

Review

# Psychological interventions with siblings of pediatric cancer patients: a systematic review

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## Abstract

**Objective:** Siblings of pediatric cancer patients have been shown to be at risk for developing emotional, behavioral, and social problems. There is a need for psychological interventions in this population. Several researchers have previously documented and evaluated their interventions with siblings. This paper aimed at reviewing the existing reports of evaluated psychological interventions with siblings of pediatric cancer patients and at outlining future directions.

**Methods:** Research was conducted on several online bibliographic databases. Articles were selected on the basis of predefined criteria. If possible, effect sizes (ES) were calculated.

**Results:** Fourteen studies representing 11 different sibling interventions met criteria for inclusion. One individual intervention, three camps, and seven groups were found. Objectives of interventions concentrated mainly on enhancing siblings' coping and improving their medical knowledge. In terms of outcome measures, most of the studies focused on psychological adjustment variables. Findings showed significant improvements in siblings' depression symptomatology, medical knowledge, and health-related quality of life. Findings were inconsistent with regard to anxiety, behavioral problems, social adjustment, self-esteem, and posttraumatic stress symptoms. Depending on the outcome variables, small to large ES were found. Satisfaction with the intervention was high in both siblings and parents.

**Conclusion:** There is tentative evidence that psychological interventions with siblings of childhood cancer patients can effectively reduce psychological maladjustment and improve medical knowledge about cancer. However, the number of studies is small, and several methodological shortcomings have to be noted. In future, more randomized controlled trials need to be conducted in larger samples to extend the evidence base.

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## Introduction

Diagnosis and treatment of childhood cancer is a stressful experience for all family members, including healthy siblings. As a result of the need to care for the ill child the entire family is forced to change daily life and routines. Understandably, the parents' attention becomes focused on their ill child, whereas healthy siblings are confronted with decreased availability of their parents [1]. Siblings may also be worried about the illness, and they have to observe their brother or sister undergo emotional and physical pain. Many siblings experience intrusive and conflicting emotions such as feelings of fear, isolation, jealousy, or guilt [2].

Most psychological research in pediatric oncology includes either patients alone or patients and mothers, with a lack of attention to siblings, fathers, or others in the child's social ecology [3]. This focus of research and literature on the sick

child may reflect the siblings' position in the family during the illness process [4]. However, the research on the psychological adjustment of siblings of cancer patients has grown over the past three decades. Houtzager *et al.* [4] reviewed the literature on siblings' adjustment to childhood cancer and found that most siblings do not seem to suffer from severe psychopathology. However, there is growing evidence indicating that siblings of children with cancer suffer significant psychological distress. *Internalizing problems* such as anxiety and depression [2,5,6] as well as *externalizing behavioral problems* [5,7] have been reported. In addition, siblings suffer from *problems at school* [8,9], *psychosomatic complaints* [5,7,10], *lower quality of life* [5,11], and *posttraumatic stress (PTS)* [12,13]. Yet, siblings have also reported some *positive effects*, including maturation, understanding, compassion, and closer family relationships [14,15].

In sum, siblings of pediatric cancer patients have been shown to be at risk for developing several

emotional, behavioral, and social problems, and most clinicians and researchers agree that there is a need for psychological interventions for this population [16,17]. However, the majority of published papers on siblings of pediatric cancer patients still report the results of descriptive and correlational research, and there is a great deal of non-empirical, anecdotal data in this field. Similar to the whole field of pediatric psycho-oncology, intervention research in siblings is rare [18]. Nevertheless, several authors have documented sibling programs and have examined the effects of the interventions. The present paper reviews empirical evidence on the effectiveness of psychological interventions with siblings by summarizing outcome variables and calculating effect sizes (ES) whenever possible. Further, the review describes objectives and settings of existing interventions and gives recommendations for future research in this field.

## Method

### Data sources and search strategy

Relevant electronic databases were searched for published empirical studies and dissertations evaluating psychological interventions with siblings of pediatric cancer patients. The databases searched were Medline, EMBASE, CINAHL, PsycINFO, Cochrane Systematic Reviews, and ProQuest Digital Dissertation. Searches were conducted in September 2008 for the period 1980–2008.

The searches were carried out using the following search terms for the patient population: (1) *sibling* (in title); (2) *pediatric, paediatric, child, adolescent*; (3) *neoplasm, oncology, cancer*. Search terms for the intervention were *intervention, camp, group, weekend, support, counsel, education, therapy* (in title or abstract). The Boolean operator 'and' was used to combine the three patient population identifiers and identifiers for intervention. The operator 'or' was used to combine identifiers within the three population areas and the intervention area.

