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Identifying and addressing the impact of exposure to maltreatment and experience in children and child serving systems of care

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Identifying and addressing the impact of exposure to maltreatment and experience in children and child serving systems of care

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Table of contents

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Editorial: Identifying and addressing the impact of exposure to maltreatment and experience in children and child serving systems of care

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KEYWORDS

trauma, systems of care, maltreatment, child, adolescent

Editorial on the Research Topic
Identifying and addressing the impact of exposure to maltreatment and experience in children and child serving systems of care

Child maltreatment is prevalent and contributes to a wide range of emotional and behavioral issues across one's lifespan. The extant literature on child maltreatment includes its epidemiology, neurobiology, clinical impacts, and related treatments. Over the last several years, increasing attention has been placed on the experiences and impacts of systems of care for children who have been exposed to maltreatment. It was in this context that Frontiers solicited the manuscripts for this Research Topic. In reviewing the work of the 12 teams who submitted manuscripts for this Research Topic, we noted several themes, each of which represents a lesson from the authors and a call for ongoing investigation into understanding how to identify and address risk factors for maltreatment, recognize those affected, and organize systems of care more effectively to provide support. Although specific works are highlighted in each lesson, a careful reading of the manuscripts in this Research Topic reflects each of the themes outlined below.

Lesson 1

Research must reflect the risks and patterns of maltreatment worldwide. Naved et al. link social determinants, including a more patriarchal culture, to the risk of exposure to violence among boys and girls. Wakuta et al. focus on traumatic interactions in school settings and Zhang et al. explore the impact of parental protection/overcontrol as a risk on the experiences of university students in China. Although not directly examining maltreatment, Au-Yeung et al. describe important work to support the well-being of Indigenous youth.

Lesson 2

Research must reflect a broader range of traumatic exposures that can contribute to emotional and behavioral problems in children and youth. In the work of Wakuta et al. the impact of teacher-student interactions and later distress is connected, with Zhang et al. linking parent-child relationships, specifically protection and overcontrol to emotional and behavioral health outcomes. Harris et al. describe the complex relationships within families with children who display problematic sexual behaviors; the authors argue for careful consideration and compassion for the experience and well-being of children who exhibit these traits, along with vigilant and comprehensive care for the recipients of these behaviors, when planning effective family-based interventions.

Lesson 3

Research must reflect the full range of outcomes related to maltreatment exposure. Wakuta et al. examine the phenomenon of Hikikomori, or severe social withdrawal, in relation to traumatic exposures in schools, while in the study by Yu et al. a relationship is observed between co-existing depression and anxiety related to ACES exposure from the UK Biobank data. Thompson and Svendsen explore the characteristics and needs of youth presenting with problematic sexual behaviors. Palmer and Dvir use an ecological systems analysis to review the impact of trauma on individuals with autism spectrum disorder (ASD) and Intellectual Disability (ID).

Lesson 4

Research and practice must continue to address stress, trauma exposure, and vulnerability to it. Au-Yeung et al. describe early efforts to bring the JoyPop phone application to Indigenous youth, who are at elevated risk for maltreatment. McTavish et al. offer a complementary, clinician-focused discussion and describe a strong case conceptualization, rather than narrower approaches, as a critical frontline tool for serving children and families engaged in child welfare.

Lesson 5

When addressing complex system issues it is challenging to engage in thinking/working collectively. Joh-Carnella et al. describe the experiences of healthcare providers and child protection teams, identifying effective communication and gaps in collaboration. Howarth et al. describe the challenges faced by teams attempting to create a core set of outcomes to measure the effectiveness of interventions for child-focused domestic abuse. Harris et al. propose using a broader lens than “perpetrator/victim” when attempting to address problematic sexual behaviors in the family setting.

Lesson 6

Effective listening is an essential element in understanding the lives and experiences of individuals. Au-Yeung et al. work with tribal councils and Indigenous youth so as to evaluate the effectiveness of the applications within JoyPop, and discover that some of their expectations about how youth would respond to certain applications are different from their assumptions. Joh-Carnella et al. use listening methods including qualitative interviews to generate themes related to the collaboration between child protection and healthcare providers. Palmer and Dvir explore the impact of communication challenges faced by children with ASD and IDD (Intellectual and Developmental Disorder) and their impact on identifying trauma exposure. Harris et al. suggest engaging all family members, including the recipients of problematic sexual behaviors, to address family needs and goals.

Lesson 7

It is possible to accomplish more than one task at a time. McGuier et al. describe the development of identification and referral pathways for post-traumatic stress disorder (PTSD) and related mental health issues within Child Advocacy Centers that had previously focused on investigating allegations of sexual abuse and other maltreatment. Joh-Carnella et al. investigation of the experiences of both child protection teams and healthcare providers points to opportunities for more effective collaboration. The case conceptualization model described by McTavish et al. identifies ways in which system partners can bring their individual expertise to achieve a deeper understanding of families impacted by violence.

Lesson 8

There is more work to be done. Each of the manuscripts in this Research Topic points to important areas of future inquiry that require robust and sustained research investments. Continued efforts to understand all aspects of the prevention, identification, and impact of child maltreatment remain critical to the health and well-being of individuals across their lifespan.

Author contributions

JS: Writing – original draft, Writing – review & editing. WM: Writing – review & editing. MK: Writing – review & editing.

Conflict of interest

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

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Understanding the roles of the healthcare and child welfare systems in promoting the safety and well-being of children

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Introduction: The accurate identification and appropriate investigation of child maltreatment is a key priority for promoting the optimal health and development of children. Healthcare providers are often well-positioned professionals to report suspected child abuse and neglect, and, therefore, interact regularly with child welfare workers. Little research has examined the relationship between these two groups of professionals.

Methods: We interviewed healthcare providers and child welfare workers in order to examine the referral and child welfare investigation processes to understand strengths and identify areas of improvement for future collaboration. Thirteen child welfare workers from child welfare agencies and eight healthcare providers from a pediatric tertiary care hospital in Ontario, Canada were interviewed to meet the study's objectives.

Results: Healthcare providers spoke about positive experiences making reports, factors impacting reporting decisions, areas for improvement (e.g., difficulties communicating, lack of collaboration, and disruption of therapeutic alliance), training, and professional roles. For interviews with child welfare workers, identified themes included healthcare professionals' perceived expertise and understanding the role of child welfare. Both groups brought up the need for increased collaboration as well as systemic barriers and legacies of harm.

Discussion: Our core finding was a reported lack of communication between the groups of professionals. Other identified barriers in collaboration included a lack of understanding of each other's roles, hesitation for healthcare providers making reports, as well as legacies of harm and systemic inequities in both institutions. Future research should build on this examination by including the voices of healthcare providers and child welfare workers to identify sustainable solutions for increased collaboration.

KEYWORDS

pediatrics, child abuse and neglect, reporting, child welfare, healthcare, Canada

1. Introduction

Child maltreatment is a significant public health concern associated with increased adverse physical health, mental health, and developmental outcomes, along with increased morbidity and mortality (1–3). Healthcare providers in Canada are mandated to report suspected child maltreatment and play an important role in the prevention, identification, and management of child maltreatment concerns (4–6). Data from the Ontario Incidence Study of Reported Child Abuse and Neglect 2018 (OIS-2018) indicate that child welfare investigations referred by healthcare professionals are more likely to be substantiated and involve more intrusive forms of child welfare involvement, compared to investigations referred by other sources (5, 7). In general, families who come into contact with the child welfare system are often struggling in multiple domains, including concerns with economic insecurity, precarious living conditions, intimate partner violence, as well as substance use and mental health issues for caregivers (7–9). Previous studies looking specifically at hospital-based referrals to child welfare have documented these household- and caregiver-related concerns (7, 10).

The child welfare and healthcare systems are in a unique and opportune position to work together to support families, provide resources aligned with their needs, and intervene in situations of suspected maltreatment. However, few studies have focused on how these systems interact to support families; extant literature tends to focus on healthcare providers' experiences, with fewer studies examining the reception of reports made by healthcare providers within the child welfare system.

Studies looking at healthcare professionals' experiences engaging child welfare have reported healthcare providers' discomfort with and lack of confidence in reporting suspected child maltreatment (11–22). For example, one study examining healthcare professionals' experiences seeking support from the child welfare system reported a lack of routine screening for maltreatment, discomfort with discussing maltreatment, and inadequate knowledge of community resources (1). A Canadian study of the experiences of resident physicians training in a variety of medical specialties (i.e., pediatrics, family medicine, emergency medicine, obstetrics and gynecology, and psychiatry) identifying and reporting child maltreatment found that residents felt they required further training on what constitutes child maltreatment and how to identify non-physical forms of maltreatment (16). Further, the authors highlighted that study participants reported confusion around the reporting process to child welfare services and what their role was following a report (16). It could be that physicians are well-positioned and yet ill-equipped to identify risk factors for child maltreatment in order to intervene early and hopefully mitigate the need for additional child welfare involvement.

Given the dearth of literature on this topic, the current paper fills an important knowledge gap with respect to interactions between the healthcare and child welfare systems. By interviewing both healthcare providers (including physicians and nurse practitioners) who make referrals to child welfare for suspected child maltreatment, as well as child welfare workers who receive these referrals, our objective was to holistically examine the referral and child welfare investigation processes to identify gaps for future intervention at both the provider and policy levels.

2. Materials and methods

2.1. Participants and sampling

We conducted two sets of interviews simultaneously to meet the study's objectives: (1) interviews with child welfare workers with experience investigating cases referred to child welfare by healthcare professionals and (2) interviews with healthcare providers (i.e., staff physicians and nurse practitioners) who had made referrals to child welfare agencies for concerns of child maltreatment. Recruitment emails were sent out to eligible staff at two large child welfare agencies (government-funded agencies that receive and respond to reports of child maltreatment) in Ontario, Canada as well as at one tertiary care children's hospital. The recruitment email instructed interested participants to contact the study team to arrange an interview. Following this initial identification of participants through purposive sampling, snowball sampling was used to recruit further participants. Specifically, following each interview, participants were asked by the research assistant conducting the interview to identify colleagues with relevant experience who might be interested in study participation. The potential participants were then sent individualized emails soliciting their interest in the study. In total, 13 child welfare workers and eight healthcare providers participated in the study.

2.2. Ethics approvals

Ethics approval for this study was obtained from the University of Toronto ethics board (protocol number: 41000) as well as from individual ethics boards of participating institutions (i.e., the tertiary care children's hospital and child welfare agencies).

2.3. Data collection

Participants arranged interviews with research assistants via email, and interviews were conducted using Microsoft Teams video conferencing (Microsoft, Redmond, WA). One research assistant was responsible for interviewing all child welfare workers who participated in the study, while interviews with healthcare providers were conducted by a separate research assistant and the project lead. The purpose of the study and potential risks were identified to the participants prior to the interview. Consent was obtained from all participants before beginning the interview, including consent to participate in the study, conduction of the interview via Microsoft Teams, the use of the transcription function within Microsoft Teams, and a separate audio recording of the interview. All but one participant consented to the use of the Microsoft Teams transcription function and audio recording. The one participant that did not give consent did allow the research assistant to take handwritten notes. In one other interview, the audio recording malfunctioned. Data from those two interviews (both with child welfare workers) were used to support themes, but direct quotes from those interviews are not used in the current paper. All other interviews were transcribed verbatim using both the Microsoft Teams transcription function and the audio recording.

Interviews were approximately 30 min in length and were conducted using a semi-structured approach. The interview guide consisted of seven questions for child welfare workers and 12 questions for healthcare providers; questions were designed to be open-ended and had designated prompts to elicit further information from participants (see Appendix for the list of interview questions). Questions for healthcare providers focused on: training specific to assessing child maltreatment, experiences and implications of reporting suspected maltreatment to child welfare, awareness of resources outside of child welfare, and suggestions for how the healthcare and child welfare systems can collaborate to better support children and families. Questions for child welfare workers focused on: reasons why healthcare worker reports might lead to more intrusive child welfare interventions, experiences working with healthcare providers, and areas of improvement for collaborating with the healthcare system (23). Data collection continued until thematic saturation was reached, meaning participants began sharing similar information and data were not resulting in new themes (24).

2.4. Demographic characteristics

Following completion of their interviews, participants were emailed a request to complete a survey that included information on their current position, experience in their respective fields, and demographic information (i.e., age, gender, and race/ethnicity). Completion of this survey was voluntary. Demographic information was provided by 12 (of a total 13) child welfare workers and six (of a total eight) healthcare providers. See Table 1 for a detailed summary of the participant demographics.

2.4.1. Healthcare providers

All six healthcare providers who completed the demographic survey were physicians. Although two nurse practitioners participated in the study, they did not complete the demographic questionnaire. Each physician identified their role as being primarily clinical, and one physician indicated they also had a leadership role. One had been practicing for 1–5 years, three had been practicing for 5–10 years, and two had been practicing for over 10 years. Most respondents (four out of six) were in the 31–40 age range, all six respondents were female, and three identified as white.

2.4.2. Child welfare workers

Half of the participating child welfare workers who responded to the demographic survey (six out of 12) primarily conducted investigations, representing the front-end of the child welfare service continuum. Eleven of the 12 survey respondents indicated they had over 10 years of experience. Most of the child welfare workers who participated in the study and provided their demographic information identified their gender as female (10 of 12 respondents) and their race as white (10 of 12 respondents).

2.5. Data analysis

We employed a constructivist thematic analysis approach for interview coding (25). A theoretical process was used, whereby the research team had an understanding of relevant literature when considering possible themes, and coding was conducted using NVivo software (25). One research assistant coded all interviews conducted

TABLE 1 Demographic characteristics of study participants.

	N	%
Child welfare workers		
Current position		
Child welfare worker – investigations	6	50%
Child welfare worker – ongoing	3	25%
Child welfare worker – other	3	25%
Primary responsibility (current position)		
Clinical	5	42%
Leadership	0	0%
Other	6	50%
Number of years of practice		
0–5 years	0	0%
5–10 years	1	8%
>10 years	11	92%
Age		
21–30 years	1	8%
31–40 years	3	25%
41–50 years	2	17%
51–60 years	4	33%
60+ years	1	8%
Prefer not to say	1	8%
Gender		
Male	2	17%
Female	10	83%
Non-binary	0	0%
Prefer not to say	0	0%
Race/ethnicity*		
White	10	83%
Latin American	1	8%
Indigenous	1	8%
Prefer not to say	1	8%
*One worker selected two categories		
Healthcare providers		
Current position		
Physician	6	100%
Nurse practitioner	0	0%
Healthcare worker – other	0	0%
Primary responsibility (current position)		
Clinical	6	100%
Leadership	1	17%
Other	0	0%
Number of years of practice		
0–5 years	1	17%
5–10 years	3	50%
>10 years	2	33%

(*Continued*)

TABLE 1 (Continued)

	N	%
Age		
21–30 years	1	17%
31–40 years	4	67%
41–50 years	1	17%
51–60 years	0	0%
Prefer not to say	0	0%
Gender		
Male	0	0%
Female	6	100%
Non-binary	0	0%
Prefer not to say	0	0%
Race/ethnicity*		
White	4	57%
Japanese	1	14%
Chinese	1	14%
Prefer not to say	1	14%
* One worker selected two categories		

with child welfare workers and a separate research assistant coded all interviews with healthcare providers. The project lead served as a secondary coder for nine interviews (four out of eight interviews with healthcare providers and five out of 13 interviews with child welfare workers). The researchers then met to discuss the identified codes, ensure they were consistent between coders, and collate these codes into relevant themes. All themes were reviewed by the study's principal investigator, who holds a PhD in Social Work, and all themes from interviews with healthcare providers were additionally reviewed by two of the study's co-investigators who are physicians. All study investigators have considerable experience in the healthcare and child welfare fields. This collaborative process ensured the trustworthiness of the analysis.

3. Results

Pertinent themes are described below.

3.1. Healthcare providers

3.1.1. Theme 1: positive experiences

3.1.1.1. Interactions with child welfare workers

Participating healthcare providers reported that, overall, their interactions with child welfare have been positive. Participants mentioned that good collaboration between healthcare providers and child welfare workers contributes to positive experiences, especially when caseworkers "feel like a part of the healthcare team" (HCW-8). In addition, participants noted child welfare involvement can positively impact children and families, particularly when workers are supportive and can connect caregivers to helpful resources such as those that address concrete needs (e.g., arranging transportation or providing cribs and car seats), as well as those aimed at meeting social needs like mental health supports and parenting classes. One participant summarized it as child welfare's ability to "mobilize systems around the family" (HCW-2).

3.1.1.2. Impact of reporting on healthcare providers' therapeutic relationships with families

Some healthcare providers (3/8) mentioned that, in certain situations, making a report to child welfare had a positive impact on their relationship with a family. One participant shared an example of a family accessing needed support as a result of child welfare involvement, as the participant stated: "once this support was in place, actually their lives really changed for the better… I feel like they see me kind of as instrumental in that improvement actually, because I made the call" (HCW-8). Participants shared they almost always informed a child's caregiver(s) that they were making a report to child welfare, with participants stating they try to be "transparent" with families about their concerns. Participants found framing a referral to child welfare "as a support rather than an accusation," (HCW-8) and stressing that they are mandated to report can help to maintain a positive therapeutic relationship (HCW-1).

3.1.2. Theme 2: factors impacting healthcare providers' confidence in reporting decisions

Healthcare providers generally reported feeling confident in their decision to make a report to child welfare when the concern clearly fell within their duty to report. One participant outlined this as "reasonable grounds to suspect that a child has been harmed or may be harmed based on the actions or inactions of a caregiver" (HCW-5). The participant went on to share that "once one has that concern for any reason…[their confidence in reporting] is there because the duty is so clear" (HCW-5). Specifically, participants reported that concerns involving hard evidence, such as injuries that lacked a "clear explanation" or were developmentally inappropriate, and instances of medical neglect clearly fell within their duty to report.

There were situations in which healthcare providers reported feeling more uncertain about their duty to report. As one participant stated, "there is that degree of uncertainty when it's not… in your face assault, right? When it's a bit more nuanced" (HCW-7). Participants indicated this complexity emerges specifically in complicated medical situations in which there are rare or unusual medical explanations that are difficult to confirm or cases with psychosocial complexities (e.g., Factitious Disorder Imposed on Another [FDIA], caregiver and adolescent conflict, and milder supervision concerns). Participants also reported that concerns about minimal child welfare response or lack of child or family benefit from child welfare involvement contributed to the feeling of uncertainty when reporting, particularly with these complex cases.

3.1.3. Theme 3: areas for improvement

3.1.3.1. Apprehension toward reporting

Understanding that child welfare involvement can be difficult and traumatic for families, healthcare providers reported sometimes feeling apprehensive about making reports. Healthcare providers highlighted how this knowledge often led them to weigh the costs and benefits of involving child welfare when deciding to make a report. One participant stated, "the challenge is deciding when I think… there's enough risk to call, and you know deciding like what the trade-offs will be" (HCW-7). Another participant referred to child welfare as a "last resort [after] having exhausted…all the other reasonable and feasible steps" (HCW-3). Once a referral is made, participants feel child welfare workers do not appreciate the thought, time, or, at times, number of people involved in making the referral.

Further, healthcare providers reported fears of threats and legal retaliation from caregivers. Participants mentioned that there is always an "awareness" of potential retaliation from caregivers, and that it is an "ongoing reality" in their line of work. Participants also described specific incidents of receiving complaints or threats of lawsuits; some stated this fear is heightened when dealing with families who are "very confrontational" and "very resourceful."

3.1.3.2. Difficulty communicating with child welfare workers

Though healthcare providers shared that, generally, interactions with child welfare workers are positive, many explained that communication between the two professions can be difficult. In addition to scheduling issues, such as workers being busy or hard to reach, most participants reported that child welfare workers' limited medical knowledge was a primary issue. One participant stated that, "the child protection team may not always have the knowledge around the medical situation in that it often takes a lot of convincing and education to relay the actual or potential concern" (HCW-2). Further, participants specifically referenced how they felt this difference in medical knowledge created different perceptions of risk between the two professions and made communicating their level of concern particularly challenging. One participant reported: "there have been times where it feels like we are kind of living in parallel universes… their perception of risk is so different…where it just feels like we are struggling to kind of connect in that way" (HCW-8).

3.1.3.3. Disappointing response or outcome following referral

Many healthcare providers described occasions on which they found the child welfare response to their referrals unexpected, frustrating or disappointing, with results that were "not as protective as one would hope" (HCW-5). In particular, there were concerns that children and families were left without supports or services following case closure. One participant mentioned that "the level of risk has to get quite high before the response that you are hoping for is actually in place" (HCW-2). Another healthcare worker described a negative response from child welfare upon making a referral, where they felt the worker tried to discourage them from making the report because it was "messy" and complex (HCW-7).

3.1.3.4. Lack of collaboration

Participants described a lack of collaboration between the healthcare and child welfare systems. While they understood that this was often due to confidentiality concerns, participants felt there were missed opportunities for healthcare providers to assist with cases. As one participant stated, "when there's a really complex case where the medical team is so willing and able to really help the workers understand the issues, the worker instead just calls the family or shows up at the door, eliminating any opportunity for collaboration" (HCW-5). Many of the participants felt that improving collaboration between the two teams would benefit patients and their families.

3.1.3.5. Disruption of therapeutic alliance

When asked about the impact calling CAS has on their relationship with families, some participants shared how the report can harm or negatively impact the relationship. One participant commented that, "once you call CAS the therapeutic alliance is kind of shot" (HCW-7). Participants cited the "lack of trust" following a report as being particularly detrimental. Participants noted that reporting tended to have a particularly negative effect when the caregivers felt they were being blamed or accused of harming their child. According to one participant, caregivers can take the report "very personally," as though it was "an attack on them and their character" (HCW-1).

Multiple participants referenced relationships really suffering in the context of FDIA. FDIA (also referred to as Caregiver Fabricated Illness, medical child abuse, or Munchausen by Proxy, among other names) is a condition in which a caregiver induces or exaggerates an illness in their child so they receive ongoing medical care and treatment (26–28). As one participant stated, when a case involves FDIA "you know there's already a bit of tension in the relationship… and then the call can…lead the relationship to deteriorate" (HCW-8). Participants noted that, since caregivers appeared well-intentioned in these cases, they were particularly difficult both to report to child welfare as well as for child welfare to intervene. Less commonly cited reasons for the breakdown of therapeutic relationships included having a family with a history of child welfare involvement and cases involving complex concerns.

3.1.4. Theme 4: training

When asked about the level of child maltreatment training they received, all participating healthcare providers reported receiving very little formal training, unless they had specialized in child abuse pediatrics. One participant shared, "during training in medical school in general pediatrics, [child maltreatment is] a very small part of the core curriculum, but everyone has some degree of exposure. It's minimal, it's not at the level of an expert. It's mainly focused on awareness and recognition" (HCW-5). Similarly for nurse practitioners, child maltreatment was a small component of the general training they received.

3.1.5. Theme 5: professional roles

Many participants referenced the distinct roles of healthcare and child welfare professionals. One participant stated, "I think that we all have different roles… it's understanding limits and boundaries and where your role as a physician starts and finishes" (HCW-3). Another worker reported, "I appreciate and understand that we are distinct entities with distinct expertise, and I have my role and they have their role" (HCW-5). Further, healthcare professionals understand that a key element of child welfare's role is their ability to conduct investigations and gather information to determine "what is right for the child in those circumstances" (HCW 3).

3.1.6. Theme 6: bias and systemic issues with access to support

The presence of systemic issues within both the healthcare and child welfare systems was often discussed. One participant mentioned the difficulty they had connecting families to community supports, while many others commented on the current lack of mental health supports available for children and caregivers. Participants acknowledged that many children and families with "multi-system" issues often "land on CAS' lap" (HCW-4). Participants further noted that child welfare is often limited with regards to how much support they can offer to families or "what their response can be" (HCW-5). One participant indicated they felt the child welfare system is not designed to address structural barriers that exist for many of the children and families it serves.

Multiple participants raised concerns regarding biases and inequities present within child welfare based on the system's history and current practices rooted in systemic racism. One participant stated, "in the past…there's been some structural and systemic problems with how particular… populations or groups are treated [in child welfare]" (HCW-8). Another participant shared how a child's background can impact a child welfare response, stating: "it is incredibly disheartening when you can anticipate what responses will be based on differing socioeconomic status and racial background" (HCW-5).

In particular, participants were concerned with the recent shift occurring in local child welfare practice toward a less invasive approach taken in investigations involving families of color in an effort to redress systemic overrepresentation of these children in child welfare systems. Healthcare providers reported concerns that children were harmed by the hesitant response of child welfare. One participant shared, "just as it is extraordinarily flat out wrong that some of the heavy-handed action from societies harmed children in the past, I also recognize that on this side there are children who are going to be harmed by inaction and a failure of society and institutions to protect them" (HCW-5). While participants stated they understood the reasons behind these policy changes (i.e., to reduce the number of children of color in care), they were worried for their patients.

Overall, participants highlighted the importance of the child welfare system being foremost oriented around protecting children and promoting their well-being. According to one participant, "the ideal is that the child welfare system is…an organization that's just devoted to the welfare of children and that…accusation element [is] sidelined into one [small branch]… as opposed to taking over the entire like way people view the child welfare system" (HCW-8).

3.1.7. Theme 7: suggestions for collaboration

When asked for suggestions on how to improve collaboration between the healthcare and child welfare systems, the most common answer was to improve communication between the two professions. While healthcare providers acknowledged the importance of protecting confidential information, participants noted that an "ongoing dialogue" would benefit children and families through increased knowledge-sharing and support. One healthcare worker mentioned that including child welfare workers in case conferences might help improve communication.

To address the concern of child welfare workers not understanding the medical science of certain cases, some participants suggested having specialized child welfare workers who deal with medical referrals. One participant believed having a group of workers in each agency that is familiar with child abuse pediatrics could help agencies understand healthcare professionals' processes and medical decision-making factors. Similarly, another participant suggested there be more consistency with child welfare workers in cases to "have the least amount of transitions possible between worker and worker and worker" (HCW 2).

3.2. Child welfare workers

3.2.1. Theme 1: healthcare professionals' perceived expertise

When asked why child welfare investigations referred by healthcare professionals are more likely to involve more intrusive child welfare involvement and substantiation, many participating child welfare workers referenced healthcare providers' perceived expertise and credibility. Some participants reported that healthcare professionals' education and medical training better position them to recognize protection concerns. One participant emphasized this by stating that healthcare professionals "know what they are doing" (CW-13), while another stated: "we trust healthcare professionals in their jobs" (CW-12). Workers also identified the nature of protection concerns as an indicator of validation; healthcare settings reportedly see more severe cases of child abuse or neglect. As a result, the instances being reported to child welfare are more serious in nature and, therefore, more likely to be substantiated, opened for ongoing services, or involve a child welfare placement.

Some workers specified that the weight attributed to healthcare professional referrals has more to do with perceived expertise than actual credibility or family circumstances. One worker shared: "child welfare has historically… viewed the opinion of, you know, quote/un-quote professionals as more legit in comparison to community referrals or other referrals" (CW-11). According to some participants, regardless of the child protection concern, healthcare professionals are seen as reliable and credible sources that have more value attributed to their report due to their professional title.

3.2.2. Theme 2: understanding the role of child welfare

Child welfare workers highlighted both positive and negative interactions with healthcare professionals, with many identifying healthcare providers' understanding of child welfare's role as the differentiating factor. Most participants stated that when healthcare professionals are more knowledgeable of child welfare workers' responsibilities, jurisdiction, and capacity, the collaboration process is more pleasant. Too often, according to study participants, child welfare is reportedly contacted by healthcare professionals for matters that could be mitigated by other services. For instance, if a doctor is concerned about a caregiver's mental health and not concerned about the safety or well-being of the child, child welfare workers reported it is more appropriate to make a referral to a community mental health service than calling a child welfare agency to make a report.

Many participants were confident that if more healthcare professionals better understood both the role of the child welfare system as a whole, as well as individual workers' roles, both fields could engage in more effective work. One participant explained: "healthcare providers are not always informed as to how we do our work and what the process might be" (CW-3). Workers suggested healthcare professionals receive more training to help facilitate this understanding. Child welfare workers were also cognizant of the parallel learning process that should take place to create improved understanding. One participant reported: "if there was better understanding on both sides of the process…then it would be a better response [to child maltreatment concerns] and probably like a more unified one" (CW-13).

3.2.3. Theme 3: need for increased collaboration

Though workers shared having positive experiences when collaborating with healthcare professionals, many reported there is room for improvement. Whether due to busy schedules or disinterest in engaging with child welfare workers after an initial report is made, healthcare professionals can be difficult to contact. Often, over the course of their investigations, child welfare workers need to speak with the healthcare professional who made the report to get details about the family and ask follow-up questions that only the reporting party can answer. As such, participants shared feeling frustrated by the lack of communication and partnership that can characterize interactions with healthcare professionals. According to participants, creating more effective collaborative relationships between the child welfare and healthcare systems starts with open communication. One worker shared: "the more communication that we have together, the more we work together as a team to support a family" (CW-4).

3.2.4. Theme 4: systemic barriers and legacies of harm

Throughout the interviews, some workers expressed concern about child welfare's legacy of harm in Canada and the impact it has on families. Participants reported that child welfare agencies are working hard to address the overrepresentation of Black and Indigenous children in care. The racism, biases, and assumptions that plague both the child welfare and healthcare systems are being challenged by child welfare workers. One participant stated: "[child welfare has] a well-known reputation. It's not a good reputation. We've worked hard to earn it, but that does not mean that we have to fit into it" (CW-2). Child welfare workers believe that the process of challenging injustices and striving to create a more equitable system also needs to be initiated in healthcare settings to maximize the impact of these systemic changes. A worker reported: "it's a whole new narrative that our agency is trying to bring about. So, I do not know what's happening in the medical field, but that's going to create quite a lot of barriers or issues if the healthcare system also does not choose to move forward with a new narrative" (CW-11).

Child welfare workers' desire to see changes in healthcare professionals' practices results from their concern for families that have had negative experiences with the healthcare system based on systemic inequities. One participant reported: "there is a certain middle class measuring stick that [healthcare professionals are] measuring their patients and our families up against. And if they do not meet that, they are very judgmental. They are very biased of different family situations" (CW-12). The expressed concerns extend beyond individual or personal biases, but rather shed light on the shortcomings of the healthcare system's current structure. A child welfare worker shared: "I really worry about some healthcare settings being able to engage with certain families" (CW-2). To address the systemic barriers that limit families' access to equitable and adequate service provision, participants strongly believed that changes must be made in both the child welfare and healthcare systems.

4. Discussion

By taking a multidisciplinary approach to data collection (i.e., by interviewing both healthcare providers and child welfare workers), we holistically examined the child welfare referral and investigation processes in situations where potential child maltreatment is identified in the healthcare setting. In doing so, we were able to identify both the strengths as well as the areas for improvement in the collaborative relationship between the healthcare and child welfare systems. Healthcare providers represent important points of contact for children as they routinely see vulnerable children including those too young to attend school and those with mental health concerns or disabilities (5, 29–32). Especially within the Canadian context of a universal healthcare system, children may be more likely to come into contact with healthcare professionals compared to other social systems and supports.

Our core thematic finding is that both groups felt the need for greater communication, collaboration, and understanding between the two professions. Although acknowledging that the healthcare and child welfare professions are demanding of workers' time, both groups shared frustrations with the perceived lack of availability of their counterparts. It could be that explicit conversations around expectations with respect to level of involvement in the investigation process from the outset could help to mitigate some of the tension experienced between professionals during child welfare investigations. The two groups of participants described the importance of understanding each other's professional roles. Appreciating the limitations of both their own and each other's ability to assess and intervene with children and families was highlighted by study participants as a key facilitator of effective partnership. Overall, the participants indicated the importance of understanding not only the distinct roles of the two professions, but also acknowledging the limitations of their own expertise and appreciating what the other professionals can do that they cannot. Both child welfare workers and healthcare providers in this study highlighted the utility of having specialized teams that were familiar with the other profession to deal specifically with concerns of child maltreatment identified in a medical setting.

Findings from our study indicate that child welfare workers perceive healthcare providers to be expert assessors of potential child maltreatment. Paradoxically, healthcare providers in our study reported a lack of specialized training in child maltreatment accompanied by a frequent lack of confidence in identifying and reporting child maltreatment in cases that are not clear-cut cases of assault. This finding is consistent with findings from another study conducted with Canadian medical residents (16), and this dearth of training represents a significant gap within medical education and training.

Nonetheless, healthcare providers need to be attuned to signs of potential child maltreatment given their unique vantage point. Early identification and thoughtful intervention have the potential to mitigate some of the downstream effects of child maltreatment. The logical first step in addressing suspected child maltreatment is making a referral to child welfare services, where trained professionals can further investigate and intervene where necessary. Indeed, participating healthcare providers cited benefits of reporting child maltreatment. Yet, consistent with findings of previous studies, healthcare providers in our study highlighted several barriers to reporting suspected child maltreatment including fear of disrupting the therapeutic relationship with the child/family, causing increased harm to the family through involving child welfare, and being threatened with legal retaliation from the family (12–14).

While healthcare providers in our study indicated there are certain circumstances in which they are more certain that a referral to child welfare should be made, they report hesitancy in other situations. As was found in other studies, healthcare providers indicated that the decision to refer to child welfare is clear in cases with evidence of physical or sexual abuse that squarely fall within the mandate to report (12, 21). On the other hand, the decision to report is more nuanced in cases with more complex medical explanations of injuries/presenting concerns or those with other elements at play such as FDIA, caregiver/teen conflict, and some supervision concerns. Extant literature similarly states that healthcare professionals tend to hesitate to refer to child welfare in cases with increased complexity (16, 21). Oftentimes, healthcare providers attempt to mobilize supports within the healthcare setting and consult many different members of the healthcare team before making a report, as they recognize and appreciate the repercussions their report may have for the family. This could be especially true in these more complex cases where the duty to report is less clear-cut.

Systemic barriers to accessing equitable care and services along with legacies of harm within both the child welfare and healthcare systems were brought up by interview participants. In particular, they spoke of the overrepresentation of Indigenous and Black children in the child welfare system. While participating child welfare workers referred to efforts being made to redress this overrepresentation, healthcare providers identified limits to these efforts and identified possible harms with inaction. Participants identified further biases against children and families with lower socioeconomic status. Ontario child welfare-involved children experiencing economic hardship are more likely to be struggling in multiple domains including having developmental concerns and academic difficulties (33). Further, these children are more likely to be involved in substantiated child welfare investigations (33). The fact that participants identified problematic responses to families struggling with economic hardship likely represents strongly embedded societal biases against these families along with systemic factors that have left them more vulnerable to situations that might warrant child welfare involvement. Other systemic concerns including access to services were brought up by participants; these represent ongoing issues with supports available to families (34–36).

4.1. Limitations

Some limitations should be considered when interpreting study results. The sample consisted of 13 child welfare and eight healthcare providers from two Ontario child welfare agencies and one large pediatric tertiary care center. The use of snowball sampling methodology resulted in the selection of participants with similar areas of expertise. That said, we did not collect information on healthcare providers' sub-specialties beyond pediatrics. It was revealed through the interviews, though, that a large proportion of the healthcare providers who participated in our study worked specifically in child abuse pediatrics meaning they had unique perspectives even when compared to other pediatric healthcare providers. In general, the use of a voluntary sample could have selected for individuals with more knowledge on or interest in the study subject matter or those with particularly positive or negative experiences with the child welfare system. One participant declining to consent to audio recording and the use of the transcription function, and an audio technical error in a separate interview decreased confidence in the quality of the transcription of those two interviews.

5. Conclusion

To our knowledge, this is the first study that incorporates voices from both the child welfare and healthcare systems in an attempt to identify areas for improvement and strengths in collaboration between these professional groups. Healthcare providers remain an important point of contact for vulnerable children and their families to access necessary services that can support and protect them, including child welfare services. That said, child welfare involvement can be very disruptive to the family unit, and our study findings demonstrate that healthcare providers do not make the decision to report to child welfare lightly. Ideally, the child welfare and healthcare systems would be complementary to each other and work synchronously to best support children and families. Unfortunately, the results of our study identify barriers to this collaborative work, including inadequate communication and understanding between the professions, hesitations in healthcare professionals' reporting, along with legacies of systemic injustices in both sectors. Several future steps were identified that might help promote improved collaboration and, therefore, more streamlined and effective services provided to children and families. Future research might include focus groups combining professionals from both groups to encourage specific interactions and feedback about how to ethically improve collaboration. Future research might also expand to include family doctors who have a different perspective compared to pediatricians. Ultimately, best practices for education, training, and collaboration in both professions need to be ascertained to promote optimal outcomes for children and families.

Data availability statement

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

Ethics statement

The studies involving human participants were reviewed and approved by University of Toronto Ethics Board. The patients/participants provided their verbal informed consent to participate in this study.

Author contributions

NJ-C and BF conceptualized the study. EL, MK-C, and NJ-C conducted the interviews, performed the interview coding, and drafted the initial manuscript. AV, JS, and DL assisted with developing initial interview questions. NJ-C, BF, EL, MK-C, AV, and JS drafted the ethics documents, recruited the participants, and reviewed all interview codes. BF obtained the funding. All authors approved the final manuscript.

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Conflict of interest

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

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Appendix

Appendix. Interview questions

Healthcare providers

1. How long have you been working as a healthcare provider?
2. What kind of training, if any, have you received for assessing child maltreatment?
3. Over the course of your career, have you noticed signs of child maltreatment or been concerned that a child is at risk for maltreatment? *If no, do not conduct interview.*

    a. If yes, describe the most recent referral you made to child welfare.

4. Describe a time where you were very certain in your decision to make a report to child welfare.
5. Describe a time when you were uncertain in your decision to make a report to child welfare.
6. Have you ever regretted your decision to make a report to child welfare? If so, please describe.
7. Describe a time when your report to child welfare positively impacted your relationship with a family.
8. Describe a time when your report to child welfare negatively impacted your relationship with a family.
9. Describe what, if any, supports you would offer to a family in the following scenarios:

    a. You suspect the family is experiencing economic hardship.
    b. You suspect concerns for the caregiver (e.g., caregiver has substance abuse concerns, has mental health concerns, is a victim/perpetrator of intimate partner violence, or lacks social supports).
    c. You suspect concerns for the child's functioning (e.g., developmentally, behaviourally, academically, etc.).
    d. You have a strong suspicion of abuse or neglect.

10. In general, how would you describe your experiences with the child welfare system?
11. How do you expect the child welfare system to support a family you refer?
12. Do you have any suggestions for how the healthcare and child welfare systems could work together to better support children and families?

Child welfare workers

1. Are you a screening or investigating worker?
2. How long have you worked as a (screening worker or investigating worker)?
3. In the past year, have you received a referral or investigated a case that was referred from a healthcare worker?
4. Data from the Ontario Incidence Study of Reported Child Abuse and Neglect 2018 indicate that investigations involving healthcare referrals are more likely to be substantiated, opened for ongoing services, or involve a child welfare placement compared to other referral sources. Do you have any insight as to why this would be the case?
5. How do you think healthcare providers can best support a family prior to making a referral to child welfare?
6. Overall, how would you describe your interactions with healthcare professionals in the context of a child welfare referral or investigation?
7. Do you have any suggestions for how the healthcare and child welfare systems could work together to better support children and families?

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Traumatic stress symptoms and PTSD risk in children served by Children's Advocacy Centers

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Purpose: Children who experience maltreatment are at high risk for posttraumatic stress disorder (PTSD). Children's Advocacy Centers (CACs) can facilitate access to treatment following maltreatment allegations. We describe PTSD symptoms and intervention decision-making for children served by CACs.

Methods: Children served by CACs in a single state were screened for PTSD symptoms using a structured mental health screening/referral protocol. CAC staff used an electronic form that provided guidance for decision-making. We examined descriptive statistics for PTSD symptoms and risk and tested associations between child characteristics and symptoms. We described CAC staff's delivery of brief interventions and referral decisions and tested associations with child characteristics and symptoms.

Results: Two thousand and three hundred fifty children completed screening between 2018 and 2020. Almost half (45.5%) exhibited traumatic stress symptoms suggesting high probability of PTSD at the time of their CAC visit. Children who identified as female or transgender male and older children were more likely to be at high risk for PTSD. Brief interventions were delivered to 66% of children, and most were referred to evidence-based trauma treatment (53.1%) or community mental health services (39.0%). Categorization as moderate or high PTSD risk was associated with a higher likelihood of brief intervention delivery and referral to trauma treatment.

Conclusion: Many children served by CACs are likely to meet criteria for PTSD at their initial visit. CAC staff demonstrated the ability to deliver brief interventions and make referrals to mental health treatment. Use of structured screening/referral protocols may improve early identification and treatment access for children experiencing PTSD symptoms.

KEYWORDS

child abuse, posttraumatic stress disorder (PTSD), traumatic stress, children's advocacy centers, screening, care process model for pediatric traumatic stress

Introduction

Child maltreatment and associated mental health problems are critical public health concerns (1–3). At least 1 in 8 and as many as 1 in 3 children experience maltreatment in their lifetime (4, 5), placing them at elevated risk for posttraumatic stress disorder (PTSD) and other mental health problems (6–9). Sexual abuse specifically is associated with more than twice the risk of PTSD and more than three times the risk of suicide attempts (6, 10).

Children's Advocacy Centers (CACs) provide coordinated interagency investigations and services after allegations of sexual abuse and other serious maltreatment (11–14). CACs are well-positioned to identify children with mental health needs and facilitate access to mental health assessment and treatment (15).

In 2018, CACs across the state of Utah began implementing a structured protocol to screen and respond to children at risk for traumatic stress symptoms and suicidality at the time of their CAC visit. This protocol, the Care Process Model for Pediatric Traumatic Stress (CPM-PTS), provides a road map of care and electronic screening and decision support tools to assist frontline staff in screening for and responding to traumatic stress symptoms and suicidality (16). Screening for traumatic stress and referrals to evidence-based trauma treatments are critical components of trauma-informed systems of care (17). Development, implementation, and use of the CPM-PTS are described by Byrne et al. (18). In this brief report, we present results from the first 2 years of CPM-PTS use in Utah CACs. We describe the prevalence of traumatic stress symptoms and PTSD risk among children served by CACs, test associations between child characteristics and symptoms, and describe CAC staff decision-making for children reporting elevated symptoms.

Methods

Setting and procedures

The current project is an observational study of the prevalence of traumatic stress symptoms in children seen in CACs in Utah between March 2018 and February 2020. Data were available from 16 CACs that implemented the CPM-PTS; four CACs were in urban areas and the rest in rural/frontier areas (19). In most CACs, the CPM-PTS was administered by staff without clinical training (12/16; 75%), most often victim advocates. The CPM-PTS was intended for use with all children between 5 and 18 years old. Child symptoms and staff decision-making were collected through two tools built into the HIPAA-compliant, web-based Research Electronic Data Capture (REDCap) platform hosted by the University of Utah (20, 21). Two REDCap tools supported CPM-PTS administration: a client/family-facing trauma screening (Pediatric Traumatic Stress Screening Tool[1]) and a staff-facing decision support (Decision Support Tool). Children/caregivers completed the screening tool during their visit on an electronic tablet device at the CAC, and screening results were inserted into the subsequent decision support REDCap form where CAC staff documented their decisions and actions. Timing and workflows were determined by individual CACs [see (18)]. All records for these analyses were de-identified. All procedures and a waiver of informed consent were approved by the University of Utah Institutional Review Board.

Participants

Participants were children and adolescents between 5 and 18 years old visiting a participating CAC during the 2-year period for an initial forensic interview (i.e., interview with the child conducted by a trained professional to elicit facts about maltreatment allegations). We included children with complete responses to the Pediatric Traumatic Stress Screening Tool; we excluded children seen solely for therapy or follow-up, children with a primary language other than English or Spanish, and children whose records were missing date, site of administration, or age.

Measures

Pediatric traumatic stress screening tool

The screening tool was available in caregiver- and youth-report versions in English and Spanish. Caregiver report was recommended for children aged 5–10 years old and self-report for children aged 11–18 years old. Before administering the screening tool, CAC staff recorded child demographics (i.e., age, gender, race, ethnicity) and the reason(s) for the CAC visit on a linked form. The screening tool captured exposure to potentially traumatic events and traumatic stress symptoms; it also included one screening question for suicidality. Potential traumatic exposures and traumatic stress symptoms were assessed with the UCLA PTSD Reaction Index Brief Form (22). Subscales of the Brief Form assess specific domains of PTSD symptoms: intrusion, avoidance, negative alternations in cognitions and mood, and arousal/reactivity. After indicating and describing recent and remote traumatic experiences, respondents rated 11 symptom frequency items on a 5-point scale from 0 "none" to 4 "most" of the time during the past month.[2] Prior studies of the UCLA Brief Form have found excellent internal consistency ($\alpha > 0.90$) and support for the measure's clinical utility in discriminating between cases with and without PTSD using a cutoff score of 21 (22). Internal consistency for this study was excellent ($\alpha = 0.92$).

Decision support for the CPM-PTS classified risk for PTSD as high (score ≥ 21), moderate (score 11–20), or low (score ≤ 10). The moderate risk category was added to identify children within this high-risk population who may benefit from psychoeducation, brief "light-touch" interventions, and/or further evaluation. A question from the Patient Health Questionnaire-Adolescent (23, 24) was used to assess risk for suicide and/or self-harm (i.e., thoughts that you would be better off dead or thoughts of hurting yourself). The CPM-PTS Pediatric Traumatic Stress Screening Tool is freely available within the published protocol (16).

Decision support tool

The Decision Support Tool guided CAC staff through a three-step process: (1) report any new maltreatment allegations and/or respond to other identified safety concerns, (2) evaluate and respond to

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[1] In this context, pediatric traumatic stress refers to any traumatic stress experienced by children; it is not specific to medical traumatic stress.

[2] An additional item from the UCLA PTSD Reaction Index assessing distressing dreams was included to identify children who may benefit from brief interventions targeting sleep; this item was not included in scoring of PTSD symptoms.

suicidality, and (3) provide brief interventions and/or referrals to mental health care (see (18) for more details). At each step, the tool suggested appropriate actions. Staff could choose to take other actions in place of and/or in addition to those suggested. Any positive response to the suicidality screening question prompted CAC staff to administer the Columbia Suicide Severity Rating Scale (25), which classified the level of suicide risk and suggested appropriate response options based on the level of risk (e.g., safety planning, facilitating immediate crisis response) [see (26)].

The Decision Support Tool suggested brief interventions and referral options based on the domain and severity of traumatic stress symptoms, prioritizing sleep problems when present, then symptoms of intrusion and/or hyperarousal/reactivity, and finally symptoms of avoidance and/or negative alterations in cognitions and mood (27–29). Brief interventions suggested for children with elevated sleep problems included the nighttime use of diaphragmatic breathing or guided imagery. Suggested brief interventions for children with elevated symptoms of intrusion and/or hyperarousal/reactivity were daytime use of diaphragmatic breathing, guided imagery, and mindfulness. Lastly, suggested brief interventions for children with elevated symptoms of avoidance and/or negative mood were caregiver-child communication, behavioral activation, and caregiver-child special time. Staff could deliver more than one brief intervention.

The Decision Support Tool also suggested referral options for families. Referral options included follow-up with primary care provider and/or referral to general community mental health services for children categorized as low risk for PTSD and referral to evidence-based trauma treatment (e.g., Trauma-Focused Cognitive-Behavioral Therapy) for those at moderate or high PTSD risk. Staff could also recommend other actions (e.g., follow-up with existing mental health provider). Staff documented their actions and decisions (e.g., delivery of brief intervention, referral to evidence-based trauma therapy) within the Decision Support Tool.

Analyses

We analyzed data for children seen for an initial forensic interview during the 2-year study period. Screening data were considered complete if at least 10 of 11 questions on the UCLA Brief Form were completed; a missing question was assigned the mean response of completed questions. We first used Pearson's chi-square tests of independence to examine differences in missing data by child and CAC characteristics (e.g., demographic characteristics, urban vs. rural CAC location) to evaluate potential bias.

We then conducted a set of analyses focused on child characteristics and symptoms. Our outcomes were total traumatic stress symptom score and PTSD risk category. Child characteristics examined were gender, race, ethnicity, age, and reason for CAC visit as indicated by staff. We also described differences in symptoms for children seen in rural vs. urban CACs. We examined descriptive statistics and used Pearson's chi-square tests and multilevel regression models to examine associations of child characteristics with outcomes. Multilevel analyses were conducted in R using the *lme4* package; other analyses were conducted in SPSS. Multilevel models included child age, gender (cisgender male vs. cisgender female or transgender male[3]), race/ethnicity (non-Hispanic white vs. minoritized group), and concern for sexual abuse (no vs. yes) along with a random effect to account for clustering within CACs. For traumatic stress symptom scores, we conducted linear mixed models fit by restricted maximum likelihood estimation. For PTSD risk category, we conducted binomial generalized linear mixed models fit by maximum likelihood.

Our next set of analyses focused on CAC staff's decision-making and responses. We described staff delivery of brief interventions and referral decisions recorded in the Decision Support Tool. We used Pearson's chi-square tests to test for differences in responses by PTSD risk category and suicidality. Binomial generalized linear mixed models were conducted to test associations of child characteristics with delivery of brief interventions and referral decisions. Models included PTSD risk category, age, gender, race/ethnicity, concern for sexual abuse, and a random effect to account for clustering within CACs.

Results

Missing data

The CPM-PTS was adopted and administered electronically by 16 CACs. On average, CACs administered the CPM-PTS to 53% of the children they served. Screening rates ranged from 10 to 100% across CACs [see (18) for more information on implementation outcomes and determinants of use]. During the 2-year period of this study, CPM-PTS administration was initiated with 2,569 children. Nine percent of these children ($n = 219$) were excluded from analyses because of missing data (75 missing age; 114 missing symptom data; 30 missing age and symptom data), resulting in an analytic sample of 2,350 children. Missing data were more common among children seen in rural CACs compared to urban CACs (11.5% vs. 6.2%, $\chi^2 = 22.77$, $p < 0.01$) and children aged 5–10 compared to adolescents aged 11–18 (6.4% vs. 3.9%, $\chi^2 = 7.06$, $p < 0.01$). There were no differences in the likelihood of missing data by child gender, race, or ethnicity. Referral decisions were documented for 83% of the children included in analyses ($n = 1,950$).

Characteristics and symptoms of children completing the CPM

Child characteristics are presented in Table 1. Children were mostly female, white, non-Hispanic, adolescents ($M = 12.96$ years, SD = 3.36), and visiting the CAC for concerns about sexual abuse. As expected, child characteristics varied across sites due to differences in the populations of their catchment area and their criteria for service. Traumatic stress symptom scores ranged from 0 to 44 ($M = 19.20$; SD = 11.60). Close to half of children (45.5%) were categorized as high

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[3] Transgender male was not included as a separate category because of the small sample size (n=6). We chose to group transgender males and cisgender females together for analyses because both are marginalized groups relative to cisgender males.

TABLE 1 PTSD risk category and child characteristics (*N*=2,350).

