

Psychosocial Outcomes in Siblings of Pediatric Cancer Patients

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### Abstract

Research in pediatric psychology has scarcely begun to investigate the psychosocial needs of siblings of pediatric cancer patients. The purpose of this literature review was to discuss the articles that have investigated the psychosocial needs of pediatric cancer patients as well as the articles that suggest promising intervention strategies to help these young individuals. The articles discussed investigated the effects on healthy-siblings during the cancer treatment, the outside factors that influence the amount of distress a sibling may suffer with, the differences in reactions and adjustment between nonbereaved and bereaved siblings, and the possible intervention strategies including increased social support, family-oriented rehabilitation programs, psychological interventions, educational interventions, and experiences in camp environments designed specifically for siblings of pediatric cancer. Much progress has been made in this domain of pediatric psychology; however, further research should be conducted to provide the maximum amount of support for siblings of pediatric cancer patients.

### Psychosocial Outcomes in Siblings of Pediatric Cancer Patients

Numerous studies have been conducted on the psychological well-being of children treated for pediatric cancer, as well as the impact their illness has on their behavior and psychological adjustment in survivorship. While these studies have proven beneficial in helping us understand these individuals, very little research has been conducted that focuses on their siblings. The research that has been conducted with the siblings of these young cancer survivors, although small in comparison to the survivors themselves, provides information that demonstrates that these individuals experience distress and may require psychosocial support. The current review paper will overview the psychological effects on the siblings during cancer treatment, the effects of the relationships on the sibling's ability to cope, the effects on the siblings after the child has completed cancer treatment, the impact on the sibling when the child has not survived treatment, as well as possible intervention strategies that have shown promise in aiding the siblings of pediatric cancer patients. By reviewing these areas of study on siblings, the field of pediatric psychology may be able to better understand the challenges these children face, predict the obstacles they may encounter, and provide families with promising strategies for healthier psychological functioning for each member of the family.

Although relatively few studies have investigated the effects of pediatric cancer on siblings, research has shown that the siblings of pediatric cancer patients experience varying forms of distress after the patient has been diagnosed with cancer. A study conducted by Houtzager, Grootenhuis, Hoekstra-Weebers, and Last (2005) examined the quality of life (QoL) of siblings of pediatric cancer patients and variables that predict QoL. They evaluated their coping strategies and referenced the quality of health (physical problems, sleep difficulties, eating issues, etc.) for each sibling prior to the diagnosis of cancer based on the child's and the

parent's statements. Their sample included siblings between the ages of seven and 18, with measures administered one month to eight weeks after the diagnosis. Various scales were used in this study to measure current QoL and coping strategies. In addition, the parents of the children were interviewed to determine the sibling's health prior to the diagnosis of pediatric cancer in the family. Houtzager et al. (2005) then compared the responses from the siblings of pediatric cancer to responses from their non-cancer associated peers.

The results of this study found that many of the siblings of pediatric cancer reported considerably impaired emotional and cognitive QoL. As well, over half of the siblings reported a significant decrease in positive emotions felt since the diagnosis in comparison to their peers who did not have a sibling diagnosed with cancer. They also found that the siblings whose parents reported them to have had health issues prior to the diagnosis were similarly reported to have more struggles with cognitive and motor functioning as well as more negative emotions after the diagnosis occurred. The researchers also found that siblings who were given more positive feedback about the likelihood of recovery coped with the diagnosis better and experienced better cognitive functioning as well as more positive emotions and less negative emotions than those who were not given explanations. Interesting differences between age and gender were also found in the study; older siblings experienced significantly more negative emotions than the younger siblings, while sisters tended to be less satisfied with their relationships with family members and peers in comparison to brothers. These results were found only one month after the diagnosis, thus there could be differences in such findings after more time has passed.

Interestingly, Houtzager, Grootenhuis, Caron, and Last (2004) had conducted a prior study investigating the psychosocial influences on siblings after two years had passed since the

diagnosis. In this study, the same age group as in the previously mentioned study was examined along with a group of peers who did not have a sibling with pediatric cancer. The sibling participants were administered two quality of life (QoL) questionnaires as well as measures of anxiety and emotional and behavioral problems. The findings from this study are interesting in comparison to the study conducted only a month or two following diagnosis. Houtzager et al. (2004) found that for the majority of individuals, most of the acute emotional distress that followed the diagnosis of cancer seems to normalize after two years passed. However, there appeared to be a subgroup of siblings who did not seem to cope as well with the illness. They discovered that nearly half of the younger siblings tended to report a lower overall QoL at this point in the survivorship process, and that nearly a third of the older siblings experienced internalizing depression, anxiety, or social withdrawal. Houtzager et al. (2004) reasoned that this may be the result of older children being more self-sufficient and able to find outside support from peers, whereas the younger siblings only have their parents and close family to turn to in their time of need. The younger siblings also tended to experience significant levels of physical health issues after the diagnosis, perhaps because the younger children are unable to fully understand the illness of the pediatric cancer patient and therefore are at a higher risk of acquiring great fears related to the illness (Houtzager et al., 2004). Although these explanations are plausible, they are speculative and further research should be conducted to investigate why some children are more resistant to the stress of having a cancer patient in the family while others are not so fortunate.

Other studies with siblings of patients with cancer have investigated other psychosocial outcomes, including posttraumatic stress symptoms. Kaplan, Kaal, Bradley, and Alderfer (2013), examined a group of sibling participants between the ages of eight and 17 whose family member

was within four to 38 months after the diagnosis of cancer. The participants completed two scales of PTSD symptoms and anxiety as well as a child depression inventory in order to measure the prevalence, common symptoms, and comorbidities. Over half of the sample scored in the moderate to severe range for PTS symptoms, reported feeling upset and avoiding the thoughts and conversations about the cancer, and were found to have extreme levels of arousal symptoms, with the symptoms interfering with daily functioning. In addition, they experienced comorbidity with anxiety and depression. Approximately 22 percent of the sample also satisfied criteria for full-blown PTSD based on the scale used in the study (Kaplan et al., 2013). The authors concluded that further research should be done in this area to find ways to prevent such symptoms as well as to find the best course of action for treating cancer-related PTS symptoms.

