

A Meta-Analysis of Psychosocial Interventions for Siblings of Youth with
Chronic Medical Conditions

By

Christina M. Amaro

Submitted to the graduate degree program in Clinical Child Psychology and the Graduate
Faculty of the University of Kansas in partial fulfillment of the requirements for the
degree of Doctor of Philosophy.

Chair: Michael C. Roberts, Ph.D., ABPP

Ric G. Steele, Ph.D., ABPP

Eric M. Vernberg, Ph.D., ABPP

Rachel M. Moore, Ph.D.

Christopher M. Johnson, Ph.D.

Date Defended: August 16, 2019

The dissertation committee for Christina M. Amaro certifies that
this is the approved version of the following dissertation:

A Meta-Analysis of Psychosocial Interventions for Siblings of Youth with
Chronic Medical Conditions

Chair: Michael C. Roberts, Ph.D., ABPP

Date Approved: August 16, 2019

Abstract

Siblings of youth with chronic medical conditions may be at risk for psychological adjustment concerns, and interventions targeting the psychosocial needs of siblings are important to address. The purpose of the current study was to conduct a meta-analysis examining psychological interventions for siblings of youth living with chronic illnesses, with specific attention to improving their adjustment and distress. To identify articles that met inclusion criteria, literature searches were conducted using several search engines including PsycINFO, PubMed, ERIC, and ProQuest Nursing & Allied Health Source. Fifteen articles across 14 studies (826 participants) were included within the current review; studies included treatment and comparison group as well as pre-test post-test designs. Study characteristics were coded and risk of bias was assessed. Interventions primarily targeted siblings of youth diagnosed with cancer. Overall, findings from the current meta-analysis revealed aggregate effect sizes that were small but significant for both distress ($g = 0.48$, 95% confidence interval [CI] [0.32, 0.67]) and adjustment ($g = 0.23$; 95% CI [0.10, 0.36]). However, findings should be viewed cautiously in light of several limitations including small sample sizes, less rigorous study methodologies (e.g., pre-test post-test designs), and potential high risk of bias. The current meta-analysis elucidates the need for further sibling intervention development, including randomized controlled trials as well as larger and more diverse samples.

Acknowledgements

They say it takes a village, and I'm incredibly grateful for mine. There are numerous individuals who have supported me throughout my graduate training, including completion of this dissertation. First, I would like to thank Michael. I have learned so much from you through courses, Stone Soup lab meetings, and our time up in Strong Hall. Thank you for opening the door to so many exciting opportunities and supporting me in various endeavors. You have also strengthened my knowledge of pediatric psychology history, and I am now well prepared for any pediatric psychology trivia game. Most importantly, thank you for being such a wonderful mentor. You have had such a positive and meaningful impact on so many individuals within clinical child and pediatric psychology, and I am so lucky that I get to be included within that network. I aspire to be the kind, patient, knowledgeable, and supportive mentor that you have always been to me.

To my committee members, thank you for taking the time to serve on my committee and for providing thoughtful comments to improve the project. To the Clinical Child Psychology Program (CCPP) faculty and affiliated members, thank you for furthering my training through coursework, practicum, and research opportunities. I am also grateful for my practicum supervisors at KU Medical Center and Children's Mercy Hospital; thank you for helping me to gain exposure to pediatric psychology within academic medical settings and strengthening my clinical skills. Lauren, Leesa, and Tammie, thank you for your dedication to the CCPP students, and your many efforts to keep the program and clinic running smoothly. To the students of CCPP, including my fellow Stone Soup lab-mates, I am so fortunate that you are not only my colleagues but

also good friends; I am glad we are on this journey together. I would also like to thank Amy for assisting with the coding needs for this project. With regards to funding for the current project, I am grateful for the support from the University of Kansas Psychology Strategic Initiative Grant.

Furthermore, I would like to thank my supervisors and mentors at Nemours who have helped me to strengthen my skills, encouraged me to further build my confidence, and supported my growth as a professional. To my cohort at Nemours, this was quite a year, and I am so fortunate that I was able to spend it with all of you. I would also like to thank the members of the Sibling Partnership for Advocacy, Research, and Care in Childhood Cancer for kindly welcoming me into the sibling community and providing a range of different opportunities to learn and contribute.

To my parents, thank you for always believing in me and encouraging me to pursue a variety of opportunities, including experiences that were completely out of my comfort zone. To my sisters, Catherine and Jacqueline, thank you for sending along words of encouragement and much needed jokes as well as taking several trips to visit; I am so lucky to have you two as sisters and friends. Furthermore, I am grateful for my extended family members for their encouragement over the years; I am so fortunate to have such a strong support system. Similarly, I am thankful for my friends, both near and far, who continually sent well wishes. Finally, thank you to my amazing partner, Anthony. I greatly appreciate your kindness and patience with me when I insisted we go to various coffeehouses so I could finish up work, as well as your encouragement of breaks when I strongly needed them.

Table of Contents

Introduction.....	1
Sibling Adjustment to Chronic Medical Conditions.....	2
Interventions for Siblings.....	4
Current Study.....	9
Method.....	9
Literature Search.....	9
Defining Intervention.....	10
Defining Outcome Variables.....	10
Inclusion/Exclusion Criteria.....	11
Identification.....	13
Included.....	13
Eligibility.....	13
Screening.....	13
Data Extraction.....	13
Statistical Analysis.....	16
Results.....	17
Description of Studies.....	17
Overall Effect Sizes.....	29
Exploratory Analyses.....	30
Risk of Bias.....	31
Publication Bias.....	32
Discussion.....	33
Limitations.....	35
Future Directions.....	37
Conclusions.....	46
References.....	47

Introduction

Utilizing a social-ecological framework (Bronfenbrenner, 1979), a child's chronic medical condition impacts not only the child, but interacts with a variety of systems, such as family, peers, school, hospitals, the neighborhood, culture, and technology (Kazak, Alderfer, & Reader, 2017). The family system is comprised of interrelated subsystems including dyadic relationships (e.g., parents, sibling-child with a chronic medical condition, parent-child) and larger subsystems (e.g., mother-father-child, mother-sibling-child with a medical condition; Kazak, 1997). Within pediatric psychology there is a long history of research examining components of the family system within the context of childhood chronic illness (Canter, Amaro, Noser, & Roberts, 2018). For example, parental distress has been linked to child distress for youth with spina bifida as well as medical procedure distress for youth undergoing treatment for cancer (e.g., Caes et al., 2014; Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004). Research has also examined the impact of family factors (e.g., parental involvement, family functioning, parental psychopathology) on various outcomes including youth adherence to treatment regimens (e.g., Hommel, Ramsey, Loiselle, & Ryan, 2017). However, much of this literature has been dedicated to the examination of parents and family functioning broadly with less attention on the psychological functioning of siblings (Kazak, Rourke, & Navsaria, 2009).

Siblings of youth with a chronic medical condition (hereinafter referred to as "siblings") may be impacted in a variety of ways. For instance, they may experience disruptions to their typical routine, changes in their role within the family system (e.g., family relationships, status within the family), and limited attention from parents who

may be attending more to the child with a chronic medical condition (e.g., DeMaso, Martini, & Cahen, 2009; Deavin, Greasley, & Dixon, 2018; Murray, 1999). During hospitalizations, siblings may be separated from their parents and sibling with a chronic medical condition and remain in the care of their relatives for extended periods of time. Siblings may experience a reduction in social opportunities, such that they need to leave immediately after school to help with responsibilities in the home or are unable to participate in extracurricular activities due to limited family finances (DeMaso et al., 2009). Further, siblings may have a narrow understanding and/or incorrect information about the chronic health condition, which may cause undue distress and fear (DeMaso et al., 2009; Deavin et al., 2018).

Sibling Adjustment to Chronic Medical Conditions

Studies have demonstrated that most siblings tend to adjust well over time and demonstrate resiliency (e.g., Kazak et al., 2017; Van Schoors, Caes, Verhofstadt, Goubert, & Alderfer, 2015). Resiliency might be influenced by a family's ability to normalize the adversity of a chronic medical condition as well as the ability to reframe the situation as something they can manage (Patterson, 1991). Systematic reviews suggest that siblings of youth diagnosed with cancer may initially experience social problems and elevated psychological symptoms closer to their siblings' diagnosis, but overall, demonstrate positive outcomes over time, such as increased maturity, empathy, and independence (Alderfer et al., 2010; Long, Lehmann et al., 2018). It also appears that siblings of youth with cancer endorse depressive and anxiety symptoms within normal limits (Long, Lehmann et al., 2018). Several aspects of family functioning (i.e., greater cohesion, support, communication, and satisfaction along with decreased conflict) are

related to enhanced sibling adjustment and resiliency in families who are impacted by chronic medical conditions, such as spina bifida and pediatric cancer (e.g., Bellin, Bentley, & Sawin, 2009; Long, Marsland, & Alderfer, 2013; Van Schoors et al., 2015; Van Schoors et al., 2017). Similarly, siblings may also experience positive changes as a result of the diagnosis, such as greater social support particularly within the school setting and closer friendships (Samson, Rourke, & Alderfer, 2016).

On the other hand, some siblings may experience difficulties with adjustment to a medical condition and demonstrate psychosocial functioning challenges. Within the pediatric oncology literature, for example, younger siblings may initially be at increased risk for somatic complaints within the first two years of diagnosis (Alderfer et al., 2010). There are increased concerns for post-traumatic stress disorder, with approximately one-fourth of siblings meeting criteria for a diagnosis, increased negative emotional reactions, family dysfunction, and possibly diminished quality of life (Alderfer et al., 2010; Long, Lehmann et al., 2018). Siblings of youth with cancer report increased school absenteeism, less time with friends, and decreased involvement in extracurricular activities (French et al., 2013; Long, Lehmann et al., 2018; Samson et al., 2016). For siblings of youth with spina bifida, behavioral problems have been associated with increased sibling conflict and diminished peer support (Bellin et al., 2009). Furthermore, siblings of youth diagnosed with inflammatory bowel disease may experience elevated emotional and behavioral difficulties and have increased worries about their sibling being teased because of their medical condition (Mackner, Sisson, & Crandall, 2004).

Across illness populations, siblings report a range of negative feelings including jealousy related to reduced parental attention (e.g., feeling unimportant, neglected, left

out), loneliness, and vulnerability as well as a range of somatic complaints, such as sleep disturbances and headaches (e.g., Batte, Watson, & Amess, 2006; Deavin et al., 2018; Knecht, Hellmers, & Metzger, 2015). There may be increased worries related to the health of their sibling with a medical condition (Batte et al., 2006; Deavin et al., 2018). Meta-analyses have been conducted to examine the effect of chronic illness on siblings' psychosocial functioning. Specifically, Sharpe and Rossiter (2002) found a small, negative effect ($d = -.20$). Providing an updated review, Vermaes and colleagues (2012) found a similar small, negative effect ($d_+ = -.10$), noting that siblings demonstrated elevated internalizing ($d_+ = .17$) and externalizing problems ($d_+ = .08$) as well as less positive self-attributes ($d_+ = -.09$). Inconsistent with findings from Sharpe and Rossiter (2002), the updated review suggested that siblings of youth with greater illness severity (e.g., life threatening illnesses, intensive daily treatments) experienced increased psychological concerns and younger siblings demonstrated less negative self-attributes (Vermaes et al., 2012). The discrepant findings may have been due to methodological differences in studies included within the analyses (Vermaes et al., 2012).

Interventions for Siblings

Although adjustment to chronic medical conditions appears to vary across siblings, a significant subset of siblings do appear to experience increased challenges and psychosocial distress; therefore, this subgroup of siblings may benefit from psychological services. As early as 1985, there was a call to evaluate psychosocial interventions for siblings (Drotar & Crawford, 1985). Recent recommendations and standards of care have indicated that, while it is important to address concerns related to the child with a chronic medical condition, the psychosocial needs of family members should also be taken into

consideration. For example, Principle 7 of the American Academy of Child and Adolescent Psychiatry Practice Parameter for youth with chronic illnesses pertains to family members (DeMaso et al., 2009). Acknowledging the potential impact chronic medical condition has on a family, the Practice Parameter suggests mental health screening for siblings. Family therapy (e.g., working with the family as a unit or working with family members, including siblings, separately) is also recommended to treat mental health concerns when indicated. Overall, the majority of these recommendations are specific to providers working directly with parents, including providing psychoeducation, addressing coping strategies, and discussing appropriate means of gradually transferring medical responsibilities from caregivers to the child with a chronic medical condition (DeMaso et al., 2009). Additionally, Principle 11 of the Practice Parameter recommends connecting families to additional community resources and services as needed. For instance, social service agencies may provide a range of resources for families including respite care, childcare services for siblings, and transportation. Organizations, such as parent support groups and national organizations for specific medical conditions, may also serve as a form of support for the family (DeMaso et al., 2009).

