-D scale has been selected as primary outcome. Other outcome measures include improvement in the severity of anxiety symptoms, of global and personal functioning, an improvement in family members' coping strategies and in the level of quality of life. It has been highlighted the importance of developing screening and treating programs for PD, and our study will use rigorous study design to evaluate the efficacy of the adaption of a well-known family psychoeducational model to the treatment of PD. The aims of present trial are to: (1) develop an informative package for pregnant women with PD; (2) promote a screening programme for PD; (3) identify those (socio-demographic and pregnancy-related environmental) factors associated with a higher risk to develop a perinatal or postnatal depression; (4) evaluate the efficacy of a new experimental psychoeducational intervention in reducing the depressive symptoms during pregnancy compared to the BTO.

Keywords: perinatal depression, women mental health, severe mental disorders, family burden, coping strategies

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INTRODUCTION

Depressive disorders represent the major cause of disability worldwide (1, 2). These disorders are prevalent in the perinatal period and about 12% of women are affected (3). However, prevalence rates of perinatal depression (PD) should be cautiously considered since this disorder is frequently underdiagnosed, mainly because patients experience delays and difficulties in help-seeking due to feelings of guilty and fear of stigmatization (4, 5). PD poses a significant burden on affected women, their families and on society at large; moreover, it carries serious long-term consequences on the mental health of the new born (6, 7). In particular, PD can have several detrimental effects including low birth weight, preterm birth, small development for gestational age, early childhood developmental delays, poor maternal fetal attachment, impairments in cognitive functioning, behavioral disturbances, and development of depressive disorders in the childhood or adolescence (8–16).

Different risk factors have been identified for PD, including low socioeconomic status (17–19), being a single mother (17, 20), poor social support (21), general life stress (17), and unplanned pregnancies (22). Indeed, PD is associated with a dysregulation and hyperactivity of the hypothalamic-pituitary-adrenal axis activity (HPA) (23), with an increased exposition to the corticotrophin-releasing hormone (CRH) during intrauterine development.

Several interventions have been developed for reducing the impact of risk factors and for preventing the development of PD (24), including professionally-based home visits, postpartum peer-based telephone support, interpersonal psychotherapy (25), and cognitive behavioral therapy (26). Although several of these interventions have been made available in clinical practice, research is needed in order to confirm their efficacy (27).

Symptoms of perinatal depression often include anxiety, irritability, sleep disturbance, low mood, and excessive concern for the child's care (28), in fact, PD is a multi-faceted and complex condition which can have heterogeneous clinical presentations (15). In particular, a significant number of women with PD experience comorbid anxiety (29, 30), obsessive-compulsive symptoms and post-traumatic stress disorders (31).

One of the main debated issues regarding the optimal prevention and treatment of perinatal depression is related to the opportunity to early detect the disorder through screening procedures (32, 33). In particular, it has been argued that screening programmes can be effective in reducing burden and disability associated with the disorder (34), although there is the risk to create false-positives (35). Screening procedures aim to detect people at high risk to develop a full-blown disorder, and therefore the clinical utility of the screening process is greatly influenced by the ability to accurately identify those patients (33–35).

However, screening programmes are considered acceptable and even desirable by most pregnant women, both depressed and non-depressed (36). Therefore, available guidelines (37, 38) recommend that healthcare professionals (including midwives, obstetricians, health visitors, and general practitioners) ask questions about past or present mental illness, family history of perinatal mental illness and previous psychiatric treatments including inpatient care. Moreover, the American Academy of Pediatrics also recommend that pediatricians screen new mothers for depression during their visits in the 6 months following childbirth (39).

According to a recent review (40), treatment of PD depends on symptom severity and functional impairment (41, 42). Cognitive behavioral therapy (43), interpersonal psychotherapy, and psychoeducation (44, 45) are usually adopted in case of mild to moderate forms of PD; while antidepressants, more often selective serotonin reuptake inhibitors (SSRI), are used for severe cases (46–51).

When available, pregnant women with PD prefer psychotherapy (52) and the effectiveness of psychological and psychosocial treatments for PD has been explored (53).

Several initiatives have been proposed worldwide in order to assess the efficacy and feasibility of psychoeducational interventions for women with PD. A recent meta-analysis by Sockel (26) found only one study (54) evaluating the efficacy of a psychoeducational programme for women with PD in improving depressive symptoms. More recently, other programmes have been proposed also in low-middle income countries (55).

Psychoeducational interventions are effective in reducing affective symptoms and the levels of stress, with low costs for the mental health department (56).

Based on these premises, the Department of Psychiatry of the University of Campania "Luigi Vanvitelli" is carrying out a study to develop and test the efficacy of a psychoeducational family intervention in pregnant women with perinatal depression and their close relatives.

To our knowledge no randomized controlled trial has been carried out so far in Italy on the effectiveness of psychoeducational interventions for women affected by perinatal depression, although psychoeducational intervention has demonstrated its efficacy in a wide range of severe mental disorders, such as bipolar disorder (57), schizophrenia (58), major depression (59), obsessive compulsive disorder (60), and eating disorders (61).

AIMS

The present trial aims to evaluate the efficacy and feasibility of a new psychoeducational family intervention compared to the Best Treatment Option (BTO) in a sample of women affected by perinatal depression and their family members. The Best Treatment Option (BTO) is provided according to the NICE guidelines (62). In particular, mild to moderate forms of depression may be addressed with self-help or psychological counseling. For moderate or severe forms of PD, high-intensity psychological intervention, or pharmacological treatments are recommended.

The secondary aims are to: (1) improve mental health literacy on the topic of perinatal depression, with a specific focus on risk and protective factors as well as on available therapeutic strategies for the management of perinatal depression; (2)

identify clinical, socio-cultural, and pregnancy-related predictive factors for the development of perinatal depression; (3) improve coping strategies and family functioning of family members participating to the intervention; (4) evaluate the long-term effects of the intervention.

METHODS

Design

This is a randomized controlled trial with two parallel arms for evaluating the efficacy and effectiveness of psychoeducational family intervention in improving depressive symptoms in patients with perinatal depression. The new psychoeducational intervention has been developed by the research staff, based on the Falloon model of psychoeducation for patients with schizophrenia and their families (63).

PATIENTS' RECRUITMENT PROCEDURE

Recruitment for the RCT will be conducted adopting a multistep strategy, as reported in **Figure 1**:

- Step 1. All pregnant women attending the outpatient unit of Gynecology and Obstetrics of the University of Campania "Luigi Vanvitelli" will be invited to participate in the study through the use of informative leaflets on perinatal depression (i.e., characteristics of the disorder, risks, long-term consequences, available treatments). Those interested in having more information will be referred to a research team member presenting a brief overview of study procedures and time commitment in order to obtain their informed consent. Once informed consent is obtained, patients will be officially enrolled in the study and will undergo the screening procedure, compiling the Edinburgh Postnatal Depression Scale (EPDS) (64).
- Step 2. Women with a score $\geq$10 at the EPDS will be invited to receive a full psychiatric assessment.
- Step 3. Patients will receive a full mental state examination by an expert clinician working at the Department of Psychiatry of the University of Campania "Luigi Vanvitelli," in order to evaluate the presence of any mental disorder.
- Step 4. Once a diagnosis of perinatal depression is made, patient's code will be given to the statistician for randomization in one of the two groups. Patients will be allocated through a randomized procedure (with a 1:1 ratio) to the experimental or to the control group.

INCLUSION CRITERIA

Starting from January 2019, patients attending the outpatient unit of Gynecology and Obstetrics of the University of Campania "Luigi Vanvitelli" will be invited to participate to the study. The eligibility criteria will be: (1) gestation age $>$ 3 months or within 3 days of childbirth; (2) age $\geq$ 18 years; (3) absence of any disabling physical condition; (4) living with a relative for at least 9 months in the last year and continuously in the last 3 months.

EXCLUSION CRITERIA

Exclusion criteria will be: (1) intellectual disability; (2) diagnosis of schizophrenia, schizoaffective disorder, delusional disorder, or other not specified psychosis-spectrum disorder; (3) having experienced depressive symptoms before pregnancy.

