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Supporting Siblings as a Standard of Care in Pediatric Oncology

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In this study, evidence is provided for supporting siblings as a standard of care in pediatric oncology. Using Medline, PsycInfo, and CINAHL, a systematic search of articles published over the past two decades about siblings of children with cancer was conducted. A total of 125 articles, which were primarily descriptive studies, were evaluated by the four investigators using Grading of Recommendations Assessment, Development, and Evaluation (GRADE) criteria. There is moderate-quality evidence, as well as support from community stakeholders, to justify a strong recommendation that siblings of children with cancer should be provided with psychosocial services and that parents and professionals are advised about how to meet siblings' needs. Pediatr Blood Cancer 2015;62:S750-S804. © 2015 Wiley Periodicals, Inc.

Key words: adjustment; cancer; intervention; pediatric; sibling

INTRODUCTION

Siblings are exposed to significant stress when a child is diagnosed with cancer. Concern about the ill child, disruptions in family roles and routines, decreased contact with family members, and additional demands for caregiving or other responsibilities in the home are common.[1,2] In some cases, siblings are also called upon to serve as a donor for stem cell transplant, which can introduce other ethical and psychosocial concerns.[3] Taken together, these unique challenges leave siblings of children with cancer at risk for acute and long-term psychosocial difficulties. However, there are no current evidence-based standards for the supportive care of siblings of children with cancer.

Although severe psychopathology is rare, several reviews suggest that some siblings exhibit symptoms of anxiety, depression, posttraumatic stress; lower quality of life and healthcare utilization; and disruption to academic and social functioning.[1,2,4,5] Most difficulties improve over the first year after diagnosis, but they may resurface or worsen with declines in the ill child's health or death.[6] Siblings can also demonstrate resilient outcomes, such as posttraumatic growth.[4,7] This variability in adjustment underscores the need for accurate screening to identify risk and protective factors and to provide appropriate services for siblings vulnerable to difficulties. Unfortunately, siblings have unmet needs and psychosocial support may be limited.[8,9] Further, in a large survey of professionals from three pediatric oncology organizations, only 25% reported that they provide psychosocial services to siblings.[10] Thus, it is critically important to establish evidence-based standards of care for siblings of children with cancer in efforts to encourage the provision of more consistent and comprehensive services for this population.

METHODS

This review was performed as a part of the collaborative effort, Standards for Psychosocial Care of Children with Cancer and Their Families. A full description of the methods used to develop each standard is in the introduction to this special issue.[11] The literature search for this standard used three databases: Medline, PsycInfo, and CINAHL (March 1, 1995–March 1, 2015). Abstract search terms included keywords related to siblings AND childhood AND cancer AND psychosocial outcomes (see Supplementary Table I). The search was limited to peer-reviewed journal articles written in English and involving human subjects. Commentaries, opinion pieces, case studies, dissertations, and unsystematic reviews were excluded.

After duplicates were removed, the titles and abstracts of 3,205 citations were screened by two authors (see Fig. 1). Empirical studies or reviews, both qualitative and quantitative, that included siblings of children (aged 18 and below) diagnosed with cancer were retained for full text review (N = 278). No research exclusively examined outcomes for sibling donors of children with cancer who received stem cell transplant. Thus, eight articles that were nonspecific to cancer but included sibling donors were retained. The reference lists of retained systematic reviews were also hand-searched, resulting in the addition of six studies. In all, 106 empirical studies (74 quantitative, 32 qualitative), 16 reviews, and three guidelines were included in this report. The preferred reporting items for systematic reviews and meta-analyses (PRISMA) flow diagram is shown in Figure 1. The study team included four doctoral level psychologists (authors of this paper). The founder and current director of *SuperSibs!* provided community stakeholder input, and at least two physicians or psychologists from the larger standards project reviewed

Psychosocial Standard of Care

Siblings of children with cancer are a psychosocially at-risk group and should be provided with appropriate supportive services. Parents and professionals should be advised about ways to anticipate and meet siblings' needs, especially when siblings are unable to visit the hospital regularly.

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RESULTS

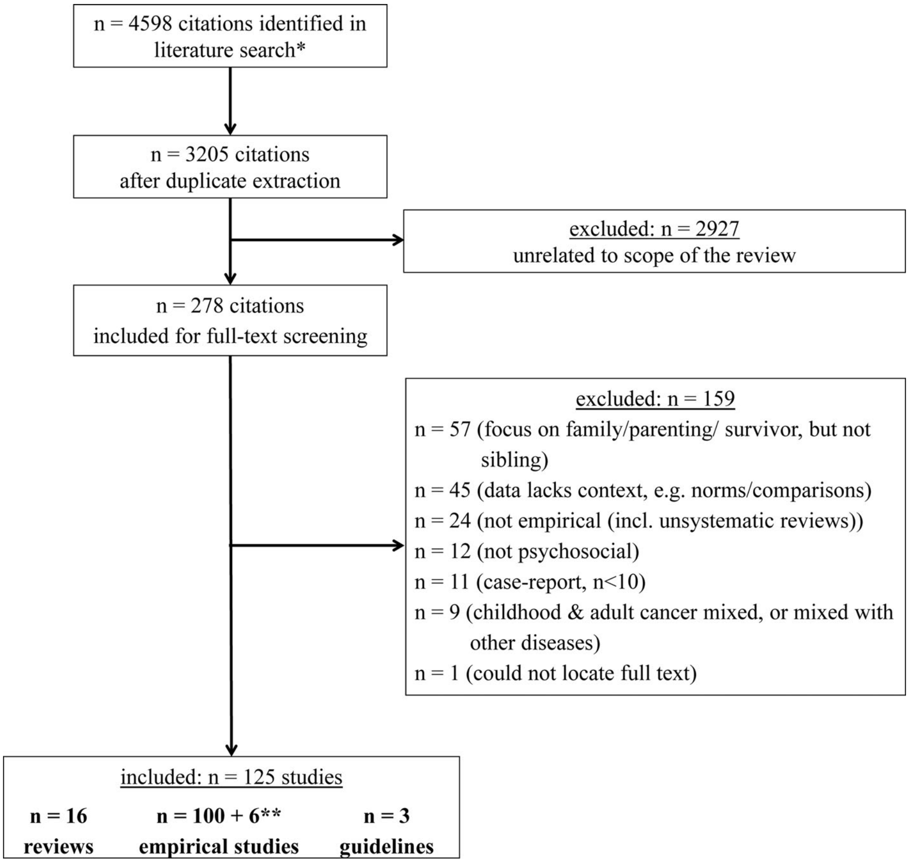
A summary of evidence is presented in Table I, indicating a strong recommendation based on the moderate quality of evidence and the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) system.[12] Detailed results from all included articles can be found in the Supplementary Table II. Several reviews have summarized the issues facing siblings of children with cancer.[1,2,4,5] Thus, selected studies are reported below to highlight examples of support for specific aspects of psychosocial care for siblings.

Sibling Stressors

When a child is diagnosed with cancer, siblings face significant disruption in their lives as evidenced by eight reviews or guidelines, six quantitative, and 11 qualitative studies. This stress includes additional demands for caregiving and other responsibilities at home, as well as experiencing diminished contact with the ill child and less attention from parents.[13–16] Challenges to maintaining normalcy and engaging in typical developmental activities also have been reported.[17,18] For example, siblings may be more likely to miss school compared to peers and fall behind academically,[18,19] although this may improve as treatment subsides.[20,21] They also describe the experience of having a brother or sister with cancer as a loss of their family's way of life and a loss of their sense of self.[1] Thus, these stressors may increase the risk for psychosocial difficulties for siblings of children with cancer.

Communication Needs

Communication with siblings is important over the course of the illness and beyond as indicated by eight reviews or guidelines, four quantitative, and eight qualitative studies. Siblings need information about the child's illness and treatment, as well as opportunities to talk about the impact of the illness on their lives and adjustment.[9,14,22] Psychosocial providers should work with the healthcare team and parents to facilitate their communication with siblings. Siblings should be updated regularly and provided with information about the disease and treatment in a developmentally appropriate manner. Based on one review, three quantitative, and five qualitative studies, siblings who serve as matched donors for stem cell transplant represent a special



* Medline: n=2709, PsycInfo: n=1035, CINAHL: n=854
** identified through bibliography search of included reviews;
(these were not identified in the literature search due to missing cancer-related key terms)

Fig. 1. Preferred reporting items for systematic reviews and meta-analyses (PRISMA) flow diagram demonstrating the article selection process.

TABLE I. Sibling Standard of Care: Summary of Evidence

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
Siblings of children with cancer are a psychosocially at-risk group and should be provided with appropriate supportive services. Parents and professionals should be advised about ways to anticipate and meet siblings' needs, especially when siblings are unable to visit the hospital regularly.	• Descriptive studies indicate family disruption and added stressors in the home increase risk for sibling difficulties. • Descriptive studies and guidelines for care suggest communication and involvement are important for siblings. • Controlled longitudinal research suggests risk for psychopathology is highest early in treatment and in the first 2 years after a child's death. • There is also evidence of resilience from qualitative and quantitative work. • A few intervention studies provide preliminary support for evidence-based practice.	• Mixed methods, qualitative, quantitative, and literature reviews. • Majority cross-sectional retrospective survey and in-depth interviews. • Replication of findings evident for descriptive studies. • Limited intervention trials.	Moderate quality given consistent findings from lower level evidence studies.	Strong recommendation given the risk–benefit ratio including significant psychosocial effects for some siblings and positive outcomes with appropriate intervention.

[1] Based on Supplementary Table II [2] types of studies, for example, RCT, cross-sectional, longitudinal, consensus, and systematic review articles [3] quality of evidence: High, moderate, low, and very low [4] strength of recommendation: Strong or weak based on GRADE criteria.[12]

circumstance in which communication, informed consent, and potential for distress should be evaluated and addressed.[3,23–25] They should be educated about tests and procedures, as well as the potential for success and/or failure of the transplant. The American Academy of Pediatrics advises attention to ethical issues associated with children who serve as donors,[26] and distress should be closely monitored by psychosocial providers during the course of planning for transplant, the procedure itself, and thereafter.

Psychosocial Impact

Evidence for the psychosocial impact of childhood cancer on siblings comes from 11 reviews or guidelines, 59 quantitative, and 12 qualitative studies. Research suggests that siblings of children with cancer are at risk for emotional and behavioral difficulties, such as anxiety, depression, and posttraumatic stress symptoms;[27,28] poorer quality of life and lower healthcare utilization;[29,30] and academic and social disruptions.[30,31] Siblings of children with cancer may also have higher rates of borderline and clinical range scores for internalizing, externalizing, and total problems (23–48%) relative to normative samples (16–17%).[30,32,33] However, psychosocial difficulties are not universal and severe psychopathology is rare.[20,21,34,35] Difficulties tend to be more common in the early phases of the illness and improve over the first year.[30,31] Some siblings can also exhibit resilient outcomes or enhanced functioning,[1,4,7] underscoring the need to identify those at risk in order to triage services. Findings are mixed, but factors such as older age, female gender, premorbid distress, lower social support, and family conflict have been linked to worse sibling outcomes in some cases.[28,35–38]

Bereaved Siblings

One review, five quantitative, and eight qualitative papers focused specifically on bereaved siblings. Siblings report a desire to be involved and informed when their brother or sister is at the end of life.[39,40] During this time, they report both positive and negative changes in themselves (e.g., sad, more mature) and their relationships with others (e.g., closer or more distant from others).[41] Bereaved siblings have been rated by both parents and teachers as having more internalizing and externalizing problems than norms or control groups,[42,43] and they can exhibit difficulties in peer relationships relative to classmates within the first 2 years of the death.[44] These concerns may diminish with time, but bereaved siblings have also reported that long-term outcomes (e.g., educational and career goals) may be affected by the loss.[6] This highlights the need to provide ongoing support to families, especially after a child has died.[45]

Supportive Care

The supportive care of siblings of children with cancer encompasses a broad spectrum of services, including assistance with family communication, psychoeducation, decision making for sibling donors, coping and prevention of psychosocial difficulties, as well as assessment and treatment of psychopathology. The intervention literature is limited but includes various attempts to address sibling needs and difficulties as evidenced by four reviews, 15 quantitative, and three qualitative papers. Most often sibling support groups or camps are described,[46–50] with only a few randomized controlled trials reported.[51,52] One review concluded that camp programs may enhance physical and emotional functioning (e.g., self-esteem),[53] while reviews including other types of interventions indicate improvements in knowledge, mood, and quality of life.[54,55] Given the preliminary nature of intervention research with siblings of children with cancer, reliance on evidence-based strategies derived from work with other populations may be necessary, especially when more severe psychopathology is evident.

