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REVIEW

The Family Stress Model in the Context of Pediatric Cancer: A Systematic Review

Christine Neugebauer[1,2] · Ann M. Mastergeorge[1]

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Abstract

A systematic review of the pediatric cancer literature was conducted to identify and summarize the variables that influence family-level psychosocial outcomes and to develop a conceptual framework based upon the Family Stress Model. PubMed, PsychInfo, Web of Science, and CINHAL databases were searched between 2008–2018 using PRISMA. Empirical studies in this review examined both uni- and bidirectional relationships between at least two members within the family (e.g., parent and child) or measured a component of family functioning (e.g., cohesion, adaptability). After screening 2815 records, 10 review studies and 52 empirical studies met the study criteria for inclusion. Variables related to family-level psychosocial outcomes were synthesized into five thematic categories: family, parent/caregiver, child with cancer, immediate and extended relationships, and socioeconomic. Potential mediating and moderating effects reported included family cohesion, family ritual, family environment, family functioning, and parenting stress and caregiver burden. Based on this review, a conceptual path model was developed to illustrate factors pertaining to pediatric cancer that may be applied to the Family Stress Model (FSM). This adaption of the FSM in pediatric cancer integrates the factors of economic stress and pediatric cancer on outcomes related to family functioning and child/sibling psychosocial functioning that are central to families dealing with cancer diagnoses and can be utilized to explicate the relative strength and influence of risk and protective factors.

Keywords Pediatric cancer · Family functioning · Family stress model · Financial stress · Systematic review

Highlights

- A conceptual framework applying the Family Stress Model displays the directional pathways influencing families experiencing a diagnosis of pediatric cancer.
- Specific financial stressors related to the cancer diagnosis contribute to economic pressure which impact parent psychological functioning.
- The Family Stress Model can be applied to expand understanding of the relationships between economic pressure and family-level outcomes as well as the many variables that mediate and moderate those relationships.

Pediatric cancer is a negative life event that triggers a state of chronic stress across the family system complicated by and not limited to fear of losing a child, sustained medical treatments and hospitalizations that impact family separation, strained caregiver roles, and financial stress (Long & Marsland, 2011; Van Schoors et al., 2017). The chronicity of this stress has been described in studies that examined the direct effect caregiver burden and chronic parenting stress have on child quality of life and adjustment (Hamner et al., 2015; Wolfe-Christensen et al., 2010). Over the past several years researchers have examined the role of family functioning within the context of pediatric cancer and identified family processes to include both risk (Pierce et al., 2017) and protective (Santos et al., 2015) factors impacting coping (Monti et al., 2017) and adjustment (Mullins et al., 2016) on

✉ Christine Neugebauer
Christine.neugebauer@ttuhsc.edu

[1] Department of Human Development and Family Sciences, Texas Tech University, Lubbock, TX, USA

[2] Department of Pediatrics, Texas Tech University Health Sciences Center, Lubbock, TX, USA

individuals as well as the family system (Van Schoors et al., 2018).

Previous reviews have explored a range of factors related to family-level outcomes in pediatric cancer including parenting stress (Vrijmoet-Wiersma et al., 2008), pediatric quality of life (Klassen et al., 2011), family adjustment (Long & Marsland, 2011), and resiliency (Van Schoors et al., 2015). Of significant consequence in this review is determining the relevance economic stress has as a key variable in studies measuring family-level influences in the pediatric cancer literature. Given the financial implications of treatment for cancer, it is noteworthy that economic stress has received limited attention in previous reviews specific to family functioning in the context of pediatric cancer (e.g., Long et al., 2018). A meta-analysis conducted by Van Schoors et al. (2017) identified a significant association between family functioning and child outcomes in families of children with cancer; however, there was no reference to the role financial burden may have on the family system. That is, the economic burden of cancer affects families at all socioeconomic levels with families at economic risk experiencing the most financial stress, and mid-level income families at risk of falling below the poverty line (Bona et al., 2014). In their review specific to financial burden for families experiencing pediatric cancer, Pelletier & Bona (2015) concluded that financial hardship impacted a high proportion of these families and identified economic burden as a strong negative influence on family functioning. High medical bills, missed time from work, unanticipated hospitalizations, and caring for an ill child at home result in lost income due to decreased hours at work or employment termination (Lau et al., 2014; Limburg et al., 2008). The financial strain associated with having a child with a catastrophic illness has been linked to increased parental depression, lowered quality of life, and decreased family well-being (Creswell et al., 2014; Miedema et al., 2008). The fact that financial and economic stress has received minimal attention in pediatric cancer research is surprising given that it has been documented in the social science literature to be a central component to overall stress levels for families (Conger & Conger, 2002).

Currently, there are a few conceptual models available to guide research on exploring family-level processes for families coping with pediatric cancer. For example, Wallander & Varni (1998) offer a transactional model of stress and coping that can be applied to families of children with chronic illness. Their model includes several disease, psychosocial, and ecological variables that serve as either risk or mitigating factors to family adjustment. Likewise, the Resiliency Model of Family Stress, Adjustment, and Adaptation (McCubbin et al., 2002) posits that family stress and hardship are natural occurrences across the family life cycle and adjustment to these stressors stems from within family relationships and external societal processes. Additionally, both the Social Ecological Post-Traumatic Stress Framework (Kazak 2007; Kazak & Baxt, 2007) and Caregiving Process and Caregiver Burden Model (Klassen et al., 2007) have been widely applied to pediatric cancer research due to how these frameworks integrate the many complex processes contributing to familial stress in this context. Despite each of these models offering strengths in understanding the dynamic influences that unfold within families who have a child diagnosed with cancer, they do not highlight financial hardship as a key component to stress nor clearly illustrate directional relationships among family-level processes. Research has established that financial stress can have deleterious effects on both family and child psychosocial outcomes (Conger & Conger, 2002); furthermore, family relationship processes have a complexity beyond unidirectional processes (Wadsworth et al., 2013).

Notwithstanding, other models are needed to better explicate the interplay between economic pressure and family-level dynamics that occur in the context of pediatric cancer. Indeed, studies in pediatric cancer confirm a link between economic pressure and poorer child health outcomes including research suggesting that families at or near the poverty line have increased mortality rates in children with cancer (Adam et al., 2008; Bona et al., 2014, 2016; Gupta et al., 2014). Hence, the Family Stress Model (FSM) may serve as a valuable framework to further our understanding regarding economic stress and its mediating and moderating interaction with parenting stress, family functioning and child developmental outcomes (Conger & Donnellan, 2007). The model emerged from a longitudinal study (Iowa Youth & Families Project) examining the psychosocial impact economic pressure had on rural Midwestern families after the crash of agricultural businesses in the region (Conger & Conger, 2002).This model demonstrates how economic pressure affects the quality of interparental relationships, which in turn impacts child outcomes. For example, a negative financial event, such as high medical bills associated with cancer treatment, increases economic pressure which contributes to parenting stress and marital conflict; this in turn may lead to harsh parenting practices which can exacerbate child behavior problems (Conger & Conger, 2002; Wadsworth et al., 2013). Indeed, while economic pressure is a key component of the FSM, it is the parent's response to this stressor that has an interaction effect with child and family adjustment. For example, when parents exhibit resilient (e.g., strengthened marital relationships; sensitive parenting) versus maladaptive (e.g., marital conflict, negative emotional reactiveness) behaviors in response to a difficult life event, the children are more likely to emotionally adjust and have favorable developmental outcomes (Conger & Conger,

2002; White et al., 2015). Thus, protective factors can buffer the negative effects of stressful events and promote family resilience. In pediatric cancer, protective factors may include social support (Jackson et al., 2009), maintaining family routines (Santos et al., 2015) and a family-centered approach to medical care (Klassen et al., 2011). Another important feature of the FSM is the application of bidirectional pathways to depict the dynamic processes that mediate and moderate family-level outcomes (Masarik & Conger, 2017). To illustrate, parenting stress associated with caring for a child with cancer has been linked to worsened health-related quality of life outcomes in children with cancer (Loiselle et al., 2016); conversely, the disease severity and treatment status of the child with cancer has a reciprocal impact on the parent's quality of life (Hutchinson et al., 2009; Salvador et al., 2015). Moreover, there are protective factors associated with resilience that can mitigate parent psychological stress and these factors can provide important information for prevention and intervention for impacted families. For instance, families who have strong social support or utilize constructive coping strategies may experience lower perceived stress which enhances family cohesion and improved child outcomes for children with cancer (Klassen et al., 2011; Monti et al., 2017). Accordingly, previous research using the FSM has shown how discrepancies in health and developmental outcomes are strongly connected to a family's socioeconomic status (Kwon & Wickrama, 2014; Scaramella et al., 2008). For example, Scaramella et al. (2008) found that families experiencing financial hardship were more likely to display lowered efficacy in parenting which contributed to more child behavior problems. Finally, a notable strength of the FSM is that it has been tested over time in multiple sociocultural contexts and stressful family events demonstrating the rigor this model has in social science research (Aytaç & Rankin, 2009; Barnett, 2008; Martin et al., 2019).

Despite the plethora of psychosocial pediatric cancer research, gaps remain in our understanding of how various factors and mechanisms directly or indirectly influence family resilience, adjustment, psychosocial risk, quality of life, and overall functioning (Klassen et al., 2011; Litzelman et al., 2013; Long & Marsland 2011; Vrijmoet-Wiersma et al., 2008). Therefore, the purpose of this systematic review is to (1) review the current pediatric cancer literature to identify and summarize variables associated with the impact of stress and family functioning on family relationships and how these factors interact as potential mediators and moderators; and (2) develop a conceptual framework grounded in the FSM to facilitate methodologies in future research to include economic pressure as a pivotal variable when examining family processes in the context of pediatric cancer.

