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Family Resilience after Pediatric Cancer Diagnosis: A Systematic Review

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Abstract

Objectives: A systematic review was conducted to: 1) investigate family resilience in the context of pediatric cancer, and 2) examine theoretical, methodological and statistical issues in this literature. Family resilience was operationalized as competent family functioning after exposure to a significant risk.

Methods: Following guidelines for systematic reviews, searches were performed using Web of Science, Pubmed, Cochrane, PsycInfo and Embase. After screening 5563 articles, 85 fulfilled inclusion criteria and were extracted for review.

Results: Findings indicated that most families are resilient, adapting well to the crisis of cancer diagnosis. However, a subset still experiences difficulties. Methodological issues in the current literature hamper strong nuanced conclusions.

Conclusions: We suggest future research with a greater focus on family resilience and factors predicting it, based upon available theory, and conducted with attention toward unit of measurement and use of appropriate statistical analyses. Improvements in research are needed to best inform family-based clinical efforts.

Keywords: pediatric cancer; oncology; family; adaptation; resilience; systematic review

Family Resilience after Pediatric Cancer Diagnosis: A Systematic Review

Pediatric cancer is an unpredictable and uncontrollable stressor that puts the diagnosed child, his/her family members, and the family as a whole at risk for adjustment difficulties (Alderfer & Kazak, 2006). Research examining individuals' responses to the challenges posed by pediatric cancer, however, reveals resilience. For example, when compared with population norms, children diagnosed with cancer typically show no greater evidence of emotional maladjustment or psychological dysfunction (Phipps, 2007; Stam, Grootenhuis, & Last, 2001). In fact, in some studies, children with cancer demonstrate better emotional functioning than comparison groups and report benefit from their experience during treatment (Phipps, Steele, Hall, & Leigh, 2001) and positive changes within themselves, their relationships, and their plans for the future after treatment (Barakat, Alderfer, & Kazak, 2006). Similarly, most siblings of children with cancer score within normal limits on standardized measures of internalizing and externalizing disorders and may display increased empathy, maturity and responsibility after cancer diagnosis (Alderfer et al., 2010; Houtzager, Grootenhuis, & Last, 1999). Most parents of children with cancer also exhibit resilience. After diagnosis, moderate levels of emotional distress, anxiety, and acute or posttraumatic stress symptoms are observed (Grootenhuis & Last, 1997; Patino-Fernandez et al., 2008), but most improve within a matter of months to levels of distress comparable to normative samples (Dolgin et al., 2007; Ljungman et al., 2014). Parents also report posttraumatic growth, for example, being more patient and having a better understanding of what is important in life after their child's successful cancer treatment (Barakat et al., 2006).

Although there is evidence of resilience after childhood cancer diagnosis for individual family members, research into the *resilience of the family system* after diagnosis of pediatric cancer is less common. Given the presumptions that a family is more than the sum of its parts (Von Bertalanffy, 1973), and that a cancer diagnosis not only affects the individuals within

the family, but also their relationships with one another and the way in which the family functions (Alderfer & Kazak, 2006), it is important to understand the impact of cancer on the family as a whole. Furthermore, given the complex medical regimens of pediatric cancer, families must be able to alter roles and responsibilities, effectively communicate, manage emotions and successfully work as a team to meet treatment demands. In short, family-level processes and outcomes are important in pediatric cancer.

While systematic reviews are available for the literature regarding family adjustment after a diagnosis of pediatric cancer (Long & Marsland, 2011; Pai et al., 2007), this work has not been conceptualized within the framework of family resilience theory. Consistent with Hilliard, Harris, & Weissberg-Benchell (2012), in which resilience was defined as achieving one or more positive outcomes despite exposure to significant risk, we defined resilient families as those that return to, sustain, or achieve competent levels of functioning in one or more domains of functioning (i.e., cohesion, adaptation, communication) after being challenged by childhood cancer. A systematic review was deemed necessary to synthesize the relevant empirical literature, which emerges across various disciplines (e.g., psychology, nursing, medicine) using divergent (i.e., qualitative and quantitative) methods. The primary aim of the review was to determine whether there is evidence of family resilience after a diagnosis of childhood cancer. The secondary aim was to examine theoretical, methodological and statistical issues in the existing literature and formulate recommendations for future family resilience research.