	N (%)				χ^2
		Risk for PTSD			
	Full sample	Low	Moderate	High	
		653 (27.8)	628 (26.7)	1,069 (45.5)	
Gender					113.78**
Female	1,701 (72.4)	401 (23.6)	418 (24.6)	882 (51.9)	
Male	619 (26.3)	247 (39.9)	200 (32.3)	172 (27.8)	
Transgender male	6 (0.3)	0 (0.0)	2 (33.3)	4 (66.7)	
Unknown[a]	24 (1.0)				
Race					32.56**
American Indian/Alaska Native	63 (2.7)	20 (31.7)	21 (33.3)	22 (34.9)	
Asian	14 (0.6)	3 (21.4)	3 (21.4)	8 (57.1)	
Black/African American	29 (1.2)	11 (37.9)	4 (13.8)	14 (48.3)	
Multiracial	72 (3.1)	16 (22.2)	26 (36.1)	30 (41.7)	
Native Hawaiian/Pacific Islander	30 (1.3)	19 (63.3)	3 (10.0)	8 (26.7)	
Other	15 (0.6)	2 (13.3)	6 (40.0)	7 (46.7)	
White	1,824 (77.6)	499 (27.4)	491 (26.9)	834 (45.7)	
Unknown[a]	303 (12.9)				
Ethnicity					1.74
Hispanic/Latinx	313 (13.3)	80 (25.6)	80 (25.6)	153 (48.9)	
Non-Hispanic/Latinx	2,037 (86.7)	573 (28.1)	548 (26.8)	916 (45.1)	
Age					106.65**
5–10 years old	639 (27.2)	261 (40.8)	190 (29.7)	188 (29.4)	
11–18 years old	1,711 (72.8)	392 (22.9)	438 (25.6)	881 (51.5)	
Reason for CAC visit[b]—concern for:					
Sexual abuse	1,698 (72.3)	430 (25.3)	436 (25.7)	832 (49.0)	32.44**
Physical abuse	402 (17.1)	125 (31.1)	128 (31.8)	149 (37.1)	14.23**
Witnessed domestic violence	150 (6.4)	55 (36.7)	53 (35.3)	42 (28.0)	19.77**
Neglect	107 (4.6)	32 (29.9)	31 (29.0)	44 (41.1)	0.86
Harmful material(s)	91 (3.9)	28 (30.8)	26 (28.6)	37 (40.7)	0.91
Witnessed crime	47 (2.0)	11 (23.4)	15 (31.9)	21 (44.7)	0.82
Other	130 (5.5)	50 (38.5)	32 (24.6)	48 (36.9)	8.10*
Location of CAC visit					25.56**
Urban CAC	1,338 (56.9)	322 (24.1)	356 (26.6)	660 (49.3)	
Rural/frontier CAC	1,012 (43.1)	331 (32.7)	272 (26.9)	409 (40.4)	

[a] Cases with unknown demographic characteristics were not included in chi-square analyses.

[b] Multiple reasons for CAC visit could be selected. Analyses compare cases with and without that reason selected.

**$p < 0.01$; *$p < 0.05$.

PTSD risk, and another quarter (26.7%) were at moderate PTSD risk. Suicidality and staff responses to suicide risk among 11–18 year-old youth are described by Shepard et al. (26). See Supplementary file 1 for information about suicidality within the full sample of 5–18 year-olds.

Table 1 shows differences in PTSD risk category by child characteristics. Children who identified as female or transgender male were more likely to be high PTSD risk than those who identified as male, and older children were more likely to be high PTSD risk than younger children. There were significant differences by race, although sample sizes for most groups were small, and no differences by ethnicity. Children seen for concerns about sexual abuse were more likely to be high PTSD risk compared to those without this concern, and those seen for concerns about physical abuse, witnessing domestic violence, or other reasons were less likely to be high PTSD risk than those without these concerns. There were significant differences between children seen in urban and rural CACs; more children seen in urban CACs were categorized as high PTSD risk and fewer children were low risk.

In a mixed linear model accounting for nesting of children within CACs, gender (female or transgender male; $b = 4.11$, $p < 0.001$) and older age ($b = 0.83$, $p < 0.001$) were associated with higher traumatic stress symptom scores; race/ethnicity and concern for sexual abuse were not significantly associated with symptom scores. Similarly, in multilevel logistic regressions predicting PTSD risk category, female/transgender male gender was associated with a greater likelihood of high PTSD risk (odds ratio = 2.30, $p < 0.001$), and older age was associated with greater likelihood of moderate (odds ratio = 1.06, $p < 0.001$) or high (odds ratio = 1.18, $p < 0.001$) PTSD risk. Race/ethnicity and concern for sexual abuse were not significantly associated with the likelihood of moderate or high PTSD risk. Full model results are presented in Supplementary file 2.

CAC staff decision making

Most children (77.4%) reported at least one elevated symptom (rating ≥ 3 on 0–4 scale), prompting CAC staff to deliver a brief intervention. Brief interventions were delivered to two-thirds (66.0%) of all children, including 85.2% of cases when a brief intervention was recommended by the CPM-PTS decision support tool. Types of brief interventions are shown in Table 2. The most frequent interventions were teaching diaphragmatic breathing, suggesting ways to improve caregiver-child communication, and teaching guided imagery. Approximately half of the children who received a brief intervention received more than one (32% received 2 interventions; 18% received 3 interventions).

Brief interventions were delivered to 22.3% of children at low risk for PTSD, 78.7% of those at moderate risk, and 85.3% of those at high risk. Children seen in rural CACs were less likely to receive an intervention than those in urban CACs (60% vs. 71%). In multilevel analyses, PTSD risk category was the strongest predictor of brief intervention delivery. Relative to those at low risk, children at moderate risk (odds ratio = 16.05, $p < 0.001$) and high risk (odds ratio = 25.40, $p < 0.001$) were substantially more likely to receive an intervention. There was considerable variation between CACs in brief intervention delivery (conditional ICC = 0.23). Child characteristics were not significantly associated with the likelihood of receiving an intervention (see Supplementary file 2).

Referral decisions are shown in Table 3. Among children with documented referral decisions (83%), most received a referral to evidence-based trauma treatment (53.1%) or community mental health services (39.0%). A small proportion were encouraged to follow up with their primary care provider (2.1%), and other options, such as following up with their existing mental health provider, were reported for the remainder (5.8%). Referrals to evidence-based trauma treatment were made for 45.5% of children at low risk for PTSD, 52.2% of those at moderate risk, and 58.4% of those at high risk. Children seen in rural CACs were less likely to receive a referral to evidence-based trauma treatment than those in urban CACs (44% vs. 60%). In multilevel analyses, PTSD risk category was significantly associated with the likelihood of receiving a referral to evidence-based trauma treatment (moderate PTSD risk odds ratio = 1.58, $p < 0.01$; high PTSD risk odds ratio = 2.24, $p < 0.001$). Again, there was considerable variation between CACs in referral decisions (conditional ICC = 0.24). Child characteristics were not significantly associated with referral decisions (see Supplementary file 2).

Discussion

We examined traumatic stress symptoms and risk for PTSD within a large sample of children seen in Children's Advocacy Centers for concerns about child maltreatment. Close to half (45.5%) of these children had a high probability of a PTSD diagnosis at the time of the CAC encounter; another quarter described moderate levels of traumatic stress symptoms that are likely to benefit from specific, trauma-focused interventions. Most children (77.4%) reported one or more elevated traumatic stress symptoms. Older children, those who identified as female or transgender male, and those who were visiting the CAC because of concerns about sexual abuse were at elevated risk. Children seen in urban CACs were more likely to be at high risk for PTSD than children seen in rural CACs, perhaps because of variations

TABLE 2 Brief intervention delivery by symptom cluster.

	Full sample (*N*=2,350)	Elevated sleep symptom(s) (*n*=1,012; 43%)	Elevated intrusion/ reactivity symptom(s) (*n*=1,493; 64%)	Elevated avoidance/ negative mood symptom(s) (*n*=1,534; 65%)
Any brief intervention	1,551 (66.0)	877 (86.7)	1,274 (85.3)	1,315 (85.7)
Diaphragmatic breathing	729 (31.0)	502 (49.6)	676 (45.3)	609 (39.7)
Guided imagery	500 (21.3)	430 (42.5)	456 (30.5)	447 (29.1)
Mindfulness	265 (11.3)	187 (18.5)	249 (16.7)	230 (15.0)
Caregiver-child communication	572 (24.3)	310 (30.6)	436 (29.2)	572 (37.3)
Behavioral activation	124 (5.3)	70 (6.9)	103 (6.9)	124 (8.1)
Caregiver-child special time	84 (3.6)	45 (4.4)	72 (4.8)	84 (5.5)
Other/unspecified	324 (13.8)	153 (15.1)	200 (13.4)	193 (12.6)

Categories are not exclusive. Children could have more than one domain with elevated symptoms and/or receive more than one brief intervention. Shaded areas indicate brief interventions recommended for each symptom cluster.

TABLE 3 Referral decisions and child PTSD risk category (N=1,950).

	N (%)				χ^2
	Follow up with PCP	Community MH	Evidence-based trauma treatment	Other	
Full sample	41 (2.1)	760 (39.0)	1,036 (53.1)	113 (5.8)	
PTSD risk					79.08**
Low	23 (4.3)	219 (40.7)	245 (45.5)	51 (9.5)	
Moderate	12 (2.3)	221 (41.5)	278 (52.2)	22 (4.1)	
High	6 (0.7)	320 (36.4)	513 (58.4)	40 (4.6)	

**$p < 0.01$.

in service criteria and the types of cases seen in different CACs. For example, child welfare or law enforcement in urban areas with higher caseloads may have more stringent eligibility criteria for CAC services, resulting in urban CACs serving children who experience more severe maltreatment.

CAC staff delivered brief interventions to most children and were especially likely to do so for children at moderate or high risk for PTSD based on screening results. Approximately half of the children who received a brief intervention received more than one intervention. Children seen in rural CACs were less likely to receive a brief intervention than those in urban CACs, and there was substantial variation in intervention delivery between CACs. This variation is likely because of differences between workflows and staff at different CACs. Our prior work has identified staff self-efficacy as a key determinant of CPM-PTS use. Ongoing training and technical assistance is needed to support CPM-PTS use and may be particularly important for supporting staff without clinical training in their delivery of brief interventions (18, 26). Overall, use of brief interventions was high, and our findings suggest that staff without clinical training can screen children for mental health needs and deliver brief interventions successfully.

Most children received a referral to evidence-based trauma treatment or community mental health services, and the likelihood of a referral to evidence-based trauma treatment was greater for those at greater risk for PTSD. Encouragingly, child gender, age, and race/ethnicity were not associated with the likelihood of a referral. Providing staff and families with specific data to drive decision-making may reduce biases in the referral process. Although referral rates were generally high, not all children at high risk for PTSD received a referral to treatment. It is likely that referral decisions are at least in part reflective of families' preferences. Staff may not make a referral if a family is already receiving services or is not interested in treatment. Use of evidence-based engagement strategies and follow-up with families may help increase treatment engagement (30).

Mental health workforce shortages and limited availability of specialty mental health services are also likely to affect referral decisions. A substantial proportion of children at elevated PTSD risk were referred to general community mental health services rather than evidence-based trauma treatment, and children seen in rural CACs were less likely to be referred to evidence-based trauma treatment than children seen in urban CACs. These findings likely reflect lack of access to therapists trained specifically in evidence-based trauma treatments, especially in rural areas. Although most CACs report access to evidence-based treatments such as Trauma-Focused Cognitive-Behavioral Therapy (15), they may not have sufficient capacity to serve all the children who could benefit. Ongoing efforts to recruit, train, and retain therapists are needed, especially in rural and frontier areas.

Limitations

Screening rates varied considerably over time and between CACs, with an average screening rate of 53% across CACs during the 2-year period [see (18)]. There may be selection biases affecting who was offered and who completed the CPM-PTS. However, our prior work suggests that variability in screening was driven primarily by CAC workflows and staff self-efficacy, not child or family characteristics (18). In addition, the demographics of our sample are broadly comparable to the population of children served (31), and our analyses accounted for nesting of children within CACs.

CPM-PTS data were entered by different reporters, including caregivers, youth, and CAC staff. Because child age is confounded with reporter, it is possible that our finding of lower symptoms in younger children may be an artifact of using caregiver vs. self-report. Both the caregiver and self-report versions of the full UCLA PTSD Reaction Index and the self-report version of the Brief Form have strong psychometrics; however, the psychometric properties of the caregiver version of the Brief Form have not yet been examined (22, 32, 33). It should also be noted that the CPM-PTS only screens for traumatic stress symptoms and suicidality, and children served by CACs may have other mental health needs not identified in screening. Children classified as high risk for PTSD or experiencing significant impairment should receive a thorough diagnostic assessment by a mental health professional.

Child demographics were entered by staff, and we do not know whether staff asked children/caregivers about their identities and entered the demographic characteristics described or if staff entered responses based only on their perceptions of children's identities. Most children were identified as non-Hispanic white (75.7%), consistent with state demographics (77.2%) (34). The very limited racial/ethnic diversity and small numbers of children from most racial/ethnic groups limits our ability to identify meaningful differences in this sample. In addition, the 'reason for CAC visit' was entered by staff based on the allegation that brought the family to the center and does not indicate whether the allegation was confirmed. It also may not correspond with the descriptions of recent and/or remote traumatic experiences provided by caregivers and youth on the CPM-PTS screening tool.

Conclusion

CACs are well-positioned to identify children with mental health needs after allegations of sexual abuse and other maltreatment and provide trauma-informed care (17). In a statewide sample of children served by CACs, we found that almost half were experiencing substantial traumatic stress symptoms and likely to meet PTSD diagnostic criteria at the time of their initial CAC visit. CAC staff, most of whom did not have clinical training, were able to use the CPM-PTS, a structured protocol, to administer a screening tool, deliver brief interventions, and make appropriate referrals to mental health treatment. CACs are a critical setting for early identification of children experiencing traumatic stress symptoms following maltreatment and can facilitate timely access to evidence-based treatments.

Data availability statement

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

Ethics statement

The studies involving human participants were reviewed and approved by the University of Utah Institutional Review Board. Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study in accordance with the national legislation and the institutional requirements.

Author contributions

EM: conceptualization, formal analysis, writing—original draft. KC: conceptualization, methodology, formal analysis, data curation, writing—review and editing. KB: data curation, project administration, writing—review and editing. LS: project administration, writing—review and editing. BK: conceptualization, methodology, writing—review and editing, supervision, funding acquisition. All authors contributed to the article and approved the submitted version.

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Conflict of interest

KC reported that her institution receives financial compensation for expert witness testimony provided in cases of suspected child abuse for which she is subpoenaed to testify.

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

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2023.1202085/full#supplementary-material

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Exploring the feasibility of a mental health application (JoyPop^{TM}) for Indigenous youth

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Objective: The purpose of the current study was to explore the acceptability and feasibility of a resilience-focused mobile application, JoyPop^{TM}, for use with Indigenous youth.

Methods: A Haudenosaunee community-based research advisory committee co-developed the research project, in accordance with OCAP^{TM} principles. Adopting a mixed-method approach, five youths from an immersion school used the JoyPop^{TM} app for four consecutive weeks, as well as completed pre-test questions and weekly usage surveys. Most participants also completed post-test questions and a semi-structured interview. Based on a semi-structured interview protocol, youth responded to questions, and the most common themes were categorized to capture the experience of using the app.

Results: All youth reported a positive impression, used the app daily, found it easy to navigate, and indicated that they would recommend it to a friend. All features were uniformly positively endorsed. There were features that youth used most often (Deep Breathing, "SquareMoves" game, and Art features) and moderately (Rate My Mood, Journaling, and SleepEase). The social connection feature, Circle of Trust, was least utilized, with youth reporting a preference for in-person problem-solving. The drop-down menu of crisis helplines was not used. Youth recommended more gaming options. In terms of cultural resonance, appreciation for the app's use of water sounds in the SleepEase feature was expressed, as was cultural consistency with the "Good Mind" perspective. Recommendations included additional nature sounds, Indigenous design elements, the inclusion of Native language words, and traditional stories.

Discussion: The JoyPop^{TM} app was positively received by Six Nations youth, and ways to ensure its cultural appropriateness were identified. Moving forward, it is recommended that Indigenous designers create a new version with community design co-creation. Additional research with various groups of Indigenous youth is warranted as a pan-Indigenous approach is not recommended.

KEYWORDS

Six Nations, youth, mental health, resilience, mHealth

1. Introduction

Indigenous Peoples are a term used to describe the First Peoples of Turtle Island (including Canada and the United States), their ancestors, descendants, and future generations. Within a Canadian context, this term is used to describe three distinct populations: the First Nations, Métis, and Inuit. As of 2021, the population of Indigenous peoples in Canada was at 1.8 million, and it is one of the fastest growing populations, growing by 56.8% from 2006 to 2021 (1). Indigenous peoples are one of the youngest populations in Canada, with 44% of the population under the age of 25 (2). Given disparities in healthcare resources among remote, semi-rural, and rural reserve communities and urban communities, it is important to consider youth sub-populations, especially Indigenous reserve communities where basic living needs (e.g., clean running water) are challenging to obtain. While the greater proportion of First Nations youth live off-reserve in urban and other locales (56%), 44% of First Nations youth live on reserve (3).

The United Nations Convention on the Rights of the Child (UNCRC), accepted by most countries in the world, signals respect for and promotion of a child's right to physical and psychological wellness (4). The United Nations Declaration on the Rights of Indigenous Peoples (UNDRIP) proclaims Indigenous People's rights to maintain and strengthen their distinctive spiritual and cultural relationship with their traditionally owned or otherwise occupied lands, territories, waters, coastal seas, and other resources (5). Health inequities experienced by Indigenous communities stem from centuries of colonization by European settlers, which systematically disrupted traditional community structures, wellness models, cultural practices, and the movement across, utilization of, and stewardship of lands (6, 7). The displacement of Indigenous peoples from their ancestral lands, the disregard of treaties, and the loss of traditional languages have resulted in the persistence of adverse contexts, impacting Indigenous health at a mental, physical, social, spiritual, and cultural level (7–10). For those on reserve, there are intersectional health adversities that impact the optimization of mental health (e.g., resource infractions, ongoing residential school-related trauma, and community violence). In Canada, there has been a persistent over-representation of Indigenous children in the country's child welfare system that serves to limit cultural connectivity (11). In a systematic review, young Indigenous women had an elevated risk of mental health problems ($OR = 1.86$) and, in the context of maternal depression, small effect sizes were found for increased risk for offspring depression (12, 13). This is significant as parental cultural connectedness was found to be a protective factor for their child's mental health problem risk (12). Despite the fact that First Nations youth face mental health challenges, according to the 2017 Aboriginal Peoples Survey, 48.9% of Indigenous youth reported excellent or very good mental health (3, 12). These authors interpreted these findings in terms of enhanced resilience efforts among Indigenous communities.

Resilience is defined by the United Nations Children's Fund (UNICEF) as "the ability of children, households, communities, and systems to anticipate, prevent, withstand, manage, and overcome cumulative stresses and shocks in ways that advance the rights of every child, with special attention to the most vulnerable and disadvantaged children" (14). To support the development of resilience, it is recommended that programming be developed, and local systems and structures be supported in the planning and delivery of public services (14). In a scoping review of Indigenous youth resilience studies, the broad guiding definition from UNICEF was considered consistent with Indigenous perspectives of interconnectedness and interdependence (15). Considering this perspective, important resilience pathways include connecting to the natural world, learning from animals, inter-generational teaching, and mentoring relationships (15). Others have defined Indigenous resilience as a long healing journey to address multiple, historical, ongoing, and current traumas. Drawing upon decolonization processes, resilience factors for Indigenous youth include opportunities for empowerment, positive cultural identity, and a future orientation (15–17). Resilience programming (e.g., creative art expression and access to Indigenous language immersion education) supports the development of the capacity to respond to and overcome risks (18–20). Given the active elements in traditional ceremonies, including learning from the land, observation, and trial-and-error learning, Indigenous youth seem to gravitate toward educational empowerment through personal expression opportunities to build self-esteem, self-identity, and skillful, healthy relationality (21). For example, in an arts-based qualitative study, three themes emerged: (1) nature as a calming place, particularly in relation to water and bodies of water; (2) nature as a metaphor for resilience, in terms of growth and renewal; and (3) nature as hope, in terms of future opportunities and positive change (22). More research is needed that supports Indigenous youth voicing what resilience strategies are found to be personally helpful, as Indigenous youth may be less likely to seek formal health services because of stigma, discrimination, concerns around anonymity, distrust of governmental services, the availability of culturally relevant and trauma-informed approaches, challenges in identifying signs and symptoms of mental health problems, and difficulty with where or how to seek help (9, 16, 23).

In a national survey of 675 Indigenous youth in the United States, it was found that 78% had regular access to a mobile phone (24). Hence, mobile applications have been identified as an accessible tool to support Indigenous youth resilience, especially in offering accessibility, affordability, and the capacity to address "in-the-moment" needs, without necessarily depending on Internet connectivity (9, 25). Research reveals that youth generally prioritize anonymity and privacy, ease of use ("look and feel"), and interactivity when engaging with mental health applications (26). The unique realities of Indigenous youth, wherein they navigate two worlds of traditional Indigenous culture and modern youth culture, have challenged researchers to identify adaptive frameworks. A two-eyed seeing approach has been popularized to accommodate both Indigenous and Western knowledge traditions (27, 28). Two-eyed seeing is rooted in Indigenous knowledge systems that emphasize non-hierarchical sharing and facilitate a "dialogue" between Western and Indigenous ways of knowing (27, 28). Seeking to harmonize these two distinct viewpoints, this approach recognizes and values the unique strengths and insights that each perspective brings (27, 28).

The JoyPop^{TM} app (see Figure 1) is an iOS English and French language mobile application designed with the

The JoyPop App

Rate My Mood

Initially prompts users to rate their happiness by sliding a wave of color up or down to indicate their happiness level. If the happiness rating is lower than 50%, the user is prompted to rate how sad, angry, or "meh" they are feeling using the same technique.

Journal

Allows the user to complete a journal entry by entering their free-flowing thoughts and emojis, or by responding to a resilience-oriented writing prompt at the top of the screen. Users can save their journal entries to the Calendar feature.

Breathing Exercises

Opens to a diagram of the body, with best-practice tips to prepare for relaxation. The user is then prompted to choose between completing a balanced breathing exercise or a relaxation breathing exercise. Users are then guided through the breathing exercise with text instructions and an animated diagram.

Art

Allows the user to doodle in color, swiping their finger across the screen as the paintbrush.

SquareMoves

A game in which multi-shaped blocks fall from the top of the screen and the user taps on the shapes to rotate them or swipe them across the screen to move them as they fall to the bottom, with the objective of forming a solid line at the bottom of the screen.

Call For Help

Allows the user to select a 24-hour helpline to call if they are experiencing distress while using the app. The user is provided with culturally-specific hotlines (eg, an indigenous-specific crisis line, LGBTQ helpline) to choose from.

Circle of Trust

Allows the user to input up to six safe, social contacts (ie, by entering their name and phone number) to call if they want to talk or are in need of support. The user can label the contact as a friend, family member, professional, or elder/mentor.

Calendar

Allows the user to reflect on previously saved journal entries by date.

FIGURE 1
Features of the JoyPop^{TM} app.

intention of promoting and enhancing resilience among youth (youthresilience.net). While it was initially produced as a potential tool for at-risk youth, particularly those with trauma backgrounds, its focus on positive emotions and actions has the potential to support a broad spectrum of users. App development was supported by professional app developers, researchers, computer sciences and health sciences university students, the Toronto Police Services high school youth leadership group (i.e., Teens Ending Abusive Relationships, T.E.A.R), and practitioners within child welfare, which is described elsewhere (25, 29). The key proposed mechanism of the app is the facilitation of emotion regulation (ER). ER skills (e.g., awareness, self-reflection, labeling, modulation, expression, and management of positive and negative emotions) develop substantially across adolescence with the advent of greater autonomy strivings, more abstract thinking, normative relational challenges (e.g., friendships, romantic partnerships, work relationships, greater autonomy strivings), and changing neurocircuitry (e.g., increased prefrontal "control" and problem-solving) (30, 31). ER reflects the developmental capacity to integrate feelings and thoughts about emotions with actions that are driven by habit, reactivity, or impulsiveness (or reflexivity) or impacted by conscious, effortful responding (or reflectivity). Disrupted ER is central to the experience of anxiety and depression, in terms of managing negative emotions and accessing positive emotions (32). The JoyPop^{TM} app was built on the assumption that resilience is a skill set that can be increased through targeting ER components, specifically skills to dampen physiological reactivity, high negative emotionality and low positive emotionality, and high cognitive load (17, 25, 29). Positivity is enhanced with a focus on positive emotionality (e.g., happiness) and personal agency (e.g., motivational messaging and encouragement for users to attend activities if their mood is rated low) (17, 25). Specifically, ER is addressed: (1) physiologically with diaphragmatic breathing (33) and quality sleep (25); (2) affectively with mood awareness and monitoring (34) and unstructured art expression (35); (3) socially, with close relationship connecting and distress line support (25); and (4) cognitively with journaling with resilience-oriented prompts (36) and focused attention gaming techniques (37).

In evaluating the JoyPop^{TM} app with adolescents who had transitioned to their first year of university, JoyPop^{TM} improved both ER and depression scores, with youth readily adopting the app into a daily or near-daily routine over the study month (29). A qualitative research study also found that the JoyPop^{TM} app was readily adopted into users' daily routines, being seen to positively start and end their day. Users appreciated the opportunities to be expressive with how they were feeling in a variety of modalities. Similar themes were found in a qualitative study by Kim et al. (38), who gathered the perspectives of adults from the Six Nations community to assess the appropriateness of JoyPop^{TM} app for Indigenous youth. These three JoyPop^{TM} studies, however, did not target Indigenous youth to provide an exploration of how the app would be received. The purpose of the current study was to explore the accessibility and feasibility of the JoyPop^{TM} app with Indigenous youth living on reserve. Specifically, we queried the opinions of the app and its features among Haudenosaunee youth of the Six Nations of the Grand River, as well as the youths' reported usage, to explore accessibility and feasibility issues.

2. Methods

To provide a methodology overview, this study is a qualitative case study based on phenomenological research. The case study is an appropriate research method for the exploration of a phenomenon within a particular context, including natural setting that is culturally relevant; the case study benefits from the undertaking of the exploration through a variety of data sources (39). Taken together, a potential process and framework may emerge that are useful for further research.

This research was conducted at Canada's largest First Nations reserve, Six Nations of the Grand River (SN), in the Great Lakes region (Ontario, Canada). SN is home to six distinct nations—Cayuga, Mohawk, Seneca, Onondaga, Oneida, and Tuscarora—and is a semi-rural community governed by the Haudenosaunee Confederacy Council and the Six Nations Elected Band Council. With an on-reserve population of 12,849 individuals, and a cumulative band membership of 28,019, this community has engaged in health science research for decades (40). Philosophical paradigms for Indigenous research necessitate the underpinning of Indigenous Nation-specific laws. Embedded within the fabric of this community is Haudenosaunee law, an embodiment of the alliance between nations under the Great Law of Peace, or *Kayannerenkó: wa* (41). One important embodiment of Haudenosaunee values and perspectives is the "Good Mind" and its correlates of acting in a "good way" and in thanksgiving to all beings, such that all come together as one (42). The Good Mind is considered that which keeps a person balanced and in harmony. It reflects an individual point of accountability to oneself, one's clan, one's community, one's nation, and, ultimately, to the natural environment (Mother Earth). The Good Mind is a physical, psychological, and spiritual journey that includes a reflective awareness of thoughts and intentions, and a way of being that is expressed through self-compassion and compassion for other beings (43). These contextual elements highlight the requirement for Western researchers to be invited into the territory (as is the custom among Nations), and the essential learning about culture to appropriately shape the research process, results, and interpretation.

2.1. Researcher reflexivity and positionality

All Six Nations authors live and/or work on the reserve and represent the Mohawk, Cayuga, Seneca, and Oneida nations. All have also attended Western universities and/or colleges and are deeply embedded in traditional culture, ceremonies, and language. Indigenous committee members engaged with Western researchers within regular meetings and on an as-needed basis, as well as providing a listing of culture-specific readings. Particularly among Western academics conducting Indigenous-focused research, reflexivity and positionality are key to validating qualitative research findings and promoting trust in co-creation. The first author, AA, is a non-Indigenous student who completed a bachelor's degree in Health Sciences from McMaster University. She now benefits from financial aid resources to attend the University of Toronto. Author DM is a non-Indigenous, South Asian student from McMaster's Department of Biochemistry. Author KB holds both settler and Indigenous ancestry from the Chippewas of Rama First Nation. Her academic background in Health and Aging from McMaster University focuses on culturally relevant youth programming in Indigenous communities. Authors AA, DM, and KB completed a nationally promoted course from the University of Alberta to better understand Indigenous culture and attended group meetings with the Six Nations Youth Mental

Wellness Committee. Author CW is a white, non-Indigenous, European ancestry settler, who benefited from Western education in obtaining a Ph.D. (Clinical Psychology). She has worked with Indigenous welfare leaders since the early 2000s and has engaged with Indigenous community members through various publicly funded research projects, including on trauma and resilience among Mi'kmaq youth living on reserve in Nova Scotia, Canada. She worked on water-related research with the Six Nations prior to forming the Six Nations of the Grand River Youth Mental Wellness Committee to specifically guide adolescent research. She attended an online visit through Canada's first residential school in Mohawk territory. Throughout the life of the current project, she was mentored by an Elder from the Six Nations in 1:1 meetings. Reflective practice and positionality discussions occurred in the context of the committee meetings and larger dissemination events.

2.2. Participants

The Six Nations of the Grand River Haudenosaunee community identified an appropriate site for youth research, which was a private, on-reserve Mohawk and Cayuga language immersion school that ranged from kindergarten to grade 12. The school's vice-principal and counselor were available to youth, parents, and teachers to respond to inquiries about the research and provide information in the Indigenous language, as needed. The study was introduced to youth with 15-min presentations delivered by a Six Nations research assistant to each grade-eligible class (i.e., grades 7 to 9). Presentations highlighted the features of the JoyPop^{TM} app, the purpose of the study, and the study's timeline. An information sheet and recruitment poster were distributed by the school counselor to all eligible students via their school email. Interested youth provided consent as per guidelines detailed in the ethics portion of this article, and consent was stored separately from data on a password-protected institutional drive.

With the goal of understanding the experience of using the app, a phenomenology approach was undertaken. Generally, this approach requires a sample size between 3 and 25 participants (44). As class sizes were small, the study could have maximally recruited 25 youth. This study recruited a group of five youth participants (2 males; 3 females), from 14 to 16 years old, with one youth at 11 years old. Four youth reported their languages as Mohawk, two as Cayuga, and one youth did not provide a response. All youths participated in the pre-questionnaire and weekly usage questionnaires, and four youths elected to complete the post-test questionnaire and complete an interview.

2.3. Data collection

Participants were assigned a study ID code to use in any data collection and asked to complete a pre-test survey. They were then given access to download the JoyPop^{TM} app directly onto school-provided iPads and were instructed to use the JoyPop^{TM} app daily for four consecutive weeks, simultaneously completing weekly surveys. The app itself has onboarding instructions, and there was a 4-min instructional video on app features and use of the app (https://www.youtube.com/watch?v=3LzYTdjZPnU&t=64s) which youth were directed to watch. Following the 4 weeks, youth were asked to complete a post-test survey and offered the opportunity to participate in an individual interview to share their experience with the JoyPop^{TM} app.

2.4. Research tools

Prior to using the app, a pre-test survey was distributed, which consisted of questions ranging from open-ended demographic-related inquiries to forced-choice questions (yes, no, or unsure) about lifestyle and closed scales using the 5-point Likert Scale assessing baseline ratings on perceived mental wellness. These were developed in conjunction with the SN Youth Mental Wellness Committee. Weekly surveys were also distributed to track the self-reported frequency of daily app usage; youth were asked how many days within the week they accessed the JoyPop^{TM} app and how frequently per day. A post-test survey was used to query youths' opinions about the app. Questions were similar to the pre-test survey, with additional forced-choice questions about the app's helpfulness and overall impressions. For those who offered to share more feedback, interviews were delivered via Zoom, a video-conferencing tool, using audio only, by a research team member. Each interview was 1 h in length and followed a semi-structured format, with two general topic questions (e.g., Would you please describe what mental wellness means to you?; What does a "good mind" (Haudenosaunee concept) mean to you?). Following these, the interview completed the previously established JoyPop^{TM} interview protocol outlined in Kim et al. (38) wherein the 4-min JoyPop^{TM} introductory video was played and participants were asked for their thoughts on the app's features, first shown in complete and then re-run with pre-specified pauses at the end of each feature description (e.g., After the Breathing Feature description on the video, the video was paused, and youth were asked, "What did you think about the Breathing feature?"). Open-ended questions guided the conversation, and youth were encouraged to bring up new topics as they wished (e.g., "Is there anything else you would like to discuss that we did not ask you about?"), and thoughts about the app (e.g., "Outside of this research study, would you use JoyPop^{TM}, why or why not?"). Following the interview, a summary of the discussion points was emailed to each youth for member-checking purposes to ensure the accuracy of the data gathered and description. This process facilitated relinquishing the expert "researcher" role to listen for youth voices (e.g., "I" statements) and reflect on how the voices fit together in terms of similar opinions and where there were unique or divergent opinions, ordering quote segments in terms of similarity based on frequency across youth participants.

2.5. Data analysis

Each consultation was transcribed by two research assistants (AA and CM). In the adult study, a coding framework was developed to facilitate content analysis, in which descriptive labels, or codes, are used to capture and summarize themes discussed by the consultants (38). Additional codes were added to ensure youth's questions and perspectives were represented. Content analysis was the selected analysis method as it allows researchers to identify cultural patterns, themes, trends, and other features from verbal, visual, or written data (45). Using the framework, each transcript underwent a separate coding process in a double-blind fashion. Inter-rater reliability scores were calculated to ensure consistency with the coding process, with inter-rater reliability exceeding 85% (46, 47).

2.6. Ethics

Several factors were in place prior to conducting this JoyPop^{TM} study with Indigenous youth living on reserve. These included first being invited by the Six Nations of the Grand River Haudenosaunee community to conduct research on JoyPop^{TM}, where the app's face validity was seen as a good fit with community emphasis on youth resilience. Our community-based trainee (KCDM) co-presented this research proposal with CW at a Canadian Institutes of Health Research Gender and Wellness Development Fair, which allowed for broader discussion with national Indigenous representatives and youth, on the rationale and structure of the JoyPop^{TM} app and its research. This facilitated the formation of a Western (CW) and Six Nations (KCDM and DMH) research co-leadership to seek funding to develop an approach to app development. The school principal joined the research team as co-applicant and provided support for schools staff to act as project consultants and facilitators. The early partnering with school staff provided important directives in terms of a cautious approach inviting youth to participate in research, using school-based communication channels to first approach parents and guardians, then present at teacher meetings, and finally, conduct in-class presentations to youth. A collaborative plan supported a budget for remuneration (i.e., gift cards directly to youth, and iPads to the school), as well as establishing dedicated school staff as part of a guiding committee.

Having secured funding, a community-based Six Nations Youth Mental Wellness Committee was established to meet regularly. The Committee member remuneration was at \$50 per 1 h meeting. Any additional member-specific meetings also occurred at this remuneration level. This committee was composed of the Indigenous Six Nations grantees and broadened to a diverse membership of health services, education, language, and culture expert adults with community leadership roles. Specifically, this committee recommended several procedural approaches: (1) first conducting a qualitative interview study of adult community members who were involved in youth services to gather information about their views about the app, their thoughts about its relevance to youth, and thoughts about cultural adaptations to the app; (2) utilizing the immersion school as a test case site and adhering to the typical school communication procedures (e.g., secure parental section on school's website); (3) maximizing privacy so that data are stored with an ID number and that no app back-end data would be collected; (4) engaging committee members in-depth about the interpretation of findings within a cultural lens; and (5) developing a post-study action plan that included storage of data within the Six Nations site.

School guidelines and committee feedback steered the recruitment process. Interested youth below the age of 16 were encouraged to review a consent form with a parent or guardian, and active guardian consent was required. For those age 16 or above who were interested, the youth was deemed as able to provide their own consent. Youth were remunerated for participation in the questionnaire and interview components via online gift cards (\$35 total).

This study followed the OCAP^{TM} (Ownership, Control, Access, and Possession) principle framework, which guides the collection, protection, use, and sharing of First Nations data and ensures caution is taken with youth-specific research (48). Ethics approval was received by Haudenosaunee Confederacy Council, Six Nations Elected Band Council's Research Ethics Committee, and McMaster Research Ethics Boards for consultation interviews and survey data collection (MREB #3728; HIREB #12572). Our advisory committee presented in Mohawk to the longhouse meeting of the Confederacy leadership, and our community-based trainee (DM) co-presented this research idea at a Canadian Institutes of Health Research Gender and Wellness Development Fair, with Indigenous co-leadership, which allowed for focus group discussion more broadly on the rationale and structure of the JoyPop^{TM} app and its research.

3. Results

3.1. Pre-test, weekly surveys, and post-test

When asked to rate their health on a 5-point Likert scale, youth rated their physical health as good ("good" or "very good") and had mixed ratings on their mental health (from "fair" to "very good"). In terms of digital technology, all youth reported using social media and communication apps daily over the past 12 months. In terms of health or wellness apps, three of the five youth reported having used them in the past.

Youths were sent an email at the end of the school week to report which days of the week the JoyPop^{TM} app was used. Youth reported that they minimally used the app once a day and maximally three times a day across the 5 days of the school week.

After 4 weeks of using the JoyPop^{TM} app, a post-test was done to determine final impressions, in which four out of five consultants participated. In response to a forced-choice question (yes, no, or unsure), all youth reported that they would recommend the JoyPop^{TM} app to a friend. In response to a question on which feature they used most often, all youth identified using the Tetris-like gaming feature, SquareMoves, most often. Youth reported using all features except the dropdown of crisis helplines and the Circle of Trust feature. In terms of an open-ended question on barriers to using the app, three out of four youths reported that forgetting to use the app interfered with the frequency of their use of the app. It should be noted that the app uses no "push notifications" to prompt use.

3.2. Qualitative interviews: main themes

As small number of youth contributed to this study, we do not use any identifiers for quotations from youth. In each section,

TABLE 1 Mental wellness: sub-codes and quote examples.

Parent code	Sub-code	Example of youth's quotes
Mental wellness	Positivity/ happiness	A good mind is someone who has like just good thoughts positive thoughts all the time and uses kind words all the time just being positive.
	Difficulty understanding emotions	Now that I'm this age I'm more bringing up how I feel and how I deal with my emotions which is why it is really hard for me to describe when I'm happy or when I'm happy and how to express it in a healthy way either so right now I think I'm learning how to express like being happy and being just emotions cause usually I just shove everything down.
	Acts of kindness	"And what are some things that you do to make sure you have a good mind in your day-to-day life?" (interviewer) "Helping people like with their needs or whatever. And like just being kind to your family members and taking care of stuff, and helping whoever."
	Personal hobbies	Let's see. I'm going to say reading again because it puts me in an exciting mood and I'm happy to read.
	Positive body language	"What are some ways that you express your happy emotions?" (interviewer) "Probably like smile a lot and laugh and if I'm really happy, I jump around, so stuff like that."
Cultural	Relationships	You could do anything and have a good mind about it you know… Talking to your elders, respect everyone you know… um… I was taught this by an elder at the long house.
	Nature	I like to spend time outside, you know… Be with nature and mother earth just to see all she created for us. Or uh what Creator created for us. All this beautiful stuff we see with your own eyes... he did that all for us you know.

examples are from different youth. The SleepEase feature, which provides support for getting into a sleep mode, was reported as being used by only one participant and, as such, is not presented below. While the Circle of Trust feature was not used by any youth, it is discussed below because it was the most valued feature *a priori* by advisory committee members.

3.2.1. Mental wellness

Sub-codes and examples are found in Table 1. Common codes, or themes, identified when discussing mental wellness included concepts of positivity/happiness, positive body language, personal hobbies, acts of kindness, and difficulty understanding emotions. All youth described positivity and happiness as important contributors to their mental wellness, and that the app could provide emotional support. Culturally, the youth recognized that a Good Mind had positive emotionality as a key feature and that the app had a positive emotional orientation. Most youths elaborated upon several examples of positive body language (e.g., being active, greeting others, smiling, laughing, movement) that represented mental wellness. Specifically, laughter was noted as a sign of spreading "good energy" around to others. Most youth indicated that personal hobbies were a part of maintaining their mental wellness and making them feel happy. These hobbies ranged from reading to dancing to sports (It is noted that youth are engaged with traditional dance lessons, with an annual Pow Wow held on reserve; furthermore, lacrosse teams and leagues are prominent). One youth noted that "I'm going to say reading, again, because it puts me in an exciting mood, and I'm happy to read." (It should be noted that the app allowed for the saving of journaling entries within a calendar feature so that youth could revisit and read what they had written).

Additionally, all youth indicated that acts of kindness were important to their concept of Good Mind as their own behavior carries responsibility toward community members. Youth expressed that these acts of kindness were rooted in caring for oneself and others. For example, three youth provided the following:

> Helping people, like, with their needs or whatever. And like just being kind to your family members, and taking care of stuff, and helping whoever.
>
> Treat people the way you want to be treated, I guess.
>
> A Good Mind is someone who has, like, just good thoughts, positive thoughts all the time, and uses kind words all the time… yeah, just being positive.

Furthermore, all youth expressed difficulty understanding their emotions at times, especially when mixed emotions are involved. Youths reflected that they are still learning to express and understand their own emotions. For example:

> Now that I'm this age, I'm more bringing up how I feel and how I deal with my emotions, which is why it is really hard for me to describe when I'm happy or how to express it in a healthy way either. So right now, I think I'm learning how to express, like, being happy and being just emotional, "cause usually I just shove everything down".

3.2.2. Cultural importance of relationships and nature

When discussing culture, themes identified by all youth included relationships and nature. All the youth mentioned the importance of relationships, and most said that their relationships with family and friends were important for maintaining their happiness and mental wellness.

> OK, so what's the visual or thought that brings a smile to your face? Probably my little cousins. And lacrosse. Family dinner.
>
> You could do anything and have a good mind about it, you know… Talking to your Elders, respect everyone you know… um… I was taught this by an Elder at the Longhouse.

All the youth identified that Nature is important to a Good Mind, bringing positivity through relationality (i.e., connected and equal to all living things and having gratitude for creation). Nature-based visuals and sounds, such as the sound of rain, thunder, crickets, and flowing waters, were found to be very positive (It should be noted that there are specific teachings attached to natural occurrences, such as thunder representing grandfather beings). Two youth examples are as follows:

> I go into these stages where all I think about is the sound of water, the sound of thunder, the sound of rain and that puts me at peace.
>
> I like to spend time outside, you know… Be with nature and Mother Earth just to see all she created for us. Or, uh, what Creator created for us. All this beautiful stuff we see with your own eyes... he did that all for us, you know.

3.2.3. Mobile application usage

Mobile Application Usage refers to the youths' general usage of any type of mobile application. The most common themes were related to games, social media, and other applications. Most youth stated that they used games to pass time and enjoyed the reward aspects (e.g., collecting coins and customizing avatars). Most youth listed applications for specific information, entertainment, and communication. Some examples are as follows:

> For particular reasons, like the weather and stuff…
>
> *Subway Surfer* just passes time. I really like that app because we can just pass time or whatever.
>
> For Instagram, I always go on the explore page, and there are a bunch of quotes slash memes about what people are feeling, and I save them, and really like them because they really have a way of 'oh, I relate to them.' That's the only way I can express my feelings. So, I really keep those in my main part of why I use Instagram. Also I get book recommendations from there. Snapchat is more just like talking to my friends from like hours away, and talking to friends down here, it's more of like a communicating app for me.

3.2.4. Joypop^{TM}

Youth opinions about the features of the JoyPop^{TM} after having used it for 4 weeks were collected. Overall, the ease of using the app and the value of the different activities were consistently positive among all youth. Youth responded to open-ended questions about the app more generally, with comments provided below on aspects of the user experience (e.g., "the look and feel"). Comments about specific features are discussed, first examining features that youth reported using most frequently (Breathing, SquareMoves, Art), followed by those used moderately (Journaling, Rate My Mood), and those used rarely (Circle of Trust). Finally, youths provided their ideas on how to improve the app in terms of aligning with culture.

3.2.4.1. App layout and user experience

Youth were asked about the aesthetics and efficiency of the app layout and designs in terms of the icons used (e.g., color scheme, smiley face on the home screen; the home screen text "Happiness starts with you"; the icon designating features on the activities page, thumbs up visuals with positive affirmation comments, prompts to go to activities from mood ratings, the diagrams supporting body relaxation cues, and sleep hygiene tips that appeared before using the breathing and sleep features, respectively). Overall, youth regarded all these positively, and none noted negative reactions. The icons and symbols were familiar to youth, and the movement across screens was considered easy to navigate, in an intuitive way. Youth did not feel they required additional instruction to use the app (beyond the Circle of Trust, as noted below), and it was an advantage to be able to use it without the need for an Internet connection. Youth liked that the app was simple, without too many features or words, and that the language and words used in the app were easy to understand. Youth comments are below.

> What do you think about the colors that are used in the app?' Well for me those are my favorite colors.
>
> I think the [icons] are pretty cool, and you can like understanding what the app part is of it. So, like, for example, the art has a little art symbol on it so you know that's the art, and like the breathing has the face breathing and stuff so like, yeah.
>
> Um, well, it's just a really fun app to use and, like, it makes all my boredom go away. Like, if I'm at my brother's lacrosse game on the rez, I can just pop that on and use it.

3.2.4.2. Breathing feature

All youth found the breathing animation to be a novel activity and appreciated that it uses steps to guide the breathing cycles and that there was more than one breathing activity. No youth reported any negative reactions; the most common sub-code found in the interviews was *Breathing Feature: Positive Reaction.* All youth reflected that, while they had been aware of breathing and how emotionality affects breathing, they had not done specific breathing exercises before. Youth noticed an immediate positive impact from focusing on their breathing. Youth found the breathing exercise options useful for relaxation and for regaining control of their breathing during stressful feelings or events.

> I thought [the breathing feature] was good. I know I hear about that stuff all the time, about people breathing . . . it relaxes them. Sort of like meditating a little bit. Yeah, it's good.
>
> The breathing exercises, I thought, were really good, because there are a lot of people, including myself, that have a hard time, when they're going through a hard time, can't get their breathing under control. So, pulling that up [on the screen] can really help. So I think that's good.
>
> Oh, um, I thought [the breathing feature] was great. It helped me clear my thoughts, you know. I mean you just relax, 'cause I was having a rough day and then it just brightened my mood, so.

3.2.4.3. SquareMoves feature

The SquareMoves Feature refers to the Tetris-like game in the app where youth tap the screen to rotate blocks and fit blocks together. When a line is completed, a graphic reward occurs, and the youth score is displayed. Youth found the feature of a game in the app to be very positive in terms of visuals, ease of use, appropriate challenge, and the experience of "fun". No youth disliked this game. Youth found that playing this game was helpful to distract from negative emotions and engage more in positive emotions by tackling the challenges (i.e., controlling the speed of the block drops) and playing in a familiar format. The most common sub-code that was found was *SquareMoves: Positive Reaction*. Youth had a positive reaction to the SquareMoves activity, which was similar to common arcade games that they had played in the past. Youth found that playing it over time could lead to relaxation.

> Oh, um, it was just a relaxing game, you know. [...] I just spent hours playing that game, I just zoned out.
>
> Yeah, I liked it.

3.2.4.4. Art feature

The art feature refers to the integrated drawing space on the app. The most common sub-code that was identified was *Art Feature: Positive Reaction*. All youth had a positive reaction to the art feature in the app, and no youth had a negative reaction. Youth mentioned that engaging in this feature was helpful in managing and expressing emotions. There were several different ways youth reported using this feature. Specific strategies included doodling, writing out emotional words, and drawing symbols and scenes to express their emotions. A few youth noted:

> I really liked it. I can, like, doodle all the stuff. Yeah, I just really like doodling and the art option.
>
> I think it's good. I'm not really good at art, but I think it's good for people that maybe they do art to get past their feelings, or if they're upset, they do art to calm down their nerves, or something like that. But I actually like art and stuff.
>
> I would just go on there and write one word about how I was feeling, and that was it. So if I was feeling sad that day I would go on there and write a big capital letter 'SAD,' and that's what I would do.

3.2.4.5. Rate my mood feature

The rate my mood feature on the JoyPop^{TM} app allows users to become more aware of their mood, starting with a query on "happiness" and then giving different negative mood options (sad and angry) and a non-committal mood ("meh"). Youth found the slide activity (sliding a wave visual up and down to reflect the mood intensity) was interesting to do. Youth liked that it was not overly complicated with emotions. They appreciated rating their different emotions across the day and reported that they could detect patterns and changes in emotions. All youth responded positively to this feature, and no youth identified negative reactions. The most common sub-code that was identified was *Mood: Positive Reaction*. Most youth enjoyed being able to name and investigate their mood and also acknowledged that their mood changed throughout the day.

> Uh, I think it's good. I think it's good that, um,.... when you say that you're sad, and you can. It actually goes into detail, and you could tell it, you're actually like mad. Or how you said you're having a "meh" day or whatever. Yeah, I think it's good.
>
> I thought that was a good one too because even myself would go on there, and it helps you feel, um, heard about what you're feeling that day because you want someone to ask you, how you're feeling today. When you don't have that, you can just go on there and rate it, and it tells you if you're in a bad mood, it tells you are you feeling okay, what's wrong. So, I think it's good for someone who needs to feel heard.
>
> Yeah. If I have a really weird day, like a "meh" day, which I have almost like, every day, at the start [of the day]. But it turns into a happy day.

3.2.4.6. Journaling feature

The journaling feature provides the user space to write down their thoughts, and seems to be a familiar activity for youth. Youth could utilize their iOS microphone options, add emojis from their phone template, or type in their text. Resilience prompts are available to write about, or youth could use the space for expressive or reflective practice. Youth reported it as a useful activity to organize their thoughts and be expressive about their emotions. All youth reported this feature as positive, and no negative reactions were identified. The most common sub-code that was found was *Journaling: Positive Reaction*. Committee members commented on this finding in terms of the value of this feature in providing a private place for self-expression. Examples from youth are:

> Journal, it's good, now, you can go in there every day. You can write how you're feeling, or about how your day went. Stuff like that. It's not going to be shared with nobody, though. People write in there and stuff like that. Yeah, it's a good thing that people can write down their feelings, and how their day went and stuff.
>
> It helped me get my thoughts out, using that. Just trying to... cause I have a lot to think about, and, yeah. It was... sometimes you gotta write what you're thinking down.
>
> The journaling... I think it's good too. Personally, I didn't use it that much, because I have my own journaling thing I do for myself, but I think that it could be good for others.

3.2.4.7. Circle of trust feature and helplines

The Circle of Trust feature allows users to input contacts of people that they trust in case of a crisis or need for support. Although this feature was highly valued as culturally consistent by committee members, youth did not use it. Youth reported not being familiar with reaching out for help outside of in-person options or ever using helplines. There was a feeling of distrust toward connecting with people who were not family, friends, or very well-known to them. The most common sub-code that was identified was *Circle of Trust: Negative Reaction*. Most youth had a negative reaction to this feature in terms of being unsure about how the information would be stored or used given the direct connection to others' contact numbers. In short, youth identified needing more information as to how their information would be protected in the app. Youth comments are as follows:

> I personally, again, that one isn't for me. I can't. It's really like my trust has to stay within this area, it can't be out like that.
>
> I didn't really know what to type there so yeah I just didn't know what to type.
>
> When it comes to mental health for me, I have to full out talk to people. Because I'm not good with technology.

3.2.4.8. Adaptations

Adaptations refer to suggestions to improve the app, with a particular focus on how to make the app more culturally relevant. Notably, the significance of cultural relevance cannot be overstated as it has been shown to have a substantial impact on the effectiveness of tools and interventions aimed at engaging Indigenous youth. Studies have demonstrated that culturally relevant interventions have found success in resonating with their intended audience and achieving desired outcomes (49, 50).

All youth had positive feedback related to the games in the app. Youth strongly favored games, which they identified as a positive way to improve feelings of happiness, and encouraged that more games be added and that game creation be a feature for Indigenous youth: "There could be more games too. As a youth, especially boys, they are more connected to games. With the sounds of it (i.e., using sounds), I feel like there can be more of those too." Most youth had Indigenous-specific feedback to improve the app. One of the suggestions was to include an Indigenous dice game played with others called Gayendowa:neh. Another suggestion was to include Indigenous language within the app and add more Native symbols as icons, such as a wampum belt, clan animals, and feathers.

