DRAFT:

Trauma-focused to Support Caregivers of Children with Severe Illness: A Caregiver-Initiated Systematic Review

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Introduction

A child's diagnosis of a life-limiting or life-threatening disease, or complex medical condition, is a known traumatizing event for family caregivers (ref). The child's prognosis and care or treatment regime, the provision and witnessing of direct medical care to the child, family financial burdens, regular complex decision-making work, and care coordination tasks all contribute to caregiver distress. Family caregivers experience significant psychological challenges, including anxiety and depression at levels equal to or greater than patients, and exceeding levels experienced by the parents of healthy children. Up to 20-30% of family caregivers may experience post-traumatic stress symptoms (PTSS) [14]. Such symptoms are associated with decreased caregiver quality of life, poorer physical health, and diminished social functioning. Moreover, within the context of life-limiting and complex pediatric illness and care, family caregivers—who most often include immediate family members, such as parents, siblings, grandparents, and legal guardians—are essential supports to their child's health and wellbeing. As such, child health outcomes be negatively impacted by impairments to caregiving ability caused by psychosocial strains to family caregivers.

These risks to caregiver and child well-being highlight the necessity for effective family caregiver support interventions and services as components of family-centered care. Trauma-informed care interventions, also known as trauma- and resiliency-informed care interventions; to highlight the strengths-based nature of the approach [20], are those interventions that directly target the prevention or mitigation of trauma-related symptoms using specialized psychological approaches, and emphasize the importance of recognizing the signs and impact of trauma when providing specialized psychological practices [9]. These interventions have the potential to improve PTSS-related outcomes in the family caregivers of seriously ill children, enabling them to feel empowered despite their traumatic experiences [9].

However, little is known of the trauma experience of caring for and/or losing a child who has a life-limiting or chronic illness—particularly in relation to the existence and effectiveness of trauma-informed care interventions for caregivers. To begin to inform the design and development of effective trauma-informed interventions that might be implemented into routine pediatric healthcare practice, an examination of the related state of the science is needed. To challenge current paradigms related to stakeholder engagement in intervention design, an identification, critique, and synthesis of both qualitative and quantitative research is required to gain a deeper understanding of how caregivers experience trauma-informed care interventions.

The overall objective of this parent-led systematic review is to synthesize the literature on the effect of trauma-informed care services for the prevention and management of traumatic stress and related symptoms in the family caregivers of children with life-limiting illness and complex medical conditions. Our primary objective is to describe the effectiveness of trauma-informed interventions on post-traumatic stress symptoms in caregivers. Our secondary objectives are to: a) describe the effectiveness of trauma-informed interventions on other trauma-related symptoms including general psychological wellness, and somatic manifestations associated with trauma in caregivers; b) examine which interventional and study methodological characteristics might explain any heterogeneity in intervention results; and c) synthesize themes describing caregivers' experiences of receiving trauma-informed care interventions, the impact of such care on family caregivers, and their suggestions for service improvement.

Methods

The aims for this review were developed by BP, who is a parent of a child with a CMC, and all aspects of the conduct of this review were family caregiver-led. The review protocol was registered on PROSPERO (CRD42022331536). Our reporting is in accordance with Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) (ref) and PRISMA-Search (PRISMA-S) (ref) checklists.

Search Strategy

Our strategy was developed by our team of family caregivers of children with CMCs, pediatric healthcare clinicians, and a health sciences librarian (MM). We searched Medline, PsycINFO, Embase, Cumulative Index of Nursing and Allied Health Literature Plus (CINAHL), the Cochrane Library, the Applied Social Sciences Index and Abstract, and Social Services Abstracts from inception to May 3, 2023 with no limitations or filters. We used a combination of subject headings and text words specific to each database to search for the concepts of traumatic stress and family caregivers. Using the Peer Review of Electronic Search Strategies (PRESS) guidelines(ref) the Ovid Medline search strategy was peer reviewed by an independent health sciences librarian prior to search translation. The search strategy is presented in the Supplementary Information. Reference lists of included studies and identified systematic reviews were also searched for potentially relevant articles. When a relevant conference abstract was located, we contacted authors up to twice to query about any associated full-text article, which we reviewed for relevance.