Method

As outlined by Eiser, Hill and Vance (2000) and the Cochrane Collaboration (Deeks, Higgins, Altman & Green, 2011), we complied with a strict scientific methodology to ensure comprehensiveness, minimal bias and reliability of our systematic review. To this end, the following consecutive steps were taken: a) formulation of the scope of the review and

research question; b) thorough literature search; c) detailed data extraction; and, d) integration of the major findings and implications. Meta-analysis was not conducted because of heterogeneity across studies in terms of sample characteristics (e.g., different stages of treatment) and outcomes measured, as well as our desire to integrate qualitative findings to ensure a comprehensive review.

Literature Search and Inclusion Criteria

Web of Science, Pubmed, PsycInfo, Cochrane and Embase were searched using keywords selected in collaboration with a library information specialist and three researchers familiar with the field (details available upon request). Studies selected for inclusion examined aspects of functioning among families of children with cancer including: a) a subjective (qualitative) or objective appraisal of changes since diagnosis (longitudinal data); or, b) standardized scores, clinically meaningful categorization of families (e.g., based upon cut-scores), or a comparison to norms or control groups. Included studies investigated families of children diagnosed with any type of cancer before age 18. Studies published in languages other than English and non-empirical articles (i.e., reviews, case reports, books, book, reviews, commentaries, practice guidelines, conference abstracts, and dissertations) were excluded. Articles concerning families of children who died from cancer were also excluded, as theirs is a distinct family experience. Reference lists of the selected papers were also reviewed to ensure inclusion of all relevant papers.

Study selection

The database search was undertaken in July of 2014, identifying 5496 unique papers. The first and second author independently screened all titles for decisions regarding exclusion with 89% agreement. Disagreements were discussed and resolved. The 1592 remaining abstracts were then screened for exclusion, with an agreement rate of 83%. Again, disagreements were discussed and resolved. Finally, the first author screened the full texts of

the remaining 427 studies for final decisions regarding inclusion. The second author screened 25% with 93% agreement. Disagreements were discussed and, if necessary, a third reviewer was consulted; 72 studies were selected. Thirteen articles were added based on reference chaining, resulting in a final set of 85 papers (see Figure 1).

Data extraction

The first and second author extracted data from the studies in a systematic and standardized way, summarizing study characteristics and general findings on abstraction sheets (available upon request). Year of publication, journal and database were recorded along with methodological characteristics such as type of design (cross-sectional or longitudinal), measures used, and sample achieved (e.g., sample size, demographics). In addition, the theoretical framework, unit of measurement used and characteristics of the statistical analyses (interdependence of data reported by multiple respondents) were evaluated. Findings of the studies were extracted by summarizing the results in a few lines. The last author reviewed the information extracted against original publications to ensure accuracy. Authors were contacted for papers and information, as needed, but unpublished data was not requested.

Results

Part I: Characteristics of the Studies in the Review

A supplementary Table summarizes the methods and findings of each reviewed study. The majority of the reviewed studies were quantitative ($n=43$; 51%); 41% used qualitative methods ($n=35$; marked in text with ^{QL}), and the rest used mixed methods designs ($n=7$; 8%; marked in text with ^{mix}). Most studies used cross-sectional designs ($n=67$; 79%). Sample size varied from 3 to 209 families (6 to 465 individuals). Among the studies with quantitative data, 20 (40%) included comparison groups and 14 (28%) used standardized norms; sixteen studies (32%) did not make comparisons but provided longitudinal data or placed families into clinically meaningful groups (e.g., based on validated cut-scores). A wide variety of cancer

diagnoses were included in the studies, with leukemia, lymphoma and brain tumors most frequently represented. Time since diagnosis ranged from one week to 18.6 years. No timeframe was reported in 8 studies (8%) and was vague (e.g., “survivors”) in 31 others (36%).

Part II: Narrative Summary of Reviewed Studies

The narrative review is organized by six relatively distinct aspects of family functioning that emerged from the literature: cohesion, conflict, adaptability, communication, family support, and overall family functioning. For each subsection, a brief explanation of the concept is given, followed by the number and types of relevant studies identified. Findings from the perspective of each family member (child with cancer, parents, siblings) are then presented separately with qualitative results described before quantitative results.

Cohesion. Cohesion refers to the emotional bond between family members (Olson, 2000). Family resilience after pediatric cancer would be evident in a balance of connectedness and separateness, with possible increases in cohesion, whereas a lack of cohesion (disengagement) or too much cohesion (enmeshment) would be considered maladaptive. This construct was addressed in 17 qualitative, 17 quantitative and 2 mixed method studies.