BARRIERS

Barriers to provide psychosocial support to siblings include limitations in (i) availability of trained psychosocial staff and community resources, (ii) staff knowledge of issues faced by siblings; (iii) access to standardized screening tools to assess sibling distress and needs, (iv) healthcare providers' access to and communication with siblings, and (vi) intervention research to inform evidence-based care. Institutions should have adequate psychosocial staff (e.g., social work, child life, and psychology) and provide education and training to increase awareness of sibling issues. Periodic assessment and provision of services across the illness spectrum is recommended. Standardized screening tools should be combined with clinical interviews to enhance assessment. Services should be sensitive to the family context and developmental level of the sibling. Partnering with parents, extended family members, and other professionals (e.g., teachers and community-based providers) to anticipate and address sibling psychosocial needs is ideal. Flexibility in location and modality of care is often necessary as contact with siblings may be restricted due to hospital policy or for practical reasons. This is especially true after a child's death. Parent proxy report or phone contact with siblings may be alternatives to in-person meetings. Knowledge of resources for siblings and community referrals are important. In addition, controlled and longitudinal research is needed that includes multiple sites, methods, and informants, particularly in the evaluation of interventions for siblings.

CONCLUSIONS

Siblings of children with cancer are a psychosocially at-risk group and should be provided with appropriate supportive services. Parents and professionals close to the sibling should be advised about ways to anticipate and meet siblings' needs, even when they are at a distance. Data from 74 quantitative, 32 qualitative, and 19 reviews or guidelines affirm this recommendation. Current evidence for this recommendation is of moderate quality given consistent findings from lower level evidence and small-scale studies. Nevertheless, this is an overall strong recommendation given the risk–benefit ratio, including significant psychosocial effects for some siblings and the positive outcomes noted from intervention. Continued research with respect to the identification of groups at risk for psychosocial difficulties and the evaluation of interventions is warranted for siblings of children with cancer.

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Authors' contribution

C.A.G. and M.A.A. were responsible for the conception and design of this standard. All authors were responsible for the collection and assembly of data, data analysis, interpretation and presentation of data, drafting of the manuscript, and critical revisions for important intellectual content. All authors approved the final version of this manuscript and take public responsibility for the content presented in this article.

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SUPPLEMENTARY INFORMATION

SUPPLEMENTAL TABLE I. Database Search Terms Used in Medline, PsycInfo, and CINAHL

1. sibling* OR sister OR sisters OR brother OR brothers OR family OR families
2. childhood OR child OR children OR adolescen* OR pediatric OR paediatric OR youth
3. cancer OR cancers OR malignan* OR tumor OR tumors OR tumour OR tumours OR neoplasm* OR sarcoma OR sarcomas OR hodgkin* OR leukaemi* OR leukemi* OR lymphom* OR non-hodgkin* OR oncolog* OR hematolo*
4. psychosocial OR psychiatric OR psycholog* OR adjustment OR adaptation OR distress OR stress OR social OR school OR anxiety OR depression OR grief OR grieving OR bereave* OR mourning OR well-being OR "quality of life"
5. 1 AND 2 AND 3 AND 4
6. limit 5 to peer-reviewed journals, published 1995–2015, English language, human, exclude dissertations [PsycInfo, CINAHL]
   limit 5 to published 1995–2015, English language, human [Medline]

SUPPLEMENTAL TABLE II. Systematic Literature Review

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Alderfer & Hodges, 2010 [1]	Cross-sectional, descriptive, comparison to norms	161 siblings (ages 8–18) & one parent each ($n = 161$), 3-38 months post-diagnosis, 51 teachers	● The percentage of siblings falling into the borderline/clinical range on the parent-report CBCL was significantly elevated compared to norms: ● Total problems: 30% vs. 16%; Internalizing problems: 28% vs. 16%; Externalizing: 23% vs. 16%; Social Competence: 28% vs. 16% ● Siblings did not differ from norms on social and school competencies (7%/8% vs. 8%) and rule-breaking (8% vs. 8%) ● The percentage of siblings reporting clinical levels of anxiety symptoms was similar to general population (18% vs. 16%), the percentage reporting clinical levels of depressive symptoms was lower (9% vs. 16%) ● Mean scores across both parent- and sibling-report were in the “normal” range with T-scores ranging from 47–53 ● 54% of siblings reported moderate to severe cancer-related PTSS ● Higher (self-rated) social support from friends, parents, teachers, classmates and others were related to fewer depressive symptoms and less rule-breaking, but unrelated to anxiety or cancer-related traumatic stress or teacher reported school functioning/behavior	+	NA	+	+	+	4
Alderfer et al., 2003 [2]	Cross-sectional, case-control	78 siblings (ages 10–20), 5.3 years post-treatment & 134 children with healthy siblings	● 49% of siblings reported mild posttraumatic stress (PTS) reactions, another 32% reported moderate to severe PTS. ● Siblings reported more symptoms of PTS than the reference group for Intrusion ($d = 0.36$), Avoidance ($d = 0.53$), and for the total score in the PTSD-Reaction Index ($d = 0.52$) ● Levels of anxiety were comparable ($d = 0.13$) ● Female siblings and those older than age 6 at diagnosis reported more symptoms of PTS on one measure, but not the other	+	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Ballard et al.,2004 [3]	Cross-sectional, descriptive	Parents from 86 families reported upon 159 siblings (ages 0–26), on and off treatment	● 41 parents (50%) said “yes” when asked “do you think your well child(ren) has or is likely to have any problems as a results of your sick child’s illness.” Parents of children off-treatment were less likely to endorse this question ● 40% (n = 33) indicated a member of the multidisciplinary team had tried to help their well child(ren) ● 25% felt written information or advice on helping their well child(ren) was adequate; 14% felt video information provided on this subject was sufficient. ● The following percentages of parents indicated willingness to involve siblings in: – 88%, teaching about what cancer is – 87%, teaching on cancer etiology – 88%, teaching on cancer treatment – 87%, hospital tour and medical equipment play – 83%, hospital play – 86%, sharing feelings – 87%, discussing ways of coping – 78%, telephone support – 78%, letter exchange – 91%, watching supportive videos ● Parents of children with a mean age under 7 and over 16 were less likely to indicate a willingness to involve siblings in the activities listed above ● 58% (n = 43) said their well child should be more involved in their sick child’s nursing care ● Parents were less likely to indicate they would attend intervention sessions that did not coincide with outpatient appointments. Parents observing/anticipating problems with siblings were more likely to indicate an ability to bring their child(ren) for intervention ● 72% of parents (n = 53) indicated that sessions designed for the whole family would be more useful than sessions designed for siblings alone	+	NA	+	+	–	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias* Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Bansal et al., 2014 [4]	Cross-sectional, case-control	Parent-report of 40 siblings, 40 children with cancer in maintenance phase, & 40 matched controls (ages 5–18)	• No significant differences between siblings and controls across HRQoL domains, i.e. emotional, social, and school health ($d = 0.15/0.59/0.03$) • No significant differences between siblings and controls across Emotional Health subscales, i.e. fear ($d = 0.20$), sadness ($d = 0.19$), anger ($d = 0.45$), sleep ($d = 0.00$), and future worries ($d = 0.54$) • No significant differences between siblings and controls across Social Health subscales, i.e. Ability to maintain friendships ($d = 0.12$), Willingness of other for friendships ($d = 0.00$), Bullied by others ($d = 0.70$), Ability to compete ($d = 0.12$) • No significant differences between siblings and controls across School Health subscales, i.e. Attentiveness ($d = 0.00$), Memory ($d = 0.00$), Homework ($d = 0.31$), Absent due to sickness ($d = 0.08$), Absent due to hospital visit ($d = 0.18$) • parents over-rated sibling HRQOL compared to self-report in physical, emotional, and school domains, but not social health	+	NA	+	+	+	4
Bansal et al., 2013 [5]	Cross-sectional, case-control	Self-report of 40 siblings, 40 children with cancer in maintenance phase, & 40 matched controls (ages 5–18)	• No significant differences between siblings and controls for total HRQOL score ($d = 0.15$) or across HRQOL domains: Emotional Health ($d = 0.28$), Social Health ($d = 0.88$), and School Health ($d = 0.06$) • Significant differences on some Emotional Health subscales, i.e. sadness ($d = 0.74$), anger ($d = 0.92$), and future worries ($d = 1.14$), but not fear ($d = 0.22$), and sleep ($d = 0.10$) • No significant differences between siblings and controls across most Social Health subscales: Ability to maintain friendships ($d = 0.00$), Willingness of other for friendships ($d = 0.00$), and Ability to compete ($d = 0.26$), but siblings were bullied more often by others ($d = 1.11$) • No significant differences between siblings and controls across School Health subscales, i.e. Attentiveness ($d = 0.23$), Memory ($d = 0.07$), Homework ($d = 0.29$), Absent due to sickness ($d = 0.06$), Absent due to hospital visit ($d = 0.37$)	+	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Barr et al., 2010 [6]	Cross-sectional, post-intervention	76 camp attending families and 86 non-attending families (N of siblings not reported)	● Mother-report on (formerly sick) children and siblings; siblings completed one questionnaire ● Study compared sibling camp attendees (who had attended camp at some time within 4 years of their brother or sister's diagnosis) vs. non-attendees ● Non-attendee siblings reported lower grades at school than attendees ● Non-attendee siblings reported poorer parental nurturing and greater parental rejection than attendee siblings, but no differences in parental monitoring	+	NA	+	–	–	4
Barrera et al., 2004 [7]	Cross-sectional, descriptive	72 siblings (ages 6–18 years) & parents, time:NR	● Siblings with higher social support reported fewer symptoms of depression ($eta^2 = .07$), anxiety ($eta^2 = .09$), and fewer behavioral problems ($eta^2 = .16$) ● Siblings with higher social support were perceived by parents as having fewer symptoms of anxiety ($eta^2 = .13$) and fewer behavioral problems ($eta^2 = .22$) ● Various interactions between age, gender and groups status are also reported	+	NA	–	+	+	6
Barrera & Atenafu, 2008 [8]	Cross-sectional, descriptive (from larger longitudinal study)	Parent-report of 33 siblings (ages 3–20) from 46 stem cell transplant survivors, 2 years post-HSCT	● Siblings' mean scores on the CBCL were within the normal range (statistical analysis not performed) with T-scores ranging from 49 to 51 for externalizing and internalizing problems respectively	+	NA	+	–	NA	4
Barrera et al., 2002 [9]	Longitudinal, descriptive	17 siblings enrolled, 12 completed the intervention. (ages 6–18)	● This study evaluated the Siblings Coping Together program, a cognitive behavioral and family systems based manualized intervention consisting of 8 weekly 2-hour sessions ● Self- and parent-report ● 33% of siblings fell into the clinical range for anxiety prior to intervention by both parent ($n = 2$) and sibling ($n = 4$) report ● 8% of siblings ($n = 1$) fell into the clinical range for depression (sibling-report) ● Levels of sibling-reported depression improved from pre- to post-intervention ($d = 0.48$), as well as levels of state anxiety ($d = 0.95$) ● Parent-reported sibling anxiety did not change ($d = 0.70$)	–	NA	+	+	–	3

