FREQUENCY AND PREDICTORS OF SIBLING PSYCHOLOGICAL AND SOMATIC DIFFICULTIES FOLLOWING PEDIATRIC CANCER DIAGNOSIS

by

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Abstract

Siblings of children with cancer encounter stressors and challenges that can lead to severe distress and a host of psychological difficulties. Factors including age, gender, and disease characteristics of the child with cancer are reported to influence sibling adjustment. The majority of research, however, is dated, inconsistent, and marred by methodological problems. Guided by the disability-stress-coping model, the study examined the: (a) frequency of sibling and parent reported symptoms of anxiety and depression, internalizing and externalizing behavior problems, and somatic problems, (b) influence of sibling, family, and disease factors on sibling adjustment, (c) moderating effects of age on the relationship between sibling factors and sibling adjustment, and (d) mediating effect of primary cognitive appraisal on the relationship between self-esteem and sibling adjustment.

One hundred and eight siblings (7-17 years; 51 males; 57 females) participated. Siblings completed the State-Trait Anxiety Inventory for Children and the Children's Depression Inventory to provide measures of sibling reported symptoms of anxiety and depression. Parents completed the Child Behavior Checklist to provide measures of parent reported internalizing behavior problems, externalizing behavior problems, and somatic problems. The communication and intrapersonal thoughts and feelings subscales of the Sibling Perception Questionnaire, completed by siblings, were used to assess perceived social support and primary cognitive appraisal. Self-esteem was assessed with the global self-worth subscale of the Self Perception Profile for Children/Adolescents, completed by siblings. Hierarchical regression analyses were

conducted to assess the direct and indirect effects of sibling, family, and disease factors on psychological outcomes.

Siblings and parents reported higher incidents of clinically significant symptoms of anxiety, internalizing behavior problems, and somatic problems than expected in a normative population. Sibling age and gender, diagnosis of the child with cancer, social support, self-esteem, and primary cognitive appraisal were significantly associated with sibling and parent reported psychological adjustment measures. Age moderated the relationship between gender, social support, and primary cognitive appraisal and several adjustment outcomes. Lastly, primary cognitive appraisal partially mediated the relationship between self-esteem and sibling reported anxiety and depression symptoms. These findings highlight the need for sibling psychosocial interventions and provide direction for the development and implementation of such groups.

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Table of Contents

List of Tables

List of Figures

List of Appendices

CHAPTER ONE

Introduction

Pediatric cancer is a life-threatening illness that often involves demanding medical treatment. Given numerous medical advances in the treatment of cancer, deaths in children due to cancer are declining and it is expected in 2009, approximately 87% of the 1300 children and adolescents diagnosed with cancer in Canada will survive the disease (Canadian Cancer Society, 2009). When children are diagnosed with and treated for cancer, they and their family members encounter major challenges and stressors related to the short- and long-term side-effects of treatment and possible disease relapse (Houtzager, Grootenhuis, & Last, 1999). Moreover, throughout treatment, families must manage repeated hospitalizations, hospital visits, financial difficulties, uncertainty about the child's prognosis, and fear of his/her possible death (Houtzager et al., 1999; McGrath, 2001; Patistea, Makrodimitri, & Panteli, 2000). These challenges and stressors can disrupt the daily lives of all family members and alter the balance within family systems (Houtzager et al., 1999).

Siblings[1] experience particularly difficult and demanding conditions and events that lead to considerable stress. Siblings face not only numerous disruptions to their daily lives including sudden and extended separations from the child with cancer and their parents, but they witness parental distress and/or physical and emotional pain in the child with cancer. They can also experience uncertainty of the future if they understand the threats of cancer and its treatment (Alderfer, Labay, & Kazak, 2003). Research has shown the disruptions and challenges encountered by siblings, in combination with the unpredictable course of the illness and feelings of uncertainty and helplessness, can lead to severe and chronic distress in siblings (Houtzager, Grootenhuis, Hoekstra-Weebers, & Last, 2005).

[1] Throughout this dissertation, *siblings* denote healthy children and adolescents whose brothers or sisters have cancer.

Given the considerable challenges and stressors siblings encounter, researchers have devoted increased attention to their adjustment. In the last 20 years, researchers have more systematically examined the psychological adjustment[2] of siblings following pediatric cancer diagnosis, treatment, and remission (Houtzager et al., 1999). Conventionally, psychological adjustment is a dynamic process including cognitive, affective, physical, and functional factors that denotes social adaptation and emotional well-being (Cimprich, 1999; Heim, Valach, & Schaffner, 1997). Although debated within the field, psychological adjustment can be assessed by examining the presence of numerous and diverse psychological and social difficulties including internalizing behavior problems (e.g., anxiety, depression), externalizing behavior problems (e.g., physical aggression, temper tantrums), social relation problems, and so on.

Both early and more recent research on the psychological adjustment of siblings suggests siblings can experience more distress than the ill child (Alderfer et al., 2003; Cairns, Clark, Smith, & Lansky, 1979; Spinetta, 1981), and Sahler, Roghmann, Carpenter, and Mulhern (1994) estimate up to 63% of siblings experience psychological difficulties at some point after diagnosis. Reviews and a meta-analysis of the psychological adjustment of siblings of children with cancer or chronic illnesses including cancer conclude siblings can demonstrate significant emotional and behavioral problems including depression and high levels of anxiety (Barlow & Ellard, 2006; Houtzager et al., 1999; Sharpe & Rossiter, 2002; Williams, 1997). Moreover, compared to controls, they generally have lower levels of psychological and cognitive adjustment (Barlow & Ellard, 2006; Houtzager et al., 1999; Sharpe & Rossiter, 2002) and can experience somatic problems such as sleeping and eating difficulties and head- and stomach-aches (Heffernan & Zanelli, 1997; Sahler et al., 1994; Williams, 1997; Zeltzer et al., 1996).

[2] The following terms will be used interchangeably throughout this dissertation to denote sibling psychological adjustment and the symptoms of anxiety and depression, internalizing and externalizing behavior problems, and/or somatic problems they encounter: psychological adjustment, psychological and somatic difficulties, sibling outcomes, and sibling adjustment.