Within pediatric oncology, Psychosocial Standards of Care were recently developed as part of an interdisciplinary collaboration (Wiener, Kazak, Noll, Patenaude, & Kupst, 2015). One of the 15 standards is directly aimed at siblings and recognizes the importance of psychosocial support for siblings, as they tend to be an at-risk group (Gerhardt, Lehmann, Long, & Alderfer, 2015). Given the unique stressors and psychosocial concerns that siblings face, this Sibling Standard of Psychosocial Care focuses on the importance of communication with siblings, especially regarding their

sibling's illness and treatment, as well as supportive care, including psychoeducation, coping strategies, and ongoing assessment and treatment (Gerhardt et al., 2015). Taken together, these recommendations demonstrate professional agreement that the psychosocial concerns of siblings should be addressed. Therefore, it is necessary to identify effective interventions that can achieve these goals of meeting the needs of siblings.

Meta-analyses have been conducted to examine the effectiveness of family-based interventions that seek to reduce psychological concerns in families with a child with a chronic medical condition. A meta-analysis conducted by Pai, Drotar, Zebracki, Moore, and Youngstrom (2006) examined interventions for children with cancer and their parents, and findings demonstrated small effects for improvements in parent distress and adjustment. Subsequently, Law, Fisher, Fales, Noel, and Eccleston (2014) conducted a meta-analysis of the literature on parent and family-based interventions for youth with chronic medical conditions. Findings indicated that these interventions had small, but significant, effects on parent behaviors which continued through follow-up (Law et al., 2014). The primary focus of these meta-analyses has been on interventions with the child with a chronic medical condition and their parents in particular. Furthermore, a recent meta-analysis examined interventions for families of youth diagnosed with cancer (Sánchez-Ega, Rubio-Aparicio, Sánchez-Meca, & Rosa-Alcázar, 2019). The analyses, which combined data from both siblings and parents, suggested positive effects on family anxiety symptoms ($d_{adj} = 0.34$) and problem-solving skills ($d_{adj} = 0.38$; Sánchez-Ega et al., 2019).

In addition to having an understanding of family-based interventions broadly, it is also important to examine the unique impact of interventions for siblings. For instance, Prchal and Landolt (2009) conducted a systematic review that examined interventions for siblings of youth with cancer. The findings indicated that many of the interventions were provided in a group format and generally produced an improvement in several psychological domains including depressive symptoms, knowledge of oncology, and health-related quality of life; however, some interventions exhibited no significant treatment effects, and there were reportedly mixed findings in other areas, such as anxiety symptoms, posttraumatic stress symptoms, and behavioral problems (Prchal & Landolt, 2009). When possible, the authors calculated effect sizes from the studies and used the interpretation guidelines provided by Cohen (1988), such that $d = 0.2$ is a small effect, $d = 0.5$ is medium, and $d = 0.8$ is large. The effect size ranges were as follows: 0.31-0.47 (small to medium) for depression, 0.36-0.98 (small to large) for anxiety, 0.35-1.45 (small to large) for behavioral problems, 0.47 (medium) for posttraumatic stress symptoms, 0.32 (small) for health-related quality of life, and 0.22-1.45 (small to large) for parent-sibling communication. Findings demonstrate that there is great variability in effect sizes within and across domains. The authors noted that, although there is tentative evidence for these treatments for siblings of youth with cancer, detailed recommendations regarding the effectiveness of interventions could not be provided at that time (Prchal & Landolt, 2009).

Two reviews have examined interventions targeting siblings more broadly, such that they examined interventions for siblings of youth with chronic medical conditions and mental health conditions. Specifically, Hartling and colleagues (2014) conducted a

systematic review examining interventions for siblings of youth with a chronic medical condition (along with one sample that included youth diagnosed with acute and chronic medical conditions) or disability (e.g., intellectual disabilities, autism spectrum disorder, attention-deficit/hyperactivity disorder, learning disabilities). Although published in 2014, their literature search only included studies published through 2008; the manuscript had been accepted for publication in 2009. The review demonstrated mixed results; specifically, improvements were displayed in areas such as anxiety, depression, self-esteem, medical knowledge, and attitudes. However, changes were not observed for disease-related fear or sibling-report of emotional symptoms or behavioral difficulties (Hartling et al., 2014). Additionally a more recent meta-analysis examined well-being interventions for siblings of youth with chronic physical or mental health conditions (e.g., learning disabilities, autism spectrum disorder, and intellectual disability; Smith, Pereira, Chan, Rose, & Shafran, 2018). Findings from pre-post intervention designs suggested increased sibling knowledge of medical conditions ($d = 0.69$) and diminished behavioral concerns ($d = -0.44$); however, improvements in behavioral concerns were not evident when examining controlled studies (Smith et al., 2018).

Although reviews have been useful in providing some evidence to suggest that interventions for siblings of youth with chronic medical conditions may improve psychosocial outcomes, there are several limitations of these prior reviews and findings on the effectiveness of interventions for siblings have been inconsistent. While siblings of youth with chronic medical conditions and other conditions, such as autism spectrum disorder and intellectual disabilities, may share similar experiences (e.g., changes in the family system), there are also many other experiences that are likely different (e.g.,

potential hospitalizations for youth with a chronic medical condition; disease-related fears). As such, interventions may address these sibling needs in different ways.

Combining data from interventions on parents and siblings may diminish the ability to fully understand the unique impact and effectiveness of interventions for siblings.

Furthermore, limited inclusion of studies on interventions for siblings of youth with only one specific chronic medical condition may not be a complete depiction of the current literature. Therefore, it may be beneficial to conduct a meta-analysis specifically examining interventions for siblings of youth with chronic medical conditions.

Current Study

Prior literature provides some preliminary evidence for providing psychosocial support for siblings given the unique stressors and psychosocial concerns that they may encounter. The current study sought to extend the literature by updating reviews on interventions targeting siblings, specifically those of youth diagnosed with a chronic medical condition, and also by quantifying the effectiveness of interventions in improving psychosocial functioning (i.e., distress and adjustment) of siblings of youth with chronic medical conditions.

Method

Literature Search

Literature searches were conducted using several search engines including PsycINFO, PubMed, ERIC, and ProQuest Nursing & Allied Health Source with the following search terms: (sibling or brother or sister) and (illness or chronic illness or disease) and (intervention or treatment or support or therapy or program or camp) and (child* or adolescen* or youth or pediatric). Limits were set on searches to exclude

articles written in any language other than English. Within the meta-analysis literature, authors have recommended working with a research librarian in order to appropriately tailor search terms for the specific search engine, as indexing systems may vary across engines (e.g., Lipsey & Wilson, 2001; Wu, Aylward, Roberts, & Evans, 2012); therefore, a research librarian at the University of Kansas was consulted. Duplicates yielded from searches were removed. Literature searches were also conducted using GoogleScholar. To identify any relevant dissertations, a search was conducted in *ProQuest Dissertations and Theses*. Backward and forward searches were conducted on included articles. After duplicates were excluded, 9,735 articles were screened for inclusion within the current review. The initial search was initiated on March 19, 2018 and completed on December 8, 2018.

Defining Intervention

For the purposes of this meta-analysis, a psychological intervention was defined as a program, support service, or therapy for siblings with the goal of improving adjustment and distress outcomes (e.g., internalizing symptoms, social support). Additionally, the intervention should consist of a structured interaction between a sibling participant and an individual facilitating the intervention (e.g., psychologist, nurse, social worker, lay volunteers, and graduate student). Given that a variety of professionals may work with siblings of youth with chronic illnesses, a variety of therapy types were included (e.g., cognitive behavioral therapy, art therapy, music therapy). However, interventions that consisted of medication were excluded.

Defining Outcome Variables

Prior meta-analyses and systematic reviews on interventions targeting siblings have noted a range of outcome variables (e.g., internalizing problems, posttraumatic stress symptoms, academic achievement, social functioning) that have been examined (e.g., Hartling et al., 2014; Prchal & Landolt, 2009; Vermaes et al., 2012). For the purposes of this meta-analysis, sibling psychological distress and adjustment were examined; this focus provides a means of being inclusive of many types of outcomes that fall under similar umbrellas, but also limits the number of outcomes being examined. This approach was utilized by Pai and colleagues (2006) in which distress was defined as “upsetting or aversive feelings or affect experienced by an individual” (e.g., anxiety symptoms, depression), whereas adjustment “was defined as skills and abilities that are related to social, occupational, and education functioning” (e.g., problem-solving skills, social skills, perceived competence, quality of life; p. 979).

Inclusion/Exclusion Criteria

Within the literature, authors have been inconsistent in their conceptualization and operationalization of the term “pediatric chronic health conditions” (van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). For the purposes of this current review, pediatric chronic medical conditions are “health or medical problems that last 3 months or more, require ongoing medical care, affect a child’s normal activities, and are associated with functional impairment” (Compas, Jaser, Reeslund, Patel, & Yarboi, 2017, p. 327). This definition is based on the systematic review conducted by van der Lee and colleagues (2007).

To be included within the study, articles met the following criteria: (a) siblings of youth with chronic medical conditions were the primary target of the intervention; (b)

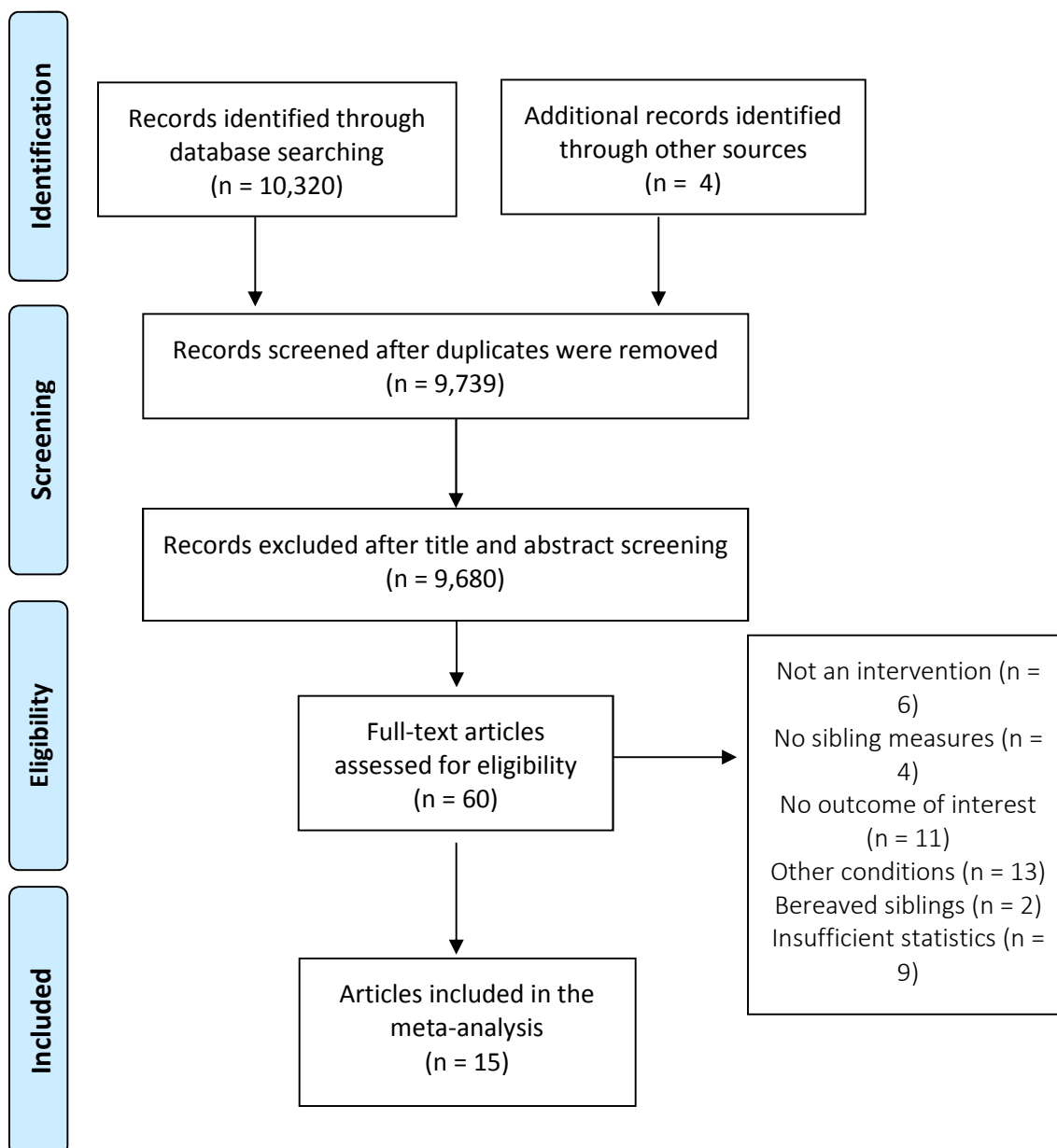
mean age of participants was 18 years old or younger at the start of the intervention; (c) included a measurable adjustment and/or distress outcome (e.g., anxiety symptoms, depressive symptoms, social skills, quality of life); (d) reported in English; and (e) reported enough information for effect sizes to be calculated. Studies were excluded for the following reasons: (a) intervention was family-based but does not provide sibling measures and/or siblings were not part of the family-based intervention; (b) intervention did not target outcome(s) of interest; (c) intervention targeted siblings of youth with conditions that are not chronic medical conditions, such as acute medical illness (e.g., acute pain, otitis media, broken bone) or neurodevelopmental disorders (e.g., autism spectrum disorder, intellectual disability), and (d) intervention primarily targeted bereaved siblings or sibling donors of hematopoietic stem cell transplant (HSCT).