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias* Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Barrera et al., 2004 [10]	Longitudinal, descriptive	42 siblings of which 40 completed the first post-intervention time point (ages 6–14)	● Measures were completed twice prior to intervention (eight weeks apart), once immediately after the final intervention session, and again 6–12 months later ● Sibling self-reports of depression ($eta^2 = .20$) and anxiety decreased ($eta^2 = .19$) ● Parent-reports of sibling anxiety decreased ($eta^2 = .35$) ● No significant intervention effect was found for parent report of sibling behavior problems	–	NA	+	+	+	3
Birenbaum, 2000 [11]	Longitudinal, descriptive, comparison to norms	Parent-report of 61 bereaved siblings (from 37 families)	● Families provided data 2 months prior to death (T1), and 0.5 (T2), 4 (T3) and 12 months (T4) after the death ● The percentage of 6–11 year old siblings displaying symptoms was greater than that in the general population across 2 or more time points for the following (out of 100) items:Argues a lot; Stubborn, sullen, irritable; Impulsive; Hyperactive; Showing off; Self-conscious; Likes to be alone; Secretive; Teases at lot; Needs to be perfect; Moody; Worrying; Unhappy, sad, depressed; Daydreams; Acts too young; Refuses to talk; Gets teased; Too dependent; Lonely; Withdrawn; ● The percentage of 12–19 year old siblings displaying symptoms was greater than that in the general population across two or more time points for the following (out of 100) items:Argues a lot; Stubborn, sullen, irritable; Disobedient at home; Impulsive; Hyperactive; Shy or timid; Showing off; Demands attention; Self-conscious; Likes to be alone; Secretive; Needs to be perfect; Can't concentrate; Moody; Worrying; Unhappy, sad, depressed; Daydreams; Acts too young; Talks too much; Poor eater; Refuses to talk; Feels unloved; Too loud; Too dependent; Cries a lot; Nightmares; Lonely; Withdrawn; Trouble sleeping	–	NA	+	–	–	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Buchbinder et al., 2011 [12]	Cross-sectional, descriptive	3083 adult siblings of long-term survivors; siblings were unborn-36y at diagnosis	• 3.8% of siblings reported elevated psychological distress (T-score≥63 on the Global Severity Index of the BSI) – 2.0% was in the clinical range for somatization – 3.3% was in the clinical range for depression – 1.1% was in the clinical range for anxiety • When entered simultaneously into multi-variable models, the following factors emerged as significant predictors of sibling distress: – Being unmarried (global distress, depression) – income < \$60,000 per year (global, somatization, depression, anxiety) – Self-reported fair/poor general health (global, somatization, depression, anxiety) – Sibling chronic health condition (somatization, anxiety) – Being younger than the survivor (global) – Survivor self-reported fair/poor general health (depression) – Survivor self-reported distress (global, depression) – Sibling age ≤ 29 at baseline (somatization only) – Being male (somatization)	+	NA	+	+	–	6
Buizer et al., 2006 [13]	Cross-sectional, case-control	Parent-report of 37 siblings of 64 children (>1year post-treatment), and 98 controls (ages 4–18)	• No significant differences between siblings and controls on the CBCL, i.e. Total Problems ($d = 0.08$), Internalizing ($d = 0.05$), and Externalizing ($d = 0.14$), as well as the subscales: withdrawn ($d = 0.28$), somatic complaints ($d = 0.05$), anxiety/depression ($d = 0.04$), social problems ($d = 0.14$), thought problems ($d = 0.33$), attention problems ($d = 0.16$), delinquent behaviors ($d = 0.22$), and aggressive behaviors ($d = 0.08$) • No significant differences between siblings and controls with regard to percentages falling outside the normal range on these scalesThere were no significant differences between siblings and controls on the Conners' Teacher Rating Scale (CTRS), regarding Total problems ($d = 0.13$), Internalizing ($d = 0.11$), Externalizing ($d = 0.07$), Acting Out ($d = 0.04$), Antisocial ($d = 0.11$), Hyperkinesia ($d = 0.14$), Anxious/withdrawn ($d = 0.21$), Social Isolation ($d = 0.10$) • No significant differences between siblings and controls in regard to percentages falling outside the normal range on the CTRS scales	+	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias* Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Chao et al., 2003 [14]	Cross-sectional, descriptive	24 children with cancer (aged 8–17) & 18 of their parents, 14.5 months post-diagnosis	● Patients and parents observed some positive changes in siblings' behaviors, such as more responsibility, more independence and greater closeness to the patient	–	NA	+	?	–	6
Cordaro et al., 2012 [15]	Cross-sectional, case-control	Parent-report of 30 siblings of children with cancer & 33 siblings of healthy children (all ages 4–18), time:NR	● Significant better scores for siblings than controls on the CBCL Total Problems, Internalizing, and Externalizing scale (data not reported) ● Siblings that were older than the child with cancer displayed more problems than those younger than the child with cancer ● Adjustment of siblings did not differ by gender, absolute age, family size, understanding of the situation or extent of changes in daily routine	+	NA	+	+	NA	4
Dolgin, Blumensohn et al., 1997 [16]	Study 1: Cross-sectional, comparison to "normal" and "clinically referred" children	Study 1: parent-report of 33 siblings (aged 6–18);33 matched "normal" children & 33 matched "clinically referred" children	Study 1: ● Siblings' scores were no different from "normal" children and significantly better than "clinically referred" children: Total Behavior Problems ($d = 0.11/1.15$), Internalizing ($d = 0.08/1.01$), Externalizing ($d = 0.25/0.85$), Withdrawn ($d = 0.23/1.07$), Somatic Complaints ($d = 0.05/0.44$), Anxiety/Depression ($d = 0.17/0.82$), Social Problems ($d = 0.16/0.72$), Thought Problems ($d = 0.10/0.88$), Attention Problems ($d = 0.10/1.18$), Delinquent Behaviors ($d = 0.00/0.91$), Aggressive Behavior ($d = 0.12/0.97$) ● The proportion of siblings who met clinical cut-offs on the measure ($n = 2$) was not significantly different from the "normal" controls ($n = 1$) and significantly less than the "clinically referred" group ($n = 15$)	+	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
	Study 2: Cross-sectional, intercultural comparisons (Israeli vs. American)	Study 2: Parent-report of 35 Israeli siblings (ages 6–18) & 35 matched American siblings, 6–42 months post-diagnosis	<u>Study 2:</u> ● no significant differences between Israeli and American siblings regarding Total Behavioral Problems ($d = 0.32$), Internalizing ($d = 0.22$), Externalizing ($d = 0.37$) ● No significant differences in the proportion of Israeli siblings (6%) and American siblings (14%) falling in the clinical range for total behavioral problems ● More family support, greater family expressiveness, and use of interpersonal support as a parental coping strategy was associated with fewer total, internalizing and externalizing problems ● Greater family conflict was associated with more total, internalizing and externalizing problems						
Dolgin, Somer et al., 1997 [17]	Longitudinal, descriptive	23 siblings (ages 6–18) and their parents, on treatment/within 6 months post-treatment	● Intervention study of 6 weekly sessions involving facilitated group discussions, art therapy, role playing, and informal social interaction ● Siblings reported a significant improvements in Interpersonal Problems ($d = 0.85$), Intrapsychic Preoccupation ($d = 1.38$), Disease-related Communication ($d = 0.60$), but not Fear ($d = 0.00$) ● Siblings reported increases in cancer-related knowledge ($d = 1.30$) and improvements in their mood state ($d = 1.27$) ● Parents reported improvements in siblings' mood state ($d = 1.87$)	–	NA	+	+	NA	3

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Eilertsen et al., 2013 [18]	Cross-sectional, descriptive	174 bereaved siblings (ages 19–33) 2–9 years post-death	● Siblings were more likely to fall into clinical ranges of anxiety … – if their need for social support was not satisfied *in the last month before* their brother or sister's death (or were too young to remember); n = 54 vs. 114 (the rest), RR = 3.6, 95% CI = 1.8-7.3 – if their need for social support was not satisfied *after* their brother or sister's death (or were too young to remember); n = 29 vs. 118, RR = 2.9, 95% CI = 1.5-5.6 – if their need for social support was not satisfied *during the past year prior to follow-up;* n = 36 vs. 136, RR = 3.8, 95% CI = 2.0-7.2 – if they reported that that others cared for them not at all or little *in the last month before* their brother or sister's death (or were too young to remember); n = 26 vs. 131 (who felt that others cared for them at least moderately), RR = 2.0, 95% CI = 1.03-4.0; no differences were found between these groups when reporting on the time periods *after* the death or *during the past year prior to follow-up* – if they shared none or less than half of their feelings about their brother or sister's death with their family (or who were too young to do so); n = 90 vs. 84 (who shared half or more), RR = 2.8, 95% CI = 1.3-6.2 – if they shared none or less than half of the difficulties occurring in their life with their family in the past year, n = 60 vs. 113, RR = 2.9, 95% CI = 1.5-5.8 ● Generally, siblings who reported that their parents did not care for them around the time of death, were more likely to fall into clinical ranges of anxiety (more detailed results are reported in the paper)	+	NA	+	–	–	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Evans & Radford, 1995 [19]	Cross-sectional, descriptive	38 siblings (ages 16–30) of survivors (incl. 5 siblings of children on active treatment)	● 23 siblings (62%) achieved five or more A-C grades at level of GCSE by age 16; the national average was 30%, no statistical comparison reported ● 18 siblings (48%) continued onto higher education; the national average was 17.3%, no statistical comparison reported ● 4 siblings (10%) felt that their education had suffered as a result of their sibling's illness ● 23 siblings (61%) had high self-esteem	–	NA	–	–	–	6
Freeman et al, 2003 [20]	Cross-sectional, descriptive	46 siblings (ages 9–30, M = 17)	● Diagnosis phase: siblings reported problems with the manner of communication with parents and doctors and a lack of information about cancer etiology; but family and social support were helpful. ● Hospitalization: Lack of information about prognosis and lack of help with schoolwork were problematic. Family, social support, and liberal visitation policies were helpful. ● Post-hospitalization: Lack of help with schoolwork was problematic. Support from families, friends, and religion was helpful	–	NA	+	+	–	6
French et al., 2013 [21]	Cross-sectional, case control	77 Siblings (ages 7–18), 4–17 years post-diagnosis & population control group	● Siblings missed significantly more school days than a population control group (9.9 vs. 5 days, $d = 0.61$) ● No differences in missed school between siblings and survivors	+	NA	+	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (***Continued***)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Gerhardt et al., 2012 [22]	Cross-sectional, case control	105 bereaved siblings (ages 8–17), 3–12 months post-death, & 311 matched classmates	● Peer acceptance ratings were similar for bereaved siblings and classmates ($d = 0.02$) ● Peers perceived bereaved boys as more sensitive-isolated and victimized than bereaved girls (*d's* = 0.45 - 0.50) ● Teachers rated siblings are more prosocial than matched comparisons ($d = 0.30$) ● Elementary school: Bereaved siblings were perceived by peers as less prosocial and more sensitive-isolated (*d's* = 0.38 - 0.65). Peers perceived bereaved siblings as less accepted and as having fewer friends than comparisons (*d's* = 0.43 - 0.40) ● Middle/high school: Peers and teachers viewed bereaved siblings as higher on leadership–popularity than comparisons (*d's* = 0.28 - 0.41)	+	NA	+	+	NA	4
Glaser et al., 1997 [23]	Cross-sectional, case control	Teachers of 21 siblings & 20 matched classmates (ages 6–15), 2–10 years post-diagnosis	● Teachers rated overall school behavior similarly for siblings and matched controls ● Teachers rated siblings to be less likely to express concern for others and less likely to openly express feelings of joy than matched controls ● Teachers rated siblings and controls equally likely to express feelings of happiness, love, anger, sadness, frustration, and confusion	–	NA	+	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Hamama et al., 2000 [24]	Cross-sectional, descriptive	62 siblings (ages 9–18), during active treatment	● Higher levels of anxiety were reported by siblings whose brother/sister was ill for a shorter amount of time ($r = -.46$) and by siblings who endorsed lower levels of self-control ($r = -.28$); when considered together, both time since diagnosis and self control were significant predictors of sibling anxiety ● Higher levels of loneliness were reported by younger siblings ($r = -.31$) and by siblings who endorsed lower levels of self-control ($r = -.24$); when considered together, only self-control was a unique predictor of sibling loneliness ● Gender-by-birth-order interaction: girls who were younger than their ill siblings had a significantly higher rate of loneliness than girls who were older than the ill child or to the older and younger boys ● Sibling anxiety and loneliness were not significantly associated with sibling sex, specific cancer diagnosis, or type of treatment	+	NA	+	+	NA	6
Hamama et al., 2008 [25]	Cross-sectional, descriptive	100 siblings (ages 8–19), time:NR	● Higher levels of anxiety were correlated with lower self-efficacy ($r = -.24$), lower self control ($r = -.44$), and higher role overload ($r = .23$) ● Higher levels of psychosomatic symptoms were correlated with lower self-efficacy ($r = -.41$), lower self control ($r = -.29$), and higher role overload ($r = .26$) ● Higher self-control was associated with lower role overload ($r = -.21$) ● Age-by-self-control interaction: self-control predicted anxiety in older sibs (ages 12–19) but not younger sibs	+	NA	+	+	NA	6
Hashemi et al., 2010 [26]	Randomized controlled trial	60 parents of siblings (ages 7–15), 23 months post-diagnosis	● Parents who participated in a 3-session educational intervention ($n = 30$) scored higher on a measure of knowledge of siblings' needs immediately after the intervention ($d = 4.35$) and 2 months later ($d = 5.17$) than those assigned to the control group ($n = 30$)	+	–	+	–	–	2