While the previously mentioned studies have proven useful in identifying specific psychosocial outcomes, Sargent et al. (1995) used interviews with siblings and their parents to gain a broader understanding of how cancer affected them and their families. To meet criteria for the study, the cancer diagnosis had to have taken place six to 42 months prior to participating. During the interviews, the participants were asked nine questions. The first eight questions concerned the siblings' thoughts and feelings towards the changes they experienced within themselves and in the family, the possible negative and positive effects of the cancer, the reactions of other siblings in the family, and the best and worst coping strategies they have found to be associated with pediatric cancer. The ninth and final question asked the sibling to provide some advice for siblings who would one day be in the same situation.

The researchers found a gender by age interaction, with each group tending to have a specific area that was more difficult for them than for the other groups. When comparing the two age groups (younger children versus older children), the older siblings tended to have more

positive feedback about the experience than the younger siblings, which was believed to be the result of differing maturity levels. The younger siblings seemed to struggle more than the older siblings when it came to responding to the questions, which could be due to lack of cognitive development to understand abstract concepts. However, a lack of such development could also lead some to argue that the younger siblings are also unable to fully understand the situation, less capable of sharing their true feelings, and may be more likely to become frustrated by the entire situation due to these contributing factors. The authors concluded that the level of maturity and the ability to manage the daily stressors that are associated with pediatric cancer are good predictors of the siblings' positive or negative outlook over the entire situation. The study also found that the group at the highest risk for problems were the younger brothers. This information is important because it allows the families to educate themselves on what to look out for and how to help the siblings by identifying the specific areas that their age group and gender tend to struggle with the most.

All of the previously reviewed articles explored domains of adjustment for siblings of pediatric cancer after the initial diagnosis. Although these results are helpful, other areas of investigation help to fully understand the influence of cancer on these individuals, the obstacles that they face, and to identify the strategies to help them. While the initial diagnosis itself can cause great distress in the cancer siblings, there are other factors that can be investigated to possibly predict the coping outcomes of the siblings of pediatric cancer including family risk factors and the child's level of empathy.

The amount of research exploring factors that lead some siblings to be more susceptible to distress after the diagnosis of cancer is minimal in comparison to other cancer studies. In one of the few studies in this area, Long, Marsland, and Alderfer (2013) investigated family risk

factors (family functioning problems, parental acceptance, psychological control, and parental PTSS) and how siblings of pediatric cancer were affected by them. To meet criteria for this study, families had to have a child with cancer currently receiving treatment or the diagnosis had to have occurred within two years prior to the study with the child still living and the child needed to have a sibling between the ages of 8 and 18. The authors used the Post-Traumatic Diagnostic Scale, the Family Assessment Device, the Child Report of Parent Behaviors Inventory, the Child Depression Inventory, the Revised Children's Manifest Anxiety Scale, and the Child PTSD Symptom Scale in this study. They then examined the associations between sibling distress, family risk factors, and other possible contributing factors for the distress, such as: the amount of time since the diagnosis, additional children in the family, the income and marital status of parents, and the sibling's age, sex, race, and birth order in relation to the child with cancer. Their results demonstrated that low income and unmarried status of the parents as well as the sibling's race (specifically non-white siblings) significantly increased the amount of distress in the sibling after diagnosis. Time since diagnosis, additional siblings, age, sex, and birth order had no significant effects on the sibling's distress level. In regards to the family risk factors that were examined in this study, the authors found that greater sibling distress was highly correlated with high parental psychological control, lower parental acceptance, and family functioning issues. However, the effects on sibling distress were strongest with family functioning problems than they were with the other two risk factors. The same trend was apparent with the PTSS experienced by parents as well. These results underscore the importance of helping parents to better monitor their own behavior and attitudes toward the illness when around their children.



The role of empathy in the context of siblings of cancer survivors has also been investigated. Labay and Walco (2004) examined the associations between the sibling's empathy, understanding of the illness, relationship with the patient, and psychological adjustment at least 3 months after diagnosis. The siblings completed measures of empathy, sibling relationship, and illness concepts while the parents reported the siblings' adjustment by filling out the Child Behavior Checklist. The results of this study found that the healthy siblings were less involved and less successful in school and social activities; however, the authors also found that the siblings with greater levels of empathy had fewer difficulties. The results of this study suggest that a sibling's empathy is a significant predictor of the child's psychological adjustment problems in that having more empathy appears to allow the sibling to be better able to accept the situation. The authors also found that while educating the child on the illness does not directly affect the sibling's adjustment, cancer knowledge does increase the sibling's empathy. They also found that the siblings in the study did tend to have more behavioral issues when their relationship with the pediatric cancer patient was considered to be more intimate due to a heightened sense of vulnerability and distress. Although this study only had 29 participants, these results help identify possible predictors of sibling distress. Such knowledge is helpful to parents so that they can be aware of and prepared to provide the needs of all of their children through facilitating empathy.

In summary, additional research needs to be conducted to uncover the underlying factors that lead some healthy siblings to struggle with adjustment. Another area of interest is examining adjustment between children whose sibling has survived cancer and those whose sibling is deceased due to the cancer. Each of the two types of siblings of pediatric cancer patients have their own adjustment issues, needs, influences, and outcomes.

For the siblings of cancer patients who have survived their illness, studies have examined their post-illness adjustment and have found that the siblings can still experience various psychosocial challenges. Alderfer, Labay, and Kazak (2003) investigated the likelihood of the siblings of pediatric cancer survivors experiencing posttraumatic stress symptoms (PTS). Participants in this study were adolescents who completed self-report measures of anxiety, possible PTS, and perceptions of the entire cancer experience. The results demonstrated that siblings of pediatric cancer survivors were significantly more prone to PTS symptoms than their peers. Nearly half of the participants in the sample reported having mild symptoms, while an additional 32% reported to have moderate to severe PTS symptoms. The authors of this article suggested that PTS may be a useful model when working with siblings of pediatric cancer, even after the patient has finished treatment. Using PTS as a model for the reactions of siblings of pediatric cancer survivors may help the psychology community to better understand the adjustment and the psychosocial needs of these children.