Studies examining bereaved siblings were excluded from the current analysis because bereaved siblings may experience a different set of challenges and interventions for them may have notable differences; this decision is consistent with prior reviews (e.g., Prchal & Landolt, 2009; Van Schoors et al., 2017). Similarly, the literature on HSCT suggests elevated psychosocial concerns (e.g., increased anxiety and posttraumatic stress symptoms; lower self-esteem) in sibling donors in comparison to nondonor siblings as well as increased challenges (e.g., increased guilt) if the transplant is not successful (Packman, Weber, Wallace, & Bugescu, 2010). Thus, intervention components may be significantly different and were excluded from the current review.

Using the inclusion and exclusion criteria, 9,739 titles and abstracts were screened. After screening, 60 full-text articles were reviewed. A total of 15 articles across 14 studies were eligible for inclusion within the current meta-analysis. See Figure 1 for

the flow diagram based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Moher et al., 2009).

Figure 1. PRISMA flow diagram for included studies.



Data Extraction

Coding of study characteristics. Studies that met inclusion criteria were coded independently by two coders (the author and another graduate student). Discrepancies were discussed until a consensus was reached. Several participant characteristics were coded, including sibling age (i.e., range, mean), sibling's medical condition, socioeconomic status, sample size and attrition, percentage of the sample that is female, and percentage of sample that is non-Caucasian. Study characteristics were also coded (i.e., parent included in treatment, publication type, publication year). The following intervention characteristics were coded: (a) intervention setting (e.g., group, individual, camp); (b) intervention duration (e.g., number of weeks); (c) who administered the intervention (e.g., psychologist, graduate student); and (d) primary objective of the intervention (e.g., increase coping, family communication, reduction of psychological symptoms, peer support).

Coding risk of bias. Risk of bias was examined based on the Cochrane Collaboration risk of bias tool (Higgins, Altman, & Sterne, 2011). Specifically, risk was categorized in the following areas: 1) random sequence generation, 2) blinding of participants and personnel, 3) incomplete outcome data, and 4) selective reporting. Each domain was rated as either low (e.g., the data suggest that a specific domain is less likely to be biased), high (e.g., methodology suggests there was a high potential for bias), or unclear (e.g., insufficient detail to determine the potential bias).

Coding and calculating effect sizes. Effect sizes were managed and calculated using the software program Comprehensive Meta-Analysis version 3 (Biostat, Englewood, NJ). Effect sizes were calculated based on a variety of study statistics

including means, standard deviations, p -values, t -statistics, and sample sizes. If studies reported both total and scaled scores, only the total scores were used to calculate effect sizes. Studies that reported nonsignificant findings but did not provide a p value were assigned $g = 0.0$ as a conservative measure, which may have underestimate the true effect size (Card, 2012). Formulas based on specific study designs and study statistics were used. For instance, mean differences and pooled standard deviations were used to calculate effect sizes for studies that included a comparison group. An equation for dependent samples was used that accounted for bias of repeated measures (Card, 2012). When calculating the standard error for repeated measure designs, the correlation coefficient, or the interindividual stability of the measure across the two time points, is required (Card, 2012). However, several of the included studies did not report this coefficient for measure(s). To avoid using a fixed value without empirical data, the manuals for measures and existing literature were examined. When feasible, the median correlation from included studies were also used (Kahana, Drotar, & Frazier, 2008). If a correlation coefficient could not be obtained using these procedures, a conservative estimate of $r = 0.7$ was used per recommendations by Rosenthal (2001).

To avoid violation of assumptions of independence, mean effect sizes were calculated for studies that reported multiple outcome measures of the same construct (e.g., changes in an anxiety measure and a measure of depression; Card, 2012; Lipsey & Wilson, 2001). For each outcome, positive effect sizes indicated improvements. To allow for comparison across studies and methodologies (i.e., pre-post design, comparison group or Randomized Controlled Trial [RCT]), all weighted mean effect sizes were converted using Hedges' g . Although similar to Cohen's d , Hedges' g is recommended for studies

with smaller sample sizes (Card, 2012; Durlak, 2009). Finally, a stem-and-leaf plot was created to identify and exclude any potential outliers (i.e., greater than three standard deviations from the mean) from analyses (Lipsey & Wilson, 2001).

Statistical Analysis

In order to account for sample sizes, study effect sizes were weighted based on standard error, such that studies with more precise, smaller standard errors (resulting from larger sample sizes) had more weight (Lipsey & Wilson, 2001). A random effects model is recommended for smaller sample sizes and was used in the current analyses (Lipsey & Wilson, 2001). An overall mean effect size, along with confidence intervals around the mean effect size, was calculated for each outcome (i.e., adjustment and distress). The magnitude of study effect sizes was interpreted according to the following guidelines: small effect (less than 0.20), medium effect (0.50-0.79), and large effect (greater than 0.80; Cohen, 1988). If zero is included within the confidence interval, the effect is not considered statistically significant.

Heterogeneity analyses were then computed using the Q-statistic. This analysis examines the variability within the sample to determine if the variation is greater than what would be expected for the standard error (e.g., due to sampling error in addition to study specific differences; Card, 2012). When a Q-statistic was significant, the I^2 index, which provides the ratio of between-study variability to the total variability, was examined. Based on recommendations, small heterogeneity is defined as $I^2 = 25\%$, medium heterogeneity as $I^2 = 50\%$, and large heterogeneity as $I^2 = 75\%$ (Huedo-Medina, Sánchez-Meca, Marín-Martínez, & Botella, 2006). To examine potential sources for differences, moderator analyses are also recommended when a Q-statistic is significant.

However, due to the limited number of studies within the current review, moderator analyses were not conducted.

Finally, publication bias is a common concern in meta-analyses, such that non-significant findings are less likely to be reported or published, and thereby not fully captured in a meta-analysis. Therefore, a fail-safe N was calculated for each outcome, which represents the number of studies with an effect size of zero that could be added to the meta-analysis before the overall mean effect sizes drops to the smallest meaningful effect size (Card, 2012).

Results

Description of Studies

The current analysis included 15 articles across 14 studies. Given multiple time points (i.e., post and follow-up) and outcome variables, 107 basic effect sizes were calculated. As previously noted, multiple effect sizes from a study were aggregated in order to prevent violations of independence. Studies were published between 1986 and 2018, with 33% ($n = 4$) of articles having been published in the last year. Four studies were conducted by the same team in Canada; the remaining studies were conducted in the following countries: Germany ($n = 2$), United States ($n = 2$), Ireland ($n = 1$), Australia ($n = 1$), Netherlands ($n = 1$), Israel ($n = 1$), South Korea ($n = 1$), and Switzerland ($n = 1$). Four articles across three studies reported on RCTs (Barrera, Atenafu, Nathan, Schulte, & Hancock, 2018; Barrera et al., 2018; Kazak et al., 2004; Prchal, Graf, Bergstaesser, & Landolt, 2012) and two interventions examined treatment versus a comparison group (Cimini, 1986; Niemitz & Goldbeck, 2018). The remainder of the studies used a pre-test

post-test intervention design. The majority of included studies were from peer-reviewed articles, whereas two of the included studies were unpublished dissertations.

Participant characteristics. The current meta-analysis included a total of 826 participants, with a mean sample size of 55 participants across the 14 studies; studies reported a range of 17 to 259 participants. Participants were ages 4 to 20 years of age. Eleven studies did not report on participant ethnicity, and 10 studies did not provide information on participant socioeconomic status (SES). The primary sibling medical condition was cancer and a range of diagnoses were reported (e.g., leukemia, neuroblastoma, solid tumors, brain tumor). Additionally, one study (Besier, Holling, Schlack, West, & Goldbeck, 2010) examined siblings of youth diagnosed with cystic fibrosis (CF), congenital heart disease (CHD), and cancer. Time since sibling medical diagnosis ranged from 1 month to 126 months; 5 studies did not report this information. See Table 1 for additional study participant characteristics.

Niemitz (2018)	Prchal (2012)	Sidhu (2006)
184	30	26
4-18	6-17	8-13
9.20 (3.21)	Intervention Median = 8.5, Control Median = 11.5	9.8
47%	41%	52%
Not reported	Not reported	Not reported
Not reported	63.33% middle SES, 36.67% upper SES	Not reported
Germany	Switzerland	Australia
Cancer	Cancer	Cancer
2-151 months	Not reported	48% diagnosed within 12 months
Remission had to be achieved but maintenance treatment permitted	Yes	100%

Dolgin (1997)	Houtzager (2001)	Jo (2018)	Kazak (2004)
23	24	17	43
7-17	7-18	7-10	10-20
11.71 (3.02)	11.3 (3.13)	9.18 (not reported)	14
48%	62.5%	47%	Not reported
Not reported	Not reported	Not reported	Not reported
Not reported	Not reported	Not reported	Median household income was in the \$50,000-\$70,000 range
Israel	The Netherlands	South Korea	United States
Cancer	Cancer	Cancer	Cancer
4-38	2-89	Less than 12 months to over 24 months	Not reported
61% on therapy/stable; 17% on therapy/unstable	54.2% in active treatment	Not reported	Completed treatment 1.10-12.16 years prior

Besani (2018)	Besier (2010)	Chung (2000)	Cimini (1986)
17	259	40	30
7-18	4-16	6-17	7-14
10.8 (not reported)	8.6 (3.3)	10.3 (3.01)	9.3 (not reported)
52%	45.6%	50%	50%
Not reported	92.3% German, 4.2% Austrian, 0.8% Turkish, 2.7% Other	Not reported	Not reported
Not reported	Not reported	Not reported	Not reported
Ireland	Germany	Canada	United States
Cancer	22.27% CF, 30.80% CHD, 47.30% Cancer	Cancer	Cancer
2-120	Not reported	1 month – 126 months	At least one year
100%	Not reported	87.5% on treatment	In treatment for at least one year

Table 1. Participant characteristics

First author (Year)	Barrera (2002)	Barrera (2005)	Barrera (2018)
Total Sample Size (N)	17	42	75
Child Age Range (years)	6-17	6-14	7-16
Child Age Mean (DS) (years)	Not reported	9.93 (2.38)	11.05 (2.5)
% female	35%	60%	45%
Ethnicity	Not reported	83% Caucasian, 5% Asian-Canadian, 5% mixed Native Canadian, 2%Asian-Canadian	54% born in Canada, 31% not born in Canada
SES	47% low, 18% medium, 24% high	Majority from middle-class SES backgrounds	22% less than 50,000, 41.3% 50.001-99,999, 36% over \$100,000
Country	Canada	Canada	Canada
Sibling Medical Condition	Cancer	Cancer	Cancer
Time Since Diagnosis (range in months)	Not reported	At least 3 months	Not reported
On treatment	76%	Not reported	46.67%

Intervention characteristics. Intervention studies reported a range of primary objectives including reducing emotional problems, providing support and information, improve quality of life, promoting more effective family functioning and coping, and increasing sibling adjustment. Although several studies did not report a specific theoretical orientation for the intervention, 7 studies reported using cognitive behavioral therapy principles. Further, 4 studies indicated that treatment components were based on family systems theory or family therapy approaches. Study interventionists varied and included psychologists, psychology trainees (i.e., graduate students, residents, and postdoctoral fellows), clinical social workers, and child life specialists. Interventions took place in several settings, including hospitals or medical centers, a camp, an inpatient rehabilitation center, and home-visits. Type of interventions consisted of individual (sibling only), group (with other siblings), and a combination of sibling-only and family-based sessions.