Inclusion and Exclusion Criteria

All team members discussed the application of eligibility criteria prior to screening and the criteria were piloted by two authors (PP and IZ). We included studies that: (1) were primary interventional or observational research studies; (2) focused on a family member providing significant care and support to a child (such as a parent), with the associated child being <25 years of age with medical complexity or a life-limiting condition; (3) examined a trauma-informed intervention, defined as one utilizing specialized psychological approaches to prevent or mitigate trauma-related symptoms; and (4) presented data on any of patient-, caregiver-, family-, and system-level outcomes associated with use of the trauma-focused intervention. We included articles published in all languages. Articles that were not published in English were translated to English by collaborators fluent in the language of writing.

Study Selection

Within Covidence (ref), two blinded reviewers independently screened the titles and abstracts (PP, IZ, or KH) and then full-text records (PP, IZ, or CC) of the obtained records. At both stages, discrepancies between reviewers were resolved by a third team member with lived experience of family caregiving (BP).

Data Extraction

A standardized REDCap-based electronic form was developed to support data abstraction and calibrated prior use with a sample of five articles. Data extraction was completed in duplicate (PP and IZ) for 24 articles with >80% agreement. Data extraction for the remaining articles was completed by one author and double-checked by the other. Any identified issues in abstraction were resolved through discussion will all team members.

Assessment of Study Quality and Risk of Bias

We used the 9-item checklist developed by Hawler et al. (ref). to appraise the quality of the included studies. This checklist was designed to accommodate the evaluation of studies utilizing heterogenous research methodologies while gauging the validity and quality of the studies. Two reviewers independently completed quality appraisal (PP and IZ), with disagreements resolved by discussion and consensus among all team members.

Data Synthesis

Given the heterogeneity in study types and methodologies, we used a narrative synthesis method to compile the results. We organized identified trauma-focused interventions according to the theoretical framework underpinning each intervention, as described by the study author or in consultation with a pediatric psychologist (AH). Study outcomes were classified into two categories: trauma symptoms and positive psychosocial outcomes. For each category of outcomes, we reported the overall frequency of observed improvements, as well as the specific improvements for each outcome and the total number of times they were measured. Remaining symptoms were unchanged after intervention implementation, unless otherwise stated. We reviewed each studies result section for qualitative data pertaining to caregiver satisfaction with the intervention or suggestion for improvement and coded these data using a conventional content analysis approach.

Results

Study Selection

Our search retrieved 31,070 articles with 15,170 unique references. Following title and abstract screening 14,967 articles were ineligible and excluded from further analysis. The remaining 203 articles were assessed as full-texts, of which 155 were excluded. A list of articles excluded at the full-text stage and rationale is available in the **Supplementary Material**. A total of 48 studies were included in the review **(Figure 1).**

Study and Sample Characteristics

The details of each study are presented in **Table 2.** Of the 48 included studies, 29 (60%) used quantitative and 19 (40%) used mixed methods methodologies. The most frequently used study design was the randomized controlled trial (n=29; 60%). Studies were conducted across 50 countries, most commonly the United States (n=20), Australia (n=6), and Sweden (n=5).

A total of 4164 caregivers were included across all studies. Family caregivers included in studies were parents (in all studies) and others including grandmothers, aunts, and siblings in a minority of studies (n=4; 83%). When reported, most caregivers were Caucasian. The average proportion of female participants across studies was 71%, with data missing from eight studies. Children's primary health conditions were cancer (n=38; 79%), congenital heart disease (n=6; 13%), cerebral palsy (n=2; 4%), and mucopolysaccharidosis type III (n=2; 4%). A total of 174 trauma-associated symptoms were evaluated with the most common being post-traumatic stress (n=28), anxiety (n=27) and depression (n=23). The most frequently measured positive psychosocial outcomes were quality of life (n=12), psychological flexibility (n=9), and benefit-finding (n=7)

Quality Assessment of Studies

Based on the Hawker checklist, 46 (96%) studies rated "high" in quality, while the remaining two studies rated "fair". The mean Hawker checklist score for studies was 33 out of 36 possible points (SD=2.47; range 24-36) (**Table 3**). Common study weaknesses were a lack of data transferability and generalizability (146/192 possible points), insufficient eligibility criteria explanation, scarce demographics details or sample size justification (166/192 possible points), and insufficient statistical data analysis explanation or reasoning (171/192 possible points).