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Hashemi et al., 2013 [27]	Randomized controlled trial	60 parents of siblings (ages 7–15), <1year post-diagnosis	• Parents who participated in a 3-session educational intervention reported higher levels of quality of life in the siblings two months after the intervention (MEAN CHANGE in QoL from Time 1 to Time 2: experimental group: $M(SD) = 28.8(11.3)$; control group: $M(SD) = 3.6(4.5)$) • This pattern of results emerged across all dimensions of quality of life (i.e., physical, autonomy, social, cognitive, motor, positive emotions, negative emotions, and total score)	+	–	+	–	–	2
Heffernan et al., 1997 [28]	Cross-sectional, descriptive	21 siblings (ages 9–18) from 17 families & their mothers, during active treatment	• Mothers: the most commonly reported change in siblings' behaviors after their brother's/sister's cancer diagnosis was "more sensitive to the needs of others"	–	NA	+	–	NA	6
Houtzager et al., 2001 [29]	Uncontrolled intervention trial, comparison to norms	24 siblings (ages 7–18), 2months - 7.5 years post-diagnosis	• This study evaluated a 5-session supportive care program • Compared to Dutch norms, baseline anxiety scores were higher for child boys (ages 7–12, $n = 3$, $d = 1.27$), child girls ($n = 3$, $d = 1.42$), and adolescent boys (ages 13–17, $n = 2$, $d = 1.27$), but not adolescent girls ($n = 4$, $d = 0.69$) • Changes in anxiety pre and post intervention: Mean anxiety scores decreased significantly between baseline (37.08) and post-intervention (33.25) • Percent in highest decile: 18 sibs (75%) had anxiety scores in the 8–10th decile at baseline, and 11 sibs (46%) had anxiety scores in the 8–10th decile post-intervention • Predictors of response to intervention: Boys and siblings of children with leukemia or lymphoma showed a bigger decrease in anxiety from pre- to post-intervention	–	–	+	–	–	3

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Houtzager et al., 2003 [30]	Longitudinal, comparison to norms	83 siblings (ages 7–18) at Time 1; 66 siblings at Time 2, 1–6 months post-diagnosis	● 1-month post-diagnosis: adolescent girls (ages 12–18, $n = 12$) reported significantly more anxiety symptoms ($d = 0.59$) and girls ($n = 21$) reported overall more internalizing problems ($d = 0.88$).Adolescents ($n = 34$) reported significantly lower quality of life than norms ($d = 0.49$). A significantly higher portion of female adolescents reported borderline/clinical levels of internalizing (48% vs. 16%), externalizing (33% vs. 16%) and total problems (43% vs. 17%) than the percentages in the normative population. ● 1-month post-diagnosis: Children (ages 7–11, $n = 44$) reported significantly lower quality of life than norms. ● 6-months post-diagnosis: Adolescent boys ($n = 15$) reported significantly lower externalizing ($d = 0.62$) and total problems ($d = 0.57$) than norms. Boys in elementary school ($n = 11$) reported significantly lower anxiety scores than norms ($d = 1.32$). A significantly higher percentage of adolescent girls (35%) reported borderline/clinical levels of internalizing and externalizing problems than norms. ● Changes from 1- to 6-months post-diagnosis: Adolescent boys showed significant reductions in externalizing symptoms ($d = 0.35$) and total problems ($d = 0.45$), and elementary-aged girls showed significant reductions in anxiety ($d = 0.44$) ● Predictors of poorer functioning for girls: older age, more stressful life events, and bro/sis diagnosed with leukemia or lymphoma. For boys: older age	+	NA	+	+	–	4
Houtzager, Grootenhuis et al., 2004 [31]	Cross-sectional, comparison to norms	103 siblings (ages 7–18), 2years post-diagnosis	● A higher percentage of school-aged siblings (age 7–11) endorsed quality of life scores below the 20th percentile (47% of siblings vs. 20% of norms) ● A higher percentage of adolescent siblings (ages 11–18) endorsed impairments in internalizing symptoms (29% of siblings vs. 16–17% of norms)	+	NA	+	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Houtzager, Oort, et al., 2004 [32]	Longitudinal, descriptive	83 siblings (ages 7–19) at 1month; 66 at 6months; 60 at 12months; & 57 at 24months post-diagnosis	● Siblings endorsed the most distress at the 1-month time point, and distress stabilized thereafter for most siblings *NOTE: See other papers by this author for additional data on sibling distress at each time point* ● Predictors of sibling adjustment problems: high family adaptability & cohesion, older age, female gender, lower optimism, fewer hospital days, higher vicarious control (i.e., relying on the expertise of the medical specialist), and parental distress	+	NA	+	+	+	4
Houtzager, Grootenhuis, Hoekstra-Weebers, & Last, 2005 [33]	Cross-sectional, comparison to norms	83 siblings at 1month post-diagnosis (ages 7–18)	● Compared to norms, a higher percentage of siblings endorsed quality of life scores below the 20th percentile on domains of physical complaints (31% vs. 20%), motor skills (32% vs. 20%), cognitive problems (37% vs. 20%), social problems (33% vs. 20%), negative emotions (33% vs. 20%), and lack of positive emotions (57% vs. 20%) ● Predictors of lower quality of life: older age, female gender, lower predictive control (positive expectations of outcome), higher interpretive control (attempt to find explanation for illness), and more pre-existing physical, sleeping, and eating problems	+	NA	+	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Houtzager, Grootenhuis, Caron, & Last, 2005 [34]	Longitudinal, descriptive	83 Siblings at 1-month and 57 at 2 years post-diagnosis (ages 7–18); & their parents	● Time 1 Adolescents (ages 12–18): siblings reported significantly more adjustment problems (higher levels of internalizing, externalizing, and total problems) and poorer quality of life (physical and emotional problems) than parental reports of sibling functioning ● Time 1 Children (ages 7–11): siblings reported significantly more physical and motor problems than parental reports of sibling functioning ● Time 2 Adolescents (ages 12–18): siblings significantly more adjustment problems (higher levels of internalizing, externalizing, and total problems), more emotional problems, and better social quality of life than parental reports of sibling functioning ● Time 2 Children (ages 7–11): siblings reported significantly more physical and motor problems and fewer positive emotions than parental reports of sibling functioning	+	NA	+	+	+	6
Hovén et al., 2013 [35]	Cross-sectional, descriptive	453 parents of siblings of adult survivors of childhood cancer (mean: 16 years post-diagnosis)	● 11% of parents endorsed a significant impact of the illness on siblings (i.e., score ≥ 2.5 on the Impact on Family Scale) ● Higher perceptions of sibling impact were associated with more severe disability, more unmet needs, and female gender of the survivor	+	NA	+	+	NA	6
Kamibeppu et al., 2010 [36]	Cross-sectional, case-control	71 siblings of survivors in (>1 year) remission (mean age = 25.6) and 584 female and 235 male controls	● Female siblings ($n = 41$) endorsed significantly lower levels of hyperarousal than controls ($d = 0.45$) ● Female siblings reported significantly higher total posttraumatic growth ($d = 0.43$), relating to others ($d = 0.51$), and appreciation of life ($d = 0.74$) ● Male siblings ($n = 30$) reported significantly higher appreciation of life ($d = 0.44$)	+	NA	+	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Kaplan et al., 2013 [37]	Cross-sectional, descriptive	125 siblings (ages 8–17) 4–38 months post-diagnosis	• One third of the sample (34%, $n = 42$) met symptom criteria for PTSD based upon questionnaire responses; when requiring perceptions of life threat and feelings of intense fear, horror or helplessness just over one fifth or the sample (22%, $n = 28$) met full criteria • 60% ($n = 75$) reported moderate to severe posttraumatic stress reactions • 90% reported at least one symptom of re-experiencing; 56% reported three or more symptoms of avoidance; 82% reported at least two symptoms of arousal – 74% of siblings with at least one PTS symptom indicated that symptoms interfere with functioning in at least one domain: General happiness (47%), Fun and hobbies (42%), School (41%), Chores or duties at home (37%), Friendships (37%), and Family relationships (36%) • Siblings meeting full criteria for PTSD ($n = 28$) had higher self-reported anxiety ($d = 1.00$) than those who did not ($n = 97$); as did those reporting moderate to severe PTS ($n = 75$, $d = 1.28$) compared to those with no or mild PTS ($n = 50$) • Siblings meeting full criteria for PTSD had higher self-reported depression ($d = 0.90$) than those who did not; as did those reporting moderate to severe PTS ($d = 0.90$) when compared to those with no or mild PTS • Only 5% of the sample ($n = 6$) fell into the clinical range for anxiety; 5 of these met criteria for PTSD • Only 3% ($n = 4$) fell into the clinical range for depression, half of these met criteria for PTSD • There were no significant differences in rates of PTSD or mean levels of PTS for male versus female siblings or for siblings of different ages	+	NA	+	+	NA	4
Kazak et al., 2004 [38]	Randomized clinical trial with wait-list control	78 siblings (ages 10–20) at baseline, 1–12 years post-treatment; 43 siblings at follow-up	• This study evaluated the Surviving Cancer Competently Intervention Program, a day-long multi-family group intervention • There were no significant intervention effects for siblings on self-reported measures of cancer-related traumatic stress or anxiety[Ratings of the paper in the following columns only reflect the exploratory data regarding siblings]	–	NA	+	+	–	2