Clearly, not all siblings suffer major psychological difficulties related to pediatric cancer. Rather, as delineated by conceptual models developed to describe the psychological adjustment of individuals to chronic illness, sibling psychological and somatic difficulties are variable and appear to be influenced by numerous factors including sibling age (Barrera, Fleming, & Khan, 2004c; Bendor, 1990; Hamama, Ronen, & Feigin, 2000; Houtzager, Grootenhuis, Caron, & Last, 2004; Sahler et al., 1994; Sargent, Sahler, Roghmann, & Mulhern, 1995; Spinetta, 1981) and gender (Alderfer et al., 2003; Barrera et al., 2004c; Hamama et al., 2000). Adjustment also appears to be influenced by disease factors[3] such as length of time since diagnosis (Cohen, Friedrich, Jaworski, Copeland, & Pendergrass, 1994; Hamama et al., 2000), familial factors including social support (Barrera et al., 2004c; Williams et al., 2002), and sibling cognitive appraisal (Sloper & While, 1996). However, empirical findings pertaining to the extent to which siblings suffer from psychological and somatic difficulties and the factors (i.e., sibling, family, and disease factors) associated with such difficulties are inconclusive and frequently dated. Moreover, although researchers have examined the direct impact various sibling, family, and disease factors have on adjustment, few have investigated the indirect relationships between these factors and sibling adjustment that are purported by conceptual models. It is important to determine which factors have a direct effect on adjustment and which factors moderate or mediate the impact of stress and the cancer experience.

Empirical support for sibling adjustment conceptual models is limited and such models are rarely employed to guide psychological adjustment research and the development and implementation of intervention groups. The key purpose of the current study, therefore, is to assess the present-day psychological adjustment and somatic difficulties of siblings and the

[3] Throughout this dissertation, *disease factors* refer to clinical disease factors of the child with cancer, such as diagnosis and length of time since diagnosis.

factors that both directly and indirectly (i.e., moderate or mediate) influence psychological and somatic problems. Guided by the disability-stress-coping model (Wallander & Varni, 1992), a multivariable model previously adapted to examine adjustment in parents (Barrera et al., 2004b) and children (Barrera, Atenafu, Andrews, & Saunders, 2007; Barrera et al., 2003) to pediatric cancer and treatment, the study examines the direct and/or indirect influence of specific sibling, family, and disease variables on sibling adjustment. Ultimately, the research attempts to establish the most salient psychological and somatic problems siblings experience, the factors associated with their difficulties, and the nature of such associations. As such, the research ascertains the possible mechanisms by which cancer affects siblings and tests the disability-stress-coping model for sibling adjustment. In turn, the research identifies those siblings who would benefit most from a sibling intervention program and those factors that must be targeted through intervention support.

Impact of Pediatric Cancer on Sibling Psychological Adjustment

As siblings face considerable stress and numerous disruptions to their daily lives, some researchers have investigated the impact of the cancer experience on their psychological adjustment. Specifically, investigators have examined the extent to which siblings experience internalizing and externalizing behavior problems and require intervention. Other researchers have attempted to identify the possible mechanisms by which the cancer experience leads to adjustment difficulties and ascertain those siblings most in need of intervention support.

Internalizing Behavior Problems

Adjustment research indicates stress and the cancer experience are related to a myriad of behavior difficulties in children including internalizing behavior problems. Interviews with parents and/or siblings were used more frequently in early research to investigate such difficulties. In general, parents reported during interviews that siblings experienced various

emotion problems including jealousy, rejection, and withdrawal (Carpenter & Sahler, 1991; Martinson, Gilliss, Collaizzo, Freeman, & Bossert, 1990). Parents also reported emotional lability (Carpenter & Sahler, 1991), loneliness, and ambivalence (Martinson et al., 1990). During interviews, siblings reported similar difficulties including anger, loneliness, rejection, withdrawal, and guilt (Chesler, Allswede, & Barbarin, 1992; Gogan & Slavin, 1981; Kramer, 1984; Martinson et al., 1984). Siblings also described feelings of sadness, anxiety (Kramer, 1984; Schuler et al., 1985), jealousy (Chesler et al., 1992; Gogan & Slavin, 1981; Martinson et al., 1990), isolation (Schuler et al., 1985), and fear of death (Walker, 1988). Moreover, siblings said they worried about their brother or sister's physical pain and/or emotional ability to cope (Chesler et al., 1992) and getting cancer themselves (Gogan & Slavin, 1981).

In more recent studies, siblings described feeling constantly worried, anxious about losing their brother or sister, and/or concerned about the side-effects of treatment and the cancer returning (Nolbris, Enskar & Hellstrom, 2007). Additionally, 74% of parents described emotional problems related to themes of jealousy/envy, worry/fear/anxiety, anger/resentment, and/or loneliness/sadness/depression (Williams et al., 2009).

Empirical findings from standardized and non-standardized questionnaires developed to assess, in part, internalizing difficulties are less consistent than interview results. Early findings from standardized parent questionnaires such as the Child Behavior Checklist (CBCL; Achenbach, 1991) suggest siblings and healthy controls do not differ on measures of internalizing behavior problems such as symptoms of anxiety and depression (Horwitz & Kazak, 1990; Sawyer, Crettenden, & Toogood, 1986; Van Dongen-Melman, De Groot, Hahlen, & Verhulst, 1995). Moreover, both parents and siblings reported low sibling anxiety state scores on the standardized State-Trait Anxiety Inventory questionnaire (STAIC; Spielberger, 1983) (Fife, Norton, & Groom, 1987). Fife and colleagues note, however, many of the siblings' questions and

behaviors were indicative of high anxiety levels and siblings and parents may have reported low anxiety as they used denial as a coping mechanism. That is, there may be a subgroup of siblings who experience and display symptoms of anxiety and, along with their parents, use denial to cope with stress from the cancer experience and maintain a sense of equilibrium (Fife et al., 1987).

Although some researchers utilized parent and child questionnaires and found siblings do not experience internalizing behavior problems, others report evidence of internalizing difficulties. For example, on a non-standardized questionnaire developed to examine parental perceptions of sibling experiences and adaptation to stress and the cancer experience, parents reported sibling adjustment problems including emotional lability and withdrawal (Carpenter & Sahler, 1991). These internalizing difficulties were reported to develop after the cancer diagnosis. Similarly, utilizing standardized parent questionnaires, Cohen and colleagues (1994) found siblings are reported to experience more internalizing behavior problems than a normative population.