Three groups reported on RCTs in which 1 compared treatment to a waitlist control (Kazak et al., 2004), 1 compared treatment to standard care (Prchal et al., 2012), and 1 compared treatment to an attention control (e.g., minimal intervention where siblings socialized with others and completed activities such as arts and crafts over 8 sessions; Barrera et al., 2018; Barrera, Atenafu, Nathan, Schulte, & Hancock, 2018). Cimini (1986) reported on a waitlist comparison group. Finally, Niemitz and Goldbeck (2018) treatment consisted of rehabilitation clinics plus 5 additional psychoeducational session in comparison to siblings who only participated in the rehabilitation clinics.

Duration of treatment varied from 1 day to 12 weeks with the number of sessions ranging from 1 to 12 sessions. For studies that did report on the length of session, sessions ranged from 50 to 120 minutes. Ten studies reported the use of a treatment manual. Five studies reported

directly involving the parent(s) as part of the intervention. Additionally, 3 studies reported including parents in a peripheral manner, such as providing in-person or phone follow up about the intervention content and to address questions as well as involvement in developing the intervention (e.g., focus group).

Measures of distress included parent and child questionnaires such as the Children's Depression Inventory (Kovacs, 1992), the University of California at Los Angeles Post-Traumatic Stress Disorder Reaction Index (Steinberg, Brymer, Decker, & Pynoos, 2004), and Spence Children's Anxiety Scale (Spence, 1998). Child- and parent-reported measures of adjustment included the Sibling-Perception Questionnaire (Sahler & Carpenter, 1989), PedsQL (Varni, Seid, & Rode, 1999), and the Behavior Assessment System for Children (Reynolds & Kamphaus, 1992). Furthermore, 8 studies reported administering follow-up measures, and follow-up assessment took place between 4 weeks to 7 months post-intervention. See Table 2 for additional intervention characteristics.

Niemitz (2018)	Prechal (2012)	Sidhu (2006)
Combination of individual and family sessions	Individual	Group
Clinics	Hospital	Camp
4 weeks	2 sessions	4 days
1 family session 60 minutes; 4 sibling only groups 30-60 minutes	50 minutes	Not reported
Yes	Yes	Involved in focus groups in the development of the camp and again at end of camp to report on any
Allied psychosocial health professionals	Psychologist	Trained group facilitators
Intervention group received the standard rehabilitation intervention as well as additional psychoeducation (e.g., parent education, medical education) and a family-based psychosocial intervention (e.g., to improve family communication, identifying feelings)	Intervention group received medical information and cognitive behavioral strategies to enhance coping with stressful situation as well as information for parents	Developmentally appropriate activity groups, outdoor adventure challenges, didactic education, therapeutic and social activities

Dolgin (1997)	Houtzager (2001)	Jo (2018)	Kazak (2004)
Group	Group	Individual	Combined siblings only and family sessions
Medical center	Medical center	Home	Hospital
6 weeks	5 sessions	12 weeks	4 sessions in 1 day
Not reported	Not reported	60 minutes	1 day (5 hours of direct therapeutic contact: 2 hours)
No	1 parent session before group started; 1 evaluative session afterward	Not reported	Yes
Clinical social worker, child life specialist, supervising psychologist	2 psychologists	Qualified art psychologist	Psychology trainees (graduate students, residents, postdoctoral fellows), psychologists, nurses, and social workers
Art therapy techniques, role playing, informal social interaction, information regarding cancer and treatment, discussion on illness impact within the family	Discussion of changes, problem-solving skills, enhance feelings of control, discussion of illness-related emotions, medical information (with oncologist), and visit of the oncology unit (with nurse)	Art therapy focused on awareness of self, expression of feelings, relationship with parents and siblings, and hope for the future	Cognitive behavioral principles to reduce distress and develop coping strategies

Besani (2018)	Besier (2010)	Chung (2000)	Cimini (1986)
Combination of individual and family	Combination of individual and family sessions	Group	Group
Pediatric oncology centre	Inpatient rehabilitation	Hospital	Medical center
1 day workshop	4 weeks	8 weeks	3 sessions
Not reported	Not reported	2 hours	Not reported
Yes	Yes	Not directly but followed up via phone and in person	No
Clinical psychologist and play specialist	Not reported	Graduate students and research assistants supervised by licensed psychologist	Graduate student
Problem-solving skills related to worries and fears; narrative techniques to assist with meaning making and emotional processing; medical education and psychoeducation	Tailored to the sibling and family, including 4 (one per week) psychoeducational group session focusing on family situation. On average, 3 exercise sessions, 1 relaxation session, and 1 supportive or psychotherapy session each week. Specific parent-child sessions also offered up to 3 times a week	Information on medical condition, psychosocial foci (e.g., family context, siblings' feelings, school), coping and problem-solving skills	Information about the hospital, pediatric cancer, and different feelings/reaction. Opportunities to ask questions and discuss with the group

Table 2. Study Intervention Characteristics

First author (Year)	Barrera (2002)		Barrera (2005)		Barrera (2018)	
Type of Intervention	Group		Group		Group	
Setting of Treatment	Hospital		Hospital		Hospital	
Duration	8 weeks		8 weeks		8 weeks	
Length of Sessions	2 hours		2 hours		120 minutes	
Parent Involvement	Not directly but followed up via phone and in person		Not reported		Not reported	
Interventionist(s)	Graduate students and research assistants supervised by licensed psychologist		Graduate students and research assistants supervised by licensed psychologist		Graduate students and research assistants, child life specialists, licensed psychologists	
Intervention Components	Medical education and psychoeducation, psychosocial foci (e.g., family context, siblings' feelings, school), cognitive behavioral strategies to enhance coping and problem-solving skills		Medical education and psychoeducation, psychosocial foci (e.g., family context, siblings' feelings, school), cognitive behavioral strategies to enhance coping and problem-solving skills		Medical education and psychoeducation, cognitive behavioral strategies to enhance problem-solving in different areas (e.g., family relationships, fears and feelings), social interactions through games and crafts	

Overall Effect Sizes

The random effects aggregate effect size for adjustment was small but significant ($g = 0.23$; 95% confidence interval [CI] [0.10, 0.36]; see Figure 2 for a forest plot), suggesting that interventions had a small, positive effect on sibling adjustment. Further analysis indicated that there was not significant heterogeneity ($Q = 11.32$, $p = 0.25$, $df = 9$), which may be due to inadequate power.

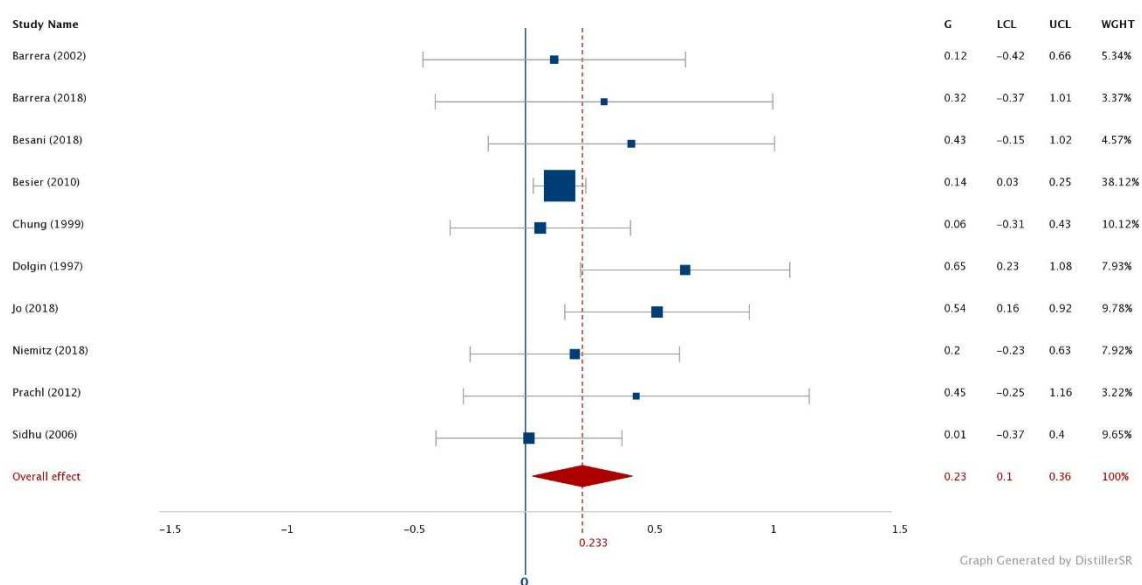


Figure 2. Forest plot of the random effects model for adjustment

Note. G = Hedges' g ; LCL = 95% confidence interval lower limit; UCL = 95% confidence interval upper limit; WGHT = random effects weight and the relative study weight is also represented in the forest plot by the size of the data marker.

Similarly, results indicated a small but significant effect on distress in siblings ($g = 0.48$, 95% CI [0.32, 0.67]; see Figure 3 for a forest plot). Heterogeneity analysis was significant ($Q = 23.03$, $p = 0.03$, $df = 12$), revealing a small to medium amount of heterogeneity ($I^2 = 47.90\%$).

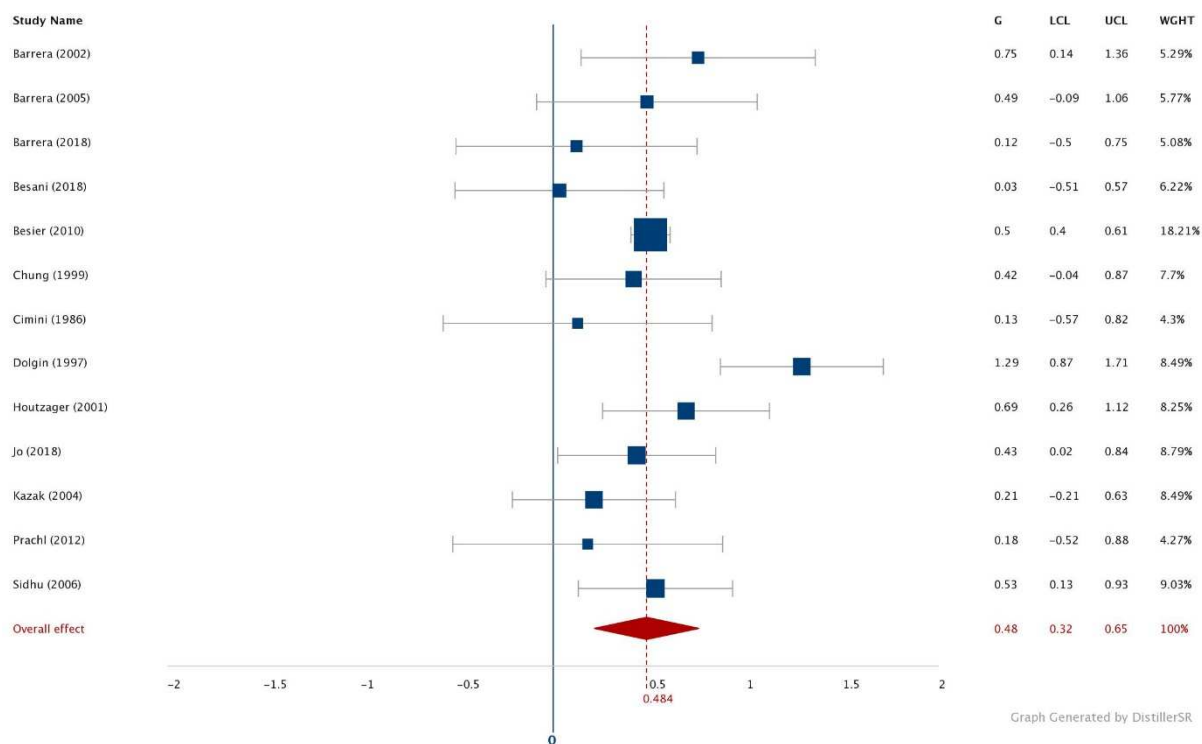


Figure 3. Forest plot of the random effects model for distress

Note. G = Hedges' g ; LCL = 95% confidence interval lower limit; UCL = 95% confidence interval upper limit; WGHT = random effects weight and the relative study weight is also represented in the forest plot by the size of the data marker.