Additional psychosocial needs of sibling survivors were evaluated using a family systems approach by Buchbinder, Casillas, and Zeltzer (2011) in their review of studies examining survivorship of siblings of pediatric cancer and their long-term psychosocial health. They reviewed 19 studies that included 24 different measures assessing the impaired psychosocial health of the siblings of pediatric cancer survivors. The first study they reviewed was the article by Alderfer, Labay, and Kazak (2003) mentioned in the previous paragraph. Collectively, the remaining articles found that siblings of pediatric cancer experienced a sense of awareness, alarm, vigilance, or relaxed vigilance (Brett & Davies, 1988), scored more outside the normal range in externalizing and internalizing behaviors than siblings of Wilms's tumor survivors (Buzier, deSonneville, van den Heuvel-Eibrink, & Veerman, 2006), had greater odds of

psychological distress, health issues, and functional impairment than their siblings (Hudson et al., 2003), and were more likely to partake in unhealthy behaviors (Larcombe, Mott, & Hunt, 2002; Lown et al., 2008; Tao, Zeltzer, Byrne, Mills, & Robison, 1998). They also found that cancer continued to have a positive or negative effect on a family 12 years after diagnosis (Koocher & O'Malley, 1981). One case study of a younger sister of a pediatric cancer patient demonstrated that themes of closeness, activity, fearfulness, anger, worry, and spirituality continued to persist 14 years after the diagnosis (Lehna, 1998). The articles also found that siblings of pediatric cancer may be more likely to develop depression and sleep problems than survivors (Mulrooney et al., 2008), especially apparent in siblings of survivors with disfigurement (Schultz et al. 2008), and female siblings may be more likely to develop lifetime major depression than male siblings (Teta, Del Po, Kasl, Meigs, Myers, & Mulvihill, 1986). Although survivors tended to have more tension, anger, depression, and confusion, their siblings were similar in scores for vigor and fatigue (Zeltzer et al., 1997) and were more likely to have similar or better health-related QoL than norms, but also tended to have lower scores in vitality and emotional role functioning (Zeltzer et al., 2008). However, other studies indicated that siblings of pediatric cancer demonstrated less physiological distress than norms, and that risk factors for depression were similar to those for the general population (Zebrack et al., 2002; Zebrack et al., 2004; Zebrack et al., 2007). Overall, the literature review concluded that family members clearly play a vital role in the adaptation of not only the cancer survivors but also of the sibling survivors. Specifically, the authors indicated that sibling survivors tend to experience psychosocial health changes in the form of anxiety, vivid memories of the traumatic experience, and strong emotions. The authors explained that new worries and concerns in the sibling that

come with survivorship of the patient may lead to greater anxiety, depression, health risk behaviors, and impaired energy in the sibling.

While the previously mentioned studies have shown that young, adolescent sibling survivors can have severe adjustment issues post-illness, a cross-sectional study conducted by Buchbinder et al. (2011) attempted to identify risk factors found in adult siblings of childhood cancer survivors. The siblings in this study participated after at least five years of surviving the cancer had passed. They completed the Brief Symptom Inventory-18, and were matched with their sibling who survived the illness. Sociodemographic and health data were examined as possible risk factors for adverse psychological effects on the adult siblings of long-term childhood cancer survivors. In general, the results of this study found that after enough time passes, siblings of long-term childhood cancer survivors are typically psychologically healthy. However, there appears to be a subgroup of these siblings who have considerably higher risk factors for psychological distress. The high-risk subgroups include: siblings who were younger than the cancer survivor (which may have led to a lack of parental attention during the cancer treatment), and siblings whose previously ill sibling has self-reported high medical and/or psychological distress. In addition, siblings of sarcoma survivors were at higher risk due to the long-term effects of the illness in comparison to those of leukemia survivors, and an unexplainable higher level of depression found in siblings of male cancer survivors than of female survivors. While it is positive to find that adult siblings of childhood cancer generally have healthy psychosocial development, it is also important to identify those subgroups of adult sibling survivors that experience risk factors for distress later in life. Understanding which groups are at a higher risk will allow the psychology community to provide the necessary tools to help such individuals become less distressed and enjoy a higher quality of life.

While there are a variety of challenges that siblings of pediatric cancer survivors face, there are additional aspects to consider for the siblings grieving a loss due to pediatric cancer. These siblings not only endure a time of distress and hope during treatment, they also experienced a highly significant loss accompanied by a multitude of psychological struggles. Barrera, Alam, D'Agostino, Nicholas, and Schneiderman (2013) conducted a longitudinal study evaluating the parental perceptions of the bereavement of children after the loss of a sibling due to pediatric cancer 6 and 18 months post-death. The areas that they investigated in this study were grief reactions post-death, what actions helped the child grieve in a more healthy fashion, new relationships with parents, and the nature of their bond with the deceased child after death. The siblings in this study varied in their reactions to the death based on their age. The younger siblings (less than 6 years old) tended to express intense, mixed emotions (such as sadness and anger), have behavioral problems, and struggled with understanding the meaning of death. The adolescent siblings, on the other hand, were more inclined to suffer academically, fear death, avoid expressing emotion around parents, and engage in risky behaviors such as consuming alcohol. The researchers also found that getting back to participating in one's normal daily activities, openly communicating within the family about the deceased child, obtaining social support from friends (for adolescent siblings), and group support (for young siblings) were beneficial in helping the sibling cope with the death of the pediatric cancer patient. They also found that the surviving siblings tended to still possess a bond with their deceased sibling, which was displayed by their actions of either using the belongings of the deceased sibling, studying in a field related to their sibling's illness, having considerable numbers of dreams about the deceased sibling, and (for younger siblings) pretend play with the sibling who is no longer there. The types of grief mentioned in this study are a cause for concern for bereaved siblings of

pediatric cancer if reactions are prolonged, and call for further investigation to provide them with strategies to cope properly.

Sveen, Eilegård, Steineck, and Kreicbergs (2014) studied the prevalence of prolonged grief among bereaved young adult siblings of pediatric cancer after 2 to 9 years had passed since the death and they examined possible contributing factors as to why their grief has been unresolved. Participants filled out a study-specific questionnaire that measured grief, descriptive and sociodemographic characteristics, anxiety and depression, and amount of social support. Slightly more than half of the participants (54%) answered that they had either worked through their grief “not at all” or “to some extent”, showing that a majority of the participants were still suffering with significant grief. The study also revealed that lack of social support and a shorter amount of time since the death were both influential factors predicting the prolonged grief that bereaved siblings of pediatric cancer may experience. The prevalence of prolonged grief among these individuals indicates a greater need for social support for these individuals and for further research to maximize the help that can be provided to those who have lost a sibling due to cancer.