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Kenney et al., 2010 [39]	Cross-sectional, descriptive	32 siblings	• Sibling reported better emotional functioning than norms (on the SF-36), $d = 0.46$ • Siblings reported less distress than norms (on the BSI), $d = 2.09$	–	NA	+	+	–	6
Kobayashi et al., 2015 [40]	Cross-sectional, descriptive, comparison to norms	13 siblings (ages 8–18), 3–35 months since hospital discharge	• Sibling self- and parent-reported quality of life did not differ from Japanese norms • Authors reported strong correlations between parent-reported family functioning and sibling-reported quality of life (no statistics provided)	–	NA	+	–	NA	4
Krull et al., 2010 [41]	Longitudinal, cross-sectional, comparison to norms	Parents of 406 siblings of (>5 year) survivors of childhood cancer	• Adolescence: 82.7% of siblings had a BMI in the underweight or normal range (vs. 84% of the normative population), 10.5% in the overweight range (vs. 10% of the normative population), and 6.8% in the obese range (vs. 5% of the normative population) • The percentage of siblings that fall in the impaired range on emotional and behavioral symptoms did not differ from that in the normative population	+	NA	–	–	+	6
Labay et al., 2004 [42]	Cross-sectional, descriptive, comparison to norms	29 siblings (ages 7–16) during active treatment	• Social competence: Siblings scored significantly below norms on activities ($d = 0.61$), social relationships ($d = 0.60$), and school performance ($d = 0.46$) • Empathy: Siblings with greater empathy reported lower externalizing and total problem scores, independent of birth order and family size. Level of empathy was related to more sophisticated understanding of the cancer treatment and prognosis • Age: Older siblings reported higher empathy ($r = .46$), less rivalry ($r = -.42$), and less conflict ($r = -.42$), and they perceived greater power in the sibling relationship ($r = .46$)	–	NA	+	+	NA	4
Lähteenmäki et al.; 2002 [43]	Cross-sectional, case-control	28 siblings (ages 8–21) and 103 classroom controls	• 7% of siblings and none of the controls started school later than normally • Siblings did not differ significantly from controls in the provision of private tutoring	–	NA	–	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Lähteenmäk et al., 2004 [44]	Longitudinal, case-control	33 siblings (ages 3–17) & 357 controls; self- & parent-report, 3 months – 1 year post-diagnosis	● Younger children (age 3–7, $n = 12$) 3-months post-diagnosis: Parents of siblings reported significantly poorer scores on "behavior in play and activities," "confrontational situations with parents," and other behavioral problems ● Younger children (age 3–7) 12-months post-diagnosis: Parents of siblings reported marginally higher conduct problems than parents of control children ● Younger children (3-7) change from time 1 to time 2: Scores were relatively stable from time 1 to time 2 ● Older children (age 8–17, $n = 21$) 3-months post-diagnosis: Siblings reported significantly higher state anxiety than controls. Parents of siblings reported significantly higher levels of conduct problems and other behavioral problems ● Older children (age 8–17) 12-months post-diagnosis: Parents of siblings reported significantly higher levels of conduct problems, impulsive-hyperactivity, learning problems, other behavioral problems, and psychosomatic problems than parents of control siblings ● Older children (age 8–17) change from time 1 to time 2: Self-reported state anxiety decreased significantly from time 1 to time 2. Levels of parent-reported behavior problems were stable from time 1 to time 2.	–	NA	+	+	NA	4
Larcombe et al., 2002 [45]	Cross sectional, case control	67 siblings (ages 18–30) of (>5 year) survivors & 184 controls	● Siblings had similar ratings on a health behavior index compared to controls ($d = 0.03$); the health behavior index encompassed smoking, alcohol consumption, recreational drug use, diet, and sun care	+	NA	–	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Long, Marsland & Alderfer, 2013 [46]	Cross-sectional, descriptive	209 siblings (ages 8–18), & 256 parents; during active treatment/ <2y post-diagnosis	● Greater distress was associated with lower parental acceptance, psychological control, and family functioning problems ($r = .19$ - $.55$; strength of associations varied by informant) ● 25% of siblings met DSM-IV criteria for PTSS, 5% for clinical levels of anxiety and 5% for depression ● High cumulative family risk (found in 21 families, 10%) predicted distress	+	NA	+	+	NA	6
Long, Alderfer et al, 2013 [47]	Cross-sectional, descriptive	30 siblings (ages 10–17); during active treatment, >6months post-diagnosis	● 30% ($n = 9$) of siblings reported moderate to severe PTSS, 7% ($n = 2$) depression, 7% anxiety ● Distress was associated with depression ($r = .89$), anxiety ($r = .90$), PTSS ($r = .88$), and age ($r = .42$) ● Siblings who were younger than the ill child had higher distress than older siblings ($d = 1.44$), even after controlling for disease and demographic factors	–	NA	+	+	NA	6
Lown et al., 2013 [48]	Cross-sectional, case-control	3034 siblings (ages 18–56) of survivors, 17y post-diagnosis	● Siblings of CCS were more likely to be heavy and risky drinkers than a national control sample (OR = 1.3 respectively) ● Age, lower education, symptoms of depression, anxiety and distress were associated with heavy alcohol use	+	NA	+	+	NA	4
Mishra et al., 2012 [49]	Cross-sectional, case-control	95 siblings (ages 9–18); during active treatment	● Children with cancer showed significantly higher levels of alexithymia than their siblings ($d = .36$), who in turn scored significantly higher than 151 healthy controls ($d = .64$)	+	NA	+	+	NA	4
Murray, 2000 [50]	Cross-sectional, descriptive	25 siblings (ages 7–12) 25 mothers; 12 months post-diagnosis	● Participants rated frequency and helpfulness of nurse interventions ● Siblings rated emotional (e.g., talking) and instrumental support (e.g., help me get involved) as most helpful interventions; while mothers rated emotional and informational (e.g., about cancer) support as most helpful	–	NA	+	+	NA	6
Murray, 2001 [51]	Cross-sectional, descriptive	50 siblings (ages 7-12); during active treatment	● Siblings who attended summer camp ($n = 22$) reported more positive self-concept scores than siblings who did not ($n = 28$), $d = 0.99$	+	NA	+	+	NA	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Murray, 2001[52]	Cross-sectional, descriptive	50 siblings (ages 7–12) 50 parents; during active treatment	● Sibling-reported most helpful interventions: encourage parents spending time with them, followed by helping parents to notice good behavior, to get involved, and to allow to visit ill sibling in hospital ● Parents rated stimulating communication most helpful	+	NA	+	+	NA	6
Packman et al., 1998 [53]	Cross-sectional, descriptive	44 siblings (ages 6–18) of BMT patients, time:NR	● No significant differences between donor siblings ($n = 21$) and nondonors ($n = 23$) on perceptions of family environment; including subscales such as communication, self-image, or emotional time[no descriptive statistics reported]	–	NA	+	+	NA	6
Packman, 2003 [54]	Cross-sectional, descriptive	42 siblings (ages 6–18) of BMT patients, time:NR	● No significant differences between donor ($n = 21$) and nondonor siblings ($n = 21$) on the emotional indicator score of the human figure drawing, but overall psychological distress was indicated[no descriptive statistics reported] ● No relationship between emotional indicator and family environment scores, psychosocial adjustment, or posttraumatic stress however, in donor siblings only, the emotional indicator was associated with posttraumatic stress ($r = .529$)	–	NA	+	+	NA	6
Packman, Fine et al., 2004 [55]	Longitudinal, descriptive	77 siblings (ages 6–17) of which 18 lost their ill sibling on study, time:NR	● No significant differences regarding PTSD, anxiety, self-esteem, or QoL between bereaved ($n = 18$) and non-bereaved siblings ● Among all siblings: PTSS, anxiety, and emotional problems decreased over the 3-month camp period ($d = 0.47/\ 0.44/0.67$), and QoL and self-esteem increased ($d = 0.32/0.43$)	+	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Packman, Gong et al., 2004; Packman, Crittenden et al., 1997 [56,57]	Cross-sectional, descriptive, qualitative	44 siblings (ages 6–18) of living BMT survivors within 2 years of transplant, 44 parents, 43 teachers	● Donor siblings ($n = 21$) reported significantly more anxiety and low self-esteem than nondonors ($n = 23$) ● No significant differences on PTSS between both groups, but about 1/3 of the whole sample reported moderate to severe levels of PTSS ● There was a trend of donor siblings being more withdrawn ($d = 0.52$) than nondonors ● Parent-report of maladaptive behaviors: no significant differences between donors and nondonors ● Teachers were more likely to report school problems among nondonors, specifically learning problems [insufficient descriptive statistics reported] ● Qualitative interviews revealed:Donors felt that they had no choice, felt lonely, felt that the whole stem cell transplant process was only minimally explained to them, they reacted with withdrawal while nondonors showed more externalizing behavior	–	NA	+	–	NA	6
Packman, 2005 [58]	Longitudinal, descriptive	77 siblings (ages 6–17) of whom 18 were bereaved, 74 parents, time:NR	● Improved HRQoL (emotional, social, school and psychosocial domain) after participation in summer camp (d's $= 0.22$ - 0.33) ● Parents did not report significant improvements (d's $= 0.01$ - 0.23) ● Findings were corroborated in qualitative interviews (3/6 themes: socializing, instillation of hope, interpersonal learning)	+	NA	+	+	+	4
Packman, 2008 [59]	Longitudinal, descriptive, qualitative	77 siblings (ages 6–17) 77 parents, time:NR	● Projective figure drawing tests indicated reductions in emotional distress ($d = 0.67$), feelings of isolation, and anxiety, as well as increased self-esteem and improvement in family environment from pre to post camp[insufficient descriptive statistics reported] ● Reduced family distress was only found in non-bereaved siblings and parents, but not bereaved	+	NA	+	–	–	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Phuphaibul & Muensa, 1999 [60]	Cross-sectional, descriptive	134 mothers of siblings (ages 6–12); 5 months - 9 years post-diagnosis	● Negative adaptive behaviors: – Increased appetite/craving ($n = 52$;38.8%) – Anger expressions ($n = 25$;18.5%) – Emotional Instability ($n = 22$;16.4%) – Anger toward the ill child ($n = 22$;16.4%) – Complaints of parental bias ($n = 13$;9.4%) – Increased separation anxiety ($n = 12$;8.9%) – Complaints of decreased parental attention ($n = 11$;8.2%) ● Positive adaptive behaviors: – Increased affect and care ($n = 69$;51.5%) – Increased self-responsibility in daily life and school ($n = 54$;40.3%) – Obedience ($n = 40$;29.9%) – Increased maturity ($n = 3$;2.3%)	+	NA	+	+	NA	6
Prchal et al., 2012 [61]	Randomized controlled pilot trial	30 siblings (ages 6–17) of newly diagnosed children	● At baseline, HRQoL was lower than the Swiss norm population, 7 siblings (23.3%) had full and 13 (43.3%) had partial PTSD ● Siblings were randomly assigned to intervention and active control group: ● Medical knowledge, HRQoL, psychological well-being, PTSS, and anxiety improved over 7 months in both groups (pre-post *d's* = 0.38/0.80/1.85/0.53/0.47 for the intervention group, $n = 14$), but interaction effects for group x time were not significant.	–	+	+	+	+	2
Rajajee et al., 2007 [62]	Cross-sectional, descriptive	34 parents (sibling age and time:NR)	● 24 children had siblings who were unaware of the diagnosis ● All parents that had well (and school-aged) siblings at home ($n = 17$) reported that school performance of the well sibling was negatively affected	–	NA	+	+	NA	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias*					Level **
				Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	
Rosenberg et al., 2015 [63]	Cross-sectional, descriptive	58 bereaved siblings (ages 18–46); 5–17years post-death	● Retrospective report of drug use and well-being at various time points: ● Use of illegal drugs increased in the first year after diagnosis compared to before sibling's diagnosis of cancer, but behaviors had declined to baseline by the time of survey ● Same pattern was reported for anxiety and depression ● Almost all siblings (88%) felt that the loss of their siblings still influenced their lives ● Personal growth was reported by numerous siblings (e.g., 36% better communicators, 43% more mature, 17% more confident than peers) ● 50% reported that their sibling's death influenced their education and career choices (in 12% in a negative, 45% in a positive way)	+	NA	+	+	NA	6
Ruegg et al., 2013 [64]	Cross-sectional, descriptive, comparison to norms	695 siblings (mean age 25) of adult (>5years) survivors	● T-scores on all subscales of the physical and mental health domains of the SF-36 ranged around the normative mean of 50 (T-scores: 48 – 56.6)	+	NA	+	+	NA	4
Sahler et al., 1997 [65]	Cross-sectional, case control	170 siblings (mean age 10), 170 mothers; 6–42 months post-diagnosis	● Siblings fell into 3 groups of adaptation: – Dysfunctional (21%), intermediate (46%), and resilient (33%) ● Overall well-being of siblings' mothers was significantly lower compared with control mothers ($d = 0.72$) ● Stratifying mothers by the sibling's adaptation status revealed that mothers with resilient siblings reported the highest well-being scores compared to the other 2 groups	+	NA	+	+	NA	6
Salavati et al. 2014 [66]	Longitudinal, descriptive	111 siblings (ages 8–17); during active treatment <1y post-diagnosis	● Sample was split into less resilient ($n = 56$) vs. more resilient ($n = 54$) siblings based on scores above or lower than 61 on the Child Depression Inventory (less resilient = higher depression) ● Improvement over the course of an 8 weekly group session program was found for less resilient siblings (i.e. higher depressed group) on pre/post tests for depression and anxiety ($d = 0.53$ respectively)	+	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias* Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Sidhu et al., 2006 [67]	Longitudinal, descriptive	26 siblings (ages 8–13); during active treatment	• At baseline: 13% of the sample was "at risk" and 26.7% in "clinical" range on the SRP (Self-Report of Personality) assessing emotional disturbance • Post 4-day intervention: clinical range: 0%, at risk: 22.2% • At two month follow-up: clinical range: $n = 1$, at risk: 28.2% • School and personal adjustment did not change over time; clinical maladjustment and anxiety improved pre- to post treatment and remained stable from post treatment to the 2-months follow-up • Interpersonal issues improved (e.g., perceptions of feeling ignored and misunderstood; not wanting to bother parents with concerns)	–	NA	+	+	+	4
Sloper & While, 1996 [68]	Cross-sectional, descriptive	99 siblings (ages 8–16), 99 parents, n for teachers not reported; 5–10 months post-diagnosis	• 28% ($n = 28$) of siblings in clinical ranges of behavioral adjustment problems; 24 of these 28 showed negative changes since diagnosis (as reported by parents and teachers) • This poor adjustment group ($n = 24$) did not differ from the well-adjusted group regarding their perceptions of own resources (e.g., support, self-esteem), but they had more negative perceptions on the interpersonal interactions and relationships scale ($d = 0.65$)	–	NA	+	+	NA	6
Tao et al., 1998 [69]	Cross-sectional, descriptive	409 siblings (ages 18–41) of adult (>5years) survivors	• 64.3% of siblings had never smoked, 35.7% ever smoked, 31.3% were ever regular smokes, and 20.3% were current smokers. • The pattern (never, former, or current smoker) of siblings was similar to cohort-specific NHIS data (collected in 1990 among people aged 18–24)	+	NA	+	+	NA	6
Van Dongen-Melman et al.,1995 [70]	Cross-sectional, case-control	60 siblings (ages 5–16) of short- (<5y) and long-term (>5y) survivors, 60 parents, 60 matched controls	• No differences between siblings and controls on emotional and behavioral problems • Differences based on gender: male siblings scored lower on extroversion compared with male controls ($n = 25$ each); female siblings had lower somatization and higher social acceptance scores ($n = 20$ each) [insufficient descriptive statistics reported]	+	NA	+	+	NA	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