INTERVENTION

Theoretical Background of the Experimental Intervention

Experimental intervention includes techniques derived from classic psychoeducation (63), motivational intervention (65, 66) and cognitive behavioral therapy (67). The intervention has been developed following the guidelines on the management of perinatal depression released by NICE (62), the American College of Obstetricians and Gynecologists (68) and the United States' Preventive Service Task Force (69). The adopted methodology included the following phases: (1) analysis of the scientific literature; (2) evaluation of available handbooks and manuals of other psychosocial interventions targeting perinatal depression; (3) focus groups with expert researchers, clinicians, users and carers, in order to identify the most relevant information to be included in the intervention; (4) development of an *ad-hoc* manual with a detailed description of each session of the intervention, booklets, and other written materials are provided to patients and relatives, whenever relevant; (5) evaluation of the feasibility of the "pilot" intervention in a small-size study.

Features of the Experimental Intervention

The intervention consists in a uni-familiar psychoeducational intervention, scheduled every 7–10 days. It includes six modules on:

1) Individual and family assessment. This module is focused on the assessment of patients' personal and social functioning, and on the identification of personal goals. Moreover, the communication skills adopted by family members are evaluated as well as the characteristics of family functioning.
2) Information on the clinical characteristics of the disorder and its treatment. In this module, information on the main clinical and epidemiological features of PD, including incidence, prevalence, long-term outcome, risk for the children, and negative impact on mother's mental health are presented. Moreover, available pharmacological and non-pharmacological treatment options are discussed, with a specific focus on the risk/benefit ratio for each approach and the importance to be compliant with the therapy. Moreover, during these sessions, the patient is invited to actively participate as the "expert," describing her own personal experience. At the end of the session, an informative booklet will be provided to patients and their family members, summarizing the main aspects discussed during the session.
3) Early warning signs. The third session is focused on the early warning signs of perinatal depression (e.g., changes in the number of sleeping hours; presence of irritability; anxiety;

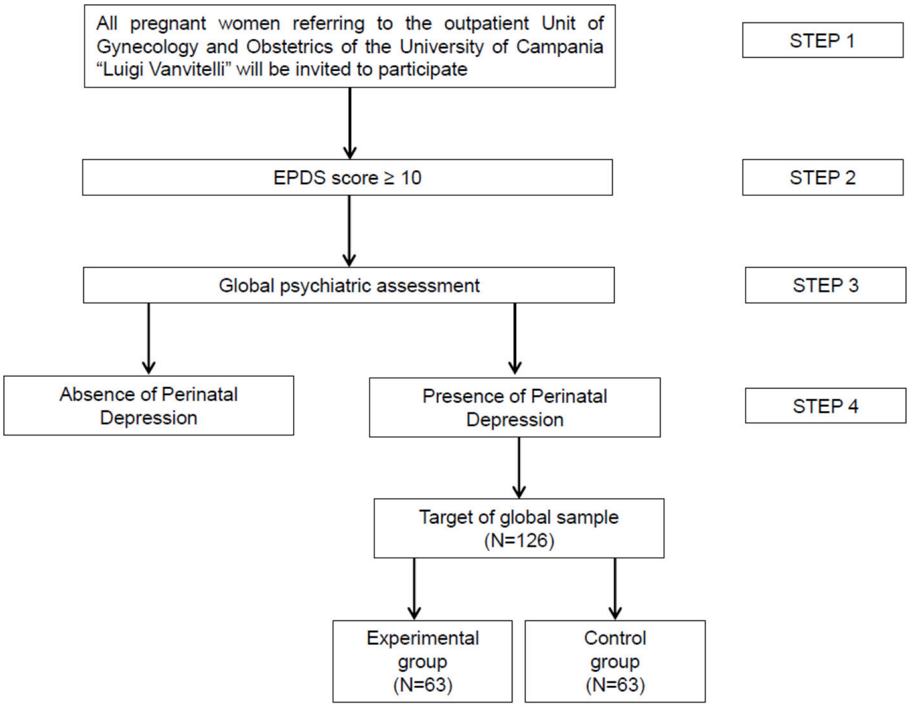


FIGURE 1 | Multi-step recruitment procedure.

etc.) and on the importance to early detect such signs. In particular, patients are requested to identify, with the support of the mental health professional, their own warning signs and to report them on the "Schedule on early warning signs." Strategies for preventing or managing crises are discussed and reported in the same schedule.

4) Management of suicidal behaviors. This module is focused on suicidal risk and on the identification of warning suicidal signs. During this module, an *ad-hoc* schedule is provided to participants and family members in order to define a plan to be used in case of necessity.
5) Communication skills sessions are focused on teaching strategies on how to express pleasant or unpleasant feelings, and on how to improve active listening. This module includes the use of role-plays and case vignettes, based on the personal experience of the patients and their family members.
6) Problem solving skills. The last session is focused on teaching problem-solving techniques. Participants are invited to define a problem in their daily routine and to list all potential strategies to solve it. During the session, participants are guided by mental health professional to develop a plan for solving the problem and discuss possible advantages and disadvantages of each possible solution.

Each session lasts about 90 min. Moreover, two or more booster sessions are planned, if needed. Sessions are developed in order to stimulate discussion and interaction among participants. Site and frequency of sessions can be adapted to families' needs and mental health professionals' duties and workloads. The intervention will be carried out at the local mental health center and provided by trained mental health professionals, the time to complete all modules will be between 42 and 60 days. An *ad-hoc* manual has been developed by the research group in order to ensure treatment fidelity. Leaflets and other written materials will be given to patients and family members, when relevant (**Table 1**).

Control Intervention

Patients allocated in the control intervention will receive the best treatment option (BTO) according to the NICE guidelines (62). At each assessment point, the BTO will be documented through the use of the Pattern of Care Schedule (PCS), an *ad-hoc* schedule filled in by the researcher together with the treating psychiatrist. PCS aims to collect all the information related to the treatments received by the patient. Moreover, patients will continue to be in contact with the treating psychiatrist and whether necessary will receive psychological counseling, both individual, or familiar.

TRAINING OF MENTAL HEALTH PROFESSIONALS

Three mental health professionals (at least one will be a psychiatrist) will receive an *ad-hoc* training course for the provision of the experimental intervention. Supervision meetings will be organized during the study period in order to ensure fidelity to the procedure.

TABLE 1 | Characteristics of the interventions.

PSYCHOEDUCATIONAL FAMILY INTERVENTION

Uni-familiar psychoeducational intervention, scheduled every 7–10 days. It consists of six modules:

- Individual and family assessment
- Information on the clinical and epidemiological characteristics of the disorder
- Early warning signs
- Management of suicidal behaviors
- Communication skills
- Problem solving skills

Each session lasts about 90 min. Moreover, two or more booster sessions will be planned, if needed. Sessions are developed in order to stimulate discussion and interaction among participants. Leaflets and other written materials will be given to patients and family members when relevant.

BEST TREATMENT OPTION (BTO)

Provided according to the NICE guidelines

- Persistent subthreshold depressive symptoms/ mild to moderate depression: self-help and psychological counseling
- History of severe depression who initially presents with mild depression: pharmacological treatment
- Moderate or severe depression: high intensity psychological intervention, or pharmacological treatments, or an integration of both interventions.

ETHICAL ISSUES

This study is being conducted in accordance with globally accepted standards of good clinical practice, in agreement with the Declaration of Helsinki and with national and local regulations. The study investigators ensure that all mental health professionals involved in the study are qualified and informed about the protocol, interventions, and trial-related duties. The study protocol has been submitted to the Ethical Review Board of the University of Campania "Luigi Vanvitelli."

ASSESSMENT TIME AND INSTRUMENTS

Researchers participating to the study are blinded to patient allocation. All patients are assessed at the following time points: baseline (T0); 3 months post-randomization (T1); 6 months post-randomization (T2); 9 months post-randomization (T3); 12 months post-randomization (T4) (**Table 2**).