> I don't really know… Just maybe add some kind of Native stuff. Like feathers or maybe clans. That would be a good thing to put in there. Put your clan and your nation in there. Maybe the animals. Like the ones that are on the earth, and the ones that are in the sky. Umm, yeah, just more Native stuff.

Most youth identified stories as an important aspect of Indigenous culture. Traditional stories were identified as something that could be integrated into the app as an activity that users could participate in, and many found these stories to be interesting and relaxing. Additionally, all youth expressed the desire to incorporate Native languages into the app and provided suggestions on how this could be achieved, such as the ability to change the language of the whole app to align with their preferred language.

> ...you know, I think stories could be good for the app. Putting little stories in there for people could be interesting. Just to get people's mind off things.
>
> ...for Haudenosaunee, I think there can be words added to it. Just little tiny words, expressing being happy or mad, on there, in our language.
>
> Maybe like a setting of different languages, like if you speak a different language you can just press it, and switch it to the language that you speak.

4. Discussion

This study took a phenomenological approach to understand the accessibility and feasibility of the JoyPop^{TM} iOS mobile application for Indigenous youth, partnering with a Haudenosaunee cultural and Mohawk and Cayuga language immersion school on the Six Nations of the Grand River. These findings are consistent with earlier JoyPop^{TM} studies which found that the app includes favorable features and designs, is well-utilized on a weekly basis, and engendered perceptions of positive learning and emotion regulation. While there is no specific method or protocol to approach research with Indigenous communities, the process across this research project was consistent with established frameworks for Indigenous-oriented research (e.g., Two-eyed seeing; OCAP principles) and is a potential model conducive to continued app research and development. By partnering and collaborating with the Six Nations of the Grand River, this study underscores the importance of involving Indigenous communities directly in the research process to ensure there is greater resonance impact and planning next steps.

Given that this feedback is from a Haudenosaunee perspective, there remains interest in a Haudenosaunee-specific resilience app that may utilize the positive features found in JoyPop^{TM} and extend in additional directions. The Good Mind concept, significant to the Haudenosaunee People, emerges as a promising thread that could be weaved, for example, more explicitly throughout all app features. This highlights that nation-specific research and resource development are important, as in other reserves, youth may connect more strongly to other cultural referents (e.g., significant historical figures and particularly animals such as fish in a coastal community and deer or bison in a land-based community). As such, a pan-Indigenous approach to a resilience app may not be consistent with all Indigenous perspectives, which may necessitate tailoring app approaches to specific cultural contexts.

Certain app features were considered unique, helpful, and engaging over time. This included the Art Feature and Breathing Feature. Art has been identified as a flexible vehicle for exploring positive and negative emotions and has been identified as a coping mechanism by Indigenous youth in the literature (51). Although the Art feature exclusively refers to drawing, the youth's positive feedback aligns with Indigenous practices and beliefs about art mediums as forms of expression, celebration, and healing (52). The Breathing feature was available for use to combat stress and negative emotionality, which was suggested in terms of ratings on overall mental health. Indigenous perspectives tend to endorse spiritual reflection and relaxation through activities like meditation (53). To align more with Indigenous youth, visual animations that utilize Indigenous teaching may be preferable to the current depiction of a circle expanding and contracting, such as symbols of trees that are honored or eagle feathers, which are used in traditional healing ceremonies (54). Given the higher overall risk for suicidality among Indigenous youth and the capacity for routine use of breathing techniques to physiologically address emotional regulation, such an activity may be important as a stand-alone activity outside of the app (33).

Youth were eager to provide recommendations to improve the app in alignment with their cultural grounding and Native languages. Integrating Indigenous cultural elements was previously emphasized among adult participants, who most frequently recommended changes in the design and layout of the JoyPop^{TM} app (38). Participants of this study also recommended adding more games to the JoyPop^{TM} app, but did not specify any type or specific game, suggesting that active engagement with incremental challenges can be harnessed toward resilience goals. This suggestion aligns with prior findings where the SquareMoves feature was among the most used and helpful features by providing a positive form of distraction (from negative emotions, such as panic, anxiety, and boredom). Games also play a significant role in Indigenous culture. For example, the game of lacrosse holds many important cultural beliefs and practices for Indigenous people. The Haudenosaunee believed that Lacrosse was the Creator's game and that the spirits used the game of lacrosse as a way to resolve conflicts in the Sky world (55). This game was then gifted to the Haudenosaunee, who now use lacrosse as a way to heal and strengthen community ties (55). Thus, the suggestion of adding more games to the JoyPop^{TM} app may highlight these underlying values and beliefs, which would further support the wellness and healing of Indigenous youth. It is noteworthy that the gaming feature, as well as the journaling and art drawing features, were used by all participants most frequently, in comparison to other features (SleepEase and Circle of Trust). Although employing distraction can be a valid tool for emotional regulation, the suggestion to incorporate more games into the JoyPop^{TM} must be considered in reference to the Indigenous principle of balance. This becomes even more significant when acknowledging the potential drawbacks associated with relying excessively on gaming for coping. Research has highlighted the risks and potential dangers associated with the overuse of distraction-based coping strategies or emotional avoidance, such as the risk of gaming addiction (56). Within the app, this is mitigated by the use of features across features to support both active reflection (Journaling) and active distraction or engagement in eye-hand coordination challenges (SquareMoves), as well as being able to rate and label emotions.

Finally, we were surprised to find the lack of resonance among the youth with the Circle of Trust feature. Previous research with Indigenous adults reported that the feature was most praised as it aligned with Indigenous values of maintaining relationships, and the pre-test similarly found that all participants valued relationships (38). This finding led to the exploration of the underlying reasons behind this lack of resonance and uncovered valuable insights that relate to the historical and socio-cultural context of colonization and technology. The negative reactions from youth and their unease around the privacy of the app might signify a broader mistrust of technology that has been inherited from historical experiences of colonization. Reactions from the youth toward the Circle of Trust feature ranged from confusion regarding usage to concerns about privacy and a sense of distrust toward person-specific information. One possible solution would be to address these issues through improved instructions. However, these responses from youth, more importantly, shed light on the remaining ties between technology and colonization, which tend to position technology as having Western-European ontologies and the legacy of unethical research practices. Technologies are often associated with unequal power dynamics, cultural assimilation, and the legacy of unethical research practices (57).

4.1. Strengths, limitations, and future directions

The study process was shepherded by a variety of consultations and ongoing commitment from the Six Nations community, which serves as the study's strength. Community members prioritized youth safety in the sequencing of research studies, and adaptations were made to ensure adherence to community guidelines (e.g., having a trusted member of the school community recruit the youth, as typically, a research assistant would recruit to avoid coercion). By adapting protocols to uphold community guidelines and preferences, this study took a community-based research approach that promoted collaboration, community wisdom, and co-ownership of research procedures (58, 59). This iterative method to the study process is particularly important when considering the history of exploitative research practices that have led to distrust.

While this research study took great care to implement collaborative community-driven research, there are other ways researchers can help rebuild trust with Indigenous communities, such as by utilizing existing Indigenous-led ethical protocols and standards that have been identified (60). Within these protocols, themes include: (1) balancing individual and collective rights, which includes discussing the intellectual property and ownership of data collected from Indigenous communities; (2) ensuring culturally grounded ethical principles. This includes incorporating a decolonized approach and the values, beliefs, and culture of the specific Indigenous community one is working with; and (3) ensuring community-driven/self-determined research so that Indigenous communities could access the data collected, analyzed, and the findings so that the research conducted could help them in further planning and secondary data analysis. The research methods and approach have to be cultivated together with collaborative discussion between communities and researchers to rebuild Indigenous peoples' trust.

A limitation of the study is its small sample size, making it difficult to draw overarching conclusions about the preferences of Indigenous youth toward the JoyPop^{TM} application. The start of the COVID-19 pandemic prior to active data collection likely contributed to the lower number of youth participating (61). A larger sample size may provide more insight into the inconsistencies between this study and the perspectives of Indigenous adults, particularly whether the Circle of Trust feature would promote relationships in alignment with Indigenous principles or foster distrust toward the app's handling of personal information. A future pilot study with a larger sample would be relevant for evaluating the generalizability of the current themes to a broader sample of youth across Six Nations (i.e., not immersion school attendees), other locales of Six Nations (in Quebec, Ontario, Canada, or New York State, US), or those living off-reserve. In short, sub-population research is essential in the app's early development phases.

The purpose of this study was to explore the accessibility and feasibility of the JoyPop^{TM} app with Six Nations youth and gain insight from participants on how app features can be adapted to be more relevant for its target population. Mental health mobile applications continue to hold promise as an mHealth intervention. With few resilience-oriented interventions, continued examination of app-based supports for enhancing resilience and health promotion seems resonant with Indigenous values and needs.

Data availability statement

The datasets presented in this article are not readily available because the dataset being Indigenous is not a public database and the Six Nations of the Grand River control access to it, based on Canadian ethics guidelines for research with Indigenous persons. Requests to access the datasets should be directed to allison.ay28@gmail.com.

Ethics statement

Ethics approval was received by Haudenosaunee Confederacy Council, Six Nations Elected Band Council's Research Ethics Committee, and McMaster Research Ethics Boards for consultation interviews and survey data collection (MREB #3728; HIREB #12572). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

Author contributions

AA-Y: Formal analysis, Investigation, Writing—original draft, Writing—review and editing. DM: Writing—original draft, Writing—review and editing. KB: Writing—original draft, Writing—review and editing. DG: Conceptualization, Writing—review and editing. KM: Conceptualization, Methodology, Validation, Writing—review and editing. DM-H: Conceptualization, Methodology, Validation, Writing—review and editing. CW: Conceptualization, Formal analysis, Investigation, Methodology, Supervision, Validation, Writing—original draft, Writing—review and editing. TG: Writing—review and editing.

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Profile and mental health characterization of childhood overprotection/overcontrol experiences among Chinese university students: a nationwide survey

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Introduction: The childhood experiences of being overprotected and overcontrolled by family members have been suggested to be potentially traumatic. However, the possible associated factors of these experiences among young people are still not well studied. This study aimed to partly fill such gaps by a relatively large, nationwide survey of Chinese university students.

Methods: A total of 5,823 university students across nine different provinces in China were included by the convenience sampling method in the data analyses. All participants completed the overprotection/overcontrol (OP/OC) subscale in a recently developed 33-item childhood trauma questionnaire (CTQ- 33). Data were also collected on all participants' socio-demographic profiles and characterization of mental health. Binary logistic regression was conducted to investigate the associated socio-demographic and psychological factors of OP/ OC.

Results: The prevalence of childhood OP/OC was estimated as 15.63% (910/5,823) based on a cutoff OP/OC subscale score of ≥13. Binary logistic regression suggested that being male, being a single child, having depression, having psychotic-like experiences, lower family functioning, and lower psychological resilience were independently associated with childhood OP/OC experiences (all corrected-$p<0.05$). The OP/OC was also positively associated with all the other trauma subtypes (abuses and neglects) in the CTQ-33, while there are both shared and unique associated factors between the OP/OC and other trauma subtypes. Post-hoc analyses suggested that OP/OC experiences were associated with depression in only females and associated with anxiety in only males.

Discussion: Our results may provide initial evidence that childhood OP/OC experiences would have negative effects on young people's mental health which merits further investigations, especially in clinical populations.

KEYWORDS

childhood trauma, overprotection, overcontrol, mental health, depression, psychotic-like experience

1. Introduction

Overprotection/overcontrol (OP/OC) behaviors were defined as behaviors in which caregivers (including parents and other family members) are overly involved in children's daily activities and experiences, often caused by excessive anxiety about the children's safety (1, 2). As suggested by past studies, multiple possible reasons may lead to OP/OC behaviors. For example, some parents exhibited fear in fulfilling their parenting responsibilities, which may in turn lead to their OP/OC (3). Furthermore, a lack of care by one parent can also lead to OP/OC behaviors by the other one (4). A prior research study has shown that perceived OP/OC from family members might limit children's development of a clear understanding of environmental dangers and might have negative effects on their mental health statuses (5, 6). For instance, perceived OP/OC experiences were suggested to be possibly associated with decreased self-efficacy and increased vulnerability to perceived threats (1), the development of childhood anxiety (3), as well as the onset of anorexia (7) in children and teenagers. In addition, OP/OC might be related to increased risks of depression, post-traumatic stress disorder (8), and suicidal behaviors (9).

In addition to the short-term negative psychological effects of OP/OC in children/teenagers as mentioned above, recent studies have also suggested that OP/OC might be developmentally traumatizing, and childhood OP/OC experiences may have long-term effects on one's mental health in early adulthood and even later life (5, 10, 11). For instance, individual recall of childhood OP/OC appears to be associated with a higher prevalence and incidence of adult psychological health problems in the general population (12–14). Some evidence suggests that childhood OP/OC experiences are related to sleep disturbance (15) and associated with difficult recovery in patients with schizophrenia (16) in adulthood. Another related study reported that overprotective support reduced stress in the short term but hindered individuals from coping with stress in the long run by weakening autonomy, especially when that support is terminated (17). For these reasons, perceived OP/OC during childhood has been regarded as a kind of traumatic experience besides the other well-known childhood trauma subtypes (e.g., abuses and neglects) and attracted attention in recent psychological studies (2, 18). Recognizing and identifying factors associated with childhood OP/OC experiences, therefore, may be valuable for improving our understanding of the developments of common mental problems and disorders, as well as finding potential targets for early interventions for mental disorders.

The current literature about possible associated factors of childhood OP/OC experiences in young people, however, is still limited in several ways. First, some previous results have reported inconsistent and even conflicting conclusions. For example, while many earlier studies as mentioned above suggested that childhood OP/OC is related to more mental problems including depressive and anxiety symptoms in later life, the opposite results were also reported, e.g., that paternal overcontrols predicted lower anxious-depressed symptoms (19). One of the potential reasons for these contradictory results may be the insufficient sample size in many of these studies; for instance, the samples in most of the previous studies range from only dozens to hundreds (20–23), which may lead to relatively low statistical power and unreliable results. Second most of the prior studies have focused on the associations between OP/OC and several common mental problems such as anxiety and depression; however, the knowledge is limited on the relationships between OP/OC and some other important socio-demographic profiles and mental health characteristics. These characteristics include, for example, psychological resilience which is defined as one's ability to recover and maintain adaptive behaviors when facing constant stress (24). There has been evidence that other subtypes of childhood trauma (e.g., abuses and neglects) could lead to a lower psychological resilience, which mediates the relationships between childhood trauma and depression in college students (25). As a kind of traumatic experiences, OP/OC experiences may be also associated with a lower psychological resilience, which remains however poorly investigated to our knowledge. Third, while most of the prior studies on OP/OC experiences were conducted in Western countries, it is relatively little known about the prevalence and associated factors of OP/OC among youths under other cultural conditions, such as in China. One possible reason for such a limitation is the lack of an easy and feasible screening tool for OP/OC experiences in the Chinese language. Nevertheless, this gap has been addressed by a recently validated Chinese version of the 33-item expanded childhood trauma questionnaire (CTQ-33) (18), and further studies on OP/OC among the young Chinese populations may be warranted.

In the current study, we aim to address the limitations raised above by performing a nationwide, large-sample survey among the young Chinese population. Specifically, a total of 5,823 Chinese university students across nine different provinces in China were included in the analyses. Data were collected on all participants' childhood OP/OC experiences, socio-demographic profiles, and characterization of mental health (e.g., psychological resilience). Logistic regression models were conducted to investigate the possible associations between childhood OP/OC experiences and other socio-demographic/psychological factors. We hope that our results will shed light on the understanding of the possible role of OP/OC in psychological health among young people.

2. Methods

2.1. Participants

A total of 5,993 Chinese university students were initially recruited in this survey using the convenience sampling method from nine universities across nine different provinces (Shandong, Jiangxi, Guangxi,

TABLE 1 Sample characteristics of the analyzed participants.

University	Located province	Number of participants	Females (%)/males	Age (mean ± SD)	Scores of CTQ-OP/OC subscale		
					Mean	SD	Mean + SD
Jining Medical University	Shandong	1,212	694 (57.26)/518	19.56 ± 1.28	8.88	3.64	12.52
Gannan Medical University	Jiangxi	3,065	1717 (56.02)/1,348	19.50 ± 1.56	9.01	3.37	12.38
Guangxi Medical University	Guangxi	317	187 (58.99)/130	19.96 ± 1.31	8.99	3.18	12.17
Guangzhou Medical University	Guangdong	85	51 (60.00)/34	19.49 ± 1.36	9.28	3.14	12.42
Hebei Medical University	Hebei	161	105 (65.22)/56	18.25 ± 0.74	8.61	3.08	11.69
Inner Mongolia Medical University	Inner Mongolia	131	100 (76.34)/31	18.73 ± 0.92	8.15	3.41	11.56
Qiqihar Medical University	Heilongjiang	298	177 (59.40)/121	18.37 ± 0.79	8.62	3.49	12.11
Central South University	Hunan	394	210 (53.30)/184	18.28 ± 1.08	9.08	3.48	12.56
Xinjiang Medical University	Xinjiang	160	102 (63.75)/58	20.01 ± 1.41	8.56	3.57	12.13
Total		5,823	3,343 (57.41)/2,480	19.36 ± 1.47	8.93	3.43	12.36

CTQ, childhood trauma questionnaire; OP/OC, overprotection/overcontrol; SD, standard deviation.

Guangdong, Hebei, Inner Mongolia, Heilongjiang, Hunan, and Xinjiang) in China (see distributions in Table 1). The survey was conducted from September 2021 to October 2021, and all students completed the survey online through a famous platform in China, "Questionnaire Star".[1] To avoid the potential confounding impacts of other clinical conditions on the results, students with a previous diagnosis of any psychiatric disorder were excluded ($n = 120$). In addition, students with missing data ($n = 47$) or over the age of 25 years ($n = 3$) were excluded. Therefore, 5,823 participants were included in the final data analyses in the current study (see Table 1 for sample characteristics). All participants and/or their guardians gave informed consent to agree to participate in this study. The research proposal was approved by the Ethics Committee of the Second Xiangya Hospital of Central South University.

2.2. Assessments

2.2.1. Socio-demographic factors

Information on the following socio-demographic factors was collected from all participants and taken into the analyses: age, sex, ethnicity (Han or minority), single-child household (yes or no), parental separation (yes or no), left-behind children experiences ("Are one or both parents have not been with the participants for at least 6 months before the age of 16 years?," yes or no), as well as family histories of mental disorders. Note that all participants with a personal history of mental disorders have been excluded from the analyses.

2.2.2. Measure of OP/OC experiences

Childhood OP/OC experiences of all participants were measured by the OP/OC subscale of the CTQ-33 (2). The CTQ-33 was expanded from the original 28-item childhood trauma questionnaire (CTQ-28) (26) with an additional OP/OC subscale and thus has six subscales measuring six different subtypes of childhood trauma experiences: emotional abuse, physical abuse, sexual abuse, physical neglect, emotional neglect, and OP/OC (2). All items in the CTQ-33 are 5-point Likert-type questions, and higher scores indicate higher levels of childhood trauma experiences. The Chinese version of the original CTQ-28 has been shown to have good reliability and validity in Chinese populations (27). The additional OP/OC subscale in the CTQ-33 has also been translated into Chinese and proved to be valid (18). In the current study, the CTQ-33 displayed good internal consistency (Cronbach's $\alpha = 0.843$).

Based on prior publications, participants with scores above the cutoff points for a particular subscale can be defined as having a particular subtype of childhood trauma experience as follows: physical abuse ≥10, emotional abuse ≥13, sexual abuse ≥8, physical neglect ≥10, and emotional neglect ≥15 (28, 29). In the present study, we intended to first classify all participants into those with and without childhood OP/OC experiences. However, to the best of our knowledge, an optimal cutoff point for the OP/OC subscale in the CTQ-33 has not been established to date. Therefore, referring to multiple published studies (30–32), we estimated the appropriate cutoff score for the OP/OC subscale based on one standard deviation (SD) above the mean score in the surveyed sample. The participants with an OP/OC subscale score higher than such a cutoff point were then defined as having childhood OP/OC experiences.

2.2.3. Self-reported depression

All participants completed the self-reported, 9-item Patient Health Questionnaire (PHQ-9) (33) to assess the severity of depressive symptoms over the past 2 weeks. The Chinese version of PHQ-9 has been validated in a previous study (34). Each item of the PHQ-9 was rated on four values ranging from 0 ("not at all") to 3 ("nearly every day"). The total score of PHQ-9 can range from 0 to 27, and the participants were regarded to have depression when the total score ≥10 (35). In the present study, the PHQ-9 displayed good internal consistency (Cronbach's $\alpha = 0.903$).

2.2.4. Self-reported anxiety

All participants completed the self-reported, 7-item Generalized Anxiety Disorder Scale (GAD-7) (36) to assess their anxiety levels during the last 2 weeks. The Chinese version of GAD-7 has shown good reliability and validity in the Chinese population (37, 38). Each item of

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[1] www.wjx.xn

the GAD-7 was rated from 0 ("not at all") to 3 ("nearly every day"). The total score of GAD-7 can range from 0 to 21, and the participants were regarded to have anxiety when the total score ≥10 (37, 38). The GAD-7 displayed good internal consistency in this sample (Cronbach's α = 0.923).

2.2.5. Psychotic-like experiences

The 15-item version of the Community Assessment of Psychic Experiences (CAPE-15) (39–41) was used to evaluate the psychotic-like experiences of all participants. The Chinese version of the CAPE has been validated and is widely used to assess psychotic-like experiences in Chinese populations (42–45). The CAPE-15 includes 15 items that measured both frequency of and distress associated with a series of common psychotic-like experiences (e.g., subclinical delusions and hallucinations). Both the frequency and distress scores of each item are rated on a four-point Likert scale. Referring to prior studies (46), the participants were regarded to have meaningful psychotic-like experiences when both the mean frequency score and mean distress score were greater than 1.5. The frequency score of each subject showed good internal consistency (Cronbach's α = 0.871).

2.2.6. Family functioning

The family functioning of each participant was measured by the Family APGAR scale (47, 48). The Chinese version of Family APGAR has been validated and widely used in previous studies (48–50). The Family APGAR scale consists of five items assessing family functioning from five dimensions: adaptation ("A"), partnership ("P"), growth ("G"), affection ("A"), and resolution ("R"). The score of each item ranges from 0 ("almost always") to 2 ("hardly ever"). Total scores of the Family APGAR scale can thus range from 0 to 10, and a relatively low family functioning can be defined by a total score ≤3 (48–50). The Chinese version of the Family APGAR in our research has good internal consistency (Cronbach's α = 0.922).

2.2.7. Psychological resilience

Each participant's psychological resilience was measured by the 10-item Connor-Davidson Resilience Scale (CD-RISC) (51), a self-administered questionnaire extracted from the original 25-item version (52). The Chinese version of CD-RISC has been validated and widely used in previous studies (24, 53, 54). In the CD-RISC, the score of each item ranges from 0 to 4 (0 = "never" to 4 = "almost always"), and the total score ranges from 0 to 40. Referring to prior research (53), the cutoff of a CD-RISC a total score of ≤25 was used to define a relatively low psychological resilience. The Chinese version of the CD-RISC in this sample has good internal consistency (Cronbach's α = 0.966).

2.3. Statistical analyses

Socio-demographic and psychological characteristics were first compared between the participants with and without childhood OP/OC experiences using descriptive statistics. Independent *t*-tests and chi-square tests were used for continuous variables (e.g., age) and categorical variables (e.g., sex), respectively.

In line with some prior studies (55), binary logistic regression analysis was then performed to investigate the possible associations between all socio-demographic/psychological factors (age, sex, years of education, ethnicity, province, single child, parental separation, left-behind experiences, family history of mental disorders, depression, anxiety, psychotic-like experiences, family functioning, and psychological resilience) and childhood OP/OC experiences after adjusting for the confounding effects of other variables. It should be noted that we took all factors into account in regression models and when investigating the relationship between OP/OC and one factor, the possible confounding effects of all the other factors have been excluded. In addition, the province (coded as dummy variables) was controlled in the analyzing models as a variable of no interest. The *p*-values were corrected across the 14 factors using the Benjamini–Hochberg false discovery rate (FDR) corrections, and a corrected *p*-value of < 0.05 was considered to be statistically significant. Moreover, since previous studies have suggested that OP/OC is highly positively correlated with all the other trauma subtypes (abuses and neglects) in the CTQ-33 (2), we did not include other subscales of the CTQ-33 in the regression model to avoid possible multicollinearity problems. Instead, we explored their relationships with OP/OC using separate models in the following supplementary analyses (see later in Section 2.4).

2.4. Supplementary analyses

Several supplementary analyses were performed in addition to the main analyses. First, we tested the relationships between OP/OC and other trauma subtypes (abuses and neglects) in the CTQ-33 using Spearman correlations. We also tested whether OP/OC and other trauma subtypes would have similar associated socio-demographic/psychological factors: here, all participants were classified into those with and without a particular subtype of childhood trauma (e.g., psychical abuse, based on the cutoffs mentioned in Section 2.2.2), and separate binary regression models were used to investigate the associated factors of such trauma subtype. Similar to the analyses on OP/OC, the statistical significance was set at an FDR-corrected *p*-value of < 0.05.

Second, considering that sex differences in mental health have been widely reported (56–58), we further explored the possible sex differences in relationships between OP/OC and other factors. Here, similar to analyses in the entire sample, the associated factors of childhood OP/OC experiences were assessed by binary logistic regression models in the male ($N = 2{,}480$) and female ($N = 3{,}343$) participants separately, and the statistical significance was still set at an FDR-corrected *p*-value of < 0.05.

2.5. Validation analysis

In the current study, we estimated an appropriate cutoff point for the OP/OC subscale at ≥13. To confirm whether the identified associated factors of OP/OC would change when using different cutoff scores, we repeated the regression analyses using two other different cutoff points ≥12 and ≥14, respectively.

3. Results

3.1. Sample characteristics and estimated cutoff points

Sample characteristics of the analyzed participants are shown in Table 1. The proportion of female participants was 57.41%

(3,343/5,823), and the average age was 19.36 years (SD = 1.47) for the entire sample.

Before the analyses, we first compared the OP/OC scores between different provinces to see whether they can be treated as a homogeneous sample. In the entire sample, the (mean + 1SD) value of the OP/OC subscale score was 12.36. Meanwhile, the (mean + 1SD) values of the OP/OC subscale scores were found to be very close across the subsamples from different provinces. Specifically, in most (7/9) of the subsamples, such values were in the range of 12.11–12.56 except in Hebei (11.69) and Inner Mongolia (11.56; Table 1). Nevertheless, these two provinces had relatively small sample size ($N = 161/131$ for Hebei and Inner Mongolia, respectively) which may bias the results. Therefore, we propose that the distributions of OP/OC scores in different provinces were very close, suggesting they can be treated as a homogeneous sample. According to these results, we also propose that an OP/OC subscale score of ≥13 may be an appropriate cutoff to classify those having and not having clinically meaningful OP/OC experiences. Such a cutoff point was also applied in the following analyses.

3.2. Group comparisons on socio-demographic and psychological characteristics

Based on the above cutoff point (OP/OC subscale score ≥ 13), the prevalence of OP/OC experiences was estimated as 15.63% (910/5,823) in the current sample. Results of the direct comparisons on all characteristics between the participants with and without OP/OC experiences were shown in Table 2. Compared with those without OP/OC, the participants with OP/OC experiences had a higher proportion of males ($p < 0.001$), a higher proportion of single child ($p = 0.031$), and a higher proportion of "left-behind" children ($p = 0.006$). Compared with those without OP/OC, the participants with OP/OC experiences are more likely to have depression, anxiety, psychotic-like experiences, low family functioning, and low psychological resilience (all $p < 0.001$).

3.3. Results of binary logistic regression analysis

As shown in Table 3 and Figure 1A, after controlling for confounding factors in the logistic regression model, the following factors remained independently associated with OP/OC experiences: being male (odds ratio 1.973, 95% confidence interval 1.685–2.311, corrected $p < 0.001$), being a single child (odds ratio 1.232, 95% confidence interval 1.033–1.471, corrected $p = 0.046$), having depression (odds ratio 1.436, 95% confidence interval 1.105–1.866, corrected $p = 0.018$), having psychotic-like experiences (odds ratio 2.231, 95% confidence interval 1.872–2.659, corrected $p < 0.001$), having low family functioning (odds ratio 3.808, 95% confidence interval 3.188–4.549, corrected $p < 0.001$), and having low psychological resilience (odds ratio 2.126, 95% confidence interval 1.799–2.511, corrected $p < 0.001$). There were no statistically significant associations between OP/OC and the following factors: province, age, years of education, ethnicity, parental separation, left-behind experiences, family history of mental disorders, and

TABLE 2 Comparisons on socio-demographic and psychological characteristics between the participants with and without childhood OP/OC experiences.

Variables	With OP/OC (*n* = 910)	Without OP/OC (*n* = 4,913)	Group comparison
Age (years, mean ± SD)	19.31 ± 1.48	19.37 ± 1.47	$t = 1.064$, $p = 0.287$
Sex	–	–	$\chi^2 = 115.786$, $p < 0.001$***
Male, *n* (%)	535 (58.79%)	1945 (39.59%)	–
Female, *n* (%)	375 (41.21%)	2,968 (60.41%)	–
Years of education (mean ± SD)	13.09 ± 1.25	13.11 ± 1.19	$t = -0.482$, $p = 0.630$
Ethnicity	–	–	$\chi^2 = 0.079$, $p = 0.779$
Han, *n* (%)	851 (93.52%)	4,582 (93.26%)	–
Minority, *n* (%)	59 (6.48%)	331 (6.74%)	–
Single child, *n* (%)	300 (32.97%)	1,444 (29.40%)	$\chi^2 = 4.679$, $p = 0.031$*
Parental separation, *n* (%)	98 (10.77%)	500 (10.18%)	$\chi^2 = 0.292$, $p = 0.589$
Left-behind experiences, *n* (%)	318 (34.95%)	1,492 (30.37%)	$\chi^2 = 7.507$, $p = 0.006$**
FHMD, *n* (%)	17 (1.87%)	78 (1.59%)	$\chi^2 = 0.376$, $p = 0.540$
Depression, *n* (%)	163 (17.91%)	298 (6.07%)	$\chi^2 = 147.806$, $p < 0.001$***
Anxiety, *n* (%)	92 (10.11%)	162 (3.30%)	$\chi^2 = 85.416$, $p < 0.001$***
Psychotic-like experiences, *n* (%)	355 (39.01%)	710 (14.45%)	$\chi^2 = 309.885$, $p < 0.001$***
Low family functioning, *n* (%)	697 (76.59%)	1835 (37.35%)	$\chi^2 = 481.149$, $p < 0.001$***
Low psychological resilience, *n* (%)	471 (51.76%)	1,065 (21.68%)	$\chi^2 = 357.747$, $p < 0.001$***
Total, *n* (%)	910 (15.63%)	4,913 (84.37%)	–

***$p < 0.001$, **$p < 0.01$, *$p < 0.05$. FHMD, family history of mental disorders; OP/OC, overprotection/overcontrol; SD, standard deviation.

anxiety (all corrected $p > 0.05$), after controlling for confounding factors.

3.4. Supplementary analyses on other trauma subtypes

As shown in Table 4, significant positive correlations were found between the OP/OC score and scores of all other trauma subtypes in the CTQ-33 (all $p < 0.001$), confirming that OP/OC is highly positively associated with the other trauma subtypes. Results of the separate binary logistic regression analyses for factors associated with other trauma subtypes are shown in Figures 1B–F and Supplementary Tables S1–S5. Generally, it was found that the OP/OC and other trauma subtypes have both shared and unique associated factors ($p < 0.05$ after corrections). For example, all trauma subtypes including OP/OC were found to be positively associated with having psychotic-like experiences, having low family functioning, and having low psychological resilience; meanwhile, parental separation was found to be associated with only the physical neglect and emotional neglect experiences (Figure 1).

3.5. Supplementary analyses on possible sex differences

Results of separate logistic regression analyses in the female or male participants are shown in Figure 2 and Supplementary Tables S6, S7. Generally, most of the associated factors of OP/OC were found to be consistent across the female and male participants (corrected $p < 0.05$ in both the two subsamples). The exceptions were that OP/OC was found to be associated with depression in only the female participants, and associated with anxiety in only the male participants (Figure 2).

3.6. Validation analysis

When using the cutoff points of OP/OC subscale score ≥12 or ≥14, 20.81% (1,212/5823) and 12.31% (717/5823) of the surveyed participants were categorized as having OP/OC experiences, respectively. The following factors were still found to be significantly associated with OP/OC when using the above different cutoff points: being male, being a single child, having depression, having psychotic-like experiences, having lower family functioning, and having lower psychological resilience (all corrected $p < 0.05$, see Supplementary Tables S8, S9).

4. Discussion

In this study, we investigated the possible associations between childhood OP/OC experiences and a series of socio-demographic and psychological factors in a nationwide sample of Chinese university students. Generally, our results suggested multiple non-modifiable (e.g., sex) and modifiable (e.g., family functioning) factors that could be independently associated with childhood OP/OC experiences. The OP/OC was also positively associated with all the other assessed trauma subtypes (abuses and neglects) in the CTQ-33. These results may provide initial evidence that childhood OP/OC experiences might have negative effects on the mental health in young populations.

In the current study, we first explored an appropriate cutoff point for the OP/OC subscale in CTQ-33 based on the statistical distributions in the surveyed sample. The cutoff was estimated at ≥13, and 15.63% (910/5823) of the surveyed participants were categorized as having OP/OC experiences according to such cutoff point. This prevalence is higher than those of physical abuse (8.59%, 500/5823), emotional abuse (8.07%, 470/5823) and sexual abuse (8.98%, 522/5823) but lower than those of physical neglect (33.52%, 1952/5823) and emotional neglect (16.26%, 947/5823) in the current sample. Note that all the identified associated factors of OP/OC were found to be unchanged when using

TABLE 3 Results of the binary logistic regression analysis for factors associated with OP/OC.

Variables	B	SE	Wald	Significance	Odds ratio	95% CI for odds ratio	
						Lower	Upper
Age	−0.080	0.046	3.055	$p = 0.149$	0.923	0.844	1.010
Male (*vs* female)	0.680	0.081	70.915	$p < 0.001$***	1.973	1.685	2.311
Years of education	0.017	0.056	0.092	$p = 0.849$	1.017	0.911	1.136
Minority (*vs* Han ethnicity)	0.150	0.175	0.738	$p = 0.507$	1.162	0.825	1.637
Single child	0.209	0.090	5.368	$p = 0.046$*	1.232	1.033	1.471
Parental separation	−0.033	0.130	0.065	$p = 0.849$	0.968	0.750	1.248
Left-behind experiences	0.131	0.088	2.215	$p = 0.218$	1.140	0.959	1.354
FHMD	−0.057	0.301	0.036	$p = 0.849$	0.944	0.524	1.702
Depression	0.362	0.134	7.323	$p = 0.018$*	1.436	1.105	1.866
Anxiety	0.247	0.172	2.067	$p = 0.218$	1.281	0.914	1.795
Psychotic-like experiences	0.803	0.089	80.433	$p < 0.001$***	2.231	1.872	2.659
Low family functioning	1.337	0.091	217.369	$p < 0.001$***	3.808	3.188	4.549
Low psychological resilience	0.754	0.085	78.765	$p < 0.001$***	2.126	1.799	2.511

The presented p values were FDR-corrected. ***$p < 0.001$, **$p < 0.01$, *$p < 0.05$. CI, confidence interval; FDR, false discovery rate; FHMD, family history of mental disorder; OP/OC, overprotection/overcontrol; SE, standard error.

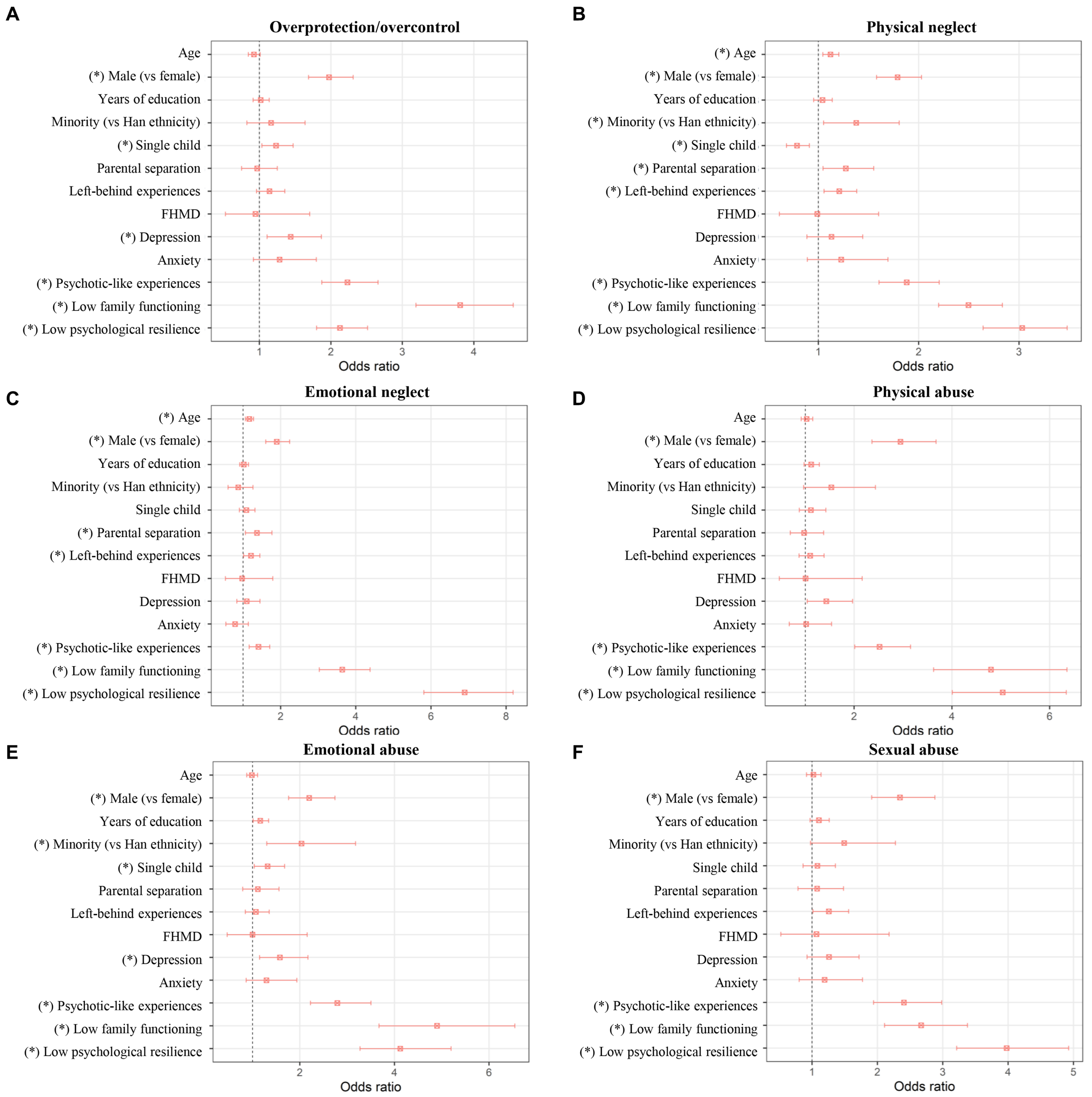

FIGURE 1
Results of separate binary logistic regression analyses for factors associated with OP/OC **(A)** and for factors associated with other trauma subtypes **(B–F)**. The odds ratios with 95% confidence intervals are presented, and the "*" indicates a significant association with corrected $p < 0.05$. FHMD, family history of mental disorder; OP/OC, overprotection/overcontrol.

TABLE 4 **Spearman correlation coefficients between the OP/OC score and scores of other trauma subtypes in the CTQ-33.**

	Physical neglect	Emotional neglect	Physical abuse	Emotional abuse	Sexual abuse
OP/OC	0.340***	0.380***	0.408***	0.454***	0.353***
Physical neglect		0.551***	0.392***	0.404***	0.370***
Emotional neglect			0.343***	0.338***	0.285***
Physical abuse				0.559***	0.574***
Emotional abuse					0.450***

CTQ-33, the 33-item childhood trauma questionnaire; OP/OC, overprotection/overcontrol; the "*" indicates a significant correlation with $p < 0.001$.

different cutoff points at ≥12 and ≥14 (see Supplementary Tables S8, S9); therefore, the main conclusions in this study are unlikely to be largely driven by different choices in cutoff points.

Using the binary logistic regression model, we found that being male and being a single child are positively associated with childhood OP/OC experiences (Table 3). The observed sex effects on OP/OC are

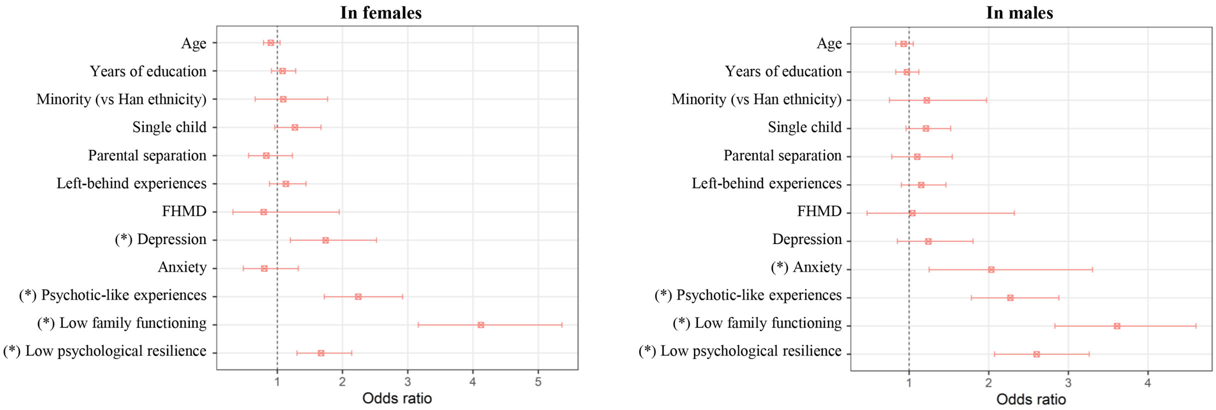

FIGURE 2
Results of separate binary logistic regression analyses for factors associated with OP/OC in the female or male participants. The odds ratios with 95% confidence intervals are presented, and the "*" indicates a significant association with corrected $p < 0.05$. FHMD, family history of mental disorder; OP/OC, overprotection/overcontrol.

partly consistent with previous research showing sex differences in perceived parenting styles (59). We propose that several biological and social factors might account for such sex differences. For example, boys are favored over girls under the traditional ideology of son preference (60), which may lead to more focus on the boys than girls in some families. For the same reason, the children which are the single child of their family might attract more attention, and even overprotective parental strategies. Notably, we did not find significant associations between parental separation and OP/OC. One possible reason might be that children can be affected differently by whether their parents' separation was amicable or conflict-ridden (61).

The regression analyses suggested that having depression is independently associated with OP/OC, even after adjusting for possible confounding effects of all other variables (Table 3). To the best of our knowledge, the findings in previously published studies are not totally consistent regarding the possible associations between OP/OC experiences and levels of depressive symptoms in later life. For example, one earlier research reported a strong association between negative parenting behaviors such as overprotection and later depressive symptom (8, 13). However, there is also research suggesting that paternal overcontrols can predict lower depressive symptoms (19). It is noteworthy that compared to most of these studies, our study has a much larger sample size and thus a higher statistical power. Therefore, this study may provide more solid evidence in support of the positive association between OP/OC and depression. In fact, multiple previous studies have also underlined OP/OC and other childhood traumas as predictors of dissociative depression alongside some linkage to the "traumatic narcissism" concept (13, 62), which are in line with our results and give a possible explanation for such relationship.

Our results also suggested that having psychotic-like experiences is independently associated with OP/OC (Table 3). To the best of our knowledge, this study is one of the first reports to suggest a positive relationship between OP/OC and psychotic-like experiences. Psychotic-like experiences are subclinical delusion-like or hallucination-like symptoms, which are related to increased risks of developing subsequent mental disorders (55). Previous studies have shown that some other subtypes of childhood trauma such as abuses and neglects would strongly increase the risks of developing schizophrenia and other psychotic disorders (63, 64), which may be presented as having psychotic-like experiences in the early stage (65). Here, our results suggest that OP/OC, as another subtype of childhood trauma, is also associated with psychotic-like experiences.

Additionally, we found that childhood OP/OC experiences are associated with lower family functioning and lower psychological resilience (Table 3). Both family dysfunction (66) and decreased psychological resilience (67) have been linked to higher risks of developing mental problems. Lower family functioning was also associated with lower wellbeing and higher risks of substance use (68, 69). These findings, together with the observed significant effects of OP/OC on depression and psychotic-like experiences, may thus highlight the unignorable negative effects of OP/OC experiences on young people's mental health.

As supplementary analyses, we have explored the possible differences in associated factors between OP/OC and other childhood trauma subtypes. It was found that some associated factors, such as having psychotic-like experiences, lower family functioning, and lower psychological resilience, were shared for all different trauma subtypes including OP/OC (Figure 1). Some differences were also found; for example, being a single child was positively associated with OP/OC but negatively associated with physical neglect; furthermore, having depression was positively associated with OP/OC and emotional abuse but not significantly associated with other trauma subtypes (Figure 1). Therefore, while being a subtype of traumatic experiences, there might be both common and unique features between the OP/OC and other trauma subtypes.

We have also explored the possible sex differences in relationships between OP/OC and other factors by performing analyses in the female and male participants separately. Generally, we found that most of the associated factors of OP/OC were consistent across the female and male participants; however, interestingly, the OP/OC experiences were associated with depression in only the female participants and associated with anxiety in only the male participants (Figure 2). There has been ample evidence for significant sex differences in multiple psychological characteristics, e.g., that females are more likely to be affected by depression (58, 70). Here, our results may partly help to further understand the sex differences in these psychological characteristics in the aspect of different influences of childhood OP/OC experiences.

This study has certain limitations. First, because of the nature of the cross-sectional survey, we are unable to establish the causality in relationships between OP/OC experiences and other factors. Therefore, further longitudinal studies are needed to address such a limitation. Second, several self-reported retrospective scales were used in this study, which may lead to memory-related biases. Third, the OP/OC experiences from one's father, mother, or other family members were not distinguished in the CTQ-33, which might have different associated socio-demographic factors and psychological effects. This limitation may be overcome by using other scales in future studies. Last, while only healthy participants were included in the current study, further studies conducted in clinical populations with mental disorders may provide more important implications for understanding the negative effects of childhood OP/OC experiences.

5. Conclusion

In conclusion, this study investigated the possible associated factors of childhood OP/OC experiences in young populations using the CTQ-33 and a relatively large, nationwide sample of Chinese university students. The main findings include that being male, being a single child, having depression, having psychotic-like experiences, having lower family functioning, and having lower psychological resilience were independently associated with childhood OP/OC experiences. The OP/OC was also positively associated with all the other trauma subtypes (abuses and neglects) in the CTQ-33; nevertheless, the OP/OC and other subtypes of trauma were found to have both shared and unique associated factors. Collectively, these results may provide initial evidence that childhood OP/OC experiences would have negative effects on young people's mental health and highlight the great value of further investigations on OP/OC especially in participants with mental disorders. The results of this survey in healthy Chinese individuals might also provide baseline reference data for potential future studies on OP/OC in clinical populations.

Data availability statement

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

Ethics statement

The studies involving humans were approved by Ethics Committee of the Second Xiangya Hospital of Central South University. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

JZ, ZW, ZL, and YL contributed to the conception and design of the study. JZ, ZW, MC, MY, LZ, MS, DL, GC, QY, HT, CA, ZL, and YL contributed to the data acquisition. JZ, ZW, and YL contributed to the analysis and interpretation of data. JZ and YL drafted the manuscript. HT, MS, and XH revised the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

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

Publisher's note

Supplementary material

The supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2023.1238254/full#supplementary-material

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Adverse childhood experiences: impacts on adult mental health and social withdrawal

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Background: Adverse childhood experiences (ACEs) have been found to negatively impact adult mental health outcomes. Numerous studies have highlighted on ACEs in family and community settings. However, few have examined the impact of ACEs in school settings, despite the potential influence on social participation. Hikikomori, characterized by severe social withdrawal, was first studied in Japan and has gained recognition in recent years. The present study aims to present the concept of ACEs specific to schools and investigate the impact of both school ACEs and traditional ACEs on adult mental health and Hikikomori.

Methods: A total of 4,000 Japanese adults, aged 20–34, were recruited through an Internet survey form. All data were obtained in October 2021. Participants answered questions regarding their ACEs in the family (10 items), school ACEs (five teacher-related items and two bullying-related items), depressive/anxiety symptoms, and Hikikomori (remaining at home for more than 6 months).

Results: A significant association with depressive/anxiety symptoms was shown in both ACEs and school ACEs. An increase of one point in the ACE scores was associated with a 24% increase in the risk of depressive/anxiety symptoms. School ACE scores also demonstrated a significant association with depressive/anxiety symptoms, with an increase of one point associated with a 44% increase in the risk of these symptoms. As for Hikikomori, a significant association was shown in the school ACEs only: a 29% increased risk of Hikikomori for every one-point increase in school ACE scores. Both school ACE scores for teacher-related and bullying-related factors revealed a significant association with Hikikomori; the rates of increased risk were 23 and 37%, respectively.

Conclusion: These results suggest that school ACEs, rather than ACEs in the family, are associated with the risk of Hikikomori. School ACEs are important for social adaptation, and reducing traumatic experiences in school settings may have the potential to prevent problems in later life, specifically in terms of social participation.

KEYWORDS

adverse childhood experiences, ACEs, school, social withdrawal, Hikikomori, bullying

1. Introduction

1.1. Adverse childhood experiences studies

ACEs encompass highly distressing events that children may experience, such as child abuse, domestic violence, and parental substance abuse (1). These ACEs are associated with illicit drug use, mental illness, and cardiovascular diseases (1). The economic burden of ACEs is substantial, estimated to be $581 billion in Europe and $748 billion in North America, with 75% of the cost incurred by individuals who have experienced two or more ACEs (1).

The ACEs study conducted by Kaiser Permanente and the Centers for Disease Control and Prevention in the United States is a representative research (2). This study found that ACEs have enduring effects on both physical and social aspects of wellbeing throughout one's life (3). Subsequent research has corroborated these findings, highlighting the impact of ACEs on health outcomes during adulthood (4–6). Thus, the prevention of ACEs has significant implications not only for personal wellbeing, but also for social stability and reduction of financial burdens. The association between ACE scores and health and social problems in adulthood is usually proportional, particularly when ACE scores are ≥4, and the risk increases dramatically (7).

The prevalence of ACEs is known to differ based on socio-economic status (8), and race (9), and changes over time (10). However, it is likely that the impact of ACEs transcends national and cultural differences (11). Consequently, the ACEs study has spread worldwide and has been conducted in various regions, including Asia. For instance, Qu et al. (12) reported that at least one adverse experience had been encountered by 51.2% of elementary and junior high school students in China. Additionally, Lin et al. (13) found that 89.9% of Chinese adults aged 45 and older had at least one ACE, with 18.0% having ≥4. Similarly, Wang et al. (14) investigated ACEs among Taiwanese youth and found that 61.6% had at least one ACE. In Japan, the prevalence of experiencing at least one ACE, among adults, is ∼27–40% (15–17). According to a review by Bellis et al. (1), research conducted in the USA indicated that individuals with at least one ACE accounted for 52–67%, while in Europe, this figure ranged from 25 to 53%. Notably, Japan exhibits a tendency toward lower ACE scores than other countries. Therefore, referencing the ACEs study when making policy decisions to support children and develop social systems would be beneficial.