Siblings report similar internalizing difficulties on non-standardized and standardized questionnaires. For example, on a non-standardized questionnaire designed to assess four areas (i.e., communication/social support, illness related issues, interpersonal relationships, and intrapersonal thoughts and feelings), siblings identified by parents as having post-diagnosis adjustment difficulties reported they protected their parents from their worries and felt isolated, ignored, and misunderstood (Carpenter & Sahler, 1991). More recent findings from standardized sibling questionnaires parallel these results and indicate, compared to a reference group, siblings experience significantly more positive emotion problems (e.g., little happiness, joy, satisfaction, enthusiasm) and negative emotion problems (e.g., depression, jealousy, anger, sadness, worry, fear) (Houtzager et al., 2005). Similarly, on standardized questionnaires, siblings reported more

emotional distress including symptoms of anxiety than a control group or the normal population (Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004; Sidhu, Passmore, & Baker, 2006).

In summary, investigations into the internalizing behavior problems of siblings revealed siblings and parents tend to report siblings experience internalizing behavior problems such as jealousy, anger, and symptoms of anxiety and depression. However, not all siblings experience internalizing behaviors that reach pathological levels and the results are somewhat inconsistent, varying with date of publication and method of data collection.

Externalizing Behavior Problems

Researchers have also investigated externalizing behaviors (e.g., acting out, aggression, non-compliance, inattention) in siblings. In one of the first studies to examine externalizing behavior problems, 81% of mothers reported during interviews that siblings exhibited difficulties including discipline problems at home and school (Powazek, Payne, Goff, Paulson, & Stagner, 1980). Results from other early research contradict this finding. For example, utilizing standardized questionnaires, Horwitz and Kazak (1990) and Sawyer and colleagues (1986) found siblings and healthy controls did not differ significantly on measures of externalizing behavior problems.

Relatively recent results from both standardized and non-standardized questionnaires completed by parents and/or siblings indicate siblings experience conduct problems and attention seeking behaviors (Cohen et al., 1994; Heffernan & Zanelli, 1997; Lahteenmaki et al., 2004). Specifically, Cohen and colleagues administered the CBCL (Achenbach, 1991) and found, compared to standardized norms, siblings are reported to exhibit significantly more externalizing behavior problems. Moreover, Heffernan and Zanelli administered non-standardized questionnaires to siblings and parents and found, following diagnosis, siblings report or are reported to exhibit new or more externalizing behavior problems including disrespect,

aggressiveness, and attention seeking behavior. Similarly, Sloper and While (1996) found approximately 24% of siblings scored in the clinical or borderline range on the behavior problems scale of the CBCL (Achenbach, 1991) and were reported by their parents and/or teachers to evidence negative behavior changes following diagnosis. These authors fail to indicate what behavioral changes were reported by parents. Contrary to these results, Van Dongen-Melman and colleagues (1995) found no differences in the behavioral adjustment of siblings and controls matched for age, gender, and socioeconomic status (SES). That is, parents reported on standardized questionnaires such as the CBCL that siblings do not exhibit significantly more externalizing behavior problems than a representative sample of the general population. Thus, in summary, research into the externalizing behavior problems of siblings is inconsistent as some siblings report or are reported to experience few difficulties and others experience diverse problems including physical and verbal aggression, inattention, and non-compliance. Similar to internalizing behavior problems research, however, research on externalizing behaviors appears to vary with publication date and/or informant.

Somatic Problems

Although somatic problems in siblings have not been studied extensively, empirical findings suggest stress and the cancer experience are related to numerous somatic difficulties. In early and more recent research, parents reported on standardized and non-standardized questionnaires siblings experienced head- and stomach-aches and sleeping and eating difficulties following cancer diagnosis (Carpenter & Sahler, 1991; Cohen et al., 1994; Heffernan & Zanelli, 1997; Powazek et al. 1980; Sahler et al., 1994; Zeltzer et al., 1996). Siblings also reported they experienced more somatic problems (e.g., sleeping and eating problems) than the normative population on standardized questionnaires (Zeltzer et al., 1996). Likewise, in recent research, 52% of 83 parents reported during interviews that siblings experienced health complaints related

to sleeping, eating, and physical functioning (Houtzager et al., 2005). Approximately 33% of parents reported siblings experienced physical complaints such as head- and stomach-aches, sickness, or other somatic symptoms (Houtazager et al., 2005). These results provide some evidence to indicate some siblings experience a host of somatic difficulties.

Summary

In summary, investigations into the effects of pediatric cancer on sibling adjustment have provided some conflicting results. Whereas the majority of empirical findings suggest siblings do not suffer from severe psychopathology, some results indicate siblings experience considerable psychological and somatic difficulties. Consequently, although research suggests most siblings adjust well, some siblings experience clinically significant internalizing and externalizing behavior problems and somatic difficulties. A critical examination of the literature, however, indicates the majority of the research is dated and many results are marred by methodological problems.

Although results from interviews tend to be consistent and suggest siblings experience a host of somatic and behavior problems, many researchers posed direct and leading questions such as, “Tell us about feelings of jealousy and guilt” (Gogan & Slavin, 1981) and “What did/do you worry about?” (Martinson et al., 1990) as opposed to general questions including, “What was it like for you when your brother/sister was diagnosed with leukemia?” (Kramer, 1984). Additionally, many siblings and parents provided retrospective accounts of the difficulties siblings faced (e.g., Gogan & Slavin, 1981). Thus, as research participants were frequently asked during interviews to recall problems encountered, often with leading questions, it is not surprising high and significant levels of psychological and somatic difficulties were often reported.

Methodological problems common to research that utilized interviews, standardized questionnaires, and/or non-standardized questionnaires pertain to sample sizes and methods of data analysis. Whereas some qualitative and quantitative studies including those of Cohen and colleagues (1994), Gogan and Slavin (1981), Schuler and colleagues (1985), and Sloper and While (1996) had larger sample sizes (i.e., between 81 and 129 participants), many researchers utilized small sample sizes. For example, of the 28 studies highlighted in a literature review by Houtzager and colleagues (1999), 17 had sample sizes of 35 or fewer participants and used tallies (e.g., Powazek et al., 1980), percentages (e.g., Fife et al., 1987), and/or thematic analyses (e.g., Chesler et al., 1991) to conclude siblings experience psychological and somatic difficulties. Moreover, many studies failed to include control or non-cancer participants and investigate whether psychological and somatic difficulties reported by parents and/or siblings differed significantly from typical siblings who did not face the cancer experience. That is, some researchers (e.g., Carpenter & Sahler, 1991; Chesler et al., 1991; Gogan & Slavin, 1981) conclude siblings experience psychological and somatic difficulties but failed to investigate and/or report on the extent to which the adjustment difficulties were atypical and thus, clinically relevant.