The following questionnaires and schedules will be used during the study:

1. The Edinburgh Postnatal Depression Scale (EPDS) (64) is a simple and short 10-items self-report screening questionnaire initially developed for use in postnatal women to improve detection of postnatal depression. The EPDS allows health professionals to detect women who might need help and require a referral for a full diagnostic assessment. EPDS has satisfactory sensitivity and specificity values and it is also sensitive to changes in severity of depression over time. For the purpose of the present study, the cut-off threshold has been set at ≥ 10 (66). In validation studies, different cut-off points have been found due to clinical, social, cultural and economic diversities (70). The cut-off score for possible depression (i.e., a positive screening result) is $\geq$13 points but, as suggested by Hewitt et al. (71), a cut-off score ≥ 10 can be used in order to identify patients with both major or minor depression. Moreover, given the observed variability in the performance of the test, the screening procedure could produce both false positives and negatives (72). Therefore, it is essential that this screening tool is complemented by the clinical judgment (73).
2. The Hamilton Depression Rating Scale (HAM-D) (74) is a 17-items questionnaire used to rate the severity of depressive symptoms such as low mood, insomnia, agitation, anxiety and weight loss. The interview and scoring take about 15 min. The score for each item ranges from 0 (not present) to 4 (extreme severity).
3. The Hamilton Anxiety Rating Scale (HAM-A) (75) is a 14-items questionnaire developed to measure the severity of anxiety symptoms, both psychic anxiety (mental agitation and psychological distress) and somatic anxiety (physical complaints related to anxiety). The score for each item ranges from 0 (not present) to 4 (extreme severity).
4. The Global Assessment of Functioning (GAF) (76) is a 100-point rating scale assessing social, occupational, and psychological functioning of adults, with higher scores indicating better level of functioning.
5. The Clinical Global Impression (CGI) (77) scale measures illness severity (CGI-S), global change (CGI-C) and therapeutic response. The CGI-S is rated on a 7-point scale, from 1 (normal) to 7 (the most severely ill patients). The CGI-C scores range from 1 (very much improved) to 7 (very much worse). Treatment response ratings should take into account both the therapeutic efficacy and the treatment-related adverse events, and range from 0 (marked improvement and no side-effects) to 4 (unchanged or worse, and side-effects outweigh the therapeutic effects). Each component of the CGI is rated separately; the instrument does not provide a global score.
6. The Manchester Short Assessment of Quality of Life (MANSA) (78) is a 17-item questionnaire assessing quality of life focusing on satisfaction in twelve aspects of life. For twelve items, the satisfaction is rated on 7-point rating scales, ranging from 1 ("could not be worse") to 7 ("could not be better"). For five items, the responses are binary ("yes" or "no"). The total MANSA score is the mean of the Likert-item scores.
7. The Family Assessment Device (FAD) (79), based on the McMaster Model of Family Functioning, consists of 7 subscales on family involvement in patient's care (affective involvement, affective responsiveness, behavioral control, communication, problem solving, and roles and general family functioning). Scores range from 1 ("healthy functioning") to 4 ("unhealthy functioning").
8. The Family Coping Questionnaire (FCQ) (80) is a self-administered 34-items questionnaire, which has shown a good reliability and external validity. Each item is rated on a 4-level scale, from 1 ("never") to 4 ("always"). The items are grouped into the following 11 subscales: information on patient's illness; positive communication toward the

TABLE 2 | Assessment tools adopted in the study's protocol.

	Screening phase	T0 (baseline)	T1 (month 3)	T2 (month 6)	T3 (month 9)	T4 (month 12)
ASSESSMENT INSTRUMENT						
Edinburgh Postnatal Depression Scale (EPDS)	x					
Hamilton Depression Rating Scale (HAM-D)		x	x	x	x	x
Hamilton Anxiety Rating Scale (HAM-A)		x	x	x	x	x
Global Assessment of Functioning (GAF)		x	x	x	x	x
Clinical Global Impression (CGI)		x	x	x	x	x
Manchester Short Assessment of Quality of Life (MANSA)		x	x	x	x	x
Family Assessment Device (FAD)		x	x	x	x	x
Family Coping Questionnaire (FCQ)		x	x	x	x	x
Socio-demographic schedule		x	x	x	x	x
Pattern of Care Schedule (PCS)		x	x	x	x	x

patient; relatives' maintenance of social interests; patient's involvement in social activities; talking with friends about the patient's condition; coercion; avoidance; resignation; use of alcohol and drugs; collusion; search for spiritual help. The FCQ is widely used for the assessment of coping strategies among relatives of people with severe mental disorders (81, 82).

9. The Pattern of Care Schedule (PCS) (80), a 40-item questionnaire on pharmacological and non-pharmacological treatments as well as on health care access made by the patient. It is compiled by the researcher in collaboration with the patient. If information is inadequate, or if the researcher is not sure about patients' reliability, other sources (e.g., treating physician, relatives, etc.) can be consulted. During the study period, the treating clinician will continue to provide the usual treatment to patient, and–if necessary–to change or adjust the pharmacological regimen. This schedule is used in order to record pharmacological treatments and doses, psychotherapeutic and psychosocial interventions (type of intervention, number of sessions) provided to patients in order to document the BTO.

BASELINE ASSESSMENTS

At baseline the following information will be collected: (1) sociodemographic data (age, nationality, educational level, marital status, employment status, number of family members, duration of the illness, time in charge at the mental health center (months), number of (voluntary and involuntary) hospitalizations, suicide attempts (numbers); (2) clinical features related to pregnancy (gestational period, previous pregnancies, *in vitro* fertilization, clinical conditions of the fetus, and the pregnant); (3) social context, such as relationship with the partner, family conflicts, socio-economic stressors, history of any psychiatric disorder (**Table 3**).

TABLE 3 | Baseline assessments.

Sociodemographic data	Clinical features related to pregnancy	Social context
• Age	• Gestational period	• Relationship with the partner
• Nationality • Educational level • Marital status	• Previous pregnancies • *In vitro* fertilization • Clinical conditions of the fetus and the pregnant	• Family conflicts • Socio-economic stressors • History of any mental disorder
• Employment status • Number of family members • Duration of the illness • Time in charge at the mental health center • Number of hospitalizations • Suicide attempts		

STATISTICAL ANALYSES

Power Analysis

In order to assess the efficacy of the experimental intervention, a power analysis was performed. As reported by Sockol et al. (83), available psychosocial interventions for perinatal depression have been found to have an overall effect size of 0.65. Therefore, the sample size has been defined selecting 0.65 as desired level of effect size, an α error set at 0.05 and power set at 0.95. Therefore, the total sample will consist of 126 patients, allocated with a 1:1 ratio to the experimental or to the control group (**Figure 1**).

Data Analysis

Differences in socio-demographic and clinical characteristics, such as severity of depressive and anxiety symptoms, quality of life, trimester of pregnancy, and personal functioning between the two groups will be evaluated using Chi-square or *T*-test for independent samples, as appropriate. Differences in coping strategies and in family accommodation reported by relatives will be evaluated with the same tests.

In order to assess the efficacy of the experimental intervention, a linear multivariable regression model will be implemented, using as main outcome the score at HAM-D at 3 months post-randomization. Moreover, several confounding variables (such as age, marital status, familiarity for mental disorders, peer support, previous pregnancy, etc.) will be entered in the model. Furthermore, predictors of response at the end of the intervention will be identified.

"Response" is generally defined as a 50% decrease in scores on Hamilton Depression Rating Scale (HAMD) (74). In this protocol, we will consider the score at the 3-month HAM-D compared with the baseline score. Therefore, the continuous variable "3-month HAM-D score" will be transformed in a binary variable (1 = response; 0 = absence of response), and it will be entered in a logistic regression multivariate model in order to identify possible predictors of positive response to the experimental intervention.

In order to evaluate differences in relatives' coping strategies according to patients' clinical features and type of relationship with the patient, a two-level model with fixed independent variables and random intercept, using a maximum likelihood estimation, will be performed. The mean score at the subscales of the FCQ will be entered in the model, adding covariates step by step in order to identify the model with indexes of best fit. The final model will be selected according to the−2 loglikelihood value, and the model with the lowest value will be choose. Estimate, standard error, 95% confidence interval, and *P*-values will be calculated.

The level of significance will be set at $p < 0.05$. All analyses will be performed using the Statistical Package for Social Science software (SPSS), version 18.0 (SPSS).