1.2. Expanded ACEs

The ACEs study initially focused on estimating the prevalence and examining the association with outcomes, but has since expanded in various directions. One key aspect of the discussion is regarding the expansion of the ACEs concept itself. The original ACE scale only encompasses experiences of physical and emotional abuse within the family; it does not include other types of ACEs such as experiences of discrimination in the community, or harm inflicted by friends outside the home. Consequently, the criteria for expanding ACEs were established by the World Health Organization (WHO), including biological relevance (i.e., eliciting a biological stress reaction), policy sensitivity, prevalence across societies (neither too high nor too low), ease and speed or measurement, and proximity to causality (18, 19). Additionally, the suggestion of adding "exposure to community violence" was proposed (20).

Cronholm et al. (20) investigated additional items for the expanded ACEs, such as witnessed violence, experienced discrimination, unsafe neighborhoods, experienced bullying, and lived in foster care. Moreover, they found that these items were also associated with poverty (20). Thurston et al. (21) focused on community-level ACEs, such as community violence and racial discrimination, highlighting the need to consider ACEs at the community level. Moreover, Masuda et al. (22) found that extra-familial ACEs, such as exposure to community violence, exhibited a stronger correlation with psychosomatic symptoms compared to intra-familial ACEs, in a study of Japanese university students. SmithBattle et al. (23) reported that the most commonly discussed expanded ACE items in prior research included exposure to community violence, economic hardship in childhood, bullying, absence or death of parent or significant others, and discrimination, with the former being the most frequently mentioned.

Furthermore, recent studies utilizing the National Study of Child Health, which has been extensively discussed, have included exposure to community violence and discrimination as additional ACE items. It is widely acknowledged that the original ACE items alone are insufficient, and further research is warranted (20).

1.3. School ACEs

Previous research has shown that the impact of ACEs cannot be solely attributed to experiences within the household, but also extends to experiences outside of the home. Sweeting et al. revealed that the effects of bullying are equally severe to those of ACEs, based on a survey conducted on a representative sample in the United States (24). Bullying, due to its impact on physical health risks similar to other ACEs, should be categorized as a new item within the ACEs framework (25).

Instances of teachers causing harm to children and the subsequent negative effects on wellbeing have been reported. Gershoff (26) stated that although physical punishment is permitted in 35% of countries, evidence suggesting that it enhances learning is non-existent. Instead, it increases the risk of dropping out and teacher or school avoidance. Research conducted by Nearchou (27) in Greece found that experiences of psychological abuse from teachers predict problematic behavior, with a significant number of children (64%) having been victims of psychological abuse from teachers.

Moreover, Monsvold et al. (28) reported that experiencing bullying victimization by teachers is associated with an increased risk of personality disorders. Delfabbro et al. (29) also highlighted that bullying by teachers occurs as frequently as bullying by peers, with low-achieving children being more vulnerable to being victimized by teachers.

These findings suggest that various harmful experiences occur in school settings, including bullying, physical punishment, inappropriate teaching methods, and harassment, which may have long-lasting negative impacts similar to ACEs in adulthood. However, previous ACEs studies have only included witnessing or experiencing community violence as an additional item to original ACEs, with questions limited to peer bullying in school settings and no inclusion of teachers or other school staff as perpetrators (30, 31).

1.4. Hikikomori or social withdrawal

It is well-known that ACEs have a negative impact on employment and work performance. Individuals with ACEs are at a higher risk of joblessness and poor work performance (32, 33). One form of social withdrawal is known as Hikikomori, which was first studied in Japan and has recently gained attention worldwide (34, 35). Hikikomori is characterized by a refusal to leave one's home or room (36, 37) and is associated with suicide, compulsive behavior, and dependent behavior (38). The prevalence of Hikikomori in Japan was estimated to be 2.05% (39), 1.9% in Hong Kong (40), and 2.3% in South Korea (41). The high prevalence of Hikikomori has negative implications for society. Kato et al. (37) proposed a hypothetical model in which family factors, such as strong maternal and weak paternal relationships, and school factors, such as a less competitive environment (yutori-kyoiku) or highly competitive atmosphere (juken war), contribute to the occurrence of Hikikomori. However, no studies have quantitatively examined whether adverse experiences in school are associated with the risk of Hikikomori. Furthermore, ACEs have been reported to increase the risk of employment problems (32, 42) and workplace bullying (43), but no studies have investigated the association between ACEs and Hikikomori.

2. About this study

In this study, we introduce the concept of "school ACEs" in addition to the original ACEs items. We recognize that teachers in school settings may have the potential to harm children, resulting in long-lasting effects. School ACEs encompass peer bullying (classmates and upper classmen) that commonly occurs in schools. This additional item is integrated within the existing ACEs item on abuse, with the distinction being that teachers are identified as the perpetrator as opposed to family members.

Although bullying has already been examined, experiences of being hurt by teachers and other school staff are likely to satisfy the criteria for ACEs expansion set by WHO (18). In Japan, corporal punishment occurs in 0.63% of elementary schools, 1.33% of junior high schools, and 3.51% of high schools despite being prohibited by law (44). Here, corporal punishment refers to physical acts such as hitting, kicking, and shoving by teachers and does not include psychological effects such as verbal violence, threats, ignoring, or negative evaluations. According to a survey by the Tokyo Metropolitan Government (45), verbal violence occurs more often than corporal punishment, and the possibility that children may be harmed by non-corporal punishment by teachers cannot be ruled out, making the examination of school ACEs crucial (45).

This study investigates the outcomes of the school ACEs as possible worsening mental health in adulthood and social withdrawal, Hikikomori.

Mental health deterioration in adulthood must be prevented as it is a societal burden and a personal or family problem (46, 47). Mental health deterioration is a representative outcome of ACEs (1, 3, 4, 12, 48), and worsening of mental health in young people is likely associated with a wide range of individual and societal issues such as marital status and household income in adulthood (46). However, the association between school ACEs and deteriorating mental health is yet to be explored.

Regarding Hikikomori, according to the occurrence model shown by Kato et al. (37), stressful life events at school or workplace cause evasive behavior, which leads to Hikikomori. The presence of school ACEs, such as bullying and receiving reprimand by teachers, within the category of stressful life events, suggests a potential association between school ACEs and Hikikomori. However, no research has been conducted to substantiate this claim.

Therefore, this study aims to assess the prevalence of ACEs and school ACEs in Japan. Furthermore, we aim to examine the association between school ACEs and the deterioration of adult mental health, as well as their association with Hikikomori.

Our research questions are as follows:

#1 What is the prevalence of ACEs and school ACEs in Japan?
#2 How are ACEs and school ACEs related to adult depressive/anxiety symptoms?
#3 How are ACEs and school ACEs related to "Hikikomori"?

3. Methods

3.1. Participants

A total of 4,000 Japanese adults, ages 20–34, were recruited through an Internet survey form (Survey Research Center Co., Ltd. Tokyo, Japan). For comparison purposes, the ages of the participants in this study were matched to those in the Cabinet Office's Survey on Hikikomori conducted in (15–39 years of age) Japan (39). However, due to ethical considerations, the target age was set at 20 and above. In addition, because the Japanese school education system differed greatly between the 35–39 and ≤34 age groups, the survey was limited to ≤34 age group. Age groups were divided into 20–24, 25–29, and 30–34, with each group recruited to ensure equal numbers and sex ratios. A total of 1,333 participants were included in the 20–24 age group (49.3% male, 49.3% female, and 1.4% non-response), 1,334 in the 25–29 age group (49.3% male, 49.3% female, and 1.4% non-response), and 1,332 in the 30–34 age group (49.6% male, 49.7% female, and 0.7% non-response).

Initially, a trap question ("Do not answer this question") was inserted into a survey to filter out respondents who are not answering honestly or carefully. Only those participants who passed the trap (i.e., those who did not respond to the item) were included. One participant who passed the trap but provided the same response to all questions including an invert scale was excluded from the analysis. All data were obtained in October 2021.

The survey was conducted anonymously. A written explanation of the survey was presented online, and consent for participation was obtained by checking the "I agree" box. Participants were rewarded with points (Japanese yen equivalent: 4 yen) that could be used online as compensation for their participation herein. The Hirosaki University Ethics Committee approved this study (reference No: 2021-011).

3.2. Measures

3.2.1. ACEs

Using the ACEs Questionnaire, childhood trauma was measured (3). The questionnaire assesses 10 types of childhood trauma. Five are personal: physical abuse, verbal abuse, sexual abuse, physical neglect, and emotional neglect. Five are family dysfunctions: a parent who is an alcoholic, a mother who is a victim of domestic violence, a family member in jail, a family member diagnosed with a mental illness, and the disappearance of a parent through divorce, death, or abandonment. Responses were binary, "yes" or "no," with the number of "yes" items being the total score.

3.2.2. School ACEs

The items were created by replacing the subject term of the five items of the ACEs questionnaire (physical abuse, verbal abuse, sexual abuse, emotional neglect, and witness of victims of violence) as follows: "Did a parent or other adult in the household ..." was replaced to "Did a teacher or other adult in school (or preschool) ...." Additionally, two items related to bullying at school were added to the school ACE items: one item related to bullying victimization by classmates and the other item related to bullying victimization by upperclassmen. Responses are binary, "yes" or "no", with the number of "yes" items being the total score.

3.2.3. Depressive/anxiety symptoms

To assess depression and anxiety, the Patient Health Questionnaire-4 (PHQ-4) was used (49, 50). This scale consists of four items rated on a four-point Likert scale: two items extracted from the PHQ-9 (51) and two from the Generalized Anxiety Disorder-7 (GAD-7) (52). The total score was calculated, and the status of mental health problems was classified into four categories: normal (0–2), mild (3–5), moderate (6–8), and severe (9–12) (49). In this study, depressive/anxiety symptoms were dichotomized according to the level of severity as follows: moderate/high (6–12), or not (0–5).

3.2.4. Hikikomori

We asked participants how often they go out, how long they have not been out including the reasons, and their current employment status. The "Guideline for the Assessment and Support of *Hikikomori*" defines *Hikikomori* as a phenomenological concept that refers to a state of avoidance of social participation (e.g., going to school, working, and socializing outside the home) as a result of various factors and remaining at home for 6 months or longer in principle (except for going out without socializing with others) (53). In accordance with this guideline, we defined a group of *Hikikomori* as follows: a person who remains at home for more than 6 months except for going out without socializing with others, who is not employed, a self-employed worker, a full-time homemaker, or a student and the reason for their current condition was not the result of illness, pregnancy, nursing care, effects of coronavirus disease 2019 (COVID-19) outbreak, or natural disasters.

3.2.5. Background factors

Information on sex, age, nationality, education, number of family members living together, and economic conditions was collated as participants' background factors. The items were almost identical to those in the Cabinet Office's Survey (39). Item categories are shown in Table 1.

3.3. Statistical analysis

First, the total scores and prevalence rates of each item for ACEs and school ACEs were calculated. Next, the correlation between each ACEs item and school ACEs were examined through a correlation analysis. Separate logistic regression analyses were then performed for the depressive/anxiety symptoms and Hikikomori as outcomes, respectively. Potential confounding factors such as sex, age, education, living conditions, and number of family members were used to control the effects of ACEs and school ACEs. Since 99.7% of the participants was Japanese nationals, nationality was not included in the model. For the school ACEs score, we used the total score in Model 1 and divided it into teacher-related and bullying-related scores in Model 2.

In addition, we examined the impact of having ACEs and school ACEs on outcomes. Outcomes were moderate with higher levels of depressive/anxiety symptoms and the Hikikomori, and logistic regression analysis was used. Exposure was having both at least one ACE and at least one school ACE, or having both ≥ 4 ACEs and at least one school ACE.

4. Results

4.1. Prevalence of ACEs, school ACEs

Except for one participant who provided the same response to all questions, all participants (n = 3,999) had no missing data and were included in the analysis. Table 1 shows the background information of the sample and the prevalence of ACEs and school ACEs. The mean score of ACEs was 0.76 [standard deviation(SD) = 1.37], and 35.9% (n = 1,436) of the entire sample had at least one ACE. In addition, 6.1% (n = 244) had an ACEs score of ≥ 4. The mean score of the school ACEs was 0.96 (SD = 1.18), and 55.1% (n = 2,202) of the entire sample had at least one ACE. When dividing the school ACEs into teacher-related (five items) and bullying-related scores (two items), 20.5% (n = 819) of the entire sample had a score of ≥ 1 on the teacher-related items, and 50.5% (n = 2,020) had a score of 1 or more on the bullying items. Participants who showed moderate or higher levels of depressive or anxiety symptoms were 16.3% (n = 653), and participants assigned to the Hikikomori group were 3.5% (n = 138). Half (n = 69) of the

TABLE 1 Demographic characteristics of participants and prevalence of ACEs.

	Total sample ($n = 3,999$)
Sex: men, n (%)	1,976 (49.4)
Women, n (%)	1,977 (49.4)
Other, n (%)	46 (1.2)
Age, mean (SD)	27.2 (4.3)
Nationality: Japanese, n (%)	3,986 (99.7)
Other, n (%)	13 (0.3)
Academic background:	
Junior high school, n (%)	77 (1.9)
High school, n (%)	797 (19.9)
Vocational school, n (%)	411 (10.3)
Junior college, n (%)	253 (6.3)
College or graduate school, n (%)	2,409 (60.2)
Other, n (%)	52 (1.3)
Life circumstances (9-point scale), mean (SD)	4.7 (1.4)
Number of family members, mean (SD)	2.7 (1.4)
ACE total score, mean (SD)	0.76 (1.37)
Emotional abuse, n (%)	503 (12.6)
Physical abuse, n (%)	390 (9.8)
Sexual abuse, n (%)	162 (4.1)
Emotional neglect, n (%)	460 (11.5)
Physical neglect, n (%)	94 (2.4)
Divorce, n (%)	716 (17.9)
Mother treated violently, n (%)	210 (5.3)
Substance abuse, n (%)	177 (4.4)
Mental illness, n (%)	298 (7.5)
Incarcerated relative, n (%)	34 (0.9)
ACE > 1 point, n (%)	1,436 (35.9)
ACE > 4 point, n (%)	244 (6.1)
School ACE total score, mean (SD)	0.96 (1.18)
School ACE teacher-related score, mean (SD)	0.32 (0.75)
School ACE bullying-related score, mean (SD)	0.64 (0.71)
Emotional abuse, n (%)	297 (7.4)
Physical abuse, n (%)	149 (3.7)
Sexual abuse, n (%)	51 (1.3)
Emotional neglect, n (%)	577 (14.4)
Friends treated violently, n (%)	208 (5.2)
Bullying victimization by classmates, n (%)	1,924 (48.1)
Bullying victimization by senior students, n (%)	632 (15.8)
School ACE > 1 point, n (%)	2,202 (55.1)
PHQ-4 total score, mean (SD); median	2.65 (3.16); 2
PHQ-4 moderate/severe group, n (%)	653 (16.3)
Hikikomori group, n (%)	138 (3.5)

Hikikomori group was assigned to the moderate or higher levels of depressive or anxiety group. This was significantly higher than the 15.1% ($n = 584$ out of $n = 3,861$) in the non-Hikikomori group ($\chi^2 = 118.6$, $p < 0.001$).

Table 2 shows the correlations between each item of ACEs and school ACEs. Within the 10 items of ACEs, strong correlations were observed between emotional abuse and physical abuse, as well as between emotional neglect and physical abuse. Within the seven items of school ACEs, moderate correlations were observed between some items; however, no strong correlations were found. The correlation coefficient between the total score of ACEs and school ACEs was 0.41, indicating a moderate correlation.

4.2. Association with mental health and Hikikomori

Table 3 shows the association between moderate or higher levels of depressive/anxiety symptoms and ACEs and school ACE scores. In Model 1, a significant association was found between ACE scores and depressive/anxiety symptoms, with a 24% increased risk of depressive/anxiety for every one-point increase in the ACEs score. School ACE scores were also significantly associated with depressive/anxiety symptoms, with a 44% increased risk for every one-point increase in the school ACEs score.

In Model 2, the risk of depressive/anxiety symptoms was significantly increased in both ACE scores and school ACE scores for teacher-related and bullying-related factors. The potential confounding factors showed that age had a significant effect, with a decreased risk of depressive/anxiety symptoms with increasing age. Additionally, more favorable life circumstances were associated with a decreased risk of depressive/anxiety symptoms.

The association between Hikikomori and ACEs and school ACE scores is shown in Table 4. In Model 1, ACE scores did not show a significant association with Hikikomori, except for school ACE scores: a 29% increased risk of Hikikomori for every one-point increase in school ACE scores. In Model 2, both school ACE scores for teacher-related and bullying-related factors showed a significant association with Hikikomori; the rates for the increased risk were 23 and 37%, respectively. Additionally, having more family members increased the risk of Hikikomori. However, a higher academic background and more favorable life circumstances were associated with a decreased risk of Hikikomori.

Of the 1,436 who had at least one ACE, 1,027 (71.5%) had at least one school ACE. Of the 244 who had ≥4 ACEs, 211 (86.5%) had at least one school ACE. With both at least one ACE and at least one school ACE, the odds ratio for depressive/anxiety symptoms was 3.59 [95% confidence interval (CI): 3.00, 4.28; $p < 0.001$] and the odds ratio for Hikikomori was 2.48 (95% CI: 1.74, 3.53; $p < 0.001$) compared with those who did not have both. With both ≥4 ACEs and at least one school ACE, the odds ratio for depressive/anxiety symptoms was 4.38 (95% CI: 3.28, 5.86; $p < 0.001$) and the odds ratio for Hikikomori was 2.11 (95% CI: 1.22, 3.65; $p = 0.01$). The impact on depressive/anxiety symptoms is particularly pronounced when both adversity experiences are cumulative.

TABLE 2 Correlations among each item of ACEs and school ACEs.

	ACEs											School ACEs							
												Teacher-related items					Bullying-related items		
	1	2	3	4	5	6	7	8	9	10	Total	11	12	13	14	15	16	17	Total
1. Emotional abuse	1																		
2. Physical abuse	0.54	1																	
3. Sexual abuse	0.18	0.16	1																
4. Emotional neglect	0.50	0.35	0.17	1															
5. Physical neglect	0.30	0.28	0.19	0.30	1														
6. Divorce	0.17	0.17	0.10	0.12	0.11	1													
7. Mother treated violently	0.38	0.50	0.13	0.23	0.27	0.15	1												
8. Substance abuse	0.26	0.20	0.19	0.22	0.22	0.16	0.22	1											
9. Mental illness	0.22	0.20	0.13	0.20	0.18	0.13	0.21	0.22	1										
10. Incarcerated relative	0.11	0.13	0.16	0.07	0.18	0.09	0.10	0.18	0.13	1									
Total score of ACEs	0.73	0.69	0.39	0.63	0.49	0.49	0.59	0.49	0.48	0.27	1								
11. Emotional abuse in school	0.26	0.23	0.13	0.22	0.22	0.05	0.20	0.09	0.13	0.12	0.30	1							
12. Physical abuse in school	0.14	0.21	0.10	0.12	0.13	0.04	0.18	0.08	0.08	0.14	0.21	0.33	1						
13. Sexual abuse in school	0.11	0.11	0.26	0.06	0.20	0.06	0.13	0.15	0.09	0.23	0.22	0.20	0.20	1					
14. Emotional neglect in school	0.25	0.16	0.14	0.31	0.15	0.08	0.13	0.11	0.12	0.06	0.29	0.37	0.18	0.13	1				
15. Friends treated violently in school	0.18	0.20	0.09	0.18	0.19	0.05	0.17	0.10	0.12	0.13	0.25	0.37	0.33	0.20	0.24	1			
16. Bullying victimization by classmates	0.20	0.18	0.08	0.18	0.10	0.06	0.11	0.09	0.12	0.03	0.22	0.19	0.09	0.03	0.20	0.18	1		
17. Bullying victimization by senior students	0.15	0.14	0.08	0.15	0.11	0.04	0.10	0.09	0.11	0.08	0.19	0.24	0.18	0.05	0.17	0.20	0.32	1	
Total score of school ACEs	0.33	0.30	0.18	0.32	0.24	0.10	0.24	0.16	0.19	0.15	0.41	0.63	0.46	0.28	0.60	0.55	0.68	0.62	1

Light gray indicates a moderate correlation, while dark gray indicates strong correlation.

TABLE 3 Effects of ACE and school ACE on depression/anxiety.

	Model 1	Model 2
	OR (95% CI)	OR (95% CI)
ACE total score	1.24 (1.17, 1.32)*	1.25 (1.17, 1.33)*
School ACE total score	1.44 (1.39, 1.55)*	
School ACE teacher-related score		1.33 (1.19, 1.49)*
School ACE bullying-related score		1.60 (1.40, 1.83)*
Sex (female)	1.07 (0.89, 1.29)	1.07 (0.89, 1.29)
Age	0.97 (0.95, 0.99)*	0.97 (0.95, 0.99)*
Academic background	0.94 (0.88, 1.01)	0.94 (0.88, 1.01)
Life circumstances	0.68 (0.64, 0.73)*	0.68 (0.64, 0.73)*
Number of family members	1.03 (0.96, 1.10)	1.03 (0.96, 1.10)

OR, odds ratio; CI, confidence interval; $^{*}p < 0.05$.
The total score of the school ACEs was used in Model 1, and it was divided into teacher-related and bullying-related scores in Model 2.

TABLE 4 Effects of ACE and school ACE on Hikikomori.

	Model 1	Model 2
	OR (95% CI)	OR (95% CI)
ACE total score	1.01 (0.89, 1.13)	1.01 (0.90, 1.14)
School ACE total score	1.29 (1.13, 1.47)*	
School ACE teacher-related score		1.23 (1.01, 1.51)*
School ACE bullying-related score		1.37 (1.06, 1.78)*
Sex (female)	0.84 (0.58, 1.21)	0.83 (0.58, 1.21)
Age	0.99 (0.95, 1.03)	0.99 (0.95, 1.03)
Academic background	0.65 (0.57, 0.74)	0.65 (0.57, 0.74)
Life circumstances	0.65 (0.58, 0.74)	0.65 (0.58, 0.74)
Number of family members	1.14 (1.01, 1.30)*	1.14 (1.01, 1.29)*

OR, odds ratio; CI, confidence interval; $^{*}p < 0.05$.
The total score of the school ACEs was used in Model 1, and it was divided into teacher-related and bullying-related scores in Model 2.

5. Discussion

The present study is one of the first few studies to integrate adverse school experiences within the ACEs framework (as school ACEs), and examine their prevalence, as well as their impact on mental health and adjustment in adulthood. Importantly, the prevalence of school ACEs was much higher than traditional ACEs, suggesting that many people have more adverse childhood experiences in school than at home. The findings of this study also reveal that school ACEs are linked to both declining mental health in adulthood and Hikikomori. Regarding the latter, no association was found in traditional ACEs such as abuse at home or family dysfunction, suggesting that school ACEs play a crucial role as contributing factors.

As responsible members of society, recognizing the significance of schools inflicting harm on children and the subsequent negative effects that persist into adulthood is necessary.

5.1. Descriptive statistics

The prevalence of school ACEs is higher in Japan than the previously reported prevalence rates of extra-familial (including ACEs at school) ACEs (22). Several factors may contribute to this difference. First, the previous study sample was limited to university students, whereas our study included a broader range of participants. Second, while the previous study examined physical violence from teachers and negative perceptions as extra-familial ACEs, our proposed concept of school ACEs encompasses additional aspects such as emotional neglect and witnessing friends being treated violently by teachers. Notably, emotional neglect has the highest prevalence within the school ACEs teacher-related items. Both emotional neglect and witnessing violence toward friends indicate an overall unsafe school environment for children, even if they have not personally experienced direct harm. These findings suggest that children can be emotionally harmed by the actions of teachers, regardless of whether they have experienced direct victimization.

The rate of bullying victimization by classmates was 48.1% and that by upper classmen was 15.8%. This is higher than the bullying victimization rate of 33.6% in Osuka et al.'s (54) survey of Japanese elementary and junior high school students. However, this can be attributed to the fact that Osuka et al.'s survey directly targeted elementary and junior high school students and only covered bullying victimization in the 3 months prior to the survey, while this survey targeted bullying victimization during the entire period before the age of 18 in adulthood.

In this survey, the prevalence of Hikikomori was 3.5%. Although simple comparisons cannot be made because of the different ages targeted, considering that the prevalence of Hikikomori reported by the Cabinet Office of Japan (39) was 2.05%, the result was 1.7-fold higher. However, another survey conducted in Tokyo reported the prevalence of Hikikomori as 4.39% (55). The prevalence of this study was closer to the latter.

The proportion of those who scored at least one ACE in this survey was 35.9%, which is similar to the results of previous studies on Asian populations, as well as previous studies in Japan (30–40%). This indicates that this survey, despite being conducted online, accurately captured the situation of ACEs in Japanese people.

The strong correlations found for some of the ACE items may indicate that multiple adverse experiences tend to overlap because the perpetrator is the same person, while the only moderate correlations among the school ACE items may have been related to the fact that teachers and classmates change every year. Nevertheless, the moderate correlations still suggest that multiple adversity experiences are more likely to be cumulative within a school, and the impact of cumulative experiences needs to be further examined.

5.2. Association with mental health

Our findings provide evidence supporting the association between ACEs and mental health problems in adulthood, which is consistent with previous research (1, 3, 4, 12, 46). The results also support the hypothesis of our study, whereby school ACEs are related to mental health problems in adulthood. Previous research has emphasized the significance of adverse experiences outside the home (22), indicating the need to expand the concept of ACEs (20). Moreover, there is supporting evidence linking bullying victimization and mental health problems in adulthood (24, 56).

Additionally, ACEs can also affect academic performance (33) and behavioral problems (57, 58), rendering children with ACEs more susceptible to teacher reprimands and bullying. Thus, considering the possibility that ACEs may play a role in the association between school-related ACEs and mental health deterioration in adulthood is necessary. It should be emphasized that regardless of the reasons, it is unacceptable for schools, whose responsibility is to safeguard children's wellbeing and enable their development, to inflict hurt and contribute to mental health problems. Therefore, a reevaluation of the behavior of teachers toward their students within school settings is therefore warranted.

5.3. Association with Hikikomori

The novel and significant finding of this study is that school ACEs have a strong association with Hikikomori, whereas ACEs within the family do not. This has not been previously reported in the literature. It highlights how schools, which should protect children's development, can actually cause harm and profoundly impact their social adaptation.

Individuals with Hikikomori have difficulty with social participation (36) and tend to feel safe at home (38). Thus, it is plausible that experiences at school (school ACEs), where students interact with society, have a substantial impact compared to ACEs within the family. Kato et al. proposed a model for the development of Hikikomori, in which the school environment triggers bullying victimization or scolding by teachers, leading to avoidance behavior (37). The influence of the family environment is reflected in personality traits and individual characteristics, but it is not considered a causative factor for Hikikomori. In this survey, conventional ACEs were not associated with Hikikomori, while school ACEs were, supporting this model.

The findings of this study reveal that experiences of bullying victimization and mistreatment by teachers increase the risk of mental health problems and further contribute to the risk of Hikikomori, thereby hampering social participation. These experiences have significant societal implications; it has the potential to add to societal burden. Kato et al. have previously highlighted the importance of addressing bullying victimization and mistreatment by teachers (37), which are now specifically identified as school ACEs. Further research and interventions are warranted to address these issues. Recent research suggests that mistreatment within schools may be a significant factor contributing to school refusal in Japan (59). A survey conducted by the Cabinet Office of Japan in 2020 listed school refusal as a prominent cause of Hikikomori (60). These findings, together with the results of this study, imply that school ACEs are associated with Hikikomori through the intermediary factor of school refusal.

In the current trend of ACE research, efforts are being exerted to identify protective factors that can mitigate the risks associated with ACEs in adulthood. For example, the framework of health outcomes of positive experiences was introduced by Sege and Harper Browne (61), which suggests that positive childhood experiences can help alleviate the impact of ACEs. Bethell et al. (62) identified positive childhood experiences (PCEs) as protective factors that work in adulthood, such as safe family environments and friendships, and seven items were established. They demonstrated that these protective factors work even for those with high ACE scores. Robles et al. (63) identified three protective factors related to the family and four related to the community that counteract the negative impact of ACEs on academic performance in school. Therefore, it is imperative for us to address school ACEs while also identifying school PCEs. Such research holds the potential to ultimately prevent societal issues such as school refusal, youth unemployment, and poverty.

6. Limitations

This study has several limitations. First, this study was conducted on a web-based survey, recruiting a total of 4,000 participants of different sexes and age groups. As the survey was conducted among survey collaborators owned by private companies, it is likely that a bias exists toward individuals who are "willing to participate in such surveys" and are "internet-friendly". However, it can be concluded that this study reflects the overall situation to a certain extent considering that web-based surveys have become prevalent and the descriptive statistics reveal no significant difference from previous survey results. Second, this survey focuses on ACEs before the age of 18, including school ACEs, which may be affected by recall bias. However, given the lack of consistently strong correlations between ACEs and school ACEs, it suggests that participants are likely differentiating and providing distinct responses for ACEs and school ACEs. Third, this survey did not measure neurodevelopmental conditions, such as autism spectrum disorder or attention deficit hyperactivity disorder, which are associated with a higher risk of bullying victimization (64, 65) and ACEs (66). Furthermore, while Hikikomori has been associated with schizophrenia, social anxiety disorder, personality disorders, and depression (36), this survey did not collect information on such mental illnesses. Therefore, the influence of neurodevelopmental and psychiatric conditions on the results of this survey remains unclear. Fourth, we included two types of bullying-related items in school ACEs: one item related to bullying victimization by classmates and the item related to bullying victimization by upperclassmen. However, these may not be considered separate experiences. Rather, it would have been more appropriate to ask about the types of bullying, such as physical, psychological, and sexual bullying, in conjunction with the teacher-related items. Future studies will need to further examine the school ACE items. Finally, this survey was conducted in 2021 during the COVID-19 pandemic, which had a significant impact on social and economic life. Therefore, COVID-19 might have affected the mental health and social withdrawal outcomes of this survey, and this confounding factor should be highlighted.

7. Conclusions

This study provides a new concept of school ACEs, which is an extension of the conventional ACEs framework. The results indicate that school ACEs may have a more serious impact on social participation than conventional ACEs. This highlights the increasing importance of schools, as places for children's social participation, in providing a safe and secure environment to ensure children's healthy development and promotion of their wellbeing.

By situating negative experiences in schools within the context of ACE research and comparing them with traditional ACEs, this study has elucidated their severity and nature. Positive experiences in schools, such as active engagement with school, school climate, and achievements, are known to have a positive impact on the future but have not been integrated into PCEs research.

Through the examination of school experiences within the framework of ACEs and PCEs research, the role of schools in the future society becomes apparent. This insight can help clarify what we should or should not provide to children. Given the substantial influence of schools on children's lives, it is essential to further explore the complexity of school ACEs while simultaneously uncovering the realm of school PCEs. Consequently, comprehensive research on school ACEs, coupled with an in-depth investigation of school PCEs in the future, is imperative.

Data availability statement

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

Ethics statement

The studies involving humans were approved by the Hirosaki University Ethics 205 Committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

MW: Conceptualization, Data curation, Funding acquisition, Investigation, Project administration, Writing—original draft, Writing—review & editing. TN: Data curation, Formal analysis, Investigation, Methodology, Validation, Writing—review & editing. YO: Writing—review & editing. NT: Writing—review & editing. MT: Writing—review & editing. MA: Writing—review & editing. TS: Writing—review & editing. TK: Writing—review & editing.

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Conflict of interest

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

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1277766/full#supplementary-material

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A conceptual analysis of system responses to the issue of problematic sexual behaviors in children and recommendations for future work in Children's Advocacy Center multidisciplinary teams

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Problematic sexual behavior (PSB) in children is a common, yet frequently misunderstood and mishandled issue facing communities. Because of the intersection of children both causing harm and being harmed, societies across the globe struggle with whether to punish or support during these times. For Children's Advocacy Centers (CACs), whose mandate it is to support victimized children, this tension is exacerbated. CACs have historically relied on identifying a "perpetrator" and "victim" when providing their services, however PSB displaying youth do not fit this classic dichotomy. Compared with other children, PSB displaying youth are more likely to experience greater incidents and types of violent childhood trauma, have increased parent instability, decreased familial support, and struggle with co-occurring mental health diagnoses. Due to the stigma and fear surrounding sexual behaviors in children and systemic barriers including varied definitions of PSB, uncertainty regarding how to respond within the context of child-serving roles, and the criminalization of children's behaviors, access to supportive services is complicated and challenging. Treatment completion rates for this population are as low as 13%, despite most methods being short-term, non-invasive, and community based. This conceptual analysis paper identifies five key themes in the literature that influence these barriers and proposes an interdisciplinary approach for CAC multidisciplinary teams (MDTs) to better support this vulnerable population.

KEYWORDS

problematic sexual behavior, Children's Advocacy Center, multidisciplinary teams, interdisciplinary teams, liberation health model, community response

Introduction

Sexuality and sexual behaviors are common and expected aspects of child development. However, the diverse ways and contexts in which these behaviors are displayed, coupled with the stigma and bias surrounding these variables, make defining what is "typical" versus "problematic" challenging (1–3). Problematic sexual behavior (PSB) is generally defined as a behavior displayed by children or youth that involves sexual body parts or acts, is outside their expected developmental stage, and causes harm to self or others (2, 4). These behaviors tend to be minimally responsive to adult redirection, involve negative emotionality such as feelings of fear, shame, or anger, occur between youth of disparate ages, sizes, or abilities, and can use force or coercion to involve other children in the behavior (4, 5). It is helpful, therefore, to consider sexual behavior in children along a continuum.

Some studies have found that as many as 80% of youth will engage in some form of sexualized play or interaction with a similarly aged peer by the time they reach adolescence (2). It is also estimated that approximately 25% of cases referred to Children's Advocacy Centers (CACs), and over one third of all cases referred to law enforcement, for concerns of sexual harm or misconduct involve youth acting out against other youth (3, 6–8). Given the tension between the commonality of children engaging in sexual behaviors during childhood and the serious implications of being labeled as causing sexual harm, it is important that professionals and communities critically evaluate these behaviors and assess how best to respond.

Most PSB responses are siloed into either the legal or mental health systems, with little regard for the family's perspective in this process (9, 10). However, problems arise when these cases are not approached from a more holistic perspective. As few as 13% of youth referred for PSB treatment ever complete their intervention, despite recidivism rates for short-term PSB treatment being as low as 2% (3, 5, 11, 12). Drawing from the liberation health framework, this discrepancy is likely reflective of issues related to historical oppression and stigma driving siloed PSB responses (13, 14). Rather than communities critically analyzing their beliefs around sexuality, reflecting on how this impacts their perceptions around addressing PSB in children, and emphasizing their strengths as a diverse and interconnected system, they continue to draw from much of the same flawed and limited perspectives.

This paper seeks to address this issue. The authors begin with a review of the historical ways in which communities have responded to PSB to provide a better contextual framework. They then discuss five key concepts identified in the literature which have facilitated harmful or ineffective practice. Lastly, the authors offer considerations for future PSB response in communities, highlighting the unique role of CACs as a critical, interdisciplinary team that is well-positioned to address and respond to this complex issue.

Note: Throughout this paper, readers will observe that the authors use the term "parent" rather than "caregiver." Drawing from the lived experience of one of the authors, while "caregiver" is often viewed as a more inclusive term for the various ways an adult can care for and raise a child, it can also be experienced as a way of othering and distancing non-biological parent–child relationships. Thus, the authors have opted to use the term "parent" to describe any person in a parenting role with a child. This includes, but is not limited to, biological parents, grandparents, aunts/uncles, and foster parents.

A review of historical responses to PSB

Examining the historical context of PSB response is vital to ethical and clinically sound practice. Although helping professionals intend to do no harm, the reality is that providing help for complex and potentially stigmatizing issues is difficult to do. Intersections of belief systems, identity, systemic oppression, and vulnerability all intertwine with help and harm, and helping professionals must critically reflect on how they contribute to this process (14–16). It is important to recognize that, historically, PSB concerns were influenced by white supremacy, homophobia, heterosexism, and firm gender binaries. Professionals and teams must recognize the impacts of these origins and how communities conceptualize this issue so they can avoid inadvertently continuing it (13, 17–21).

1940s–1950s

Studies involving sexual behaviors in children first emerged in professional literature in the 1940s. "Appropriate" sexuality was defined almost exclusively by dominant western European, Christian values (13, 21–23). People who displayed overt sexual behaviors, were attracted to individuals of the same gender, or dressed or acted outside of ascribed gender norms were labeled as deviants who were too "ill" or "dangerous" to live in society (18, 22, 24). This belief was furthered by the first publication of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM) in 1952, which labeled homosexuality as a form of "Sexual deviation" and placed it under the same diagnostic category of as "transvestism, pedophilia, fetishism and sexual sadism (including rape, sexual assault, mutilation)" [(25), p. 39; (26)]. Sexuality and diversity were both seen as threatening to the safety and morality of a community, and those who did not align with what was set forth by society were subjected to harmful treatments, institutionalization, and even criminal charges (1, 22, 27).

1960s–1970s

During the 1960s and 1970s there was a pivotal societal change that allowed the diverse spectrum of sexuality to be considered. Writings like the 'Kinsey Report' argued that all people—including women and children—experience a range of sexual behaviors and experiences, and professionals were challenged to better define what constituted a "typical" versus "concerning" behavior (27–30). With this growing understanding that children were sexual beings, fear and questions arose about the connection between PSB and adults who sexually harm (24, 30, 31). However, rather than addressing these questions, communities responded by either institutionalizing youth displaying PSB for indefinite periods of time or ignoring the issue altogether in hopes that such behaviors were a "phase" the child would grow out of (24, 32). There was little thought or regard for the long-term implications of this response, and present day research shows that this type of practice ultimately placed youth at greater risk of both PSB and future harm (6, 33).

1980s–1990s

Due to the long history of the punitive response to PSB through the legal system, families were increasingly fearful and wary of seeking help from professionals (34, 35). Youth with PSB were labeled as "super-predators," which resulted in stigmatization of this population (1, 7, 36). This made it difficult for researchers and practitioners to obtain accurate data to guide their decision making (34, 37). Rather than having reliable, longitudinal data from the children and families struggling with PSB to inform care, professionals were forced to rely on arrest and court records that were severely limited and inaccurate (35).

Professionals came together for the first time in 1987 to address these challenges, and they created a national task force to address PSB in children (38). The result was a unanimous call for better research and a more structured response to support treatment efforts (35, 37). With improved assessment and response tools, clinicians and advocates for this population could begin to identify risk factors for PSB and collect more accurate data to support their claim that treatment with children could be successful (34, 35, 39).

2000s

Although the history of PSB was marked by fear that these youth were doomed to become adults who caused harm, the research of the 2000s presented a very different picture. It was demonstrated that youth who struggled with PSB were more likely to be victims of violence themselves and to have co-occurring difficulties in the areas of emotion regulation, social skills, and other mental health diagnoses (6, 40, 41). Furthermore, the types of sexual behaviors displayed were vastly diverse, which meant that addressing the child's needs and treatment responses had to be individualized to the child's unique context (2, 36, 42). Treatment that specifically engaged the family unit was shown to have positive results, with recidivism rates ranging between 2 and 10% (40, 43, 44).

Present day PSB response and the role of the CAC

Professionals and communities continue to work to address PSB in a variety of ways. Because research emphasizes the importance of responding to PSB in a clear, unified, family-centered manner, CACs are increasingly identified as a logical entity to facilitate this work (3, 10, 45). There are over 1,100 CACs around the world designed to keep children safe and centered within an MDT response (10, 46, 47). Teams of professionals, including law enforcement, District Attorney staff, child welfare workers, medical providers, and mental health professionals, all come together to ensure that the needs of vulnerable children are met in a way that does not cause further trauma or confusion. However, one of the challenges in addressing PSB concerns in this setting is that CACs were developed from a "victim/perpetrator" framework. Youth with PSB do not fit into this classic duality, and therefore, a different approach is needed.

Recent international CAC PSB research shows that teams often feel they are being "disloyal" to the child who is the recipient (e.g., the "victim") of the PSB behavior if they provide support to the displayer of the behavior (e.g., the "perpetrator") (3, 45). However, attempting to classify the children in this way can do more harm than good. PSB in children is a very different issue from child sexual abuse perpetrated by an adult, and children who struggle with PSB respond well to education, positive relationships, behavioral modifications, and treatment (48–50). Given that a great deal of PSB occurs within family units, typical responses of simply separating the children is not only difficult to do, but it often causes additional harm for both youth because the child who is the recipient of the PSB often feels a sense of guilt and loss over their sibling's removal (3, 45, 50).

Recent literature supports the ways in which CACs can address these gaps. In 2020, Sites and Widdifield published a white paper report titled, 'Children with Problematic Sexual Behavior: Recommendations for the Multidisciplinary Team and Children's Advocacy Center Response' (10). In this report, the authors highlight the strengths of the CAC model and how multidisciplinary teams can support all children involved in issues of PSB. They suggest that, with small changes to pre-existing protocols, increased mental health provider involvement, and inclusion of families in the conversation, CACs can continue their work of supporting all vulnerable children (3, 10).

Key concepts

Throughout the authors' cumulative experience working with families, they repeatedly identified several challenges when addressing PSB. To better understand this phenomenon, they conducted an extensive review of historical and present day literature, ranging from 1943 to 2023. This served to validate the authors' observations, and the authors subsequently categorized these reoccurring themes into five main concepts: Difficulty defining PSB, use of short-sighted safety responses, disregard for the intersection of PSB with other needs, lack of parent involvement and engagement, and siloed responses. It is important to note that these identified gaps and challenges, while difficult, also provide opportunities for CAC MDTs to improve service delivery to this special population.

Difficulty defining PSB

Attempting to define problematic or harmful sexual behavior is a difficult task. Although it is widely accepted that PSB is a sexual behavior displayed by a child that is outside their expected developmental trajectory and causes harm to self or others, there are a multitude of nuances that make defining this problem challenging (17, 51). Factors like whether the displaying child has reached puberty, the age of the other child(ren) impacted by the behavior, the type of behavior displayed, any use of additional forms of violence, and parent attunement and response (2). Furthermore, adults tasked with protecting children often struggle with their own emotions and sense of safety related to childhood sexuality and sexual behaviors, which contributes to the challenge of establishing a clear and unbiased definition of PSB (1, 9, 42).

Despite these complexities, one of the most common ways PSB is categorized is by distinguishing between whether the youth displaying the behavior is categorized as a "child" (a youth under the age of 12 years) or an "adolescent" (12 years or older) (3, 17, 51). The impact of puberty, sexual gratification, and the desire for sexual relationships are important influencing factors in both the displaying of PSB and its treatment in adolescence (2, 33, 50). Societal factors, including the role of the legal system and risk of prosecution are also key influencers behind the push to use age as a component of assessment (3, 52).

Studies have found that a state or country's legal age of consent and/or prosecution influence, if not determine, whether support is given to a child (3, 7, 52). Although age allows for clearer definitions in matters of the law, communities are cautioned against placing too much weight on this one factor when assessing level of risk or severity related to PSB (33, 51). Instead, communities are encouraged to think more critically about PSB and to use individualized, developmentally based assessment tools that can provide context to the behavior and the child (9, 42, 43). This not only provides teams with a deeper understanding of the factors influencing the development of PSB, but also provides valuable insight in how to address and respond to it.

Use of short-sighted safety responses

Historically, most PSB interventions focused on physically separating the displaying child from their families and communities. Because these youth were viewed as a potential threat to society and to other children, heavy emphasis was placed on ensuring safety in the most clear-cut way possible (13, 19, 36, 53). This included removing the child from their home environment and placing them in juvenile justice or residential treatment facilities, and/or placing them on sex offender registries. However, there are long-lasting and potentially devastating consequences to this approach.

Research demonstrates that use of sex offender registries and "dishwasher" treatment—or treatment where a youth is removed from their environment to be "fixed" and returned to their original environment upon "completion"—is not only ineffective but harmful (1, 6, 19, 53, 54). Placing children in residential facilities that have higher concentrations of youth struggling with PSB and other significant needs, and decreased access to parental supports and connections, puts them at higher risk of both future victimization and continued displays of PSB (6, 33, 55). While some children require intensive, inpatient treatment to address highly intrusive and violent displays of PSB, most youth do not fall into this category. Most youth respond well to short-term, community-based therapy that includes and supports parents in the safety planning and behavior modification process (10, 17, 50). Additionally, because of the high level of success with therapy, placing youth on sex offender registries which follow them well into adulthood has been found to do little more than exacerbate challenges for the youth and their family (19, 36, 53).

Conversely, working with the family to assess and respond to PSB concerns allows teams to better understand the concerns and behaviors within the context of the child's environment and to address them in a more timely and effective manner (44, 48, 50). This approach also provides insight into the family's protective factors, which can be used to further support the child and ensure that safety needs are met. Because PSB is frequently rooted in trauma and relationship difficulties, working to support the child within the context of relationships has immense value (45, 48). Therefore, when teams decide to isolate children from, or even within, their environment, they must critically examine the implications of such a choice. While this may provide an immediate, short-term solution to PSB in the community, research suggests it does little to support long-term safety and healing for families when done outside of a comprehensive and developmentally sensitive manner (7, 36, 53).

Disregard for the intersection of PSB with other needs

Children who struggle with PSB often have additional intersecting needs that make supporting them both important and challenging. One of the earliest intersections identified in the literature is the connection between PSB and prior victimization (2, 17). While it is important to note that upwards of 95% of children who have experienced sexual abuse do not go on to display PSB, youth who display PSB are significantly more likely to be victimized in this way (6, 9). Furthermore, research suggests that a child's risk for PSB increases with the number and types of victimizations a child experiences—particularly when violence is involved, as with physical abuse and domestic violence (33, 52).

Youth placed in foster care or congregate care settings, or who are involved with the juvenile justice system, are also at greater risk for both displaying PSB and being impacted by the PSB of other youth (6, 9). One possible reason for this is that youth in care are less likely to benefit from protective factors like parent connection and guidance. Attuned parents can both alleviate trauma symptoms and provide supervision when concerning behaviors are identified, both of which are key in addressing and preventing PSB (2, 33). Thus, the compounding factor of early childhood trauma and reduced parental protection and support increases the likelihood of PSB in children (6, 33).

Youth with PSB are also more likely to struggle with co-occurring behavioral and mental health difficulties. Issues related to social and emotional awareness, impulse control, and self-regulation commonly intersect with PSB (2, 9, 17). Therefore, treatment responses must be comprehensive in their approach (10, 43). They should include a combination of psycho-sexual education, social skill building, self-regulation techniques, and trauma processing (33, 50, 51). Likewise, responses that are interdisciplinary in nature—spanning the boundaries of family members, educators, mental health providers, and legal and medical systems—have also been found to be beneficial in addressing the complexities of this population (3, 7, 12).

Lack of parent involvement and engagement

The impact of parents on PSB is well documented in the literature. Parent involvement and responsiveness has been found to be one of the key protective factors in both the development of PSB, as well as in promoting successful treatment outcomes (12, 48, 51). Therapeutic responses that include parent skill building around behavior management, boundary setting, and communication were found to be among the highest predictors of successful treatment outcomes (2, 3, 44, 50). This suggests that empowering parents in their ability to both address the behaviors and improve their relationship with their child are mutually beneficial to addressing the problem of PSB.

However, psychosocial influences of fear, stigma, guilt, and generational trauma have powerful influence over a parent's receptiveness to discussions of PSB and safety planning (12, 48). Because PSB involves harm to a child, a child welfare report and District Attorney referral are often made following any disclosure or discovery of PSB (3, 10, 45). The report filed to the child welfare office is typically documented as a concern of 'parental neglect', and the referral to the District Attorney is typically for allegations of 'child sexual abuse.' Although there are important reasons behind these protocols, including the need to ensure the safety of children and to connect families with emergency assessments and support, this experience frequently leaves families feeling far from supported or empowered.

Parents often report feeling judged, confused, powerless, and isolated following a system response to PSB (12, 45, 48). Rather than families finding the support and clarity they need from their interactions with child-serving professionals, they frequently experience these agencies as a threat to their family and to their child's future safety. This results in increased defensiveness and resistance to engagement and provides valuable insight into recent research findings which demonstrate that treatment completion rates for this population are as low as 13%, despite even lower recidivism rates (11, 12).

One way the literature suggests addressing these challenges is by consciously partnering with parents and including them in the PSB response process (2, 48). This allows parents to better understand the issue of PSB and provides them with tools to address it (10, 50). This approach also increases trust between the families and the professionals positioned to help them. Community multidisciplinary teams should draw from the growing body of literature which has demonstrated PSB treatment to be highly successful, and therefore can provide families with a sense of hope for the future. Because of the level of stigma continuing to surround issues of PSB and child sexual behaviors, teams must be willing to explore a family's fears and challenges in order to build an open, trusting, and collaborative working relationship.

Siloed responses

Because PSB is a unique and complex issue, a diverse group of perspectives is required to address it. As early as the 1960's, PSB practitioners have leaned on the resources and skills of their colleagues in other fields to help support youth who display PSB with great success (24). For example, one single outpatient therapist could not feasibly provide weekly treatment, assess safety in the home, and ensure families followed through on all recommendations put forth by the courts. However, in partnering with local probation officers, the therapist could remain in the role of mental health practitioner, knowing that various aspects of the family's needs were being met by other professionals. Furthermore, in working collaboratively, the therapist received vital information from the probation officer about how the family was doing and whether progress was being made. This benefit was reciprocal in nature, as the probation officer also benefited from the therapist's clinical opinion regarding the child's progress in treatment. Thus, children and families were better served through this collaborative and integrated approach.

CACs are a prime example of this collaborative approach. CAC MDTs meet regularly to ensure that all team members working with a family have the same information regarding concerns identified, steps taken to ensure safety, and next steps needed to support the child (10, 45). While this is undoubtedly beneficial in ensuring clear communication between partnering teams and systems, there are limitations to this way of practice. MDTs maintain distinct boundaries around their roles and communication with one another. Each discipline speaks to their own work, and typically has unique goals and agendas related to their professional role in a child's case (56). For example, law enforcement and District Attorney team members focus on upholding their role as investigators and prosecutors of crimes against children, whereas mental health providers focus on their role in providing ongoing support and treatment. While each role has valuable contributions, in isolation, they do not accurately reflect the whole picture.

It is the intertwining of interdisciplinary perspectives that contributes to optimal outcomes. Rather than having separate goals amongst the MDT, interdisciplinary teams strive for a shared common goal and purpose (57, 58). While this has the potential to result in conflict and disagreements amongst team members, the interdisciplinary framework acknowledges this shared approach as a means of ensuring that issues are being addressed in a holistic manner (56, 59, 60). Rather than teams remaining siloed, with their own values and biases, they are pushed to deconstruct their ideas and see what help or hinderance they provide to the process and to the family.

To assist in this process, interdisciplinary practice emphasizes inclusion of the lived experience perspective. Lived experience offers teams invaluable insight into the issues that their clients face and bring to light any barriers or challenges that arise (9, 54, 61). It also helps teams to address the ongoing challenge regarding the stigma of PSB and the difficulty of families to trust and engage in the process (12, 48). Through access to people who have previously been through the process of PSB identification, response, and treatment, families can be reassured that healing is both real and possible.

Considerations for an initial support and stabilization response for CAC MDTs

In light of these five key concepts, the authors offer three perspectives for CAC MDT members to consider when responding to initial PSB concerns. Rooted in the liberation health framework, the authors seek to demonstrate how their own interdisciplinary collaboration has helped to support families and one another during a time of PSB response, which has been shown to be instrumental in determining whether families successfully engage in PSB community supports (10).