Lastly, as previously indicated, the majority of research in the field is dated and conducted at a time when the prognosis of the child with cancer was often poor and parents were encouraged to protect siblings from information about cancer, treatment, and the child's prognosis (Bluebond-Langner, 1978; Evans, 1968; Slavin, O'Malley, Koocher, & Foster, 1982; Share, 1972). Consequently, many adjustment difficulties frequently reported in early literature may reflect a lack of cancer knowledge in siblings and higher levels of stress, fear of the child's death, uncertainty, and family disruption common to siblings at that time. Given dated research and the aforementioned methodological problems of research that utilized interviews,

standardized questionnaires, and non-standardized questionnaires, the conclusions made by many researchers must be viewed with caution. Specifically, the extent to which research findings are reliable and generalizable to present-day siblings is questionable. Thus, it is imperative research address the aforementioned limitations and conduct a rigorous and current investigation of sibling psychological and somatic difficulties.

Factors Affecting Sibling Adjustment

Research into the impact of stress and the cancer experience on siblings suggests some siblings experience psychological and/or somatic difficulties. As previously discussed, however, results are inconsistent. In effort to account for the variation in research findings and identify siblings most in need of intervention support, some investigators have explored the relationship between sibling psychological adjustment and various sibling, family, and disease factors. Although many factors have received little empirical consideration and the results are variable, research suggests factors including time since diagnosis (e.g., Cohen et al., 1994; Hamama et al., 2000), sibling age (e.g., Barrera et al., 2004c; Bendor, 1990; Hamama et al., 2000; Houtzager et al., 2004; Sahler et al., 1994; Sargent et al., 1995; Spinetta, 1981; Van Dongen-Melman et al., 1995), and sibling gender (e.g., Alderfer et al., 2003; Barrera et al., 2004c; Hamama et al., 2000; Van Dongen-Melman et al., 1995) are associated with sibling psychological adjustment. Adjustment also appears to be influenced by social support (Barrera et al., 2004c; Williams et al., 2002) and sibling cognitive appraisal (Sloper & While, 1996).

Disease Factors

Few researchers have examined the impact of the diagnosis and prognosis of the child with cancer on sibling adjustment and published results are somewhat mixed (Houtzager et al., 1999). For example, diagnosis, prognosis, and time since diagnosis were not associated with emotional and behavioral problems in siblings, as reported by parents on standardized

questionnaires (Sahler et al., 1994). Similarly, utilizing standardized questionnaires, Cohen and colleagues (1994), Hamama and colleagues (2000), and Sloper and While (1996) found no relationship between diagnosis and sibling internalizing and/or externalizing behavior problems and Fife and colleagues (1987) found prognosis was not related to sibling anxiety. Contrarily, others have found some evidence for a direct relationship between diagnosis and sibling adjustment. Specifically, parents reported during interviews that cancer severity was associated with some positive effects (Barbarin, Sargent, Sahler, & Carpenter, 1995). Specifically, siblings of children with poorer prognoses were described as more mature, responsible, and independent (Barbarin et al., 1995). Additionally, Sloper and While (1996) found siblings with poor adjustment (i.e., borderline or clinical scores on the behavior problems scale of the CBCL), as compared to those with good adjustment, had siblings who spent more nights in the hospital. Sloper and While also found the relationship between prognosis and adjustment demonstrated a trend towards significance as siblings with poor adjustment were somewhat more likely to have a brother or sister with a poor prognosis.

The influence of time since diagnosis on adjustment has received more empirical consideration. An early examination that utilized standardized questionnaires and interviews found once treatment was complete, siblings evidenced similar emotional and behavioral adjustment as a group of randomly selected age-matched healthy peers (Van Dongen-Melman et al., 1995). Similarly, on a standardized questionnaire, mothers reported fewer internalizing and externalizing behavior difficulties with increased time since diagnosis (Cohen et al., 1994). Additionally, relatively recent research that utilized standardized questionnaires found recent diagnosis, as compared to longer time since diagnosis, was more highly associated with greater sibling anxiety (Hamama et al., 2000). At one month after diagnosis, Houtzager and colleagues (2005) found siblings both 7 to 11 years of age and 12 to 18 years of age experienced more

positive emotion problems (e.g., little happiness and enthusiasm) and negative emotion problems (e.g., anger, sadness, worry) than a normative population. The majority of these difficulties were absent at two years post-diagnosis (Houtzager et al., 2004).

Although the previous findings suggest adjustment difficulties decrease with increased time since diagnosis, some researchers have recently found sibling adjustment difficulties are prolonged. For example, Barrera and Atenafu (2008) found at two years post-bone marrow transplant, siblings had more internalizing behavior difficulties than survivors. Additional recent research that employed standardized questionnaires found, as compared to siblings of children without serious medical conditions, siblings of children with cancer reported more symptoms of posttraumatic stress, with a third reporting moderate to severe reactions (Alderfer et al., 2003). Moreover, a recent study that examined the drinking patterns of pediatric cancer survivors and siblings as adults found adult siblings consumed more alcohol than survivors and a peer population (Lown et al., 2008). Lown and colleagues suggest siblings may encounter stressors and demands that put them at risk for early and longer-term problematic alcohol use. Their results also indicate the cancer experience may be a risk factor for long-term adjustment difficulties that adult siblings attempt to manage with alcohol consumption.

In conclusion, literature pertaining to the relationships between diagnosis, prognosis, and length of time since diagnosis and sibling psychological adjustment is varied and inconclusive. Although the majority of research has utilized standardized questionnaires and suggests diagnosis and prognosis are not related to sibling internalizing and externalizing behavior problems, there is some empirical evidence suggesting increased time since diagnosis is related to decreased internalizing and externalizing behavior problems. Although some support comes from research that examined the direct relationship between time since diagnosis and sibling adjustment difficulties (e.g., Cohen et al., 1994; Hamama et al., 2000), other support comes from

research that examined sibling outcomes at various times after diagnosis. For example, whereas Van Dongen-Melman and colleagues (1995) examined psychological adjustment after cessation of treatment and found no internalizing or externalizing behavior problems, Houtzager and colleagues (2005) examined adjustment one month after diagnosis and found evidence of adjustment difficulties and impaired quality of life. Although important to the field's understanding of the relationship between time since diagnosis and sibling adjustment problems, such research can also explain, in part, why investigations into sibling adjustment produce conflicting findings. That is, results of sibling adjustment research may be inconsistent as time since diagnosis varies between studies and not all investigations include and/or examine participants with different lengths of time since diagnosis.