The initial literature search yielded 449 hits with 431 articles and 18 dissertations. In a pre-selection process, completed by the first author, titles and abstracts were screened for the existence of psychological interventions. Thirty-five articles and dissertations remained. First and second authors checked the full text of pre-selected studies for inclusion criteria according to a standardized checklist and obtained agreement in case of uncertainty. Additionally, reference lists of relevant studies and reviews were examined to identify other pertinent articles. Doctoral dissertations were examined only if they were accessible in full text.

### Study selection

Under examination were studies evaluating the effect of interventions aimed at improving the psychological adjustment of siblings of childhood cancer patients. Studies were included if they met the following criteria: (a) *type of studies*: evaluation of an intervention with quantifiable data about the effect of the intervention, (b) *type of outcome measures*: standardized and validated outcome measures with self- or proxy report of psychological adjustment and/or quality of life and/or satisfaction with intervention and/or medical knowledge, (c) *types of participants*: children and adolescents aged 0–18 years, sister or brother diagnosed with childhood cancer, not more than 50% bereaved children in the sample, sample size at least 10, and (d) *type of intervention*: standardized and sibling-specific intervention program, content of intervention described, specific for childhood cancer. Sibling programs with a majority of bereaved children were excluded because we assumed that they focus mainly on grief and loss rather than on coping with specific sibling stressors. If necessary, corresponding authors were contacted for additional information or clarification of the text. Reasons for exclusion were recorded (Table 1).

**Table 1.** Excluded studies

| Study                     | Reason for exclusion  |
|---------------------------|---|
| Adams-Greenly et al. [19] | 1   |
| Ballard [20]              | 1; 2  |
| Bedway and Hartkopf       | 1; 2; 3   |
| Smith [21]                |   |
| Bordeur [22]              | 1; 2; 4; intervention unclear   |
| Bendor [23]               | 1; 2: age group up to 22 years  |
| Carpenter et al. [24]     | Same intervention, data set, and outcome variables as Sahler and Carpenter [54]   |
| Creed et al. [26]         | 1; only bereaved children   |
| Cunningham et al. [27]    | 1   |
| Fanos et al. [28]         | 1; 3 (percentage of childhood cancer patients unclear); age unclear   |
| Kazak et al. [29]         | Sample size too small   |
| Kinrade [30]              | 1   |
| Kramer and Moore [31]     | 1   |
| Lobato and Kao [32]       | 3   |
| Lobato and Kao [33]       | 3 (cancer population 1.4%)  |
| Olin [34]                 | Outcome measure: stress measured by cortisol concentration in children's saliva, unclear if sibling-specific intervention |
| Packman et al. [35]       | 1   |
| Ruffin et al. [36]        | 1; 4  |
| Simms et al. [37]         | 1; 2  |
| Wellisch et al. [38]      | Unclear if sibling-specific part existent   |
| Williams et al. [39]      | 3 (cancer population 36.4%)   |
| Williams et al. [40]      | 3 (cancer population 8.7%)  |

1, Outcome measure not standardized/validated. 2, Intervention not standardized. 3, Intervention not specific for childhood cancer. 4, Intervention not sibling specific.

Most papers (62%) had to be excluded because outcome measures were not standardized and validated.

### Data extraction

Data were extracted by the main author from each publication into a standardized data collection form. Information about study design, sample size, age, and months since diagnosis were recorded. Further, information about the specific sibling interventions (setting, number of sessions) and about outcome variables were extracted. We subdivided outcome variables into variables assessing psychological adjustment (depression, anxiety, social adjustment, self-esteem, behavioral problems, and PTS symptoms) and quality of life. In addition, we examined findings based on the Sibling Perception Questionnaire (SPQ) by Carpenter and Sahler [9], a widely used measure for siblings' responses to childhood cancer. The SPQ was developed to assess school-aged siblings' responses in the three domains *perceptions of impact of the illness* (subscales intrapersonal responses, interpersonal relationships, fear of disease, and communication about the illness), *affective responses* (positive and negative mood), and *medical knowledge* (instrumental and identification). There are no normative data available for this measure as yet, but the instrument is being used increasingly in studies with siblings of children with cancer [41]. Finally, siblings' and parents' satisfaction with the intervention program was an outcome variable examined (all self-developed scales).