In qualitative work, *children with cancer* spoke about the illness drawing the family closer (Clarke-Steffen, 1997^{QL}; Enskar, Carlsson, Golsater, & Hamrin, 1997a^{QL}; Woodgate & Degner, 2003^{QL}). In quantitative work, many children reported strengthened bonds with parents (Kvist, Rajantie, Kvist, & Siimes, 1991). In three studies, significantly greater levels of family cohesion were found as compared to control groups/standardized norms both during and after treatment (Beek, Shappin, Gooskens, Huisman, & Jongmans, 2014; Cornman, 1993; Trask et al., 2003) and four additional studies of families who were off-treatment found no differences in child-reported cohesion in comparison to control groups/standardized norms (Kazak, Christakis, Alderfer & Coiro, 1994; Kazak & Meadows, 1989; Madan-Swain et al.,

1994; Pelcovitz et al., 1998), all suggesting resilience. Only one study reported lower levels of survivor-reported cohesion among families of children with cancer than a normative sample; 40% of these teen survivors characterized their families as disengaged (Rait et al., 1992).

In qualitative studies, *parents* (Arabiati, Al Jabery, Abdelkader & Mahadeen, 2013^{QL}; Brody & Simmons, 2007^{QL}; Clarke-Steffen, 1997^{QL}; Koch, 1985^{QL}; Neil-Urban & Jones, 2002^{QL}; Nicholas et al., 2009^{QL}; Norberg & Steneby, 2009^{QL}; Quin, 2004^{mix}; Rocha-Garcia, Alvarez Del Rio, Hernández-Peña, 2003^{QL}; Sloper, 1996^{mix}; Woodgate & Degner, 2003^{QL}) often indicated that family cohesion was strengthened by the illness, sometimes with a tendency toward enmeshment (Velasco, Ddvila de Cortazar & Covarrubias-Espinoza, 1983^{QL}), though a minority indicated the opposite in one study (Sloper, 1996^{mix}). The bond between parents and the patient was specifically noted as becoming stronger (Kvist et al., 1991; Nicholas et al., 2009^{QL}; Norberg & Steneby, 2009^{QL}) but a minority of parents indicated that relationships with siblings became weaker (Kvist et al., 1991; Quin, 2004^{mix}). Thirteen quantitative studies compared parent-reported cohesion to norms/controls. Two studies, one with repeated assessments within 9 months post-diagnosis (Varni, Katz, Colegrove, & Dolgin, 1996) and a second studying families toward the end of treatment and beyond (Cornman, 1993) found mean levels of parent-reported cohesion to be greater than norms. Nine additional studies with samples ranging from newly diagnosed families through long-term survivors (Beek et al., 2014; Carlson-Green, Morris, & Krawiecki, 1995; Cohen, Friedrich, Jaworski, Copeland, & Pendergrass, 1994; Greenberg, Kazak & Meadows, 1989; Horwitz & Kazak, 1990; Kazak et al., 1994, Kazak & Meadows, 1989; Madan-Swain et al., 1994; Manne et al., 1995) found no differences between families of children with cancer and control groups/standardized norms. All of these studies suggest resilience. Finally, two studies suggested lower levels of cohesion among families of children with cancer, one investigating children on treatment (Morris et al., 1997) and a second of families post-treatment (Rosenberg

et al., 2014). Overall, findings generally point toward resilient outcomes (i.e., sustained or improved cohesion) from the parental perspective.

In qualitative studies, *siblings* also reported increased cohesion within the family (Chesler, Allswede, & Barbarin, 1991^{QL}; Clarke-Steffen, 1997^{QL}; Koch, 1985^{QL}; Prchal & Landolt, 2012^{QL}; Sargent et al., 1995^{QL}; Sloper, 2000^{QL}; Wiener et al., 2008^{QL}; Woodgate & Degner, 2003^{QL}; Woodgate, 2006a^{QL}). One quantitative study found sibling-rated cohesion to be greater than norms (Cornman, 1993), also suggesting resilience. However, increased closeness was not always perceived as being inclusive of the siblings (Chesler et al., 1991^{QL}).

In summary, most studies provide evidence for family resilience within the domain of cohesion after diagnosis of pediatric cancer, though siblings may experience being at the periphery of the family. We found no similarities among the few studies that suggested less cohesion among families of children with cancer nor any systematic differences between these studies and those suggesting resilience in terms of sample characteristics (e.g., diagnosis, time since diagnosis, treatment status, child age, country of origin) or methodology (e.g., respondent, measure, sample size, comparison group).