Exploratory Analyses

Although moderator analyses were not possible given the limited number of included studies, exploratory analyses were conducted to examine possible differences in study methodologies (i.e., studies that solely used a pre-test, post-test design versus studies that included a comparison or control group). This approach has been used in a prior meta-analysis where moderator analyses were not possible due to the limited studies that met inclusion criteria (Kichline & Cushing, 2019).

Three studies that included a comparison group provided a measure of adjustment, and results demonstrated an aggregate effect size of $g = 0.36$ (95% CI [0.01, 0.72]), suggesting that these interventions had a small but significant effect on sibling adjustment. However, heterogeneity was not significant ($Q = 0.62, p = 0.73, df = 2$), which may have resulted from the inclusion of very few studies in this exploratory analysis. Four studies reported on distress outcomes and results indicated a small and nonsignificant effect ($g = 0.17; 95\% \text{ CI } [-0.11, 0.46]$) on sibling distress. This finding differed from the overall analysis, which suggested a small and significant effect of interventions on sibling distress. Further analyses revealed no significant heterogeneity ($Q = 0.08, p = 0.99, df = 3$). Again, it is important to view these findings cautiously, as these analyses are exploratory in nature and only include a very limited number of studies.

Regarding studies with pre-post designs, 7 examined adjustment and findings revealed a small but significant effect ($g = 0.24; 95\% \text{ CI } [0.07, 0.42]$), suggesting that pre-post design interventions had a small, positive effect on sibling adjustment. Further analyses revealed no significant heterogeneity ($Q = 10.61, p = 0.10, df = 6$). Additionally, 9 pre-post studies examined distress. Results indicated a medium and significant effect ($g = 0.57; 95\% \text{ CI } [0.38, 0.76]$) on sibling distress, as measured by pre-post intervention designs. Findings also demonstrated significant heterogeneity ($Q = 17.397; p = 0.03; df = 8$), revealing a medium amount of heterogeneity ($I^2 = 53.99\%$), which was consistent with analysis from all of the intervention studies.

Risk of Bias

The majority of studies was coded as having potentially high risk of bias regarding random sequence generation (71%) and blinding of participants and personnel (85%), which is

consistent with the high number of uncontrolled trials within the current analyses. Sixty-four percent of studies were rated low on attrition bias. Similarly, 78% demonstrated low reporting bias, with 2 studies (14%) rated as unclear and 1 study rated as high (2%). See Figure 4 for representation of risk of bias across studies.

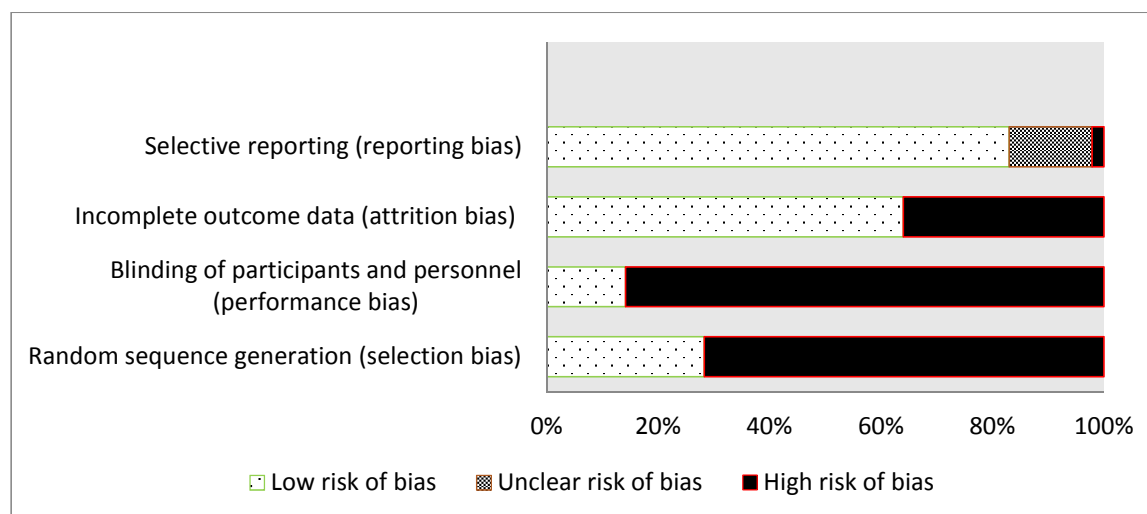


Figure 4. Representation of risk of bias across studies

Publication Bias

A modified fail-safe N equation proposed by Orwin (1983) was used to examine publication bias, and the formula identified the number of studies with null findings that would need to exist to reduce the overall mean effect size for each outcome. For instance, based on the overall aggregate effect size for distress, 19 studies with an average effect size of 0 would need to exist to reduce the current findings to an effect size of 0.2, whereas 50 studies would be needed to reduce the effect size to 0.1. In terms of the overall effect size for adjustment, approximately 13 undiscovered studies would be needed to reduce the effect size to 0.1.

Fail-safe Ns were also calculated for results of the exploratory analyses. To reduce the effect size to 0.1 for pre-post intervention studies, 10 studies reporting on adjustment and 42 studies reporting on distress (17 to reduce to an effect size of 0.2) would need to be discovered.

With regards to interventions with a comparison group, 8 studies with null adjustment results and only 3 with null distress results would be needed to reduce the effect size to 0.1.

Discussion

The purpose of the current meta-analysis was to quantify the effectiveness of interventions on improving adjustment and distress outcomes for siblings of youth with chronic medical conditions. The present study sought to provide an updated review on interventions for siblings as well as extend the literature by focusing on interventions specifically for siblings of youth with chronic medical conditions. Aggregate effect sizes on the complete sample suggest small but significant effect sizes of interventions on sibling adjustment ($g = 0.23$) and distress ($g = 0.48$). These findings should be viewed cautiously in light of several limitations, including high number of pre-post intervention designs and increased risk of bias. Results from exploratory analyses, which focused on study methodology (i.e., pre-post design; treatment versus comparison group), varied such that pre-post design studies yielded a small effect on adjustment but a medium effect on sibling distress. Findings from studies with a comparison group suggested a small but significant effect on adjustment; findings were small and not significant for distress. There is a large difference in the effect of distress depending on the intervention design. It is plausible that pre-post designs overestimate the effect of distress, whereas interventions using a RCT design suggest that distress is less impacted by interventions. However, results from the exploratory analyses should be viewed cautiously given the small number of studies included in each analysis, particularly in the analyses examining the use of a control or comparison group.

Regarding prior reviews, there was some overlap between included articles (e.g., 3 to 7 articles across the reviews), and overall findings of the present meta-analysis are consistent with results from prior reviews. For instance, a recent meta-analysis that examined interventions for

both parents and siblings of youth diagnosed with cancer similarly demonstrated small effect sizes for anxiety symptoms and problem-solving skills (Sánchez-Ega et al., 2019). Another recent meta-analysis on siblings of youth with chronic medical conditions and developmental disabilities by Smith and colleagues (2019) demonstrated a small and significant effect of interventions on sibling internalizing symptoms, which is consistent with the overall aggregate effect size of distress in the present meta-analysis. For interventions specifically for siblings of children diagnosed with cancer, Prchal and Landolt (2009) provided individual study effect sizes when feasible. In some areas, such as anxiety symptoms ($d = 0.36-0.98$) and quality of life ($d = 0.22-1.45$), effect sizes varied from small to large. This result is contradictory to the present meta-analysis which generally demonstrated small aggregate effect sizes.

A strength of the current meta-analysis is the inclusion of broad outcomes of sibling adjustment and distress, which permitted the ability to examine several similar types of measures while also limiting the number of outcomes examined. This approach builds on current reviews, which primarily focused on internalizing and externalizing symptoms. The examination of broader sibling outcomes, such as adjustment and distress, may be beneficial because the majority of siblings do not display clinically significant concerns of psychopathology, but are still at risk for range of negative outcomes, such as poor quality of life, school difficulties, and changes in family functioning (e.g., Alderfer et al., 2010; Gerhardt et al., 2015). Furthermore, examining the effectiveness of these interventions is particularly important given recent guidelines promoting supports and interventions for siblings of youth with chronic medical conditions (e.g., DeMaso et al., 2009; Gerhardt et al., 2015).

It can be useful to convert effect sizes of intervention studies into clinically meaningful units, or number needed to treat (NNT), which quantifies the number of patients a clinician can

expect to treat before obtaining favorable outcomes in one patient in comparison to those in a control group (e.g., Citrome, 2008; Magnusson, 2014). Based on the overall aggregate effect sizes, the NNT is 6.01 for sibling distress and 16.51 for adjustment. However, according to findings from the exploratory analyses from studies using a comparison group, a larger volume of siblings would be needed to be treated before seeing superior improvements in adjustment (10.63 siblings) and in distress (34.3 siblings).

In addition to NNT, Magnusson (2014) provides other metrics to assist in the interpretation of effect sizes. For instance, Cohen's U_3 provides the percentage "of the treatment group [that] will be above the mean of the control group" (Magnusson, 2014). Given the overall aggregate effect sizes, this percentage would be 58% for adjustment and 66% for distress. Furthermore, findings suggest that there is 84.15% to 92.03 overlap between treatment and control groups for distress and adjustment, respectively. Magnusson (2014) also provides the probability of superiority, which provides the "chance a person picked at random from the treatment group will have a higher score than a person picked at random from the control group." For the present findings, the probability of superiority for distress is 61.14%, whereas adjustment is 55.62%. Taken together, these findings suggest challenges with treatment outcomes; limitations of the current literature are presented below.

Limitations

One limitation of the current meta-analysis is the small number of studies that met inclusion criteria, such that there were only 15 articles across 14 studies. While analyses revealed significant heterogeneity in the aggregate effect size for distress, due to the limited number of included studies, potential moderator analyses of treatment effects could not be examined. Additionally, the limited number of studies in the current analyses may have reduced power to

detect significant heterogeneity results for the adjustment outcome. In the future, with additional intervention studies, moderator analyses could provide information about potential unique factors that contribute to improving outcomes in sibling interventions.

The current findings are limited by high risk of bias, particularly in the areas of random sequence generation (i.e., selection bias) and blinding of participants and personnel (i.e., performance bias). The use of RCTs could potentially decrease risk of bias by implementing more rigorous methodologies, including randomizing participants into treatment or control groups as well as blinding participants and staff. Regarding potential publication bias, it is important to note that fail-safe N s ranged from 13 to 50 for the overall aggregate effect sizes for all study designs as well as 3 to 42 for exploratory analyses focusing on either pre-post or comparison group intervention design. Given the comprehensiveness of the literature search, it is less likely that as many as 13 studies with average effect sizes of 0 were missing from inclusion in the present meta-analysis. However, only 3 studies with average effect sizes of 0 related to distress outcomes would be needed to overturn the exploratory analyses focusing on comparison groups. This appears less of a far reach given that two unpublished dissertations were discovered and included within the current meta-analyses.

Another potential limitation is the generalizability of the included interventions. With the exception of Besier (2010), which included siblings of youth diagnosed with CHD, CF, and cancer, the other interventions solely consisted of siblings of youth diagnosed with cancer. Although findings suggest that interventions have a small but significant impact on outcomes for siblings of youth diagnosed with cancer, there is limited evidence to suggest that these findings generalize to siblings impacted by other chronic medical conditions. Core components of these interventions (e.g., cognitive-behavioral strategies to enhance problem solving; social support

from peers) plausibly could be used with siblings with tailoring of other, more cancer-specific components (e.g., information regarding the medical condition). For instance, Besier and colleagues (2010) reported tailored treatment to the sibling and family, and components consisted of relaxation strategies, psychoeducational sessions, and parent-child therapy appointments. However, additional research is needed to provide evidence. Furthermore, in light of these limitations, several considerations for future studies are provided below.