Method

The purpose of this review was to synthesize findings from both quantitative and qualitative studies and theoretically integrate them into a conceptual framework based upon the FSM to guide future research. As such, we did not perform a quality assessment of the studies nor a meta-analysis. To maintain theoretical validity in our aim of theory-building research, our review necessitated inclusion of diverse methodologies to inform the phenomenon of study.

We followed the PRISMA methodology for reporting systematic reviews (Liberati et al., 2009) as illustrated in Fig. 1. We searched both the psychology and health science literature (i.e., PubMed, PsychInfo, Web of Science, and CINHAL) within a ten-year time frame, 2008–2018. To provide feasibility in analyzing the large body of literature in the field, we narrowed the scope of our review to a ten-year period which we deemed an acceptable period to inform our research aim. We applied the following search terms to identify relevant articles pertaining to family-level outcomes in pediatric cancer: (Pediatric OR children) AND (cancer OR oncology) AND (family OR caregivers) AND (stress OR functioning). We defined family-level outcomes according to Patterson (2002) in which "a minimum of two family members must be involved…[and] the outcome must measure the product of the family relationship" (p. 234). As such, articles needed to measure either uni- or bidirectional familial relationships (e.g., parent and child) or measure a component of family functioning. Key variables of interest included pediatric cancer, family functioning, and stress. We included studies that utilized a measure specific to family functioning (e.g., Family Environment Scale) or any component of family functioning (e.g., cohesion, adaptability). We operationalized stress to comprise measures addressing parenting stress (e.g., Parenting Stress Index), caregiver burden or stress (e.g., Caregiver Burden Scale-R), or emotional distress (e.g., Parent Psychosocial Distress in Cancer). In addition, because post-traumatic stress disorder has a time duration criterion for onset, we included studies that assessed post-traumatic symptoms as an indicator of stress but excluded studies specifically examining post-traumatic stress disorder. Inclusion criteria consisted of peer-reviewed articles, systematic reviews, quantitative and qualitative studies. Studies contained in our review focused on families of children with cancer diagnosed 18-years or younger, children who were currently in treatment or within one-year of completing treatment, and measured a family process or a component of stress as it related to directional family influences (e.g., caregiver burden on child adjustment). We excluded publications that included other serious health conditions (e.g., cystic fibrosis), participants diagnosed over the age of 18, children off-treatment for more

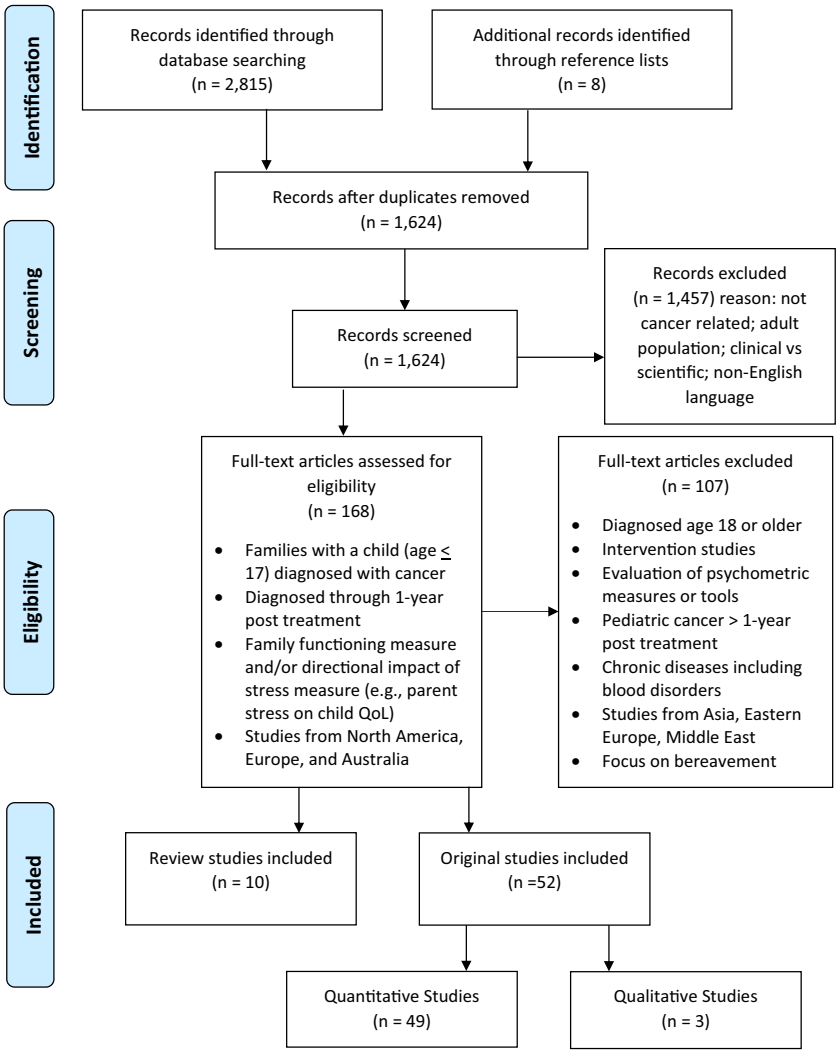

Fig. 1 PRISMA flow diagram demonstrating phases of the systematic review

than 1-year, or studies that did not examine directional family-level processes.

Data extraction elicited a total of 2815 publications. Figure 1 displays detailed explanation of the record extraction process. The first author completed extraction and evaluation procedures and using an iterative method, the second author reevaluated these procedures. To strengthen inter-judge reliability, a third reviewer reexamined a random sample for exclusion and inclusion accuracy based upon the established rating criteria. Inter-rater agreement was 82%. Final extraction and intra-judge discrepancies were confirmed through discussions until consensus was obtained between co-authors.

Detailed coding for each record included the following information: author, year published, study design, sample, age of participants, methodology, theoretical framework, independent and dependent variables, analytical strategy, measures, and findings. In synthesizing the multiple factors impacting family-level outcomes and directional influences, we extracted all independent and dependent variables used in the studies (e.g., child behavior, caregiver burden, family cohesion) and clustered them into categories using a qualitative approach to determine thematic categories. Variables were extracted from each study, written on index cards, and placed into groupings that were then classified into broader categories according to relatedness using an iterative method. For example, family risk and family cohesion both related to the broader concept of family functioning. Discussions occurred between authors to discern decisions regarding into which categories to place variables and identify categories salient to the purposes of this review. Frequency of variables within categories informed relevancy to the findings in this review. For example, two studies (Klassen et al., 2011; Rosenberg et al., 2014) measured variables related to medical care service delivery (e.g., family-centered care provision) which was determined insufficient to substantiate inclusion in our summary despite the potential influence these factors may have on family adjustment. This process elicited a total of five thematic categories with four related to family-level

processes (i.e., family-level factors, parent/caregiver influences, child with cancer illness & disease factors, and immediate and extended family relationships) and one related to socioeconomic considerations.

Results

Synthesis of Review Studies

In our search, ten review articles addressed pediatric cancer and family-level outcomes. Table 1 summarizes methods and findings of these reviews. In particular, three articles specifically addressed the impact of pediatric cancer on family functioning (Long & Marsland, 2011; Van Schoors et al., 2015, 2017), two investigated parenting stress or caregiver burden (Sultan et al., 2016; Vrijmoet-Wiersma et al., 2008), three summarized findings related to sibling adjustment (Alderfer et al., 2010; Long et al., 2018; Zegaczewski et al., 2016), one focused on quality of life for the child with cancer (Klassen et al., 2011), and one provided a qualitative review on family members experiences in the context of pediatric cancer (Mu et al., 2015). In general, these reviews offered insight into the many factors that potentially improve or worsen family-level outcomes including parenting stress, caregiver strain, family support, and family cohesiveness. Disease and illness-related factors such as time since diagnosis, treatment intensity, and type of cancer were also shown to have an influencing effect on adjustment, functioning, quality of life, and coping (e.g, Klassen et al., 2011). Family structure, demographic characteristics, and cultural processes were less examined and revealed inconsistent findings on family outcomes (Sultan et al., 2016; Vrijmoet-Wiersma et al., 2008).

Overall, these review papers shared universal interpretations in identifying limitations in the literature specific to pediatric cancer and family-level outcomes. Further, the methodologies reported commonly lacked scientific rigor due to non-randomized samples, cross-sectional designs, lack of comparison groups, and timing of measurements (e.g., Sultan et al., 2016; Van Schoors et al., 2015). For instance, sample sizes were relatively small and often heterogeneous in combining cancer types, time duration since diagnosis, and treatment intensity into the same groups making it difficult to effectively interpret findings. Further, these reviews identified extensive variation in the psychometric measures assessing family functioning and stress, noting infrequencies in adopting clear operational definitions (Vrijmoet-Wiersma et al., 2008). For example, family functioning was often defined broadly without distinguishing which components, (e.g., cohesion, adaptability) specifically reflected the construct (Van Schoors et al., 2015). Half of the reviews did not address nor identify mediating or moderating variables (e.g., Van Schoors et al., 2015; Zegaczewski et al., 2016). Understanding which specific mediators or moderators impact family-level outcomes in pediatric cancer is essential to developing best-practices and policies to decrease risk and promote resilience (Long et al., 2018). Another substantial concern was the widespread lack of a theoretical framework to guide research on this topic. Van Schoors et al. (2015) found approximately 84% of the studies included in their review did not use a theoretical model to guide their methodology. We identified three reviews omitting any theoretical foundation to their synthesized approach or analytical summary. Studies ungrounded in theory pose both methodological weaknesses and limitations to the meaningfulness of the findings.