Conflict. Family conflict is openly expressed anger and discord among family members (Moos & Moos, 1994). Family resilience after pediatric cancer diagnosis would be evident if there were no increase in the amount of family conflict over time or in comparison to norms/controls. This construct was addressed in two qualitative and 12 quantitative studies.

Four quantitative studies compared family conflict reported by *children with cancer* to control groups/standardized norms, and findings were mixed. One of these studies indicated less child-reported conflict in families of children with cancer off-treatment compared to norms (Beek et al., 2014) and a second study indicated no difference between two such groups (Brown, Madan-Swain, & Lambert, 2003), both suggesting resilience. However, two additional studies indicated more child-reported conflict among families of children in

treatment (Manne & Miller, 1998) and a sample including those on maintenance or off-therapy (Cornman, 1993) when compared to norms or controls.

In two qualitative studies, *parents* of children with cancer reported themes of family conflict across the illness trajectory (Patterson, Holm & Gurney, 2004^{QL}; Shortman et al., 2013^{QL}) and two quantitative studies, one with families of children on treatment and a second with families in maintenance or off-therapy indicated more parent-reported conflict compared to norms and controls (Cornman, 1993; Morris et al., 1997). However, seven studies, with samples ranging from one month through at least five years post-diagnosis, indicated no differences between families of children with cancer and norms or controls on measures of conflict (Ach et al., 2013; Brown et al., 2003; Greenberg et al., 1989; Kronenberger et al., 1998; Noll et al., 1995; Varni et al., 1996) and two studies indicated less conflict for families of children with cancer, one studying families on treatment (Gerhardt et al., 2007) and the second studying families off-treatment (Beek et al., 2014), all suggesting resilience. One study assessing sibling-reported family conflict in families off active therapy indicated greater levels of conflict when compared with norms (Cornman, 1993).

In summary, reports of increased conflict were not found in samples exclusively consisting of off-treatment families suggesting long-term resilience in this domain; however some conflicting results did arise when samples included families on treatment (including maintenance). Conflict was reported across both qualitative and quantitative studies and across family members, but not consistently. Sample characteristics (e.g., diagnosis, age of child; country of origin) and aspects of study design (e.g., measure, sample size) did not seem associated with outcome. It seems that being on treatment may be a risk factor for conflict.

Adaptability. Adaptability is the amount of malleability in the family's leadership, role relationships and relationship rules (Olson, 2000). Well-functioning, resilient families would balance structure and flexibility after cancer diagnosis, possibly increasing in

adaptability; poorly functioning families would be rigid (i.e., not enough adaptability) or chaotic (i.e., too much; Olson, 2000). This construct was addressed in 11 quantitative studies.

Six studies assessed adaptability from the perspective of the *child with cancer*. One, with a sample combining families on- and off-treatment revealed a greater family adaptability than norms (Trask et al., 2003) and five involving off-treatment families tended to find no differences in comparison to norms or controls (Kazak et al., 1994; Kazak & Meadows, 1989; Madan-Swain et al., 1994; Pelcovitz et al., 1998; Rait et al., 1992), suggesting resilience.

In regard to *parent-reported* adaptability, three studies of families of children on treatment (Cohen et al., 1994; Horwitz & Kazak, 1990; Manne et al., 1995) and two studies of families of children off treatment (Kazak et al., 1994; Kazak & Meadows, 1989) found no differences from norms or controls in level of adaptability. An additional study found a higher degree of adaptability among families of survivors compared to norms (Rosenberg et al., 2014). All of these studies suggest resilience. However, one study of newly diagnosed families found that mothers tended to characterize their families as chaotic (Perricone et al., 2012), a second study of families on treatment found that a greater percentage of families of children with cancer than controls fell at the extremes for adaptability (i.e., either chaotic or rigid; Horwitz & Kazak, 1990) and a third study of families off-treatment noted that mothers were more likely than controls to characterize their families as rigid (Madan-Swain et al., 1994). No studies were found assessing family adaptability from the perspective of siblings.

Overall, it seems that most families of children with cancer are not different from norms/controls in terms of adaptability indicating resilience in this domain. While it is possible that a greater percentage adopt a chaotic way of functioning (near diagnosis) or a rigid style (during treatment and beyond), this may be a minority of families.

Communication. Communication, or the interchange of thoughts, feelings, experiences and information within the family, is generally believed to be an important

component of family functioning that can foster adaptation (Olson, 2000). Resilient families would maintain or increase communication within the family in response to cancer and their communication patterns would be open, clear, and effective. This construct was addressed in four qualitative studies, three mixed methods studies, and nine quantitative studies.