Future Directions

Tiered approach to sibling interventions. First, it may be beneficial to use the Pediatric Psychological Preventative Health Model (PPPHM; Kazak, 2006) when developing and examining sibling interventions. This three-tiered conceptual model is rooted in socioecological and public health frameworks, and in terms of applying the approach to siblings specifically, is designed to be applied to every sibling entering the health care system. Therefore, each sibling's psychosocial needs and risks would be assessed and then the results of the assessment would determine which tier the individual fits in, and thus, which services are appropriate for that individual (i.e., universal, targeted, or clinical/treatment; see Kazak, 2006 for a figure). The model provides tailoring of sibling supports given risk and treatment needs, rather than providing a "one-size-fits all" approach.

At the base of the pyramid is the Universal group, such that it consists of the largest group of people and denotes that families, including siblings, "are distressed but resilient" (Kazak, 2006, p. 385). In this way, siblings in this category would likely benefit from general information and support. For siblings in this tier, the development and evaluation of psychoeducational materials, such as handouts or booklets, may be useful. For instance, Oberoi and colleagues (2019) described a community-academic partnership that engaged sibling

stakeholders to determine unmet support needs. These data then informed the revision of existing support resources, including mailed materials consisting of age-appropriate content (e.g., sibling stories, coloring activities).

On the second tier of the pyramid is the Targeted group, which represents targeted care for siblings “displaying acute distress as well as presenting with risk factors” (Kazak, 2006, p. 385). It will be beneficial to determine which sibling interventions could be administered to siblings falling in this Targeted group. Perhaps a brief one to two session intervention would be useful at addressing concerns in this group. Finally, the top of the pyramid consists of siblings in the Clinical/Treatment category, such that they demonstrate “high risk for ongoing distress” (e.g., elevated symptoms of anxiety or depression; Kazak, 2006, p. 385). Perhaps siblings in this group would benefit from a longer, more intensive treatment that is similar to traditional cognitive-behavioral therapy. Overall, additional research could help determine the most appropriate supports for each level.

Taking this approach, psychologists would most likely see siblings in the Clinical/Treatment group, who are at the top of the pyramid and at highest risk. Qualitative findings from the present meta-analysis demonstrated that the interventionists were from multiple disciplines, including psychology, child life, and social work; however, psychology was the most predominant discipline reported. Within a hospital setting, it is common for siblings to receive services from providers such as child life, social work, art therapists, and music therapists. Psychologists might partner with other psychosocial providers to further evaluate and build upon existing sibling programs and services, which may not be fully represented in the literature at present. Additionally, it will be important for future research to target siblings presenting with significant distress in interventions conducted by psychology providers.

Sibling psychosocial screening. Furthermore, psychosocial screening may help to more consistently monitor and identify sibling risk, as well as appropriately place siblings into treatment categories. For instance, not every sibling may need intensive services offered in the “Clinical/Treatment” but may benefit from “Targeted” services. For instance, it may be beneficial to use a psychosocial screener such as the Psychological Assessment Tool (PAT; Kazak et al., 2018). Indeed, one study in the current meta-analysis (Besani et al., 2018) reported using the PAT; however, it was not necessarily used to categorize siblings into specific treatment categories but rather as an outcome measure of sibling psychosocial risk. While the PAT was initially designed to be used within pediatric oncology (Kazak et al., 2011), it was designed to be generalizable to other illness groups (Kazak et al., 2017) and has been adapted for use with several pediatric illness populations, such as congenital heart disease (Hearps et al., 2014), chronic pain (Woods & Ostrowski-Delahanty, 2017), organ transplantation (Pai, Tackett, Ittenbach, & Goebel, 2012), and sickle cell disease (Reader et al., 2017). The PAT includes a sibling problems subscale, which assess for problems exhibited by sibling, requesting that the parent note if it is an issue for a sibling by check “yes” or “no” to each item (Pai et al., 2008). The current version also includes items to assess for sibling aggression and suicidal ideation as well as to determine if the sibling is currently prescribed medication for behavioral concerns (Kazak et al., 2018).

Expanding upon the PAT, a sibling-specific screening module was recently developed (Long, Pariseau et al., 2018). Although the sibling psychosocial risk screener retained many of the items from the sibling problem subscale of the PAT, it also allows parents to answer questions about each sibling, includes new items (e.g., reactions to cancer, social-ecological factors), and takes into account time since diagnosis. Specifically, the measure includes an initial

screeners to be administered at the time of diagnosis as well as a follow-up screener for 3 or more months after diagnosis (Long, Pariseau et al., 2018). Similar to previous adaptations to the PAT, it would likely be important to expand beyond pediatric cancer and further examine the use of similar sibling-specific screening modules for siblings of youth with a range of medical conditions.

Implementing such sibling psychosocial screeners are not without challenges. For instance, access to healthy siblings can be difficult, as they are not frequently presenting to the hospital or doctor's visit with the child who is diagnosed with an illness (Gerhardt et al., 2015). Because of this "invisibility," members of the medical team may not check in about their psychosocial functioning. Further, health care professionals may become aware of sibling psychosocial functioning only after parents verbally report significantly elevated concerns (Franklin, Patterson, Allison, Rosso-Buckton, & Walczak, 2018). Further, many of the existing screeners rely on parent-report of concerns, and sibling-report is often not captured. Another concern is what to do with the information obtained from the sibling specific screener when the child with a chronic illness is the identified patient at the hospital. For instance, would the sibling need to have their own electronic medical record, and how would services be billed? Sibling challenges may occur at different time points; therefore, how can providers adequately monitor siblings? In addition, a potential concern with the routine screenings is what does the team do if a child does screen positive? Thus, it may be important to have established interventions in place for the sibling to access as well as a protocol if safety concerns arise (e.g., social work engaging in safety planning for endorsed suicidal ideation). Screening could take place in other settings where siblings have a greater presence, such as school or primary care. Teachers and primary care physicians could monitor concerns such as high school absenteeism, decline in grades,

and/or withdrawal from peer activities. The field would benefit from further problem-solving around these challenges and potential barriers to implementing routine screenings. Overall, routine sibling psychosocial screenings could serve as a preventative approach and a way to potentially better identify sibling needs and concerns.

Additional measures. If the field is using a tiered treatment approach proposed by the PPPHM, additional outcome measures could be considered. As noted by the model and consistent with the sibling literature, a smaller portion of siblings are meeting criteria for psychopathology; therefore, the majority of siblings would likely fall into the “Universal” and “Targeted” categories. However, the majority of the studies included within the current review primarily examined psychosocial distress, including anxiety and depressive symptoms. Specifically, several studies in the current review reported that siblings’ scores at baseline were within normal limits; therefore, there was little room for improvement from the intervention given ceiling effects. These findings are also consistent with studies demonstrating that siblings of youth diagnosed with cancer fall within normal limits on measures of anxiety and depression, for example (e.g., Long, Lehmann et al., 2018). This is not to say that these outcomes are not important to monitor and target, especially given that these are going to be elevated for a subset of siblings (e.g., approximately one-fourth of siblings of youth with cancer meet criteria for PTSD; Long, Lehmann et al., 2018). However, it may be equally important to consider other, possibly more meaningful and sensitive, measures that would be relevant to the majority of siblings.

As suggested by Barrera and colleagues (2018), studies may need to utilize other measures of internalizing and/or externalizing symptoms that would be better able to detect subtle subclinical changes. Moving away from internalizing symptoms, assessing the

psychosocial unmet needs of siblings, such as the Sibling Cancer Needs Instrument (SCNI; Patterson et al., 2014), could be useful in determining which services would be most appropriate. Based on findings from a prior systematic review on siblings of youth with cancer (Long, Lehmann et al., 2018), professionals may want to target and examine other areas of functioning, such as social support and family functioning.

Intervention design. As previously mentioned, the majority of studies included in the present meta-analysis employed pre-post designs. This situation limits the ability to determine if observed outcomes are due to the intervention or rather occur merely due to the passage of time. According to established guidelines (e.g., Chambless & Hollon, 1998; American Psychological Association Task Force on Psychological Intervention Guidelines, 1995), RCTs are often considered to be the gold standard in determining the efficacy and effectiveness of interventions. Furthermore, RCT intervention findings need to be replicated by at least two independent teams (Chambless & Hollon, 1998). Although providing a rigorous methodology, RCTs are not without challenges, including the need for large sample sizes. This necessity can be particularly concerning for sibling research in which it can be difficult to recruit and retain participants. To overcome this issue, researchers may consider multi-site collaborations to increase sample sizes. Additionally, clinical researchers will want to consider the use of adaptive interventions, such as sequential multiple assignment randomized trials, which can help to answer several remaining questions, including optimal timing, type, and duration of supports for siblings (Noser, Cushing, McGrady, Amaro, & Huffhines, 2017).

Intervention settings. Several studies reported challenges with recruitment due to a variety of reasons, including scheduling conflicts and transportation issues (e.g., Barrera et al., 2018; Besani et al., 2018). Because the majority of studies within the current meta-analysis

reported intervention delivery in a hospital-based setting, future studies should examine other settings for intervention implementation for potential effectiveness. For instance, pediatric psychologists are well-suited to provide interventions within primary care, which is the medical home where the majority of youth present for care (e.g., Stancin & Perrin, 2014; Stancin, Sturm, & Ramirez, 2014). Interventions for youth with chronic illnesses have been developed for use in primary care (e.g., Mitchell, Amaro, & Steele, 2016), and interventions for siblings could also be developed and implemented within this setting. Primary care services have been shown to reduce stigma often associated with behavioral health services (Stancin, Sturm et al., 2014) and may reduce barriers to access that may be encountered by bringing a sibling to a specialty clinic at an academic medical setting.

Another way to address these concerns is through the use of eHealth (electronic) and mHealth (mobile) technologies. Broadly defined, eHealth interventions use technology to improve the delivery of care for youth and families (Palermo & Wilson, 2009). There are several benefits for the use of eHealth interventions, such as reducing barriers to treatment by offering more flexibility, permitting families to access the intervention at a more convenient time and location, which might minimize challenges with study recruitment and attrition (Canter, Christofferson, Scialla, & Kazak, 2019; Cushing, 2017). Prior eHealth interventions for families of youth with chronic illnesses have used a mix of designs, such that some interventions have only been delivered online whereas others have also included telemedicine sessions with an interventionist (Canter et al., 2019). Researchers developing interventions for siblings may consider using eHealth technologies. For instance, content could be made available online or a number of sessions could be conducted with an interventionist via telemedicine. Siblings have also reported the need to connect with other siblings who share their experiences (Long,

Lehmann et al., 2018). Consequently, researchers using eHealth technologies could consider an option for videoconferencing or additional in-person opportunities to facilitate a group.

Other sibling populations. Some interventions have been developed for both siblings of youth with chronic medical conditions (in addition to pediatric cancer) and siblings of youth with developmental disabilities (e.g., Lobato & Kao, 2002, 2005). However, there is not consensus in the literature about whether a single intervention is beneficial for all siblings or if intervention components need to be condition specific (e.g., Smith et al., 2018; Tudor & Lerner, 2015). For instance, there may be benefits in providing a universal intervention for a siblings of both youth with chronic illnesses and developmental disabilities. For instance, a portion of siblings of youth with disabilities may experience emotional and behavioral difficulties (e.g., Giallo, Gavidia-Payne, Minette, & Kapoor, 2012; Rossiter & Sharpe, 2001), and it is possible that siblings of youth with various diagnoses may benefit from the same treatment components (e.g., social support through a group format, problem-solving skills). However, it is also possible that separate interventions are required, as siblings might present with unique challenges and needs depending on their siblings' condition. For example, siblings of youth with developmental disabilities may be faced with issues related to the quality of the sibling relationship (e.g., Stoneman, 2005), whereas siblings of youth with chronic illnesses may need to manage recurrent doctor visits and hospitalizations as well as potential concerns with life-threatening illnesses (e.g., Sharpe & Rossiter, 2002). Therefore, interventions targeting siblings of youth with medical conditions potentially may need to incorporate psychoeducation on hospitalizations and the medical condition(s). Similarly, other treatment modifications could be necessary for siblings of youth with developmental disabilities and/or other mental health conditions. Overall, the field

would benefit from clarity on this issue to determine the need for a single intervention versus distinct interventions to adequately address the needs of siblings.