Salient to our focus, we found half of the studies (N = 5) specifically identified economic burden or financial strain as risk factors to family-level outcomes in pediatric cancer. For instance, financial concerns contributed to parenting stress and depressive symptoms (Vrijmoet-Wiersma et al., 2008) as well as disruptions to family functioning (Long & Marsland, 2011; Mu et al., 2015) and sibling adjustment (Long et al., 2018). On the other hand, lower socioeconomic status did not appear to impact quality of life outcomes for the child with cancer (Klassen, et al., 2011). Rising medical bills and potential job loss are a fundamental reality for many parents who have a child with cancer, which can add to their psychological distress and pose challenges to overall family adjustment (Bona et al., 2016; Long & Marsland, 2011).

Synthesis of Empirical Studies

Table 2 displays a detailed overview of the 52 empirical studies specifying research design, sample, analytical approach, measures, theoretical framework, and outcomes for each study. As indicated in Table 2, these studies employed a range of analytical approaches, including various types of regression analyses, to determine direct effects, mediation, and moderation. Less than one-fifth of the studies utilized more sophisticated analyses (e.g., multilevel modeling, path analysis) in their models. In synthesizing the theoretical underpinnings for the studies included in this review, nearly 60% were grounded by a theoretical framework. Socio-ecological and the transactional stress & coping models were the most prevalent theoretical frameworks followed by fifteen others (e.g., family systems, allostatic load). In summarizing the research designs, 54% ($n = 28$) were cross-sectional, 8% ($n = 4$) were cross-sectional with comparison groups, 33% ($n = 17$) were longitudinal, and 5% ($n = 3$) of studies used a qualitative methodology. Table 3 outlines the five thematic categories and their relationship to outcomes and predictive influences as indicated in our review. Two thematic categories were

Table 1 Summary of review studies

Author (Yr)	Method	Family-level outcome	Aim	Theoretical model(s)	Databases	Search terms	Synthesized findings	Conclusions/ recommendations
Alderfer et al. (2010)	Systematic review	Sibling adjustment	The psychosocial impact of childhood cancer on siblings	Post-traumatic stress framework; family systems; developmental social ecological	PsycINFO, MEDLINE/ PubMed, CINAHL	(Sibling* or sister or brother or twin or family), (cancer or malignancy or tumor or oncology*) and (pediatric or child*)	Five themes related to sibling psychosocial adjustment: –Psychological functioning –Family functioning –Social and school functioning –Somatic complaints –Resilience/growth	Siblings do not seem to demonstrate elevated psychosocial risk; a subset of siblings demonstrate PTSS, negative emotional reactions, and lower quality of life
Klassen et al. (2011)	Systematic review	Child quality of life	Identify quality of life factors to identify gaps in knowledge and areas for future research	Did not use an explicit model to guide research; compated findings to quality of life theoretical model for children with epilepsy	MEDLINE, CINHAL, EMBASE, PsycINFO, Cancerlit, Sociological abstracts	Quality of life, health-related quality of life, quality adjusted life years, health status, functional status, well-being, or patient-reported outcome	Summarized quality of life factors into five areas: –Cancer variables (type, relapse status) –Cancer treatment variables –Treatment-related symptoms –Child factors –Family factors (including SES)	Noted that most research related to child quality of life focused on illness and treatment variables with less attention to child and family variables
Long & Marsland (2011)	Systematic review	Family adjustment	Examined whether family functioning, marital quality, and parenting are associated with child adjustment	Risk and resistance model; developmental psychopathology; Kazak's social ecology; pediatric medical traumatic stress; circumplex model	PsycINFO, PubMED, CINAHL	(Pediatric or child*) and (neoplasm* or cancer) and (family or marital or marriage or parent*)	Quantitative studies: –Mixed findings on family functioning, marital quality, and parenting stress Qualitative studies: –Family-level stressors, including income stress, –Parent perceptions of changes in marital relationship	Widespread variability in how families adjust within the context of pediatric cancer; recommend future research to use transactional frameworks
Long et al. (2018)	Systematic review	Sibling adjustment	Understanding of sibling adjustment to pediatric cancer including risk/ resiliency factors	Not specified	Pubmed, CINAHL, PsycINFO	Multiple search terms for the categories: siblings, cancer, and psychosocial functioning	–Over 50% of siblings report problems with family functioning –Poor family functioning associated with greater sibling distress –Family risk factors were most impactful to sibling distress	Recommend interventions to foster family communication & sibling support; research to identify mechanisms & moderators of sibling adjustment
Mu et al. (2015)	Qualitative systematic review	Family members' experience with childhood cancer	Understand family members' experiences over the first-year post diagnosis of childhood cancer	Not specified	CINAHL, PUBME, ProQuest Dissertations and Theses, and Chinese electronic periodical services	Keyword by phenomena of interest, type of participant, and type of study	–Family loss and turmoil surrounding diagnosis of cancer –Sense of courage and hope for mutual responsibility –Family support promoted resilience –Health team communication promoted understanding of illness –Theme of positive attitude and planning for the future	Exist limited information on siblings' experiences
Sultan et al. (2016)	Systematic review	Parent distress	Identify factors and outcomes on parent distress in families of pediatric cancer	Not specified	PubMed, CINHAL, PsycINFO	MeSH terms for parents, MeSH terms for distress and MeSH terms for cancer	–Distress-related illness/ treatment, demographic and psychological factors, coping style, parent view of child adjustment, stressful events, and family functioning	–Examine modifiable factors –Increase range of research designs –Expand attention of distress to family outcomes

Table 1 (continued)

Author (Yr)	Method	Family-level outcome	Aim	Theoretical model(s)	Databases	Search terms	Synthesized findings	Conclusions/ recommendations
Van Schoors et al. (2015)	Systematic review	Family resilience	Determine if evidence exists on family resilience after diagnosis of pediatric cancer	Family resilience framework	Web of Science, PubMed, PsycINFO, Cochrane, Embasse		Summarized six aspects of family functioning related to resilience: –Cohesion –Family Support –Conflict –Communication –Adaptability –Overall family functioning	Future research: –Adopt family resilience model –Involve whole family over time –Mixed-methods designs –Homogenous samples –Identify mechanisms of resilience
Van Schoors et al. (2017)	Meta-analysis	Association between family functioning and child adjustment	Provide analysis, summary, and commentary on evidence regarding association between family functioning and child adjustment to pediatric cancer	Social ecology; Double ABCX model; disability stress coping model; FAAR; circumplex model, adolescence resilience model; process model of stress and coping; family resilience process model	Web of Science, Pubmed, PsycInfo, Cochrane, Embase	(Cancer OR tumor OR malignancy OR oncolog*) AND (child* OR pediatric) AND (family OR parental), AND (psycholog* OR adaptation OR adjustment)	–Family functioning strongly associated with child adjustment –Greater cohesion associated with child adjustment –Greater family expressiveness –Greater family support associated with better child adjustment –Conflict associated with poorer child adjustment	–Need for theory to underlie research –Designs that integrate curvilinear models of analysis –Conceptualization of measurement of constructs
Vrijmoet-Wiersma et al. (2008)	Review	Parent distress	Describe prevalence and nature of parental strain according to disease factors, gender, and risk/protective factors	Lazarus stress and coping theory	PubMed, PsychINFOCINAHL	Parent, mother, father, stress reaction, psychological stress, adaptation, coping strategy, neoplasm/ psychology, and pediatric cancer	–Salient themes of parent distress included uncertainty, anxiety, depressive symptoms, and PTSS –Risk factors included lower education and SES, high caregiver demand, and premorbid problems –Protective factors included coping, social support (moderating effect), family functioning	–Family functioning operationalized as both a predictor and outcome variable –Conceptualization of parental stress and timing of measures varies considerably –Highest distress observed around time of diagnosis
Zegaczewski et al. (2016)	Integrative review	Sibling adjustment including family factors	Examine factors associated with healthy siblings' psychosocial adjustment	Pediatric psychosocial preventative health model	CINAHL, PubMed	Siblings, pediatrics, children, neoplasms, and psychosocial adaptation	Factors that correlated with sibling adjustment: –Sibling characteristics (e.g., age, gender) –Perceived social support (e.g., family) –Family tension increased distress –Perceived contextual factors (e.g., role overload, family adaptability)	Limitations with poor sample size: –Studies did not consider stage of cancer or treatment intensity of ill child