Four quantitative studies addressed expressiveness/communication within the family from the perspective of the *child with cancer*, all involving samples off active treatment. Greater expressiveness was reported in two of these (Beek et al., 2014; Cornman, 1993) and no differences were reported in a third (Madan-Swain et al., 1994), suggesting resilience. In the fourth study more than 60% of a sample of adolescent survivors endorsed ‘unhealthy’ family communication patterns, characterized as vague and with masked intent (Alderfer, Navsaria & Kazak, 2009). It is unknown if this rate is different from families without cancer.

In a qualitative study, nearly 70% of mothers reported an open communication style with their children (Clarke, Sheppard & Eiser, 2008^{QL}). In two quantitative studies, *parents of children with cancer* reported more expressiveness within their families, as compared to norms, both during (Varni et al., 1996) and after active treatment (Cornman, 1993) and in six additional studies with both on- and off-treatment samples, no differences were found for expressiveness (Beek et al, 2014; Morris et al., 1997) or communication (Greenberg et al., 1989; Kazak et al., 1997; Madan-Swain et al., 1994; Streisand, Tercyak, & Kazak, 2003). All of these studies suggest resilience. However, in two studies with researchers characterizing communication patterns within families of children with cancer, 59% of newly diagnosed families were found to share minimal (40%) or ambiguous (19%) information (Clarke, Davies, Jenney, Glaser & Eiser, 2005^{QL}) and only about 30% of off-treatment families evidenced effective communication patterns (Adduci et al., 2012^{mix}). An additional study found that about one third of parents of survivors rated their family communication patterns

post-treatment as “unhealthy” (Alderfer et al., 2009). These latter studies did not include control groups, so it is

unclear whether these rates are unique to families of children with cancer.

Five studies assessed *siblings*’ perceptions of family communication. Across three qualitative studies, most siblings reported being well-informed and satisfied with communication within their family (Havermans & Eiser, 1994^{mix}; Prchal & Landolt, 2012^{QL}; Sloper, 2000^{QL}). About two thirds of siblings in one sample, however, did want more information sooner (Sloper, 2000^{QL}) and a minority across two other samples reported becoming tired of hearing about cancer when months into or after treatment (Havermans & Eiser, 1994^{mix}; Prchal & Landolt, 2012^{QL}). A fourth study, conducted in China, reported that 60% of the siblings in their sample claimed not to have a chance to talk about the illness with their parents or sick brother/sister during treatment (Wang & Martinson, 1996^{mix}). This finding may be culturally-specific. In one quantitative study from the United States, siblings’ reports of expressiveness within their off-active-treatment families exceeded norms (Cornman, 1993).

In summary, when compared to norms/controls, children with cancer, their parents and siblings reported equal or increased communication/expressiveness within their families, suggesting resilience. However, observations of families, classification based upon cut-scores and comments of siblings provide some evidence for poor communication patterns; it is unclear if the rates of these patterns are typical. Finally, cultural differences may be important in this domain of family functioning.

Family Support. Family support refers to assistance, encouragement and caring from the family received or perceived by an individual (Walsh, 1998). Resilient families would be expected to maintain or increase support in response to cancer. Family support was addressed in 14 qualitative, two mixed method and 15 quantitative studies.

In qualitative studies, *children with cancer* reported that family support was very important in helping them get through cancer (Enskar et al., 1997b^{QL}; Havermans & Eiser, 1994^{mix}; Kyngas et al., 2001^{QL}; McGrath, Paton & Huff, 2005^{QL}; Ritchie, 2001^{QL}; Woodgate & Degner, 2003^{QL}; Woodgate, 2006b^{QL}). In fact, in one qualitative (Enskar et al., 1997a^{QL}) and three quantitative studies, they reported support or satisfaction with support from family/parents as being greater than that from any other source (i.e., friends, teachers; Kazak et al., 1994; Nichols, 1995; Trask et al., 2003). Three studies indicated that adolescents with cancer (Brown et al., 2003; Haluska, Jessee, & Nagy, 2002) - and specifically those undergoing haematopoietic progenitor cell transplant (Barrera, Andrews, Burnes & Atenafu, 2007) - reported more parental support than controls/norms and three including those on- and off-treatment found no differences (Kazak & Meadows, 1989; Manne & Miller, 1998; Wesley, Zelikovsky & Schwartz, 2013), generally indicating resilience.