Looking at chronic medical conditions specifically, the sibling interventions included within the present meta-analysis were predominantly focused on siblings of youth diagnosed with cancer. This situation is not surprising given that oncology has been a consistent focus of research within pediatric psychology (Canter et al., 2018). In comparison to other illness populations, pediatric oncology may also receive additional grant funding, which could impact research efforts and the development of sibling interventions in other populations where funding may be more sparse. However, the prior literature demonstrates that there are also considerable adjustment difficulties in siblings of youth with other medical conditions, such as diabetes and seizures (e.g., Vermaes et al., 2012). Furthermore, other chronic illnesses can be life-threatening and require several hospitalizations and surgeries, which can impact families and siblings. For instance, siblings of youth diagnosed with hypoplastic left heart syndrome, which is a form of congenital heart disease, have demonstrated poor sibling adjustment, and interventions for siblings and families may be beneficial (Caris et al., 2018). Therefore, future studies should further explore the effectiveness of interventions for siblings of other chronic medical conditions.

Even within pediatric oncology, sibling supports are less frequently implemented in comparison to the other Pediatric Oncology Psychosocial Standards of Care (Scialla et al., 2018). Future clinical researchers will want to identify possible barriers that reduce or prevent institutional adherence to the Sibling Standard of Psychosocial Care, including the provision of sibling support services (Gerhardt et al., 2015). Additional advocacy efforts may be essential to fully integrate siblings of youth with chronic medical conditions into family-centered care so that sibling needs are also adequately addressed (Shelton, 1999).

Conclusions

As with other areas where insufficient research has been conducted to understand a pediatric psychology situation, so too does the area of siblings require further development of conceptualizations and empirical investigation. The present meta-analysis provides an initial examination of interventions targeting sibling adjustment and distress. The present findings should be viewed cautiously in light of the small number of included studies, large use of pre-post design methodology, and potential high risk of bias. Approximately one-third of the studies included were published within the last year, possibly demonstrating recent growth within sibling research. As interventions continue to be developed for siblings of youth with chronic medical conditions, employing more methodologically strong designs (e.g., RCTs, larger sample sizes, examination of long-term outcomes) that also consider a range of additional factors (e.g., potential use of different measures, inclusion of other chronic medical populations) will be especially important.

References

References marked with an asterisk indicate studies included in the meta-analysis.

Alderfer, M. A., Long, K. A., Lown, E. A., Marsland, A. L., Ostrowski, N. L., Hock, J.

M., & Ering, L. J. (2010). Psychosocial adjustment of siblings of children with cancer: A systematic review. *Psycho-Oncology, 19*, 789-805.

American Psychological Association Task Force on Psychological Intervention

Guidelines. (1995). *Template for developing guidelines: Interventions for mental disorders and psychological aspects of physical disorders*. Washington, DC: American Psychological Association.

*Barrera, M., Atenafu, E., Nathan, P. C., Schulte, F., & Hancock, K. (2018). Depression and quality of life in siblings of children with cancer after group intervention participation: A randomized control trial. *Journal of Pediatric Psychology, 43*, 1093-1103.

*Barrera, M., Atenafu, E. G., Schulte, F., Nathan, P. C., Hancock, K., & Saleh, A. (2018). A randomized controlled trial of a group intervention for siblings of children with cancer: Changes in symptoms of anxiety in siblings and caregivers. *Psycho-Oncology, 27*, 1629-1634.

*Barrera, M., Chung, J. Y. Y., & Fleming, C. F. (2005). A group intervention for siblings of pediatric cancer patients. *Journal of Psychosocial Oncology, 22*, 21-39.

*Barrera, M., Chung, J. Y. Y., Greenberg, M., & Fleming, C. (2002). Preliminary investigation of a group intervention for siblings of pediatric cancer patients. *Children's Health Care, 31*, 131-142.

Batte, S., Watson, A. R., & Amess, K. (2006). The effects of chronic renal failure on

- siblings. *Pediatric Nephrology*, 21, 246-250.
- Bellin, M. H., Bentley, K. J., & Sawin, K. J. (2009). Factors associated with the psychological and behavioral adjustment of siblings of youths with spina bifida. *Families, Systems, and Health*, 27, 1-15.
- *Besani, C., McCusker, C., Higgins, A., & McCarthy, A. (2018). A family-based intervention to promote adjustment in siblings of children with cancer: A pilot study. *Psycho-Oncology*, 27, 2052-2055.
- *Besier, T., Holling, H., Schlack, R., West, C., & Goldbeck, L. (2010). Impact of a family-oriented rehabilitation programme on behavioural and emotional problems in health siblings of chronically ill children. *Child: Care, Health and Development*, 36, 686-695.
- Bronfenbrenner, U. (1979). *The ecology of human development*. Cambridge, MA: Harvard University of Press.
- Caes, L., Goubert, L., Devos, P., Verlooy, J., Benoit, Y., & Vervoort, T. (2014). The relationship between parental catastrophizing about child pain and distress in response to medical procedures in the context of childhood cancer treatment: A longitudinal analysis. *Journal of Pediatric Psychology*, 39, 677-686.
- Canter, K. S., Amaro, C. M., Noser, A. E., & Roberts, M. C. (2018). Historical Analysis: The *Journal of Pediatric Psychology* from 1976 to 2015. *Journal of Pediatric Psychology*, 43, 21-30.
- Canter, K. S., Christofferson, J., Scialla, M. A., & Kazak, A. E. (2019). Technology-focused family interventions in pediatric chronic illness: A systematic review. *Journal of Clinical Psychology in Medical Settings*, 26, 68-87.

- Card, N. A. (2012). *Applied meta-analysis for social science research*. New York: Guilford Press.
- Caris, E. C., Dempster, N., Wernovsky, G., Miao, Y., Moore-Clingenpeel, M., Neely, T., ... Cua, C. L. (2018). Perception scores of siblings and parents of children with hypoplastic left heart syndrome. *Congenital Heart Disease, 13*, 528-532.
- *Cimini, M. D. (1986). *An examination of behavior problems in the siblings of pediatric cancer patients and an evaluation of an information/support group* (Doctoral dissertation). Retrieved from ProQuest Dissertations and Theses. (303449654).
- Citrome, L. (2008). Compelling or irrelevant? Using number needed to treat can help decide. *Acta Psychiatrica Scandinavica, 117*, 412-419.
- Chambless, D. L., & Hollon, S. D. (1998). Defining empirically supported therapies. *Journal of Consulting and Clinical Psychology, 66*, 7-18.
- *Chung, J. Y. Y. (2000). *Siblings of children with cancer: Opening the opportunities for participation* (Doctoral dissertation). Retrieved from ProQuest Dissertations and Theses. (304589818).
- Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). Hillsdale, NJ: Erlbaum.
- Compas, B. E., Jaser, S. S., Reeslund, K., Patel, N., & Yarboi, J. (2017). Neurocognitive deficits in children with chronic health conditions. *American Psychologist, 72*, 326-338.
- Comprehensive Meta-Analysis (Version 3) [Computer software]. (2017). Englewood, NJ: Biostat.
- Cushing, C. C. (2017). eHealth applications in pediatric psychology. In M. C. Roberts &

- R. G. Steele (Eds.), *Handbook of pediatric psychology* (5th ed., pp. 201-211). New York: Guilford Press.
- Deavin, A., Greasley, P., & Dixon, C. (2018). Children's perspectives on living with a sibling with a chronic illness. *Pediatrics*, *142*, 142(2), e20174151.
- DeMaso, D. R., Martini, D. R., & Cahen, L. A. (2009). Practice parameter for the psychiatric assessment and management of physically ill children and adolescents. *Journal of the American Academy of Child & Adolescent Psychiatry*, *48*, 213-233.
- *Dolgin, M. J., Somer, E., Zaidel, N., & Zaizov, R. (1997). A structured group intervention for siblings of children with cancer. *Journal of Child and Adolescent Group Therapy*, *7*, 3-18.
- Drotar, D., & Crawford, P. (1985). Psychological adaptation of siblings of chronically ill children: Research and practice implications. *Journal of Developmental and Behavioral Pediatrics*, *6*, 355-361.
- Durlak, J. A. (2009). How to select, calculate, and interpret effect sizes. *Journal of Pediatric Psychology*, *34*, 917-928.
- Franklin, M., Patterson, P., Allison, K. R., Rosso-Buckton, A., & Walczak, A. (2018). An invisible patient: Healthcare professionals' perspectives on caring for adolescents and young adults who have a sibling with cancer. *European Journal of Cancer Care*, *27*, e12970.
- French, A. E., Tsangaris, E., Barrera, M., Guger, S., Brown, R.,...Nathan, P. C. (2013). School attendance in childhood cancer survivors and their siblings. *The Journal of Pediatrics*, *162*, 160-165.
- Friedman, D., Holmbeck, G. N., Jandasek, B., Zukerman, J., & Abad, M. (2004). Parental

- functioning in families of preadolescents with spina bifida: Longitudinal implications for child adjustment. *Journal of Family Psychology*, *18*, 609-619.
- Gerhardt, C. A., Lehmann, V., Long, K. A., & Alderfer, M. A. (2015). Supporting siblings as a standard of care in pediatric oncology. *Pediatric Blood & Cancer*, *62*, S750-S804.
- Giallo, R., Gavidia-Payne, S., Minett, B., & Kapoor, A. (2012). Sibling voices: The self-reported mental health of siblings of children with a disability. *Clinical Psychologist*, *16*, 36-43.
- Hartling, L., Milne, A., Tjosvold, L., Wrightson, D., Gallivan, J., & Newton, A. S. (2014). A systematic review of interventions to support siblings of children with chronic illness or disability. *Journal of Paediatrics and Child Health*, *50*, E26-E38.
- Hearps, S. J., McCarthy, M. C., Muscara, F., Hearps, S. J., Burke, K., Jones, B., & Anderson, V. A. (2014). Psychosocial risk in families of infants undergoing surgery for a serious congenital heart disease. *Cardiology in the Young*, *24*, 632-639.
- Higgins, J. P. T., Altman, D. G., & Sterne J. A. C. (2011). Chapter 8: Assessing risk of bias in included studies. In J. P. T. Higgins & S. Green (Eds.), *Cochrane Handbook for Systematic Reviews of Interventions* Version 5.1.0. Retrieved from www.handbook.cochrane.org.
- Hommel, K. A., Ramsey, R. R., Loiselle, K., & Ryan, J. L. (2017). Adherence to pediatric treatment regimens. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of pediatric psychology* (5th ed., pp. 119-133). New York: Guilford Press.
- *Houtzager, B. A., Grootenhuis, M. A., & Last, B. F. (2001). Supportive groups for

siblings of pediatric oncology patients: Impact on anxiety. *Psycho-Oncology*, *10*, 315-324.

Huedo-Medina, T. B., Sánchez-Meca, J., Marín-Martínez, F., & Botella, J. (2006).

Assessing heterogeneity in meta-analysis: Q statistic or I^2 index? *Psychological Methods*, *11*, 193–206.

*Jo, M. J., Hong, S., & Park, H. R. (2018). Effects of art intervention program for siblings of children with cancer: A pilot study. *Journal of Pediatric Oncology Nursing*, *35*, 178-187.

Kahana, S., Drotar, D., & Frazier, T. (2008). Meta-analysis of psychological interventions to promote adherence to treatment in pediatric chronic health conditions. *Journal of Pediatric Psychology*, *33*, 590-611.

Kazak, A. E. (1997). A contextual family/systems approach to pediatric psychology: Introduction to the special issue. *Journal of Pediatric Psychology*, *22*, 141-148.

Kazak, A. E. (2006). Pediatric psychosocial preventative health model (PPPHM): Research, practice, and collaboration in pediatric family systems medicine. *Families, Systems, & Health*, *24*, 381-395.

Kazak, A. E., Alderfer, M. A., & Reader, S. K. (2017). Families and other systems in pediatric psychology. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of pediatric psychology* (5th ed., pp. 566-579). New York: Guilford Press.

*Kazak, A. E., Alderfer, M. A., Streisand, R., Simms, S., Rourke, M. T., Barakat, L. P., ..

Gallagher, P., & Cnaan, A. (2004). Treatment of posttraumatic stress symptoms in adolescent survivors of childhood cancer and their families: A randomized clinical trial. *Journal of Family Psychology*, *18*, 493-504.