Table 2 Summary of original studies

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Barakat et al. (2010)	Cross-sectional	Adolescents with cancer and parent ($n = 102$)	Family Adjustment and Adaptation Response Model	Family Assessment Device	Regression	Child quality of life	Family functioning and parent child relationship quality predicted QOL; SES was not a predictor for QOL
Bemis et al. (2015)	Cross-sectional	Mothers ($n = 318$) and their children with cancer ($n = 151$)	Socio-Ecological	Perceived Stress Scale; Responses to Stress Questionnaire-Pediatric Cancer Version	Regression	Mothers' and children's psychological distress	SES was positively correlated with psychological distress, but effect was no longer significant when controlling for levels of stress
Bennett et al. (2013)	Cross-sectional	Caregivers of children with cancer ($n = 37$)	Lazarus and Folkman Stress and Appraisal	Parenting Stress Index—SF	Multiple regression	Parenting stress	Locus of control and coping style contributed to parenting stress; child behavior problems and time since diagnosis were not significant predictors
Colletti et al. (2008)	Cross-sectional	Parents of children with cancer ($n = 62$)	Transactional Stress and Coping Model	Parenting Stress Index—SF	Hierarchical Regression	Child adjustment	Higher parenting stress associated with poorer child behavioral and emotional adjustment; parental overprotection did not appear to influence child adjustment
D'Urso et al. (2019)	Cross-sectional	Children with cancer ($n = 34$) and siblings ($n = 26$)	PTSD Framework	Family Assessment Device: Impact of Events Scale-R	Hierarchical linear regression	Child & sibling PTSS	Lack of association between child or sibling PTSS and parent report of family functioning
Erker et al. (2018)	Cross-sectional	Children with cancer ($n = 160$) and siblings ($n = 105$)	Not specified	PROMIS Pediatric Family Relations Measure	Mann–Whitney test	Family relationships	Worse family relationships for siblings of patients on-treatment; relationships worsened when siblings had depressive symptoms; demographic factors (gender and age) and diagnosis influenced relationships
Fedele et al. (2011)	Longitudinal	Mothers of children with cancer ($n = 22$)	Not specified	Parenting Stress Index—SF	ANCOVA	Child adjustment	Parenting stress, overprotection, and perceived child vulnerability at T1 was associated with child adjustment at T2
Fladeboe et al. (2018a, b)	Longitudinal	Families of children with cancer ($n = 117$)	Engfer's Spillover Framework	Parenting Questionnaire (conflict scale)	Cross-lagged models	Parent–child conflict	Poor marital adjustment at diagnosis significantly linked to increased parent–child conflict at 6-month post diagnosis but not at 12 months

Table 2 (continued)

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Fladeboe et al. (2018a, b)	Longitudinal	Families of children with cancer ($n = 103$)	Family Systems	Negative Life Events Scale for Children; Treatment-Related Events Questionnaire	Multi-level modeling	Sibling conflict	All measures of stress (financial, general, cancer-related) were significantly associated with increased sibling conflict at 12 months since treatment
Gage-Bouchard et al. (2013)	Cross-sectional	Parents of children with cancer ($n = 60$)	Thompson's Transactional Stress and Coping Model	Family Environment Scale	Linear mixed models	Caregiver coping; family cohesion and conflict	Income was not a predictor of caregiver coping style; education and gender were predictors for active coping strategies; SES not significant predictor on family functioning factors
Hamner et al. (2015)	Cross-sectional	Parents of children with cancer ($n = 43$)	Not specified	Everyday Stressors Index	Hierarchical linear regression	Pediatric QoL	Parent chronic stress contributed to worsened child quality of life outcomes
Hoven et al. (2008)	Cross-sectional comparison	Mothers ($n = 182$) and fathers ($n = 139$) of children with cancer ($n = 190$)	Not specified	Parental Psychosocial Distress in Cancer	ANOVA	Parent distress	Parents of children with complicated cancer showed higher distress compared to parents of children with less complicated cancer
Hutchinson et al. (2009)	Cross-sectional comparison	Caregivers of children w/ brain tumors on ($n = 47$) and off tx ($n = 43$)	Not specified	Brief Symptom Inventory; Impact of Event Scale; Parent Experience of Child Illness; Caregiver Strain Questionnaire	MANOVA	General and caregiver stress	Off-treatment caregivers reported significantly lower general distress; both groups reported similar caregiver strain
Jackson et al. (2009)	Longitudinal mixed methods	Parents of children w/ brain tumors ($n = 88$)	Resiliency Model of Family Stress, Adjustment, and Adaptation	FACES-II	Multivariate analysis	Family adjustment	No association between level of coping and social support; family adaptability and cohesion correlated with coping patterns at different time points
Jobe-Shields et al. (2009)	Cross-sectional	Children awaiting stem-cell transplant and their parents ($n = 146$)	Social-Ecological	Family Environment Scale	Multiple regression	Child distress	Parent symptoms of depression, family cohesion, and family expressiveness were significant predictors of child-reported distress as well as intensity of distress
Karlson et al. (2013)	Longitudinal	Caregivers of children with cancer ($n = 163$)	Not specified	Psychosocial Assessment Tool (PAT 2.0)	Multilevel modeling	Family psychosocial risk	Caregivers with lower education and more financial difficulties were found to be at significantly greater psychosocial risk

Table 2 (continued)

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Katz et al. (2018)	Longitudinal	Families of children with cancer ($n = 60$)	Family Systems	Parenting Questionnaire—Conflict Scale; Sibling Relationship Questionnaire—Conflict Scale	Multilevel modeling	Family relationship quality	Generally, families showed stability in relationship quality over time; within-family variability observed indicating couple distress more likely to occur early in treatment; parent–child and sibling conflict more likely to occur later in treatment
Kelada et al. (2019)	Cross-sectional	Grandparents of children with cancer ($n = 89$) and healthy comparison ($n = 133$)	Not specified	Family Assessment Device	Multilevel modeling	Family functioning	Grandparents in cancer group reported worse family functioning compared to healthy comparison; poor family functioning associated with time since diagnosis, grandparent caregiving role, and distance from child with cancer
Klassen et al. (2011)	Cross-sectional	Parents of children with cancer ($n = 411$)	Caregiving Stress Process Model	Family Assessment Device; Care of My Child with Cancer Questionnaire	Structural equation modeling	Parent health-related QOL	Care-team provision, caregiver strain, and self-perception predicted parent psychological health; caregiver strain and social support predicted parent physical health; higher income associated with reduced caregiver strain
Kunin-Batson et al. (2016)	Longitudinal	Parents of children with cancer ($n = 160$)	Not specified	Family Assessment Device	Multinomial logistic regression	Child psychosocial functioning	Poor family functioning was a significant predictor of poor child psychosocial functioning
Labrell et al. (2019)	Cross-sectional	Mother of children with malignant brain tumors ($n = 35$)	Not specified	Family Assessment Device; Pediatric Inventory for Parents; State-Trait Anxiety Index	Generalized linear models	Maternal distress	Maternal anxiety negatively correlated with family cohesion; maternal distress higher in mothers of younger children with brain tumors
Litzelman et al. (2013)	Cross-sectional comparison	Parents of children with cancer ($n = 71$) and without cancer ($n = 135$)	Social-Ecological Systems Theory	FACES-IV; Impact on the Family Scale	Multivariate analysis	Pediatric QOL	Lower income families who had a child with cancer associated with worse Pedi QOL compared to low income families without a child with cancer; the effect significantly weakened when family burden was added to the model

Table 2 (continued)

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Litzelman et al. (2011)	Cross-sectional	Parents of children with cancer ($n = 80$)	Not specified	Perceived Stress Scale	Regression	Parent QOL	Child's limitations and active treatment status associated with worse parent QOL but association was mediated by caregiver burden and stress
Loiselle et al. (2016)	Longitudinal	Caregivers of children with HSCT ($n = 90$)	Not specified	Psychosocial Assessment Tool (PAT 2.0)	Multilevel modeling	Child HRQOL	Increased caregiver stress at baseline time point predicted worsened HRQOL at 3–9 months; as a covariate, income not found to be a factor impacting HRQOL
Long et al. (2013)	Cross-sectional	Siblings ($n = 209$) and their parents	Developmental Psychopathology Framework	Family Assessment Device; Post-Traumatic Stress Diagnostic Scale	Multiple regression	Sibling adjustment	Greater sibling distress associated with problems with family functioning and parent psychological control
Marcoux et al. (2012)	Longitudinal	Parents of children with ALL ($n = 138$)	Not specified	Family Well-Being Assessment	Multilevel modeling	Child behavior	Familial stress a risk factor for child internalizing behavior problems at 3-month post diagnosis
Mitchell et al. (2016)	Longitudinal	Parents of children with ALL ($n = 160$)	Not specified	Family Assessment Device	Univariate longitudinal logistic regression	Child QOL	Poor family functioning a risk factor for emotional and social impairment; findings independent of income or maternal education
Molzon et al. (2018)	Cross-sectional	Caregivers of children with cancer ($n = 150$)	Transactional Stress and Coping Model; Kazak Social and Ecological Model	Brief Symptom Inventory	Path analysis	Child QOL	Caregiver distress, impacted by family income and perceived barriers to care, had a direct negative impact on child quality of life
Monti et al. (2017)	Longitudinal	Parents and their children with cancer ($n = 166$)	Kliewer Socialization Coping Model	Responses to Stress Questionnaire-Pediatric Cancer version	Hierarchical multiple regression	Child coping	Parents' coping style & depressive symptoms predicted child coping at T1; fathers' secondary coping predicted child secondary coping across time
Mullins et al. (2016)	Cross-sectional	Parents of children with cancer ($n = 138$)	Transactional Stress and Coping Model	Parent Stress Index—SF; Impact of Events Scale-R; Brief Symptom Inventory	Path analysis	Parent adjustment and Child QOL	Negative parent illness appraisals associated with poor parent adjustment linked to child QOL; indirect effect of parenting stress between parent illness appraisal and child QOL