STEPWISE PROCEDURE

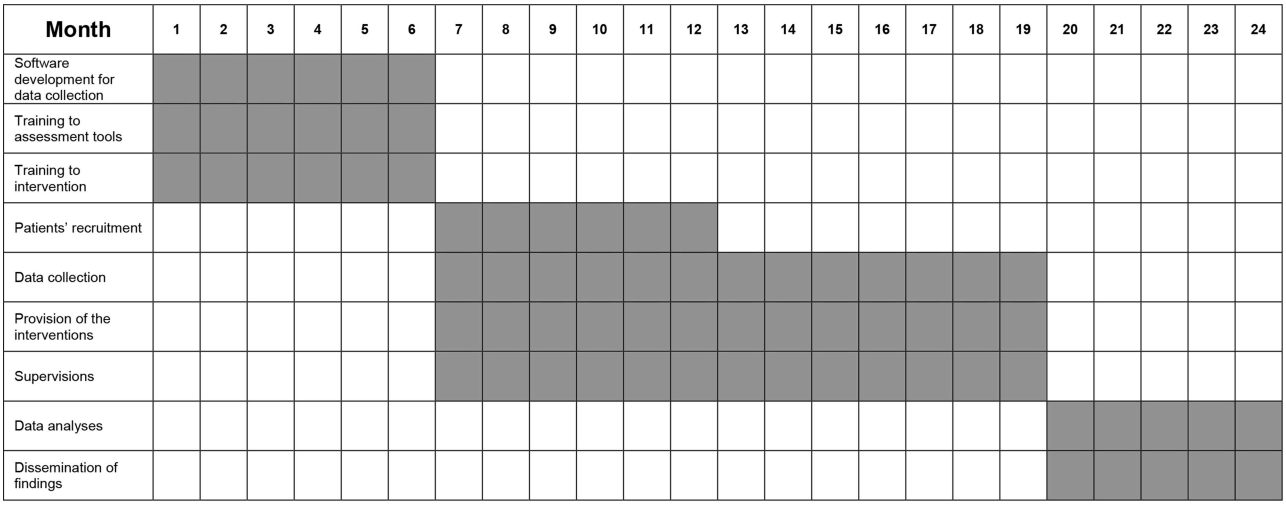
From month 1 to 6, the following phases have been carried out: (1) development of the work plan for data collection; (2) training course for mental health professionals on the interventions; (3) training course for researchers on assessment tools. From month 7 to 12, patients' recruitment will be implemented. From month 7 to 18, interventions will be provided to participating patients. From month 7 to 20, patients' follow-up assessments are being made. From month 20 to 24, statistical analyses will be performed as well as findings will be disseminated through scientific papers, posters and conference participation. A specific plan for dissemination will be developed, including institutional newsletters, press and media release, flyers, training course throughout the national context in order to promote the implementation of the intervention on a large scale. Moreover, users and carers will be involved in the process of dissemination organizing thematic conferences for presenting the characteristics of the intervention.

The stepwise procedure is summarized in **Figure 2**.

ANTICIPATED RESULTS

Primary Outcome

The severity of depressive symptoms, evaluating at the HAM-D, has been selected as primary outcome since it is a reliable index and it has already been used in previous studies on the efficacy of psychosocial interventions (84). In particular, the primary outcome is the reduction of at least 50% of the HAM-D scoring at 3 months post-randomization (T1). The work hypothesis is that BTO plus experimental intervention will be



Month	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24
Software development for data collection	■	■	■	■	■	■																		
Training to assessment tools	■	■	■	■	■	■																		
Training to intervention	■	■	■	■	■	■																		
Patients' recruitment							■	■	■	■	■	■												
Data collection							■	■	■	■	■	■	■	■	■	■	■	■	■					
Provision of the interventions							■	■	■	■	■	■	■	■	■	■	■	■	■					
Supervisions							■	■	■	■	■	■	■	■	■	■	■	■	■					
Data analyses																				■	■	■	■	■
Dissemination of findings																				■	■	■	■	■

FIGURE 2 | Stepwise procedure.

more effective than the BTO alone in reducing the severity of depressive symptomatology.

Secondary Outcome

The secondary outcomes include an improvement in the severity of anxiety symptoms (evaluating at the HAM-A), of global functioning and personal functioning (evaluated at the CGI and at the GAF, respectively), an improvement in family members' coping strategies (i.e., an improvement in "problem-oriented" strategies reduction); in the level of quality of life (evaluated at the MANSA) and of family functioning.

DISCUSSION

Pregnancy is a stressful period for women, which can be further complicated by an early postnatal hospital discharge (85, 86). Risk factors for PD include previous mental disorders, low socioeconomic status, being a single mother, general life stress (12), unwanted and unplanned pregnancies (22), conflicts with the partner, lack of support, limited information, and stigma (87).

Despite many scientific associations and task forces, including the United States' Preventive Services Task Force (USPSTF) (88), the National Institute for Health and Care Excellence (62), the Canadian Task Force on Preventive Health Care (CTFPHC) (89) and the American College of Obstetricians and Gynecologists (68), have highlighted the importance of developing screening and treating programs for PD, there are still many unsolved issues, such as efficacy and the availability of those programs, whose efficacy has been demonstrated by RCTs.

One of the main strengths of our study is represented by the adaption of a well-known family psychoeducational model to the treatment of perinatal depression. In particular, the Falloon psychoeducational intervention was developed for the community management of schizophrenia (63, 90) and has been subsequently adapted to the management of major depression (59, 91) and bipolar disorder (57, 92–94). Perinatal depression may represent an ideal target for this kind of interventions, since it is highly dependent from stressful life events and from family context. Moreover, peripartum is a period of women's life in which pharmacological treatments should be used very cautiously and only in a limited situations. Another strength of our study is the rigorous methodology adopted, which will allow us to understand the impact of the experimental intervention on patient's outcome through a randomized controlled approach. Moreover, the main outcome is the reduction of depressive symptoms at 3-months after randomization. This is an ambitious outcome, since many confounding variables can impact on it. In particular, in some cases the childbirth represents itself an event associated with a reduction of depressive symptoms (95), thus biasing the effectiveness of the experimental intervention. Moreover, other contextual factors (such as support by family members, changes in the daily routine following the childbirth, or starting a pharmacological treatment) can have an impact on the long-term outcome. We aim to control for such variables using a statistical procedure, in order to accommodate for the impact of all these confounding variables.

Furthermore, the postnatal psychoeducation programme is suitable for clinical use as it is relatively brief and can be delivered by postnatal unit nurses and midwives after a short period of training.

Another strength of this study is the use of the EPDS as screening measure, which will allow comparisons with data from other international studies.

In order to detect patients affected by both major and minor depression, the cut-off at EPDS of $\geq$10 has been selected (71) and it will have to be complemented by the clinical judgment (96).

The assessment tools have been selected on the basis of their wide use in clinical practice and in clinical trials (84). Therefore, the adoption of these instruments will give us the opportunity to compare our findings with those of other previous studies. Moreover, the HAM-A and HAM-D are well-known, validated, reliable, and easy to use scales, and have been selected in order to not overburden the researchers and mental health professionals involved in the study with a specific training on the use of assessment tools (84).

Our study has some limitations. The first is the fact that the study will be carried out in one center only, with a consequent reduced generalizability of findings. However, this can be considered as a pilot study representing the basis for a larger multicentric study that our research group is planning.

Another possible limitation is the exclusion from the study of women with psychotic disorders or women with depressive disorders before pregnancy. This methodological choice was due to the need to assess and treat only patients with an onset of depression during pregnancy. Another possible limitation is the lack of a standardized pharmacological treatment for women that will be recruited for the RCT phase. Despite a standardized pharmacological treatment could be useful to assess more precisely the efficacy of the experimental psychoeducational intervention, we decided not to standardize the pharmacological treatment in order to treat women with PD under ordinary conditions. In any case, the pharmacological management of pregnant women will follow the NICE guidelines (62).

CONCLUSIONS

Perinatal depression represents a serious threat for mental health, also considering the detrimental consequences for children. Therefore, it is an ethical imperative to identify new strategies for adequately treat such conditions and reduce the long-term negative impact on the mothers as well as their babies and family members. We hope that the study we will carry on will help to improve the clinical, psychosocial, and family management of perinatal depression.