The comparison of ES across different studies makes it possible to judge the strength of the finding across different interventions, sample characteristics, or methods. Unfortunately, the data to be analyzed for computing ES are often not readily available from the study reports. Of the reviewed studies only two [42,43] reported ES in their original papers. Pre/post-ES (Cohen's *d*) were calculated as the within-group difference between pre-intervention and post-intervention means divided by the weighted pooled pre/post-standard deviation for the intervention group. In addition, between-group ES were computed whenever possible, comparing the difference between pre/post-mean for the experimental and the comparison group divided by the pooled standard deviation. All ES were calculated by the present author from the data given in the research reports, whenever the necessary information were available. Positive ES represent improvements in the desired direction. Cohen [44] suggests that an ES of 0.2 is indicative of a small, 0.5 of a medium, and 0.8 of a large effect.

## Results

Fourteen studies (10 published articles and 4 dissertations) met the inclusion criteria, representing 11 different sibling interventions. Two studies [43,45] are based on the same data set but present other outcome variables. The main characteristics of the included studies are summarized in Table 2. ES are presented in Table 3.

### Description of studies

#### Origin

The studies reviewed were published between 1986 and 2005. Eight publications are from the United States [25,43,45,46,49,50,53,54], three from Canada [42,47,48], and one each from Israel [51], the Netherlands [52], and Australia [41], respectively.

#### Study design

Ten studies used pre/post-intervention designs. Only three publications are based on randomized controlled trials (RCTs) to evaluate the intervention [46,49,53]. One study provided a non-randomized control group in addition to the pre/post-evaluation [25].

#### Sample characteristics

The sample sizes varied from 11 to 90 siblings with an age range from 6 to 20 years. All studies but one assessed the effect of the intervention exclusively among siblings. Kazak *et al.* [53] evaluated an intervention with cancer survivors and their families including siblings, but they used sibling-specific material when working with siblings. Time since onset of cancer in the ill child varied within and between all studies from months to several years. Five publications did not report time since diagnosis [25,42,43,45,47].

### Types of interventions

#### Setting

Seven of the 11 different intervention programs used a group setting [25,42,47–53], with 3–10 sessions and group size from 4 to 12 siblings. Three interventions were provided in a camp format [41,43,45,54] with the camp duration of 4–7 days. Only one intervention was designed as an individual intervention [46]. In two intervention programs parents were involved [50,53].

#### Lead professional

The interventions were provided by a variety of professionals, including psychologists [42,43, 45,47–49,52], nurses [46], or interdisciplinary teams [25,41,50,51,53,54].

**Table 2.** Summary of studies included in the review

| Study                                   | Design  | N          | Age (years) | Months since diagnosis | Setting | Sessions | Intervention objective                          | Outcome measures  |
|---|---------|------------|-------------|------------------------|---------|----------|---|---|
| Atherton [46]                           | RCT     | 18 (9/9)   | 8–13        | < 6                    | Single  | 2        | Medical knowledge, coping                       | = anxiety (s) <sup>b</sup>  |
| Barrera <i>et al.</i> <sup>1</sup> [47] | pp      | 12         | 6–17        | —                      | Group   | 8        | Medical knowledge, coping                       | +depression (s) <sup>a</sup><br>+anxiety (s)/ = anxiety (p) <sup>b</sup><br>= behavior (s, p) <sup>kj</sup><br>+SPQ (s)/ = SPQ (p) <sup>o</sup><br>+satisfaction (s, p) <sup>q</sup>                            |
| Barrera <i>et al.</i> <sup>1</sup> [42] | pp      | 47         | 6–14        | —                      | Group   | 8        | Medical knowledge, coping                       | +depression (s) <sup>a</sup><br>+anxiety (s, p) <sup>b</sup><br>= behavior (s)/+behavior (p) <sup>kj</sup><br>+satisfaction (s, p) <sup>q</sup>   |
| Chung <sup>1</sup> [48]                 | pp      | 25         | 6–17        | 1–128                  | Group   | 8        | Medical knowledge, coping                       | +depression (s) <sup>a</sup><br>= anxiety (s)/+anxiety (p) <sup>b</sup><br>= self-esteem (s) <sup>g</sup><br>= behavior (p) <sup>j</sup><br>+SPQ (s)/–SPQ (p) <sup>o</sup><br>+satisfaction (s, p) <sup>q</sup> |
| Cimini [49]                             | pp; RCT | 30 (15/15) | 7–14        | < 12                   | Group   | 3        | Medical knowledge, coping                       | +depression (s) <sup>a</sup><br>+anxiety (s) <sup>cd</sup><br>= behavior (p) <sup>j</sup><br>+satisfaction (s) <sup>pq</sup>  |
| Dennis [50]                             | pp      | 11         | 8–13        | 3–36                   | Group   | 10       | Medical knowledge, coping, family communication | = anxiety (s, p) <sup>c</sup><br>= self-esteem (s) <sup>h</sup><br>+SPQ (s) <sup>o</sup><br>+satisfaction (s) <sup>pq</sup>   |
| Dolgin <i>et al.</i> [51]               | pp      | 24         | 7–17        | 4–48                   | Group   | 6        | Medical knowledge, coping, family communication | +SPQ (s) <sup>o</sup><br>+satisfaction (s) <sup>q</sup>   |
| Heiney <i>et al.</i> [25]               | pp; ct  | 14         | 9–15        | —                      | Group   | 7        | Medical knowledge, coping                       | = social adjustment (s) <sup>f</sup><br>+satisfaction (s) <sup>q</sup>  |
| Houtzager <i>et al.</i> [52]            | pp      | 24         | 7–18        | 2–89                   | Group   | 5        | Medical knowledge, coping                       | +anxiety (s) <sup>b</sup>   |
| Kazak <i>et al.</i> [53]                | RCT     | 43 (19/24) | 10–20       | 12–146                 | Group   | 4        | Coping, family communication, PTS reduction     | = anxiety (s) <sup>c</sup><br>= PTS (s) <sup>i</sup>  |
| Packman <i>et al.</i> <sup>2</sup> [43] | pp      | 77         | 6–17        | —                      | Camp    | 1 week   | Coping, recreation                              | +anxiety (s) <sup>c</sup><br>+self-esteem (s) <sup>i</sup><br>+PTS (s) <sup>m</sup><br>+HRQoL (s) <sup>n</sup>  |
| Packman <i>et al.</i> <sup>2</sup> [45] | pp      | 77         | 6–17        | —                      | Camp    | 1 week   | Coping, recreation                              | +HRQoL (s)/ = HRQoL (p) <sup>n</sup>  |