Appraisal of our systematic review was conducted using the 16-item AMSTAR 2 checklist, with results shown in **Table 4.** Of the listed criteria, n=10 was fully met, n=1 was partially met, n=2 was not met, and n=3 was not applicable.

Intervention Characteristics

Across all studies, a total of 52 interventions were evaluated. Descriptions of interventions and their effectiveness can be found in **Table 5**.

Administration modality and interventionalist

Interventions were digitally administered (n=17), conducted within a hospital (n=15), conducted at a cancer support organization (n=1), in-person with no further specifications (n=1), and via administration methods (n=4). Ten studies did not specify the location of intervention administration. Interventions were primarily administered by clinicians (n=33), including psychologists, therapists, nurses, or social workers. Other interventionalists were graduate students (n=7), research team members (n=1), unspecified interventionists (n=5), or collaborative groups of multiple professionals (n=4).

Outcomes by Intervention Type

Problem Solving Skills Therapy

Eleven interventions used a problem-solving skills framework to teach cognitive, behavioural and social coping strategies to family caregivers(refs). Outcome improvements were reported for 25 of 31 (81%) trauma symptoms assessed across these studies, particularly for anxiety (n=2/3), depression (n=6/7), illness uncertainty (n=1/1), mood (n=7/7), posttraumatic stress (n=8/9), and worry (n=1/1). Of the 13 positive psychosocial outcomes assessed in these studies, 9 (69%) improved. These improvements were in benefit-finding (n=1/1), coping (n=1/1), and the acquisition of problem-solving skills (n=7/8). Four interventions used a problem-solving framework to teach medical management. Of seven trauma symptoms assessed, improvements were seen in 5 (71%) symptoms, including anxiety (n=2/2), depression (n=2/2), and stress (n=1/2). In the two instances where in quality of life was measured, it improved once (50%).

Overall, 12 problem solving skills-focused interventions improved all symptoms measured; 9 of which focused on cognitive and behavioural coping and 3 focused on teaching medical management for the child.

Cognitive Behavioural Therapy

Twelve interventions were underpinned by a CBT framework. Of the 37 trauma symptoms measured, 16 (43%) improved post-intervention. These improvements were for anxiety (n=3/8), avoidance (n=1/1), depression (n=3/7), insomnia (n=1/1), posttraumatic stress (n=5/6), rumination (n=1/ 2), stress (n=1/6) and worry (n=1/1). Improvements were also seen in five of nine (55.5%) positive psychosocial outcomes, which were resilience (n=1/1), coping (n=1 /2), posttraumatic growth (n=1/1), quality of life (n=2/4). No improvement in any symptom was reported in three CBT-focused interventions. In contrast, four interventions showed improvements in all symptoms measured; one of which incorporated mindfulness-based techniques and one which utilized a resilience-based theory to CBT delivery.

Family Therapy

Three interventions were based on the principles of family therapy. These interventions were not associated with improvements in any of the four trauma symptoms measured. A single positive psychosocial outcome was measured, post-traumatic growth, which improved. Six interventions combination of family therapy and cognitive behavioural therapy. A total of 19 trauma symptoms were measured. Improvements were seen for 11 (58%) symptoms, which were anxiety (n=4/6), distress (n=3/6), posttraumatic stress (n=3/5), and psychosocial risk (n=1/1). The effectiveness of these six interventions appeared to vary by caregiver sex or caregiving role. Only fathers experienced improved anxiety (n=1/6), distress (n=1/6), and posttraumatic stress (n=1/5), whereas, in a separate study, only mothers experienced improved distress (n=1/6). Worsened avoidance only in fathers was reported in a single study. All three positive psychosocial outcomes measured were improved, including family functioning and relationships (n=2/2) and quality of life (n=1/1). A single study evaluated family therapy in conjunction with acceptance and commitment therapy. Improvements were seen for two of three (67%) trauma symptoms measured, namely depression (n=1/1) and stress (n=1/1), with no change in caregiver anxiety (n=1/1). Overall, three interventions were not associated with trauma-related changes for any evaluated symptoms and three interventions were associated with improvements in all symptoms.