AUTHOR CONTRIBUTIONS

LS, VC, ML, MT, GS, and AF designed the study and wrote the protocol. AF, VDV, ML, and LS organized the training and supervision for mental health professionals. ADC, GF, VC, FZ, and VG performed the literature search. GS developed the plan of statistical analyses. LS, AF, MT, and ML coordinated the study.

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Promotion of Wellbeing for Children of Parents With Mental Illness: A Model Protocol for Research and Intervention

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Background: The main objective of this project is to create a research and intervention model to promote large-scale implementation and evaluations of generic very brief interventions for children of parents with mental disorders (COPMI). Feasible interventions for COPMI aged 0–18 years are highly needed, as this is a large high-risk group in society. Reducing behavioral problems and enhancing wellbeing for families with parents affected by any mental disorder are important preventive initiatives. One key prevention strategy is to reduce the risk and expression of psychopathology in children and to promote wellbeing. The present model protocol offers an intervention for children of parents with mental disorders internationally based on a model already implemented in the Netherlands and Norway.

Methods: Participants will be parents receiving treatment in mental health services in participating countries and their minor children aged 6–18 years. Participants should be randomized into an intervention group or control group. Data should be retrieved from electronic patient journals (demographics, DSM 5/ICD-10, SCID, MINI) as well as from assessment measures administered at baseline and follow-up, including the KIDSCREEN-27, Strengths and Difficulties Questionnaire (SDQ), Parents' Evaluations of Developmental Status (PEDS), Parenting Sense of Competence (PSOC), Resilience Scale for Adolescence (READ), Guilt and Shame Questionnaire for Adolescents of Parents with Mental Illness (GSQ-APMI), Mental Health Literacy Scale, and Parent–Child Communication Scale.

Results: The hypothesis is that there will be improvements of child behavioral and emotional problems, and outcomes in the project will be reported in terms of parent´s diagnosis, child behavioral and emotional problems, child wellbeing, family communication and functioning, as well as participants' satisfaction.

Discussion: This multi-site international protocol will focus the attention of European scientific and policy makers toward COPMI. This young segment of the population is presently almost completely neglected in most European health policies, despite having a large burden of disability and being at risk of transgenerational transmission of psychopathology. We will further discuss the feasibility of a very brief intervention aiming at preventing mental disorders in young people.

Keywords: children of parents with mental disorders, mental health care for adults, risk and protective factors, wellbeing, prevention

INTRODUCTION

Mental disorders in parents are a major biological and environmental risk factor to which many young people are exposed. About 15–23% of children live with a parent with a mental disorder worldwide (1, 2). These children are 5.2 times more at risk of depression and 3.7 times more at risk of anxiety disorders compared to their peers (3). Children of parents with mental disorders are also at risk of poorer intellectual and social outcomes (4), affect dysregulation (5), behavioral problems (6), impaired attention, and reduced overall adaptive functioning (7). Research has also shown that children of parents with mental disorders have higher rates of substance abuse and multiple diagnoses (8), as well as a lower occupational status. The transmission of parental psychopathology to children can lead to similar (transgenerational equi-finality) as well as different (transgenerational multi-finality) clinical outcomes than their parent's diagnosis (9–11). The risk factors and adverse outcomes create an amplifying vicious cycle, mandating preventative action (12).

Living with a parent suffering from a mental disorder may imply the exposure to a variety of risk conditions, including: a) an adverse family environment characterized by poor parenting, high stress reactivity, emotional vulnerability, and compromised family functioning; b) experience of guilt, stigma, shame and loneliness, and perceptions of lacking social support and social acceptance; and c) the reversal of care-giving ("parentification") (13). According to Hosman and colleagues (14), the adverse outcomes for these children are the result of a complex interplay between three systemic levels (parent, child, and parent–child relationship) and potential risk factors (such as parents' mental disorder, lack of social support, poor financial conditions, marital discord, etc.). Research has shown that impaired parent–child interaction is the environmental risk factor that explains most variance in child psychological problems (27.2%) (15).

However, through carefully designed interventions addressing this interplay, one can enhance wellbeing for families with parents suffering from a mental disorder. Interventions aiming at reducing the impact of risk factors within the family context and strengthening children's ability to cope are promising, although the results in terms of effect size seem still of limited amplitude. A meta-analysis, from 2012 on 13 trials on preventive interventions for mental disorders in offspring, indicated how the risk of developing parents' mental disorder could be decreased by 40% (16). In a recent meta-analysis, 50 randomized controlled trials on preventive interventions for mental disorders in offspring showed small, but significant effect sizes, stable over 12-month follow-up, for programs enhancing the mother–child interaction (17). Interventions addressing parents and children jointly produced overall larger effects. Nevertheless, the conclusion of this review was that there is a scarcity of high-quality studies that effectively reduce the high risk of COPMI for the development of mental disorders.

Targeting children at risk and approaching them through their parents, rather than waiting until they become adults, will strengthen the reach of youngsters at their most critical point for the onset of mental disorders (18). Some research has explored experiences of youngsters living in these families and their expressed needs (19), and results show that minor children want more information about their parent's disorder and practical support related to coping with the family situation. Moreover, children of parents with a mental disorder should be expected to be as heterogeneous as the group of their parents. In order to understand their life lived experiences and needs, health professionals and social workers therefore need to listen to them. Preventive intervention could provide more complete and effective care provision for parents already in care (20). Including a whole family approach could increase treatment outcomes in adult mental health care as well as patient satisfaction with service providers. For these reasons, healthcare systems seriously need to consider preventive programs for children of mentally ill parents.

As suggested by a pioneering Australian example, it is imperative that such programs are offered to the public once their effectiveness has been established in terms of immediate and long-term outcomes (21), through both quantitative and qualitative data (22). Interventions should be designed to reduce the impact from stress and lack of quality care within the family context (related to poor or absent communication, poor understanding of disorders, insufficient parental care, lack of clinical support, or negative attitudes toward help-seeking, etc.), as well as to strengthening children's ability to cope by informing them of their parents' mental disorder and to supply both emotional and social support.

Inspired by Beardslee and colleagues (23) in the US, various programs have been developed in Australia, Canada, Finland, the Netherlands, and Norway in order to promote youth mental health and reduce risk factors linked to living with parents affected by a mental disorder (24), mostly affective disorders. Family Talk (25) was the first structured family-based preventive intervention, and because of its demonstrated efficacy, this program has been implemented in various countries worldwide (26). As an alternative to family approaches, other programs offer group-based cognitive–behavioral preventive interventions (27) and treatment coordination tailored for individual families (28). Web programs have also been tested. Programs vary in length and include very brief interventions made up of 1–3 sessions (26, 29). Some interventions focus only on parents instead of the whole family (30).

There is a great heterogeneity in the preventive programs for children as well as in their documented efficacy (17, 31). The common component across these preventive initiatives is the provision of psychosocial education on how to cope with parental mental disorder to families and children. However, further evaluation is required to examine what interventions work and for whom (e.g., sample characteristics), and through which mechanisms (e.g., program components and fidelity). Risk screening seems to be necessary to ascertain the type and intensity of support that best meets the risk profiles and needs of individual children and families (16).

At odds with the promising preventive initiatives in this field, many countries in Europe neglect these children's needs or have no structured responses in terms of dedicated mental healthcare pathways or programs. The lack of prevention in many EU countries may stem from culture-mediated lack of consideration of the service users' families (including siblings, partners, and children) in mental healthcare settings, and hence individual- or illness-centered care and treatment models. In addition, specific economic, organizational, and political issues may contribute to the lack of a child focus (i.e., resource distribution may be limited, poor or unavailable protocols for intervention, lack of collaboration between adult mental health services and the rest of the healthcare social care and lack of educational resources).

In 2015, Giovanni de Girolamo, M.D., initiated a joint application for funding from Horizon 2020 named PROCHILD. He created an international consortium with the aim to develop an international multi-site research project to evaluate and compare the effects of a very brief intervention across the nations collaborating in the consortium. The members of the consortium were experienced researchers and clinicians in the field of parental mental disorder and their children. The research and intervention model protocol described in this article is derived out of the PROCHILD protocol.