Drawing from the medical provider perspective, Child Abuse Pediatrician Dr. Sasha Svendsen suggests a role for the medical provider within the CAC initial response to assess the behavior in the context of typical childhood sexual development, which not only helps to decrease the stigma associated with this topic, but also allows for a more neutral space to explore and reinforce body safety, body autonomy, and healthy boundaries. Drawing from the social work perspective, clinical social worker Dr. Mary Harris discusses how CAC MDTs can develop on-going critical dialogue and reflection to improve awareness of the biases and silos impacting family engagement and successful PSB outcomes. And finally, drawing from the lived experience perspective, parent and peer support professional Diane Lanni shares her experience as a caregiver of multiple youth who have struggled with PSB and as someone who has engaged with child-serving systems to address it. Ms. Lanni discusses the power of humanizing the issue of PSB and including families in the response process.

Theoretical framework

The liberation health framework is a radical social justice theory that sees value in bringing groups of people together to tackle difficult problems (62–64). Rather than certified "professionals" being seen as the experts on a person or community's situation, the liberation health model posits that all people—particularly those with lived experience—have important knowledge to contribute to fully understand and explore issues (14, 15, 65). In working collaboratively, better solutions come about, which leads to a more just, equitable, and healthy society (62, 64, 66). In drawing from a group of diverse perspectives, the entirety of the problem can be better defined and addressed.

Reflective of social work's person-in-environment perspective, the liberation health framework acknowledges that the issues people face, as well as their strengths, occur within an important context (14, 64, 67). Although people are unique individuals, they exist within a larger historical and societal framework of intersecting identities and structural forces that impact their ability to succeed or struggle in life. Seeing issues in this way allows for recognition that inequities, oppression, and a variety of -isms have significant influences and far reaching impacts (15, 64, 68).

In the case of PSB, the liberation health model helps to frame things like the discrepancies between the high rates of incomplete treatment and low rates of recidivism following treatment as a symptom of larger structural and societal barriers. When powerful child welfare and protection systems exert their dominance by problematizing the child and using threats of removal and legal charges to force families to act, rather than acknowledging the influences of trauma, prejudice, and lack of community resources on PSB development, families cannot help but respond in fear and retreat (19, 64, 69). Rather than the family and system coming together to critically examine how PSB concerns came about, the groups become siloed into opposing camps. This ultimately negatively impacts both the discovery of the problem and its resolution because, according to the liberation health framework, there is a connection between the issues and the solutions (14, 63, 64).

The authors drew from the liberation health framework as a way of contextualizing the challenges of PSB and providing a way forward for communities. It was important to choose a theory which would not only provide insights into the challenges of PSB—a daily reality for the authors—but that the framework would provide tangible and unique solutions as well. Early influence of this theory on this paper can be found in the authors' decision to perform a chronological literature review. Liberation health purports that complex social justice issues are often rooted in historical oppression, and gaining an awareness of that fact is a crucial first step to understanding how to address it (15, 63, 66).

Because liberation health posits that issues and solutions go hand in hand, the authors also utilized this framework as a way of deconstructing the five key concepts/challenges identified in the literature and the authors' experiences to find solutions. If isolation and siloed power lead to issues of unjust PSB response and continued barriers to support and treatment, then the solution to such things, according to liberation health, is inclusion, collaboration, and empowerment (15, 63).

In the final section of this paper, the authors draw from this theoretical framework to provide "considerations" for future practice rather than "recommendations." The liberation health framework posits that a key feature of liberatory practice is that people and communities must be empowered to think and make choices for themselves and their own unique context (14, 15, 66, 68). This is also supported in the PSB literature. PSB and the families and children who struggle with it are diverse and have unique needs and strengths. A "one size fits all" model often does more harm than good. This framework allows the authors to protect against such things, while still pushing communities and professionals to take actionable steps toward change.

Considerations from the medical perspective

It is important to understand that sexual behavior occurs along a continuum, ranging from typical and expected to abusive and violent. When determining whether or not a sexual behavior is problematic, there are generally three factors to consider that can help to characterize the behavior along the spectrum. They are: (1) the frequency of the behavior, (2) developmental factors involved, and (3) the level of harm to all the children involved (2, 17). Furthermore, as previously noted, the relationship of the behavior within the context of the child's overall development and their environment must also be considered.

By (first) exploring this issue from the developmental perspective of the medical lens, this complex and often emotionally charged topic can be initially assessed in a more neutral, objective, and non-threatening way (2). Children and their parents have unique relationships with their medical providers, particularly if a medical home has been established for continued routine care. This is an ongoing, trusted relationship, in which providing anticipatory guidance to the parent about the child's growth and development is paramount. Consideration of the behavior within the context of typical, expected sexual development and health provides a de-stigmatized and family-centered approach, which allows for various aspects of the behavior to be discussed in order to fully understand the context of the behavior being displayed (2). This includes exploration of any sexual abuse or other trauma history, exposure to sexually explicit content, as well as various parenting or cultural practices. By approaching this topic from the medical perspective, medical professionals can help children and parents to understand that sexual development consists of more than just hair growth and other physical body changes. This conversation allows an important opportunity for the medical professional to introduce and reinforce the concepts of body autonomy, body safety, boundaries, and healthy relationships.

When a child's problematic sexualized behaviors are approached through a similar inquiry and triage process as other medical and developmental concerns, it allows space for honest dialogue and initial assessment, whereby the medical professional obtains initial information, asks clarifying questions, and ultimately decides on action plan based on immediate identified needs. By no means does this process replace the need for a more comprehensive assessment of the situation and treatment planning, it merely serves as an initial step to address and assess the concern at hand. It then requires coordinated follow-up with the community interdisciplinary team, where a more comprehensive assessment will be provided (10). By highlighting the role of the medical provider in the early triage process once a concern has been identified, the children who truly need the specialized, yet limited resources can be more accurately identified, as well as those children who do not. The impact of this first initial step would greatly impact the already overwhelmed and under-resourced mental health services available for children who display problematic sexualized behaviors (10).

Interactions with medical professionals, particularly during yearly well visits, has the potential to provide children and parents with a model for how to have conversations about body safety and body autonomy, which can then be reinforced between the parent and the child at home. These conversations represent a critical step in the early stages of the process, while the child and parent await the larger, more comprehensive assessment. This modeling of communication also serves to empower parents with an actionable step and provides them with a sense of agency while awaiting connection to longer-term services.

Initial supportive and non-punitive responses from medical professionals may help to overcome some of the barriers associated with this issue and will hopefully promote a sense of support and encouragement for parents to engage in additional communications with other interdisciplinary team members. The way in which the child-serving professionals of the interdisciplinary team frame and approach this issue with children and their parents is crucial for parent engagement in the larger process, which research has demonstrated to be the single most pivotal factor for a child to stop engaging in problematic sexual behavior and to support them to make more positive and healthy choices (10, 40). In all conversations with parents, this topic must be humanized and the context of the behavior must be considered. Everyone must understand that the behavior does not define the child or their future. By helping children and parents to understand that the behavior is the problem, and not that the child is the problem, children and families can develop resilience factors and promote a strengths-based approach to safer, more healthy choices in the future.

Considerations from the social work perspective

Critical reflection and dialogue are important steps to addressing complex issues as a helping professional. Despite many good intentions, research shows this is not enough (19, 36). Social workers and other helping professionals sit in places of power over vulnerable clients, especially in fields like child welfare, so careful work must be done to ensure this power is not used to cause further harm. Professionals need to be aware of the many intersecting influences that impact clients and the concerns they face. Issues like bias, stigma, oppression, and other injustices are often invisible at first look, but immensely impactful on the lives of these youth (1, 19).

Intersections of children both causing and experiencing harm also leave many adults, both professional and not, feeling frightened and unsure where to turn. It is important for teams to critically examine why sexual behaviors in children trigger such intense reactions. Despite the relatively common experience of sexual play amongst children, and the knowledge that children respond well to clear and consistent designations of body and relationship boundaries, adults continue to struggle with their own perceptions and beliefs around what constitutes "expected" and "safe" behavior. This is problematic when thinking about PSB response because it places children at risk of being overly or erroneously labeled as having a problem, based on whether the behavior falls outside the professional's own experience or set of values (1, 13, 42).

One way teams can combat this is by leaning on one another for collaboration and discussion. CAC MDTs were designed to support children, but they can also provide immense support to the adults working within them. In having a space to discuss cases and safety plan, MDT members can dialogue about their concerns and experiences, and receive feedback. This give and take of perspectives provides teams with a robust knowledge and understanding of the issues facing their communities (56, 57, 60). It also protects against the centering of one dominant opinion over another and helps teams to see the various factors that have led to the concern. For issues of PSB, this means teams can see their clients in a more holistic way, and ensure their needs and experiences remain the focus of the MDT response rather than the beliefs of any one team member.

However, for this type of critical practice to take place, it is important that all people involved in the PSB response be represented in the conversation—including the families. Although this can be challenging for some professionals because systems of power often work behind the scenes and in silos as a way of protecting themselves and maintaining control, including families and other perspectives in the conversation is vital (57). Parents provide unique and invaluable insight into the child and the struggles they are facing. By including families in the conversation, teams obtain a much clearer picture of what is happening and how to proceed (44). Creating a space of open and honest dialogue reduces the risk that families—and teams—feel like they must defend themselves against one another—one of the most common barriers facing PSB response. In seeing that all team members add value to the conversation and have a common goal of achieving health, wellbeing, and safety for all children, teams can make great strides in the work of PSB response and change the trajectory for this special population.

Considerations from the lived experience perspective

PSB often occurs within the child's home but regardless of where the behavior occurred, parents often feel responsible and fear the judgment of society (3, 12, 70). "What will people think?" is a common worry. Parents may hear folks say, "they learned that somewhere" and worry that not only will their child be judged, but their entire family and parenting will be called into question. Fear and shame make it challenging for parents to reach out for the help they need for their child or children. When parents are the caregivers to both the displayer and recipient of PSB, there are additional emotions and a concern for how to support each child (3, 45). The emotional toll is heavy, and families need support in real time to prevent further emotional harm to all family members. Systems, understandably so, are focused on the immediate safety of children, but for parents, psychological safety is paramount. The basic questions of, "Is it safe to tell?" and "Who is a safe person to tell?," are at the forefront of most parents' minds.

Family engagement begins with creating an environment where everyone feels safe to be open with discussing what occurred and agreeing to a safety plan (10). Yet, are all members of a multidisciplinary team safe? For parents, they worry that accepting help is an admission of guilt. Seeking help is frightening, especially for marginalized individuals who may already feel distrust with systems. Even when families bravely try to seek help for their children, many do not know where to go. “Who do I call?” “How much do I tell?” “Do I begin with the police or the hospital or a therapist?” “Is someone going to show up on my doorstep and take my child away?”

As mentioned, the barriers to engagement with parents are many, yet we know that parent engagement is critical for successful outcomes for children (2, 12). PSB by its very nature is personal, and since the damage occurred in a relationship, healing must occur in the form of healthy relationships. Families must be empowered as the agents of change. All family members should be a part of the collaborative effort to understand what has happened and agree to an action plan. Considering the emotional vulnerability of parents, it is prudent to design the team engagement with attention to parent needs.

One way to facilitate the psychological safety of parents is the use of a peer support person. Family members are more likely to trust information from someone who has been in their shoes. Use of peer supports for parents does more than just support the parent. A peer support person can act as a critical link for information between team members. Peer supports act as “cultural brokers,” as they are comfortable navigating in both professional and familial settings (71, 72). They can often express the chief concerns of parents in a way that promotes understanding and reduces shame and stigma. Peers assume the role of an emotional container for the parent, allowing the parent to process their own thoughts and feelings, thereby giving space for needed safety planning and communication with other team members.

The time waiting for professional help can been painfully long for parents. Knowing there are actions steps to take while awaiting services can help reduce the stress and anxiety of that time period. Can my child stay at home? How can I prevent a recurrence of harm? Who do I reach out to when new household rules are broken? Should we talk about what happened or should we wait in silence for the professionals? How do I educate myself and others about how to handle PSB? Who needs to know? What do I do if there are more disclosures or behaviors? Parents may be afraid to ask many of these questions and will need an empathetic ear on the team to help them bravely ask for help. Resources should be readily available to give to caretakers when these questions arise. Parents need to know they are not alone while awaiting therapeutic services in a way that recognizes everyone involved, not just the impacted child.

Conclusion

Despite the issue of PSB being present in the literature for over 80 years, communities continue to struggle with many of its basic tenets. Defining what constitutes a behavior as problematic or harmful, effectively addressing the behavior, understanding the various intersecting factors that influence its development, including parents in the conversation, and valuing the power of interdisciplinary work, all coincide to make this a complex but important topic to consider and deconstruct. The liberation health framework offers a way to understanding this issue and to address it in a more holistic, inclusive, and socially just manner.

Through use of the liberation health framework, the authors critically examined the historical context of PSB and identified five key concepts and challenges communities face when attempting to address this issue. These challenges were then deconstructed and used to identify opportunities for change. The authors then offered their three, unique perspectives on how CAC MDTs can address this issue in their own communities. Reflective of the liberation health perspective, the goal of this paper was not to provide a concrete response model to be replicated by all. Rather, the authors focused on empowering communities and interdisciplinary professionals to find their own, unique way of responding to PSB concerns. In working in this way, the authors sought to ensure the issues of the past would come to light, be addressed, and result in long-term healing and thriving for all families.

Author contributions

MH: Writing – original draft, Writing – review & editing. DL: Writing – original draft, Writing – review & editing. SS: Writing – original draft, Writing – review & editing.

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Conflict of interest

Publisher’s note

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A descriptive analysis of children seeking medical attention for problematic sexualized behavior

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Introduction: Problematic Sexualized Behavior (PSB) in children is an increasingly prevalent and often misunderstood issue that impacts the well-being of children. Quantifying the numbers of affected children is challenging due to inconsistencies in how these children are identified, as well as misperceptions surrounding the issue and lack of a coordinated community response.

Methods: In this single-center retrospective case review, we attempt to report data on child demographics and case characteristics for children presenting to one Child Protection Program (CPP) medical specialty team for concern of PSB.

Results: A total of 224 children were identified as having engaged in PSB during the study period. 110 (49%) of these children were referred to the CPP for medical evaluation and medically triaged by the team. The remaining 114 children (51%) were identified through the medical triage of the presenting cases as having engaged in PSB with the index children, but were not referred to CPP for medical care themselves. The majority of children who were referred (69%) were the recipient of the PSB, compared to being the displayer of the behavior (20%). Of the recipient cases, the child displaying PSB was also referred to the CPP only 14.5% of the time.

Discussion: These results highlight that the number of children presenting for medical evaluation with concern of PSB is a significant underestimation of the prevalence of PSB in the community. This notable gap in identification of children engaging in PSB prevents service delivery for these children, including medical evaluation. The results also demonstrate that children displaying PSB were disproportionately missing from care and represent a specific area of missed opportunity for intervention and support by medical professionals.

KEYWORDS

problematic sexualized behaviors, child abuse, sexual abuse, child protection, neglect

Introduction

Problematic Sexualized Behavior (PSB) in children is an increasingly prevalent and often misunderstood issue that impacts the well-being of children. There is no single sexual behavior that has been identified as pathognomonic for a sexual behavior problem, thus making recognition and understanding of PSB challenging (1). It is important to consider several characteristics about the behavior when distinguishing between typical vs. problematic sexual behavior, including the frequency of the behavior, the child's developmental stage, and the level of harm involved (2). PSB is commonly defined as behaviors in children ages 12 years and younger that involve sexual body parts, are developmentally inappropriate, and may be harmful to themselves or others (2, 3).

Quantifying the number of affected children is challenging due to inconsistencies in how these children are identified (4). Estimates from Children's Advocacy Centers (CACs) across the

United States suggest that 20–25% of cases served by CACs involve youth acting out against another child (5). This number likely underestimates the total number of children impacted annually, as many are never brought to the attention of CACs or other community agencies. While evidence suggests that PSB occurs internationally, the available research is heavily concentrated in middle to high income countries, including the United States, United Kingdom, and Canada (6, 7).

The existing literature on PSB in children suggests that there may be risk factors associated with these behaviors. One common misperception is the assumption that all children displaying PSB have been sexually abused in the past. While sexual abuse has been shown to be a risk factor for displaying PSB, the prevalence of prior sexual abuse among PSB cases was found to be as low as 38% (2, 8, 9). Several other risk factors for PSB have been demonstrated to occur with significantly greater prevalence. For example, interpersonal violence was found in 68% of children presenting with PSB in the same study referenced above (9). A history of physical abuse was also identified in 47% of PSB cases (9). Other risk factors documented in the literature include high parental stress, neglect, and exposure to sexually explicit media (1, 8–13). A review by Elkovitch et al. (14) highlighted how the intersection of several risk factors plays a more impactful role in the development of PSB than any one risk factor (14). Research is needed to further characterize these risk factors and assess their impact on PSB.

Another misperception surrounding this topic is that PSB is comparable to sexual crimes committed by older adolescents and adults. Children who display PSB with other children are often labeled as "perpetrators," and viewed through the criminal lens, or considered victims of sexual abuse, although neither may be true (5, 12). The Survey on Youth with Problematic Sexual Behaviors also found that 67.8% of professionals in child-serving roles perceived children who displayed PSB to be similar to adult sexual offenders (15). Recent literature demonstrates that criminalizing the behavior and using punitive responses with children displaying PSB does a disservice to these children, who are in fact no more likely to perpetrate sexual abuse as adults when provided appropriate therapeutic intervention (16). These misinformed perceptions negatively impact the treatment of children affected by PSB and their ability to access care.

Further compounding these challenges is the lack of a coordinated community response or national standard of care to address these cases and support families. Not uncommonly, due to state legal regulations, responding to concerns of PSB falls beyond the scope of the agencies typically charged with keeping children safe, such as child welfare and law enforcement (4, 12). As a result, families often do not know who to turn to for help. The Survey on Youth with Problematic Sexual Behaviors conducted by the National Children's Alliance in 2020 found that among members of CAC's nationwide, 35.3% of respondents reported that their communities do not have a structured or consistent response to children affected by PSB (15). A recent qualitative study supported this, finding that many of the community agencies involved in addressing PSB in youth lack coordinated policies or a standardized response, resulting in a fragmented approach and limited ability to identify youth affected by PSB (17).

The challenges to effectively address PSB suggest that there is an increased role for medical providers to play in the community response to this issue. Medical providers, including pediatricians, mental health providers and child abuse specialists, are in an optimal position to provide education and resources to families, including education on child development and normative sexual behaviors, and referrals for therapeutic intervention (2). Problematic Sexualized Behavior- Cognitive Behavioral Therapy (PSB-CBT) is one such intervention that has demonstrated success with this patient population, with evidence of a ten-year recidivism rate of 2% for children under 12 years of age (16, 17). Despite this role for medical providers, there is limited research available to describe the medical evaluation of children affected by PSB. The available literature on PSB is concentrated mental health evaluation and treatment and the community response to these children.

This single-center retrospective case review aims to report descriptive data on child demographics and case characteristics for children presenting to one Child Protection Program (CPP) medical specialty team for concern of PSB.

Methods

This study was conducted at an urban, hospital-based Child Protection Program (CPP) medical specialty team in Massachusetts, United States. Children are referred to the CPP through a variety of sources when there is concern for child maltreatment, including medical professionals (emergency department providers, primary care providers, hospitalists), mental health providers, the Department of Children and Family (DCF), the District Attorney's office, and directly by caregivers. All referrals received by the CPP for medical triage are reviewed by a CPP social worker and a CPP medical provider to determine medical recommendations.

Chart review was conducted for all children medically triaged by CPP with concern of PSB during the period of 1/01/2020–12/31/2021. Cases were included in this analysis if they met the inclusion criteria outlined in Figure 1. Age < 16 years old was determined to be the age for inclusion in this study. While PSB is typically defined in the literature as occurring in children <12 years, the age of <16 years is used in practice in the community where this study was conducted as a threshold for how cases are assessed and addressed through resource provision.

For each index case referred to and medically triaged by CPP, data was extracted on child characteristics (age, developmental delay, and history of abuse), characteristics of the problematic sexual behavior (child's role, types of behaviors, number of children involved, and CPP referral status of involved children) and the outcome of the medical triage. The number of children reported to be involved in the behavior with the index child but not referred to CPP for medical triage was recorded for each case as "contacts not referred". The total number of children identified was calculated as the sum of the children referred to CPP and the contacts not referred.

The child's role was characterized as displayer, recipient, both or unknown. The role of "Displayer" was used to identify the child exhibiting the problematic sexualized behavior, while the role of

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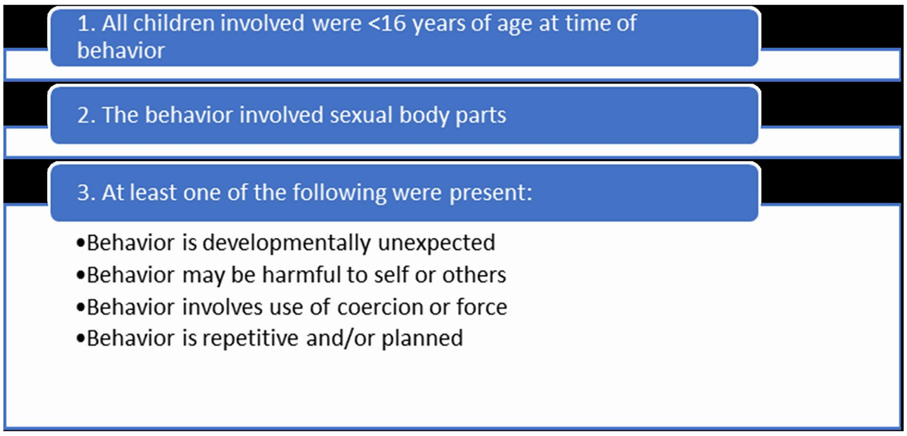
Abbreviations: PSB, Problematic Sexualized Behaviors; CAC, Children's Advocacy Center; PSB-CBT, Problematic Sexualized Behavior- Cognitive Behavioral Therapy; CPP, Child Protection Program; DCF, Department of Children and Families.



1. All children involved were <16 years of age at time of behavior

2. The behavior involved sexual body parts

3. At least one of the following were present:

- Behavior is developmentally unexpected
- Behavior may be harmful to self or others
- Behavior involves use of coercion or force
- Behavior is repetitive and/or planned

FIGURE 1
Inclusion criteria.

"Recipient" was used to identify the child on whom the behavior was performed. If a child both displayed and was the recipient of the behavior, their role in the behavior was designated as "Both". This included children who were the recipient of one behavior, who then displayed a different behavior at a different point in time. The designation of "Unknown" was assigned in cases where all children involved appeared to be participating in the problematic behaviors equally without a clear initiator (for example, in sexualized play).

For each case, the outcome of the medical triage was recorded. This included whether comprehensive medical evaluation was recommended, and if this evaluation was completed by CPP or a different medical provider.

For the purpose of this study, "developmental delay" was defined as documentation of a cognitive developmental delay in the electronic medical record. A complete developmental history is routinely assessed as part of a CPP clinic medical evaluation. Visit documentation in the medical record was reviewed for children seen in the CPP clinic to determine if the child had previously been diagnosed with cognitive delay. If the child was not seen in the CPP clinic for complete medical evaluation, a review of available medical records was conducted to assess for a documented history of a diagnosis of cognitive delay. Diagnosis of developmental delay was marked as unknown if the child did not have a complete medical history with recent well child examination documented in the medical record.

The child's history of abuse was marked as "previous abuse suspected" in situations where a history of physical or sexual abuse or neglect was disclosed at the time of initial triage or CPP clinic medical evaluation. In cases where the child was in DCF custody at time of triage, a history of abuse was presumed. History of abuse was excluded if the child received a complete medical evaluation by the CPP team and no history of abuse was identified, or if full medical record was available for review and did not include a history of abuse. All other cases were categorized as unknown.

Cases were excluded from the analysis if the problematic behavior identified was exclusively sending sexual images. While this behavior does meet the inclusion criteria used for this study, inconsistent tracking of this particular behavior over the study period would make its inclusion a source of error. Cases referred to CPP by the District Attorney's office as part of routine forensic interview follow up were also excluded from the analysis. These cases were not routinely tracked regarding PSB concerns, and therefore, were excluded from the analysis.

Risk ratios were calculated to assess for a relationship between each of the hypothesized risk factors (history of abuse, developmental delay) and the outcome of displaying PSB. Cases with unknown history of abuse and unknown history of developmental delay were excluded from the calculation of risk ratios. Chi-square test for independence was calculated using 2 degrees of freedom and 5% level of significance for the exposures of history of abuse, no history of abuse and unknown history of abuse and the outcomes of displaying PSB and being the recipient of PSB. The outcomes of "both" displaying and being the recipient of PSB and "unknown" role in the PSB behavior were excluded from the chi-square analysis due to having fewer than 5 observations per cell.

This study was determined by the Institutional Review Board to not include human subject research. It was therefore not subject to Institutional Review Board approval.

Results

A total of 110 index children were referred to the CPP for medical triage for a presenting concern of PSB. Through the medical triage process, an additional 114 children were identified as contacts of the index children. These contacts engaged in the PSB but were not referred to CPP for medical triage themselves. 51% of the 224 children in total who were identified as engaging in PSB, including both index children and contacts, were therefore not referred for medical specialty care.

Medical evaluation was recommended as the outcome of the medical triage for 81 (73.6%) of the 110 children referred to the CPP, but only 44 (40%) were medically evaluated by the CPP specialty team. An additional 22% received medical evaluation elsewhere, such as at their PCP. 10% of children were recommended to receive medical evaluation but were not evaluated by any medical provider (see Table 1).

Only 22 (20%) of the 110 children referred to the CPP were the displayer of the behavior, compared to 76 (69%) the recipients of the behavior (see Table 1). Of the recipient cases, the child displaying PSB was also referred to the CPP for medical evaluation 14.5% of the time.

Characteristics of children identified as having been affected by PSB are shown in Table 2. The mean age was 1.2 years older for children displaying PSB compared to the recipients of PSB. A presumed history of abuse was found in 45.5% of children identified as displaying PSB, compared to only 25% of children identified as the recipient of PSB.

There was no statistically significant association identified between history of abuse or developmental delay and having displayed PSB. Chi-square test for independence found the outcomes of displaying or being the recipient of PSB to be independent from history of abuse.

TABLE 1 Children identified as being involved in problematic sexualized behavior.

	n	%
Total children identified	224	
Referred to CPP	110	49.1
Displayer	22	20.0
Recipient	76	69.1
Both	5	4.5
Unknown	7	6.4
Contacts not referred	114	50.9
Medical follow up of children referred to CPP	110	
Medical evaluation recommended	81	73.6
Medical evaluation completed by external provider	25	22.7
Medical evaluation completed by CPP	44	40.0

Discussion

These results highlight that the number of children who seek medical evaluation for concern of PSB is a gross underestimation of the overall prevalence of PSB in the community, thus highlighting a significant gap in identification and service delivery for these children. Less than half of the children identified as being affected by PSB were referred to CPP. Though the majority of these identified cases were recommended to receive medical evaluation, less than half of these children were medically evaluated by CPP.

The results demonstrate that children displaying PSB represent a particular area of missed opportunity for assessment, education, and intervention. Displayers are disproportionately left out of care, despite having the most potential benefit through targeted therapeutic interventions such as Problematic Sexualized Behavior- Cognitive Behavioral Therapy (PSB-CBT) (3).

These results support the existing literature in suggesting that a history of abuse is a likely risk factor for displaying PSB, with a presumed history of abuse in nearly half of the identified displayers of PSB (2, 8, 9). Future research is needed to further characterize risk factors for both displaying and being the recipient of PSB, including delineating which types of previous exposure to child abuse and/or violence most strongly correlates with increased risk.

Limitations

Several limitations are inherent to the study's design as a retrospective case review. There is a high percentage of cases with unknown variables, such as unknown history of abuse, which may be significantly reduced in future research with data collected prospectively. Misclassification bias is possible due to this missing documentation as well as inaccuracies within medical records. This bias may affect the identification of cases, as well as the identification

TABLE 2 Characteristics of children displaying and receiving problematic sexualized behavior.

	Displayer (*n* = 22)	Recipient (*n* = 76)	Both (*n* = 5)	Unknown (*n* = 7)
Mean age (yrs.)	8.8	7.5	6.0	10.3
Age Max	16.0	15.0	8.0	15.0
Age Min	3.0	1.5	3.0	8.0
Developmental delay (%)				
Yes	13.6%	11.8%	0.0%	28.6%
No	68.2%	64.5%	80.0%	57.1%
Unknown	18.2%	23.7%	20.0%	14.3%
History of abuse (%)				
Previous abuse suspected	45.5%	25.0%	20.0%	57.1%
No known history of abuse	27.3%	44.7%	60.0%	0.0%
History of abuse unknown	27.3%	30.3%	20.0%	42.9%
Ave. # Children involved but not referred	0.6	1.2	1.4	0.4
Displayers referred (%)				
Yes	N/A	14.5%	20.0%	57.1%
No	N/A	85.5%	80.0%	42.9%
Ave. # recipients per displayer	1.3	N/A	N/A	N/A

of several of the risk factors for PSB discussed (i.e., developmental delay, history of abuse).

Another limitation of this study is the need to exclude certain subsets of cases due to inconsistent tracking throughout the duration of the study period. One such subset of cases includes those referred to CPP by the District Attorney's office as part of routine forensic interview follow up. These cases were not routinely tracked regarding PSB concerns, and therefore, were excluded from the analysis. As mentioned previously, another subset of cases excluded from analysis was cases where the problematic behavior identified was the sending of sexual images. CPP's process for managing these cases changed within the study period, resulting in inconsistent and variable tracking of these cases.

Conclusion

The inconsistency within and across communities in how cases of PSB are addressed by child-serving professionals is likely contributing to the high percentage of children not referred for medical evaluation. Systems level changes in how children impacted by PSB are identified and referred for services across community agencies, as well as the development of a national standard of care, is needed to address these missed opportunities. This will require a coordinated, multidisciplinary response involving child welfare agencies and investigatory agencies, as well as medical and mental health providers.

Data availability statement

The data analyzed in this study is subject to the following licenses/restrictions: protected information. Requests to access these datasets should be directed to kara.thompson@UMASSMEMORIAL.ORG.

Ethics statement

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements.

Author contributions

KT: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. SS: Conceptualization, Supervision, Writing – review & editing.

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Conflict of interest

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

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Variety ACEs and risk of developing anxiety, depression, or anxiety-depression co-morbidity: the 2006–2022 UK Biobank data

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Objectives: Adverse childhood experiences (ACEs) and anxiety-depression co-morbidity are attracting widespread attention. Previous studies have shown the relationship between individual psychiatric disorders and ACEs. This study will analyze the correlation between anxiety-depression co-morbidity and different levels of ACEs.

Methods: Seven categories of ACE and four classifications of psychiatric disorders were defined in a sample of 126,064 participants identified by the UK Biobank from 2006–2022, and correlations were investigated using logistic regression models. Then, to explore nonlinear relationships, restricted spline models were developed to examine differences in sex and age across cohorts ($n = 126{,}064$ for the full cohort and $n = 121{,}934$ for the European cohort). Finally, the impact of the category of ACEs on psychiatric disorders was examined.

Results: After controlling for confounders, ACEs scores showed dose-dependent relationships with depression, anxiety, anxiety-depression co-morbidity, and at least one (any of the first three outcomes) in all models. ACEs with different scores were significantly positively correlated with the four psychiatric disorders classifications, with the highest odds of anxiety-depression co-morbidity (odds ratio [OR]=4.87, 95% confidence intervals [CI]: 4.37~5.43), $p = 6.08 \times 10^{-178}$. In the restricted cubic spline models, the risk was relatively flat for females at ACEs=0–1 and males at ACEs=0–2/3 (except in males, where ACEs were associated with a lower risk of anxiety, all other psychiatric disorders had an increased risk of morbidity after risk smoothing). In addition, the risk of having anxiety, depression, anxiety-depression co-morbidity, and at least one of these disorders varies with each category of ACEs.

Conclusion: The prevalence of anxiety-depression comorbidity was highest across ACE scores after controlling for confounding factors and had a significant effect on each category of ACEs.

KEYWORDS

ACEs, anxiety-depression co-morbidity, UK Biobank, restricted cubic spline, sex

1 Introduction

Originally, the definition of Adverse Childhood Experiences (ACEs) was limited to the experience of domestic abuse or the presence of family dysfunction during childhood (1); however, the definition has now been expanded to include other related factors such as community dysfunction and peer dysfunction (2), which can lead to many negative impacts on the organism in adulthood, such as psychiatric disorders and common chronic diseases (3). According to the Centers for Disease Control and Prevention (CDC), the local Child Protective Services (CPS) counted 686,000 children in the United States who were injured as a result of ACEs (3). Most previous studies have used the ACE questionnaire developed by Felitti to detect 3 categories of child maltreatment (including physical, verbal, and sexual abuse) and 4 categories of family dysfunction (exposure to substance abuse, psychiatric disorders, violent treatment of mother or stepmother, and criminal behavior in the home) in children under or equal to 18 years of age (1). Subsequently, studies have expanded the measurement of adversity at the social dimension (4) and the ACEs International Questionnaire (ACE-IQ) has included social factors (5). Over the past 20 years, research on ACEs has focused primarily on North America (6). However, because of its widespread damage to adult mental health, addiction, and life expectancy (7, 8), many other countries (including Europe) have begun to study ACE (9).

In recent years, the Epidemiological Research Center Depression Scale (CES-D) has been reported to have high detection rates in countries such as Italy (37%) and Spain (49%), which has prompted us to study psychiatric disorders in European countries (10). Early studies have found a strong association between depression and suicidal ideation (11, 12), with approximately 58% of patients with major depressive episodes reporting suicidal thoughts (13), which suggests that the serious risk posed by psychiatric disorders to an individual's health cannot be ignored. Recent studies have shown that current evidence does not support the hypothesis that depression is caused by reduced serotonin activity or concentration (14). However, in the case of ACE, early adverse emotions associated with it alter normal psychological development, leading to psychologization (15) and mood disorders (16), triggering a biological stress response leading to effects on the hypothalamus-pituitary–adrenal (HPA) axis (17–19) that stimulate cortisol secretion from the adrenal cortex, which persists at high levels for long periods placing individuals at an increased risk for depression and anxiety. The development of psychiatric disorders may also be associated with the cumulative number of ACEs (18). Recent studies have found: that early ACEs worsen psychiatric problems in children (20) and the middle old-age (21); there is a dose-dependent relationship between ACEs and the development of chronic diseases or other risky behaviors in adulthood (22), also including psychiatric disorders (23). Of these, depression or anxiety disorders are the most common, and in some literature, it has been shown that the prevalence of depression is usually higher than that of anxiety (18, 19, 24). However, in the specific category of ACE, anxiety is more closely related to sexual or physical abuse, and depression is more closely related to emotional abuse (25).

Many previous studies have examined the relationship between ACEs and anxiety or depression, but to our knowledge, fewer studies focused on examining the relationship between anxiety-depression comorbidity (the co-occurrence of anxiety and depressive symptoms in individuals) (3, 19, 23, 26, 27). By the ICD-10 criteria, anxious depression not only requires the former condition to be met, but neither manifestation is the primary symptom. And if both manifestations can reasonably be diagnosed separately it cannot be said to be anxious depression (28). From reading the literature we have learned that, compared to non-anxious depression, anxious depression suffers from increased clinical symptoms, more frequent depressive episodes, more pronounced symptoms, and even an increased risk of suicide (28).

Therefore, the present study was designed to test the three hypotheses we proposed. First, we wanted to examine whether people who experience ACE are at increased risk for anxiety-depression co-morbidity, anxiety, depression, or at least one of these (any of the first three outcomes); and whether anxiety-depression co-morbidity is most strongly associated with ACE. In addition, we used restricted triple spline methods to assess the dose–response relationship between ACEs and psychiatric disorders in different sex groups and performed sensitivity analyses in European cohort. Also, we explored the dose–response relationship between ACEs and psychiatric disorders in different age groups. Finally, we examined the relationship between the categories of ACE and anxiety-depression comorbidity, anxiety, depression, or at least one of these (any of the first three outcomes).

2 Materials and methods

2.1 Data sources

This study used data from UK Biobank, an ongoing prospective population-based cohort study,[1] the aim is to accurately and comprehensively assess environmental, psychosocial, genetic, and non-genetic factors related to exposure and outcomes and further analyze their relationships. From 2006 to 2010, 500,000 people aged 37–73 were recruited from 22 assessment centers in the UK, including touch screens, physical measures, and biological sampling (29).

To investigate whether the categories of ACEs and different scores led to an increased risk of developing new cases of psychiatric disorders in the UK Biobank database, we established exclusion criteria for all participants: (1) those who had been lost to follow-up by 2022 for any reason, (2) participants lacking information on what was defined as an ACE, and (3) those who had a confirmed diagnosis of psychiatric disorders prior to recruitment, resulting in the inclusion of 126,064 participants (55,481 males and 70,583 females). The detailed process of participant selection is shown in Figure 1.

2.1.1 Ascertainment of ACEs

First, the content of the Conflict Tactics Scale (CTS) (30) includes detailed questions about emotional abuse, physical abuse, and domestic

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Abbreviations: ACEs, Adverse childhood experiences; CDC, Centers for Disease Control and Prevention; CPS, Child Protective Services; AS, anxiety sensitivity; CES-D, depression scale; CTS, Conflict Tactics Scale; CTQ, Child Trauma Questionnaire; IPAQ activity group, International Physical Activity Questionnaire activity group.

[1] www.ukbiobank.ac.uk/resources/

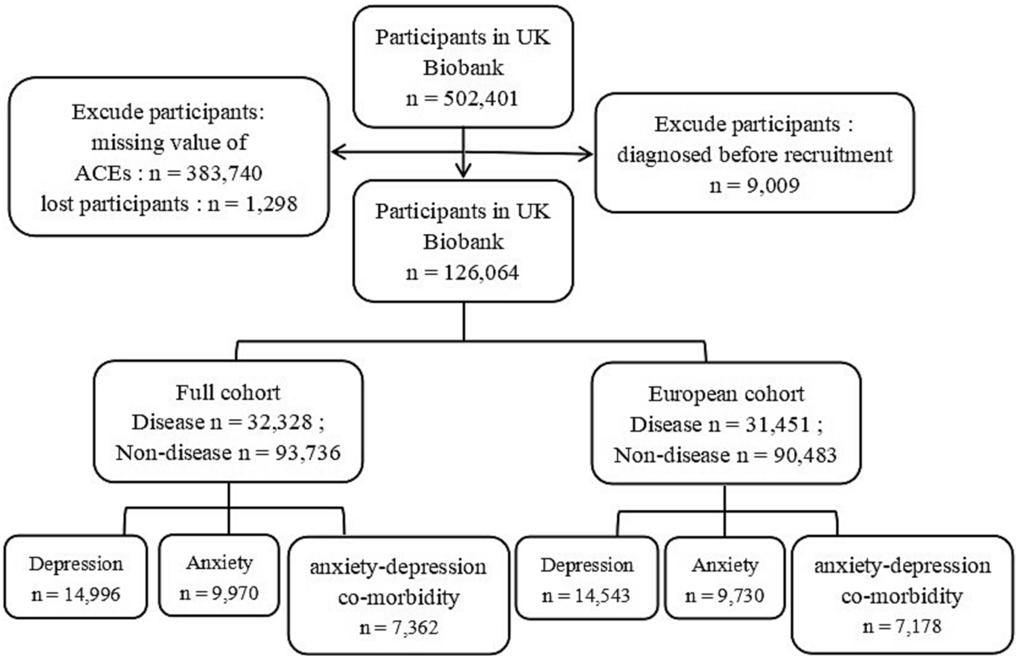

FIGURE 1
Flow chart for filtering participants in UK Biobank.

violence; second, the Child Trauma Questionnaire (CTQ) (31) includes detailed questions that measure emotional and physical neglect, with some items being reverse-scored. Simultaneously, this is all relative to children under 18. We used an ACE questionnaire (32) based on the above questions, which then corresponded to the variables scale of the UK Biobank, while early life factors, family history reports, and traumatic events in online mental health questionnaires were assessed. Ultimately, we included seven types of ACEs in this study (felt hated by family members as a child, physically abused by family as a child, felt loved as a child, sexually molested as a child, someone to take to the doctor when needed as a child, maternal smoking around birth, and having a family history of psychosis). It's worth noting that feeling loved as a child and someone to take to the doctor when needed as a child are reverse scores, which we re-coded for analysis (30, 31). The above ACEs were created as a binary variable (0 = no, 1 = yes). The seven types were then combined to calculate the total number of ACEs, yielding a range of 0–7 scores, which were divided into five groups (0, 1, 2, 3, ≥ 4) based on the total number in subsequent analysis (32). In addition, we describe the corresponding categories of ACEs in the ACE questionnaire in Supplementary Table S1.

2.1.2 Ascertainment of outcome

The outcomes in this study were depression, anxiety, anxiety-depression co-morbidity, or at least one (any of the first three outcomes), and they were all defined as binary variables (0 = no illness, 1 = illness). We determined the number of participants based on admission data ICD-10 (main conditions of anxiety disorders, bipolar disorder, depression, and recurrent depression) (33, 34), and self-reported disease diagnoses (non-cancer disease codes) recorded in the database. Addresses for admission data and reasons for admission were obtained by linking to records from Health Event Statistics (England and Wales) and Scottish Morbidity Records (Scotland). Specific information can be found online.[2] A record of the diagnosis of psychiatric disorders is provided in Supplementary Table S2. And anxiety-depression co-morbidity is defined as the simultaneous occurrence of anxiety and depressive diagnosis in an individual. It is important to note that the diagnoses of psychiatric disorders that we defined as new cases all occurred after the 2006–2010 recruitment through the July 19, 2022 cutoff.

2.1.3 Ascertainment of confounders

Demographic information about the covariates of 126,064 participants in the UK Biobank from 2006–2022 was studied for age (years), sex (0 = female, 1 = male), smoking status (0 = never, 1 = previous, 2 = current), alcohol drinker status (0 = never, 1 = previous, 2 = current), education (1 = university degree, 2 = below university or other professional qualifications, 3 = none of the above), International Physical Activity Questionnaire (IPAQ) activity group (0 = low, 1 = medium, 2 = high), ethnic background (1 = European, 2 = European or Asian or African mixed race, 3 = Asian, 4 = African, 5 = others), BMI (1 ≤ 18.5, 2 = 18.5–24.9, 3 = 25–29.9, 4 ≥ 30), Townsend Deprivation Index (TDI) (35) at recruitment, which represents socioeconomic status. The above confounders are included in this study.

2.2 Statistical analysis

First, descriptive statistics were performed on the participants' baseline characteristics and outcome variables. Continuous variables

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[2] https://biobank.ctsu.ox.ac.uk/crystal/field.cgi?id=41270

were expressed as means (standard deviation, [SD]), and categorical variables were expressed as frequencies (percentages). In addition, after stratification by ACEs scores, chi-square and Kruskal-Wallis H tests were used to check the significance of differences between participants. Logistic regression models were constructed to assess the relationship between ACE scores and depression, anxiety disorders, anxiety-depression co-morbidity, or at least one of these. Next, to test the robustness of the above relationships, we first tested the prevalence of ACE and psychiatric disorders across sex groups (n = 70,583 for females and n = 55,481 for males). Then, we constructed restricted cubic spline models (with 4 nodes at the 25th, 50th, 75th, and 95th quartiles) to test for differences in ACE in the full and European populations (36). Finally, we examined the correlation between the category of ACEs and each outcome variable. For missing values of variables, we performed multiple interpolations using the MICE package (37), and the proportion of missing data is described in Supplementary Table S3. The level of statistical significance was determined as 95% and bilateral ($p < 0.05$), and correlations were expressed as OR with 95% confidence intervals (CI). All the above analyses were performed in R software (R 2.4.1).

3 Results

3.1 Baseline comparison

The baseline characteristics are shown in Table 1. Overall, ACEs occurred in 55.59% of all participants aged 38–73. ACEs scores of 1, 2, 3, and ≥ 4 accounted for 33.79, 14.42, 5.37, and 2.01%, respectively. Also, all variables were associated with ACEs scores and all differences were significant ($p < 0.001$). Meanwhile, among all ACEs scores, females (55.99%) had more ACEs than males (44.01%) in the full cohort. Compared to other ethnic background groups, the number of Europeans is the highest (96.72%). Among other variables, most of the incidence of ACEs occurred in categories below college education (79.68%), history of alcohol consumption (94.83%), and overweight (41.31%). From the content of ACEs score and psychiatric disorders, it was clear that depression (24.64%), anxiety (10.21%), anxiety-depression co-morbidity (20.06%), and at least one (54.91%) accounted for the highest proportion when the ACEs score ≥ 4.

3.2 ACEs with psychiatric disorders risk in the full cohort

As shown in Table 2, a positive correlation was found between the ACEs score and outcome variables. When ACEs score ≥ 4, the outcome variables in descending order of psychiatric disorders risk were anxiety-depression co-morbidity (OR = 4.87, 95% CI: 4.37 ~ 5.43), at least one (OR = 3.90, 95% CI: 3.59 ~ 4.23), depression (OR = 2.54, 95% CI: 2.31 ~ 2.80) and anxiety (OR = 1.38, 95% CI: 1.21 ~ 1.58). Smoking status, alcohol consumption, and TDI (except for anxiety) were all positively correlated with psychiatric disorders, while IPAQ physical activity was negatively correlated with psychiatric disorders. Compared with other participants in the same group, obesity (BMI ≥ 30) had a higher correlation with depression (OR = 1.75, 95% CI: 1.37–2.26) and at least one (OR = 0.21, 95% CI: 1.02–1.43). Meanwhile, when an individual has a below college degree, there is a higher correlation with anxiety (OR = 1.13, 95% CI: 1.06–1.20), anxiety-depression co-morbidity (OR = 1.09, 95% CI: 1.01–1.18), and at least one (OR = 1.06, 95% CI: 1.02–1.10).

3.3 Prevalence of ACEs in different groups

Overall, there was a significant difference in prevalence between males and females in the different subgroups of ACEs ($p < 0.001$). 44.01% of males and 55.99% of females participated in the study (Figure 2). The prevalence of ACEs in the female (56.03%) group was higher than that in the male (43.97%) group ($p < 0.001$) (Figure 2).

3.4 ACEs with psychiatric disorders risk at different sex in full and European cohort

Figure 3 shows results from the restricted cubic spline models for the relationship between categories of ACE and outcome variables in the full cohort, grouped by sex and controlling for confounders. In several groups, there was a dose-dependent relationship between ACEs and outcome variables ($p < 0.05$ for non-linear tests of depression and at least one, $p > 0.05$ for non-linear tests of anxiety and anxiety-depression co-morbidity) and higher for females than males. Results for females showed that ACE = 0 was linked with the lowest odds ratio of incident depression (OR = 0.77, 95% CI: 0.74 ~ 0.79), anxiety (OR = 0.90, 95% CI: 0.86 ~ 0.93), anxiety-depression co-morbidity (OR = 0.70, 95% CI: 0.67 ~ 0.73) and at least one (OR = 0.77, 95% CI: 0.74 ~ 0.80). The risk of depression (OR = 0.88, 95% CI: 0.83 ~ 0.93) in males was relatively flat at ACEs = 0–3, while the risk of anxiety-depression co-morbidity (OR = 0.85, 95% CI: 0.80 ~ 0.90) and at least one (OR = 0.73, 95% CI: 0.70 ~ 0.75) at ACEs = 0–3 relatively flat, followed by a gradual increase. Hereafter, the risk ratios for anxiety in males were all less than 1, with

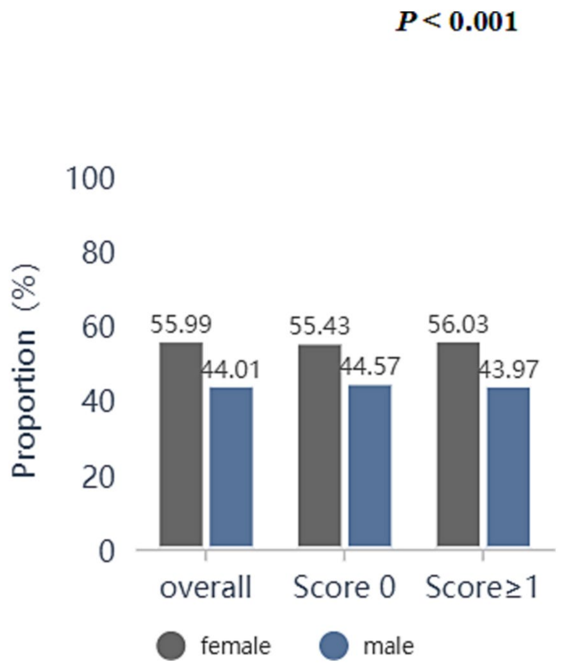


FIGURE 2
Prevalence of ACEs stratified by sex in the 2006–2022 UK Biobank.

TABLE 1 The characteristics of the selected participants grading according to different ACEs scores in UK Biobank.