Acceptance and Commitment Therapy

Four interventions utilized the principles of ACT. Thirteen trauma symptoms were measured, and seven (54 %) were improved, including posttraumatic stress (n=3/3), stress (n=2/4), anxiety (n=1/3), and depression (n=1/3). Of the 12 positive psychosocial outcomes measured, 11 (92%) were improved. These were mindfulness (n=2/2), psychological flexibility (n=2/3), acceptance (n=1/1), parental adjustment (n=2/2), and psychological functioning (n=3/3). No intervention related improvement in well-being was observed. Two ACT interventions were not associated with any change in trauma-related symptoms and two interventions were associated with improvements in all evaluated symptoms.

Guided Self-Help

Of the included interventions, three used a guided self-help intervention framework. Across studies, 9 trauma symptoms were assessed. All symptoms (100%) improved, which encompassed posttraumatic stress (n=3/3), depression (n=3/3), anxiety (n=2/2), rumination (n=1/1). A single positive psychosocial outcome, behavioural acceptance of difficult thoughts/emotions (n=1/1), was measured and showed improvements (100.0%).

Written Disclosure

Of the eight trauma symptoms measured, only posttraumatic stress (13%) was improved following written disclosure. Worsening of symptoms was observed once (13%) in the case of physical health. No change was observed in the single positive psychosocial outcome measured, quality of life.

Eye Movement Desensitization and Reprocessing

Two interventions used EMDR. Of the six trauma symptoms assessed, all (100%) improved post-intervention. Improvements were for posttraumatic stress (n=2/2), distress (n=2/2), parenting stress (n=1/1), and comorbid psychological symptoms (n=1/1).

Other Interventions

Four distinct intervention were evaluated.

Intervention Satisfaction and Improvement Suggestions

Fourteen studies reported qualitative findings pertaining to intervention satisfaction. Caregivers across nine studies (6$%) reported high satisfaction whereas, caregivers in five separate studies (36%) reported mixed, however generally positive, experiences with the trauma-focused intervention. High satisfaction was often associated with online, remote, or asynchronous intervention formats and interventions that included peer support and the normalization of experienced emotions. Five studies (31%) reported qualitative caregiver suggestions for intervention improvement. Suggestions were for increased intervention duration and frequency, additional online intervention components, and the use of group-based activities.

Discussion

Interventions and Effectiveness

This review identified promising groups of interventions that have the potential to mitigate trauma-related symptoms experienced by family caregivers of children with CMCs and LLIs. All interventions included in this review were those that utilized a trauma-informed approach to care. These interventions have the advantage of minimizing caregiver experiences of distress while also providing emotional support to family members, effective coping strategies, and guidance to facilitate recovery. Collectively, these interventions utilize a strengths-based approach, emphasizing an individual's personal strengths and abilities to overcome adversity, rather than utilizing a traditional medical model which may pathologize individuals.

While intervention effectiveness is promising, results showed mixed effects, in agreement with other systematic reviews evaluating the effects of psychosocial interventions on the mental health of family members of children with medical complexities and chronic illness. This heterogeneity in findings may be accounted for by the methodology used by included studies. As we aimed to evaluate a broad spectrum of interventions; including those at the initial and final stages of testing and those reporting caregiver experiences; we included studies of any design. The sample size of the included studies was small, leading to difficulties in generalizing individual study results, and may account for the variability in results reaching statistical significance. Further, multiple studies assessed the same outcomes, however, a vast array of measurement tools were used.

-Heterogeneity in results (population characteristics: illness uncertainty, parental age, child age, child prognosis)

Future Research

Although our review identified a vast number of interventions, interventions supporting family caregivers of diverse demographic characteristics, including caregiver gender, role and race, as well as child diagnosis, were limited.