By investigating difficulties at only one point after diagnosis, researchers do not determine whether adjustment difficulties remain constant or decrease over time. For example, although Alderfer and colleagues (2003) report post-traumatic symptoms in siblings, these researchers did not investigate whether symptoms vary with time since diagnosis. Moreover, although there is evidence suggesting sibling adjustment difficulties wane over time and are not related to diagnosis and prognosis, most researchers do not examine whether these disease factors are related to sibling somatic problems and if there is an interrelationship between disease characteristics and sibling psychological and somatic difficulties. That is, few researchers examine whether the relationship between diagnosis and sibling adjustment is influenced by sibling and family factors such age, SES, social support, and cognitive appraisal. Such research is required to better understand the inconsistency in previous research and the present-day adjustment of siblings.

Sibling Age

Although largely dated, research into the relationship between age and psychological adjustment suggests siblings of different ages respond to stress and the cancer experience in different ways (Houtzager et al., 1999). In one of the earliest studies to examine the relationship between age and adjustment, researchers utilized sibling questionnaires and projective tests and found both younger (i.e., 4 to 6 year olds) and older (i.e., 7 to 12 year olds) siblings reported more adaptation difficulties than their brothers or sisters with cancer (Spinetta, 1981). Whereas younger siblings reported lower self-esteem and negative self-concept, older siblings reported more symptoms of anxiety and depression (Spinetta, 1981). Different psychological adjustment difficulties have also been reported by latency (i.e., 6 to 12 years old) and adolescent (i.e., 13 to 18 years old) siblings during interviews (Bendor, 1990). Whereas latency siblings reported adjustment problems including feelings of isolation, anxiety, low self-esteem, and internalized hostility, adolescents raised issues including reluctance to communicate with their parents and excessive concern for their parents and the child with cancer (Bendor, 1990).

In examining the relationship between sibling age and psychological adjustment difficulties, some researchers have examined whether the prevalence of adjustment difficulties experienced by younger (e.g., latency) and older (e.g., adolescent) siblings vary significantly. Although some researchers have employed standardized questionnaires and found age is not related to psychological adjustment difficulties including internalizing and externalizing behavior problems (e.g., Horwitz & Kazak, 1990), the literature generally suggests there are significant differences in the psychological difficulties experienced by siblings of different ages. For instance, during retrospective interviews, siblings aged 6 to 10 years reported feeling jealous more often than siblings aged 0 to 5 years and 11 to 21 years (Gogan & Slavin, 1981). Similarly, during interviews and on standardized questionnaires, siblings between the ages of 6 and 12 and their parents reported more symptoms of anxiety and depression and more externalizing behavior

problems than older siblings (Sahler et al., 1994; Sargent et al., 1995). Also during interviews, parents and older siblings (average age = 13.5 years old) reported fewer emotional problems (e.g., anxiety, low self-image, and feelings of isolation) than younger siblings (average age = 8.7 years old) (Schuler et al., 1985).

More recent research also supports statistically significant age differences. For example, adolescent siblings (i.e., 14 to 18 years old) reported fewer symptoms of anxiety than younger siblings (i.e., 9 to 13 years old) on standardized questionnaires (Hamama et al., 2000), and older siblings (i.e., 14 to 18 years old) were reported by their parents during interviews to display significantly more positive effects including increased maturity, supportiveness, and independence than younger siblings (i.e., 10 to 13 years old) (Barbarin et al., 1995). Additionally, on self-report standardized questionnaires, siblings aged 7 to 11 reported poorer emotional and social quality of life than a normative group (Houtzager et al., 2004). In contrast, there were no differences in adolescent (i.e., 12 to 18 years old) quality of life, although they reported more internalizing problems than the normative group (Houtzager et al., 2004).

In summary, the majority of research suggests both latency and adolescent siblings can experience psychological adjustment difficulties, but may evidence them in different manners. Although results can vary, younger siblings appear to report more symptoms of anxiety, jealousy and lower quality of life and self-esteem. Contrarily, adolescents report more positive effects and concern for their parents and brothers or sisters with cancer. However, researchers have typically failed to examine the relationship between sibling age and somatic difficulties. Moreover, many of the results are dated and few researchers have explored the interaction between sibling age, gender, and psychological adjustment.

Sibling Gender

Less is known about the psychological differences between male and female siblings. On a standardized post-traumatic stress questionnaire, female siblings reported greater adjustment difficulties including more intrusive thoughts, avoidance, and hyperarousal than male siblings (Alderfer et al., 2003). Additionally, adolescent females reported more symptoms of depression than adolescent males on a standardized questionnaire (Barrera et al., 2004c) and on a standardized self-report questionnaire, male siblings reported significantly more behavior problems (including internalizing and externalizing behaviors) than females (Sahler et al., 1994). Researchers including Sahler and colleagues also found significant age by gender interactions. Specifically, younger males (i.e., 4 to 11 years old) reported significantly more behavior problems (including internalizing and externalizing behavior problems) than older males (i.e., 12 to 17 years old) and both younger and older females (Sahler et al., 1994). Moreover, younger females experienced the greatest increase in behavior problems after diagnosis (Sahler et al., 1994) and in comparison to male and older female siblings, younger female siblings reported more feelings of loneliness on standardized questionnaires (Hamama et al., 2000).

In contrast, other researchers suggest the psychological and somatic difficulties of male and female siblings do not differ. For instance, Barbarin and colleagues (1995) interviewed parents and found the frequency and type of internalizing and externalizing behavior problems experienced by siblings after diagnosis were not related to gender. Utilizing a standardized questionnaire completed by parents, Sloper and While (1996) also found sibling gender was not related to the presence of psychological adjustment problems. Additionally, both male and female siblings were reported by parents on a standardized questionnaire to experience significantly higher somatization problems than nonclinical norms (Zeltzer et al., 1996).