Table 2 (continued)

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Myers et al. (2014)	Longitudinal	Primary caregiver of children with ALL ($n = 159$)	Not specified	Family Assessment Device	Logistic and multivariate regression	Child psychosocial adjustment	Poor family functioning increased child anxiety and depressive symptoms; income not found related to child outcomes
Okado et al. (2014)	Cross-sectional comparison	Children w cancer and caregivers ($n = 255$) and healthy comparisons ($n = 142$)	Allostatic Load	Brief Symptom Inventory; Impact of Events Scale-R	Hierarchical regression	Child distress	Parent distress linked with child distress in cancer group only; child exposure to life events moderated this relationship
Patino-Fernandez et al. (2008)	Cross-sectional	Mothers ($n = 129$) and fathers ($n = 72$) of children with cancer	Not specified	Family Environment Scale –Acute Stress Disorder Scale; STAI-Y	Multiple regression	Parent stress	General anxiety a strong predictor of acute stress in both mothers and fathers; family functioning was not a significant predictor of stress
Penn et al. (2009)	Longitudinal	Children with brain tumors ($n = 35$) and their parents	Risk-Resistance Theoretical Framework	Impact on Families Scale; Family Assessment Device; Family Support Scale	ANOVA and multiple regression	Child HRQOL	Tumor site and poor child HRQOL at 1-month post diagnosis was highest predictor for poor child and parent-report of child HRQL; family functioning modulated concurrent HRQL
Perricone et al. (2012)	Cross-sectional	Mother of children with ALL ($n = 34$)	Not specified	FACES-II	Spearman correlation	Family functioning	Orienting coping strategy positively correlated with cohesion and adaptability; spiritual coping negatively correlated with adaptability
Peterson et al. (2018)	Longitudinal - repeated measure	Parents ($n = 73$) of children diagnosed with craniopharyn-gioma	Not specified	Brief Symptom Inventory	Linear mixed models	Parent distress	Parent distress closely associated with parent perceptions of the child with cancer's level of cognitive impairment
Pierce et al. (2017)	Cross- sectional	Parents of children with cancer ($n = 67$)	Pediatric Preventative Psychosocial Health Model	Pediatric Assessment Tool; Distress Thermometer; Posttraumatic Stress Disorder Checklist	Hierarchical multiple regression	Child HRQOL	Family risk and caregiver stress significantly predicted poorer total child HRQL; Caregiver distress independently predicted physical HRQOL and family risk independently predicted psychosocial HRQOL

Table 2 (continued)

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Prchal & Landolt (2012)	Qualitative	Siblings of children with cancer ($n = 7$)	Not specified	Interview included sibling report of what happened in the family post diagnosis	Content analysis	Sibling adjustment	Siblings reported difficulties due to absence of parents and dealing with ill-sibling's suffering; siblings reported increased family cohesion
Roddenberry & Renk (2008)	Cross- sectional	Mothers ($n = 47$), fathers ($n = 16$) and children with cancer ($n = 19$)	Not specified	Parenting Stress Index—SF	Regression	Child QOL	Significant relationships between maternal distress and child externalizing behavior problems and paternal stress associated with child internalizing and externalizing behavior problems
Rodriguez et al. (2012)	Cross -sectional	Parents of children with cancer ($n = 106$)	Tend and Befriend Model of Stress	Perceived Stress Scale; Responses to Stress Questionnaire-Pediatric Cancer Version	ANOVA and multiple regression	Parenting stress and child stress	Mothers reported higher stress compared to fathers for younger children with cancer; caregiver stress highest compared to other stressors; income negatively associated with parental stress
Rosenberg et al. (2014)	Cross- sectional	Parents of children with cancer ($n = 96$)	Not specified	FACES	Logistic regression	Resilience	Parents of children with cancer with lower resilience resources were associated with higher distress and lower family functioning
Ryan et al. (2013)	Cross-sectional	Parents of children with cancer ($n = 73$)	Transactional Stress and Coping Model	Parenting Stress Index—SF	ANCOVA	Child adjustment	Parenting stress differentiated by income level but not social status; child behavior problems influenced by lower income
Salvador et al. (2015)	Cross- sectional	Parents of children with cancer ($n = 277$)	Social-Ecological Framework	Caregiver Burden Scale-R	MANOVA and mediation analysis	Parent illness perception and parent QOL	Parents' perceptions of illness negatively correlated with parents' QOL which was moderated by child treatment status
Santos et al. (2015)	Cross-sectional	Children with cancer ($n = 389$) and their parents	Social-Ecological and Transactional Models of Development	Family Environment Scale	Structural equation modeling	Child QOL	Higher involvement in family rituals associated with family cohesion and hope mediated higher QOL
Santos et al. (2017)	Longitudinal	Children with cancer ($n = 58$) and one parent	Attachment Theory	Family Environment Scale	Hierarchical Multiple Regression	Family cohesion	Parental avoidance attachment predicted lower family ritual meaning and family cohesion over time; ritual meaning mediated relationship

Table 2 (continued)

Author (Yr)	Design	Sample	Theoretical framework	Measures (stress or family functioning)	Statistical analysis	Outcome variable	Findings related to family-level outcomes
Santos et al. (2018)	Qualitative	Mothers of children with cancer ($n = 19$)	Social-Ecological	Semi-structured interviews focused on family rituals	Grounded Theory	Family rituals	Identified themes related to both loss and transformation as strategies to family adaptation to changes in family rituals in the context of pediatric cancer diagnosis
Van Der Geest et al. (2014)	Cross-sectional	Parents ($n = 123$) and their children ($n = 67$) with cancer	Not specified	Parenting Stress Index	Correlation and mediation analysis	Parent mood and child behavior	Parenting stress strongly correlated with increased child behavior problems; parenting stress mediated the effect between father's negative mood and child behavior problems
Van Schoors et al. (2018)	Qualitative	Couples with a child with leukemia ($n = 10$)	Social-Ecological; Double ABCX	1:1 interviews and Family System Test	Multi interview analysis	Family Functioning	Family functioning impacted by change specific to conflicting dynamics (simultaneous cohesion and fragmentation) in the context of pediatric cancer
Wesley et al. (2013)	Cross-sectional	Adolescents with cancer ($n = 102$)	Not specified	Family Assessment Device	Hierarchical multiple regression	Adolescent adjustment	Poor family functioning associated with higher perceived social support from friends and family
Willard et al. (2016)	Longitudinal	Parents of children with retinoblastoma ($n = 92$)	Not specified	Parenting Stress Index	ANOVA and linear models	Parenting stress and child functioning	Child-directed parenting stress had a small effect in predicting decline in child cognitive functioning over time
Wolfe-Christensen et al. (2010)	Cross- sectional	Mothers of children with cancer ($n = 36$)	Transactional Stress and Coping Model	Parenting Stress Index	Hierarchical regression	Child adjustment	Parenting stress a predictor of child adjustment and moderated relationship between caregiver demand and child internalizing problems
Zheng et al. (2018)	Longitudinal	Parents of children with ALL ($n = 594$)	Not specified	Family Assessment Device	Multivariate modeling	Child QOL	Poor family functioning predicted poorer child QOL

Table 3 Thematic categories and related outcomes and predictive influences

Thematic category	Influences outcomes on:	Influenced by:
Family functioning		
	–Child QoL –Child and parent adjustment –Child behavior –Child and parent distress –Sibling adjustment	–Marital relationship quality –Socioeconomic status –Number of children in home –Family coping
Parent/caregiver		
	–Child QoL –Child adjustment –Child behavior –Child coping –Parent mental health	–Locus of control –Parent coping style –Financial stress –Caregiver role strain –Child disease factors
Child with cancer:		
Psychosocial characteristics		–Parent stress –Family functioning
Disease factors	–Parent mental health –Parent distress –Caregiver burden –Parent QoL	
Immediate and extended family relationships:		
Sibling		–Caregiver education –Financial strain –Parental absence –Number of children in home –Treatment status –Time since diagnosis –Family functioning
Grandparents	–Family functioning	
Socioeconomic factors		
	–Child QoL –Child behavior –Sibling conflict –Child and parent distress –Parent stress –Caregiver burden –Family psychosocial risk	

divided into two subcategories with child factors including both psychosocial characteristics and disease factors and immediate and extended relationships including both siblings and grandparents. Descriptive findings of the five thematic categories are presented below and include the following: family-level factors, parent/caregiver influences, child with cancer: psychosocial and disease factors, immediate and extended family relationships, and socioeconomic considerations.

Family-level factors

Given our inclusion criteria, it is not surprising that most of our studies included a construct related to family functioning or other family-level variables. Studies varied in their operational definition of family functioning depending upon what components were measured. For example, Gage-Bouchard et al. (2013) measured family cohesion and conflict, Jackson et al. (2009) measured family adjustment, and Wesley et al. (2013) examined overall general functioning. Family functioning was most frequently the independent or predictor variable for various outcomes including child quality of life (Mitchell et al., 2016), child psychosocial adjustment (e.g., Jobe-Shields et al., 2009), parent quality of life (Santos, et al., 2015), parenting stress (e.g., Patino-Fernandez et al., 2008) and sibling adjustment (e.g., D'Urso et al., 2019). Family functioning was less commonly measured as an outcome variable (e.g., Santos et al., 2017).