				Study Bias*					
Study	Design	Sample	Findings	Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Wang & Martinson, 1996 [71]	Longitudinal, descriptive, qualitative	45 siblings (ages 7–16), during active treatment; 30 siblings at 1 year follow-up	• Self-concept, behavioral problems, and family cohesion did not significantly change within 1 year • 3 major stressors: – Inadequate knowledge, manifested in fear of contagion/hereditary ($n = 13$, 28% at baseline, and 5 (16%) at the 1 year follow-up), 5 children had no knowledge about the ill sibling having cancer – Reduced communication: 27 siblings (60%) reported not having the chance to talk about the disease with their parents or the ill sibling. Parent did not want to talk about it or thought the siblings were too young – Insufficient family support: siblings observed physical side effects and negative emotions in the ill child, and described the mother as the saddest one in the family; • 50% of the siblings themselves felt sad and coped with it by crying, going for a walk, thinking about complicated things, being alone	–	NA	+	+	+	4
Wegener Lund et al, 2013 [72]	Cross-sectional, case-control	13105 siblings of survivors (age not reported), 0–19y post-diagnosis	• Siblings were overall not at an increased risk for mental disorders compared to the general population (HR = 0.99/1.01 for brothers/sisters) • Younger siblings (at the time of diagnosis) were at increased risks (highest risk for sibling not even born HR = 1.11/1.59 for brothers/sisters), while those older than 15 by the time of diagnosis were at decreased risks than the general population (HR = 0.79/0.83 for brothers/sisters) • Most common diagnoses: depression, anxiety, neurodevelopmental disorder, emotional and behavioral disorders, personality disorders • Number of hospital contacts were not different for siblings based on the type of diagnosis of the ill child	+	NA	+	+	NA	4
Wellish et al, 2006 [73]	Longitudinal	35 siblings (ages 7–17), 9–166 months post-diagnosis	• No significant changes in affective problems (e.g., negative mood, interpersonal issues), behavioral problems, or activities for siblings over time from pre- to post-camp to 6 month follow-up ($d's = 0.04$ -0.14)	–	NA	+	+	+	4

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 1: Quantitative Studies

Study	Design	Sample	Findings	Study Bias* Adequate Sample Size	Appropriate Blinding	No selective reporting	Appropriate analyses	Incomplete data addressed	Level **
Wengenroth et al, 2014 [74]	Cross-sectional, descriptive	500 siblings of adult (>5y) survivors (ages 20–40)	• No direct comparisons between siblings and healthy controls, but being in a life partnership/ marriage was associated with older age (OR:1.18), and being female (OR: 2.22) among siblings	+	NA	+	+	NA	6
Zeltzer, Dolgin et al., 1996 [75]	Cross-sectional, case-control, comparison to norms	254 siblings (ages 5–18) & their parents; 4–42 months post-diagnosis	• Parents rated siblings as less healthy than parents of controls; • Siblings themselves reported more troubles sleeping and eating than controls[insufficient descriptive statistics reported] • At the same time, parents were less likely to take their children to a physician than controls (for stomachaches, nervousness, or chest pain) • Compared to norm scores, sibling reported higher somatization than the nonclinical norms (*d's* = 0.05-0.54), but lower than clinical normative samples (*d's* = 0.41-0.66). • Health risk behaviors were assessed during interviews with siblings aged 12–18: 8 siblings (35%) reported alcohol use more than once a month and 6 (26%) to tobacco use • General health was rated as similar among siblings and controls (above 90% as excellent)	+	NA	+	+	NA	4
Zelter, Lu et al., 2008 [76]	Cross-sectional, descriptive, comparison to norms	388 Siblings (>18) of adult survivors (Mdn = 23 years post-diagnosis)	• Significant lower scores on depression, anxiety, and somatization than norms (T-scores: 47, 46, and 47 respectively vs. 50 of the norm) • Significant differences for physical functioning (55 vs. 53), general health (53 vs. 51), vitality (45 vs. 49), and mental health (56 vs. 48) • significant higher satisfaction with life (7.35 vs. 6.9) [no SD's reported]	+	NA	+	+	NA	4

* study meets criteria (+); study does not meet criteria (−); criterion not applicable to study (NA);
Criteria: *Adequate sample size*: n>50; *Appropriate blinding*: in case of RCTs or randomized studies; *No selective reporting*: reporting comprehensive, clearly described; *Appropriate analyses*: analyses were appropriate to aims/research questions and sample size; *Incomplete data addressed*: issues with follow-up or missing data clearly described
** 1 = systematic review or meta-analysis of controlled studies; 2 = individual experimental studies (randomized clinical trial); 3 = quasi-experimental studies (non-randomized); 4 = non-experimental studies (case-control, cohort); 5 = systematic reviews of descriptive or qualitative studies; 6 = individual descriptive or qualitative study; 7 = opinions of respected authorities and expert committees;
time:NR = time since diagnosis/end of treatment not reported;
Note: If studies reported sufficient descriptive statistics, Cohen's *d* effect sizes were calculated (If studies reported other effect sizes, these are reported here; i.e., eta^2, HR, OR)

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias*: Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	Level **
Alderfer et al., 2010 [77]	Systematic review	65 empirical papers on the psychosocial adjustment of siblings (1997-2008)	● Siblings do not experience elevated levels of psychiatric symptoms (i.e., behavioral problems, anxiety disorders, or depression); however, the percentage of siblings at risk/clinical ranges is elevated in some samples typically soon after diagnosis. ● Siblings commonly report negative emotional reactions, an absence of positive emotions, and symptoms of cancer-related post-traumatic stress ● Qualitative studies indicate that siblings experience a loss of attention and status within the family, changes in family roles and relationships, and disruptions in the sense of security a family typically provides after their brother or sister is diagnosed with cancer ● Quantitative studies are scant and do not provide information on whether the family environment of siblings differs from the general population ● There is some evidence for siblings experiencing school problems within 2 years of diagnosis ● Social problems (e.g., friendships, withdrawal, aggression) are rarely documented among siblings, but when they are, they are noted within 2 years of diagnosis ● Somatic complaints and physical functioning do not seem to be poorer for most siblings; however, siblings in early childhood may be at some risk for somatic distress within 2 years of diagnosis ● Two positive themes have emerged across qualitative studies: (a) increased responsibility, independence, and maturity of siblings; and, (b) increased empathy, sensitivity, and compassion ● Adolescent siblings seem to show the poorest adjustment compared to adults, school age, and preschool children but such data is preliminary ● Female siblings may exhibit more distress than males ● There are many methodological limitations of the current evidence base	+	+	+	+	+	5

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias*: Right papers included	Study Bias*: Relevant studies included	Study Bias*: Quality assessed	Study Bias*: Reasonably combined results	Study Bias*: Important outcomes considered	Level **
Birenbaum, 1995 [78]	Integrative research review	18 research articles and dissertations conducted by nurses (1983-1993)	Three broad themes were identified in family research in pediatric oncology nursing: ● (1) Parenting concerns: siblings are noted (in one study) to be concerned about the absence of parenting ● (2) Family communication: one study is reviewed indicating that open communication within the family around the time of a child's death is associated with fewer behavioral problems in the siblings ● (3) Stress, coping and adjustment: one study is reviewed that indicates siblings report significant life-events stress scores ● Pediatric oncology nursing research regarding the family is limited based upon methods currently used (e.g., poor measures, single reporters, cross-sectional designs)	+	+	–	–	+	5
Duran, 2013 [79]	Narrative synthesis of qualitative and quantitative research	35 studies produced between (1975-2010)	Five main themes were distilled across the papers regarding posttraumatic growth in childhood cancer survivors and their families: ● (1) making sense of the cancer experience: siblings are noted to report that their brother or sister has" a special purpose in life" because they were cured of cancer; others report that the event was random ● (2) appreciation of life (not reported regarding siblings) ● (3) greater self-knowledge: siblings were reported to note that the survivors are now more courageous, more self-confident, or more conscious of taking care of their health, that mothers became stronger dealing with cancer and that they, too, gained inner strength and self-confidence ● (4) positive attitudes toward family (not reported regarding siblings) ● (5) a desire to "give back to society" and help others ● It was also noted that data regarding healthy siblings' positive experience were very scarce	+	+	+	+	+	5

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias*					Level **
				Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	
Houtzager et al., 1999 [80]	Systematic review	35 empirical papers on the psychosocial adjustment of siblings; (1979-1997), excluding bereavement	● Emotional reactions: Some studies found that siblings experience elevated distress (posttraumatic stress, anxiety, and general adaptation problems), but findings are mixed ● Behavioral adjustment & social competence: There are frequent mixed findings, though larger, more recent studies found more behavioral and social problems in siblings than control groups. Behavioral and social problems appear to decline over time. Prevalence of sibling adjustment problems was estimated to be 25–69%. ● School-related problems: Multiple studies noted school problems in siblings, including absenteeism, poorer academic performance, and school-related distress. Two studies noted poorer school functioning in siblings than patients. ● Physical effects: Siblings' physical responses include headaches, stomachaches, trouble sleeping, and trouble eating; however, findings are mixed regarding whether levels of physical symptoms are higher in siblings than norms/controls. ● Positive effects: Many siblings show positive effects of the cancer, including enhanced maturation/growth, sensitivity, empathy, supportiveness, responsibility, caring, compassion, and independence ● Psychosocial variables related to adjustment: Siblings tended to show more problems adapting to a brother's/sister's cancer if they had pre-existing behavioral, emotional, and/or family problems (i.e., present before the child's cancer diagnosis); perceived the illness to be more threatening and/or to result in more social or family effects; and had fewer coping resources (i.e., open and honest communication about the cancer, adequate information about the illness and treatment, and involvement in the treatment process).	+	+	+	+	+	5

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias*					Level **
				Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	
			• Socio-demographic variables related to adjustment: mixed findings regarding sibling age, though several studies suggest that the nature of sibling adjustment varies by age. Siblings may show better adjustment when they come from larger families (i.e., with more siblings) with higher SES. • Family variables related to sibling adjustment: Siblings may show better adjustment in the presence of high parental social support and better parental wellbeing. • Illness-related variables related to sibling adjustment: Few research studies have reported on associations between illness characteristics and sibling outcomes.						
Long & Marsland, 2011 [81]	Systematic review of qualitative and quantitative studies	17 studies about family adjustment (1996-2009)	• Psychosocial impact on siblings is poorly understood • Cancer does not necessarily lead to increased sibling psychopathology • Siblings fall into average levels of psychopathology symptoms, but the percentage of siblings falling into clinical ranges is greater than that expected • No study on role of family functioning on sibling adjustment	+	+	−	+	+	5
Martinuik et al., 2014 [82]	Systematic review	20 studies of camp programs for survivors and siblings (2001-2013)	• Camps can help siblings explore their interests and enhance physical and emotional skills (incl. self-esteem)	+	+	+	+	+	5

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias*					Level **
				Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	
Masera et al., 1999 [83]	Guideline for health care professionals working with terminally ill children and their families		● The parents, the siblings, and the ill child (depending on their ages and level of development) should be included in the final decision-making processes ● Encourage post death follow-up visits for parents and siblings, reflecting on the medical history of the child and acknowledging the ongoing needs of parents, siblings, and other family members (e.g., grandparents). ● Offer bereavement counselling after the death of a child (for parents and siblings), encourage participation in self-help groups	NA	NA	NA	NA	NA	7
Mavrides & Pao, 2014 [84]	Narrative review of qualitative and quantitative studies	Unknown N of studies related to pediatric oncology (2000-2013)	● Siblings are likely to feel that their needs are less important, while they experiences intense emotions (i.e. shock, helplessness, or sadness) ● Feelings of jealousy, anger, withdrawal, isolation, disruptions in normal routines, school problems are common	–	–	–	–	+	5
Meyler et al., 2010 [85]	Systematic review	21 studies of family-based interventions for childhood cancer (1980-2008)	● Describes four studies that included siblings ● Literature on family-based interventions is too limited and too much variability among intervention to make informed judgments regarding particular interventions ● Effectiveness of family-based interventions: findings are encouraging, with the ● Majority of interventions being associated with positive effects for participants	+	+	+	+	+	5
O'Brien et al., 2009 [86]	Systematic review of quantitative studies	17 studies on the impact of childhood illnesses on siblings (2000-2008)	● 6/17 studies included siblings of children with cancer ● Conflicting results: some studies reported well adjustment in siblings; others found high levels of posttraumatic and psychological stress, suggesting the need for identification of those at risk and prove mental health services ● Female siblings may be specifically affected	+	+	–	–	+	5