Additional research indicates that bereaved siblings of pediatric cancer suffer from a number of adjustment difficulties.

The psychological health of bereaved siblings of pediatric cancer was investigated by Eilegård, Steineck, Nyberg, and Kreicbergs (2013), who specifically evaluated levels of self-esteem, difficulty with sleep, personal maturity, depression, and anxiety in bereaved siblings 2 to 9 years post-death and a control group of non-bereaved siblings. The researchers used the Hospital Anxiety and Depression Scale to measure each of the mentioned aspects of psychological health in the 393 participants. The results revealed that bereaved siblings reported

to have significantly lower self-esteem, more difficulties falling asleep, and lower levels of personal maturity. The researchers suggest these problems occurred due to the belief that these children “almost become invisible” during the illness and during their parents’ personal post-death grief. Surprisingly, there was not a significant difference between the two groups in their levels of depression or anxiety; however, there was a tendency for the bereaved brothers to have higher anxiety levels.

In regards to changes that a bereaved sibling may face after a pediatric cancer death, Foster et al. (2012) examined the frequency of changes in siblings life post-death based on semi-structured interviews with 36 mothers, 24 fathers, and 39 siblings. The changes that were investigated among the siblings were personal changes (personalities, academics, goals in life, activities/interests) and their relationships with peers and family members. The results found that 69% of the siblings experienced personal changes, 47% had changes in their relationships with family members and peers, and only 21% claimed to have experienced no change attributed to the death of the pediatric cancer patient. Interestingly, these changes could be positive or negative for the sibling. The changes in personality included greater maturity (15%), becoming more withdrawn (15%), becoming more compassionate (14%), increased sadness (11%), increases in anger (11%), and more fearful of death (3%). Twenty-three percent of the participants reported positive or negative changes in academics (there was no distinction between the two types of change in the article). Siblings also reported changes in their life priorities (15%) and were motivated and inspired by their deceased sibling (9%). Seven percent of the participants stated that they either were more open to new activities and interests or they became uninterested in activities including those that they enjoyed prior to the death. As for the relationships of the siblings, 21% experienced changes in peer relationships, 20% reported closer

family relationships, 18% experienced a role change in the family (oldest, youngest, or only child), and 5% reported more distant family relationships. While this study would have benefitted from more accurately separating percentages of the positive and negative personal and relational changes in bereaved siblings, the researchers express that such changes usually occur for these individuals and that they could go either way depending on their relationships and temperaments. Knowing that these changes occur can prepare families for the possibilities after the death of a child due to cancer.

Another study investigating changes in bereaved siblings of pediatric cancer was conducted by Martinson and Campos (1991). Focusing on a much later time period than most studies, they collected their data 7 to 9 years after death by interviewing 31 siblings who were between the ages of 10 and 19 years at the time of death. Participants in this study also experienced the death at home rather than in the hospital, which allowed the siblings' to have more contact with the dying sibling. Using a semi-structured questionnaire designed specifically for the study, the researchers examined changes in family life, emotional support, coping, family communication, memories of the deceased sibling, and the experience at the time of death. The study had three categories to identify the siblings' experiences with death: positive legacy (15 siblings), mixed legacy (11), and negative legacy (5). The type of legacy signifies that they had a positive, negative, or mixed outlook on the death and the experiences that came with it. Those with a positive outlook expressed having pleasure in the time they had with their sibling, were able to share the experience with others through talking about their thoughts and feelings, and frequently recalled memories of the deceased sibling. Those with a negative outlook reported having a hard time connecting with the sibling during their illness, were withdrawn from their sources of support, and avoided recalling memories. Those with a mixed outlook identified their



family as a social support, but they were unable to connect any positive outcomes with the death of the sibling. The study revealed that 16% of the participants (after 7 to 9 years had passed) felt that the experiences from the death were continuing to have negative impacts on their lives. While many of the participants felt that the in-home care of the sibling was beneficial towards their relationships with the ill sibling, the study suggests that such treatment is not significantly better than treatment in the hospital. Although the parents are not physically separated from the family to care for the ill child, they may still be unable to attend to the needs of the healthy child due to the demands of caregiving for the ill child, parental anxiety over the predicted death, or ignorance of the amount of needs of the healthy sibling that are being ignored. In conclusion, the researchers suggest that parents become aware that adolescents can be uncomfortable with parental expressions of grief and to attempt to reestablish communication with the well-sibling before the death if possible. They also suggest that health care providers attempt to communicate with the adolescent siblings in addition to the parents, noting that when a physician took the time to address the specific needs of the child instead of the needs of the family as a whole there was a lasting impression that aided in the coping of the healthy sibling. While in-home care for pediatric cancer may appear to be a beneficial option for the entire family, parents need to be aware that it may not address every issue potentially experienced by the sibling. Parents need to be aware that their well-children still need a relationship with them as well.

Relationships can be greatly influenced by the death of a pediatric cancer patient. The relationships found in the previously mentioned study by Barrera et al.'s (2013) between the sibling and the parents were greatly influenced by the death of a child due to cancer, however the relationships branched off into different directions. The types of relationships that emerged after the death were positive relationship with parent, wanting more attention from parent, parent-

blaming, and concern for mother's (primary care-taker) well-being. Some relationships between surviving siblings and parents were improved because the death opened communication between the two to express thoughts and feelings. Some of the siblings (typically those younger) expressed that though they were sad, they were also happy because they were getting the attention that their ill sibling had acquired from the parents. Younger siblings also tended to have a difficult relationship with the parent and would blame them for what happened due to their lack of understanding and difficulties relating to one another. Adolescents, on the other hand, tended to fulfill a caretaker role and show concern and compassion toward their mothers who had been the primary care-taker ever since the diagnosis. These changed relationships between sibling and parent show how significantly the death of the pediatric patient can affect the lives of these individuals.