|                           |    |    |      |         |      |        |                                       |  |
|---------------------------|----|----|------|---------|------|--------|---------------------------------------|--|
| Sahler and Carpenter [54] | pp | 90 | 6–17 | 4–156   | Camp | 5 days | Medical knowledge, coping, recreation | +SPQ (s) <sup>o</sup>  |
| Sidhu [41]                | pp | 26 | 8–13 | App. 12 | Camp | 4 days | Medical knowledge, coping, recreation | +anxiety (s) <sup>a</sup><br>+social adjustment (s) <sup>b</sup><br>+self-esteem (s) <sup>c</sup><br>+SPQ (s) <sup>o</sup> |

1, Same intervention but different data set; 2, same intervention and data set but different outcome variables; RCT, randomized controlled trial; ct, controlled trial; pp, pre- and post-intervention assessment; s, self-report; p, proxy report; + indicates significant improvement in the desired direction compared with control/pre-assessment; -- indicates significant impairment compared with control/pre-assessment; = indicates no significant difference between intervention and control group or no significant change from pre- to post-assessment. <sup>a</sup>Children's Depression Inventory (CDI) [55]; <sup>b</sup>State-Trait Anxiety Inventory for Children (STAIC) [56]; <sup>c</sup>Revised Children's Manifest Anxiety Scale (RCMAS) [57]; <sup>d</sup>Fear Survey Schedule for Children Revised (FSSC-R) [75]; <sup>e</sup>Self-Report of Personality (SRP), Subtest of Behavior Assessment System for Children (BASC) [58]; <sup>f</sup>Social Adjustment Scale (SAS) [59]; <sup>g</sup>Self-Perception Profile for Children (SPPC) [60]; <sup>h</sup>Piers-Harris Children's Self-Concept Scale [61]; <sup>i</sup>Rosenberg Self-Esteem Scale (RSE) [62]; <sup>j</sup>Child Behavior Checklist [63]; <sup>k</sup>Youth Self-Report (YSR) [63]; <sup>l</sup>Post-Traumatic Stress Disorder Reaction Index (PTSD-RI) [66]; <sup>m</sup>UCLA PTSD Index [64]; <sup>n</sup>Pediatric Quality of Life Inventory (PedsQL) [65]; <sup>o</sup>Sibling Perception Questionnaire (SPQ) [9]; <sup>p</sup>No psychometric evaluation; <sup>q</sup>Self-developed scales.