In qualitative studies, *parents* also reported that family support was important in the context of cancer (Beltrao, Vasconcelos, Pontes, & Albuquerque, 2007^{QL}; Brody & Simmons, 2007^{QL}; Enskar et al., 1997b^{QL}; Enskar, Carlsson, Golsater, Hamrin, & Kreuger, 1997c^{QL}; Greenberg & Meadows, 1992^{QL}; Jackson et al., 2008^{mix}; McGrath et al., 2005^{QL}; Nicholas et al., 2009^{QL}; Shortman et al., 2013^{QL}; Woodgate & Degner, 2003^{QL}). Five studies comparing parents of cancer survivors to controls or norms with samples both on- and off-treatment indicated no differences in level of family support (Brown et al., 2003; Gerhardt et al., 2007; Kronenberger et al., 1998; Noll et al., 1995) suggesting resilience. Only one study, focused on parents of brain tumor survivors one to five years post-treatment, found levels of support lower than controls; this was for mothers' but not fathers' reports (Ach et al, 2013).

Finally, *siblings* also reported that family support was important in coping with cancer (Havermans & Eiser, 1994^{mix}; Sloper, 2000^{QL}; Woodgate & Degner, 2003^{QL}). In one quantitative study, siblings' ratings of parental support were not different from norms (Barrera

et al., 2007), suggesting resilience. Interestingly, friends and teachers were also frequently reported as support providers (Havermans & Eiser, 1994^{mix}; Sloper, 2000^{QL}). In one study, siblings rated support from parents as less important and available than support from friends and equal to that of teachers on both of these dimensions (Alderfer & Hodges, 2010).

Across studies, children with cancer, their parents and siblings all reported that family support helped them cope with the cancer experience. Children with cancer consistently rated support within the family as being equal to or greater than norms/controls, suggesting resilience in this domain. Parents tended to report this too with one exception – mothers of brain tumor survivors. Late effects and the associated demands placed on mothers in this specific population may raise their support needs, so this finding may be important clinically. Finally, studies of siblings indicated that support from outside the family is also important and readily available to them.

General Family Functioning. Resilient families would maintain or improve upon their general functioning patterns after cancer diagnosis. Perceptions of general family functioning across dimensions and domains was addressed in 20 qualitative, two mixed methods and 17 quantitative studies. Some studies assessing this construct combined data across family members. These findings are presented before data regarding individual family members' perspectives.

Qualitative studies combining *data across family members* revealed a shift in priorities and focus on the ill child that resulted in family disruption and loss of normal family life during treatment (Bjork, Wiebe, & Hakkstrom, 2009^{QL}; Clarke-Steffen, 1997^{QL}; Koch, 1985^{QL}; McGrath et al., 2005^{QL}), as well as a struggle post-treatment to return to normality (Bjork, Nordstrom, Wiebe, & Hallstrom, 2011^{QL}). In one study, when asked about the impact of surviving cancer, about 10% of adolescent *survivors* reported general family functioning difficulties (Greenberg & Meadows, 1991^{QL}). *Parents* specifically reported disruption of the

family, stress between family members, and trouble balancing family needs including those of siblings (Arabiat et al., 2013^{QL}; Bjork et al., 2009^{QL}; Brody & Simmons, 2007^{QL}; Enskar et al., 1997c^{QL}; Ferrell, Rhiner, Shapiro, & Dierkes, 1994^{QL}; Patterson et al., , 2004^{QL}; Quin, 2004^{mix}; Rocha-Garcia et al., 2003^{QL}; Sidhu, Passmore, & Baker, 2005^{QL}; Sloper et al., 1996^{QL}; Ward-Smith, Kirk, Hetherington, & Hubble, 2005^{QL}). *Siblings* reported disrupted family routines, being separated from the family due to treatment, and a general loss of family life (Chesler et al., 1991^{QL}; Prchal & Landolt, 2012^{QL}; Sargent et al., 1995^{QL}; Sloper, 2000^{QL}; Woodgate, 2006a^{QL}).

Three quantitative studies combined *data across family members* to assess family functioning. The first found that families of children with cancer, at least two years post-diagnosis, were functioning similarly to control families across a range of areas (e.g., cohesion, communication, consideration, satisfaction; Sawyer, Crettenden & Toogood, 1986). The second analyzed data from mothers, fathers and survivors (not nested within family), and found that 41% of the sample characterized their family as well functioning (high cohesion, high expressive, low conflict), 46% placed their family in a moderate range, and 13% reported poor functioning (low cohesion, low expressiveness, high conflict); 26% of families had at least one member reporting poor functioning (Ozono et al., 2007). In a third study, using a family mean across parents and survivors, 35% of families were found to score in the “unhealthy” range for general functioning (Alderfer et al., 2009). It is unknown whether these percentages are similar for families of children without cancer.