Interventions mainly targeted caregivers identifying as mothers. Female individuals account for majority of caregivers, which holds true in the context of pediatric illness. Literature indicates a need for interventions specific to this population as mothers of children with chronic and/or life-limiting illnesses tend to report elevated levels of post-traumatic stress, anxiety and depression. However, fathers remain understudied in pediatric samples, often due to convenience sampling and beliefs that they are disinterested in such services. Therefore, future studies should aim to recruit and investigate intervention effects on fathers. Doing so is especially important as adverse psychological symptoms experienced by one parent have been established as reciprocally influencing the other.

Most caregivers identified in the studies included were those who were Caucasian, as well as parents of the ill child. Family caregivers from minority ethnic groups tend to experience greater traumatic stress and may have greater potential to benefit from such interventions, however, they are not adequately represented and are thus understudied. Therefore, future research efforts should aim to investigate the effects of interventions in individuals of differing race and ethnicities. Additionally, it is important to recognize that household familial structure and thus child rearing and caregiving differs not only at the individual level, but also across different cultures and geographic regions. For instance, children in developing countries often co-reside with grandparents as families are dependent on their help, and rural areas have more children per household. It is therefore important for future research to establish intervention efficacy across caregivers who vary in their relationship to the ill child, to better understand how to treat the family as a unit.

Family caregivers of children with cancer were most often targeted by interventions. While a diagnosis of cancer is given to approximately 300,000 children globally and is known to have significant negative psychosocial effects on caregivers, it is important to recognize that other complex medical conditions also affect many pediatric patients. For instance, a retrospective cohort study identified pulmonary-respiratory, neurologic, cardiovascular and metabolic conditions to be more prevalent pediatric medical complexities as compared to other malignancies, identifying a diverse population of medically complex patients who rely on family members for caregiving.

Limitations

This study has several limitations.

Our review only includes peer-reviewed literature identified by our database search, as it was not feasible to conduct a grey literature search. As a search of the grey literature often increases the likelihood of identifying unpublished studies with negative or null results, our review may have inadvertently omitted other relevant findings, thus limiting the comprehensiveness of our review and conclusions that may be drawn.

Further, completion of risk of bias assessment using the 9-item Hawker et al (2002) checklist indicated study quality was generally high. This tool was selected to accommodate the vast array of study designs included in our review; however, it was not explicit in addressing important methodological features of included studies such as confounding and selection bias in non-randomized designs, and participant blinding and unconcealed allocation in randomized designs. As these features were not adequately assessed, this may have led to an overestimation of the quality of the studies included, and findings should therefore be interpreted with caution.

Implications for Practice

As evident in our review, trauma-informed interventions have substantial promise in alleviating post-traumatic stress and other trauma-related symptoms in family caregivers of children with CMCs and LLIs. To implement such interventions into clinical practice, it is critical that clinicians receive thorough training to better identify such symptoms and routine screening measures are established to identify caregivers who will benefit most from receiving such interventions. This proactive approach will allow for more timely implementation of these resources to not only mitigate, but also potentially prevent these symptoms from occurring.

Additionally, our review identified interventions targeting and effective in distinct populations of caregivers, such as caregivers differing in sex, gender, age and child diagnosis. Therefore, stratification of interventions to specific subsets of family caregivers may aid in optimization of intervention effectiveness.

As our review identified a wide variation of existing interventions, each slightly varying in content and delivery, usual constraints impeding caregiver participation in such interventions can potentially be alleviated. Frequent barriers to mental health services for parents and siblings of children with special health care needs include inconvenient service times and locations for 12.3% and 8.7% of caregivers, respectively. Our review identified interventions differing in location of intervention administration (e.g., online asynchronous, at pediatric health care center etc.) and intervention frequency and duration, offering multiple viable options for caregivers whose access to such services may otherwise be restricted. The variation in identified interventions may also mitigate logistical challenges in implementing such services into clinical practice, as differing levels of interventionist involvement and required training may address concerns related to the number of personnel involved, required space in clinical settings, and other costs associated with implementation.