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias*					Level **
				Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	
Packman et al, 2010 [87]	Review of qualitative and quantitative studies	Studies on the impact of HSCT on the whole family (time line not reported)	● Sibling donors are at increased risk for emotional disturbances (e.g., PTSS, anxiety, low self-esteem) ● Behavior problems more likely among donor vs. nondonor siblings ● Donor siblings are likely to feel lonely, not fully understand the donation process, and feel a lack of attention from parents ● Unsuccessful transplants can result in feelings of guilt ● Social support and developmentally appropriate provision of information can be specifically helpful for both donor and nondonor siblings	+	+	–	+	+	5
Pedro et al., 2008 [88]	Integrative review	15 studies on emotional support for families (1996-2006)	● Emotional, instrumental, and informational support are most beneficial for siblings ● Reduced support is associated with psychosocial problems, such as resentment, jealousy, anxiety, depression, behavioral problems	+	+	–	+	+	5
Prchal & Landolt, 2009 [89]	Systematic review	14 studies on psychosocial interventions for siblings (1980-2008)	● Included 14 studies about 11 different interventions (3 camp/7 group/1 individual) ● Interventions were focused on enhancing (a) medical knowledge, (b) coping, (c) family communication, (d) reducing PTSS, € providing peer support ● Generally improvements were found for depression, negative mood, medical knowledge, and HRQoL ● Findings are inconclusive regarding changes in anxiety, behavior problems, social adjustment, PTSS, and self-esteem over the course of sibling programs ● Most interventions could not demonstrate an improvement in communication about illness ● Siblings and parents were generally highly satisfied with programs ● Surprising that individual settings are so rare, since programs need to be developmentally appropriate and relevant to the individual sibling	+	+	–	+	+	5

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

				Study Bias*					
Study	Design	Sample†	Findings	Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	Level **
Scott et al., 2003 [90]	Systematic review	5 intervention studies for siblings (1966-2003)	● Four studies evaluated structured group interventions, finding increased knowledge after participation, understanding, reductions in interpersonal problems, preoccupation, and increased disease-related communication, mood ● One study evaluated support group sessions (vs. a group without intervention) finding no significant differences in social adjustment between both groups ● generally, evidence is too weak based on 5 studies	+	+	+	+	+	5
Spinetta et al., 1999 [91]	Guideline for assisting siblings		Issue → recommendations: ● (1) Feelings of isolation → address the need for support of siblings with parents; encourage parents in how to involve siblings from the beginning ● (2) Need for communication → parents should be encouraged to: Discuss sibling's own day, include siblings in discussion about diagnosis and treatment/continue to update sibling, bring siblings to hospital discussions should take the developmental stage of the sibling into account; topics for discussion: feelings of guilt, fears, school problems, somatic complaints, acting out, loss of childhood, loss of playmate, loss of family cohesion, changes, additional chores, anger, uncertainty about the future; support groups can be helpful ● Specific phases of the treatment may need specific interventions/support	NA	NA	NA	NA	NA	7

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

				Study Bias*					
Study	Design	Sample†	Findings	Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	Level **
Van Dongen-Melman, 2000 [92]	Recommendation/guideline for psychosocial care for families		● Siblings seem least effected (compared to ill child, mothers, and fathers) ● Studies about potential late effects in siblings are sparse (4 studies cited); it is indicated that although siblings have emotional problems during treatment, they are not at a heightened risk for (psychosocial) late effects, but individual siblings/subgroups may have lingering concerns and problems (which are usually bound to the relationship with their parents and position within the family) ● If problems are apparent, they don't seem to decline over time ● Short-term counseling facilitating communication could help alleviate problems ● Help parents address their relationship with the well siblings (re-entry into normal life after treatment completion with surviving and well children)	NA	NA	NA	NA	NA	7
Wilkins & Woodgate, 2005 [93]	Systematic review of qualitative studies	27 studies about sibling's experiences (1979-2005)	● Included 26 qualitative studies about siblings; findings were categorized into 3 themes: – Changing lives (losses, gains) – Intense feelings (sadness, loneliness, rejection, anxiety, anger, jealousy, guilt) – Unmet needs (family communication, information, involvement in care of the sick sibling, support to maintain interests and activities) ● Critique of the qualitative studies: Lack of conceptualization, research design, research methods	+	+	+	+	+	5

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 2: Reviews and guidelines

Study	Design	Sample†	Findings	Study Bias* Right papers included	Relevant studies included	Quality assessed	Reasonably combined results	Important outcomes considered	Level **
Wilkins & Woodgate, 2006 [94]	Systematic review	Several studies used for a concept analysis (1960-2006)	● Concept analysis of the meaning of "transition" for siblings ● Transition should be viewed as a process-oriented phenomenon in response to change ● Usually studies focused on transition in response to a specific event (i.e. diagnosis or death): ● It seems especially difficult for siblings to adapt to having an ill sibling, and to sudden and extended separations from the ill sibling and parents (after diagnosis) ● In case of death, siblings can experience as very individual grief process and usually need to overcome the transition from loving a brother/sister in presence to loving them in absence	+	+	−	+	+	5
Williams et al., 2014 [95]	Systematic review	19 nursing studies on perspective of parental caregiving	● Healthy siblings are significantly affected by upheavals from normalcy during a cancer diagnosis ● Sibling are often separated from parents and ill child; and experience a lack of attention ● Parents described needing grandparents or friends to care for the healthy siblings	+	+	−	+	+	5

† number of studies refers to the studies included in the review, but does not necessarily reflect the number of studies about siblings
* study meets criteria (+); study does not meet criteria (−); criterion not applicable to study (NA);
Right papers included: right types of papers were included; *Relevant studies included*: important, relevant studies to the scope of the review were included; *Quality assessed*: included studies were appropriately assessed for their quality; *Reasonably combined results*: appropriate to combine results in the presented way; *Important outcomes considered*
** 1 = systematic review or meta-analysis of controlled studies; 2 = individual experimental studies (randomized clinical trial); 3 = quasi-experimental studies (non-randomized); 4 = non-experimental studies (case-control, cohort); 5 = systematic reviews of descriptive or qualitative studies; 6 = individual descriptive or qualitative study; 7 = opinions of respected authorities and expert committees

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias*: Research question clear	Study Bias*: Approach justified	Study Bias*: Context described	Study Bias*: Appropriate sampling	Study Bias*: Data collection/ analyses appropriate	Level of Evidence**
Barbarin et al., 1995 [96]	Cross-sectional, part of a larger mixed methods study, interviews	179 parents (of 179 families) of 254 siblings (ages 4–18), 6–42 months post-diagnosis	● Before diagnosis, 40% of siblings were reported to have problems; after diagnosis, 60% of siblings were reported to have problems Problems of general adaptation increased from 2.8% to 13.8 % Internalizing problems increased from 7.5 to 14% Externalizing problems increased from 4% to 12% Social problems decreased from 8% to 1.6% Academic difficulties decreased from 9.5% to 6% Sibling conflict increased from 2.8% to 4% ● Of the 137 siblings who had difficulties before the diagnosis, 21% improved after the diagnosis, 45% did not change and 34% became worse ● Of the 141 siblings who had no problems prior to the diagnosis, 56% developed problems ● In regard to independence, maturity, and supportiveness, 56% of siblings were reported to improve, 34% did not change, and 10% declined ● Sibling sex, birth order, and gender were unrelated to frequency and type of problems before and after diagnosis ● Siblings over age 10 compared to those under age 10, first born siblings compared to latter born siblings and siblings of children with more severe disease were more likely to increase in maturity and independence	+	+	−	−	+	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias*					Level of Evidence**
				Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	
Barrera et al., 2013 [97]	Longitudinal, interviews	25 bereaved parents at 6 months & 15 at18 months post-death (sibling age not reported)	● Siblings expressed grief through missing the deceased child, expressing mixed emotions including sadness and anger, behavioral problems (temper tantrums for young siblings; risky behaviors for teens), and through difficulties with daily functioning (e.g., school work). ● Moving on with life, talking about the deceased child, social support from friends and groups support were seen as helpful ● Many parents across sibling ages reported positive, open relationships. However, some parents of young siblings indicated that demands for parental attention increased and that parents were sometimes blamed for the child's death. Some older siblings were reported to become overly concerned for their mothers' well-being ● Siblings showed a continued bond with the deceased child through using the deceased belongings, choosing to study something related to illness, dreaming about the deceased, including the deceased in pretend play, and scapegoating the deceased	+	+	−	−	+	6

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	 Approach justified	 Context described	 Appropriate sampling	 Data collection/ analyses appropriate	Level of Evidence**
Foster et al., 2012 [98]	Cross-sectional data from a larger mixed methods longitudinal study, interviews	36 mothers, 24 fathers, 39 siblings (ages 8–17) of 40 bereaved families, 6–19 months post-death	● 68% of participants reported personal changes in siblings: – Changes in sibling's personality (e.g., increased maturity, compassion, withdrawal, anger, sadness, fearfulness) noted by 61%, 54%, and 44% of mothers, fathers, and siblings respectively – Changes in sibling's attitude toward and interest in school work (both positive and negative examples) noted by 28%, 13%, and 26% of mothers, fathers, and siblings – Changes in the goals and life perspective of the sibling (e.g., motivation, life priorities) reported by 8%, 17%, and 21% of mothers, fathers, and siblings – Changes in sibling's activities and interests (both increased and decreased interest) reported by 8%, 4%, and 8% of mothers, fathers, and siblings ● 47% of all participants noted changes in siblings' relationships with family members and peers: – Changes in the dynamics of the sibling's peer relationships (both positive and negative) 14%, 13%, and 33% of mothers, fathers, and siblings – Increased closeness of family relationships was reported by 19%, 17%, and 23%; while 3% of mothers, 4% of fathers, and 8% of siblings reported decreased closeness – Changes in the sibling's role within the family was noted by 25%, 8%, and 18% of mothers, fathers, and siblings ● 21% of participants (17% of mothers, 13% of fathers, and 15% of siblings) reported no changes in siblings attributed to the death	+	+	–	–	+	6

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	Level of Evidence**
Dighe et al., 2008 [99]	Cross-sectional, interviews	20 parents of children with incurable cancers	• Parents rarely told siblings about the patient's illness • Siblings had unmet information needs	+	–	–	–	–	6
Freeman et al., 2000 [100]	Cross-sectional, interviews, focus groups	19 siblings (ages >5), 9–48 months post-diagnosis	• Younger siblings (< 10 years; interviews) reported helping their affected brother/sister, feeling scared, having less time with parents and less homework help, and having to cancel family activities • Older siblings (≥ 10 years; focus groups) reported jealousy, changes in peer and community support, health-related fears, over-protection, and empathy	+	+	+	+	–	6
Jenholt et al., 2014 [101]	Longitudinal, semi-structured interviews	14 siblings (ages 9–22)	• 16–28 weeks person-centered intervention for siblings • Pre-intervention: 5 themes included – grasping for knowledge about cancer, – thinking for hours and having nightmares, – experiencing physical pain, – bring emotional in several ways, – waiting for a normal, good life despite the uncertain future • Post-intervention: Siblings reported having more knowledge about cancer and indicated partial improvement of sleep problems, physical pain, and strong emotions. They continued to wait for a better future	+	+	+	–	–	6
Leavitt, 1999 [102]	Longitudinal, semi-structured interviews	18 parents, 4–6 months & 1 year post-diagnosis	• Ill child is family priority and parents noted their lack of attention to healthy siblings, poor school performance, moodiness, hypersensitivity, and social withdrawal	+	+	–	+	–	6
Long, 2015 [103]	Cross-sectional, semi-structured interviews	30 siblings (ages 10–17), 7–33 months post-diagnosis & on treatment	• Two-part process of realizing the seriousness of cancer and taking action for stabilization; themes include: • Knowing something is wrong, figuring out the meaning of cancer, adapting to changes, handling emotional reactions	+	+	+	+	+	6

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SUPPLEMENTAL TABLE II. **(*Continued*)**

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	Level of Evidence**
MacLeod et al., 2003 [104]	Cross-sectional, semi-structured interviews	8 siblings & 7 bereaved sibling donors (ages 12–28), 2–15 years post-HSCT	● Main themes: – Perceptions of “no choice” – Importance of psychological aspects related to an HSCT (e.g., fear) ● Themes reported by siblings in successful HSCTs ($n = 8$): – Donation had positive impact on many life domains (e.g., relationships, world view, positive feelings of self, insight into illness), and negative effects were rarely reported ● Themes reported by siblings in unsuccessful HSCTs ($n = 7$): – Feelings of guilt and blame, anger, hard not to feel responsible for the death	+	+	+	+	+	6
Murray, 1999 [105]	Cross-sectional, triangulation, in-depth interviews	25 nurses	● Nurses acknowledged the need for emotional support in siblings, but also barriers for nurses, such as workload, time, access to siblings, and bureaucracy	+	+	+	+	+	6
Murray, 2002 [106]	Cross-sectional, written response to 2 open-ended questions	43 siblings (ages 7–12), during active treatment	● Siblings reported need for: – emotional support (42%; $n = 18$) – instrumental support (28%; $n = 12$) – informational support (21%, $n = 9$) – appraisal support (9%, $n = 4$)	+	+	–	+	+	6
Nolbris & Hellstroem, 2005 [107]	Cross-sectional, semi-structured interviews	10 bereaved siblings (ages 4–21), 1.5-6 years post-death	● Siblings expressed several problems and concerns: – Dissatisfaction with information and support ($n = 10$) – Loneliness ($n = 6$) – Reactions (anxiety, anger, jealousy, $n = 8$) – Places of refuge (e.g., cope, school, friends, $n = 10$) – Mourning ($n = 10$) – Memories (e.g., time at hospital, visiting grave, festivals, $n = 10$)	+	+	+	–	+	6