	Level	Overall	ACEs Score					*p* value
			Score 0	Score 1	Score 2	Score 3	Score 4	
N (%)		126,064	55,984 (44.41)	42,595 (33.79)	18,179 (14.42)	6,769 (5.37)	2,537 (2.01)	
Age (Mean ± SD)		55.852 (7.77)	56.36 (7.89)	55.93 (7.61)	55.05 (7.64)	54.16 (7.57)	53.72 (7.60)	<0.001
Sex (%)	Female	70,583 (55.99)	31,034 (55.43)	23,288 (54.67)	10,294 (56.63)	4,182 (61.78)	1,785 (70.36)	<0.001
	Male	55,481 (44.01)	24,950 (44.57)	19,307 (45.33)	7,885 (43.37)	2,587 (38.22)	752 (29.64)	
Smoking status (%)	Never	73,539 (58.33)	34,591 (61.79)	24,783 (58.18)	9,669 (53.19)	3,321 (49.06)	1,175 (46.31)	<0.001
	Previous	43,783 (34.73)	18,232 (32.57)	14,826 (34.81)	6,966 (38.32)	2,737 (40.43)	1,022 (40.28)	
	Current	8,742 (6.93)	3,161 (5.65)	2,986 (7.01)	1,544 (8.49)	711 (10.50)	340 (13.40)	
Alcohol drinker status (%)	Never	3,545 (2.81)	1740 (3.11)	1,120 (2.63)	427 (2.35)	173 (2.56)	85 (3.35)	<0.001
	Previous	2,968 (2.35)	1,060 (1.89)	986 (2.31)	509 (2.80)	275 (4.06)	138 (5.44)	
	Current	119,551 (94.83)	53,184 (95.00)	40,489 (95.06)	17,243 (94.85)	6,321 (93.38)	2,314 (91.21)	
Ethnic background (%)	European	121,934 (96.72)	54,384 (97.14)	41,279 (96.91)	17,439 (95.93)	6,426 (94.93)	2,406 (94.84)	<0.001
	European or Asian or African mixed race	634 (0.50)	175 (0.31)	205 (0.48)	130 (0.72)	83 (1.23)	41 (1.62)	
	Asian	1,185 (0.94)	552 (0.99)	359 (0.84)	183 (1.01)	65 (0.96)	26 (1.02)	
	African	953 (0.76)	349 (0.62)	291 (0.68)	191 (1.05)	92 (1.36)	30 (1.18)	
	Others	1,358 (1.08)	524 (0.94)	461 (1.08)	236 (1.30)	103 (1.52)	34 (1.34)	
IPAQ activity group (%)	Low	19,376 (17.93)	8,526 (17.85)	6,525 (17.83)	2,880 (18.34)	1,025 (17.52)	420 (19.29)	<0.001
	Moderate	46,529 (43.05)	20,943 (43.84)	15,797 (43.18)	6,569 (41.83)	2,382 (40.72)	838 (38.49)	
	High	42,187 (39.03)	18,304 (38.31)	14,266 (38.99)	6,255 (39.83)	2,443 (41.76)	919 (42.21)	
BMI (%)	≤ 18.5	710 (0.56)	372 (0.67)	207 (0.49)	86 (0.47)	32 (0.47)	13 (0.51)	<0.001
	18.5–24.9	49,057 (39.01)	23,310 (41.72)	16,004 (37.66)	6,632 (36.58)	2,315 (34.29)	796 (31.49)	
	25–29.9	51,951 (41.31)	22,926 (41.03)	17,821 (41.94)	7,472 (41.21)	2,759 (40.86)	973 (38.49)	
	≥ 30	24,053 (19.10)	9,262 (16.56)	8,459 (19.89)	3,940 (21.51)	1,646 (24.36)	746 (29.49)	
TDI (Mean ± SD)		−1.759 (2.801)	−1.960 (2.688)	−1.767 (2.785)	−1.484 (2.920)	−1.167 (3.084)	−0.724 (3.240)	<0.001
Education (%)	University	17,384 (13.82)	7,896 (14.14)	5,819 (13.69)	2,441 (13.45)	906 (13.42)	322 (12.72)	<0.001
	Below university or other professional qualifications	100,235 (79.68)	44,643 (79.92)	33,775 (79.44)	14,481 (79.80)	5,341 (79.14)	1995 (78.79)	
	None of the above	8,184 (6.51)	3,322 (5.95)	2,921 (6.87)	1,224 (6.75)	502 (7.44)	215 (8.49)	
Depression (%)	No	111,068 (88.10)	50,765 (90.68)	37,589 (88.25)	15,314 (84.24)	5,488 (81.08)	1912 (75.36)	<0.001
	Yes	14,996 (11.90)	5,219 (9.32)	5,006 (11.75)	2,865 (15.76)	1,281 (18.92)	625 (24.64)	
Anxiety (%)	No	116,094 (92.09)	51,906 (92.72)	39,165 (91.95)	16,606 (91.35)	6,139 (90.69)	2,278 (89.79)	<0.001
	Yes	9,970 (7.91)	4,078 (7.28)	3,430 (8.05)	1,573 (8.65)	630 (9.31)	259 (10.21)	
Anxiety-depression co-morbidity (%)	No	118,702 (94.16)	53,808 (96.11)	40,300 (94.61)	16,602 (91.33)	5,964 (88.11)	2028 (79.94)	<0.001
	Yes	7,362 (5.84)	2,176 (3.89)	2,295 (5.39)	1,577 (8.67)	805 (11.89)	509 (20.06)	
At least one (%)	No	93,736 (74.36)	44,511 (79.51)	31,864 (74.81)	12,164 (66.91)	4,053 (59.88)	1,144 (45.09)	<0.001
	Yes	32,328 (25.64)	11,473 (20.49)	10,731 (25.19)	6,015 (33.09)	2,716 (40.12)	1,393 (54.91)	

ACEs, adverse childhood experiences; IPAQ, International Physical Activity Questionnaire; BMI, body mass index; TDI, Townsend Deprivation Index.

the highest result being 0.96 (95% CI: 0.76 ~ 1.26) at ACEs = 7. Besides, females (OR = 9.52, 95% CI: 7.81 ~ 11.61) and males (OR = 5.57, 95% CI: 4.53 ~ 6.86) with ACEs score of 7 has the highest risk ratio for anxiety-depression co-morbidity. Comparative results by sex in the European cohort and by age in the full cohort are shown in Supplementary Figures S1, S2.

3.5 Category of ACEs with psychiatric disorders risk in the full cohort

Figure 4 shows the logistic regression results between the category of ACEs and the outcome variables, also controlling for confounders. Overall, most of the results were significant, but the

TABLE 2 Logistic regression models for the relationship between ACE scores and outcomes.

Variables	Depression OR (95% CI)	Anxiety OR (95% CI)	Anxiety-depression co-morbidity OR (95% CI)	At least one OR (95% CI)
ACE Score	Reference	Reference	Reference	Reference
Score 1	**1.26 (1.21, 1.31)** ***	**1.12 (1.07, 1.17)** ***	**1.36 (1.28, 1.45)** ***	**1.28 (1.24, 1.32)** ***
Score 2	**1.69 (1.61, 1.78)** ***	**1.21 (1.14, 1.28)** ***	**2.16 (2.02, 2.32)** ***	**1.82 (1.75, 1.88)** ***
Score 3	**1.98 (1.85, 2.12)** ***	**1.29 (1.18, 1.41)** ***	**2.86 (2.62, 3.12)** ***	**2.32 (2.20, 2.45)** ***
Score ≥ 4	**2.54 (2.31, 2.80)** ***	**1.38 (1.21, 1.58)** ***	**4.87 (4.37, 5.43)** ***	**3.90 (3.59, 4.23)** ***
Age	**0.99 (0.98, 0.99)** ***	**1.01 (1.01, 1.02)** ***	**0.98 (0.97, 0.98)** ***	**0.99 (0.98, 0.99)** ***
Sex				
Female	Reference	Reference	Reference	Reference
Male	**0.57 (0.55, 0.59)** ***	**0.66 (0.63, 0.69)** ***	**0.58 (0.55, 0.62)** ***	**0.53 (0.52, 0.55)** ***
Smoking status				
Never	Reference	Reference	Reference	Reference
Previous	**1.23 (1.18, 1.27)** ***	**1.16 (1.11, 1.20)** ***	**1.27 (1.21, 1.34)** ***	**1.28 (1.24, 1.31)** ***
Current	**1.53 (1.43, 1.62)** ***	1.08 (0.99, 1.72)	**1.54 (1.42, 1.68)** ***	**1.53 (1.46, 1.61)** ***
Alcohol drinker status				
Never	Reference	Reference	Reference	Reference
Previous	**1.29 (1.12, 1.49)** ***	**1.32 (1.11, 1.58)** *	**1.66 (1.38, 2.00)** ***	**1.59 (1.42, 1.77)** ***
Current	0.99 (1.89, 1.10)	1.08 (0.95, 1.23)	0.92 (0.80, 1.07)	1.00 (0.92, 1.08)
Ethnic background				
White	Reference	Reference	Reference	Reference
Mixed	0.92 (0.73, 1.15)	**0.71 (0.51, 0.99)** *	0.78 (0.57, 1.06)	**0.78 (0.65, 0.93)** **
China	**0.77 (0.63, 0.94)** *	**0.76 (0.59, 0.98)** *	**0.48 (0.34, 1.67)** ***	**0.64 (0.55, 0.75)** ***
Black	**0.51 (0.40, 0.64)** ***	**0.70 (0.53, 0.93)** *	**0.46 (0.33, 0.63)** ***	**0.47 (0.40, 0.56)** ***
Others	0.95 (0.81, 1.12)	**0.79 (0.63, 0.99)** *	**0.69 (0.54, 0.89)** **	**0.80 (0.70, 0.91)** **
IPAQ activity group				
Low	Reference	Reference	Reference	Reference
Moderate	**0.95 (0.90, 1.00)** *	0.97 (0.91, 1.03)	**0.87 (0.81, 0.93)** ***	**0.92 (0.88, 0.96)** ***
High	**0.89 (0.84, 0.94)** ***	**0.93 (0.88, 0.99)** *	**0.82 (0.77, 0.88)** ***	**0.86 (0.83, 0.90)** ***
Education				
University	Reference	Reference	Reference	Reference
Below university or other professional qualifications	0.98 (0.93, 1.03)	**1.13 (1.06, 1.20)** **	**1.09 (1.01, 1.18)** **	**1.06 (1.02, 1.10)** **
None of the above	0.93 (0.86, 1.02)	**1.11 (1.00, 1.22)** *	1.11 (0.99, 1.24)	1.03 (0.97, 1.10)
BMI				
≤ 18.5	Reference	Reference	Reference	Reference
18.5–24.9	1.24 (0.97, 1.60)	1.04 (0.80, 1.36)	**0.61 (0.47, 0.80)** **	0.96 (0.81, 1.14)
25–29.9	**1.39 (1.08, 1.78)** *	0.99 (0.76, 1.30)	**0.68 (0.53, 0.89)** **	1.03 (0.87, 1.22)
≥ 30	**1.75 (1.37, 2.26)** ***	0.94 (0.72, 1.23)	0.77 (0.59, 1.00)	**1.21 (1.02, 1.43)** *
TDI	**1.02 (1.02, 1.03)** ***	1.00 (1.00, 1.01)	**1.03 (1.02, 1.04)** ***	**1.03 (1.02, 1.03)** ***

It was adjusted for age, sex, smoking status, alcohol drinker status, ethnic background, IPAQ activity group, education, BMI, and TDI among 126,064 participants (full cohort). ACEs, adverse childhood experiences; IPAQ, International Physical Activity Questionnaire; BMI, body mass index; TDI, Townsend Deprivation Index; boldface indicates statistical significance (*$0.01 < p$ value < 0.05, **$0.001 < p$ value < 0.01, ***p value < 0.001).

correlation between being taken to the doctor when needed as a child and depression or anxiety, the correlation between maternal smoking around birth and anxiety, and the correlation between being sexually molested as a child and anxiety were not significant ($p > 0.05$). It was found that the rates of anxiety-depression co-morbidity were always the highest. Compared to other categories of ACEs, in the OR values of all diseases (in addition to anxiety), felt loved as a child (reverse rating) was the highest value, in the following order (from left to right in Figure 4), OR = 1.92 (95% CI: 1.69 ~ 2.17), OR = 2.88 (95% CI: 2.51 ~ 1.31), OR = 2.53 (95% CI: 2.28 ~ 2.81). Those with a family history of psychiatric disorders (OR = 1.24, 95% CI: 1.17 ~ 1.31) had the greatest values when suffering from anxiety. Feeling hated by family members as a child was associated with depression (OR = 1.65, 95% CI: 1.58 ~ 1.72), anxiety (OR = 1.23, 95% CI: 1.17 ~ 1.30), anxiety-depression co-morbidity (OR = 2.19, 95% CI: 2.07 ~ 2.31) and at least one (OR = 1.92, 95% CI: 1.85 ~ 1.98) increased probability showed a positive correlation. Physically abused by family as a child was associated with depression (OR = 1.35, 95% CI: 1.30 ~ 1.41), anxiety (OR = 1.14, 95% CI: 1.08 ~ 1.20), anxiety-depression co-morbidity (OR = 1.60, 95% CI: 1.52 ~ 1.69) and at least one (OR = 1.46, 95% CI: 1.41 ~ 1.51) were significantly associated with increased odds of prevalence. Sexually molested as a child was significantly associated with depression (OR = 1.46, 95% CI: 1.38 ~ 1.54), anxiety-depression co-morbidity (OR = 1.78, 95% CI: 1.66 ~ 1.91), and at least one (OR = 1.58, 95% CI: 1.51 ~ 1.64). An increased odds for someone to take to the doctor when needed as a child (reverse rating) was associated with anxiety-depression co-morbidity (OR = 1.78, 95% CI: 1.66 ~ 1.91) and at least one (OR = 1.58, 95% CI: 1.51 ~ 1.64). Having a family history of psychosis was associated with increased odds of having depression (OR = 1.63, 95% CI: 1.57 ~ 1.71), anxiety-depression co-morbidity (OR = 2.26, 95% CI: 2.14 ~ 2.39), and at least one (OR = 1.92, 95% CI: 1.86 ~ 1.99). Maternal smoking at birth was significantly associated with depression (OR = 1.12, 95% CI: 1.08 ~ 1.16), anxiety-depression co-morbidity (OR = 1.18, 95% CI: 1.12 ~ 1.24), and at least one (OR = 1.13, 95% CI: 1.10 ~ 1.16).

4 Discussion

This study used a large, diverse, and multicultural dataset from the UK Biobank to discover the dose–response relationship between ACE, depression, anxiety, at least one, and comorbidity of anxiety and depression through our research, thus validating the previous research some findings in the relevant literature. Considering the difference in race and age, we further verified the above relationship, and the final result showed that the prevalence of anxiety-depression co-morbidity was the highest. We also suggested significant

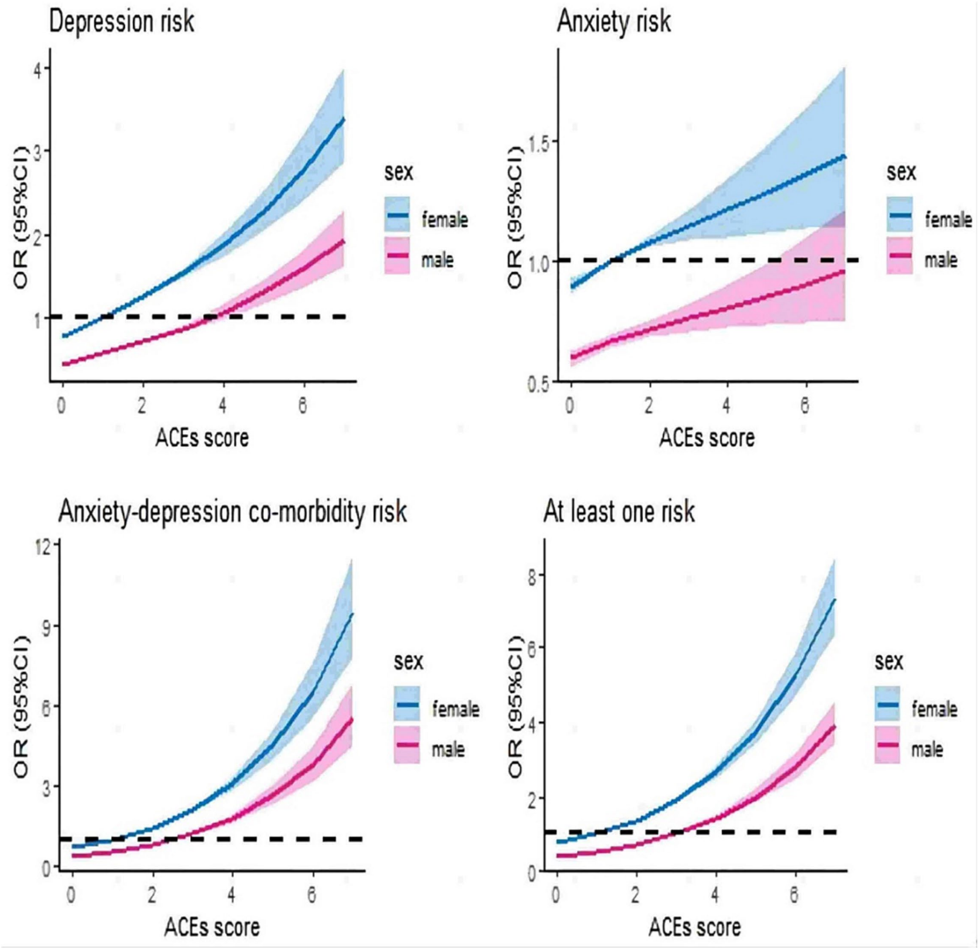
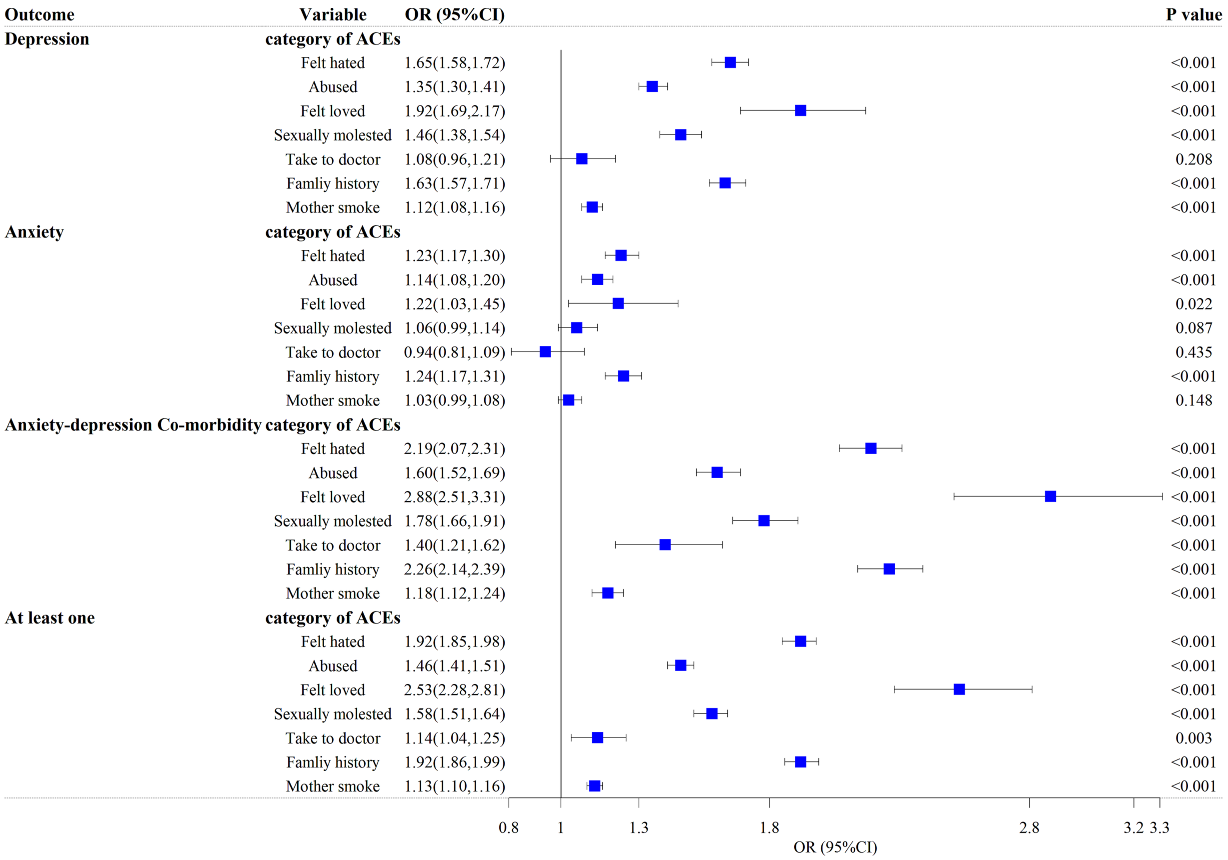


FIGURE 3
Restricted cubic spline models for relationship between ACEs and outcomes at different sex groups (full cohort). It was adjusted for age, sex, smoking status, alcohol drinker status, ethnic background, IPAQ activity group, education, BMI and TDI among 126,064 participants (full cohort), grouped according to sex.



Outcome	Variable	OR (95%CI)	P value
Depression	category of ACEs		
	Felt hated	1.65(1.58,1.72)	<0.001
	Abused	1.35(1.30,1.41)	<0.001
	Felt loved	1.92(1.69,2.17)	<0.001
	Sexually molested	1.46(1.38,1.54)	<0.001
	Take to doctor	1.08(0.96,1.21)	0.208
	Famliy history	1.63(1.57,1.71)	<0.001
	Mother smoke	1.12(1.08,1.16)	<0.001
Anxiety	category of ACEs		
	Felt hated	1.23(1.17,1.30)	<0.001
	Abused	1.14(1.08,1.20)	<0.001
	Felt loved	1.22(1.03,1.45)	0.022
	Sexually molested	1.06(0.99,1.14)	0.087
	Take to doctor	0.94(0.81,1.09)	0.435
	Famliy history	1.24(1.17,1.31)	<0.001
	Mother smoke	1.03(0.99,1.08)	0.148
Anxiety-depression Co-morbidity	category of ACEs		
	Felt hated	2.19(2.07,2.31)	<0.001
	Abused	1.60(1.52,1.69)	<0.001
	Felt loved	2.88(2.51,3.31)	<0.001
	Sexually molested	1.78(1.66,1.91)	<0.001
	Take to doctor	1.40(1.21,1.62)	<0.001
	Famliy history	2.26(2.14,2.39)	<0.001
	Mother smoke	1.18(1.12,1.24)	<0.001
At least one	category of ACEs		
	Felt hated	1.92(1.85,1.98)	<0.001
	Abused	1.46(1.41,1.51)	<0.001
	Felt loved	2.53(2.28,2.81)	<0.001
	Sexually molested	1.58(1.51,1.64)	<0.001
	Take to doctor	1.14(1.04,1.25)	0.003
	Famliy history	1.92(1.86,1.99)	<0.001
	Mother smoke	1.13(1.10,1.16)	<0.001

FIGURE 4

Logistic regression model for the relationship between individual categories of ACE and outcomes. It was adjusted for age, sex, smoking status, alcohol drinker status, ethnic background, IPAQ activity group, education, BMI and TDI among 126,064 participants (full cohort). Felt hated, felt hated by family member as a child; Abused, physically abused by family as a child; Felt loved, felt loved as a child; Sexually molested, sexually molested as a child; Take to doctor, someone to take to doctor when needed as a child; Family history, having a family history of psychosis; Mother smoke, maternal smoking around birth.

differences between categories of ACEs and anxiety-depression co-morbidity, controlling for relevant demographic and socioeconomic factors.

In this study, 55.59% reported at least one ACE, which is within the range reported in the literature (46.4–79.5%) (3). As expected from our first hypothesis, in some literature (21, 22, 26) ACEs increase the probability of adult exposure to psychiatric disorders and risky behaviors. In the present study, ACEs were highest when anxiety-depression co-morbidity was present. Although the number of anxiety-depression co-morbidity is less than depression, it had the highest value in the results because it had both anxiety and depression symptoms compared to a single psychiatric disorder. Another point mentioned is the research on the correlation between ACEs and psychiatric disorders, female, smoking history, low education, drinking history, overweight or obesity, and TDI are all risk factors; frequent physical activity is a protective factor. The above results are the same as those of this literature (38–40). As early ACE-induced mood changes trigger a biological stress response, it leads to an impact on the HPA axis, stimulating the adrenal cortex to secrete cortisol at persistently high levels for a prolonged period, placing the individual at an increased risk of developing depression and anxiety disorders (24). The higher risk of females in this compared to males may be due to the fact that females themselves have higher cortisol levels than males; both smoking and drinking are the results of compensation for bad childhood behaviors (41). TDI indices tend to reflect socio-economic levels at the regional level (42), with higher scores indicating poorer areas (35), which may put pressure on parents to make children more vulnerable to ACEs. Previous research has shown that social background is an important factor influencing ACEs (43). For example, in a study based on a representative sample of the German population, participants from West Germany/foreign countries were at a higher risk of experiencing ACEs compared to East Germany, where state-directed child care is available (44). Ethnicity may be limited by the area of data collection, with Europeans having a higher prevalence in comparison to other ethnicities.

As expected from our second hypothesis, our results show that the dose-dependent relationship between ACEs and psychiatric disorders differed between participants of different sex and ages. We also selected the European group due to the largely white population. In addition to cortisol, Robert C. Whitaker's study of the interaction between ACEs and depression or anxiety disorders and sex in U.S. adults suggests that the synergistic effect of ACEs and females on anxiety or depression is greater than the separate effects of these two factors (26).

As expected from our third hypothesis, our results showed significant differences between the category of ACEs and anxiety-depression co-morbidity. Feeling loved in childhood (this reverse score belongs to emotional neglect) was highest in depression and anxiety-depression co-morbidity. Family history of psychiatric disorders was highest in anxiety disorders. However, there were also nonsignificant categories, which suggest that the mechanisms of expression of each ACE may have different implications for a single psychiatric disorder. Mechanisms under a single expression: different types of abuse can reactively alter the HPA, thus impairing the emergency attachment system and leading to varying degrees of mood disturbance and increased or decreased cortisol concentrations (41). The lack of significance mentioned above may also be due to the significant difference between the number of patients and non-patients, resulting in low statistical power. It is worth noting that since at the outset, our assumptions for the definition of at least one was any of depression, anxiety, and anxiety-depression co-morbidity, taking into account the number of people with the condition became larger, the risk of overlap between the disorders (45) (the shared risk of the two disorders, not their co-morbidities) increased, and the effect of unmeasured factors on the results. There may be similarities between the anxiety-depression co-morbidity and its coefficients. Interestingly, the coefficient of anxiety-depression co-morbidity was the highest because of the coexistence of depressive and anxiety symptoms compared to other disorders, although the number was relatively small.

The strength of this study design was that based on a well-established large cohort from Europe, controlled the confounding factors related to ACE and psychiatric disorders, studied the correlation between each category of ACEs or scores and the comorbidity of common psychological diseases, and most of the results are significant. This suggests that the public should be concerned not only about the risky behaviors (smoking, drinking, etc.) and individual diseases (hypertension, depression, etc.) caused by ACEs in adulthood, but also about the harms caused by the anxiety-depression co-morbidity. Of course, this study also has limitations. First, with respect to disease, self-reported non-cancer diseases were selected in our study section to determine prevalence, which may introduce recall and measurement bias, and the order of diseases after recruitment has not been considered; Second, in terms of confounders, chronic diseases, and genetic factors were not considered; Third, in terms of the independent variable, the ACE scale is not absolutely suitable for European, we did not consider the prevalence of different ACEs combinations and individual ACEs are not graded.

5 Conclusion

In conclusion, although there are many previous studies on the relationship between single psychiatric disorders and ACEs, we should be aware that the potential impact of comorbidity cannot be ignored. The present study showed that with an increase in the number of ACEs or the manifestation of a single ACE, participants had a higher probability of anxiety-depression comorbidity. Therefore, only early intervention of adverse life factors, protection of the emergency attachment system, and control of cortisol hormones can prevent and control public mental health and improve the quality of life.

Data availability statement

Publicly available datasets were analyzed in this study. This data can be found here: this study used the UK Biobank resource with the application ID 88159. Researchers can access the UK Biobank by applying to the UK Biobank official website (https://www.ukbiobank.ac.uk/).

Ethics statement

The UK Biobank database has been approved by the Research Tissue Bank (RTB) with the North West Multicentre Research Ethics Committee (MREC) consent, meaning that each participant does not have to sign a separate consent form and proceeds directly under the approval of the RTB.

Author contributions

KW, YJ, and LH put forward the idea of this research. PZ obtained data. ZJ and PY clean up the dataset and analyze the data. PY and CZ explained the results of data analysis and wrote a manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2023.1233981/full#supplementary-material

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Multi-level analysis of the determinants of physical domestic violence against children using longitudinal data from MINIMat mother–child cohort in Bangladesh

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Objectives: Despite high levels of physical violence against children (VAC) globally (40–50%), the literature on the determinants of VAC remains inconclusive. Most of the literature on this topic is based on cross-sectional data, and the multi-level nature of the drivers of VAC is widely ignored. This leads to model specification problems and an inability to draw causal inferences. Moreover, despite the higher prevalence of VAC in low-and middle-income countries, studies from high income countries dominate the field. We examined the determinants of physical domestic VAC to address these gaps in the literature.

Methods: Data were collected between 2001 and 2020 from 762 mother–child dyads recruited in the Maternal and Infant Nutrition Interventions in Matlab (MINIMat) study in Bangladesh. We conducted multi-level logistic regression analyses to identify the determinants of physical domestic VAC.

Results: Prevalence of physical domestic violence against girls (69%) and boys (62%) was extremely high. Community-level prevalence of physical domestic VAC increased the likelihood of physical domestic VAC at the individual level across gender (girls - OR-5.66; 95% CI- 3.11-10.32; boys - OR-7.67; CI- 3.95-14.91). While physical domestic violence against mothers was not associated with physical domestic violence against girls, it reduced the likelihood of such violence against boys by 47%. Having 3 or more siblings predicted physical domestic violence against girls (OR-1.97; 95% CI- 1.01-3.81 for 3 siblings; OR-4.58; 95% CI- 2.12-9.90 for 4 or more siblings), but not against boys. While girls in Hindu families were more likely to experience this violence, the boys were not. Mother's education, employment non-governmental organization (NGO) participation and, household wealth did not predict this violence against any gender.

Conclusion: We contend that physical domestic violence against mothers reflects an emphasized patriarchal culture in a family where a boy is less likely to experience physical domestic violence. Social norms and social learning theories explain the greater likelihood of a child experiencing physical domestic violence in a village with a higher level of such violence. We conclude that social norms around physical domestic VAC and patriarchal culture need to be changed to effectively address this violence.

KEYWORDS

violence against children, violence against mother, multi-level analysis, rural Bangladesh, gender

1 Introduction

Violence against children (VAC) is a global public health, human rights, and development issue. Globally, 50% of children aged 2–17 experience violence (1). According to UNICEF, around 63% of children ages 2–14 are regularly exposed to physical violence by their caregivers (2). Another systematic review representing 171 countries reports that between 40 and 50% of girls and boys aged 2–14 experienced physical violence in the past month by a caregiver or household member (3). Overall, a higher proportion of boys reported experiencing physical violence than girls (4).

According to the literature, factors commonly associated with VAC are age, sex, mothers' experiences of violence, the mental health of the perpetrator, childhood trauma of the perpetrator, household poverty, and food insecurity (5–10). Although an ecological framework is widely acknowledged to explain VAC (11–15), appropriate analytical methods are often not used to identify the determinants of VAC. Evidence suggesting a clustering of VAC at the community level with rigid social and gender norms endorsing violence and gender inequality significantly contributing to VAC (5–7, 16, 17). Unfortunately, studies often ignore such broader social context (18).

Low- and middle-income countries (LMIC) report relatively higher prevalence of VAC compared to high-income countries (HIC). Most of the literature on this topic, however, comes from the latter (19). According to the nationally representative Multiple Indicator Cluster Survey (MICS) survey conducted in 2019, VAC is pervasive in Bangladesh with 65% of the children aged 1–14 years being ever exposed to physical violence (20). These high rates were accompanied by 35% of the caretaker sample holding the belief that physical punishment is essential for component of child rearing (20). In a study conducted by Mamun et al. in 2022, one in two parents of 10 to 19-year-old children endorsed child beating (21).

According to the Bangladesh Adolescent Health and Well-Being Survey (22) the pattern of physical violence against adolescents is gendered, with a higher proportion of boys reporting it during the last 12 months compared to girls (26% vs. 20%). The same source reports that physical violence against girls was most commonly perpetrated by family members, while the main perpetrators of physical violence against boys were their peers, followed by family members.

There is a paucity of literature on the determinants of physical domestic violence against children in Bangladesh. To our knowledge, the studies exploring correlates of physical VAC in Bangladesh included all perpetrators, regardless of their relationship with the child (23) despite the fact that the drivers of domestic physical violence against children are not likely to be exactly the same as those driving VAC by other perpetrators. These studies recognize that gender is a potential contributor to VAC and thus include gender as an independent variable in the model. This, however, is not enough for identifying correlates of VAC against boys and girls, which are likely to be different. Another serious limitation of these studies is that multi-level modeling appropriate for identifying determinants of outcomes explained by ecological conceptual framework was not used in any of them. Further, these studies were based on cross-sectional data, which inhibited drawing any causal inference. We attempt to address these gaps in the literature by examining the determinants of physical domestic violence against boys and girls using multi-level logistic regression analysis of longitudinal data collected between 2001 and 2020 as part of the Maternal and Infant Nutrition Interventions in Matlab (MINIMat) study conducted in the south-east of Bangladesh.

2 Methods

2.1 Study setting, design, and participants

This study was embedded in a larger longitudinal study well-known as the MINIMat trial (Maternal and infant nutrition interventions, reg#ISRCTN16581394). The details of the study are described elsewhere (24). Briefly, the MINIMat trial is a population-based food and micronutrient supplementation trial for pregnant women. The trial was conducted in Matlab, a predominantly rural sub-district of Bangladesh, where icddr,b (an international research organization based in Bangladesh), has been running a Health and Demographic Surveillance System (HDSS) since 1966. From November 2001 to October 2003, all pregnant women from the HDSS area were invited to participate in the MINIMat trial. The enrolled pregnant women ($n = 4{,}436$) were randomized into two types of food and three types of micronutrient supplementation groups following a two-by-three factorial design. Women were interviewed monthly at home and in the clinics at 14, 19, and 30 weeks of gestation. After delivery of the index child, the mother–child dyads were followed up relatively intensively for two years and later with a greater interval. This analysis includes data collected during pregnancy and at 10- and 18-year follow ups.

Socio-demographic data were collected from the mothers during a household visit at enrolment as well as in follow up interviews. A team of trained paramedics interviewed women at the clinic during the 30th week of gestation regarding their experience of domestic violence (DV). Among the recruited pregnant women, 3,504 completed the DV assessment (Figure 1). The main reasons for loss to follow up were: fetal loss, out-migration, and withdrawal of consent to participate in the study.

Only mothers whose children were born between April 2002 and June 2003, representing a one calendar year birth cohort, were invited to participate in the 10-year follow-up interviews ($n = 1{,}356$) (25). Women were interviewed again regarding their experience of DV during the interim period, using the same standard questionnaire. The women who completed the DV module during pregnancy and the 10-year follow-up were approached for an interview in the 18-year follow-up conducted during 2020–2021. Among them, 1,126 women were successfully interviewed.

The survey of the index children at the 18-year follow up included a module on VAC. A total of 1,001 children completed the interview, resulting in a total of 940 mother–child dyads for this analysis. Since the experience of violence radically differed between married and unmarried children (26), this analysis focuses only on unmarried children. Only singletons were included in the analysis. Thus, we derived a total of 762 mother–child dyads for our analyses, with 422 male and 340 female children (Figure 1).

2.2 Measures

2.2.1 Outcome variables

The outcome variable was lifetime exposure to physical domestic violence among girls and boys. Physical domestic VAC was measured using the 17-item International Society for the Prevention of Child Abuse and Neglect (ISPCAN) Child Abuse Screening Tools (ICAST) (27). Examples of items include slapping, kicking, pulling hair, twisting ears etc. We validated this scale using exploratory factor

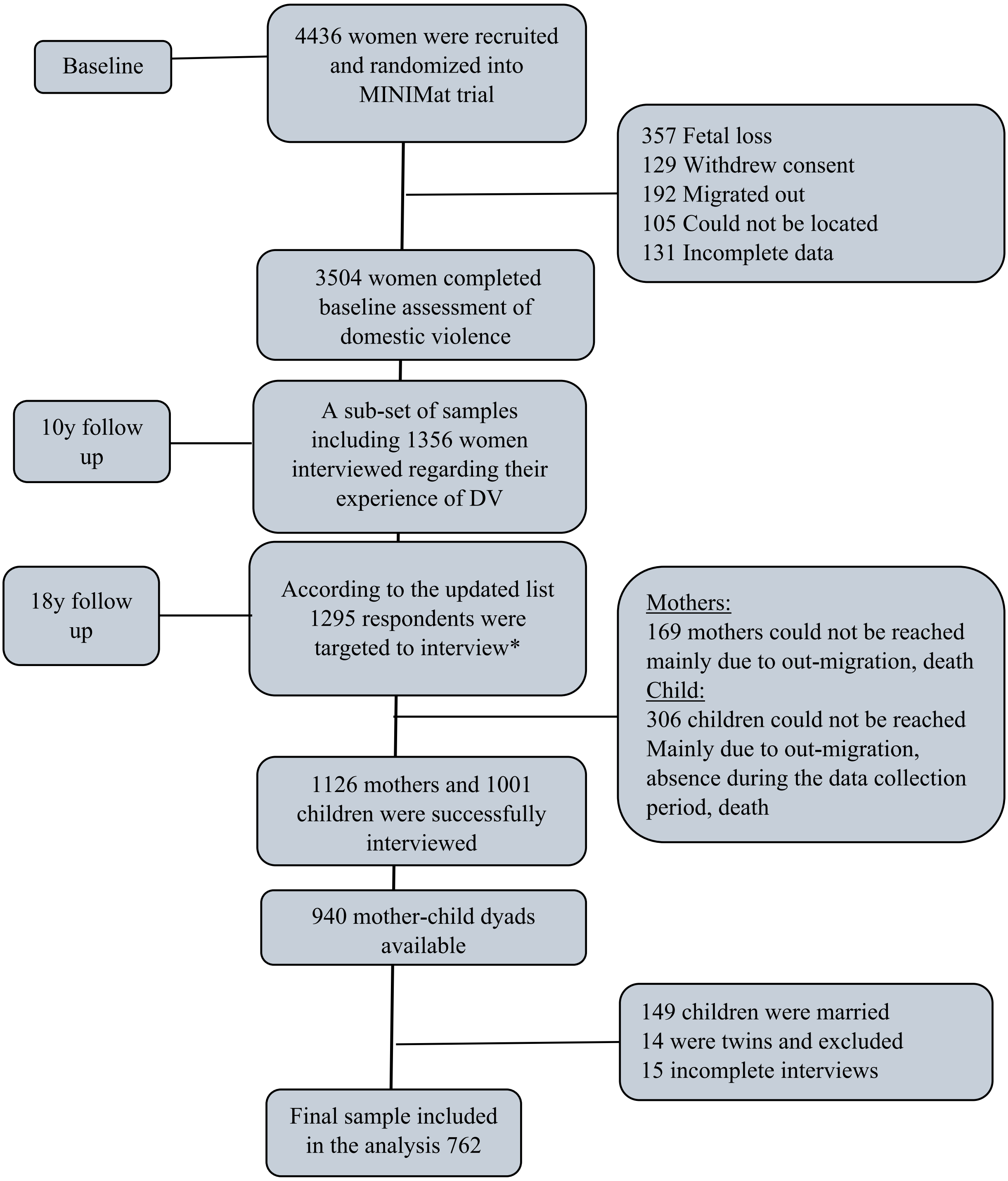

FIGURE 1
CONSORT flow diagram of the number of MINIMat Mother and children in three rounds of data collection between 2001 and 2020. *The list of MINIMat mother and children were updated at 14 years.

analysis (EFA). We performed Q-type EFA, which calculates the factors from the individual responses.

The validated scale retained eight items (e.g., slapping, beating) (See Figure 2) (Cronbach's alpha = 0.70 and KMO = 0.80). A positive response to any of these items was considered as indicating that the child was exposed to lifetime physical domestic violence and was coded as '1 = Yes', otherwise as '0 = No'.

2.2.2 Exposures

In selecting the exposure variables, we considered previous literature, the availability of relevant MINIMat data, and significant bivariate associations between the outcome and exposure variables. To ensure temporality, to the extent possible, we leveraged the longitudinal nature of the data and used lagged variables as covariates. Where such data from previous time point/s were not available we used time invariant exposure

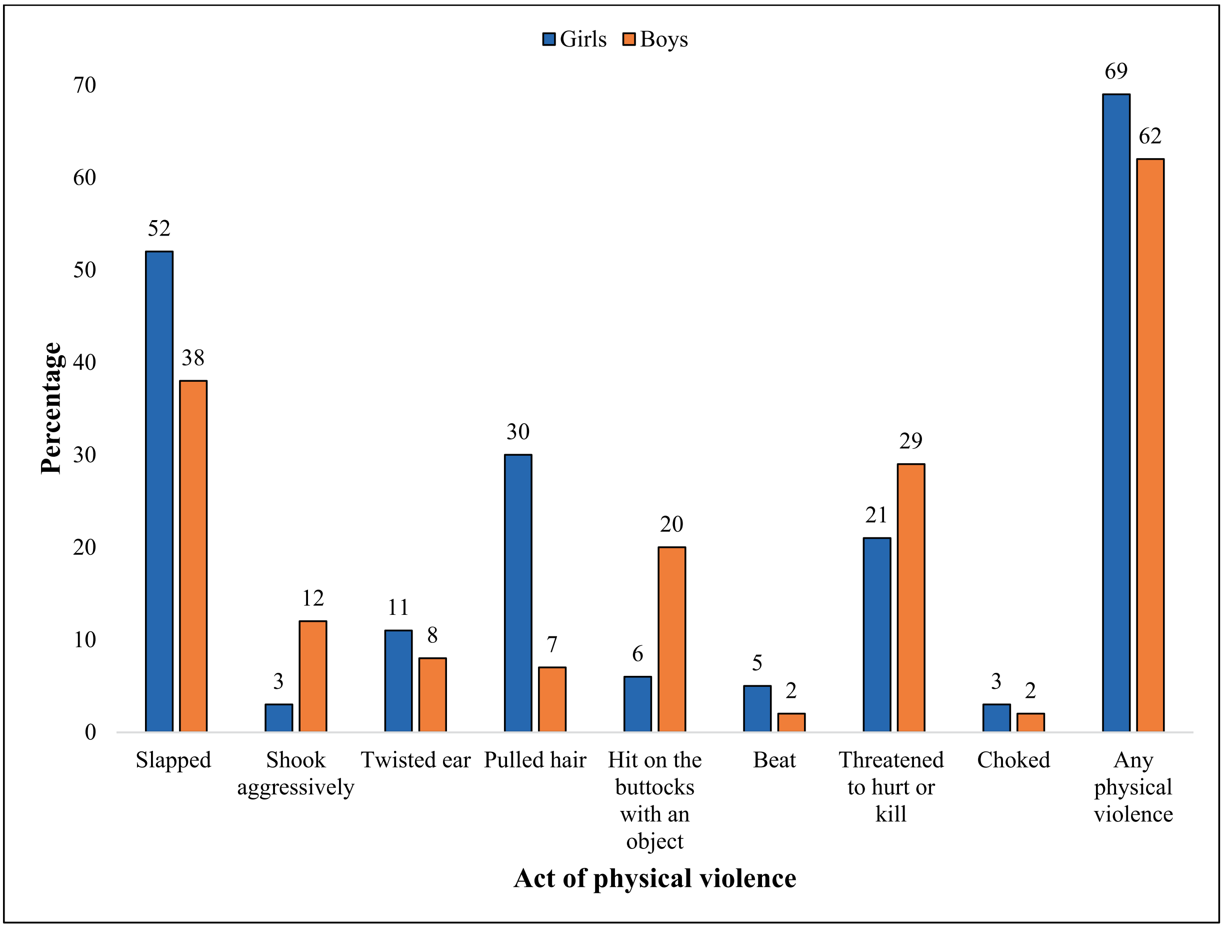

FIGURE 2
Exposure to lifetime physical domestic violence by act and sex (Boys, *N* = 422; Girls, *N* = 340).

variables. We used the measurements that had the lowest missing values and inconsistencies at the three time points. In this paper, we refer to the survey during pregnancy as T1, the 10-year follow up as T2, and the 18-year follow up as T3. The number of siblings (including he/she) of a child (T2) was coded as '1', if she had 1–2 siblings; '2', if she had 3 siblings; and '3, if, she had 4 or more siblings'.

2.2.2.1 Mother's characteristics

Mother's education at T3 was coded as, '1' for no education; '2' for 1–5 years of education; '3' for '6–10 years of education; and '4' for more than '>10 years of education. In the context of Bangladesh, particularly in rural areas, female education usually stops with marriage. Since the education of a woman is usually time invariant, we used it as a proxy for education at an earlier time point in adulthood.

A mother not being employed at T1 was coded as '0' and '1' otherwise. Not participating in any Non-governmental organization (NGO) at T2 was coded as '1'; participation in the microcredit program only as '2'; participation in other types of NGO as '3'; participation in both types of NGOs as '4'.

A modified version of the conflict tactic scale (28) was used to measure the mother's lifetime experience of physical DV (T1). A total of seven items (e.g., slapping, kicking, choking, or burning) were used to measure physical DV. A woman responding positively to any of these questions was treated as exposed to physical DV and coded as '1', otherwise as '0'.

2.2.2.2 Household characteristics

An extended household at T1 was coded as '1' and a nuclear household as '0'. Household wealth quintiles at T1 were derived by dividing the household asset scores obtained from principal component analysis into five categories. The categories were as follows: poor (1), lower middle (2), middle (3), upper middle (4), and rich (5). Families pursuing Islam at T3 were coded as '0' and Hinduism as '1'.

2.2.2.3 Community characteristics

The prevalence rate of physical domestic violence against girls and boys at the community-level at T3 was calculated separately for boys and girls using the same procedure. First, the number of individuals exposed to physical domestic violence in a village was calculated, divided by the sample size in the village, and then multiplied by 100. For 31–40% of the villages, the rate of community-level physical domestic VAC was more than 75%. We have coded villages with such high rates of physical domestic VAC as '1' and otherwise as '0'.

2.3 Statistical analyses

Descriptive analyses were performed to describe the background characteristics of the study participants and the prevalence of physical domestic VAC. We examined differences between the background characteristics of girls and boys using chi-square tests for categorical variables and t-tests for continuous variables. Multi-level regression analyses were conducted separately for the boy and girl samples. At first, a null model (Model 1) was developed to estimate the community level variance to justify using the multi-level logistic regression model. The intra-cluster correlation (ICC) value was estimated at 0.18 and 0.03 for boys and girls, respectively, implying that community-level factors can explain 18% for boys and 3% for girls of the total variation in physical domestic VAC. Second, in Model 2, individual-level factors were incorporated. Finally, in model 3, community level variables were included. All the analyses were performed using STATA version 15, and the significance level for all statistical tests was set at 5%.

2.4 Ethical considerations

All rounds of the MINIMat trial (PR-2000-025, PR-12022, and PR-19101) were approved by icddr,b's institutional review board. The third round was additionally approved by Swedish Research Ethics Authority (# 2021–00523).

3 Results

3.1 Characteristics of the study sample

Table 1 shows the characteristics of the study participants by sex. The mean age for both sexes was 17.5 years. Around 20% of the mothers had no education, and only 8% had education beyond ten years. The number of siblings was significantly higher among mothers of girls compared to those of boys. About 8% of the mothers were employed. Regardless of the sex of the index child, approximately, 66% of the mothers were NGO members. A significantly higher proportion of boys' mothers reported lifetime physical DV when interviewed in the pregnancy with the index child, compared to the mothers of girls (25% vs. 17%). About 38% of the mothers came from a nuclear family at T1. Household socio-economic status was significantly different for the boys and the girls with a higher proportion of girls coming from better off families. The samples were predominantly Muslim (85%). A higher proportion of the girls (40%) came from a community with high prevalence of physical domestic violence against girls (i.e., >=75%), compared to proportion of boys (31%) living in a community with high prevalence of physical domestic violence against boys.

As shown in Figure 2, the prevalence of lifetime physical domestic violence was 69% among the girls and 63% among the boys. Slapping was the most common act of physical domestic violence across sexes (38–52%) and choking – the least common (2–3%). A higher proportion of girls experienced all moderate acts of physical domestic VAC (e.g., such as slapping, shaking, ear and hair pulling). Exposure to beatings was more common among girls than boys. The two acts to which boys were more exposed to than the girls were spanking (20% vs. 6%) and experiencing threats to hurt or kill them (29% vs. 21%).

3.2 The determinants of lifetime physical domestic violence against children

Table 2 presents the results of the multi-level logistic regression analyses of the determinants of physical domestic VAC by sex. Judging by the size of the Akaike information criterion (AIC) in the three sets of models run for each sex, it is evident that the Model 3, where both individual/household-and community-level factors were included, shows the best fit for both girl and boy samples. The ICC in the final model for the girls was reduced from 0.08 in model 2 to 1.80e-34 which implied that the prevalence of physical domestic violence against girls in the community explained almost all the community-level variations in the physical domestic violence among girls. The ICC values in the three sets of models run on the boys' sample (0.18 in Model 2 vs. 0.04 in Model 3) showed that in Model 3, 14% of the community-level variations in physical domestic violence against boys could be explained by the community-level prevalence of physical domestic violence against them. Clearly, Model 3 provided the best estimates of the determinants of physical domestic VAC for each sex.

Model 3 shows that some household-level factors also predicted physical domestic violence against boys and girls. Thus, the risks of physical domestic violence increased with the number of siblings in the girl sample. Thus, compared to the girls who had 1–2 siblings, the girls who had three, or four or more siblings were more likely to experience physical domestic violence (OR-1.97; 95% CI- 1.01-3.81 in case of 3 siblings; OR-4.58; 95% CI- 2.12-9.90 in case of 4 or more siblings). The number of siblings had no effect, however, on the boys' exposure to this violence. Mother's experience of lifetime physical DV up to pregnancy with the index child did not affect girls' exposure to this violence, while it reduced the risk of physical domestic violence among boys by 47% (OR-0.53; 95% CI- 0.31-0.92). Living in an extended family decreased the risks of physical domestic violence among boys 46% (OR-0.54; 95% CI- 0.33-0.89), while it did not affect the girls. Girls from Hindu families were at three times higher risk of being physically abused by family members compared to their Muslim counterparts (OR-2.99; 95% CI- 1.25–7.20). Religion, did not have any impact on the boy's exposure to physical domestic violence.

In communities where the prevalence of physical domestic violence was 75% or more among girls, the likelihood of physical domestic violence was six times higher among girls (OR-5.66; 95% CI- 3.11-10.32). It was eight times higher among boys (OR-7.67; 95% CI- 3.95-14.91) in communities with 75% or higher prevalence among boys compared to communities with a lower prevalence rate.

4 Discussion

Our findings show higher prevalence of physical domestic violence among boys and girls in this sample (65%) compared to many other countries (9, 29). While the prevalence of physical domestic violence among boys is commonly reported to be higher than among girls (5), our findings show the opposite picture. We argue that this is not surprising given the patriarchal setting characterized by strong son preference and male privilege (30).

Our findings offer a deeper insight into the predictors of physical domestic violence against children by fitting separate models for boys and girls and by performing multi-level analyses. Thus, while the previous literature suggests that large family size (31) and greater number of siblings (32) increase the likelihood of VAC, our findings show that having a higher number of siblings increased the likelihood of physical domestic violence among girls, but not among boys. While the first may be due to increased stress on household resources and particularly on the mother's time in juggling household responsibilities, the latter may highlight the privileged position of a son.

TABLE 1 Background Characteristics of the sample by child sex, $N = 762$.

	All sample, % (*n*) (*N* = 762)	Girls, % (*n*) (*N* = 340)	Boys, % (*n*) (*N* = 422)	*p*-value*
Child characteristics				
Mean age (SD, range)	17.46 (0.51, 16–18)	17.46 (0.52, 16–18)	17.46 (0.51, 16–18)	0.968
Lifetime exposure of children to physical domestic VAC	64.83 (494)	68.53 (233)	61.85 (261)	0.055
Number of siblings				
1–2	31.50 (240)	29.41 (100)	33.18 (140)	
3	39.76 (303)	35.00 (119)	43.60 (184)	0.001
4 and above	28.74 (219)	35.59 (121)	23.22 (98)	
Mother's characteristics				
Mother's Education				
No education	20.21 (154)	20.88 (71)	19.67 (83)	
1–5 years	34.91 (266)	32.94 (112)	36.49 (154)	0.790
6–10 years	37.14 (283)	38.24 (130)	36.26 (153)	
11–12 years	7.74 (59)	7.94 (27)	7.58 (32)	
Mother's employment status				
Yes	8.01 (61)	6.18 (21)	9.48 (40)	0.095
No	91.9 (701)	93.82 (319)	90.52 (382)	
NGO membership				
None	33.86 (258)	34.41 (117)	33.41 (141)	
Micro-credit only	19.29 (147)	17.06 (58)	21.09 (89)	0.451
Other NGO	28.22 (215)	30.29 (103)	26.54 (112)	
Both	18.64 (142)	18.24 (62)	18.96 (80)	
Mother's exposure to physical DV	21.39 (163)	16.76 (57)	25.12 (106)	0.005
Household characteristics				
Family structure, Nuclear	38.19 (291)	36.47 (124)	39.57 (167)	0.381
Wealth index				
Poor	20.21 (154)	20.00 (68)	20.38 (86)	
Lower middle	22.31 (170)	20.29 (69)	23.93 (101)	
Middle	23.75 (181)	22.65 (77)	24.64 (104)	0.022
Upper middle	17.19 (131)	15.59 (53)	18.48 (78)	
Rich	16.54 (126)	21.47 (73)	12.56 (53)	
Religion				
Muslim	85.04 (648)	85.00 (289)	85.07 (359)	0.978
Hindu	14.96 (114)	15.00 (51)	14.93 (63)	
Community-level characteristics				
Rate of physical domestic VAC >75%, %	35.17 (268)	40.00 (136)	31.28 (132)	0.012

**p*-values are based on a t-test for continuous variables and $\chi2$ tests for categorical variables, comparing frequencies of variables by gender.