**Table 3.** Effect sizes *d* and mean effect sizes for measures of depression, anxiety, behavioral problems (behavior), posttraumatic stress (PTS), health-related quality of life (HRQoL), and subscales of the Sibling Perception Questionnaire (SPQ)

|                                      | Depression |  | Anxiety |      | Behavior |      | PTS  |  | HRQoL |      | SPQ intrapersonal |       | SPQ interpersonal |      | SPQ communication |      | SPQ fear of disease |       |
|--------------------------------------|------------|--|---------|------|----------|------|------|--|-------|------|-------------------|-------|-------------------|------|-------------------|------|---------------------|-------|
|                                      | s          |  | s       | p    | s        | p    | s    |  | s     | p    | s                 | p     | s                 | p    | s                 | p    | s                   | p     |
| Barrera <i>et al.</i> [47]           | 0.47       |  | 0.98    | 0.68 | 1.45     | 0.35 |      |  |       |      | 1.12              | -0.11 | 0.62              | 0.05 | 0.31              | 1.45 | 1.0                 | -0.19 |
| Barrera <i>et al.</i> [42]           | 0.40       |  | 0.48    | 0.69 |          |      |      |  |       |      |                   |       |                   |      |                   |      |                     |       |
| Chung [48]                           | 0.31       |  | 0.36    | 0.95 |          | 0.39 |      |  |       |      | 0.65              | 0.19  | 0.11              | 0.11 | 0.22              | 0.27 | 0.48                | -0.48 |
| Dolgin <i>et al.</i> [51]            |            |  |         |      |          |      |      |  |       |      | 1.37              |       | 0.87              |      | 0.57              |      | 0.0                 |       |
| Kazak <i>et al.</i> [53]             |            |  | 0.50    |      |          |      |      |  |       |      |                   |       |                   |      |                   |      |                     |       |
| Packman <i>et al.</i> [43,45]        |            |  | 0.44    |      |          |      | 0.47 |  | 0.32  | 0.12 |                   |       |                   |      |                   |      |                     |       |
| Mean effect size of outcome variable | 0.40       |  | 0.55    | 0.77 | 1.45     | 0.37 | 0.47 |  | 0.32  | 0.12 | 1.05              | 0.04  | 0.53              | 0.08 | 0.37              | 0.86 | 0.49                | -0.34 |

s, self-report; p, parent report.

### Theoretical basis

Description of interventions in the selected publications varied considerably with respect to details, content, and procedure. Various theoretical frameworks were given as basis for the sibling intervention. Some authors designed their program on the basis of psychotherapeutic conceptual models such as CBT [42,47,48], group therapy [25,50], or family therapy [29,53]. Others conducted a pre-intervention survey about sibling's needs [41,51] or chose their specific content areas based on clinical and research literature [43,45,46,49,54].

### Objectives

To examine specific types of intervention, we divided the reviewed studies into five categories according to the objectives of the program: Enhancement of (1) medical knowledge, (2) coping, and (3) family communication, (4) reduction of PTS, and (5) providing peer support through the experience of recreational activities. Various types of intervention objectives were found. Enhancement of coping with sibling-specific stressors and emotions was an important objective in all selected intervention programs. Nine of the examined interventions focused also on improvement of medical knowledge [25,41,42,46–52,54], that is, siblings were provided with information about pediatric cancer and its treatment. Three intervention programs tried to achieve better family communication [50,51,53]; one program aimed to reduce PTS [53]. Finally, three interventions, all in a camp setting, had their major focus on recreational activities [41,43,45,54] in order to provide siblings with peer support.

### Time since onset of cancer

Three programs defined time since onset of cancer in the ill child as an inclusion criterion for participating siblings. The intervention presented by Cimini [49] required the patient to be under treatment for at least 1 year. Kazak *et al.* [53] wanted patients to have completed treatment for at least 1 year. Only the intervention evaluated by Sidhu *et al.* [41] targeted siblings of children with cancer in an early stage of medical treatment and therefore has a preventive character.

### Effectiveness of interventions

#### Psychological adjustment

Self-reported *depression* was assessed in four studies [42,47–49], including one RCT study [49], all of them using the Children's Depression Inventory (CDI) [55]. The results suggest a significant reduction in siblings' depressive symptoms after the intervention. All the corresponding studies were provided in a group format, and three

are based on the same intervention program [42,47,48]. ES range from 0.31 to 0.47, indicating a small beneficial effect. No study assessed proxy-reported depression.