The family environment and bereaved siblings of pediatric cancer were investigated by Davies (1988), who used a semi-structured interview, the Family Environment Scale, and the Child Behavior Checklist to compare 34 families who had lost a child to cancer compared to normal families and distressed families. The normal families group included families from different geographical locations, from single-parent to multigenerational families, families from ethnic minority groups, and families whose children varied in developmental stages. The distressed families included those of alcohol abuse, general psychiatric patients, or children in crises situations such as children who had run away from home, were identified as delinquents, or were being placed into a foster home. The results of the study indicated that bereaved families with a larger number of surviving children were more likely to participate in social and recreational activities as well as have a more structured living environment with more boundaries and rules to follow. However, they also found more conflict in these homes. The scores from the

Family Environment Scale indicated that bereaved families were considered to be closer to normal families and had lower conflict scores in comparison to the distressed families. These results are interesting because although the bereaved families lost a child, they do not contend with the constant stresses that may accumulate in distressed families. Thus, they are better able to move on and to grow from the experience if they have a supportive family environment. The study also revealed that the bereaved families who were more cohesive, active and religious had surviving children who were less likely to have behavioral problems up to 3 years after the death due to pediatric cancer.

Understanding that there are differences in the reactions of siblings of pediatric cancer survivors and of bereaved siblings allows the field of pediatric psychology to pinpoint specific areas of intervention to help each type of sibling. In the following section, studies that focus on possible psychosocial strategies to aid these individuals will be discussed.

Strong social support has been suggested to benefit siblings of pediatric cancer. Barrera, Fleming, and Khan (2004) examined the role of emotional social support in the psychological adjustment of siblings of pediatric cancer. This included two types of groups of siblings of children who were being treated for cancer at the time: one group consisting of siblings who had been referred for behavioral problems and a control group of siblings of pediatric cancer who had not been referred. Each group of siblings completed measures of depression and anxiety, behavior problems, and amount of emotional social support. One parent per sibling also completed measures of the child's anxiety and behavior. The results of this study indicated that the children who reported having more social support had significantly fewer symptoms of depression and anxiety as well as fewer behavioral problems than those who felt they had less social support. This study did not distinguish which type of social support the siblings were

experiencing, but it does reveal that social support of any kind appears helpful in protecting these individuals from psychological distress without significant differences between the siblings' age or gender.

Bendor (1990) also investigated the impact of social support on siblings of pediatric cancer in the context of a sibling support group. The siblings were split into two multisession groups in this study: one for 5 siblings between the ages of 8 and 13 years and another for 4 siblings between the ages of 14 and 19 years. The purpose of the support groups was to allow the siblings to identify, express, and master their feelings of anxiety and isolation in an atmosphere where such feelings are understood. The findings of this study suggest that significant contributing factors for the feelings of anxiety and isolation in the younger group were the siblings' perception of parental focus on the ill child and lack of attention to needs, anger, loneliness, and vulnerability. The contributing factors for such feelings in the adolescent group were a fear of death, vulnerability, burden with additional responsibilities, inner conflict to pursue their own needs versus being there for their parents and ill sibling, a struggle with caring for the ill-sibling, and feeling a need to be extra sensitive on the subject of the ill-sibling. The author of the study offered recommendations for preventing the common psychosocial impairments of siblings of pediatric cancer. The suggestions include: 1) redirecting parental attention to the well-siblings rather than focusing solely on the ill-child, 2) advising parents on how to meet the emotional needs of the well-siblings, 3) having the health care team develop supportive relationships with the siblings in addition to the parents as opposed to the family as a whole, and 4) helping the parents discuss death with the well-siblings (especially younger siblings who struggle to grasp such concepts). They also suggested conducting vigorous post-

death follow-up sessions for well-siblings and educating the community on the social support they can provide with non-traditional support, such as scholarships and transportation.

Another study focused on the impact that family-oriented strategies have on the coping and distress levels of siblings of chronically ill children. Besier, Hölling, Schlack, West, and Goldbeck (2010) evaluated the impact of a 4-week family-oriented rehabilitation program for families of children with cancer, cystic fibrosis and congenital heart disease on siblings' level of behavioral and emotional problems as well as the association between such problems and the siblings' quality of life. The family-oriented rehabilitation program consisted of physical (i.e., massages, medication, physiotherapy) and psychosocial (i.e., psycho-educative groups, psychological counseling, art therapy, relaxation techniques) treatments that varied based on the family's needs. The study consisted of 259 well-sibling participants between the ages of 4 and 16 years who completed self-report measures of their quality of life, while their parents completed the Strengths and Difficulties Questionnaire. Both measures were administered at the time of admission and discharge from the clinic as well as 6 months after discharge. Participants from the study were matched with a control group from the general population based on age, gender and educational backgrounds of the parents. The results indicated that the family-oriented treatment approach was significantly effective in reducing behavioral and emotional problems among the well-siblings, and it had a lasting positive effect on the mental well-being of the siblings 6 months after being released from the program. The areas of improvement for these siblings included fewer behavioral problems, fewer emotional problems, less hyperactivity/inattention, and increased pro-social behaviors which overall reduced the levels of total challenges. While the siblings in this study varied in the patients' chronic illnesses, siblings of pediatric cancer were included, and the significance of the results demonstrate that the family-

oriented strategies were beneficial for each type of sibling including those of pediatric cancer. Further research should be conducted on the effects of a family-oriented rehabilitation program on the siblings of specifically from families with a pediatric cancer patient to strengthen support for this treatment.