The aim of the present project is to provide a research and intervention model protocol to evaluate a very brief intervention for children of mentally ill parents adapted for a large-scale implementation. The intervention is based on an existing preventive strategy already adopted in the Netherlands and Norway called the "Child Talks" (29, 32, 33). The intention is to contribute to the current state of the art, by providing a model protocol for a feasible and widely replicable preventive intervention. This is feasible as it makes it possible to:

1. Implement a structured and brief preventive intervention targeted at families with children aged 6–18 years old and one or both parents suffering from mental disorders.
2. Evaluate the efficacy of a very brief intervention compared with treatment as usual.

METHOD

Participants

Participants should be diagnosed parents of minor children aged 6–18 years old where parents are receiving outpatient or inpatient treatment at mental health services for any mental disorders. A total of two to three treatment clinics in each participating country should serve as intervention sites. The minimum final sample size for each experimental condition should be N = 82.

Recruitment

The primary investigator of an international study should establish an international consortium with researchers from the COPMI field represented. Each of the participating researchers should have access to relevant clinical sites *via* university hospitals and clinics. In each country, there should be cluster randomization (site-randomization) of at least two to three participating adult services. Participants should be recruited through adult mental health services where parents are in treatment, inviting patients and their families to take part in the study as part of the treatment offered. Recruitment should be supported by all health care and social workers in each site, and posters and flyers should be available to inform parents. The intervention to be evaluated should be randomly assigned to the experimental or control condition (see **Figure 1** for details).

Inclusion criteria: Families should have at least one parent in treatment for any psychiatric disorder (according to DSM 5 or ICD-10 diagnostic criteria) and at least one child aged 6–18 years.

Exclusion criteria: Parents with current substance or alcohol addiction, who are acutely ill or actively suicidal parents, and parents with serious physical co-morbidities should be excluded, as these families need more intense interventions. Parents who are not able to provide consent/assent due to language or other difficulties and parents with an IQ ≤ 70 or indication of intellectual impairment should be excluded. Exclusion criteria regarding the child(ren) include ongoing psychotherapy and/or pharmacotherapy led by a Child and Adolescent Mental Health Service (CAMHS), as well as children in foster care or custody (even with grandparents).

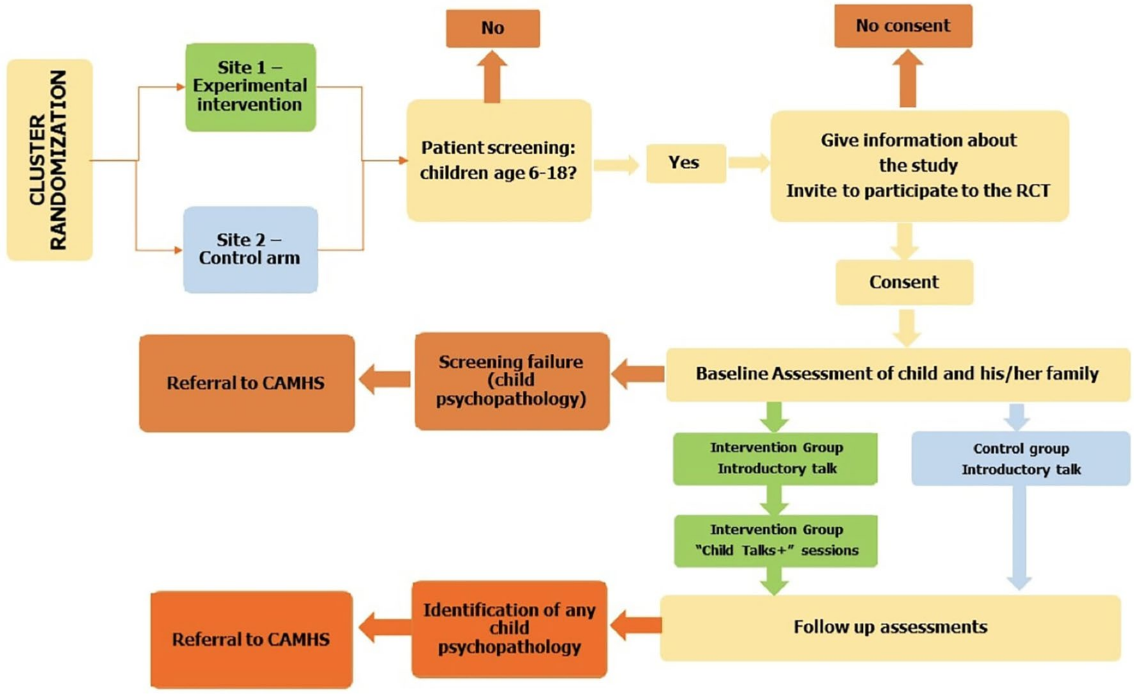

FIGURE 1 | Study design flowchart.

Procedure

A coordinator should explain the study in details to the potential participants and request their consent to contact the rest of the family. Subsequently, consent should be obtained from the participating family members. Participation by the whole family should be encouraged, although intervention can start even when only part of the family (at least one parent and one child) is willing to accept. In order for children (under the age of legal consent) to take part in the study, the parents' consent is required.

Participants should be provided with a calendar-based sessions/assessment schedule and receive reminders for their appointments. For the intervention group, two facilitators (trained in the intervention strategies) should conduct each session. Trained personnel should conduct assessments for all groups.

Design

The design of the research is a cluster-randomized controlled trial evaluating the effects of an improved version of the Child Talk Intervention called Child Talk+. In each country, the participating adult services (clusters) should be randomized into to the following experimental conditions:

Experimental group. After the baseline assessment (T0), recruited patients/families in participating services should be given the manualized "CHILD TALK+" intervention (four weekly 45-minute sessions, with trained facilitators); see **Figure 2** for the outline. After the intervention, assessment should be repeated (T1). Parents and children should be followed up at 6 (T2), 12 (T3), and 18 (T4) months. The contents and language of sessions should be adapted to the type of parental disorder and the age group of the child(ren) (6–12 or 13–18 years).

Control group. After the baseline assessment (T0), participants should receive information session about the mental health of children living with a parent affected by mental disorder, including risk and resiliency factors for the child(ren). The rationale for this experimental condition is that several European countries have policies, which mandates health personnel to identify and provide care for children of parents with mental disorder. A true control condition with no information would force health personnel in such countries to break the laws, and therefore a control condition including information about the risks for the children is necessary to include such countries. After the introductory session, postintervention assessment should be conducted (T1). Participants should be followed up at 6 (T2), 12 (T3), and 18 (T4) months. Support material should be available, but with no active role of facilitators in transferring this information into family practice.

Randomization. Cluster randomization will buffer against contamination effects, and each cluster will only deliver intervention/no intervention representing one experimental condition. The trial should be conducted according to Good Clinical Practice, reported according to CONSORT guidelines, analyzed according to SPIRIT guidelines, and registered at www.clinicaltrials.gov.

Intervention

Child Talks+ is an intervention where the mental health workers talk with the family about the situation of the children and their needs when a parent struggles with mental health issues. This intervention was developed in the Netherlands (32) and has been part of regular practice for two decades there. The updated intervention Child Talk+ comes with a manual that describes the process of carrying out four separate conversations/meetings; two initial conversations/meetings with the patient and possibly his/her partner, followed by two conversations/meetings with the patient (and partner) and the children involved (34).

Meeting 1. The initial meeting is preferably to be conducted with both parents/caregivers. In the beginning of the meeting, the purpose of conversation one should be explained to the parents/caregivers. The mental health workers should talk to the parents about the potential consequences of the mental disorder on their children and the family life. Parents should be given relevant information about the potential impact on the children, and they should be informed about possible protective factors.

Meeting 2. The second meeting is also conducted with both parents/caregivers. The purpose of this meeting is to supervise the parents and inform them about how they can discuss mental disorder at home. Examples on how to address these issues should be given, and the mental health workers should practice with the parents on how to talk to children about mental disorder, for instance, through role-play. At the end of meeting two, preparations for the next meeting should be done together with the parents/caregivers.