Several studies examined the impact of family functioning on quality of life and psychosocial adjustment in children with cancer. Although one study (Penn et al., 2009) did not find any association between family functioning and quality of life outcomes, both cross-sectional (Barakat et al., 2010; Pierce et al., 2017) and longitudinal studies have linked poor family functioning to lower quality of life in children and adolescents with cancer (e.g., Zheng et al., 2018; Mitchell et al., 2016). Santos et al. (2015) suggested that family cohesion, in addition to hope, had a beneficial mediating effect between family ritual meaning and child quality of life outcomes. In addition, a qualitative study examining family rituals, Santos et al. (2018) identified themes related to loss and transformation as processes in how families adapt family rituals, suggesting that family rituals may serve to buffer the stressors related to separation and chaos experienced upon diagnosis. Family functioning has also been shown to have both direct and mediating risk and protective influences on child psychosocial adjustment, including risks to emotional adjustment (Kunin-Batson et al., 2016; Myers et al., 2014) and increased internalizing behaviors (Marcoux et al., 2012), with increased family cohesion and expressiveness buffering child distress (Jobe-Shields et al., 2009). Conversely, poor family functioning had a different effect on adolescents with cancer who reported increased social support and connection to peers indicating the importance peers have during adolescence (Wesley et al., 2013). Overall, the evidence indicates that family functioning including, cohesion, family risk, and family ritual have potential influences on the child with cancer's quality of life, although variance as to how and to

what degree family functioning impacts child adjustment may be dependent on the child's developmental status, sensitivity to familial stress, and level of family cohesion.

Three studies examined family functioning in relationship to parent quality of life and adjustment outcomes. Klassen et al. (2011) defined parental quality of life in terms of physical health and functioning and found that family functioning, including family coping and support, served as mediating factors in buffering the deleterious effects caregiver burden had on parent physical health for those parents. Poor family functioning was found to be a significant predictor to parenting distress (Labrell et al., 2019), although Patino-Fernandez et al. (2008) did not find a significant relation linking family functioning to caregiver acute stress. More research in this area is necessary to confirm family functioning's influence on parent quality of life and distress and its potential mediating role in caregiver burden.

Eight studies included in our review measured family functioning as an outcome variable. As indicated in Table 2, longitudinal studies link increased family risk to more children in the home (Karlson et al., 2013), couple distress and family conflict (Katz et al., 2018), and poor marital relationship quality (Fladeboe et al., 2018a). Financial strain, lower education (Karlson et al., 2013), and socioeconomic status (Gage-Bouchard et al., 2013) also negatively impacted family functioning outcomes such as cohesion. Using semi-structured interviews, Van Schoors et al. (2018) identified concurrent themes of increased family cohesion and fragmentation of the family unit as conflicting processes impacting family adaptation to the crisis of pediatric cancer. Two studies indicated that constructive coping strategies served as mitigating factors in strengthening family adaptability and cohesion (Jackson et al., 2009; Perricone et al., 2012). In examining predictors to family functioning, Santos et al. (2017) found that avoidant parent romantic attachment was associated with lower family ritual meaning and cohesion. Upon further analysis, lowered family ritual meaning seemed to function as a mediating mechanism between higher avoidant attachment at time one and lowered cohesion at time two. Overall, research indicates that family functioning may be influenced by level of education, financial strain, family coping styles, parental romantic attachment, and adaptive processes, but more evidence is needed to validate these findings.

Of these studies, only five highlighted the impact financial stress has on the family system with each offering a different perspective on its influence. For instance, Van Schoors et al. (2018) briefly mentioned families adjusting employment to manage child caregiving needs while Klassen et al. (2011) considered financial strain as an additive factor to the concept of caregiver strain. Additionally, Karlson et al. (2013) identified financial burden to be a key variable contributing to many family risk outcomes (e.g., beliefs on ability of family to cope), whereas Gage-Bouchard et al. (2013) found educational attainment more predictive to family coping compared to financial resources. Only one study acknowledged a potential protective factor (i.e., family ritual meaning) to the impact of socioeconomic risk on families coping with pediatric cancer (Santos et al., 2015). In summary, limited research exists linking the impact of financial stress on family functioning in the context of pediatric cancer, and of those studies (e.g., Karlson et al., 2013), there appears to be variability in how this factor is integrated into existing research methodologies.

Parent/caregiver influences

We identified five distinct subcategories evaluating parent/caregiver factors: stress/distress (including caregiver burden), psychological factors (anxiety, depression, mood), coping, quality of life, and parenting style. Significant predictors contributing to parenting stress/distress included loss of control and an accepting responsibility (self-blame) coping style (Bennett et al., 2013), financial factors (e.g., Bemis et al., 2015), and caregiver strain pertaining to role functions (Rodriguez et al., 2012). The degree of parenting stress was also compared between groups of families with cancer to identify potential modifying disease factors such as treatment status (Hutchinson et al., 2009), complexity of disease (Hoven et al., 2008), and cancer-related cognitive impairments (Peterson et al., 2018). Together, these findings suggest that multiple stressors contribute to parenting stress including socioeconomic status, coping response, and disease factors.

Parenting stress/distress appeared as an independent variable in more than a quarter of the articles reviewed and often functioned as a mediator or moderator related to child or parent outcomes. Parenting stress was a direct predictor in negatively influencing child quality of life outcomes including physical functioning (Willard et al., 2016), overall psychosocial functioning (e.g., Hamner et al., 2015), child behavioral and social adjustment (Colletti et al., 2008), and child quality of life (e.g., Loiselle et al., 2016). Mediation effects of parenting stress linked fathers' negative mood to child behavior problems (Van Der Geest et al., 2014) as well as the relation between parent negative illness appraisals and worsened child quality of life (Mullins et al., 2016). Caregiver burden, a construct different yet related to parenting stress, mediated the relation between child treatment status and parent mental health, worsening parent outcomes (Litzelman et al., 2011). In comparing families with and without cancer, parenting stress was associated with child distress only in the cancer group which was found to be

significantly moderated by stressful life events such as poverty and family violence (Okado et al., 2014). Parenting stress has also moderated child outcomes, with fewer child behavior problems observed under conditions of less parenting stress (Wolfe-Christensen et al., 2010). Seemingly, parenting stress plays a direct role on the ill-child's psychosocial and quality of life outcomes while also potentially mediating parent mental health. Nevertheless, the impact that parenting stress has on siblings' well-being and overall family functioning has yet to be understood.

Some empirical studies measured the impact of other parent-related influences on child relationships: parent psychological states (anxiety, depression), coping style, and parental overprotection and control. Parental anxiety and depression have both been linked to lower pediatric quality of life (Penn et al., 2009; Roddenberry & Renk, 2008), child-reported distress (Jobe-Shields et al., 2009) and secondary coping in children with cancer, a constructive coping style conducive to adaptability and adjustment (Monti et al., 2017). Further, parents who were less likely to utilize constructive coping strategies were significantly more likely to report increased anxiety and depression symptoms in their children with cancer (Kunin-Batson et al., 2016; Myers et al., 2014). While parental overprotection and control may occur immediately following a child's cancer diagnosis, this parenting behavior seems to lessen over time (Fedele et al., 2011) and does not seem to negatively impact child adjustment (Colletti et al., 2008). Research examining interaction effects among parental response to pediatric cancer (i.e., parent emotional state, coping strategy, and parenting style) and their influences on family and child outcomes may further inform effective interventions to help parents constructively adapt to this stressful event.

Despite the many studies examining parent factors on family-level processes, few addressed the role financial stress can have on family outcomes. Some studies used measures that included financial stress as a component of caregiver burden (e.g., Hutchinson et al., 2009; Rodriguez et al., 2012; Wolfe-Christensen et al., 2010), which made it challenging to discern the unique contribution economic pressure may have had on parent and child outcomes. To illustrate, both Loiselle et al. (2016) and Bemis et al. (2015) found financial burden to function as an additive stressor for parents of children with cancer which worsened parent and child psychosocial functioning. Thus, financial stress, specific to having a child diagnosed with cancer, seems to intensify the physical and emotional toll for these parents.

Child with cancer: psychosocial adjustment and disease factors

We consolidated variables pertaining to the ill-child into one of two subcategories: child functioning/adjustment and illness/disease factors. Child functioning and adjustment was commonly measured using psychometric instruments quantifying child internalizing and externalizing behaviors such as the Child Behavior Checklist (e.g. Marcoux et al., 2012). Other indicators of child functioning included assessing child depression, anxiety, social skills, emotional distress or posttraumatic stress. Further, most studies measured child functioning/adjustment as an outcome variable (e.g., Okado et al., 2014). Illness and disease variables encompassed a range of factors including time since diagnosis, illness severity, intensity of treatment, cancer type, prognosis, relapse status, age at diagnosis, treatment protocol, treatment status (on or off treatment), and child's health status (e.g., physical symptoms). While many studies included these factors as covariates (e.g., Van Der Geest et al., 2014), we synthesized findings only from those studies where disease influences were key variables.

In our search, parent stress and family functioning often influenced child emotional and behavioral factors. For example, increased parenting stress was associated with increased child psychosocial behavioral maladjustment (Colletti et al., 2008) and deterioration over time in child cognitive and adaptive functioning (Willard et al., 2016). Research also indicates that parenting stress moderated the link between caregiver demand and child internalizing behavior (Wolfe-Christensen et al., 2010) or functioned as a mediator between father's negative mood and child behavior problems (Van Der Geest et al., 2014). In a cross-sectional comparison design, parenting stress predicted child distress but only in the cancer group, and the effect was moderated by children's exposure to cumulative stressful events (Okado et al., 2014). Longitudinal studies have shown familial stress and poor family functioning as predicting risk for child behavior problems (Marcoux et al., 2012) as well as increased child depression and anxiety during the first year of cancer (Kunin-Batson et al., 2016; Myers et al., 2014). One study found several variables (i.e., parent depression, family cohesion, and family expressiveness) to predict intensity of child distress (Jobe-Shields et al., 2009). Clearly, both parenting stress and poor family functioning have been identified as significant factors in predicting overall functioning of children with cancer, although gaps exist in knowing to what extent child functioning influences parenting stress and family adjustment.