Although researchers have utilized large sample sizes and standardized questionnaires and found, in general, male siblings experience more externalizing behavior problems and

females experience more internalizing behavior problems, empirical findings are limited and somewhat contradictory. Additionally, few researchers have examined the relationship between gender and somatic problems and how other factors (e.g., age) impact the relationship between gender and adjustment.

Family Characteristics

Although studied less extensively, family variables such as SES also appear to influence the psychological adjustment of siblings. For instance, some investigators have found SES negatively correlates with sibling adjustment difficulties including internalizing and externalizing problems (Cohen et al., 1994; Sahler et al., 1994; Sloper & While, 1996; Williams, et al., 2002; Zebrack et al., 2002). That is, siblings from families with lower SES (e.g., lower family income) experience more psychological problems, as reported by parents on standardized questionnaires. These results suggest high family income and SES are protective factors for psychological adjustment difficulties (Houtzager et al., 1999). Specifically, Houtzager and colleagues propose having the financial means to overcome medical costs and restrictions caused by the illness likely reduces distress in the family, and therefore, siblings.

There is also recent evidence indicating social support is related to sibling psychological adjustment. For example, Barrera and colleagues (2004c) found siblings between the ages of 6 and 18 years with high social support (as revealed by scores on a non-standardized self-report questionnaire) reported fewer behavior problems and symptoms of depression and anxiety on standardized questionnaires than those with low social support. Moreover, Carpenter and Sahler (1991) found siblings with post-diagnosis difficulties (e.g., emotional lability, attention seeking behavior, decreased academic performance) rated themselves as significantly more negative on inter-personal measures (e.g., feel ignored by others, unwanted, and misunderstood) than those without post-diagnosis difficulties. Furthermore, Williams and colleagues (2002) found

perceived social support of siblings of children with various chronic and congenital conditions including cancer was linked to various behavior problems, as reported by parents on a standardized questionnaire. Although investigations into the impact of social support on sibling adjustment is minimal and preliminary, research with children or adolescents with cancer found social support from parents, teachers, and class peers predicted lower psychological distress (Varni, Katz, Colegrove, & Dolgin, 1994) and social support reduced perceived stress in children one year after diagnosis (Varni & Katz, 1997). Moreover, researchers outside the area of pediatric oncology have consistently documented positive effects of high perceived social support on the psychological adjustment of children and adolescents in the presence of stress (e.g., Cauce, Felner, & Primavera, 1982; Demaray & Malecki, 2002; DuBois, Felner, Brand, Adan, & Evans, 1992; Licitra-Kleckler & Waas, 1993; Rueger, Malecki, & Demaray, 2008).

The majority of the aforementioned results are based on research that employed standardized questionnaires and utilized large sample sizes. Moreover, several researchers included both siblings and parents as informants and found similar reports with both informants (e.g., Barrera et al., 2004c). As such, the literature provides some solid evidence for a direct relationship between family factors such as SES and social support and sibling psychological difficulties. However, research into the impact of family factors on somatic problems is lacking and few researchers have examined what factors may influence the relationship between family factors and sibling psychological and somatic problems.

Cognitive Appraisal

When a child is diagnosed with cancer, siblings experience a host of difficult and challenging experiences including physical and emotional pain in the child with cancer, parental distress, uncertainty, decreased attention, and disrupted routines. These experiences are noted to be a source of stress for siblings (Barbarin et al., 1995; Lazarus & Folkman, 1984; Sloper &

While, 1996). Researchers including Lazarus and Folkman and Barbarin and colleagues indicate cognitive appraisal is a form of stress processing that influences how siblings perceive the stress and ultimately, adjust. Although cognitive appraisal is referred to as a single process by some researchers (e.g., Sloper & While, 1996), cognitive appraisal is typically differentiated into primary cognitive appraisal and secondary cognitive appraisal. Whereas primary cognitive appraisal is how one conceptualizes the threat of stress (e.g., illness) on his/her well-being, secondary cognitive appraisal is the evaluation of the coping resources available to manage the demands of the stress (Jenkins & Pargament, 1988; Juth, Smyth, & Santuzzi, 2008; Samsonn & Siam, 2008). For siblings, therefore, primary appraisal is the evaluation of whether the cancer experience presents stressors and demands that threaten their well-being. Secondary appraisal is the evaluation of whether they have the resources to cope with the stressors and challenges and ultimately, what they think can be done to overcome or manage the stress (Folkman, Lazarus, Gruen, & DeLongis, 1986a). When siblings believe the cancer experience threatens their well-being and they identify actions to manage the stress, siblings subsequently engage in coping. That is, siblings employ cognitive and behavioral strategies to manage (i.e., reduce, minimize) the stress and demands associated with the cancer experience (Folkman, et al., 1986a) and prevent psychological problems. Theoretically, therefore, primary and secondary cognitive appraisal are key and preliminary processes that influence the coping and thus, the psychological adjustment, of sibling (Folkman et al., 1986a; Wallander & Varni, 1992).

As primary cognitive appraisal is proposed to be a form of stress processing that accounts for the relationship between stress of the cancer experience and sibling psychological adjustment (Folkman et al., 1986a; Wallander & Varni, 1992), a few researchers have assessed if and how sibling cognitive appraisal and psychological adjustment are related. Using a non-standardized questionnaire developed to assess siblings' perceptions of the pediatric cancer experience, Sloper

and While (1996) found negative cognitive appraisals (e.g., "I feel mad about my brother/sister's illness," "I worry about my bother/sister's illness," "My bother/sister's illness makes me sad") are associated with adjustment difficulties including borderline and clinically significant internalizing and/or externalizing behavior problems. Moreover, Sloper and While found cognitive appraisal is more strongly associated with sibling psychological adjustment than disease, demographic, and social support variables. Given these preliminary findings and the proposed importance of cognitive appraisal to adjustment, research into the relationship between sibling cognitive appraisal and psychological and somatic difficulties is necessary. As primary cognitive appraisal is proposed to be a step of stress processing that precedes and influences coping efforts, examining the relationship between primary appraisal and sibling adjustment is a logical first step. However, to more precisely understand how and why sibling psychological adjustment varies, research must also examine how primary cognitive appraisal varies with factors such as age and indirectly influences sibling psychological adjustment. Such findings will ultimately guide and support future research into the relationships between cognitive appraisal, coping, and adjustment and provide direction for the development and implementation of sibling intervention programs.