Research has also briefly investigated psychological interventions with siblings of pediatric cancer. Prchal, Graf, Bergstraesser, and Landolt (2012) evaluated the effectiveness of a psychological intervention on the adjustment of siblings of pediatric cancer. The intervention took place within the first 2 months after the cancer diagnosis. Participants included 30 siblings between the ages of 6 and 17 years who were randomly assigned into either the intervention group or the control group who received standard psychosocial care. The standard psychosocial care provided to the control group consisted of meetings with the psycho-oncologist on the ward who was primarily responsible for the ill-child and for informing the parents of updates in the treatment; however, they also met with the siblings if it was considered necessary. The siblings in the intervention group individually participated in two sessions, each 50 minutes long and separated by two weeks, in which a clinical psychologist guided the siblings and their parents through medical information, strategies for coping with stressful situations, and additional information for parents. The medical information that was provided in the intervention for siblings included understanding body functions, the location of the cancer and the mechanisms of that location and of the cancer, and the cancer treatment the patient was undergoing. The medical information was provided to increase the siblings' feelings of control over the situation in the hopes that it might reduce feelings of anxiety as well as increase social competence. In order to provide coping strategies for stressful situations, the siblings were encouraged to identify the three main stressors that may arise which were then discussed in detail and relevant coping

strategies were practiced. When difficulties with identifying and/or discussing the three significant stressors occurred, cognitive behavioral therapy was conducted. The parents were also informed of the siblings responses so that they could provide maximum parental support in the areas mentioned. Each parent also received a psychoeducational booklet that provided further information over the psychosocial adjustment challenges that are common in siblings of pediatric cancer as well as recommendations to effectively support these children during such a difficult time. The siblings and their parents in both groups completed assessments at 4 to 6 weeks (prior to the interventions), 4 months, and 7 months after the diagnosis.

Outcomes measured in this study included medical knowledge, social support, health-related QoL, posttraumatic stress symptoms, anxiety, socio-economic status, and medical variables including intensity of treatment, medical complications, and health-related restrictions. Although the results of this study did not reveal a significant impact on health-related QoL, posttraumatic stress symptoms, or anxiety, they did suggest that a two-session psychological intervention program positively impacted the psychological well-being, social support, and medical knowledge of siblings of pediatric cancer. The researchers in this study imply that obtaining age appropriate medical knowledge is highly beneficial for siblings of pediatric cancer patients as a better understanding of the illness and the treatment process provides a stronger sense of control by preventing their imaginations from distorting the reality of the condition that would otherwise be misunderstood.

Adams-Greenly, Shiminski-Maher, McGowan, and Meyers (1986) sought to understand the positive effects of acquiring medical knowledge in siblings of pediatric cancer through a workshop called Specially Important Brothers and Sisters (SIBS). The SIBS workshop was offered once every 6 months (typically during school holidays) to siblings of pediatric cancer

between the ages of 6 and 17 years (divided into 2 age appropriate groups) whose ill-sibling is receiving or has received treatment within a year prior to the program. The intention of the workshop was to provide medical education, encourage expression of difficult thoughts and feelings, and emphasize successful coping strategies through three segments. The first segment is an education segment in which a physician and a nurse lead a discussion with the siblings about cancer and its treatment in a classroom-type setting, followed by a hands-on tour of the radiation oncology, the operating and recovery rooms, the Pediatric Day Hospital in which discussion and questions are still highly encouraged. After lunch in the hospital cafeteria, the second segment of the program (the support segment) began. This segment begins with a viewing of a video called "Coping with Cancer: The Sibling Perspective" in which four siblings at varying ages share how they have coped with being a sibling of a pediatric cancer patient. This video is intended to provide relatable stories, stimulate discussion of troubling feelings that are common, and demonstrate that successful coping is possible for siblings of pediatric cancer. Themes that are common in the discussions following the video include: changes in family relationships, jealousy, resentment, anger, responsibility, guilt, school problems, fear, worry, sadness, love, and concern. The siblings also discuss their own coping strategies to help fellow siblings who are struggling. The discussion following the video is led by a social worker who emphasizes the importance of family support to the siblings and advises them to talk about their feelings with their parents. The third and final segment is the recreation segment, in which the two age groups are reunited for non-competitive games that foster a sense of cohesion. The siblings are also encouraged to collect each other's contact information. During this segment, a social worker and nurse meet with the parents to show them a video and to discuss the common themes of the support segment while encouraging questions from the parents. A month after the workshop,



parents received a questionnaire in the mail assessing the effects of the workshop on their well-child's adjustment to the cancer diagnosis, achieving an 81% response rate. The results of this questionnaire were extremely positive: 96% reported the workshop had a "very positive" or "positive" effect, 90% reported increased cancer and treatment knowledge, 86% reported the sibling learned of others having the same experience, 85% reported increased sense of importance, 70% reported the sibling as more comfortable in the hospital and with the staff, 65% reported the sibling was more comfortable talking about the cancer, 60% reported better ways of coping, 60% were more verbal about their feelings, and 58% had become less resentful or angry. In conclusion, the SIBS workshop appears to provide a beneficial experience for siblings of pediatric cancer.

Although the research concentrating on the psychosocial needs of siblings of pediatric cancer has had few studies conducted, research on the effects of summer camp experiences for helping siblings of pediatric cancer has been more extensive. Murray (2001) examined the effects of attending such a summer camp on the self-concept of siblings of pediatric cancer. Participants in this study included 50 siblings of pediatric cancer between the ages of 7 and 12 years; 44% attended a summer camp for siblings of pediatric cancer while the remaining 56% did not. The Personal Attribute Inventory for Children (PAIC) scale was used to measure each sibling's level of self-concept. The results of this study revealed that the siblings who had attended summer camp also had significantly higher scores on the PAIC self-concept scale. Packman et al. (2005) also examined the positive effects of summer camp on siblings of pediatric cancer. In their study, they assessed the health-related QoL of 77 siblings between the ages of 6 and 17 years who attended a one-week session at a SIBS summer camp program at Camp Okizu. Siblings and parents completed quantitative and qualitative assessments of health-related QoL

prior to and after attending the camp. The quantitative assessment used in this study was the Pediatric Quality of Life Inventory (PedsQL); the qualitative assessments used were the Sibling Qualitative Interview and the Camp Okizu Satisfaction Surveys. The results revealed that the siblings reported significantly improved health-related QoL; however, the reports of the parents did not match in significance until their reports were controlled for the responses of bereaved parents who reported the impact as considerably less positive than the nonbereaved parents. Conversely, the qualitative measures revealed a significant improvement in the health-related QoL of siblings of pediatric cancer patients.