Child characteristics specific to the disease process itself have been included as factors relevant to outcomes on parents, family unit, and the child with cancer. In stratifying groups according to time since diagnosis, Klassen et al. (2011) identified a higher number of risk factors to parent health for those with a child in the first year since diagnosis. However, time since diagnosis was not associated with parenting stress (Bennett et al., 2013) or distress in mothers

and their children with cancer (Bemis et al., 2015). Hutchinson et al. (2009) observed that caregivers of children with cancer off treatment showed lower distress compared to those caregivers whose children were on treatment. Active treatment status also moderated higher levels of caregiver burden and worsened parent quality of life (Salvador et al., 2015). Conversely, Litzelman et al. (2011) found this association no longer significant in their study when adding parenting stress to the model. Treatment status and time since diagnosis seem to have direct and moderating effects on caregiver functioning although risks of additional disease factors on family-level outcomes warrant further investigation.

Like previous categories in this review, few studies related child or disease variables to financial stress as influencing or mediating factors. For instance, one study examining psychosocial adjustment of the child with cancer considered financial stress as a potential component to child outcomes (Wolfe-Christensen et al., 2010); however, the study did not include financial stress as a stand-alone variable but instead, measured it as an element of caregiver burden. Likewise, studies exploring the impact of child characteristics specific to the disease process on familial caregivers referenced financial stress only in relation to caregiver strain or burden (e.g., Hutchinson et al., 2009; Klassen et al., 2011; Salvador et al., 2015). Thus, evidence on the role financial stress has in relation to child adjustment or child disease characteristics is limited to its interconnection with caregiver burden.

Impact of cancer on immediate and extended family relationships

The results from our review found several studies that examined interactional processes involving siblings of children with cancer and one study that explored the impact of pediatric cancer on grandparents. While family functioning did not seem to impact post-traumatic stress symptoms for siblings (D'Urso et al., 2019), poor family functioning contributed to greater sibling distress with additional non-specific risk factors further strengthening that relationship (Long et al., 2013). Likewise, siblings in families with more children living in the home, less parent education, and greater financial pressure were at increased risk for problems as measured by the Psychosocial Assessment Tool 2.0 (Karlson et al., 2013). Recently, studies linked sibling conflict to patient treatment status and sibling symptoms of depression (Erker et al., 2018), stress including financial, general, and cancer-related (Fladeboe et al., 2018b), and time since diagnosis (Katz et al., 2018). Using a qualitative methodology, Prchal & Landolt (2012) conducted semi-structured interviews with siblings of children with cancer and found that all siblings expressed having some degree of distress associated with parental absence due to the ill child's frequent hospitalizations. Interestingly, six out of the seven siblings interviewed in this study perceived their families as having more cohesion, which mirrors the findings stated previously by Van Schoors et al. (2018). In examining family-level outcomes from the perspective of grandparents, Kelada et al. (2019) found time since diagnosis, grandparents in caregiver roles, and distance from family worsened overall family functioning. The paucity of existing research exploring pediatric cancer's impact on extended family relationships leaves much to be learned in how these relationships interrelate and mutually influence parents, the child with cancer, and the family unit.

Despite brief mention of the impact financial stress (e.g., Katz et al., 2018) or lower socioeconomic status (e.g., Erker et al., 2018) may have on family relationships, only two studies specifically highlighted the influence of financial stress on immediate or extended family relationships. Of those studies, more financial stress in the family seemed to contribute to increased sibling conflict during the first year of treatment (Fladeboe et al., 2018b) as well as problems with sibling psychosocial adjustment (Karlson et al., 2013). As such, financial stress seems to foster potential challenges for siblings of children with cancer (e.g., Karlson et al., 2013) but has not been examined on other relationships beyond the immediate family.

Socioeconomic Considerations in Families with a Pediatric Cancer Diagnosis

Of the studies that included socioeconomic factors as stand-alone variables, there were conflicting results. That is, some studies indicated socioeconomic status as contributing to poorer outcomes, whereas other studies did not show any significant association. As indicated in Table 2, families at economic risk showed significantly greater child behavior problems (Ryan et al., 2013), sibling conflict (Fladeboe et al., 2018b), as well as increased caregiver-related stress (Molzon et al., 2018) and caregiver strain (Klassen et al., 2011). Karlson et al. (2013) identified that both lower education and financial strain contributed to increased family psychosocial risk. Litzelman et al. (2011) noticed that families with lower income showed worse child quality of life outcomes but the effect was accounted for by family burden. Bemis et al. (2015) also found that while both independent and cumulative factors of socioeconomic status predicted maternal and child distress, this effect was significantly attenuated when accounting for cancer-related stress. That is, cancer-related stress and burden may serve as key processes mediating the link between socioeconomic status and poor child outcomes. In sum, not all studies supported the premise

that families with less relative income are at greater psychosocial risk. For example, both Barakat et al. (2010) and Penn et al. (2009) did not find socioeconomic status as a predictor to child health-related quality of life. Income was also not significantly associated with child psychosocial adjustment (Myers et al., 2014) or caregiver coping style and family functioning (Gage-Bouchard et al., 2013). It is apparent across these studies that a specific theoretical family framework conducive to including economic pressure and financial strain may help researchers more effectively examine the socioeconomic disparities that can occur in families experiencing pediatric cancer.

Discussion

The evidence in this review strongly supports the intersecting role family functioning has on mitigating or worsening psychosocial outcomes for children with cancer and their parents. For example, families with strong cohesion demonstrate less distress in children and siblings with cancer (Jobe-Shields et al., 2009; Prchal & Landolt, 2012) whereas poor family functioning negatively impacts child quality of life (Mitchell et al., 2016; Zheng et al., 2018). Additionally, strong associations were found linking parent psychological functioning, such as stress and emotional states, to child outcomes, including quality of life (e.g., Loiselle et al., 2016; Molzon et al., 2018) and adjustment (e.g., Colletti et al., 2008; Wolfe-Christensen et al., 2010). Moreover, it is noteworthy that few studies in our review included both parenting stress and family functioning as key variables examining child outcomes since they both appear to have strong influences on child functioning and behavior (e.g., Barakat et al., 2010; Patino-Fernandez et al., 2008). While these findings confirm results from previous reviews (e.g., Sultan et al., 2016; Van Schoors et al., 2017), our synthesis illuminated specific gaps inhibiting our understanding of family-level processes in pediatric cancer. For instance, all studies identified through this review restricted investigation to unidirectional relationship processes (e.g., parenting stress influencing child behavior) rather than simultaneous examination of potential bidirectional associations (e.g., child behavior influencing parenting stress). Likewise, child psychosocial functioning was measured only as an outcome variable excluding the possibility that child functioning may also influence parent, sibling, or family processes (e.g., Kunin-Batson et al., 2016; Ryan et al., 2013). Thus, future studies should explore the possible multi-directional influences among parents, children with cancer, siblings, and overall family functioning that may provide critical insights into the complex mechanisms underlying family dynamics as they unfold in the context of pediatric cancer (Long & Marsland, 2011; Pettit & Arsiwalla, 2008). Specifically, Pettit & Arsiwalla (2008) suggest utilizing mixed-method research designs as well as multi-informants as means to improve studying these dynamic reciprocal relationships. Further, longitudinal designs that follow families throughout the pediatric cancer experience will inform how families change and adapt over time thus providing guidance on where interventions would be most needed (Vrijmoet-Wiersma et al., 2008; Zheng et al., 2018). In sum, current studies in pediatric cancer have largely been limited to examining unidirectional processes and future studies are needed to identify what bidirectional influences exist and how these reciprocal influences function both within and across time (Long & Marsland, 2011; Pettit & Arsiwalla, 2008).

Equally important to our findings, our review indicated rarely did studies include financial stress or economic pressure as a stand-alone variable or considered the impact financial stress may have on family relationship processes (e.g., Bemis et al., 2015; Karlson et al., 2013). Moreover, of these studies, a handful combined financial stress within the construct of caregiver burden (e.g. Klassen et al., 2011; Rodriguez et al., 2012) which limits our understanding about the ways this specific type of stress impacts family relationships and family functioning when compounded with other stressors. For example, the stress of not having enough money to pay medical bills is qualitatively different than the stress associated with the emotional and physical toll in caring for a child actively undergoing chemotherapy. While we acknowledge that economic strain is not the only factor contributing to parent distress, studies that can ascertain its role in family-level processes in the broader context of pediatric cancer are necessary to inform interventions and policies to minimize family risk and health disparities (Adam et al., 2008; Tsimicalis et al., 2013; Warner et al., 2015). Accordingly, theoretical frameworks that are conducive to using sophisticated statistical modeling across time can demonstrate how economic strain directly and indirectly influences the family system (Barnett, 2008; MacCallum & Austin, 2000). Thus, our findings support the need to employ a conceptual model that imparts both economic burden and family functioning as core constructs, while offering a visual framework outlining directional interactional influences within the family.