- Kazak, A., Barakat, L., Ditaranto, S., Biros, D., Hwang, W. T., Beele, D., ... Reilly, A., (2011). Screening for psychosocial risk at cancer diagnosis: The Psychosocial Assessment Tool (PAT). *Journal of Pediatric Hematology and Oncology*, 33, 289-294.
- Kazak, A. E., Hwang, W. T., Chen, F. F., Askins, M. A., Carlson, O., Argueta-Ortiz, F., & Barakat, L. P. (2018). Screening for family psychosocial risk in pediatric cancer: Validation of the Psychosocial Assessment Tool (PAT) Version 3. *Journal of Pediatric Psychology*, 43, 737-748.
- Kazak, A. E., Rourke, M. T., & Navsaria, N. (2009). Families and other systems in pediatric psychology. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of pediatric psychology* (4th ed., pp. 656-671). New York: Guilford Press.
- Kichline, T., & Cushing, C. C. (2019). A systematic review and quantitative analysis on the impact of aerobic exercise on pain intensity in children with chronic pain. *Children's Health Care*, 48, 244-261.
- Knecht, C., Hellmers, C., & Metzging, S. (2015). The perspective of siblings of children with chronic illness: A literature review. *Journal of Pediatric Nursing*, 30, 102-116.
- Kovacs, M. (1992). *Children's depression inventory*. North Tonawanda, NY: Multi-Health Systems, Inc.
- Law, E. F., Fisher, E., Fales, J., Noel, M., & Eccleston, C. (2014). Systematic review and meta-analysis of parent and family-based interventions for children and adolescents with chronic medical conditions. *Journal of Pediatric Psychology*, 39, 866-886.
- Lipsey, M. W., & Wilson, D. (2001). *Practical meta-analysis*. Thousand Oaks, CA: Sage.
- Lobato, D., & Kao, B. T. (2002). Integrated sibling-parent group intervention to improve

- sibling knowledge and adjustment to chronic illness and disability. *Journal of Pediatric Psychology*, 8, 711-716.
- Lobato, D., & Kao, B. T. (2005). Brief report: Family-based group intervention for young siblings of children with chronic illness and developmental disability. *Journal of Pediatric Psychology*, 30, 678-682.
- Long, K. A., Lehmann, V., Gerhardt, C. A., Carpenter, A. L., Marsland, A. L., & Alderfer, M. A. (2018). Psychosocial functioning and risk factors among siblings of children with cancer: An updated systematic review. *Psycho-Oncology*, 17, 1467-1479.
- Long, K. A., Marsland, A. L., & Alderfer, M. A. (2013). Cumulative family risk predicts sibling adjustment to childhood cancer. *Cancer*, 119, 2503-2510.
- Long, K. A., Pariseau, E. M., Muriel, A. C., Chu, A., Kazak, A. E., & Alderfer, M. A. (2018). Development of a psychosocial risk screener for siblings of children with cancer: Incorporating the perspectives of parents. *Journal of Pediatric Psychology*, 43, 693-701.
- Mackner, L. M., Sisson, D. P., & Crandall, W. V. (2004). Review: Psychosocial issues in pediatric inflammatory bowel disease. *Journal of Pediatric Psychology*, 29, 243-257.
- Magnusson, K. (2014). Interpreting Cohen's d effect size: An interactive visualization. Retrieved from <http://rpsychologist.com/d3/cohend/>
- Mitchell, T. B., Amaro, C. M., & Steele, R. G. (2016). Pediatric weight management interventions in primary care settings: A meta-analysis. *Health Psychology*, 35, 704-713.
- Moher, D., Liberati, A., Tetzlaff, J., Altman, D. G., & The PRISMA Group. (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Medicine*, 6(6), e1000097.
- Murray, J. S. (1999). Siblings of children with cancer: A review of the literature. *Journal*

of Pediatric Oncology Nursing, 16, 25-34.

- *Niemitz, N., & Goldbeck, L. (2018). Outcomes of an enhancement study with additional psychoeducational sessions for healthy siblings of a child with cancer during inpatient family-oriented rehabilitation. *Psycho-Oncology, 27, 892-899.*
- Noser, A. E., Cushing, C. C., McGrady, M. E., Amaro, C. M., & Huffhines, L. P. (2017). Adaptive intervention designs in pediatric psychology: The promise of sequential multiple assignment randomized trials of pediatric interventions. *Clinical Practice in Pediatric Psychology, 5, 170-179.*
- Oberoi, A. R., Towry, L., Eilenberg, J. S., Lun, P., Lerro, G., & Long, K. A. (2019). Supporting siblings of children with cancer through a community-academic partnership. *Clinical Practice in Pediatric Psychology.*
- Orwin, R. G. (1983). A fail-safe N for effect size in meta-analysis. *Journal of Educational Statistics, 8, 157-159.*
- Packman, W., Weber, S., Wallace, J., & Bugescu, N. (2010). Psychological effects of hematopoietic SCT on pediatric patients, siblings and parents: A review. *Bone Marrow Transplantation, 45, 1134-1146.*
- Pai, A. L. H., Drotar, D., Zebracki, K., Moore, M., & Youngstrom, E. (2006). A meta-analysis of the effects of psychological interventions in pediatric oncology and outcomes of psychological distress and adjustment. *Journal of Pediatric Psychology, 31, 978-988.*
- Pai, A. L. H., Patino-Fernandez, A. M., McSherry, M., Beele, D., Alderfer, M. A., Reilly, A. T., Hwang, W. T., & Kazak, A. E. (2008). The Psychosocial Assessment Tool (PAT2.0): Psychometric properties of a screener for psychosocial distress in families of children newly diagnosed with cancer. *Journal of Pediatric Psychology, 33, 50-62.*

- Pai, A., Tackkett, A., Ittenbach, R., & Goebel, J. (2012). Psychosocial Assessment Tool 2.0_General: Validity of a psychosocial risk screener in a pediatric kidney transplant sample. *Pediatric Transplantation, 16*, 92-98.
- Palermo, T. M., & Wilson, A. C. (2009). eHealth applications in pediatric psychology. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of pediatric psychology* (4th ed., pp. 227–237). New York: Guilford Press.
- Patterson, J. M. (1991). Family resilience to the challenge of a child's disability. *Pediatric Annals, 20*, 491-499.
- Patterson, P., McDonald, F. E. J., Butow, P., White, K. J., Costa, D. S. J., Millar, B., ... & Cohn, R. J. (2014). Psychometric evaluation of the Sibling Cancer Needs Instrument (SCNI): An instrument to assess the psychosocial unmet needs of young people who are siblings of cancer patients. *Support Care Cancer, 22*, 653-665.
- *Prchal, A., Graf, A., Bergstraesser, E., & Landolt, M. A. (2012). A two-session psychological intervention for siblings of pediatric cancer patients: A randomized controlled pilot trial. *Child and Adolescent Psychiatry and Mental Health, 6*, 9.
- Prchal, A., & Landolt, M. A. (2009). Psychological interventions with siblings of pediatric cancer patients: A systematic review. *Psycho-Oncology, 18*, 1241-1251.
- Reader, S., Ruppe, N., Deatrick, J., Rash-Ellis, D., Wadman, J., Miller, R., & Kazak, A. (2017). Caregiver perspectives on family psychosocial risks and resiliencies in pediatric sickle cell disease: Informing the adaptation of the Psychosocial Assessment Tool (PAT). *Clinical Practice in Pediatric Psychology, 5*, 330-341.
- Reynolds, C. R., & Kamphaus, R. W. (1992). *Manual behavior assessment system for children*. USA: American Guidance Service, 1992.

- Rosenthal, R. (2001). *Meta analytic procedures for social research* (rev. ed.). Newbury Park, CA: Sage.
- Rossiter, L., & Sharpe, D. (2001). The siblings of individuals with mental retardation: A quantitative integration of the literature. *Journal of Child and Family Studies, 10*, 65-84.
- Sahler, O. J. Z., & Carpenter, P. J. (1989). Evaluation of a camp program for siblings of children with cancer. *American Journal of Disease in Childhood, 143*, 690-696.
- Samson, K., Rourke, M. T., & Alderfer, M. A. (2016). A qualitative analysis of the impact of childhood cancer on the lives of siblings at school, in extracurricular activities, and with friends. *Clinical Practice in Pediatric Psychology, 4*, 362-372.
- Sánchez-Egea, R., Rubio-Aparicio, M., Sánchez-Meca, J., & Rosa-Alcázar, A. (2019). Psychological treatment for family members of children with cancer: A systematic review and meta-analysis. *Psycho-Oncology, 28*, 960-969.
- Scialla, M. A., Canter, K. S., Chen, F. F., Kolb, E. A., Sandler, E., Wiener, L., & Kazak, A. E. (2018). Delivery of care consistent with the Psychosocial Standards in Pediatric Cancer: Current practices in the United States. *Pediatric Blood & Cancer, 65*(3), e26869.
- Sharpe, D., & Rossiter, L. (2002). Siblings of children with a chronic illness: A meta-analysis. *Journal of Pediatric Psychology, 27*, 699-710.
- Shelton, T. L. (1999). Family-centered care in pediatric practice: When and how. *Journal of Developmental and Behavioral Pediatrics, 20*, 117-119.
- *Sidhu, R., Passmore, A., & Baker, D. (2006). The effectiveness of a peer support camp for siblings of children with cancer. *Pediatric Blood & Cancer, 47*, 580-588.
- Smith, M. M., Pereira, S. P., Chan, L., Rose, C., & Shafran, R. (2018). Impact of well-

- being interventions for siblings of children and young people with a chronic physical or mental health condition: A systematic review and meta-analysis. *Clinical Child and Family Psychology Review*, 21, 246-265.
- Spence, S. H. (1998). A measure of anxiety symptoms among children. *Behaviour Research and Therapy*, 36, 545-566.
- Stancin, T., & Perrin, E. C. (2014). Psychologists and pediatricians: Opportunities for collaboration in primary care. *American Psychologist*, 69, 332-343.
- Stancin, T., Sturm, L., & Ramirez, L. Y. (2014). Pediatric psychology practice in primary care settings. In M. C. Roberts, B. S. Aylward, & Y. P. Wu (Eds.), *Clinical practice in pediatric psychology* (pp. 78-92). New York: Guilford Press.
- Steinberg, A.M., Brymer, M.J., Decker, K.B., & Pynoos, R.S. (2004). The University of California at Los Angeles Post-Traumatic Stress Disorder Reaction Index. *Current Psychiatry Reports*, 6, 96-100.
- Stoneman, Z. (2005). Siblings of children with disabilities: Research themes. *Mental Retardation*, 43, 339-350.
- Tudor, M. E., & Lerner, M. D. (2015). Intervention and support for siblings of youth with developmental disabilities: A systematic Review. *Clinical Child and Family Psychology Review*, 18, 1-23.
- van der Lee, J. H., Mokkink, L. B., Grootenhuis, M. A., Heymans, H. S., & Offringa, M. (2007). Definitions and measurement of chronic health conditions in childhood: A systematic review. *JAMA: Journal of the American Medical Association*, 297, 2741–2751.
- Van Schoors, M., Caes, L., Knoble, N. B., Goubert, L., Verhofstadt, L. L., & Alderfer,

- M. A. (2017). Systematic review: Associations between family functioning and child adjustment after pediatric cancer diagnosis: A meta-analysis. *Journal of Pediatric Psychology, 42*, 6-18.
- Van Schoors, M., Caes, L., Verhofstadt, L., Goubert, L., & Alderfer, M. A. (2015). Systematic review: Family resilience after pediatric cancer diagnosis. *Journal of Pediatric Psychology, 40*, 856-868.
- Varni, J., Seid, M., & Rode, C. (1999). The PedsQL: Measurement model for the pediatric quality of life inventory. *Medical Care, 37*, 126–139.
- Vermaes, I. P. R., van Susante, A. M. J., & van Bakel, H. J. A. (2012). Psychological functioning of siblings in families of children with chronic health conditions: A meta-analysis. *Journal of Pediatric Psychology, 37*, 166-184.
- Wiener, L., Kazak, A. E., Noll, R. B., Patenaude, A. F., & Kupst, M. J. (2015). Interdisciplinary collaboration in standards of psychosocial care. *Pediatric Blood & Cancer, 62*, S425.
- Woods, K., & Ostrowski-Delahanty, S. (2017). Psychometric properties of the Psychosocial Assessment Tool-Chronic Pain version in families of children with headache. *Journal of Child Neurology, 32*, 766-773.
- Wu, Y. P., Aylward, B. S., Roberts, M. C., & Evans, S. C. (2012). Searching the scientific literature: Implications for quantitative and qualitative reviews. *Clinical Psychology Review, 32*, 552-557.