Meeting 3. In meeting three, the children also participate, together with the parents. The main purpose of this conversation is to get an overview of how the children are coping with the situation. The children's experiences should be discussed. Another important purpose of this meeting is to give emotional support and to provide information about the parent's mental disorder to the children (by the parents supported by the mental health worker) as prepared in meeting two. If the children have any questions, they should be answered by the parents or the mental health workers.

Meeting 4. The final meeting is with both parents/caregivers and the children if possible. The purpose of this meeting is to sum up and evaluate the previous meetings. Any questions the family may have should be addressed. The possible next steps for the family should also be discussed, and the mental health workers should assist the family in order to seek additional support if necessary. The intervention is illustrated in **Figure 2**.

The intervention allows the parents/patients to describe their children's resources and vulnerability and to participate in planning how they want their child to be informed of the family situation. The intervention includes the children through questions about their understanding and experiences of the family situation, and the children's view of what may improve their situation.

Intervention Integrity

The professionals should follow the manual for the intervention and complete standard checklists (logbook) for each session to ensure this.

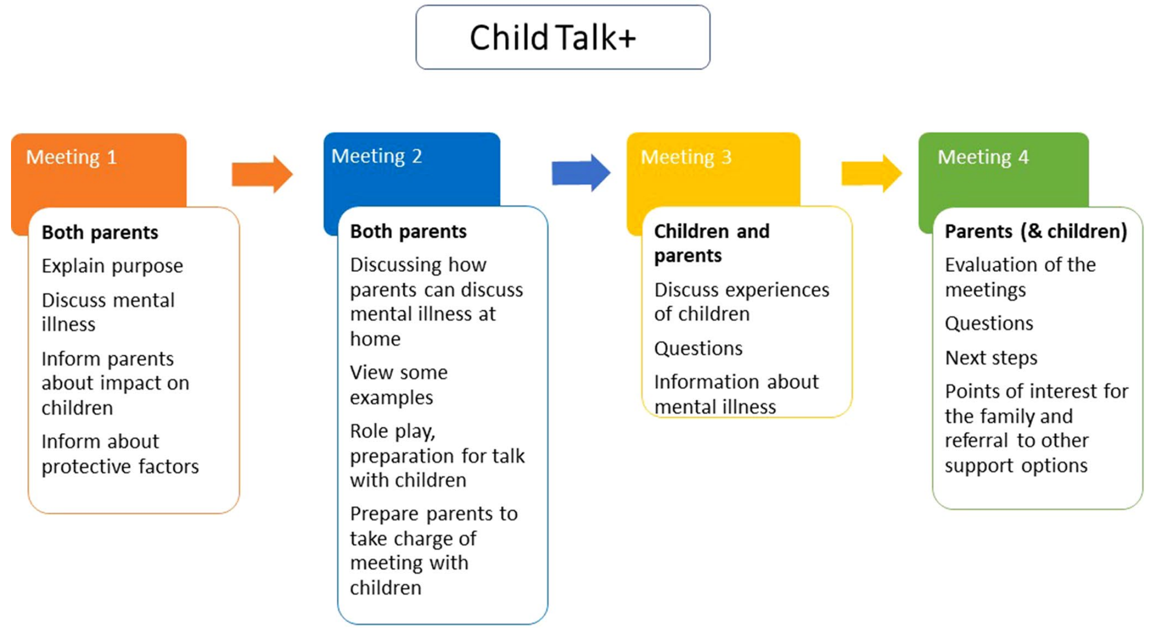

FIGURE 2 | Intervention outline.

Training

Training for facilitators should be provided according to manualized procedures. A set of booklets and slides should be created as consultation materials for both intervention and control conditions.

Measures

Multiple informants, including the children themselves, will contribute to tap a variety of areas at baseline, postintervention, and follow-up assessments. All assessments will be conducted individually with each subject, and these include: child wellbeing, child resilience, feelings of guilt and shame, child problems and development status, parents' evaluations of child developmental status, parent–child communication, child mental health literacy, and parenting competence. The following measures are suggested:

Sociodemographic variables: Parental gender, age, marital state, living situation, education, work, income, as well as parental diagnosis and severity of parental mental disorder. Diagnosis can be retrieved from clinical records. Diagnoses should be based on structured clinical interviews as SCID or MINI (35) and be codified with DMS IV, DSM 5 (36, 37), or ICD-10 (38). Severity of parental mental disorder parent should be retrieved from clinical records or GAF score (36). Children's baseline characteristics should include age, gender, living with mentally ill parent or not, total number of siblings, and educational attainment.

The health-related quality of life (KIDSCREEN-27) (39). The KIDSCREEN-27 is a measure for health-related quality of life for children from 8–18 years of age. It contains 27 items building five subscales: physical wellbeing, psychological wellbeing, autonomy and parents, social support and peers, and school environment. A 5-point Likert response scale is used in all subscales. All scores are reported as T-values, with higher scores indicating higher health-related quality of life. KIDSCREEN-27 was found to be a reliable and valid measure of quality of life in children and adolescents (40). Answering the KIDSCREEN-27 requires 10–15 minutes.

The Resilience Scale for Adolescence (READ) (41). READ is a self-report questionnaire measuring resilience: the ability to handle stress and negative experiences. READ is a 28-item scale with positively formulated items organized in five subscales: personal competence, social competence, social support, family cohesion, and structured style. Statements are answered on a 5-point Likert scale from 1 (completely disagree) to 5 (completely agree). Higher scores indicate higher degrees of protective characteristics associated with resilience within each domain. Subscale scores are summarized into a total score for resilience. It takes 5 minutes to complete the questionnaire. READ shows adequate psychometric properties and promising validity when correlated with measures of mental difficulties (42).

Guilt and Shame Questionnaire for Adolescents of Parents with Mental Illness (GSQ-APMI; 3). This questionnaire includes 10 items, 5 items measuring shame, and 5 measuring guilt. Adolescents are to answer how often they have experienced feelings of guilt and shame, with answers on a 5-point Likert scale from ranging from 0 (never) to 5 (always). Reliability scores were found adequate in a previous study (43).

The Strengths and Difficulties Questionnaire (SDQ) (44). SDQ is a brief behavioral screening questionnaire for children aged 3–16 years old. The scale is composed of 25 items, divided between 5 scales: emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial behavior. Statements are answered on a 3-point Likert scale ranging from not true, somewhat true, to certainly true. Reliability scores have been found adequate in previous studies (45).

Parents' Evaluations of Child Developmental Status (PEDS) (46). PEDS is a 10-question measure. The first item is an open-ended question where parents describe any concerns they may have about their children in terms of behavior, learning, and development. In the following eight questions, the parents consider whether they have concerns in each developmental domain, and the final question probes any additional concerns. PEDS determines whether children are at a) high risk for developmental problems, b) moderate risk for developmental and/or mental health problems, c) limited risk but in need of in-office advice, or d) limited/no risk (47). Reliability scores have been found adequate in previous studies.

Parent–Child Communication Scale (48). The Parent–Child Communication Scale consists of one scale for children and one for parents. The child report consists of 10 items measuring children's perceptions of their primary caregiver's openness to communication. Statements are answered on a 5-point Likert scale from 1 ("almost never") to 5 ("almost always"). The parent report reflects the child's perception of the primary caregiver's effort to maintain open communication with him/her. The child communication scale reflects the frequency with which the child communicates his/her feelings and problems with the primary caregiver.

Children's Mental Health Literacy Scale (49). The scale examines children's knowledge of mental disorder, recovery, and stigma. The scale consists of multiple choice questions developed for children of a parent with a mental disorder. The scale is currently being tested and will be ready for dissemination during the next year.

Parenting Sense of Competence (PSOC) (50). The PSOC is a 16-item measure intended to assess parents' beliefs that they are capable of doing a good job parenting their child. It is comprised of two subscales and is rated on a 6-point scale from 1 ("strongly agree") to 6 ("strongly disagree"). The efficacy subscale measures parents perceived competency (e.g., "being a parent is manageable, and my problems are easily solved"), while the satisfaction subscale measures parental satisfaction (e.g., "being a parent makes me tense and anxious"). Research on the PSOC has demonstrated adequate reliability and validity when used with parents of young children (50).

User satisfaction. The satisfaction of healthcare users should be evaluated by a scale tapping into issues related to challenges of being a parent with mental health problems.