Summary

In summary, siblings of children with cancer experience numerous stressors and disruptions in their daily lives and research suggests, in general, siblings experience psychological and somatic difficulties that may or may not reach pathological levels. Such difficulties include externalizing behavior problems (e.g., discipline problems, attention seeking behavior, aggressiveness), internalizing behavior problems (e.g., symptoms of depression and anxiety), lowered quality of life, and somatic problems (e.g., head- and stomach-aches, sleeping difficulties). However, there is great variability in the findings, likely due to methodological

differences (e.g., data collection techniques and instruments, informants) and limitations including small sample sizes, wide age ranges, and varying lengths of time since diagnosis. Moreover, much of the research was conducted when many childhood cancers had poor prognoses. As there have been many advances in the treatment of pediatric cancer and current survival rates are approximately 85% (Canadian Cancer Society, 2009), many empirical findings may not accurately reflect the extent to which siblings currently experience psychological and somatic difficulties. Furthermore, research results suggest numerous sibling (e.g., age, gender, cognitive appraisal), family (e.g., SES, social support), and disease (e.g., diagnosis and time since diagnosis) factors influence sibling adjustment and explain, in part, the variability in adjustment findings. Whereas the effect of factors such as age and SES on adjustment has been frequently examined, the direct and indirect effects of other factors (e.g., gender, cognitive appraisal, social support) have received less empirical consideration and/or lack consistent empirical support. Moreover, despite the development of conceptual models that attempt to explain psychological adjustment to chronic childhood illnesses and guide research and the development of interventions, few researchers and clinicians have utilized conceptual models to investigate the psychological and somatic difficulties of siblings.

Conceptual Model

Informed by research and theory, conceptual models identifying the influence of chronic illness on individuals and families were created to guide research and inform the development and implementation of intervention programs. The disability-stress-coping model (Wallander & Varni, 1992), a multivariable model informed by family systems theory and Bronfenbrenner's (1979) social ecology model, was developed to conceptualize the impact of illness-related stress on the adjustment of families and individual family members. Family systems theory posits family members are part of an interactive, interdependent, and dynamic network in which the

behavior of each individual family member (e.g., parents, siblings) impacts that of other members (Minuchin, 1988). Moreover, based on the social ecology model (Bronfenbrenner, 1979), a child and family are part of a larger system including extended family, societal factors, social networks, and culture. An individual's development, adjustment, and functioning, therefore, are affected by a plethora of factors including intrapersonal characteristics, the presence of family members, family subsystems (e.g., parents), and the larger settings and contexts in which he/she functions (e.g., school, hospital).

Wallander and Varni's (1992) disability-stress-coping model posits chronic disability or illness is a strain and source of stress that persistently interferes with an individual's performance of everyday activities and requires continual readjustment. Reflecting family systems theory and the social ecology model (Bronfenbrenner, 1979), this model predicts adjustment to chronic conditions is influenced by numerous personal and environmental factors termed risk and resistance factors. Whereas risk factors are sources of stress primarily responsible for causing adjustment difficulties, resistance factors buffer the impact of stress on adjustment by directly and/or indirectly influencing the risk-adjustment relationship (Wallander & Varni, 1992). That is, risk factors may cause stress and put individuals at risk for psychological problems and resistance factors may help manage the stress and prevent or reduce adjustment difficulties.

Developed to describe the adjustment of individuals to chronic illness and guide the development and implementation of support programs for these individuals, the disability-stress-coping model (Wallander & Varni, 1992) was recently employed to examine and describe the adjustment of parents and children with cancer (e.g., Barrera et al., 2003; Barrera et al., 2004b; Barrera et al., 2007). Thus, as proposed by this model, when a child is diagnosed with cancer, sibling adjustment is negatively impacted by three sources of stress: (a) ill child clinical factors (e.g., diagnosis, time since diagnosis, treatment status), (b) functional dependence in activities of

daily living (e.g., functional limitations, self sufficiency), and (c) psychological stressors (e.g., major life events, daily hassles). The resistance factors thought to influence the impact of these stressors include: (a) sibling characteristics (e.g., age, gender, temperament), (b) family factors (e.g., social support, SES, adjustment of family members), and (c) stress processing (e.g., cognitive appraisal, coping). Thus, according to the model, factors such as diagnosis and time since diagnosis may put siblings at increased risk for psychological difficulties and factors including age, social support, and cognitive appraisal directly or indirectly influence adjustment to reduce or minimize psychological difficulties.

Aims and Hypotheses

Previous research indicates a small population of siblings experience clinically significant adjustment difficulties. Moreover, sibling, family, and disease factors are reported to directly and indirectly influence sibling psychological adjustment. However, results are inconclusive and the majority of research is dated and atheoretical. Furthermore, although the disability-stress-coping model (Wallander & Varni, 1992) proposes variables including self-esteem and cognitive appraisal are directly and indirectly associated with psychological adjustment, these variables have received little empirical consideration.

Guided by the disability-stress-coping model (Wallander & Varni, 1992), the study investigates the present-day frequency of psychological and somatic difficulties in siblings and the influence of specific sibling, family, and disease factors on such problems. As the model is comprehensive, testing the entire model in a single study is not feasible (Thompson & Gustafson, 1996). The current study, therefore, examines a component of the model (Figure 1). This component was previously employed to examine the adjustment of parents (Barrera et al., 2004b) and children with cancer (Barrera et al., 2007; Barrera et al., 2003) to cancer diagnosis and treatment. Specifically, the study examines the direct and/or indirect impact of sibling

factors (i.e., age, gender, self-esteem, primary cognitive appraisal), family characteristics (i.e., SES, social support), and disease factors (i.e., diagnosis, days since diagnosis) on sibling psychological and somatic difficulties. In turn, the research strives to identify possible mechanisms through which cancer influences siblings and validate segments of the disability-stress-coping model (Wallander & Varni, 1992). Moreover, the research aims to ascertain the need for sibling intervention and which variables should be targeted through intervention to enhance sibling adjustment.

Aim 1: Examine Sibling Psychological and Somatic Difficulties

The first aim of the study is to describe the distribution of psychological problems (i.e., symptoms of anxiety and depression, internalizing and externalizing behavior problems) and somatic difficulties siblings experience, as reported by siblings and their parents.