A study conducted in Agartala, India by Deb & Modak suggests that extended family protects children against physical domestic violence (33). This, however, was not substantiated by another study conducted in Jammu, India (34). Our findings are more nuanced and show that the extended family protected the boys against physical domestic violence, but not the girls. This may be explained by the following. Marriages are patrilocal in Bangladesh. When a female gets married, she usually joins an extended marital family. Eventually most of the extended families split to form nuclear families (35). It is plausible that representative/s of the older generation in an extended household hold great power, and at the same time they may hold more tightly on to patriarchal ideologies and practices that tend to protect boys from being physically abused by family members, but not girls. Differences in findings from different settings may suggest importance

TABLE 2 **Determinants of lifetime physical domestic violence against children aged 16–18 years: results from the multi-level logistic regression models, *N* = 762.**

	Girls (*N* = 340)			Boys (*N* = 422)		
Variables	Model 1 (Null model)	Model 2 (Null + individual/ household level covariates)	Model 3 (Null + individual/ household+ village level covariates)	Model 1 (Null model)	Model 2 (Null + individual/ household level covariates)	Model 3 (Null + individual/ household+ village level covariates)
	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Individual/ household level variables						
Mother's Education (T3)						
No education (ref)						
1–5 years		1.37 (0.63–2.98)	1.32 (0.60–2.90)		1.75 (0.92–3.32)	1.57 (0.83–2.96)
6–10 years		2.04 (0.82–5.08)	2.15 (0.86–5.41)		1.19 (0.58–2.43)	1.01 (0.49–2.05)
11–12 years		2.86 (0.71–11.50)	2.66 (0.63–11.17)		2.23 (0.63–6.51)	1.71 (0.55–5.33)
Number of siblings (T2)						
1–2 (ref)						
3		1.85 (0.97–3.53)	1.97 (1.01–3.81)*		1.65 (0.95–2.85)	1.61 (0.94–2.78)
4 or more		4.18 (1.94–8.99)*	4.58 (2.12–9.90)*		2.08 (1.04–4.17)*	1.85 (0.93–3.67)
Mother's employment status (T1)						
No (ref)						
Yes		0.90 (0.30–2.67)	1.01 (0.34–3.00)		1.61 (0.70–3.68)	1.54 (0.68–3.46)
Mother's NGO participation (T2)						
None (ref)						
Microcredit program only		1.67 (0.75–3.72)	1.44 (0.65–3.18)		0.75 (0.39–1.43)	0.60 (0.31–1.16)
Other NGO		0.97 (0.43–1.76)	0.95 (0.49–1.82)		1.14 (0.62–2.08)	1.04 (0.58–1.88)
Both		1.39 (0.65–2.98)	1.36 (0.63–2.91)		0.67 (0.35–1.29)	0.58 (0.30–1.12)
Lifetime physical domestic violence against mothers (T1)						
No (ref)						
Yes		0.87 (0.42–1.76)	0.82 (0.40–1.68)		0.48 (0.28–0.84)*	0.53 (0.31–0.92)*
Family structure (T1)						
Nuclear (ref)						
Extended		0.89 (0.49–1.59)	0.80 (0.44–1.44)		0.59 (0.36–0.96)*	0.54 (0.33–0.89)*
Wealth index (T1)						
Poor (ref)						
Lower middle		1.80 (0.77–4.25)	1.90 (0.80–4.51)		0.69 (0.34–1.38)	0.75 (0.38–1.50)
Middle		2.17 (0.88–5.30)	2.11 (0.86–5.22)		1.03 (0.49–2.16)	1.20 (0.57–2.51)
Upper middle		1.10 (0.41–2.99)	1.04 (0.38–2.83)		0.94 (0.41–2.15)	1.03 (0.45–2.34)

(Continued)

TABLE 2 (Continued)

	Girls (*N* = 340)			Boys (*N* = 422)		
Variables	Model 1 (Null model)	Model 2 (Null + individual/ household level covariates)	Model 3 (Null + individual/ household+ village level covariates)	Model 1 (Null model)	Model 2 (Null + individual/ household level covariates)	Model 3 (Null + individual/ household+ village level covariates)
	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Rich		1.25 (0.44–3.56)	1.14 (0.39–3.30)		1.17 (0.45–3.05)	1.20 (0.46–3.12)
Religion (T3)						
Muslim (ref)						
Hindu		3.51 (1.36–9.04)*	2.99 (1.25–7.20)*		0.97 (0.46–2.08)	1.10 (0.54–2.21)
Community level variable						
Prevalence of physical domestic VAC (T3)						
Lower rates (ref)						
Higher rates			5.66 (3.11–10.32)*			7.67 (3.95–14.91)*
Random effect						
Estimate (Village level variation)	0.09	0.29	5.92e-34	0.72	0.70	0.14
ICC	0.03	0.08	1.80e-34	0.18	0.18	0.04
Intercept	2.21 (1.71–2.86) *	0.38 (0.13–1.11)	0.21 (0.07–0.60)*	1.70 (1.23–2.35)*	0.96 (0.37–2.49)	0.59 (0.23–1.53)
Model statistic						
AIC	427.02	429.10	393.21	546.47	554.81	516.36

*$p < 0.05$.

of the contextual differences. More importantly, our findings clearly show that the same factor may have differential effect on physical domestic violence against different genders and thus, results of analyses pooling both genders might may mask a different reality.

In contrast to many studies conducted both in developed and developing countries, poverty (6, 18, 36) and maternal education did not come out as predictors of physical domestic VAC in our study. This may indicate that this violence actually cuts across all households and all maternal education categories in this low educated patriarchal context dominated by age and gender hierarchies.

The finding that the Hindu girls were at higher risk of physical domestic violence compared to Muslims may be explained by the fact that as a minority group, Hindu families may face greater challenges in protecting the girls' chastity linked family honour. Thus, they may be more likely to subject the girls to physical abuse for the purpose of controlling and disciplining them (37).

The literature presents compelling evidence on the intersections between violence against women and VAC (6, 37). Our findings are, however, nuanced and support the existing literature only partially. In contrast to the previous literature that suggests that violence against women increases the likelihood of VAC (38, 39), we have found an effect of violence against mothers on physical domestic violence among boys, but not among girls. Moreover, the relationship between the two found in this study contradicts the literature. Thus, violence against the mother in a family actually reduced physical domestic violence among boys. Our findings suggest that the nature of interactions between physical domestic violence against mothers and domestic VAC may be context specific and may not go in the same direction across settings. We argue that families where women are physically abused, practice emphasized patriarchy. Thus, in these families, sons were more privileged and, accordingly, were protected against physical domestic violence. The likelihood of physical abuse of girls in such families did not increase, but neither did it reduce as in case of the boys. Thus, it is important to underline that physical domestic violence against mothers is embedded in gender inequality, which in turn generates greater gender inequality in how male and female children are treated in the family.

Our results underline the importance of multi-level modeling of determinants of physical domestic VAC showing that almost one-fifth of the variations were explained by the community-level factors among the boys. This echoes claims made by other researchers (10). We find that community-level rates of physical domestic violence actually explain this variation almost in its entirety. Thus, 75% or higher prevalence of physical domestic VAC in the community increased the likelihood of this violence across genders (8 times for boys and 6 times for girls). The high magnitude of this effect of community level physical domestic VAC on individual boys and girls, is noteworthy. This finding is in line with social norms (40) and social learning theory (41). According to social norms theory an individual in a particular social gendered context learns to define, imitate, and receive reinforcement for his/her behaviors from the larger society/community (42). As Hall suggests violence is a socially learned behaviour and individuals exposed to violence are more likely to perpetrate it. This highlights the importance of addressing social norms around domestic VAC in the community, which are largely contributing to physical domestic violence against both boys and girls.

This study suffers from some limitations. Violence always tends to be underreported and VAC reported by children is no exception (43). Underreporting may vary by gender, which may introduce measurement errors and compromise comparability across gender. It is noteworthy, that our study is one of the very few studies in Bangladesh that collecting data on VAC directly from the children. This is a strength of our study since there is evidence that underreporting of VAC is likely to be higher when data are collected from the parents (44). Our study carefully followed strategies for enhancing disclosure of violence by ensuring confidentiality, taking interviews in private and in a non-judgemental manner using validated standard tools. The data on lifetime physical domestic VAC were collected retrospectively in this study, which raises concerns regarding recall bias.

This analysis included both primary and secondary data. Consequently, the choice of covariates was constrained by the availability of information. However, strengths of this study include more in-depth and nuanced understanding of how different factors predict physical domestic violence against boys and girls. The findings clearly demonstrate that the same factor may have different effect on the outcome when separate models are run for boys and girls. This finding highlights that it is critical to conduct gender segregated analyses of predictors of VAC so that the nuances introduced by gender can be captured. Methodological strengths of this study include as well use of longitudinal data and the careful choice of covariates, paying attention to the temporality of the events. Moreover, the use of multi-level modeling enabled us to come up with robust estimates and allowed us to explain the variations to a large extent. Further, findings from this study indicate that in this setting with very widespread physical domestic violence among boys and girls, it is absolutely necessary to address violence conducive social norms so that VAC in the home is reduced. It is also important to address gender inequality and ensure equal treatment for both boys and girls.

Data availability statement

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

Ethics statement

The studies involving humans were approved by icddr,b's institutional review board and Swedish Research Ethics Authority. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin because we collected data on respondents violence. We did not want to keep any signature that can identify the respondents. Therefore we took oral consent.

Author contributions

The study was conceived by RTN and designed by RTN and KP. RTN, JFA, KP, and SZ contributed to model construction. The data were analysed by JFA under the guidance of RTN. The manuscript was drafted by RTN, JFA, and KP and critically reviewed for important intellectual content by all authors. JFA attests that all listed authors meet authorship criteria and that no others meeting the criteria have been omitted. The guarantor (RTN) accepts full responsibility for the work, she accessed the data, and controlled the decision to publish. All authors contributed to the article and approved the submitted version.

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Conflict of interest

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

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1185130/full#supplementary-material

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Case conceptualization in child welfare: an underused resource to improve child, family, and provider outcomes

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Case conceptualization, formally known as case formulation, is one tool that assists in determining the best course of action for children and families experiencing family violence that has been under-utilized in child welfare. In this article we present a step-by-step case conceptualization process that considers the child welfare context. We then present a hypothetical case example of a 10-year-old child referred by a child welfare worker to evidence-based treatment for mental health and behavioural concerns. Mental health services are not helpful for the child and further consultation is enlisted. To more effectively guide intervention and treatment planning and ultimately improve outcomes for the child, we present case conceptualization as a process that incorporates relevant aspects of the child and family's history and circumstance. We conclude with a succinct case conceptualization and treatment plan to show how the prognosis of the child can be improved when case conceptualization is employed.

KEYWORDS

child maltreatment, case conceptualization, child welfare, child mental health, intimate partner violence, family violence

1 Introduction

Children and families coming into contact with child welfare can represent some of the most complex cases for healthcare and social service providers to support, often due to interlocking socioecological factors, such as poverty, poor integration of services, high turnover of child welfare workers, poor family function, and intergenerational histories of trauma (1–14). Outcomes for children experiencing maltreatment are often framed as "gloomy" (15) and to date it is unclear if contact with child welfare improves child and family outcomes (9, 16). While a variety of evidence-based services for children and families exist, it can be challenging to determine the best course of action for treatment or support due to the complex needs of such children and families. Case conceptualization, formally known as case formulation, is one tool that can assist providers in determining the best course of action for supporting children and families that has been under-utilized in child welfare. As case conceptualization is an important clinical tool that can assist in improving client/patient outcomes (17) as well as provider outcomes (18, 19) [e.g., increased tolerance for uncertainty (20)], this commentary offers a succinct summary of the components of case conceptualization and gives examples of its application to child welfare. First, we discuss the importance of applying trauma- and violence- informed care principles to child welfare responses and the need for children involved in child welfare to undergo a comprehensive assessment in order to identify their needs and appropriate services. Second, definitions and important components of case conceptualizations are described. Third, a case example, 'Rose', is presented where evidence-based services are offered to a child; however, as the unique needs of the child and family were not first conceptualized in this case example, these services did not improve the child's outcomes. The commentary then moves to a reconsideration of the case example to illustrate how to carefully tailor interventions to the unique circumstances of the child and family.

2 The importance of trauma- and violence-informed care

There are several important principles that inform safe and effective work with children and families involved with child welfare. Providers need to consider these principles at all phases of assessment and intervention, including when doing a case conceptualization. Some of these principles include the need for support and services to be evidence-based; tailored to the specific family/child including to the child's age and stage of development; culturally sensitive; trauma-informed; comprehensive; and strengths-based (21). For example, principles of trauma-informed practice are increasingly incorporated into core competencies for healthcare and social service providers supporting children and families involved in child welfare (22–24). Trauma-informed care is a "whole system organizational change process which seeks to embed theoretically coherent models of practice across diverse settings and roles, including child welfare, family support, justice, mental health and education" (25). Trauma-informed principles have been incorporated into child welfare in a variety of ways, including through workforce development (e.g., training staff to understand the impact of trauma), service delivery (e.g., recognizing and integrating the child's trauma history in case planning), and organizational change (e.g., increasing collaboration and information sharing) (25). There have been several critiques of trauma-informed care. For example, some authors suggest that it has a "relatively narrow definition of trauma that implicitly emphasizes violence between individuals," it emphasizes "medical environments at the exclusion of others (e.g., legal systems, social services, educational systems, economic structures)," and it has an "implicit assumption that trauma affects everyone in the same way" (26). Some authors suggest that a lack of prioritization of these concerns has contributed to the pathologizing of individuals who have experienced interpersonal and structural violence (27).

In response to such criticisms, trauma- and violence-informed care was developed to draw attention to historic, intergenerational, and structural violence. Trauma- and violence-informed care "extends the trauma-informed care framework with the addition of 'violence' to emphasize the association between trauma and violence" (28, 29). Trauma- and violence-informed care addresses individual and broader systems-level aspects of the care encounter, including the environment of the care encounter (e.g., is the environment safe and welcoming?), the approach the provider uses (e.g., is the provider trained in how to respond safely to family violence?), and the provider's response to the patient in the encounter (28). Trauma- and violence-informed care is intersectional, in that it considers an individual's past and current experiences of trauma and how they intersect with past and current experiences of systematic or structural violence (e.g., racism, colonialism) (29, 30). Trauma- and violence-informed care is also attentive to the additive effects of trauma, in that it emphasizes how past, current, interpersonal, and structural violence can overlap to produce significant, negative health consequences (29, 30).

Principles of trauma- and violence-informed care can inform the approach to the case conceptualization, including 1) the recognition by providers of the high prevalence of family violence [defined as violence, abuse, conflict or neglect by a family member toward a family member that is associated with poor health (31)] and its impact on child well-being, 2) an emphasis by providers on the need to reduce possible victimization and future harm experienced by the child in their family and in their interactions with the provider; and 3) attention to physical, emotional, and cultural safety in all interactions between providers and clients (28). For example, for children involved in child welfare who are referred to mental health services, it is important to consider if past, current, and ongoing experiences of family violence may be influencing behavioural problems and if children have physical and psychological safety in their home environment in order to benefit from mental health support at the time. Many healthcare and social service providers are comfortable making referrals to counseling or psychiatric support for psychological and behavioural goals. However, this may not address the root causes of the child's concerning behaviours and may not be in the best interest of the child if there is not physical and psychological safety within the family context (e.g., quality of relationships, family dynamics) at the time of service provision. For a case conceptualization and future treatment to be effective, children must first undergo a comprehensive assessment.

3 Comprehensive assessments for children involved in child welfare

The purpose of a comprehensive assessment is to identify key aspects of a child's life and to consider evidence-based referrals that reflect the unique needs of the child, including their age and developmental stage (32). A trauma- and violence informed approach to comprehensive assessment would include attention to the environment within which the comprehensive assessment takes place (e.g., is the assessment environment safe and welcoming?), the provider's approach to assessment before the encounter occurs (e.g., is the provider trained in how to provide a safe response to disclosures of family violence, are they culturally respectful, are they aware of the intersections between experiences of structural and interpersonal violence?), and the provider's specific responses to children and families in the assessment (e.g., do they establish rapport with the child and family, do they consider their physical proximity when speaking with a child and family, do they ask the child's preferred name, do they explain the limits of confidentiality in a developmentally appropriate way, are they non-judgmental, do they respect the inherent dignity of the child and family?) (28).

A comprehensive assessment should take into account the child's home situation (e.g., description of people in the family, present living situation, extended kinship networks, any non-traditional familial relationships); their general development (e.g., cognitive, emotional, physical); their education (e.g., school, grades, teachers); their involvement with activities (e.g., recreation); their mental health, including questions about symptoms (including post-traumatic stress) and general functioning (e.g., well-being, sleep, physical health, peer relationships); their experiences of family violence and other adversities; and any other relevant aspects of their lives (e.g., substance use, sexual health) (28). With regard to asking about exposure to maltreatment, care should be taken not to overlap with the role of the child welfare worker; if the child discloses previously unreported information about suspected maltreatment, investigation of this information will typically be undertaken by the child welfare worker. Assessments should also consider relevant information about people of importance to the child (e.g., their siblings and their main caregivers' personal, social, and health history; other caring adults; friendships). Assessments should enable providers to communicate with children alone, as well as time to observe them with their primary caregiver(s) (28). Gathering this information is necessary for developing an accurate case conceptualization (33). In many cases, referral to health services is informed by an assessment of an individual; however, especially when child welfare is involved, providers must understand family dynamics, including how the child interacts with their siblings and caregivers and the caregivers' and siblings' own histories. From this assessment, providers will be better able to assess the suitability of services and supports for the child in the context of their family.

4 Case conceptualization: definition and components

After a comprehensive assessment has been completed, healthcare and social service providers can build a case conceptualization incorporating all aspects of their interactions with the child or family, as well as any additional collateral information (for example, from teachers). Case conceptualization can be defined as "a clinical strategy for obtaining and organizing information about a client, explaining the client's situation and maladaptive patterns, guiding and focusing treatment, anticipating challenges and barriers, and preparing for successful termination" (34). It is essentially a comprehensive map for treatment and support, which includes an overview of the child or family's concerning symptoms and behaviours (diagnostic formulation); how these symptoms and behaviours came to be and are understood (clinical formulation); an understanding of the role of culture in the case (cultural formulation); and a treatment or support map that links specific child/caregiver problems with evidence-based services or supports (treatment formulation) (35, 36).

4.1 What symptoms and behaviours are the child or family struggling with (diagnostic assessment/formulation)?

When children and families present with behavioural and/or emotional problems, a diagnostic assessment/formulation is often prioritized in most healthcare and social service settings (including child welfare). This typically entails a description of the client's presenting situation (e.g., concerning symptoms and behaviours) and any factors that prompt these concerns (35). A diagnostic assessment often leads to one or more diagnoses based on the Diagnostic and Statistical Manual of Mental Disorders (DSM). Many children involved in child welfare meet diagnostic criteria for a range of diagnoses, such as oppositional defiant disorder (ODD), conduct disorder, major depressive disorder, and/or reactive attachment disorder (37). Behaviours associated with these diagnoses are understandable given their experiences in unstable, chaotic, or unsafe home environments (38). In other instances, however, children exposed to family violence may not meet full diagnostic criteria (e.g., post-traumatic stress disorder) yet struggle significantly. In either situation, case conceptualization provides a much richer understanding of what contributes to, and maintains, a child's emotional or behavioural difficulties. While diagnoses may be useful or necessary for accessing specific services, they are often limited to a summary of a child's symptoms. A comprehensive case conceptualization subsumes any such diagnoses and better informs treatment planning, as it helps the provider shift from description to explanation and understanding.

For example, many children who experience child maltreatment, including exposure to intimate partner violence, have complex changes to brain development (e.g., impaired stress response), cognition (e.g., language delays, problems with concentration), behaviour (e.g., poor self-regulation, social withdrawal), mental health (e.g., depression, anxiety), relationships (e.g., poor understanding of social interactions), emotions (e.g., difficulty controlling emotions), and physical health (e.g., sleep disorders) (39–43). As referenced above, many maltreatment-related behavioural, emotional, and relational changes that children experience overlap with other common diagnoses, such as ODD (see Table 1). For example, the National Institute for Health and Care Excellence details over 70 indicators of child maltreatment. Behavioural indicators like "markedly oppositional behaviour," emotional indicators like "repeated, extreme or sustained emotional responses," and relationship indicators like "coercive controlling behaviour towards parents or carers" are closely related to the diagnostic criteria for ODD. Signs and symptoms of other disorders also overlap with maltreatment symptoms. For example, signs of attention deficit hyperactivity disorder (ADHD) (e.g., agitation, poor self-esteem, difficulties concentrating, and difficulties with work, school and sleep) are common in children who have experienced maltreatment (32, 38, 46, 47). When maltreatment-related symptoms are not recognized or treated inappropriately—for example, if symptoms are inappropriately treated with pharmacological interventions—the relational injuries that are underlying the child's symptoms are not addressed (38). Following from a comprehensive assessment, such behaviours can be better understood as traumatic stress reactions given exposures to family violence. Developmental trauma disorder has long been proposed by clinicians and researchers as a way to capture the clinical presentation of children who have been exposed to chronic interpersonal trauma (48–50).

In addition, child welfare workers may become aware of parental mental health concerns with symptoms consistent with disorders, such as borderline personality disorder or narcissistic personality disorder. While understanding a person's diagnosis may be helpful, building a treatment plan from a diagnosis only is an example of "backward reasoning," which involves creating a hypothesis about treatment and then scanning to find supporting data (51). This reasoning involves a narrowing of options and does not effectively

TABLE 1 Comparing indicators of maltreatment (44) to ODD diagnostic criteria (45).

Area of child development	Examples of signs of child maltreatment (44)	Examples of diagnostic criteria for ODD (45)
Behaviour	• "Markedly oppositional behaviour" • "Aggressive, oppositional"	• "Often actively defies or refuses to comply with requests from authority figures or with rules"
Emotions	• "Extreme distress" • "Lack of ability to understand and recognize emotions" • "Repeated, extreme or sustained emotional responses" • "Anger or frustration expressed as a temper tantrum in a school-aged child"	• "Is often touchy or easily annoyed" • "Often loses temper" • "Is often angry and resentful"
Relationships	• "Coercive controlling behaviour towards parents or carers"	• "Often argues with authority figures or, for children and adolescents, with adults"

address the unique aspects of the client's life. A more helpful strategy in case planning is to use "forward reasoning," or to use specific incidents presented by the client (or observed by the practitioner) to develop hypotheses (51). For example, if during the course of the comprehensive assessment, it was observed that a mother and her children were reluctant to speak in front of a father who appeared agitated and directed most of the conversation, and the clinician was aware of a history of reports to child welfare for intimate partner violence (IPV), the clinician might surmise that various safety strategies were needed early on when working with this family. The clinician would need to prioritize assessing the safety of the mother and the children before specific clinical services or supports could be offered. In this example, the clinician might also assume that couples' therapy was contraindicated given potential safety concerns arising from past (and potentially current) IPV (52).

4.2 Why did it happen to this child or family (clinical assessment/formulation)?

An essential component of a case conceptualization is a clinical formulation, which investigates what happened to this child to explain the "why" of the child or family's presenting concerns, given their history or current life stressors. Attention to the *why* of behaviour is a principle of trauma- and violence-informed care, as given the high prevalence of family violence it is important to consider how child and parent behaviours make sense given their potential history of family violence (29). Clinical formulations provide an explanation of a client's behaviour, usually through a particular theoretical lens, such as through a bio-psycho-social-spiritual lens or a cognitive-behavioural lens. The discussion below does not represent an exhaustive list of the theoretical frameworks/lenses that can inform a conceptualization but provides examples that may be helpful to characterize how case conceptualization must attend to both broad (structural) concerns as well as individual concerns. While not discussed in the present manuscript, other theoretical lenses for case conceptualization are available, such as the Attachment, Self-Regulation, Competency (ARC) model, are available (33, 36, 51, 53, 54).

A socioecological (also sometimes referred to as a critical ecological or ecological) lens is especially relevant when undertaking a case conceptualization for children and families involved in child welfare, as this model is commonly used in violence prevention research to outline the range of factors influencing risk and prevention of violence (55). This model, for example, attends to risk and protective factors at the societal level (e.g., societal norms regarding physical punishment); community level (e.g., availability of coordinated services for children and families); institutional level (e.g., level of support from child welfare workers); relationship-level (e.g., caregiver-child attunement); and individual level (e.g., caregiver mental health concerns). For example, a recent systematic scoping review found that youth (15+) and adults of colour accessing sexual assault services experienced many barriers to care, including a lack of access to diverse staff at sexual assault services and experiences of discrimination/racism from white service providers (56). Absence of culturally safe services at the community level is a risk to children and families, in terms of effective treatment planning; providers also have an opportunity to advocate for meaningful services for children, youth, and families in these circumstances. The socioecological model can also help to balance individual concerns (e.g., caregiver mental health concerns) and structural concerns (e.g., housing instability, experiences of racism). As outlined in the literature, child welfare scholars are often split across the individual-structural divide (i.e., authors tend to exclusively adhere to one theoretical lens or the other), which offers only a partial assessment of client problems and capabilities (57). For example, in discussing children's experiences of family violence some authors focus exclusively on structural determinants, such as racist legislation or inadequate government funding for programming, without discussing the impact of parental mental health concerns on children whereas other authors focus exclusively on programs to address parenting behaviours without attending to structural factors affecting parents. Both lenses are required for effective case conceptualization for child welfare cases.

While individual providers may not be able to effect immediate change in factors at the societal, community, or institutional level, it is important for providers to have awareness of these factors. At the individual/relationship level, or in direct clinical work with children and families, the bio-psycho-social-spiritual model remains an important model for case conceptualization as it is largely atheoretical (meaning it can be used across diverse professions that are applying different theoretical lenses) (51). It is also broad enough to cover biological concerns (the domain of healthcare providers such as physicians and nurses), psychological concerns (the domain of psychologists and psychotherapists), and social domains (the domain of teachers, recreational professionals, and advocates), as well as the intersections between these domains. The bio-psycho-social-spiritual model can help lessen the tendency for case conceptualizations to be overly biomedical or overly psychological. As such the bio-psycho-social-spiritual model may represent a useful framework for undertaking comprehensive assessments in child welfare. For example, children involved with child welfare tend to receive more pharmacological interventions than their peers (58, 59), even though in many cases it is not recommended or is even contraindicated. Also, as discussed above, many providers feel comfortable making referrals to mental health services but overlook consideration of children's need for positive social supports, such as recreational camps or tutoring.

Socioecological and bio-psycho-social-spiritual factors can be considered across the "5 Ps," or across precipitants, perpetuants, predisposing factors, and protective factors that inform the client's presenting pattern. The 5 Ps are commonly used in clinical formulations to account for the client's pattern of functioning, or when considering (in)flexible and (in)effective ways that the client is thinking, perceiving, and acting. For example:

- Presenting factors attend to the child's attentional and behavioural issues, such as physical aggression toward peers, distractibility, and resistance to following instructions.
- Precipitant factors include triggers that bring about the client's presenting concern. For example, if a child's behavioural problems at school were triggered by peers yelling, this would give us important information about the child's vulnerability to loud noises or would cause us to wonder if the child was exposed to loud noises in their environment (e.g., yelling in the home).
- Perpetuating factors maintain the client's presentation. For example, if a child's teacher lacked skills for understanding how trauma can manifest in concerning behaviours, they may not have a compassionate and understanding response to the child's behaviours, unintentionally helping to maintain the behaviour. School can be an important resource for children with experiences of violence, as children may look to schools as a "safe haven" free from violence, a predictable setting in comparison to their "chaotic" and "unpredictable" homelife, and a place where it is possible to develop safe and supportive relationships with teachers and peers (60).
- Predisposing factors include bio-psycho-social-spiritual factors that contribute to the client's presentation. For example, if a child is aware of past or current IPV in their home or had experienced harsh discipline from their caregivers, this could have significant negative effects on their development, including changes in brain development, a decreased capacity for emotional regulation, and increased threat sensitivity (61, 62).
- Protective factors include bio-psycho-social-spiritual factors that lead to adaptive presentation and functioning (e.g., curiosity, spirituality, high-quality daycare or school environments). For example, a teacher's care and concern for a child could be a protective factor. Other significant resilience factors have been found for children experiencing maltreatment, such as family and peer factors (e.g., maternal sensitivity, close mother–child relationship, friendship, and social support) (8).

Clinical formulations can draw on a variety of theories to understand the client's unique pattern of functioning, such as theories about stages of change, psychodynamic theories about personality development, theories about family functioning, and considerations of attachment theory (36). For example, it is important to consider the client's readiness for treatment, or to consider the stages of change, and to tailor treatment accordingly (63). Consider a caregiver who has been referred to an evidence-based parenting program for substantiated physical abuse of their child. The treatment trajectory and prognosis would be very different for this caregiver depending on if they were in the precontemplation phase of change (they do not consider their behaviour to be a problem and do not feel they need to change) as compared to the preparation phase of change (they have made a commitment to change their behaviour, which they consider problematic, and they may even have identified steps towards change). Some child welfare interventions have begun to acknowledge stages of changes by modifying interventions to explicitly address client readiness and stages of change (64); this represents a generalized strategy that may or may not be needed depending on the specific client.

In addition to considerations of stages of change, it is important to consider hierarchies of power in families, such as gender and intergenerational power, especially when family violence is a concern. Researchers attending to hierarchies of power discuss the need to address safety and protection of children; empowerment and safety of women and 2SLGBTQ+ persons; and responsibility and accountability for those using violence in their relationship (65). When there are concerns about family violence in the home, a provider's attention to hierarchies of power can help them to transparently suggest avenues for action when supports and services are not working to address safety in the home. For example, Humphreys (65) has outlined avenues of action when there is a conflict in needs between children experiencing safety concerns in the home, women experiencing IPV, and men using violence in relationships:

> For example, should there be a dilemma between the principle of child safety and that of the empowerment and safety of women, which even after high level support is unable to be addressed, then the safety of children remains paramount due to their level of vulnerability. Similarly, if there is a conflict of interest or resourcing pressures, the safety and empowerment of women needs to be placed as a priority over potential work with men.

For children involved in child welfare who are experiencing current maltreatment, including exposure to IPV, treatment planning might first involve addressing safety in the home, for example, by working to increase caregivers' supportive and safe behaviours. This could entail involving caregivers in an evidence-based parenting program with motivational interviewing components (66). Parallel work could be done with a caregiver who is exposed to IPV to assess their safety and refer them to evidence-based resources to address their past or current experiences of IPV [e.g., structured advocacy interventions or support for any symptoms resultant from past experiences of violence (52)]. This work may be important to do before a child is referred to evidence-based services for their emotional and behavioural problems, as these concerns may be the direct result of lack of safety in the home. If work to increase safety in the home is not successful, and safety continues to be a concern for the involved children, additional approaches will need to be considered to prioritize child safety, including the potential for high-quality out-of-home care placement, such as kinship or foster care.

4.3 What role does culture play (cultural formulation)?

A cultural formulation answers the question "what role does culture play" by analyzing salient cultural factors, such as level of acculturation and stress (51). Effective cultural formulation is essential in child welfare, especially given the overrepresentation of certain racial, ethnic, and cultural groups, such as Black and Indigenous families (67, 68). Cultural considerations can be considered across the bio-psycho-social-spiritual framework, including:

- Biological concerns (e.g., any particular health concerns common in the family's history; any biological impacts of intergenerational or racial trauma, such as worsening of chronic illness),
- Psychological concerns [e.g., social or cultural identity; cultural explanations, or culturally influenced beliefs about the client's presenting concern (51)],
- Social concerns [e.g., cultural stress; acculturation, or level of adaptation to dominant culture; any culturally-influenced stress or protective factors; a history of intergenerational or racial trauma; cultural expectations of parenting (69)], and
- Spiritual concerns (e.g., spirituality as a protective factor and/or as a source of cultural/racial persecution).

Using a trauma- and violence-informed care lens, cultural awareness can include attention to ways to increase cultural safety in the environment, approach, and provider response (28). For example, in terms of the environment, some Indigenous clients may appreciate access to an Elder in the service organization or through a referral (70). In terms of the approach, it is important for providers to be aware of a client's potential experiences of social or cultural violence, discrimination, stigmatization, or oppression (e.g., feeling misunderstood; misjudged related to social/cultural identity; direct experiences of discrimination, stigma, oppression, exclusion, ostracization, or being devalued; experiences of microaggressions; difficulties assimilating). This might involve a provider undertaking training about historical violence, for example, as summarized in the Truth and Reconciliation Commission of Canada Calls to Action (71). It can be desirable for providers to make efforts to increase working relationships with social and cultural community leaders and organizations, to increase the provider's own awareness of culturally appropriate and available services in the community (72). Such awareness will enable the provider to facilitate and tailor referrals to appropriate services when indicated. In terms of the provider response in the care encounter, it is important for providers to explore and understand the client's cultural explanation, or culturally influenced beliefs about the client's presenting concern (51). Understanding cultural explanations of the client's presentation is an important part of treatment planning (73), as divergent client and provider understanding of the 5 Ps can lead to treatment 'poorness of fit' between provider and client goals, as well as poor treatment prognosis (34).

4.4 How can what happened and its impact be addressed (treatment formulation)?

A treatment formulation answers the question "how can it be changed" by specifying a map for treatment planning. Treatment formulations address the focus, goals, strategies, and interventions of treatment, as well as treatment obstacles and prognosis. Treatment focus addresses the direction of treatment; it is akin to the metaphor of a map, which shows the best route to achieve a desirable treatment outcome (51). The map for a bio-psycho-social-spiritual approach addresses concerning situations that prompted or were exacerbated by biological, psychological, social, or spiritual vulnerabilities. For children involved in child welfare who are experiencing ongoing safety concerns, the treatment focus may involve a continual reorientation to safety in the home and services and supports to address caregivers' biological, psychological, social, and spiritual vulnerabilities. Treatment goals are realistic, measurable, and achievable; a goal can represent the final destination on the map or can involve small stops on the map as the client moves towards the final treatment outcome. An example of a goal in a parenting class would be to observe an increase in parental nurturing and responsive behaviours, such as praising positive child behaviours, reflecting appropriate speech of children, or letting children lead conversations. Even in situations where treatment is mandated, goals should be mutually agreed upon by the provider and client. This may involve engagement to first address how services can meet both provider and client goals. Goals can be short-term (e.g., symptom reduction, increased adaptive functioning) or longer-term (e.g., pattern change) (51).

Treatment strategy refers to "the action plan for focusing specific interventions to achieve a more adaptive pattern" (51); it is akin to selecting the best route and vehicle to achieve the treatment goal. A *treatment strategy* involves the selection of appropriate *treatment interventions*, or actions designed to positively impact the client's issue or problem. For example, where there is safety in the relationship between caregivers (e.g., no current or recent concerns about IPV), parent–child interaction therapy is an evidence-based intervention for children with externalizing problems who have a history of physical abuse or neglect. Usually this involves specific treatment interventions that teach the importance of child-directed interactions, including specific skills for caregivers to do more of (e.g., praise, reflect appropriate emotional response, imitate appropriate play, describe appropriate behaviour, and enthusiasm) (74).

Treatment formulation also considers treatment obstacles and prognosis. One test of an effective case conceptualization is its ability to predict the most likely obstacles and challenges (51), such as difficulties with engagement, ambivalence, and alliance. For example, as discussed above, it is important to assess the client's readiness for change and to choose appropriate engagement strategies based on their level of engagement. It is also important to assess for practical barriers to engagement, such as lack of transportation, and to problem solve with clients. Second, many clients have ambivalence about services and treatments, with part of them moving towards change and another part of them resisting change. Practitioners need strategies to support clients in moving towards change, such as those offered by motivational interviewing (75). Third, while a working alliance is a consistent mediator of change (76, 77), including in contexts where treatment is mandated [for example, by child welfare (78)], difficulties with alliance between providers and clients should be anticipated and problem-solved in child welfare work. Improving alliance between providers and clients may involve provider skills (e.g., their ability to have conversations that repair alliance); alliance is also impacted by structural concerns [e.g., child welfare workers' alliance with clients is better when there are dedicated family coordinators with low caseloads (79)].

Long-term outcomes for a child involved with child welfare can be linked to a number of factors, including child safety (recurrence of maltreatment, serious injuries/deaths), child well-being (school performance, including grade level and graduation; child behaviour; criminal justice involvement), permanence (placement rate, moves in care, time to achieving permanent placement); and family and community support (family moves, parenting capacity, ethno-cultural placement matching) (80). Even when there are intertwined factors that suggest a poor prognosis for a child, providers still have a meaningful opportunity to improve safety in the child's home in order to hopefully prevent future experiences of maltreatment and more serious behavioural concerns.

5 A case example

Below we present a case example of a child who was referred for mental health services by a child welfare worker, which is a common type of service referral made by child welfare workers.

Rose is a 10-year-old girl who was referred by a child welfare worker for treatment of mental health and behavioural concerns. Child welfare was initially contacted by a school principal after a teacher raised concerns about Rose's hygiene, inadequate lunches, and excessive sleepiness at school. The school personnel had tried to address these concerns with the parents, but the parents were not responsive and after raising these concerns Rose was increasingly absent from school. There were significant concerns about Rose's behaviour at school, including physical aggression toward peers, distractibility, and resistance to following instructions; she was also easily startled and became irritable when class discussion was happening. The teacher questioned whether Rose had ADHD. Child welfare had been involved with the family for approximately six months after Rose was increasingly absent from school without explanation. Rose has five other siblings who were also struggling. Child welfare had made referrals for services. Rose's parents were encouraged to have Rose seen by her family physician regarding her attentional and behavioural problems and she was referred for individual counseling to address her behaviours. The counselor working with Rose expressed concerns for Rose's development including difficulty identifying and communicating her feelings and needs, and a tendency to portray herself negatively. For example, when asked why she thought she had come to see the provider, Rose responded "I was bad." After 12 sessions with the counselor, Rose's functioning and behavioural concerns at school had not improved. The counselor was concerned and sought supervision for the case. Individual sessions had focused on helping Rose to identify and communicate her needs and cognitively restructure her maladaptive thoughts and beliefs. Although such intervention strategies can be helpful for many children with similar emotional and behavioural concerns, they were not effective for Rose.

This case example illustrates how case prognosis can be poor for clients when key elements of the case are missing from the case conceptualization, such as sibling and family dynamics. When key elements of the case are considered, the likelihood of a positive treatment outcome is increased. Additional aspects that are important to consider in Rose's case are discussed below.

Here is an example of additional information that was revealed about Rose's living situation during a comprehensive assessment with Rose and her caregivers:

- *Description of people in the family, present living situation, extended kinship networks, any non-traditional familial relationships*: Rose's father, Jake, identifies as Canadian/White with British heritage and her mother, Jalen, identifies as Southeast Asian. Jalen and Jake have 6 children in total between the ages of 3 and 12. The older children, including Rose, identify as Canadian/White. The family lives in a small, 3-bedroom apartment in the city and are isolated from family and community support due to frequent moves. Jake previously worked in construction but lost his job 3 years ago and has been working part-time as a painter since then. Jalen works part-time as a server in a local restaurant.
- *Developmental history*: Rose was born a month prematurely and had some early language delay that subsequently improved after beginning daycare.
- Education: Rose is academically behind her peers in terms of grades and social skills. Her teachers reported that she is quiet and "spaced out" but that she reacts violently towards peers ("goes from 0 to 100") when she experiences loud noises by peers or in the classroom. Her teachers struggle to connect with Rose; they worry that Rose never smiles. They also reported that she misses a lot of school.
- *Involvement with activities*: Rose was not currently involved in any extra-curricular activities. Her parents have indicated that they do not have the resources and also identified they do not have time to take Rose to after-school activities.
- *Emotional/behavioural/psychological functioning*: Rose was identified as having difficulty communicating her feelings and a tendency to portray her role in family interactions negatively ("I was bad"). Concerns were identified that Rose exhibited symptoms of post-traumatic stress including hypervigilance, intrusive thoughts, sleep disturbances, emotional dysregulation, and dissociation ("spacing out"). During an individual interview with Rose, she disclosed fear about her mother's well-being, as well as experiences of emotional abuse and neglect.
- *Information about caregivers and siblings*: The family had been referred to child welfare in the past because of concerns about IPV and Jake's threats to hit his wife and the children. The family was considered a flight risk by child welfare, as they have had a history of moving when child welfare has become more intrusive. Three of Rose's siblings have been referred to a pediatrician or a mental health professional, including psychiatrists, in the past and several have been diagnosed with ODD and ADHD and prescribed psychotropic medications. During the comprehensive assessment, Jake insisted that the child welfare worker had no authority in his family and that he has been doing his best for them. In a separate meeting with Rose's mother, Jalen, she appeared to experience difficulty communicating about family relationships, and was vague in her responses. Jalen reported that Jake sometimes got "out of hand" but that he was doing the best

for the family. Jalen disclosed being slapped across the face by Jake as recently as a year ago when Jake was intoxicated and expressed worry about Jake's increasing reliance on alcohol since his job loss 3 years ago. Jake has previously been involved with psychiatric services, however he refused to provide additional details about the nature of these services (including any diagnoses). During an individual interview, Jake acknowledged feeling anxious and depressed about finances; he also discussed how he used alcohol to cope with stress. Jake also acknowledged previously hitting Jalen several times and expressed regret and a desire to never hit her again.

- *Observing Rose with her caregivers*: Rose, her siblings, and mother/father were observed together. In this meeting, Rose, her siblings, and the mother were all silent and reluctant to speak even when spoken to directly. Jake appeared agitated and directed most of the conversation, glaring at his children and wife and speaking for them when they were called upon by the provider. During this meeting Jake expressed sentiments of male privilege, such as discussing women's role to listen and serve.

These additional details will be integrated into a clinical formulation of the case, as discussed below.

5.1 Clinical example revisited

Below we present a brief clinical formulation and treatment formulation statement for Rose based on the information from the comprehensive assessment.

Based on parent and teacher reports, Rose has a longstanding history of difficulties in functioning at school and within peer relationships. Her challenges with attention in the classroom were attributed by her pediatrician to a diagnosis of ADHD, and her aggressive interactions with peers led to a diagnosis of ODD. Even though Rose's symptoms are reflected in these diagnoses, there are many aspects that are overlooked without a trauma- and violence-informed case conceptualization. Through a comprehensive assessment that provided Rose the opportunity to discuss her family relationships, Rose disclosed a history of longstanding fear about her mother's wellbeing, as well as chronic emotional abuse and neglect from both parents. In individual interviews with each parent, Jalen spoke about being slapped across the face by Jake as recently as a year ago when Jake was intoxicated. She spoke about his problem with alcohol use that had increased following the loss of a full-time job 3 years earlier. In his interview, Jake initially denied any problems in the family, but subsequently spoke about feeling increasingly anxious and depressed about finances and using alcohol to cope. He acknowledged hitting Jalen on several occasions, and wanting to ensure this did not happen again, but had not told anyone including his family physician about his abusive behaviour.

To develop an understanding of Rose's issues, we need to consider her presenting symptoms in the context of her life experiences, including her relationships with caregivers. Rose has been exposed over many years to IPV and has experienced emotional abuse and neglect. Her symptoms of ADHD and ODD, as well as physical problems, including difficulty sleeping, can best be understood by considering the principles of trauma- and violence-informed care. Rose's presenting symptoms can be understood through the lens of complex trauma, which refers to experiences of multiple, traumatic events (e.g., exposure to maltreatment, loss of family relationships; inconsistent parenting etc.), specifically in the context of the child's primary caregiving relationship(s). A child's emotional well-being depends largely upon having a relationship with a caregiver who serves as a source of safety, security, and support. When a caregiver is perceived to be inconsistent, absent, or frightening during the early years of a child's life, the child's ability to tolerate and manage strong emotions, deal with daily stressors, develop self-confidence, and learn the foundations of relationships is compromised. When the parent is the source of the threat or maltreatment, then children learn to mistrust others and the world. As such, their capacity to develop emotional regulation skills is compromised, as the caregiver is not available to assist the child in regulating their emotions.

Children who experience maltreatment, including IPV exposure, can feel that they have to be on guard all the time; they often manifest this hypervigilance through distraction at school, impulsivity and aggression. Given Rose's family life and experiences, her symptoms can be understood as adaptive responses to a maladaptive home environment. As such, a key priority in treatment planning involves preventing ongoing exposure to these experiences.

The foundation for Rose's recovery is first and foremost a stable, nurturing parenting environment that is responsive to her needs and can provide her with opportunities for growth and development in a physically and psychologically safe context. While Rose requires ongoing support and assistance, it is essential that the trauma to which she has been exposed is prevented from recurring.

To address Rose's needs, it is essential that the family needs are also considered and addressed. Specifically, it will be important to engage the parents in services to address their own respective experiences of trauma and mental health issues. Additionally, it will be important to work with the parents to increase their capacity to interact with Rose and her siblings in ways that are safe and supportive and avoid threatening or harmful behaviours. To do so, the parents need to acknowledge their role and responsibility in the harm suffered by their children, and there needs to be ongoing assessment of the parenting that Rose and her siblings are receiving. Connecting the parents to an in-home evidenced-based trauma and attachment-informed parenting program would be beneficial. This can only occur however, if Jake refrains from any further violence toward Jalen, and engages with substance use treatment and ongoing involvement with child welfare to assess the risk of violence in the home. Given the history of IPV, it is important for Jalen to have access to ongoing assessment of her safety and resources and support to address her past experiences of IPV. Given Jake's patriarchal assumptions about men's and women's roles, it would be important to assess if these sentiments mirror his or Jalen's understanding of gender and how this does or does not relate to their cultural heritage. It would also be important to assess if Jalen experiences cultural barriers to seeking help.

If safety is achieved in the family, work to support Rose's individual and specific needs (e.g., traumatic stress symptom reduction, trauma processing, developing skills to manage difficult feelings, etc.) and experiences (including at school) may be indicated, including support to increase her tolerance of loud noises and work to support her success in classwork (e.g., tutoring).

If work to increase safety in the home is not successful, and safety continues to be a concern for Rose and her siblings, additional services need to be considered to prioritize child safety, including the potential for high-quality out-of-home care placement, such as kinship or foster care.

Children and family benefit from social and community supports. Rose's family's low socioeconomic status and social identity likely also influence their engagement and access to support and services. Given the family's relative social isolation, consideration should be given to increasing the family's opportunities to connect to additional supports and activities in the community which can be protective and increase safety for all members of the family. For instance, children's successful participation in social/recreational activities can be beneficial and therapeutic as children are provided opportunities to develop their skills and talents, experience membership, a positive self-concept and self-esteem. Additionally, having access to healthcare providers, ideally in a community-based healthcare team that is trauma- and violence-informed, would be supportive not only to Rose, but also to her siblings and her parents. School can also be an important resource for children with experiences of violence. Rose's teachers have identified their concerns for Rose. The school's interest and care for Rose can be protective. It will be important for the school to have some understanding of Rose's experience of trauma and how to support her within the context of trauma- and violence-informed care. Additionally, it is important for the parents' relationship with the school to be facilitated and supported. This will likely require intervention (advocacy, psycho-education) from the family's primary service provider.

Finally, access to services and supports for the family must be considered in the context of larger structural issues and barriers. For instance, this family has experienced inadequate and inconsistent housing and struggles financially. It will be essential to work with the family to ensure they have access to financial resources they are entitled to and to support and advocate for access to adequate and safe housing and transit/transportation. To be effective, service providers will need to develop collaborative working relationships with the parents. If these structural barriers are not addressed, the likelihood that individual interventions outlined above will be effective are significantly reduced.

5.2 Case commentary

In this example, the case conceptualization guides treatment planning and implementation, in order to prioritize creating safety in the family environment before addressing Rose's mental health symptoms. The prioritization of safety is an essential component of treatment for clients with experiences of complex trauma (81). The case prognosis could be considered poor given the following factors: the family has been involved repeatedly with child welfare with the most recent opening occurring 6 months ago; Rose has poor school performance and struggles with post-traumatic stress; the family has a tendency to move; and the father does not appear ready to examine his abusive interactions and potential IPV in the home. However, progress is more likely to occur when experiences of family violence are understood and prioritized in treatment planning. Understanding principles of trauma- and violence-informed care can also help to put Rose's physical, emotional, and social symptoms into context: they make sense considering her chronic experiences of maltreatment. Case conceptualization, including a bio-psycho-social-spiritual assessment, offers a strategy and rationale for sequencing interventions. By outlining the case conceptualization and treatment plan we make explicit our assumptions as providers, which we can evaluate in the future, based on treatment progress or barriers. For example, if the family's housing issues are addressed, Jake's substance use problems are managed appropriately, safety is achieved in the home (i.e., absence of IPV or child maltreatment), and Rose's symptoms still persist, then we can begin to formulate additional factors that may be influencing her presentation.

6 Conclusion

In this paper we have shown how principles of trauma- and violence-informed care, comprehensive assessment, and case conceptualization can guide treatment planning and implementation, in order to best assess, address, and prioritize biological, psychological, social, and spiritual elements of the child and family. Case conceptualization in child welfare is warranted because of the inherent complexity of presenting cases. Further work in this area could evaluate ideal theoretical frameworks for developing effective and clinically useful case conceptualizations, as well the potential benefits of interdisciplinary case conceptualization.

Ethics statement

Written informed consent was not obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article because while this paper is written like a case report, it is a hypothetical case based on common cases presented in the literature.

Author contributions

JRM: Conceptualization, Writing – original draft, Writing – review & editing. AM: Conceptualization, Writing – review & editing. AN: Conceptualization, Writing – review & editing. AP: Conceptualization, Writing – review & editing. CM: Conceptualization, Writing – review & editing. HLM: Conceptualization, Resources, Writing – review & editing.

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Conflict of interest

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

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Trauma and psychosocial adversity in youth with autism spectrum disorder and intellectual disability

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Traumatic experiences contribute significantly to behavioral and mood dysregulation syndromes presenting for treatment to behavioral health settings. Individuals with Autism Spectrum Disorder (ASD), Intellectual Disability (ID) and developmental delay experience traumatic events more frequently than their typically developing peers. However, measures used to identify trauma related disorders and treatment thereof are based on typically developing individuals. Regardless of the baseline characteristics of individuals who experience trauma, trauma exposure is the result of multiple interdependent environmental, social, and familial characteristics. We used the "ecological systems analysis approach" to structure our review of the impact of trauma on those with ASD and ID. In addition, the COVID-19 pandemic which exposed the global population to a collective trauma, has also catalyzed investigations into the challenges faced by members of society most dependent on social services. Children with ASD and ID were among those vulnerable individuals, and the COVID-19 pandemic has allowed researchers to better understand the impact of a collective trauma on those individuals. It is imperative that we understand current research and recommendations for identifying and treating trauma-related disorders in individuals with developmental disorders to best inform clinical practice and directions for future research in this area.

KEYWORDS
autism spectrum disorder, intellectual disability, trauma, post-traumatic stress disorder, child and adolescent psychiatry

Introduction

Developmental disability impacts individuals in multiple spheres of social, educational, vocational, and personal functioning. Presentations of developmental disabilities are diverse, and rates in the United States have consistently increased in recent years (1). Intellectual disabilities (ID) are defined by deficits in intellectual functions and adaptive functioning with onset during childhood (2) While there are multiple clinical presentations of ID, the DSM-V categorizes ID into four severity levels (mild, moderate, severe, and profound) and three domains (conceptual, social, and practical) (2).

Autism spectrum disorder (ASD) is a type of developmental disability that is of particular interest due to its increasing prevalence and significant overlap with ID (3). The prevalence of ID among individuals with ASD is reported to be 22.9 (95%CI [17.7-29.2]) (4). ASD is characterized by persistent deficits in social communication and social interaction across multiple contexts resulting in 1. Deficits in social-emotional reciprocity, 2. Deficits in nonverbal communicative behaviors used for social interaction and 3. Deficits in developing, maintaining, and understanding relationships (2). Criteria for ASD also includes restricted repetitive patterns of behavior, interests, or activities (Ibid).