Explicating directional influences is warranted in the following areas: (1) the need to more clearly define family functioning descriptions across studies; (2) the need for consistent theory-driven research; (3) the need for identifying key mediating and moderating factors; and (4) the importance of confirming results through robust methodological designs and analytic approaches. First, family functioning is a broad construct with many elements (e.g., cohesion, adaptability) and studies from our review

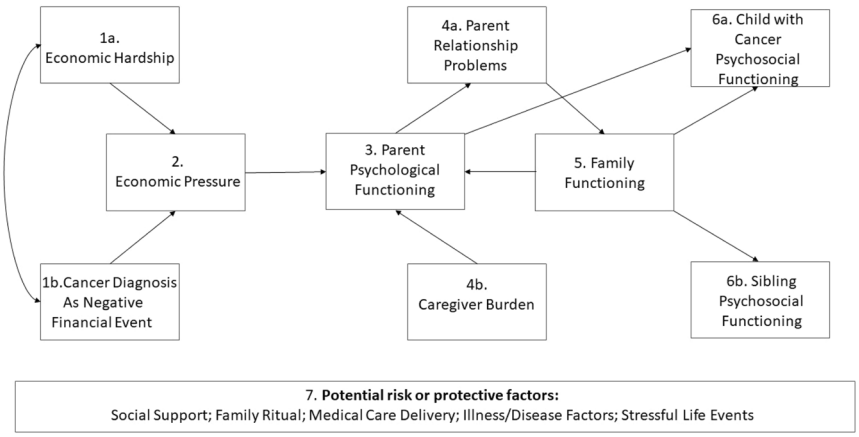

Fig. 2 A conceptual model of the Family Stress Model in the context of pediatric cancer. This model illustrates the directional processes impacting families who have a child with cancer and the potential risk and protective factors that may mediate or moderate those processes. The model is an adaptation of the Family Stress Model to guide future research and explicate a family's experience with pediatric cancer

demonstrated wide variation in both operationalization and instrumentation making it challenging to discern theoretical validation. For example, in examining family functioning on child quality of life outcomes, Mitchell et al. (2016) used the Family Assessment Device whereas Santos et al. (2015) used the Family Environment Scale. Hence, future studies may consider creating a latent construct for family functioning using several indicators to reflect the complexity of this construct while also minimizing measurement error (MacCallum & Austin, 2000). Next, we noticed that approximately 60% of the studies in our review were guided by a specified theoretical framework. Although this percentage is higher than previous findings (e.g.,Van Schoors et al., 2015) and may indicate a promising trend, the concern still remains that research ungrounded in theory continues to proceed. Third, our findings were inconclusive in substantiating moderating effects or mediating relationships between variables due to few studies measuring these effects as well as use of poor study designs to measure mediation processes (e.g., Mullins et al., 2016). To illustrate, Van Der Geest et al. (2014) identified parenting stress to mediate the relationship between parent mood and child behavior problems; however, the study used a cross-sectional design thus obstructing the ability to fully determine temporal ordering of pathways which is needed for true mediation (Maxwell & Cole, 2007). Finally, similar to previous reviews, the findings from this review reflected mixed results as to the degree to which certain variables (e.g., parenting stress, disease factors) influenced child, parent, and family outcomes (e.g., Sultan et al., 2016). For instance, Klassen et al. (2011) found that the first year of cancer posed increased health risk to parents, yet Bennett et al. (2013) did not find time since diagnosis to be an influencing factor on parenting stress. Although our review did not rate the methodological quality of the research, many studies were limited to using cross-sectional designs, heterogenous samples, non-randomization, or less sophisticated analytical techniques such as ANOVA and multiple regression. Therefore, in order to empirically validate the complexities of the family experience in pediatric cancer, future investigations should be randomized, employ longitudinal designs with rigorous analytical methods (e.g., structural equation modeling), and apply a strong theoretical framework that is optimal for testing both moderating and mediating effects (Masarik & Conger, 2017; Maxwell & Cole, 2007).

Family Stress Model Applications to Pediatric Cancer

To address the gaps identified from this review, we propose using the FSM (Conger & Conger, 2002) as a theoretical framework for studying families of children with cancer. A key advantage to applying the FSM in this context is the theoretical robustness of this time-tested model and its integration of economic pressure as a primary process in influencing parent psychosocial functioning and subsequently, child and family outcomes (Martin et al., 2019; Masarik & Conger, 2017). To guide our efforts, Fig. 2 provides an integrated conceptualization including the family-level directional relationships based upon this review with the FSM, as adapted from Masarik & Conger (2017). To illustrate this theoretical model, we present a case example (i.e., two-parent working class family with four children) outlining the sequential processes contained in Fig. 2. The hypothetical example depicted in the model begins when a child in the family is diagnosed with cancer (Box 1b) which compounds the economic hardship (Box 1a) currently experienced within this family where both parents work to manage daily expenses. Subsequently, the family now experiences the financial stress associated with the economic pressure caused by medical bills, the job loss of one parent to care for the child with cancer, as well as expenses necessary to travel for clinic appointments and hospitalizations. Central to the FSM, economic pressure directly impacts parent psychological functioning (Box 3) and in this case example, manifests symptoms of maternal

anxiety and paternal depression. The parents' negative emotional states contribute to anxiety in the child with cancer (Box 6a) which causes the child to exhibit increased behavioral problems and poor coping skills. Simultaneously, these parents experience distress related to the uncertainty of their child's prognosis which adds to the physical and emotional burden associated with caring for their child with cancer (Box 4b). Consequently, their psychological functioning is impacted by both economic pressure and cancer-related stressors; thus, the couple's relationship quality begins to deteriorate, and marital conflict arises (Box 4a). Meanwhile, the couple's discordant interactions spill over into the family system disrupting family routines and causing diminished cohesion (Box 5). As family functioning worsens it has a resonating impact on all subsystems within the family including parent psychological functioning (Box 3), sibling psychosocial behavior and adjustment (Box 6b) and likewise, the psychosocial functioning (e.g., health-related quality of life) of the child with cancer (Box 6a). Overall, family functioning in this context serves a pivotal role in how all members of the family system psychologically respond to this stressful event.

While our case example demonstrated the potential negative consequences on families experiencing cancer, there are a range of factors found from our review that may buffer or exacerbate any of the above described pathways. Hence, at the base of our model (Box 7) is a list of potential risk (e.g., stressful life events) or protective influences (e.g., social support, family ritual) that should be examined as either mediating or moderating variables within this conceptual model. For instance, perhaps the family in our hypothetical example made the commitment to continue family game night as well as other meaningful family routines to maintain a sense of normalcy and family connection. In doing so, these family rituals mitigated parent distress which in turn enhanced the ability in the child with cancer to develop better coping skills. Generally, this conceptual model is intended to be a starting point for examining family-level processes in pediatric cancer using core tenets of the FSM as this model has been used extensively to understand pathways in families experiencing economic pressure (Martin et al., 2019; Masarik & Conger, 2017). In addition to the proposed pathways outlined in Fig. 2, this model additionally allows for identifying and investigating new pathways for family-level processes in pediatric cancer. For example, studies could explore whether parenting stress directly influences family functioning and if family functioning mediates parent–sibling relationships. Further, studies on healthy siblings (e.g., Brody, 2004) have shown the importance sibling relationships have on development, yet sibling relationships have yet to be sufficiently examined in pediatric cancer research. Finally, this model could be applied to different family structures (e.g., single-parent families, stepfamilies) as well as families of different cultural and ethnic backgrounds.

Our application of the FSM to family processes has practical implications for clinicians, researchers, and policy makers. Clinicians may find the application of this model useful to inform their practice by understanding the complexities of economic pressure and the impact of financial pressure experienced by many families dealing with pediatric cancer. That is, openly discussing financial stress with parents early in the diagnosis phase can allow social workers or other members of the treatment team to seek available resources to mitigate some of the financial strain. Finally, this review calls attention to the impact health care and employment policies has on the financial ramifications for families facing pediatric cancer. As such, this conceptual model may be utilized at both an instructional and pedagogical level as a construct to guide explanation of the complex and interrelated psychosocial reactions among families coping with pediatric cancer.

This systematic review has a few limitations. First, our review was not exhaustive—it covered ten years, and exclusion of some studies focusing on children beyond treatment phase may have limited capturing a comprehensive view. Second, our interpretation is constrained to the methodological variance within the quality of studies reviewed, the imbalance between quantitative and qualitative methodologies, and the greater number of cross-sectional studies extracted. Third, while our decisions on categorizing variables was rigorously discerned, an element of bias cannot be completely ruled out. Finally, our search terms were specific to the aims of this review thus, our search did not include all terms relevant to the family stress model including those specific to parenting styles and practices which have been shown to mediate family adjustment. As such, future studies guided by the family stress model should consider parenting approaches as a key variable to child outcomes in pediatric cancer. Despite these few limitations, we believe our review was comprehensive and systematic in revealing themes to conceptualize family-level processes using the FSM.

The present systematic review aimed to identify and summarize the multifaceted factors that impact familial relationships and family functioning within the context of pediatric cancer and integrate them into a conceptual framework based upon the FSM. This conceptual model may expand previous pediatric cancer research by depicting directional paths and relationships between economic pressure, parent psychological functioning, and child outcomes as well as the many variables that mediate and moderate those relationships.

Compliance with Ethical Standards

Conflict of Interest The authors declare no competing interests.

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