Another study by Packman et al. (2008) at the same SIBS camp evaluated the differences in effectiveness between the bereaved and nonbereaved siblings of pediatric cancer by administering projective measures of emotional distress and family environment prior to attending camp and three months after. The projective measures used in this study were the Human Figure Drawing (HFD) and the Kinetic Family Drawing-Revised (KFD-R). Participants in this study included 77 siblings between the ages of 6 and 17 years, 18 of which were bereaved siblings. The parents of the siblings also completed the KFD-R. Both bereaved and nonbereaved siblings' emotional distress scores significantly decreased between the pre- and post-assessments. Nonbereaved siblings and parents also showed significant improvement in their family environments; bereaved siblings and parents also showed some improvement, but it was not statistically significant.

To address this issue of bereavement, another study was conducted at a camp exclusively for bereaved siblings of pediatric cancer. Creed, Ruffin, and Ward (2001) describe the positive impact of Camp New Horizons on bereaved siblings of pediatric cancer. Camp New Horizons is a weekend camp for siblings between the ages of 6 and 18 years who lost their ill-sibling in the

last 3 years. The siblings are split into age appropriate groups, and they learn about the grief process to reduce their feelings of isolation, how to express grief in a healthy manner, and how to move forward in the grief process. In this study, there were 19 participants between the ages of 6 and 15 years who were split into 2 age groups. The siblings and parents completed evaluations on the effectiveness of the camp. Although the evaluations did not provide data for statistical comparison, the qualitative feedback was positive from both campers and parents. The feedback from the evaluations indicated that the camp validated the feelings of the campers, improved open communication, and provided lasting friendships with peers going through the same experience. The camp atmosphere, paired with a supportive peer environment, appeared to provide a beneficial social support in helping the sibling cope with the diagnosis, and possibly the loss, of the pediatric cancer patient.

Summary. The studies that have been discussed in this review suggest that siblings of pediatric cancer present with numerous psychosocial needs. While many of the articles proposed promising intervention strategies, there is still very little research in this domain to indicate that they are empirically supported. The result of this review would also argue that while cancer patients receive considerable attention in pediatric psychology literature, their siblings do not. Siblings of pediatric cancer encounter significant distress and negative emotions, and their social life, family life, and school work can be negatively impacted without proper intervention from the pediatric health community. Pediatric psychology is on a promising path to helping these children adjust to and cope with the stress of pediatric cancer. Future research should focus on the effects of intervention strategies on bereaved and nonbereaved siblings separately and they

should also continue to develop valid instruments to use specifically for evaluating siblings of pediatric cancer.

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## Appendix A

Table I Overview of the Sibling Survivor Research Findings

from the Literature Review by Buchbinder, Casillas, and Zeltzer (2011)

Author	Objectives	Methods	Sample Characteristics	Results
Alderfer et al. (2003)	To study the behavioral and psychosocial health with respect to posttraumatic stress symptoms (PTSS) in siblings of childhood cancer survivors	Quantitative Cross-sectional Posttraumatic Stress Disorder Reaction Index, Revised Children's Manifest Anxiety Scale, Impact of Events Scale-Revised, Assessment of Life Threat and Treatment Intensity Questionnaire	78 siblings with and average age of 14 years and an average period postdiagnosis of 5 years	Mild posttraumatic stress reactions observed in half of the sample. One third of the sample scored positive for moderate to severe PTSS. There were more PTSS in siblings when compared with norms
Brett and Davies (1988)		Qualitative Semistructured Interviews	4 survivors, 7 parents, and 10 siblings. Survivors were a minimum of 5 years postdiagnosis	Sibling appraisals of the cancer experience were divided into 4 stages, including awareness, alarm, vigilance, and relaxed vigilance. In relaxed vigilance, siblings were certain about longterm survival; however, there were new doubts/questions. Siblings with more time in vigilance had apprehensions that decreased with time
Buzier et al. (2006)	To study the behavioral functioning of survivors of childhood acute lymphoblastic leukemia and Wilms's tumor	Quantitative Cross-sectional Childhood behavior checklist (CBCL), Conner's Rating Scales (CRS)	199 survivors of acute lymphoblastic leukemia or Wilms's tumor and 37 siblings with a median age of 10.5 years. Survivors were an average period of at least 5 years postdiagnosis. 98 healthy school children served as controls	On the CBCL, siblings had more scores outside normal range compared with Wilms's tumor survivors with a focus on internalizing problems although these were not statistically significant. CRS findings were similar although focused on

				externalizing problems and also not statistically significant
Hudson et al. (2003)	To study the health status in survivors of childhood cancer compared with their siblings	Quantitative Cross-sectional Brief Symptom Inventory 18 to assess mental health Questions assessing general health, functional status, and limitations of activity were adapted from the National Health Interview Survey and the Brief Risk Factor Surveillance System	9,535 survivors of an average of 17.4 years postdiagnosis and 2,916 of their randomly selected nearest age living siblings were studied. The mean age of the siblings was 29.2 years	Greater odds of psychological distress noted in survivors when compared with siblings for all diagnostic categories apart from survivors of Wilms's tumor and neuroblastoma. Greater odds of health and functional impairment were noted in survivors when compared with siblings across domains
Koocher and O'Malley (1981)	To assess the psychological consequences among childhood cancer survivors and their families	Qualitative Semistructured interviews	117 survivors and their families, including 101 siblings (mean age 17.8 years) from 55 families ages 8 to 28 born prior to diagnosis with an average time of 12.49 years postdiagnosis	Cancer continues to have impact. Emotional concerns were common (e.g., feeling left out) and positive experiences were possible (e.g., closeness of family). Anger can be resolved and normal relationships can be restored
Larcombe et al. (2002)	To describe the health behaviors of survivors of childhood cancer compared with sex- and age matched controls	Quantitative Cross-sectional Health behaviors were measured using a questionnaire focusing on alcohol use, tobacco use, drug use, diet, and skin protection behaviors	178 survivors were diagnosed at least 5 years prior to study entry, 184 peers, and 67 siblings with a mean age of 26 years were compared	Compared with siblings, survivors and peers were less likely to be currently drinking or binge drinking, using tobacco, or using drugs, although none of these differences were statistically significant. Siblings and controls had greater unhealthy behaviors when compared with survivors
Lehna (1998)	To gain a better understanding of the recall and long-term effects of a younger sister's diagnosis of cancer	Qualitative Semistructured interviews	1 sibling of a leukemia survivor who is currently 22 years of age and was 8 years of age at the time of the diagnosis	Themes of closeness, activity, fearfulness, anger, worry, and spirituality persisted
Lown et al. (2008)	To describe the	Quantitative	3,034 siblings of 5	Compared with