Trauma-related disorders including Post Traumatic Stress Disorder (PTSD) require exposure to a trauma to meet criteria for diagnosis (2). Per the DSM-V, for individuals older than six years of age, trauma is defined as exposure to actual or threatened death, serious injury, or sexual violence. For individuals younger than six years of age, trauma can include the knowledge that actual or threatened death, serious injury or sexual violence occurred to a caregiver (2). Outside of the definitions laid out by the DSM-V, it has been shown clinically that chronic neglect of basic needs, especially during early developmental periods, can lead to a PTSD presentation (5). Broadly, child maltreatment encompasses multiple categories of abuse and neglect, including physical assault, psychological aggression, and neglect (6), or family interpersonal violence (IPV), nonfamily IPV, non-IPV trauma, separation/loss, acts of commission, acts of omission, contact trauma and noncontact trauma (5). Providers have posited concerns that events that may seem trivial to others can be traumatic for individuals with ASD and ID (7–9). Given the broad definitions of trauma and maltreatment and the impact of multifactorial individual vulnerability on the risk of developing a trauma associated disorder, this review will discuss the impact of traumatic experiences on those with ASD and ID more broadly.

Exposure to trauma and maltreatment is higher among children with intellectual disability (10) (11). A 2019 population-based sample using a linked cohort between the Department of Social Services and the Autism and Developmental Disabilities Monitoring (ADDM) network found that a diagnosis of ASD alone increased the risk of maltreatment with an OR of 1.86 [1.36, 2.52]; a diagnosis of ASD and ID increased the risk further with an OR of 2.35 [1.77,3.12]; and a diagnosis of ID alone increased the risk of maltreatment the most with an OR of 2.45 [2.09, 2.88] (12). Authors considered the possibility that this discrepancy highlights the concerns that deficits in social communication in children with ASD, and/or possible biases among caseworkers may present additional challenges to detecting maltreatment in this vulnerable population (12, 13). Elsewhere, the possibility that ASD-associated deficits in social cognition including naivete, poor social boundaries, and difficulty detecting a violation of social rules or inappropriate behavior may also contribute to higher risks of victimization while also increasing their vulnerability to interpersonal manipulation which may also be a barrier to reporting maltreatment in this population (14). Despite the prevalence of maltreatment in this population, the co-occurrence of PTSD with ASD and ID has not been well described in the literature (4, 8, 14, 15).

In their 2011 paper authors Algood et al. presented a review on the maltreatment of children with developmental disabilities using an ecological systems analysis. Ecological systems analysis allows maltreatment to be understood as an outcome of a complex and interactive set of interdependent systems. Algood's review is organized by systems the following way: Social-demographic characteristics, Age, Gender, Special Education, Microsystem, Parent-Child Relationship, Domestic violence, Ecosystem, Parenting Stress, Parents' Social Support, Area of Residence, and Macrosystem. Seeing as children with ASD and ID account for a significant number of those receiving community services and educational supports and constitute a large portion of those most vulnerable to maltreatment, authors concluded that contemporaneous policies impacting systems caring for children with disabilities must also consider their impact on the ecosystem which predisposes this vulnerable population to abuse (16, 17).

The Centers for Medicaid and Medicare Home and Community Based (HCBS) Settings Rule was passed in 2011, stipulating that community-based long-term services and support be provided to individuals meeting criteria for developmental disabilities, and that states must follow this rule by March 17, 2023 (Autismsociety.org, accessed 8/11/2023). Between 2011 and 2023, investigations into the trends of adversity faced by individuals with ASD and ID progressed. In addition, the COVID-19 pandemic which presented a global challenge transecting the ecological domains has given researchers the opportunity to study the interplay of those domains. Therefore, we present an updated ecological analysis of maltreatment faced by children with developmental disabilities, and specifically ASD, to help guide clinical practice and future policy decisions.

Socio-demographic characteristics

In a 2023 secondary analysis of characteristics of children with ASD using existing medical records in the Autism and Developmental Disabilities Monitoring Network (ADDM Network) collected in 2020, the Centers for Disease Control and Prevention reported that overall, ASD prevalence per 1,000 children aged eight years in the study population was 27.6 or one in 36, with overall ASD prevalence of 43.0 among boys and 11.4 among girls. The prevalence of ID alone was reported to be 11.8 per 1,000 and 37.9% of children with ASD aged eight years and above were classified as having intellectual disability as noted in their developmental evaluation by a qualified professional, or an intellectual quotient (IQ) score ≤ 70 (3).

This data also indicated that prevalence of ASD in children aged eight years in the ADDM Network differed among racial and ethnic groups. Prevalence of ASD among white children (24.3) was lower than prevalence among Black, Hispanic, or Asian/Pacific Islander children (29.3, 31.6, and 33.4, respectively) (3). Additionally, girls with ASD were more likely to be classified as having ID compared with boys with ASD (42.1% versus 36.9%), and Black children were more likely than Hispanic and white children to be classified as having intellectual disability (50.8%, 34.9%, and 31.8%, respectively) (3).

Socio-economic status also has an impact on the ability of families to manage stress. During the COVID-19 pandemic it was found that pre-pandemic poverty was significantly directly linked to caregivers' emotional distress, and employment decrease was significantly directly related to household children's behavioral problems (18).

Special education

Special education is a critical component of treatment and offers important support to families with children with ASD. Therefore, it is not surprising that withdrawal of these supports during the COVID-19 Pandemic resulted in increased difficulties for children and their families. Latzer et al. surveyed 31 families in Israel with children with ASD who lost access to a specialized education system offering full day classes six days per week during COVID-19 lockdown. While the survey used did not specifically ask about maltreatment or abuse, all parents surveyed indicated that they did not have the knowledge or means to provide for their children's developmental needs without the professional support offered by school. The loss of expertise, therapies, physical space, and changes to routine contributed to increased repetitive behaviors and developmental regression, overall increasing the difficulty experienced by families (19).

The strain on families caring for children with ASD alone when services had previously been provided by a team of professionals could have significant consequences for the child's experience of maltreatment. In April 2020, investigators in Hong Kong surveyed 417 children with special education needs (SEN) and 25,427 typically developing (TD) children studying at mainstream schools (6). Among the children with SEN, 19.18% had physical disabilities, 20.38% had ID, 45.8% had mental disorders (e.g. ASD, attention deficit hyperactivity disorder), 24.22% had other disabilities including global developmental delay, isolated significant delay in motor/language skills, or syndromal/genetic disorder, 7.91% had visual impairments and 5.76% had hearing impairments (6). During COVID-19 school closures, investigators found that children with special education needs had significantly more emotional and behavioral difficulties across all aspects than typically developing peers ($p<0.01$) and experienced poorer overall quality of life (68.05 vs. 80.65, $p< 0.01$) (6).

While rates of child maltreatment in typically developing children were not reported by authors, they found that 23.5% of children with SEN had at least one episode of severe physical assault and 1.9% experienced very severe physical assault, while 80.5% were victims of psychological aggression and 28.7% suffered from neglect during the pandemic (6). Compared to maltreatment prior to the pandemic, relative risk of physical assault among SEN children was 1.19 (χ^2 = 9.938, p=0.01) and psychological aggression was 1.50 (χ^2 = 54.604, p=0.01) (6). While it has been reported that risk of child maltreatment (CM) increased for all children during the pandemic, the epidemiological measurement of CM rates has been complicated by a decrease in CM allegations due to school closures during the pandemic and loss of contact with mandated reporters in the education system, and there are no studies on this topic that exclude children with ASD or ID (20). The ability to compare rates of CM among TD children and children with ID and ASD is therefore limited. However, the risk of maltreatment among children with SEN increased significantly during the pandemic (6).

****Microsystem** "The relations between the developing person and environment in a direct setting where the person is embedded" – (21).

Parental stress and parent-child relationship

Studies of parent-child relationships are limited as they focus on heterosexual couples and require parents and children to live together in order to be included, which excludes families where parents live separately or who are not in heterosexual relationships (22, 23). Despite this, understanding the current literature is useful in understanding family dynamics as a significant factor in exposure to domestic violence.

A 2020 study set out to examine the relationship between parenting stress and the emotional quality of the parent-child relationship using the Five-Minute Speech Sample (FMSS) in 150 families of children with ASD aged 5-12 years (23). Parenting stress was measured using the Burden Interview and results indicated that parenting stress and depressive symptoms in mothers were negatively associated with FMSS Warmth and positively associated with FMSS Criticism toward the child with ASD. In fathers, FMSS Warmth toward the child with ASD was negatively associated with mother's level of parenting stress (Ibid). While fathers' FMSS Warmth was correlated to mother's level of parenting stress, mothers' FMSS Warmth was not mediated by fathers' parenting stress. In this study, authors also reported that 20-56% of parents of children with ASD report a clinically significant level of depressive symptoms relative to 7-29% of parents with children with other types of disabilities, and 8-19% of parents of typically developing children (23). A 2014 investigation of parent-child relationship quality and parental depression in heterosexual couples which did not control for the presence of ASD or ID, showed that while mothers' depressive symptoms were associated with lower father-child relationship quality, father's daily depressive symptoms were associated with higher mother-child relationship quality (22). It is notable that father-child relationship stress was consistently shown to be mediated by mother's level of stress or depressive symptoms regardless of the presence of ASD in the child, yet father's stress does not impact the mother-child relationship the same way (22, 23).

While there may be common trends in the dynamics between families of children with and without ASD, it is well established in the literature that parents of children with ASD report higher levels of parenting stress when compared with parents of typically developing children (23). In their 2021 study, Hickey et al. demonstrated that mother and father level of parenting stress is positively correlated to both parents' ratings of ASD symptoms and behavior problems and father level of parenting stress is significantly higher if the child with ASD is male and if the father is white, non-Hispanic (23).

Of note, while ID co-occurred in 34.4% of the 150 participating families in the 2021 study by Hickey et. al., no additional analysis was provided comparing parental stress in this subgroup. It has been previously shown that families of children with autism report significant parental stress at a rate of at least 45%, more than double the rate reported by families of children with other developmental disabilities. (24).

In the context of the COVID-19 pandemic, parental stress continued to be reported as higher among parents of children with SEN. In the 2020 Hong Kong study, parents of children with SEN reported significantly higher parental stress compared to parents of TD children (46.41 vs. 43.36, $p< 0.01$) (6). This trend was shown again in a 2022 study that compared parental stress before COVID-19 and during among families with young children, utilizing reference samples for comparison using data collected by the Berlin-based market research company INFO Marktforschungsinstitut (25). While parental stress was generally significantly higher than pre-COVID-19 levels (M=36.93, SD=10.45, range 18-71 vs. pre-COVID-19 M=34.72, SD = 10.63, range = 18 = 70; $t(1023) = 12.474$, $p<0.001$) with a small effect size (d=0.21), it was significantly lower than parental stress in a clinical reference sample of parents in treatment for their child's behavior problems (a population included in children with SEN) where parental stress was measured n=51, M=43.2, SD=9.1 (6, 25).

****Exosystem** "interactions between two or more settings, of which one is the immediate setting"- (21).

During the COVID-19 pandemic, parents took on the role of full-time educators, caregivers, while managing jobs, financial stress, and their own health challenges (19). Studies investigating trends in parental stress and occurrence of adverse childhood experiences (ACE) showed that of the 6.5% of families surveyed who reported a lifetime occurrence of ACE, 34.8% reported an increase in occurrence during the pandemic (17.6% no change, 47.5% decrease) with the highest lifetime occurrence for children witnessing domestic violence (n=332, 32.4%) and for verbal emotional abuse against children (n=332, 32.4%) (25). Parents who reported an increase in ACE also reported higher pandemic-related stress, poorer parental outcomes, with the largest effect sizes observed for parental stress (25). Further investigation of parental stress and trends in domestic violence show that inability to meet financial obligations and loss of social supports during the COVID-19 pandemic are most associated with increase report of family stress and domestic violence (26).

Domestic violence

As described in the introduction, children with ASD and ID experience significantly higher rates of reported and substantiated maltreatment, including exposure to domestic violence, when compared to their typically developing peers (12).

There is evidence to suggest that the COVID-19 pandemic increased occurrences of domestic violence in families of typically developing children and those raising children with ASD (26). Lockdowns, school closures, and social distancing presented unique challenges to families during the COVID-19 pandemic. Utilizing the Canadian Perspective Survey Series, Beland et al. surveyed 4,627 individuals to better understand the mechanism contributing to domestic violence trends during the COVID-19 pandemic. Authors showed that a family's level of concern about their ability to meet financial obligations and essential needs and need to maintain social ties were most positively associated with concerns regarding domestic violence and family stress from confinement (26). In a study conducted in Italy, 25% of parents surveyed reported that one parent had to quit their job (26.1% of mothers, 27.5% of fathers) to take care of their child with ASD and 94% of study participants reported that the COVID-19 pandemic was financially difficult for them (27).

During the COVID-19 pandemic, caregivers of children with special healthcare needs exhibited more emotional distress and reported higher levels of household children's behavioral problems than caregivers of children without special healthcare needs (18). Tso and colleagues found that increased parental stress during the COVID-19 pandemic was associated with increased likelihood of child maltreatment in children with SEN including psychical assault (r=0.237, p<0.05), severe physical assault (r=0.195, p<0.05), psychological aggression (r=0.363, p<0.01), and neglect (r=0.293, p<0.01). Authors reported that when comparing maltreatment rates in SEN children prior to the COVID-19 pandemic, they found significant increases in the rates of physical assault (59.8% vs. 71.2%, p<0.01) and psychological aggression (53.7% vs 80.5%, p<0.01) during the COVID-19 pandemic (6).

Parents' social support

Parental wellbeing informs the risk of childhood maltreatment and parents of children with ID or ASD rely on community resources, school, medical professionals, and therapies in order to maintain routine, improve functioning in the community, and allow parents greater satisfaction in their relationships with their children. Access to these services can be limited by availability, affordability, transportation, among other factors which were all exacerbated by the COVID-19 pandemic limiting availability of in-person supports to families of children with SEN.

In many cases, families lost their entire support systems during the COVID-19 pandemic (19, 28). A global review consistently showed that loss of services and support systems increased parental stress for parents of children with ASD, and that accessing alternative support systems through relatives, or through emergency health services, allowed parents to reduce their psychological distress (28). In contrast, a 2021 national survey of US families, found that while emotional social support was correlated to reduced caregiver emotional distress and decreased behavioral problems for children, this effect was not observed in households with children who have special healthcare needs (18).

Identifying post-traumatic stress disorder in children with ASD

Exposure to trauma is not commensurate with a trauma reactive disorder, such as PTSD. In the general population, PTSD has a prevalence of approximately 6-8%, and in groups with high exposure to severe psychological trauma, such as combat veterans, refugees, and victims of assault, its prevalence can reach 25% (29). PTSD is also not the only psychiatric sequela of trauma, as individuals may go on to develop other disorders including reactive attachment disorder, disinhibited social engagement disorder, acute stress disorder as well as dissociative and adjustment disorders following a traumatic experience (2). Clinically observed sequelae of trauma in children has also been understood to present as developmental trauma disorder (DTD), a diagnosis not included in the DSM-V but understood to integrate developmental psychopathology, attachment and relational capacity, emotion, and intellectual functioning of the child in the setting of multiple trauma exposures during the early developmental period (5). For the purposes of this review and treatment recommendations, we will focus on PTSD identification and treatment.

The diagnosis of PTSD requires that an individual meet diagnostic criterion and depends on the evaluator's ability to elicit a history supporting an appropriate diagnosis. The DSM-5 criteria for PTSD include exposure to actual or threatened death, serious injury, or sexual violence; at least one intrusion symptom associated with the traumatic event, persistent avoidance of stimuli associated with the traumatic event, negative alterations in cognitions and mood associated with the traumatic event, and marked alterations in arousal and reactivity associated with the traumatic event for a minimum of one month (2). In children six years of age or younger, exposure can include learning that a traumatic event occurred to a parent or caregiving figure, and negative alterations in cognitions include constriction of play and reduction in the expression of positive emotions (2).

ASD is characterized by persistent deficits in social communication and social interaction across multiple contexts (2). Given these deficits in social communication, individuals with ASD may be challenged to describe the symptoms and history required to be diagnosed with PTSD (13). The diagnosis of PTSD in children with ASD, therefore, requires careful clinical observation and understanding of how the symptoms described in the DSM-5 criteria will present in these individuals. Rating scales and structured diagnostic interviews may aid in the proper diagnosis of PTSD. For example, the Child PTSD Symptom Scale for DSM-5 (CPSS-5) is a 27 item scale intended to reveal the presence of PTSD symptoms in children ages 8-18 based on self-report by the child or a caregiver and is based on the DSM-5 criteria (30). This scale is limited by the ability of the child or caregiver to accurately complete the scale and depends on their ability to accurately identify and report symptoms, which requires emotional awareness and verbal abilities that are often deficient in children with ASD (13). In practice, when assessing an individual with ASD who has experienced trauma, it is imperative to speak clearly and slowly with a gentle tone, and to frame questions in a way that they are accessible to the patient so as not to increase anxiety. Part of this effort may be to utilize all options for assistance with communication including American Sign Language, visual cues, professional liaison, or tablet (14).

Not only do the deficits in social communication experienced by individuals with ASD increase the difficulty of reporting symptoms of PTSD, some symptoms of ASD can also appear similar to symptoms of PTSD (Figure 1) (9). While individuals with PTSD can experience intrusions such as flashbacks and dissociation, individuals with ASD can experience self-dialoguing and scripting which can present similarly to intrusions (31). Individuals with PTSD will experience negative mood and cognition. Meanwhile, individuals with ASD will demonstrate reduced reciprocity, stereotyped language and reduced spontaneous play (8, 13). Individuals with PTSD may also exhibit alterations in arousal and reactivity. Individuals with ASD can present as irritable or with aggressive mood, self-injury, and sleep disturbance independent from the presence or absence of PTSD (31).

In addition, co-occurring ASD and PTSD may present with overlapping symptoms (9). For example, individuals with ASD and PTSD may experience recurrent and intrusive recollections which manifest in repetitive play (8). They may demonstrate avoidance or emotional numbing with efforts to avoid trauma reminders, decreased interest in participating in previously enjoyed activities, and restricted affect (8). Hyperarousal may present simply with sleep disturbance, angry outbursts, difficulty concentrating, hypervigilance, or increased startle reaction (8). It has also been observed that young children with ASD and PTSD may manifest: new aggression, oppositional behavior, regression in developmental skills (toileting, speech), new separation anxiety, new fear not obviously related to traumatic event (dark, going to bathroom alone).Diagnostic challenges to identifying PTSD in children with ASD are further complicated by the high rate of psychiatric co-occurrence in individuals with ASD (4). Mutluer et. al., reviewed research on the prevalence of psychiatric co-occurrence in children and adolescents with ASD and their results are summarized in Table 1. Most reviewed studies focused on diagnoses in early childhood and latency age with a significant gap identified in prevalence studies focused on adolescents. Most significantly, ADHD was found to have the highest rates of co-occurrence in individuals with ASD at 26.2% (4), which can further complicate the presentation of alterations in arousal and reactivity in these individuals.

Given the high level of diagnostic complexity in individuals with ASD and co-occurring psychiatric disorders (4), it is recommended that clinicians caring for these children work in collaboration with parents and a range of providers including primary care clinicians, speech pathologist, occupational therapists, and teachers or school counselors. Obtaining collateral information from those providers allows clinicians to incorporate multiple perspectives of the child's functioning and offers opportunities to provide psychoeducation on responses to trauma. In addition, trauma-focused cognitive behavioral therapy (TF-CBT) is one evidence-based trauma-specific intervention that can be adapted to treat individuals with ASD (14). Eye Movement

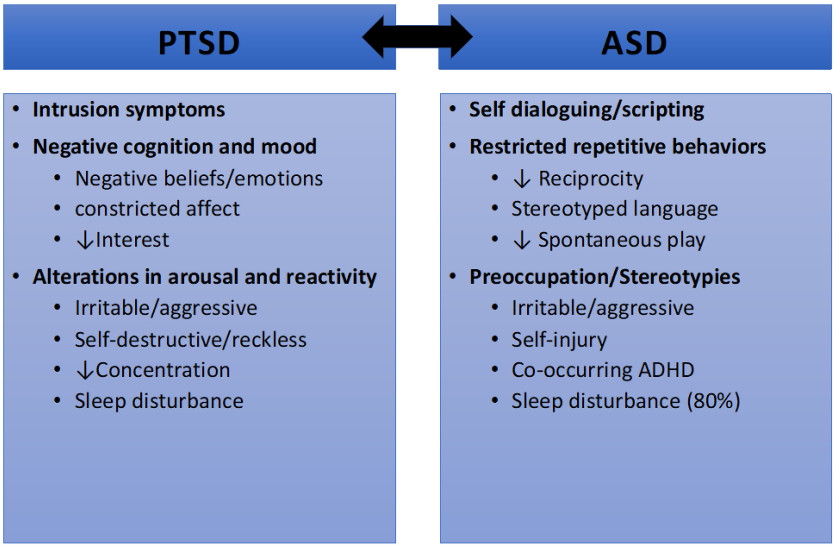

FIGURE 1
Symptomatic overlap between PTSD and ASD (8).

Desensitization and Reprocessing (EMDR) therapy is another preferred method for treating PTSD in the general population. One study examining the use of EMDR in adults with ASD and a history of trauma did show significant improvement in PTSD symptoms when compared to treatment as usual (Impact of Event Scale-Revised: d=1.16), however study limitations included small sample size (n=27), lack of control group, inability to blind participants, and researcher bias, as the therapists providing the intervention also completed the measures (32). The use of EMDR in children and adolescents with PTSD has been shown to reduce scores on the PTSD symptom scale from 60 ± 8.7 to 24 ± 10.1, p= 0.001) in this age group, though the study was limited by small sample size (n=30), absence of a control group and lack of follow-up measurements beyond six weeks (33). Despite the limitations of these studies, it is possible the EMDR would be beneficial in children and adolescents with co-occurring ASD and PTSD, though further research would be necessary (32, 33).

Disaster response and intervention

The COVID-19 pandemic impacted community resources, access to programs, and interrupted supports for families, presenting a natural disaster that threatened the physical safety of the global population. Vulnerable populations such as people with disabilities, developmental, behavioral, and mental health disorders were at higher risk for poor physical and mental health outcomes that resulted from the COVID-19 pandemic and public health measures to address it (www.cdc.gov/disasters/covid-19, 9/1/2023) (27).

The impact of disasters on children with autism is not well-studied. Valenti et al. published a 2011 study that examined the adaptive behavior of participants with ASD one year after their exposure to the 2009 earthquake in L'Aquila, Italy compared with an unexposed peer group with ASD. The researchers showed that adaptive behavior in the exposed individuals declined during the first months after the earthquake (p<0.01). The COVID-19 pandemic was unique from other natural disasters because of the wide exposure to its impact. An ongoing Yehuda Science Foundation COVID-19 study suggested that mental health consequences are primarily found among adults with the most direct exposures to the impact of the COVID-19 pandemic. There are no current parallel studies focused on mental health outcomes for children (34).

Studies of children exposed to prolonged war note that no single exposure alone can determine whether a child will later develop PTSD. However, exposures that lead to multiple other exposures (i.e.: siege exposure) can place the child at risk for all trauma-related outcomes (35). As shown in the ecological analysis above, children and adolescents with ASD and/or ID are at an increased risk of trauma exposure, financial strain, loss of community supports, all of which make them more vulnerable to trauma that can lead to PTSD. There are ongoing efforts to study the global predictors of mental health outcomes for children during the pandemic, however an analysis of these risk factors is not currently available (36). Even so, it is clear that this vulnerable population of children requires special attention so that they receive accurate mental health diagnosis and appropriate treatment (35).

In the case of a natural disaster, immediate intensive post-disaster intervention has been shown to allow children and adolescents with ASD to trend toward recovery of adaptive functioning (37). With this in mind, children with ASD and ID would benefit from collaborative care where changes in behavior are discussed and shared with providers. Evidence-based treatment for children with ASD and PTSD include trauma-focused cognitive behavior therapy that is adjusted to the developmental and skill level of the patient (14). This

TABLE 1 Psychiatric co-morbidity with ASD prevalence and clinical presentation (4).

Psychiatric Co-Morbidity	Prevalence of Co-Morbidity in Children with ASD	Clinical Presentation of Co-occurrence with ASD
Intellectual Disability	22.9%	Defined in terms of measurement tools, WISC-4 and FSIQ, however did not incorporate overall adaptive functioning and IQ scores alone cannot point to the severity level of ID. Authors noted that there is "poor testability" of subjects with comorbid ASD, which led to significant variance in results. Adaptive functioning, meanwhile, was not reported.
Attention-Deficit Hyperactivity Disorder	26.2%	Inattention, hyperactivity-impulsivity, impairments in activities of daily living, social adaptation, behavior problems
Internalizing Disorders Anxiety Disorder Depression	11.1% 2.7%	Social communication problems, sensory aversions, disruptive emotional dysregulation, inflexible adherence to routines, difficulty tolerating change.
Sleep disorder	19.7%	Heightened daytime cognitive, adaptive, and behavioral problems.
Disruptive Disorder	7%	Oppositional defiant disorder, conduct disorder, and disruptive behavior problems.
Bipolar Disorder	2%	Prevalence increased as older age groups were included in the analysis.
Obsessive-Compulsive Disorder	1.8%	Restrictive repetitive behaviors associated with ASD tend to be ego-syntonic compared to ego-dystonic nature of OCD symptoms.
Psychosis	0.6% (1.1% among adolescents)	Behavioral phenotypes of known genetic conditions such as 22q11 deletion syndrome possibly connected to greater likelihood for the identification of psychosis.

intervention is best received with collaboration among all providers for the patient as well as family, school, and primary care physicians.

Conclusion

While children with ASD and ID have long been understood to experience higher rates of trauma and maltreatment, the detection of PTSD among other sequelae of trauma exposure has remained diagnostically challenging in this population. The recent COVID-19 pandemic presented a challenge to all vulnerable populations and provides the opportunity to better understand stress response as well as the importance of community supports for families of children with ASD. Given the significant stressor heralded by the COVID-19 pandemic, loss of supports, family stress, and increased risk of domestic violence during this period, special attention to mental health sequelae will be essential in identifying PTSD in the clinical setting and providing appropriate treatment. Further investigation to better understand risk factors, prevalence, and treatment of trauma reactive disorders in this population will be essential.

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Harmonising outcome measurement for child focused domestic abuse interventions. Reflections on the development and implementation of a core outcome set

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There is appetite in the UK to better measure the impact of domestic violence and abuse (DVA) interventions on children. The spread of outcomes-based commissioning means outcome measurement is no longer just the territory of academic researchers but is now firmly within the purview of practitioners and policy makers. However, outcomes measured in trials only partially represent the views of those delivering and using services with respect to how success should be defined and captured. Even within trials there is huge inconsistency in the definition and measurement of important endpoints. This yields a body of evidence that is difficult to make sense of, defeating the ends for which it was produced – to improve the response to children and families who have experienced abuse. Development of Core Outcome Sets (COS) is seen as a solution to this problem, by establishing consensus across key stakeholder groups regarding a minimum standard for outcome measurement in trials, and increasingly in service delivery contexts. To date COS development has addressed outcomes relating to health conditions or interventions, with limited application to public health challenges. We reflect on our efforts to develop a COS to evaluate psychosocial interventions for children and families experiencing DVA. We highlight the value of COS development as a mechanism for improving evidence quality and the response to families experiencing abuse. Finally, we make recommendations to researchers and COS guideline developers to support this broader application of COS methodology.

KEYWORDS

domestic abuse, child maltreatment, core outcome sets, interventions, family health

1 Introduction

Domestic violence and abuse (DVA) is threatening behaviour, violence or abuse between adults aged 16 years and over who are relatives, partners or ex-partners (1). It is a breach of human rights as well as a major public health problem (2). It can occur in any relationship regardless of gender or sexual orientation, although women, transgender and gender non-binary persons are at increased risk of experiencing IPV. It is widely acknowledged that children's exposure to DVA is widespread and can lead to serious and long-term negative consequences, stretching across all domains of health and development (3–6). This has resulted in government policies to ensure that health and social care services respond to and safeguard children (and their families) who might be at high risk of or have experienced DVA (7–10) However, there is scant high-quality evidence about which interventions are effective and for whom, in which circumstances (11–13).

The current evidence base is limited partly because of the range of outcomes and measures used in DVA evaluations (11, 13, 14). This makes comparing the evidence between and across interventions more difficult. This issue also impacts practice-based research, where funders have been able to draw limited conclusions about the value of multi-million programmes of work (15). Consequently, regardless of the context in which research or evaluation is undertaken, decision makers are unable to draw on evidence to steer decisions about what services to commission. If the point of research is to create real world impact, then this represents a huge waste of resources (16).

More fundamentally, the outcomes measured in intervention studies - particularly trials - do not always reflect concepts of success for those who use, deliver or pay for interventions (17, 18). Typically, outcomes measured in trials reflect the priorities of researchers and are only a partial reflection of what is important to other stakeholders. Since the goal of intervention studies is to understand which interventions benefit individuals, families, and communities, it is crucial that the outcomes measured reflect their priorities. Outcomes also need to be relevant to policymakers and service providers, so that effective interventions are funded and commissioned (17).

One approach to harmonising outcome measurement, whilst bringing together stakeholder priorities on what to measure, is to develop a core outcome set (COS). This is a standardised set of outcomes that researchers, providers, service users, and commissioners agree is important to evaluate the success of an intervention for a health condition or in this case, a complex public health challenge (19). The COS is then measured and reported, as a minimum standard in trials and evaluations and ideally practice-based monitoring too (20, 21). The aim is to improve research practice and reduce wastage, by increasing consistency and reducing reporting bias (where only favourable outcome effects are reported) and ensuring the views of all relevant stakeholders influence outcome selection. While the number of COSs being developed has increased (21), studies have focused on COS development for specific medical conditions, pharmacological, or surgical interventions delivered by healthcare professionals. By contrast, there has been less focus on the development of COSs in relation to public health problems like IPV that typically require complex, multi-agency responses.

Driven by our own experiences of trying to synthesise trial evidence to draw meaningful conclusions about effectiveness (13), as well as an increasing appetite for outcomes measurement amongst UK policy makers, in 2019 we set out to develop to develop two discrete COSs for psychosocial interventions aimed at improving outcomes for children and families at risk or with experience of (1) child maltreatment (CM) or (2) DVA. This saw us attempt to take a health-focused method and extend and adapt it to yield outcomes sets that i) were meaningful to the full breadth of psychosocial interventions on offer to these populations of children and families, as well as the multitude of systems and professionals (beyond health) involved in delivering the response, and ii) privileged the views of people with lived experience of abuse with respect to how the success of interventions should be defined.

In this paper we reflect on key aspects of the project so that others might be able to benefit from our learnings and consider ways of supporting COS development in fields beyond health. We focus specifically on development of the DVA-COS, as recent acknowledgement of children as primary victims of DVA (rather than secondary victims) in the UK has driven a strong policy 'pull' for this work, meaning it is more advanced.

2 Reflections

2.1 A broad scope

We set out to produce an outcome set that could be used to evaluate (in practice or research contexts) any interventions delivered to children or family members, with the aim of improving outcomes for children (<19 years) with experience of, or at risk of experiencing DVA. It is worth restating that a COS is intended as a *minimum standard* and that other outcomes specific to a given programme or population, can be measured alongside.

The scope for our work was necessarily broad to ensure its relevance to the range of interventions on offer which purport to enhance outcomes from children experiencing DVA, as well as the range of stakeholder groups and settings involved in responding to this group (11, 12, 22, 23). On this point we were met with sustained resistance from intervention developers and academic colleagues alike. They argued that different programmes would be characterized by different theories of change, and therefore it would not be possible to 'prescribe' a set of outcomes that could be relevant to all interventions. We responded to this argument in several ways. First of all, it presumes available interventions are carefully theorized and described, with clear links drawn between the components of the intervention and intended outcomes. However, DVA interventions are often poorly described with no explicit link to theory, or between activity and outcomes (13, 24). Second, the aims of programmes are often similar, and therefore it is plausible that programmes seek to change similar outcomes, even where mechanisms of change are different (24). Third, the field is already to some extent evaluating effectiveness against some common outcomes – for example, internalising and externalising behaviours– sometimes with no clear theoretical rationale for doing so. Moreover, these outcomes are defined by researchers, privilege measurement of mental health symptoms and diagnoses, and overlook other important aspects of functioning that are important to children and their families (23, 25).

We found it important to emphasise that a COS is intended as a *minimum standard*, with no expectation that an intervention should bring about change in *all* outcomes included in a COS. By articulating the mechanisms through which change in any outcomes are expected to be achieved, it can be made explicit why changes in some outcomes may not be plausible. Understanding which outcomes are *not* changed by a given intervention is just as informative as understanding those which are, in terms of guiding decisions about commissioning and selection. We also challenged developers (and sometimes our academic colleagues) to consider what it would mean in terms of an intervention's relevance to this population if it would have no plausible effect on *any* of the outcomes included in the final COS. The involvement of multiple stakeholder groups, particularly those with lived experience, and the use of consensus methods to select outcomes, added weight to this line of argument.

2.2 Involvement of key stakeholder groups in outcome elicitation and prioritisation

We are applied researchers, and, in this tradition, we sought to involve key stakeholder groups in all stages of the work. We were surprised at the enthusiasm of those with lived, service delivery and strategic experience to contribute to what could have seemed to be a 'dry' and methodologically focused endeavour. The DVA sector in the UK is chronically underfunded and we anticipated some 'push back' about the use of public funds for this upstream work that could have felt removed from direct service delivery. And whilst there were points of tension, overwhelmingly there was agreement that this was worthwhile and much needed work. We think that the project's policy relevance and a general focus on outcomes-based commissioning and evidence informed decision making contributed to stakeholder receptiveness. This was reflected in study participation (see Figure 1 for a summary of stakeholder involvement at each stage of the study) – in our final two consensus workshops a quarter of participants were policy and commissioning stakeholders from a range of central government departments and local authorities. [The remaining 77% was fairly evenly split between survivors, statutory and non-governmental practitioners, and academics. See (18)].

We were less successful in engaging researchers, particularly those outside the UK. We are taking steps to increase awareness of the COS amongst research communities however there is a risk that it is seen as UK specific and less relevant to international colleagues. As most trials are conducted outside of the UK, this may limit its impact on the unification of outcomes measured in effectiveness studies. Having said this, the study was funded by the National

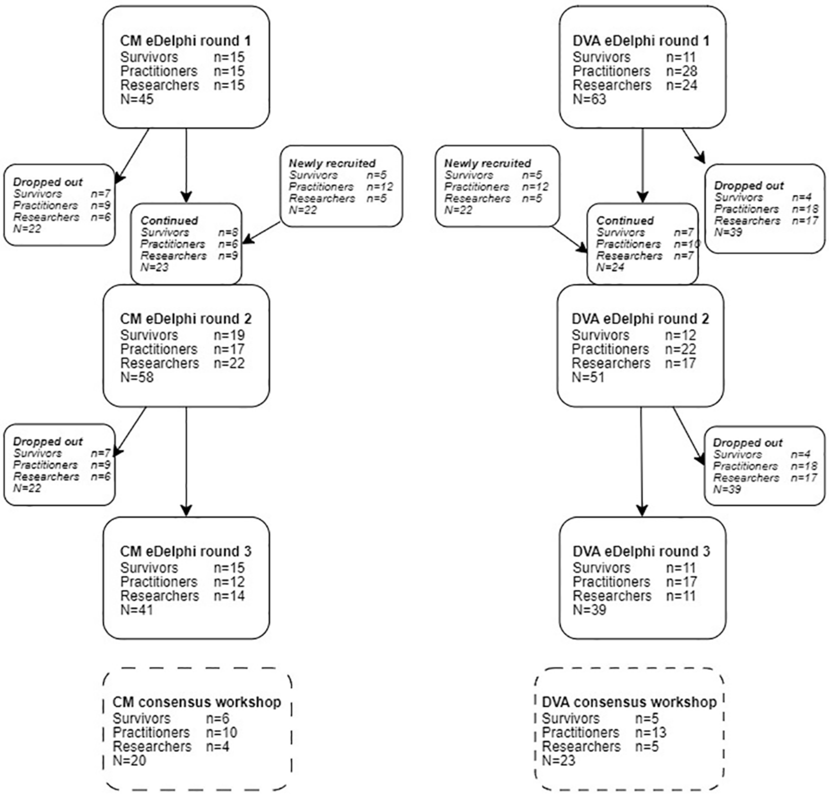

FIGURE 1
Consensus process participant flow chart. Reproduced from: Powell C, Feder G, Gilbert R, Paulauskaite L, Szilassy E, Woodman J, et al. Child and family-focused interventions for child maltreatment and domestic abuse: development of core outcome sets. BMJ Open [Internet]. 2022;12(9): e064397. Available from: https://bmjopen.bmj.com/content/12/9/e064397 No changes were made and re-use permitted under CC BY. Published by BMJ. https://creativecommons.org/licenses/by/4.0/.

Institute of Health and Care Research to support development of the UK evidence base, and so we need to be realistic about our ability to gain traction with international researchers, although others have shown this is possible (21).

In developing our initial protocol, we found there was limited guidance on involving patients, service users or members of the public in COS development or in multistakeholder consensus studies more generally (26). This perhaps reflects the more limited involvement of patients or members of the public in COS development (21, 27). We held an in-person workshop at the beginning of the COS development process, where the aim was to bring together survivors, practitioners and researchers to discuss and define outcomes, generate outcomes as a group and understand how different participants might prioritise outcomes. However, as others have flagged, we encountered some challenges in the bringing together of multiple groups for this purpose (28). We explore these in more detail in a forthcoming publication (26) but we learned that workshops involving trauma survivors and relevant professionals could be distressing without proper planning and support. We were able to use this learning to inform the planning of the final multi-stakeholder consensus meeting held at the end of the process (see below).

Whilst it is becoming more common to involve public representatives in outcome elicitation processes, it is less common to involve them in prioritisation of outcomes (29). To this end we used a modified e-Delphi study design (30). We thought that it could be more difficult to recruit survivors than professionals and this could result in survivor voices being lost in a mixed panel. In anticipation of this, we ran separate e-Delphi studies for each stakeholder panel. This enabled us to track recruitment more closely and recruit additional participants in the second round where needed. To orientate the other panel members to survivor viewpoints, we provided feedback on the survivor panel ratings (for each item) to participants in the researcher and practitioner panels (along with the standard information on group and individuals' ratings), although not vice versa. Outcomes were identified for discussion at the consensus meeting only if there was agreement across all three groups that an outcome was important (i.e. those rated important by only two groups for example were not taken forwards). For full details of participant flow and outcomes prioritized by each group, see (18).

Concerns about response burden (31) meant that we deviated from our initial protocol, in which we intended for participants to rate the importance of individual outcomes across successive rounds of the consensus process. Instead, in the first round of the Delphi we asked participants to rate the importance of outcome domains [groupings of thematically similar outcomes, see (18)], eliminating all outcomes associated with low-ranking domains. This of course may have resulted in exclusion of some important individual outcomes, but this felt like a necessary measure to ensure our methods were inclusive and realistic with respect to people's time. We gave the opportunity for feedback throughout the e-Delphi surveys, and we aimed to implement possible changes as quickly as possible throughout the process to widen inclusion. Early feedback from the first survey round suggested the mode of delivery and the language used in survey excluded some survivor participants. To mitigate this, we offered additional support to survivors to complete the survey by phone or email in subsequent rounds. This involved a researcher carrying out the survey over the phone, offering clarifying explanations where needed, and inputting responses into the software for the participant, or sending the survey as a word document attached in an email for participants who struggled to access the software. A researcher then entered the data into the software for the participant.

In reflecting on the study there are important learnings that will inform our future endeavours and may be helpful to others looking to involve people with lived experience of the topic at hand, in their development process. Primarily, researchers should not overlook the potential to cause harm through the research process, particularly when working with vulnerable groups such as those with lived experience of abuse, mental health difficulties and bereavement. The marginalisation or exclusion of individuals or groups in the COS development should not be underestimated as a source of harm. Work is currently underway to explore in more detail harms associated with the development process (32). However COS developers could usefully draw on the extensive mental health research co-production and co-design literatures (33, 34), which highlight the importance of knowledge-based practice and lived experience (35) to improve consensus processes, and acknowledge and mitigate power differences between researchers and service providers (36).

Second, full involvement of survivors in multi-method consensus research, alongside researchers and professionals, requires substantial reflection and planning that extends beyond current guidance on COS development or involvement work more generally. It takes time and money, and this should be factored into research budgets. There is need for specific guidance to support this aspect of the COS development process, including principles as basic as reminding researchers and practitioners how to behave and communicate in multi-stakeholder workshops (28).

Thirdly, it is also worth considering specific measures to ensure that the survivor/patient/service user voice is not lost or diluted through the consensus process. We found the input of a lived experience advisory group to be invaluable from this respect, although again this support needs to be properly resourced from the outset. The approach of running separate Delphi studies and providing feedback on survivor ratings seemed to work well although it significantly increased the resource required and there is only limited evidence that this approach enhances other stakeholder views of service user/patient perspectives (37). We support the call for more empirical research on the best ways to support public involvement in COS development.

Finally, we urge researchers committed to involving patient groups of public representatives to be flexible in their approach – parts of the process set out in the protocol may need to change to facilitate or maintain involvement. This should be encouraged, and deviations transparently reported so that others may benefit from learnings.

2.3 Use of a range of evidence sources to identify candidate outcomes

Whilst COS guidance places primacy on conducting a systematic review of trials to identify candidate outcomes, it was necessary for us to draw on a wider range of evidence sources including qualitative and grey literature. Although rigorous, systematic reviews of intervention studies may not include outcomes that survivors see as important. Outcomes in trials are more likely to be in line with research and clinician priorities (17, 38), survivor priorities for outcomes are more likely to be reported in qualitative or grey literature. Thus, a focus solely on trials potentially excludes outcomes of importance to survivors. Our review (39) found that more candidate outcomes were identified in the grey and qualitative literature than the trial literature, and that these outcomes were more nuanced. The inclusion of diverse evidence sources has a direct impact on the final selection of outcomes. In our two COSs, three out of the final eight (unique) outcomes were *only* identified in the grey and qualitative literature. Current guidance needs updating to reflect the importance of evidence sources beyond trials, particularly when the COS may be applicable to marginalised groups whose views may not be well understood or reflected in published research.

2.4 The final consensus statement

We used a professional facilitator to help us plan and deliver our final meeting, which was held online during the pandemic, and included representatives from all key stakeholder groups (26). We also paid for the services of a trained counsellor who was available during and following the meeting to respond to any distress experienced by participants. Both were key to the meeting's success which we gauged not just by the output, but from the feedback we received from participants regarding the respectful and inclusive nature of the debate.

During the meeting we sought to reduce the shortlist of outcomes established by the Delphi study to a list of five. Previous discussions with service delivery stakeholders highlighted feasibility of the COS would be impeded if the set was too large. The final DVA-COS is reported in full elsewhere (18) but included: 1) child emotional health and wellbeing; 2) feelings of safety; 3) freedom to go about daily life; 4) family relationships; 5) caregiver emotional health and wellbeing. It is notable that one of the outcomes (freedom to go about daily life) has not yet been measured in quantitative research, suggesting the process did its job in identifying overlooked outcomes that are important to the users of evidence.

It was also significant the COS included adult and child wellbeing, and that these outcomes were favoured over measurement of mental health (it was possible to include both outcomes in the COS). Research highlights that wellbeing and mental health are separate, although overlapping constructs (40), and that wellbeing outcomes, capturing the extent to which an individual is flourishing, are less often measured in trials relative to mental health outcomes, which are concerned with deficits and distress (17, 41). This finding resonates with early discussions with lived experience experts who expressed a desire for research to capture impact in a more holistic, hopeful, and forwards looking way, rather than by reduction to clinical symptoms and diagnoses, which they saw as overly deficit focused. That said, more work is required to further define outcomes included in the COS (as well as identify measurement tools) to enhance conceptual clarity and reduce potential for misunderstanding between researchers and practitioners (42).

2.5 Resource

For the reasons outlined above, the costings and to some extent the time frame were higher and longer than other projects listed on COMET and NIHR websites. Complex COS development needs to be adequately and realistically resourced, particularly when thinking about vulnerable groups or any work that has a broad scope and necessarily involves a range of stakeholder groups.

2.6 Implementation

One of the key aims of COS development is reduction in research wastage, however a COS study itself is a waste if nobody uses the output (16). Whilst few (relative to the number of COSs) uptake studies have been undertaken, synthesis of available evidence shows use in trials and systematic reviews to be low (16). Key reasons for this include lack of researcher awareness and understanding about relevant COS, a lack of precision in the definition of outcome domains, a lack of consensus on how to measure outcomes included in the COS, and concerns about a lack of stakeholder (including patient/public) involvement in the development process.

Whilst we were proactive in involving key stakeholder groups from the outset of the study, as noted above, we were less successful in engaging researchers in the process, particularly those from outside the UK. There is a risk here that lack of awareness, or a perceived lack of relevance to our international colleagues may prevent uptake of the COS by trialists and other academic researchers. This may be compounded by the fact that much DVA research, particularly with respect to children, seems to be undertaken outside health, in disciplines such as psychology, social work and social policy. Therefore, the COS, as the product of a health method, may be perceived as less relevant by researchers in other disciplines. We acknowledge we need to do more active work (vs passive dissemination) to increase awareness of the research community. However, this takes time and money that as, yet we are still to secure. Our funding only supported the development of what to measure and did not include funds for the 'how', which is significant given this is one of the key barriers to implementation (16, 43).

Demand for the COS amongst service commissioners and providers, facilitated by significant policy developments in the UK, has highlighted the (often cited) tension between policy and practice and research (44). A recent programme of government funding for services for children affected by domestic abuse stipulated that programmes would only be considered eligible for funding if they were able to map how interventions may facilitate change in each of the five outcomes included in the COS and agreed to evaluation of impact against the five outcomes. As researchers, this is the type of impact that we are striving for, and such endorsement of a COS can positively influence uptake (45). However, the desire for immediate policy implementation has been a challenge to our desire to run a properly resourced and rigorous measurement tool selection process aligned with current guidance (44). In order to respond to this 'pull' for evidence we have needed to undertake some (very) rapid interim work with a group of service providers to identify measurement tools that are 'good enough' to support evaluation of a specific programme of work.

We were able to build on previous work to map the COS against practice-based measurement tools commonly used in practice (46). However, this work could not identify measures for three of the outcomes (safety, family relationships and freedom to go about everyday life) that were acceptable to both service users and providers as well as psychometrically sound. Feedback was that tools were deficit focused, sometimes traumatising to complete, and too narrow in focus. Our subsequent searches, although broader in scope, concurred. We found few measurement tools that had been developed specifically to measure outcomes for this population, and little evidence that general tools had been validated for use with children and families experiencing DVA (22, 47). 'Freedom to go about everyday life' was a concept that was not captured by any of the tools we reviewed; it seems highly likely that work will be needed to further define this outcome and develop a relevant measurement instrument.

We have come up with an interim set of tools to measure four outcomes in the COS, but there is a risk that this interim way suite of measures is shared and becomes adopted as the final recommendations on measurement, when we have not yet been able to carry out a thorough selection and consensus process, and there is no suitable measure of one of the outcomes. There are no easy answers to this paradoxical situation – as applied researchers we strive to create the 'pull' for evidence, but this may mean offering up incomplete findings that become embedded into practice and difficult to update. This could be mitigated to some extent however, if funders committed to funding both the 'what' and the 'how' of COS development and were realistic in terms of what this will cost. This requires giving explicit permission to researchers to apply for larger amounts than have been awarded for COS development thus far. In fields such as our own, where the funding for intervention development and service delivery often comes from large charitable organisations, development of better ways to measure outcomes cannot simply be an academic led and funded endeavour. There needs to be a much stronger commitment from these bodies to fund and implement core outcomes work as a mechanism to improve the response to children and families though evidence informed decision making.

3 Conclusions and recommendations

Although the origins of COS development are rooted in health research, we found this to be an appropriate method for addressing disparate outcome measurement in relation to child focused DVA interventions, whilst also reflecting the perspectives of survivors of abuse as well as other evidence stakeholder. If we can facilitate uptake, we feel there is genuine potential, albeit some way down the line, to improve the service response to children and families experiencing CM and DVA through better evidence informed decision making about what works.

Given COS development has much to offer other disciplines looking to unify outcome measurement within and between academic and practice-based contexts, we suggest current guidance is updated to reflect this wider application. This could be achieved with additional examples and case studies, and explicit acknowledgement of the utility to disciplines beyond health.

We also suggest inclusion of practical guidance to support the full and meaningful involvement of members of the public, particularly those with lived experience of the topic at hand. Finally, we think that much greater emphasis should be given to the use of diverse evidence sources beyond trials, with recognition that this may be particularly important when working on problems that impact underrepresented and marginalised groups.

We recommend that researchers and funders are realistic about the time and money that is required to undertake a development process that represents the views of all important evidence stakeholders (through involvement and review of evidence). To maximise its value and to make it most meaningful, we recommend that involvement work begins as early as possible and draws on a range of methods across the development process (workshops, written updates, informational videos, briefings).

Finally, we advocate strongly that funders commit to funding both the 'what' and the 'how' of core outcome development. It is a false economy to fund only the identification of key outcomes without developing consensus on which tools should be used to measure them. Indeed, it may contribute to research wastage.

Data availability statement

Data are available upon reasonable request. Please contact the corresponding author or unit manager (cpru.data@ucl.ac.uk) with enquiries about the data used in this study.

Ethics statement

Ethics approval was provided by University College London's Research Ethics Committee for involving research participants (17893/001 & 002) and we were guided by a steering group of eight professionals. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

EH: Conceptualization, Investigation, Methodology, Supervision, Writing – original draft. GF: Conceptualization, Funding acquisition, Investigation, Methodology, Writing –

review & editing. CB: Funding acquisition, Investigation, Writing – review & editing. CP: Data curation, Formal analysis, Investigation, Methodology, Project administration, Software, Supervision, Writing – original draft, Writing – review & editing.

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Conflict of interest

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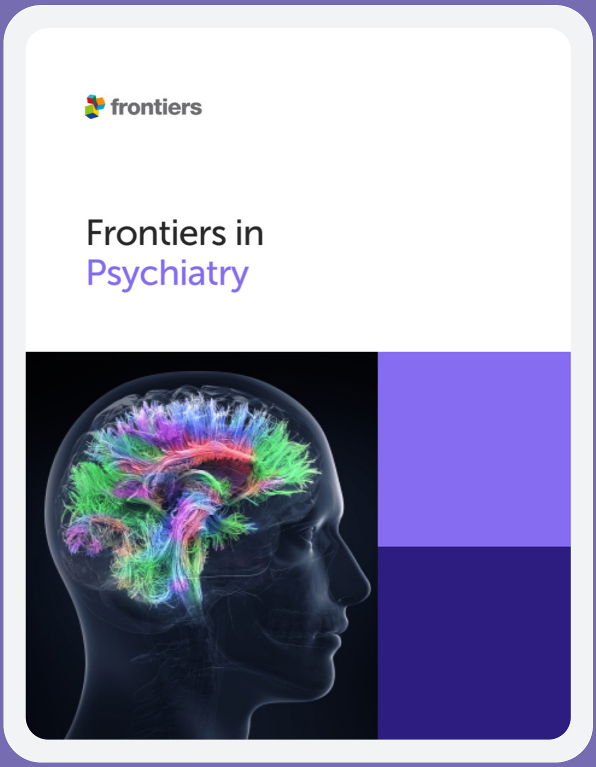

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