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias*					Level of Evidence**
				Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	
Nolbris et al., 2010 [108]	Cross-sectional, semi-structured interviews	9 siblings & 6 bereaved siblings (ages 8–19), 6 months -11.7 years post-diagnosis	● Evaluation of a sibling support group identified 3 broad categories: – Belonging to a group (e.g., talking about situation, helpful to hear other's feelings) – Importance as a member of the group (e.g., allowed to talk or remain quiet) – Therapeutic support helpful to recall and understand memories	+	+	+	−	+	6
O'Shea et al., 2012 [109]	Cross-sectional, semi-structured interviews, focus groups	13 nurses	● Sibling needs: – Getting attention, – Wanting to know, – Wanting to help, – Wanting a normal routine	+	+	+	−	+	6
Packman et al., 1997 [110]	Cross-sectional, semi-structured interviews	44 siblings (ages 6–18), 44 parents, 34 months post BMT	● Sibling donors ($n = 21$) were most likely to feel proud, but also nervous, curious, and scared before transplant, while after transplant they were most likely to report feelings like being relieved, proud, calm, or lonely (not needed anymore) ● The majority of donors did not feel like they had a choice, and older donors emphasized the importance of being involved in the decision-making process ● A minority of these donors ($n = 6$, 29%) talked to someone about these feelings, and handled stress with withdrawal; however, 81% would do it again ● Nondonors ($n = 23$) were most likely to feel calm, scared, relieved, sad, or nervous for not being picked to donate marrow; again only a minority ($n = 8$, 33%) talked about these feelings with someone ● Siblings in both groups felt most stressed at the onset of ill siblings' disease ● Parents recommended seeking support (in the form of support groups or camps) for siblings	−	+	−	+	+	6

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	Level of Evidence**
Pelletier et al., 2014 [111]	Cross-sectional, interviews	44 siblings (ages 9–22; incl. half siblings, & cousins), pre-HSCT	● Siblings (half-siblings and cousins) reported the following themes: – Believing HSCT is the best chance for cure: $n = 15$ (34%) – Accepting the physician's recommendation: $n = 15$ (34%) – Desiring a more normal and better quality of life: $n = 19$ (43%) – Trusting in God: $n = 11$ (25%) – Seeking advice or consultation: $n = 13$ (30%) – Fearing death without HSCT: $n = 8$ (18%) – Doing personal research: $n = 6$ (14%) – Having a perfect match: $n = 5$ (11%) – Trusting the physician: $n = 3$ (7%) – Expecting a better future: $n = 1$ (2%) – HSCT being part of the upfront treatment plan: $n = 1$ (2%) – Hearing of HSCT success in others: $n = 1$ (2%)	+	+	–	+	–	6
Quin, 2004 [112]	Cross-sectional, interviews	38 siblings (ages 3–21), 74 mothers, 46 fathers, >2 years post treatment	● Positive and negative effects on siblings ● Changes in siblings: fear, resentment, attention-seeking, guilt, worry, independence, protectiveness in relation to the ill sibling, problems at school, bed-wetting, and increased physical ailments ● Some parents felt that the sibling had suffered more than the ill child ● Siblings were asked what they would ask for if they had three magical wishes → wide range of wishes including material changes, meeting famous people, special holidays, ambitions, and 19% related to cancer (e.g., that the illness never happens again)	+	+	–	+	–	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	 Approach justified	 Context described	 Appropriate sampling	 Data collection/ analyses appropriate	Level of Evidence**
Roberson, 2010 [113]	Cross-sectional, semi-structured interviews and observation	25 siblings observed during camp; interviews with 4 siblings of survivors off treatment & 4 bereaved (ages 9–14)	● Types of social support identified by sibling campers: – Emotional support (encouragement, expressing emotions) – Informational support (clarification about ill sibling's condition) – Instrumental support (others being helpful, gaining coping skills) – Appraisal support (relate to others at camp) – Social comparison (sense of belonging)	+	+	+	−	+	6
Sargent et al., 1995 [114]	Cross-sectional, structured interviews	254 siblings (ages 5–18), 6–42 months post-diagnosis	● Biggest change for sibling since diagnosis: being more compassionate and caring (16%), received less attention (14%), family separations and disruptions (13%), no changes (13%) ● Biggest change for family: separation and disruptions (18%), family became closer (16%) ● Worst thing about having an ill sibling: treatment and effects (39%), fear of death (14%) ● Anything good because of cancer?: nothing positive (23%), closer to family members (19%) ● Has cancer affected healthy sibling more/same/or less than ill sibling?: affected the same (53%), self more affected than ill sibling (27%) ● What have you handled best?: helping ill sibling/family (27%), controlling own feelings (13%), understanding and accepting (8%) ● What have you handled lest well/ worst?: treatment and effects (24%), own negative feelings (20%) ● Answers differed by age, but not gender; older siblings were more likely to report positive changes than younger siblings	+	+	+	+	+	6
Sirkia et al., 2000 [115]	Cross-sectional, structured interview (94 questions)	70 bereaved parents, 1–8 years post-death	● Study compared parents-reports of siblings after terminal care ($n = 77$) vs. 34 siblings who had an ill sibling dying during active anticancer therapy ● 14 (18%) siblings in the terminal care group vs. 11 (32%) siblings in the active therapy group had problems related to fear, anxiety, school, behavior, and friends	+	−	−	+	−	6

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SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	Level of Evidence**
Sloper, 2000 [116]	Longitudinal, semi-structured interviews	94 siblings (ages 8–16), time 1: 5–10 months post-diagnosis, time 2: 9–15 months later	● 98% of siblings reported 1 or more problems 6 months after diagnosis: loss of attention and status; loss of own and families' usual activities and routines; loss of certainty and security; and loss of companionship of the ill child ● Most problems had resolved 18 months after diagnosis (2/3 felt they were not treated differently than their ill sibling) ● Important resources: supportive relationships, information about illness and treatment, continuing with own interests and activities ● Positive gains: maturation, understanding and compassion, closer family relations	+	−	−	+	+	6
Steele et al., 2013 [117]	Cross-sectional, semi-structured interviews	39 bereaved siblings (ages 8–17), and 60 parents from 40 families, 6–19 months post-death	● 13 siblings (33%) had no advice for health care professionals ● Siblings provided advice about how medical teams could communicate more effectively with them and noted the need to be included in a developmentally appropriate manner. ● Siblings also echoed the statement that their brothers and sisters want to be treated like normal kids ● Siblings emphasized the importance of visiting their brother or sister in the hospital.	+	+	−	+	+	6
Thompson et al., 2011 [118]	Cross-sectional, semi-structured interviews	39 bereaved siblings (ages 8–17) & 60 parents from 40 families, 6–19 months post-death	● Grief is very individual/personal ● Parents and siblings key to managing grief: social support (especially family as source of support), self-expression, faith/religion, and memories ● Important to be open and honest with one's feelings and find an outlet to express them	+	+	−	+	+	6

(*Continued*)

SUPPLEMENTAL TABLE II. (***Continued***)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	 Approach justified	 Context described	 Appropriate sampling	 Data collection/ analyses appropriate	Level of Evidence**
von Essen & Enskar, 2002 [119]	Cross-sectional, interviews with 2 open-ended questions	97 parents, 105 nurses, >1 month post-diagnosis	● What caring aspects are important for siblings? – Social competence ($n = 43$ parents; $n = 50$ nurses) – Participation ($n = 45$; 45) – Information ($n = 45$; 31) – Emotional support ($n = 22$; 19) – Amusement ($n = 20$; 18) – Family life ($n = 20$; 16) – Do not know ($n = 7$; 27) ● What help does the sibling need outside the hospital? – Emotional support ($n = 35$; 30) – No help ($n = 35$; 19) – Fair attention ($n = 22$; 21) – Family life ($n = 21$; 20) ● Practical support ($n = 18$; 6)	+	+	−	+	+	6
Wakefield et al., 2013 [120]	Cross-sectional, semi-structured interviews	15 siblings (ages 14–38), 13–88 months post-diagnosis	● Sources of support after treatment completion: – Parents were rated as primary source of support ($n = 14$) – Along with sibling-sibling relationship – Friends ($n = 12$) ● In addition, positive appraisal for: hospital staff ($n = 2$), oncologist ($n = 1$), camp ($n = 4$), make a wish foundation ($n = 9$), Redkite ($n = 2$)	+	−	−	+	+	6
Wiener et al., 2008 [121]	Cross-sectional, structured interviews	14 sibling donors (ages 9–28), 4–48 months after stem cell donation	Main themes: ● Preparation for donation: 64% of siblings were informed by parent and 86% received specific information by medical staff about stem cell process; 1/3 preferred more extensive preparation ● Anxiety varied; younger donors (<18) were less likely to be anxious; donors were usually anxious about pain due to various procedures ● Transplant outcome ● Life changes: positive changes reported by 67% (e.g., closer relation with ill siblings, parents, sense of pride for donating)	+	+	−	+	−	6

(*Continued*)

SUPPLEMENTAL TABLE II. (***Continued***)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias* Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	Level of Evidence**
Williams et al., 2009 [122]	Cross-sectional, written answers to 1 open-ended question	29 parents (of siblings ages 6–15), time:NR	"Cancer in one of my children has had the following effects on the brother/sister without cancer." ● 5 negative themes: jealousy, worry, anger, depressed, school problems ● 3 positive: family closeness, greater sensitivity, growth	+	−	−	−	+	6
Woodgate & Degner, 2004 [123]	Cross-sectional, interviews, observation	39 families at various stages (sibling age and time:NR)	● Siblings often teased the ill child when they complained and thought they were 'putting on an act' to get extra attention. ● Siblings needed to restrict extracurricular activities ● Siblings would do whatever they could to make their ill sibling feel more comfortable, but 'taking care' of ill children became the priority of mothers, although many fathers and siblings wanted to be more involved. Fathers and siblings experienced a sense of loss ● Fathers and siblings also reported sadness, and that they had to give up some normal family activities. They realized though that their support was important for the mother and ill child and viewed their support as necessary to protect the ill child from more suffering and the family from falling apart	+	+	−	+	+	6
Woodgate, 2006 [124]	Cross-sectional, interviews, observation	39 families at various stages (sibling age and time:NR)	● During interviews: any question about siblings' experiences often resulted in them talking about their ill sibling ● All siblings had experienced the same significant loss: loss of relationships ● Within the family; they felt like outsiders, watching their family from a distance. ● It seemed like their sense of self did not matter or exist. ● Siblings were only concerned with the ill child's needs and would put these before their own ● Siblings downplayed their contribution to helping their ill brother or sister and expressed a sense of loss about their lack of involvement in the care	+	+	+	+	+	6

(*Continued*)

SUPPLEMENTAL TABLE II. (*Continued*)

Part 3: Qualitative Studies

Study	Design	Sample	Findings	Study Bias*					Level of Evidence**
				Research question clear	Approach justified	Context described	Appropriate sampling	Data collection/ analyses appropriate	
Woodgate, 2006 [125]	Cross-sectional, interviews, and observation	30 siblings (ages 6–21), time:NR	● Cancer was experienced as a different way of being within the family which included a loss of a family way of life and loss of self ● The different way of being in the family was underlined by 3 themes: – committing to keeping my family together – being present – enduring sadness	+	+	+	+	+	6

* study meets criteria (+); study does not meet criteria (−); criterion not applicable to study (NA);

Research question clear: the research question/aim of the study was clearly stated; *Approach justified*: the qualitative approach was clearly justified; *Context described*: the study context and role of the researcher(s) was clearly described; *Appropriate sampling*: the sampling strategy was appropriate for the research question/aim of the study; *Data collection/analyses appropriate*: the method of data collection and subsequent analyses were clearly described, and adequate for the scope of the study;

**1 = systematic review or meta-analysis of controlled studies; 2 = individual experimental studies (randomized clinical trial); 3 = quasi-experimental studies (non-randomized); 4 = non-experimental studies (case-control, cohort); 5 = systematic reviews of descriptive or qualitative studies; 6 = individual descriptive or qualitative study; 7 = opinions of respected authorities and expert committees

time:NR = time since diagnosis/end of treatment not reported

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