Ten publications used self-reported *anxiety* as an outcome measure. The majority of studies assessed anxiety using either the State-Trait Anxiety Inventory for Children (STAIC) [56] or the Revised Children's Manifest Anxiety Scale (RCMAS) [57]. Six publications (including one RCT) found a reduction in self-reported anxiety due to the intervention [42,43,47,49,52]; four studies (including two RCTs) did not find significant differences [46,47,50,53]. No conclusive differences between studies with or without anxiety reduction could be found concerning intervention setting or number of subjects. Proxy-reported anxiety decreased in two studies [42,48] and showed no significant differences in two other studies [47,50]. All four studies were in group format and used a pre/post-intervention design; however, the two with a decrease of anxiety included more subjects. ES for anxiety among all studies allowing calculation varied from small to large (ES 0.36–0.98).

Two articles evaluated self-reported *social adjustment* before and after the intervention. No significant improvement could be found by Heiney *et al.* [25], using a non-equivalent control group, whereas the other study by Sidhu *et al.* [41] reported improvement between pre- and post-measurements. No ES were available for measures of social adjustment.

Sibling-rated *self-esteem* was examined in four publications. Two using a camp format found enhanced self-esteem ratings after participation in a sibling intervention [41,43]. Two studies in group format reported no significant change [48,50]. Sample sizes were larger in the studies with enhanced self-esteem. No ES were available for measures of self-esteem.

Self-reported and proxy-reported *behavioral problems* were assessed in four publications, all using the Child Behavior Checklist for parents or the Youth Self-Report (YSR) for adolescent siblings' reports [63]. With the exception of Barrera *et al.* [42], who found a significant reduction in parents' report of behavior problems from post-intervention to follow-up, none of the other studies [47–49], including one RCT, found any significant changes in siblings' behavioral adjustment, and only trends in the desired direction were reported. Little data were available for calculating ES (ES 0.35–1.45). The very large ES of 1.45 for siblings' self-reported behavior assessed by Barrera *et al.* [47] must be interpreted with caution, because this figure is based on a subset of only seven siblings.

Two of the reviewed studies used self-reported PTS as outcome variables [43,53] assessed using

either the Child Post-Traumatic Stress Disorder Reaction Index [66] or the UCLA PTSD Index [64]. Findings were inconsistent. Whereas Packman *et al.* [43] presented a significant reduction in PTS symptoms from pre to post in their camp intervention, no changes could be found in the RCT by Kazak *et al.* [53], who conducted a group intervention. ES could be computed only for one study [43] showing a small effect (ES 0.47).

Eleven of the 14 reports focused mainly on psychological adjustment variables as outcome measures. Proxy-raters were parents in all cases. Whereas the majority of studies report significant changes in at least one of the observed adjustment variables, four studies did not find changes in siblings' psychological adjustment after treatment [25,46,50,53].

### Health-related quality of life

The influence of the intervention on siblings' quality of life was examined in two publications [43,45], both using the Pediatric Quality of Life Inventory (PedsQL) [65], an instrument measuring *health-related quality of life* (HRQoL) in children and adolescents. Both studies showed significant improvements in self-reported HRQoL from pre- to post-camp. Packman *et al.* [45] found significant changes in proxy reports from non-bereaved parents. Calculated ES (data from only one study available) were small (ES 0.32) for self-reports and showed no effect for parents' reports.

### Sibling Perception Questionnaire

The SPQ [9] was used in six publications, and all of them found significant changes in the desired direction in at least one of the SPQ subscales. Only two of the studies [51,54] used all three domains of the SPQ, whereas others applied only the domain 'perception of impact of illness' [41,47,48] or the 'medical knowledge' domain [50]. Siblings' *affective responses* (positive and negative mood) improved significantly after the intervention in sibling and parent reports [51,54]. Results also indicated significant beneficial effects in *medical knowledge* as reported by siblings [50,51,54]. Regarding the domain *perception of impact of the illness* the findings are inconsistent. In the sibling-rated *intrapersonal* subscale, a measure of how illness affects the individual, significant improvements after intervention could be demonstrated in four [47,48,51,54] out of five studies. Parent reports showed no significant amelioration in intrapersonal responses to illness [47,48]. The available ES reinforced these results with medium to large ES for sibling-reported intrapersonal responses (ES 0.65–1.37) and with no effect in parent-reported intrapersonal responses. In the *interpersonal* subscale, a measure of interactions

and relationships with others, three publications found no significant changes in sibling reports [47,48,54], and two found significant improvement [47,53]. Parent reports again showed no improvement after the intervention [47,48]. ES for self-reported interpersonal relationships showed a wide range from no effect to large intervention effects (ES 0.11–0.87); based on parent reports, ES were negligible. Concerning the subscale *communication about illness* only one study found significant improvements in sibling-reported communication [51]; four other publications failed to find a significant change [41,47,48,54]. Barrera *et al.* [47] found significant improvements in parents' appraisal of communication, but Chung [48] in turn could not find such improvements. ES for sibling and parental report of communication were small to large (ES 0.22–1.45). Finally, evaluation results concerning the SPQ subscale *fear of disease* in siblings were inconsistent among the studies. Remarkable is a significant increase in siblings' fear of disease reported by parents in the study by Chung [48]. According to this, ES varied considerably in self-reported fear of disease (ES 0–1) and showed no effect or a negative effect in proxy reports.