Turning to perceptions of *children with cancer*, across quantitative studies, including children on- and off- treatment, ratings of family functioning were no different from norms/controls, suggesting resilience (Foley, Barakat, Herman-Liu, Radcliffe & Molloy, 2000; Madan-Swain, Sexson, Brown & Ragab, 1993; Wesley et al., 2013; Yonemoto, et al., 2009),

Studies of *parents* also show no differences in general family functioning compared to norms/control groups, both during and after treatment (Foley et al., 2000; Kazak et al., 1997; Noll et al., 1995; Peterson, Cousino, Donohue, Schmidt, & Gurney, 2012; Sawyer, Antoniou, Toogood & Rice, 1997; Sawyer, Antoniou, Rice & Baghurst, 2000; Streisand et al., 2003) with relative stability across time from diagnosis through four years post-diagnosis reported in longitudinal studies (Fife, Norton & Groom, 1987; Sawyer et al., 1997; Sawyer et al., 2000). However, a subset does report problems. “Unhealthy” family functioning was reported by 26-38% of parents within 3 years of diagnosis (Long, Marsland & Alderfer, 2013), 20% of parents on average 3.5 years post-diagnosis (Martin et al., 2012), and 24%-38% of parents off-treatment (Alderfer et al., 2009; Peterson et al., 2012). Also, 11% of parents of long-term survivors reported problems with family harmony (Seaver et al., 1994^{mix}).

One study compared *sibling* ratings of general family functioning during treatment to controls and found no differences (Madan-Swain et al., 1993); however 47% of siblings in a second study of families within three years of diagnosis reported “unhealthy” general family functioning (Long et al., 2013). It is unclear if this percentage is different from norms.

In summary, qualitative research clearly indicates that childhood cancer disrupts the functioning of the family in various ways; however, for most families their general functioning, even in this time of stress, is within normal limits and similar to controls, suggesting resilience. Because control groups have not been consistently used, it is unclear whether the size of the subset of families experiencing “unhealthy” functioning is atypical.

Part III: Evaluation of the Literature

Theoretical considerations. In the majority of the studies ($n = 71, 84\%$), no theoretical framework was specified as guiding the research questions or selection of the variables. Failure to use theoretical frameworks risks limiting progression of the field, as advances cannot be made if theories go untested and unrevised.

Measurement considerations. Even though the included studies focused upon family-level constructs, only 5 studies (6%) measured family functioning from the perspective of all immediate family members. In fact, more than half of the studies ($n = 45$; 53%) used a single family member as the reporter. Because the *unit of interest* should harmonize with the unit of measurement (Weber, 2011), one could argue that studies with a single informant did not adequately assess family functioning. Discrepancies in perceptions of family functioning across family members (e.g., Alderfer et al., 2009; Peterson et al., 2012) speak to the need to collect data from multiple family members, including siblings, to best capture this construct.

Statistical considerations. In studies with data arising from multiple members within the same family, the interdependence of data within the family needs to be considered. Ignoring the dependency violates statistical assumptions of commonly used statistical approaches, generating inadequate test statistics (e.g., t or F), degrees of freedom and statistical significance values (i.e., the p value; Kenny, 1995). The majority of studies in our review ($n = 70$, 82%) avoided this issue through research design (e.g., qualitative analyses; single informant). Of the remaining studies, 11 (13%) avoided the issue by performing separate analyses for different family members without combining their data. Only 4 (5%) took the interdependence into account through creating a summary score across respondents or by using appropriate statistical techniques to account for the dependency (e.g., multilevel modeling, actor-partner model).

Overall Quality. In addition to the issues mentioned above, certain characteristics of the research base make it particularly difficult to draw strong conclusions. For example, heterogeneity across and within studies in regard to sample characteristics and operationalizations of family functioning presents barriers to conducting meaningful meta-analysis. With rare exception, studies have small heterogeneous samples and gather data at a single time point, precluding identification of factors that may reliably predict which families

experience the greatest difficulties meeting the challenges of pediatric cancer. While some studies used adequately sized demographically-matched control groups, these studies typically focused on comparing mean levels of functioning as opposed to comparing the percentages of families falling within dysfunctional ranges on the measures used, potentially masking important differences between groups on variables where both high and low scores may be problematic. Furthermore, nearly all studies relied on self-report of family functioning despite known drawbacks associated with this method (Schwarz, Groves & Schuman, 1998). Observational assessment of family interactions could be indispensable in furthering our understanding of family-level adaptation in response to childhood cancer.