Sample Size and Statistical Power

Based on the results in Prchal et al. (51), the delta score, pre-post 4 months, of KIDSCREEN-27 in a single group (similar to an experimental group with four meetings) is equal to 4.28. Therefore, in a precautionary way, we hypothesized a lower difference pre-post of about 2.5 between the experimental group (four meetings) and the control group (one meeting). Considering the longitudinal design of this model study protocol, we hypothesized a preventive correlation between two evaluations (baseline-post treatment) of 0.5 so that the pooled SD of the scores changes will be 4.90. With such data and using a two-tailed paired t-test with confidence level of 95% and a power of 0.8, the estimated sample size is N = 60 for each group (experimental and control group). Considering a drop-out rate of 25%, the minimum sample size is of N = 160 (80 per group). Given that all children of the same sick parent can be involved in the trial, we also have to consider the ICC for the intra-family correlation. Although this ICC is likely to be lower, we hypothesized an ICC of 0.04. The average number of children in European families is quite variable, ranging from 1.2 in Portugal to 2.0 in France (2014 data). Since we also have to account for non-European countries, we hypothesize an average number of children per family of 1.5. The design effect in this case will be 1.02, increasing the sample size to about N = 164 (82 children per group). Finally, this sample size should be adjusted for the multicenter design of the study. DE is defined (52) as: $DE = 1+(N/m-1)*ICC$, where m is the number of sites, and ICC is the intra-class correlation coefficient (e.g., ICC = 0.03). Therefore, the minimum final sample size for each group will be equal to $N = 82*DE$.

Statistical Analysis

Group comparisons on demographic variables can be carried out using ANOVA or chi-square tests, depending on whether the variables are continuous or categorical.

We suggest to test three specific questions about group differences over time: 1) Are there any group differences in change from pre- to post-interventions? 2) Are there any group differences in change from pre-intervention to follow-up? And 3) are there any group differences in change from post-intervention to follow-up? Rausch, Maxwell, and Kelley (53) argue that these specific questions should be analyzed using ANCOVA, controlling for the pre-score in all analyses to maximize power. We therefore suggest using ANCOVA and to use the pre-score as covariate in all analyses (53). In order for the ANCOVA to be valid, there should be no treatment group differences on pre-intervention measures. In order to test whether the intervention and control group were different at pre-intervention measures, we suggest using a one-way ANOVA. Effect sizes should be calculated according to suggested methods (54).

Ethical Considerations

Each participating country should apply for ethical approval of the project from their relevant Regional ethics committees. There are several ethical dilemmas, which could be discussed in such application: a) what are the norms to which parents with mental disorders feel obliged to follow, when they try to be good parents; b) how far are they able to follow these norms, or when and why do they fail to follow them; and c) what are the cost of not intervening to stop the transgenerational transfer of mental disorder.

In cases of screening failure due to the child(ren)'s overt psychopathology or in the event that during the trial, or at follow-up assessment, serious concerns about the child(ren)'s psychopathology should emerge; the research team should act according to country-specific guidelines and norms, as agreed with regional ethical committees (i.e., referral to Child and Adolescent Mental Health Services).

Dissemination

Data from projects using the present protocol should be shared and available for researchers *via* accepted data repositories. Principles for storing data in national databases and cross-national exchange should follow relevant laws and guidelines. Results should be published internationally and presented at conferences, as well as disseminated *via* international research groups and collaboratives.

RESULTS

Outcomes should be reported in terms of demographics, parent's diagnosis, child behavioral and emotional problems, child wellbeing, family communication and functioning, as well as user satisfaction. The expectation would be to find greater and more sustained improvements for these outcomes in the intervention group than in the control condition, and test results should be presented.

DISCUSSION

Research projects based on the model study protocol presented in this paper will draw the attention of scientific and political communities toward children of mentally ill parents. This young segment of the population is presently almost completely neglected in most European national health policies, despite carrying the largest burden of disability in this age group. On a daily basis, this population faces social, biological, and environmental risk factors threatening their mental health.

The European context is not homogeneous, but only very few countries (e.g., Netherlands Norway, Sweden, Portugal, and Finland) are currently implementing protocols for children with parents affected by mental disorders. The rest of Europe implements at most, less structured initiatives in a few areas, with no data available in terms of evaluated clinical effectiveness (and costs). A universal approach has never been fully adopted. Thus, our model protocol is consistent and complementary as it aims to implement and test a very brief intervention for children of parents with mental disorders to prevent the onset of mental ill health, reduce disability and symptomatology, and ameliorate developmental disruption. Furthermore, other universal interventions may also be tested using this protocol.

The advance of the current state of the art is twofold. First, for those countries where the intervention has already been implemented, this study will i) offer the chance to test its efficacy through the replication of the results and ii) assess its external validity, exporting the model to other countries. Secondly, countries currently with no structured intervention will be able to implement it in their own health services, testing its efficacy, and evaluating its feasibility. The result of any national or international research projects based on the present model protocol could make a platform for establishing guidelines to be disseminated throughout Europe and beyond. Such guidelines would contribute to: a) recognize the needs of selective prevention initiatives, b) define the minimum requirements in formulating a protocol in each country, and c) ensure greater homogeneity of health policies in Europe and worldwide, regarding prevention and support for all minor children with parents suffering from severe mental disorders.

Improving youth wellbeing and good health is a key issue of current EU and WHO policies. The Child Talk+ intervention is primarily oriented toward promoting positive mental health for young people whose parents suffer from mental disorders. It aims at boosting existing service models. Interventions like this one will create an innovative, evidence-based intervention platform, which will provide a solid basis to improve existing healthcare systems. The present model protocol also opens the possibility to evaluate the sustainability, feasibility, and cost-effectiveness of a very brief intervention in different sites and countries. Results from such projects will inform policy makers and foster decisions that will increase the cost-effectiveness of care, thus improving the therapeutic management of patients with young children and help define preventive strategies for children's wellbeing.

In particular, research projects based on the present model protocol will address the expected impact, as follows:

Improved support and parenting among mentally ill parents. By introducing evidence-based intervention strategies, improving early recognition of children's ill health and malleable risk factors, this RCT will support a family approach in the current adult mental service. The intervention will decrease the potential impact of risk factors related to stressful environmental stimuli within the family, such as poor or absent communication of mental suffering, poor understanding of symptoms or disorders, insufficient parental care, and lack of clinical support or help-seeking. Patients will receive acknowledgement for their parenting role and support in their effort to take good care of their children although they have mental health problems. Its replicability will assist in creating a 21st century clinical framework including neglected children's needs and to promote improved integration of healthcare.

Improved wellbeing among COPMI through supportive interventions that will help them cope with a demanding family environment (created by the fragility and complexity of a parent with mental disorder). This will enhance individual resources to cope with these stressors by providing information about parents' mental disorder and giving families emotional and social support. It will also reduce negative experiences and negative emotions that could lead to feelings of intense discomfort. By limiting the negative effects of prolonged exposure to stress, the medium- and long-term impact will decrease the probability that youngsters' psychological distress leads to mental disorder.

Establish preventative strategies favoring the mental dimension of healthy childhood by allowing youngsters to meet age-specific developmental goals. Research projects based on this model protocol will aid policy makers and service providers at local, national, and international levels concerning the policy, strategic, clinical, and organizational changes needed to ensure appropriate primary and secondary prevention programs. It will also help integrate treatment for this under-served clinical population, promoting the mental and physical dimensions of healthy childhood.

Establish recommendations for the integrated treatment of patients with mental disorders and their children. Research projects based on this model protocol will foster the development of recommendations for training programs targeted at clinicians and allied professionals on how to improve mental health care for patients who are also parents, as well as good collaboration between AMHS and CAMHS. Local variations in service structures, healthcare provision, and clinician training will be taken into account.

AUTHOR CONTRIBUTIONS

All authors have, CR, KD, GS, CL, TA, FS, AY, PC, RM, TS, MB, AS, GP and GG, contributed in the conceptualization of the model protocol with regard to the design and methods, as well as having read and commented on the manuscript text. CR, KD, GS, and CL have put the paper into writing. All authors have approved of the final version of the paper.

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