### Satisfaction with intervention

Satisfaction measures are used to obtain the participants' feedback about the value and quality of an intervention program. Seven studies applied quantitative measures of satisfaction, all administered at the final session or at follow-up [25,42,47–51]. In four studies data were collected from both parents and siblings [42,47–49]. Overall, four different satisfaction instruments were used. Only the instrument used by Barrera *et al.* [42,47] and Chung [48] was tested with regard to its reliability and validity. Overall, satisfaction measures revealed in both siblings and parents a generally high level of satisfaction with the interventions provided. In addition to the quantitative evaluation, three studies conducted a qualitative analysis of intervention effects on the basis of semi-structured interviews with siblings and parents [45,48] or follow-up focus groups with parents [41]. On these qualitative measures both parents and siblings reported a clearly positive impact of the sibling intervention, thus underlining the results from quantitative satisfaction surveys. Findings from the qualitative evaluation suggested increased communication between siblings and better medical knowledge about cancer in siblings as well as a decreased sense of isolation [48]. Packman *et al.* [45] found that the majority of responses emphasized the importance of group cohesiveness, shared personal experiences, socializing, and the relieving of emotions by expressing one's feelings.



### Predictors of intervention effects

In seven studies predictors of intervention effects were examined [42,43,45,47,49,52,54]. They discuss the influence of age, sex, diagnosis, bereavement, number of camp attendances, or pre-existing behavioral problems on intervention effects. Houtzager *et al.* [52] found that anxiety in siblings of children with leukemia or lymphoma decreased more after group participation than anxiety in siblings of children with a solid or a brain tumor. Barrera *et al.* [42] identified age and gender as crucial factors influencing the effect of treatment, indicating that young boys are the most receptive to their intervention with greatest reduction in depression. In comparison, Cimini [49] examined the impact of age and gender on measures of anxiety and depression and could not find any differences. Sahler and Carpenter [54] found that with respect to medical knowledge, their camp seemed to be most beneficial to younger and returning campers. Further, they reported that siblings with behavioral problems seemed to benefit most from attendance at a camp. Packman *et al.* [43] found that there was a slightly higher benefit for first-time campers regarding PTS and HRQoL compared with siblings who had participated in a camp previously. Finally, Packman *et al.* [45] revealed that bereaved parents reported siblings' HRQoL as less positively influenced by a camp as compared with non-bereaved parents.

## Discussion

### Summary of results

This study aimed at reviewing existing reports on the effects of psychological interventions with siblings of pediatric cancer patients. Fourteen studies representing 11 different sibling interventions met criteria for inclusion. The reviewed studies included 10 pre/post-evaluations, 3 RCTs, and 1 non-randomized control group design. Three interventions were conducted in camp settings and seven in group settings; one study described an individual intervention.

Looking at the intervention setting, it may be surprising that individual interventions are so rare, since sibling programs need to be developmentally appropriate and relevant in addressing personal concerns and individualized cancer-related information. These goals seem to be best met in an individual context in which one can emphasize on the needs of the sibling. On the other hand, the use of a group or camp format can give siblings the sense that they are not alone, and meeting others with similar experiences can itself have a positive impact on psychological adjustment. Further, the use of a group or camp format is cost effective. Unfortunately, the existing studies do not allow a

comparison of the efficacy of group versus individual format. The only single intervention included in the review showed no changes in psychological adjustment. In other fields of clinical child psychology, group and individual treatments could be shown to be equally effective in reducing children's affective symptoms [67,68].

Outcome evaluations revealed improvements in depressive symptomatology, negative mood, medical knowledge, and HRQoL in siblings following the intervention. Findings were inconsistent with regard to anxiety, behavioral problems, social adjustment, self-esteem, PTS symptoms, and perception of the illness. Four out of 14 studies found no quantitative change in outcome variables after the intervention. These four studies represent different study designs, settings, and intervention objectives.