Discussion

This systematic review provides general evidence of family resilience after a pediatric cancer diagnosis; however, more work is needed to best understand this phenomenon. While we are starting to acknowledge and understand individual strengths, less is known about family-level strengths after the experience of pediatric cancer (i.e., family resilience). To further this field of inquiry, future work should be theory-based, match the unit of measurement with the unit of interest (i.e., include all/many family members) and utilize appropriate statistical methods to nest data from family members within families.

The conclusions of this review are hampered by a few factors. We considered families as resilient if they returned to, sustained, or achieved competent levels of functioning after childhood cancer diagnosis. However, data regarding the functioning of the family prior to cancer, longitudinal data examining changes in the family over time after diagnosis and criteria for judging whether the functioning of the family is “competent” were rarely available. We frequently relied upon comparisons between families of children with cancer and controls/norms to determine resilience; however, one could argue that competent functioning in the context of pediatric illness may be different from the functioning of

families in which the children are healthy (see Alderfer & Stanley, 2012). For example, perhaps a more enmeshed or rigid pattern of functioning is adaptive in the face of cancer, at least for a certain period of time following diagnosis (Olson, 2000).

Our ability to draw conclusions about the resilience of families facing pediatric cancer was also hampered by the relative lack of studies using this framework as a basis for their research approach. In fact, other conceptualizations of family resilience could not be applied to the existing literature. For example, various family resilience theories (McCubbin & McCubbin, 1988; Patterson, 2002; Rolland & Walsh, 2006) do not see family functioning as the outcome of interest. Instead, family functioning is conceptualized as the process or means through which families achieve resilience. In these models, other outcomes are evidence of resilience, such as the family's ability to successfully meet future challenges (Rolland & Walsh, 2006), to maintain the family unit or to promote the development of individual members (Patterson, 2002). These outcomes are rarely, if ever, assessed in the context of pediatric cancer diagnoses. Other definitions of resilience in the context of pediatric illness, such as successful management of illness and treatment demands (Mitchell et al., 2004), are rarely examined as an outcome of family-level processes (e.g., effective communication).

Suggestions for Future Research

To parallel the movement toward conceptualizing individual responses after pediatric cancer within a resilience framework, future research regarding family-level responses to pediatric cancer needs to adopt family resilience models. This change would require research to involve multiple members within families, assessed over time. More homogenous samples or samples large enough to examine heterogeneity (e.g., time since diagnosis, age of children) are recommended. Mixed qualitative and quantitative methods, along with observational methods, are needed to assess the full range of relevant constructs including objective and subjective characterization of the demands of pediatric cancer, capabilities, characteristics,

and key processes of functioning within the family, and short and long-term family-level outcomes indicative of resilience. Research aimed at uncovering factors capable of identifying those families who might struggle to achieve resilience and isolating the mechanisms underlying family resilience would be most helpful for informing intervention.

Implications for clinical practice

Despite gaps in the current literature, adoption of a family resilience framework and the findings of our review have implications for clinical practice. First, attention should be focused upon the impact of cancer on the functioning of the family and family functioning should be routinely assessed in this population. Some families may need assistance in rallying their resources, developing a shared perspective of their experience and working together effectively to meet the demands of cancer. Such difficulties may simultaneously jeopardize cancer treatment and important longer-term family outcomes. Relevant empirically-based family-level intervention approaches are described in the literature (e.g., Rolland & Walsh, 2006; Saltzman, Pynoos, Lester, Layne & Beardslee, 2013). Second, clinical work with families should be sensitive to possible cultural differences, should consider the family within its larger socio-ecological context, and attend to subgroups that might be at elevated risk (e.g., families of children with brain tumors). Finally, based on our review, conflict within the family during treatment and communication with and support of siblings may be areas of common difficulty for families of children with cancer that should specifically be assessed and addressed as needed.

Conclusion

There is evidence for family resilience after pediatric cancer in many domains of family functioning but there are subsets of families that may struggle with the challenges posed by pediatric cancer. Limitations in the current literature hamper strong conclusions regarding the overall impact of pediatric cancer on family resiliency. Further research conducted from

the perspective of family resilience theory is needed to close gaps in the literature, refine family resilience models and provide clear guidelines for integrating these data into clinical practice.

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Fig. I Flow chart