	alcohol consumption among adult survivors of childhood cancer compared with sibling controls and peers	Cross-sectional Measures of alcohol consumption, demographics, cancer diagnosis, treatment, and psychosocial factors. Demographic data, health data, and diagnosis/treatment data are included	year (or greater) survivors of childhood cancer. 4,774 peers from National Alcohol Survey	siblings, survivors and peers were less likely to be current, risky, or heavy drinkers
Mulrooney et al. (2008)	To examine the prevalence of and risk factors for fatigue and sleep problems in longterm survivors of childhood cancer	Quantitative Cross-sectional Functional Assessment of Chronic Illness Therapy-Fatigue, Pittsburgh Sleep Quality Index, Epworth Sleepiness Scale. Demographic data, health data, and diagnosis/treatment data are included	369 siblings of 5 year (or greater) survivors of childhood cancer	Depression was higher in siblings compared with survivors, 15.4 % and 8.1% respectively. Depressed siblings were 9.5 times more likely to be fatigued and 8.9 times more likely to have sleep problems. Fatigued siblings had lower mean scores than survivors with respect to vitality and mental health functioning subscales of the Short Form 36
Sawyer et al. (1986)	To assess the prevalence of emotional and behavioral problems in children treated for leukemia	Quantitative Child Behavior Checklist, Rutter B2 Behavioral Scale for Teachers, Family Concept Inventory	42 survivors, 42 control, 56 siblings of survivors, and 54 siblings of controls Mean age of participants was between 10 and 11 years. Survivors were a mean of 5.67 years postdiagnosis	Survivors demonstrated greater behavioral, externalizing, and internalizing problems, and less social competence. There were no significant differences between siblings of survivors and siblings of controls
Schultz et al. (2007)	To evaluate the behavioral and social outcomes in adolescent survivors of childhood cancer	Quantitative Cross-sectional Parent-reported scores on the Behavioral Problem Index Demographic data, health data, and diagnosis/treatment data included	649 siblings with a mean age of 14.9 years (range 12- 17 years) and all siblings of 5 year (or greater) survivors of childhood cancer	Sibling of survivors with limb disfigurement had 3 times the risk of developing depression and anxiety. These siblings also had 2 times the risk of developing antisocial behavior

Tao et al. (1998)	To describe the tobacco consumption among adult survivors of childhood acute lymphoblastic leukemia compared with sibling controls	Quantitative Cross-sectional Measures of smoking behaviors and tobacco consumption, demographics, cancer diagnosis, treatment, and psychosocial factors	592 survivors of acute lymphoblastic leukemia (95% of sample surviving at least 5 years) and 409 siblings with a mean age of 25.2 years (matched on full sibling, gender, and nearest age)	Compared with siblings, survivors were less likely to be current smokers, to have ever smoked, and to ever become regular daily smokers. Smoking patterns of siblings were similar to cohort specific data from the National Health Interview Survey
Teta et al. (1986)	To investigate longterm psychosocial impact of childhood cancer on survivors and their siblings	Qualitative Semistructured interviews Schedule of affective disorders psychological rating scale and research diagnostic criteria	587 siblings of childhood cancer survivors from Connecticut Tumor Registry surviving 5 or more years postdiagnosis	Prevalence of lifetime major depression in siblings was 12% for males and 24% for females, which was similar to survivors and norms. A greater percentage of siblings cited family health as a reason for depression
Van Dongen-Melman et al. (1995)	To study the late consequences of the cancer experience for siblings	Quantitative Cross-sectional Child Behavioral Checklist, Self Perception Profile for Children, Amsterdam Biographic Questionnaire for Children, Children's Depression Scale	60 siblings of ages 5 to 16 years (median 10 years) of survivors of non-Hodgkin lymphoma, leukemia, rhabdomyosarcoma. Sibling age at diagnosis 0 to 11 years (median 4 years)	Only analysis focusing on survivors greater than 5 years postdiagnosis: higher problem scores on parent reported somatic complaints
Zebrack et al. (2002), Zebrack et al. (2004), and Zebrack et al. (2007)	To study the psychological outcome of longterm survivors of childhood cancer (1) 2002—leukemia, Hodgkin's Disease, and non-Hodgkin's lymphoma (2) 2004—brain tumor (3) 2007—solid tumors and compare them with a sibling control group	Quantitative Cross-sectional Questionnaire assessing symptoms associated with depression, somatization, and anxiety. Demographic data, health data, and diagnosis/treatment data were included	(1) 2002—Sibling control sample of 2,565, aged 18 to 56 years (median 28 years) (2) 2004—Sibling control sample of 2,817, aged 18 to 56 years (mean 29.4 years) (3) 2007—Sibling control sample of 2,925, aged 18 to 56 years (mean 29.5 years) All siblings of 5-year (or greater) survivors of childhood cancer	Overall siblings demonstrated less psychological distress than norms Risk factors for depression were similar to the general population (female gender, lack of high school education, low income, and lack of employment)

Zeltzer et al. (1997)	To examine the emotional or mood state of survivors of childhood acute lymphoblastic leukemia compared with their siblings	Quantitative Cross-sectional Profile of Mood States	580 survivors of acute lymphoblastic leukemia (95% of sample surviving at least 5 years) and 396 siblings with a mean age of 25.2 years (matched on full sibling, gender, and nearest age)	Profile of Mood States score was worse in survivors compared with siblings, including domains of tension, depression, anger, and confusion. Vigor and fatigue were similar between siblings and survivors
Zeltzer et al. (2008)	To assess the psychological outcomes, health related QOL, and life satisfaction of adult survivors of childhood cancer and compare them with a sibling control group	Quantitative Cross-sectional Brief Symptom Inventory-18, Short Form 36, and Cantril's Ladder of Life	388 siblings of 5 year (or greater) survivors of childhood cancer	Siblings reported similar or better quality of life than U.S. norms; however, the vitality and emotional role functioning of sibling survivors was lower than population norms