These findings need to be interpreted with caution because just a minority of studies used control groups to evaluate their outcome. However, by only looking at results of RCTs, findings are still inconsistent. Two out of three RCTs found no significant changes in their outcome variables [46,53]; one RCT reported amelioration in depressive symptoms and anxiety post-intervention [49].

The reduction of depressive symptoms and amelioration of mood state in the present review leads to the assumption that sibling interventions have a positive influence on siblings' emotional condition. However, the ES for depressive symptoms were rather small. Based on the assumption that siblings that are provided with accurate medical information cope with and adapt to the situation more effectively [69,70], enhancement of medical knowledge was an important goal in the majority of the intervention programs reviewed. Only three studies assessed siblings' knowledge before and after the intervention, but they all found significant improvement on this variable. Increase in medical knowledge can also be found after interventions for siblings of children with a chronic illness or a disability [32,40]. There is tentative evidence that by gaining information about illness and treatment, siblings may experience enhanced feelings of control and therefore feel more secure and less anxious [71,72]. In the present review anxiety was the outcome measure most often used, but only half of the studies found an anxiety reduction post-intervention.

Depending on outcome variables different ES were found. Out of 37 ES calculated, 28 showed small to large effects ( $ES \geq 0.20$ ) and 8 showed no effect of intervention. Only one negative effect was found [48]. The issue of parent-reported significant increase in fear of cancer found by Chung [48] might be a result of the siblings discussing their fears of cancer with their parents more openly after their participation in the sibling intervention. This explanation was supported by qualitative analyses



conducted [48]. In general, fewer proxy reports were given, and parents tend to estimate intervention effect less positively as compared with siblings.

Despite great diversity in their design, objectives, and lead professionals, sibling interventions produced positive effects on psychological adjustment, siblings' knowledge, and HRQoL. With respect to siblings' and parents' satisfaction with the intervention, the bulk of evidence is quite positive. The overall results of the present review support the efficacy of sibling interventions and reveal no significant negative effects.

### Limitations of previous studies

There are several methodological issues that limit generalization of the findings. First, 10 out of 14 publications evaluated intervention effects with a sample size smaller than 50. Several researchers complained that they could not recruit a larger number of participants due to the small number of potential subjects available in this population. Second, only a minority of studies used RCTs even though this is the gold standard for intervention evaluations [73,74]. A problem with the widely used pre/post-design is the fact that positive changes of outcome measures may be due to natural effects of time or maturation rather than intervention effects. Several researchers explained the absence of a control group citing ethical and practical reasons. With regard to the ethical concerns of not providing siblings in a control group with the opportunity to participate in an intervention, there is still the possibility of a waiting list design or two assessments before providing the intervention. Third, few authors reported ES, and some did not even deliver relevant data in their report to calculate ES; thus, comparison across different types of interventions is difficult. Fourth, the great variety of outcome measures and instruments make comparisons quite difficult, and predictors have been assessed only unsystematically. Fifth, most of the involved sibling interventions are complex and have multiple components, but none of the study designs allows identification of the most effective components of the intervention. And, finally, only two studies [41,42] conducted follow-up assessments for evidence of the consistency of the intervention effect. However, long-term follow-ups would be useful to see whether the benefits remained after termination of the program.

Concerning intervention timing, further issues should be raised. Many of the studies used a broad inclusion criterion for the length of time since onset of cancer or do not take into account time since diagnosis at all. This way interventions cannot be tailored to needs and stressors associated with particular stages of treatment (initial time period after diagnosis, end of treatment, relapse, and so

on). Regarding intervention documentation, several of the reviewed studies did not describe their interventions sufficiently (for example, detailed program content, theoretical basis, existence of a manual, or lead professional are not specified). This lack of documentation limits the possibility of replication and comparability.

### Directions for future research

Based on the results of this review, several recommendations can be made regarding future research in this field. Intervention studies should increase in their methodological quality and be conducted in randomized controlled designs. To overcome the problem of small sample sizes more multisite collaborative studies should be conducted. Sibling programs should be manualized and theory based. Relationships among the dependent variables and covariates (age, gender, diagnosis, for example) should be examined further, and there is a need to distinguish content components of the intervention and evaluate them apart. Finally, direct comparison between different settings (individual versus group), their psychological outcome, and their cost effectiveness are needed.

In sum this review showed tentative evidence that psychological interventions with siblings of childhood cancer patients can effectively reduce psychological maladjustment and improve medical knowledge about cancer. However, detailed recommendations regarding most effective intervention objectives or settings cannot yet be made.

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