Hypothesis 1: siblings and parents will report higher levels of symptoms of anxiety and depression, internalizing behavior problems, externalizing behavior problems, and somatic problems than the normative population.

Aim 2: Examine Direct Effects of Sibling, Family, and Disease Factors on Sibling Psychological and Somatic Difficulties

The second aim of the study is to examine the direct influence of sibling (i.e., age, gender, self-esteem, primary cognitive appraisal), family (i.e., social support, SES), and disease (i.e., diagnosis, days since diagnosis) factors on sibling psychological problems (i.e., symptoms of anxiety and depression, internalizing and externalizing behavior problems) and somatic difficulties (Figure 2).

Hypothesis 2A: age predicts sibling psychological adjustment, with increased age associated with fewer externalizing behavior and somatic problems.

Hypothesis 2B: gender predicts sibling psychological and somatic difficulties, with male siblings demonstrating fewer somatic problems and internalizing behavior problems (i.e., symptoms of anxiety and depression) than female siblings.

Hypothesis 2C: SES predicts sibling psychological adjustment, with higher SES associated with fewer symptoms of internalizing behaviors, externalizing behaviors, and somatic problems.

Hypothesis 2D: disease factors (i.e., diagnosis and days since diagnosis) predict sibling psychological outcomes, with more recent diagnoses and diagnoses such as brain tumors that are often associated with poorer prognoses, more side- and late-effects, and family disruption (Houtzager et al. 2001) associated with more psychological and somatic difficulties.

Hypothesis 2E: sibling cognitive appraisal, self-esteem, and social support predict sibling psychological adjustment, with higher self-esteem and perceived social support and positive cognitive appraisal associated with fewer psychological and somatic problems.

Aim 3: Examine Moderating Effects of Age

Guided by Baron and Kenny's (1986) conceptualization and statistical considerations of moderation, the third aim of the study is to examine the moderating effects of age and identify whether age influences the direction and/or strength of the relationship between predictor variables (i.e., gender, social support, primary cognitive appraisal) and sibling adjustment.

Hypothesis 3A: sibling age moderates the relationship between sibling gender and sibling psychological and somatic difficulties (Figure 3). Specifically, the psychological and somatic difficulties of male and female siblings will vary, but only for older siblings.

Hypothesis 3B: sibling age moderates the relationship between both primary cognitive appraisal and social support and sibling adjustment (Figure 3), with high perceived social support

and positive cognitive appraisal associated with fewer adjustment difficulties in older, but not younger, siblings.

Aim 4: Examine Mediating Effects of Primary Cognitive Appraisal

The fourth aim of the study is to examine the influence of primary cognitive appraisal on the relationship between sibling self-esteem and sibling psychological and somatic problems (Figure 4).

Hypothesis 4: primary cognitive appraisal mediates (i.e., accounts for) the effects of self-esteem on sibling psychological and somatic difficulties. That is, primary cognitive appraisal will be directly related to both self-esteem (e.g., siblings with high self-esteem will be more likely to have positive primary cognitive appraisals) and sibling adjustment (e.g., siblings with positive primary cognitive appraisal will be more likely to experience fewer psychological and somatic difficulties) and it will be primary cognitive appraisal, rather than self-esteem, that influences and accounts for variation in sibling adjustment.

CHAPTER TWO

Method

Participants

The participants were 108 siblings who were referred for intervention services offered at Sick Kids Hospital as part of a larger research project, due to parental concerns of behavioral and emotional difficulties. Inclusion criteria for this study were: was a sibling of a child with cancer who was treated at Sick Kids and was not considered palliative at the time of the sibling's participation; was between 7 and 17 years of age; and was considered healthy and without any diagnosed developmental or neurological disorders. Data used in this study were collected prior to the siblings' participation in the sibling intervention groups.

The participants included 51 males and 57 females. Table 1 presents demographic and disease characteristics of the child diagnosed with cancer. The mean age of the participants was 10.4 (SD = 2.5). Examination of the demographic factors revealed the majority of the participants' siblings were diagnosed with leukemia, lymphoma, or other blood disorders (60%). The average length of time since diagnosis was 538 days (SD = 649) and ranged from 16 to 3800 days. The SES composition of the participants, as denoted by the highest level of education completed by the siblings' mothers, indicated most mothers completed some post-secondary education (77%). The education level of 8 mothers was unreported.

Table 1

Demographic Characteristics of Participants and Disease Factors of Children with Cancer

	Frequencies	%
Gender		
Males	51	47
Females	57	53
Age		
Seven - nine	46	43
Ten - twelve	41	38
Thirteen - seventeen	21	19
Diagnosis of child with cancer		
Leukemia/Lymphoma/Other blood disorders	65	60
Brain tumor	14	13
Other tumor	29	27
Days since diagnosis		
1-365	56	52
366-730	34	32
731-1095	7	6
1096-1460	4	4
$\geq$ 1461	7	6
Socioeconomic status		
Grade 9-13	23	23
College/University	77	77

Procedure

Ethics approval for the present study was obtained from the Research Ethics Board at Sick Kids on March 27, 2008 and the Office of Research Ethics at the University of Toronto on April 29, 2008.

Siblings were referred by staff (i.e., nurses, physicians, social workers) in the Haematology/Oncology Department to participate in the group intervention. Siblings were screened for inclusion criteria and their parents were informed about the research intervention group during a pre-intervention session with a trained psychology student or staff member. Siblings who met inclusion criteria and siblings and parents who consented to participation completed a packet of questionnaires. The parent packet contained a questionnaire to obtain

demographic information and questionnaires to assess for symptoms of anxiety and internalizing behaviors, externalizing behaviors, and somatic problems. Siblings received packets containing questionnaires to assess symptoms of anxiety and depression, perceived social support, self-esteem, and primary cognitive appraisal. Reading assistance was provided as appropriate. Each sibling received a unique identification number and to ensure confidentiality, this number was used on all questionnaires and data.

Measures

Outcome Measures

The following outcome measures were completed to examine sibling internalizing behaviors (i.e., symptoms of depression and anxiety), externalizing behaviors, and somatic problems:

1. The *Children's Depression Inventory* (CDI; Kovacs, 1992). The CDI is a 27-item scale that assessed sibling reported symptoms of depression. It has an acceptable test-retest reliability coefficient (.83 at 3 weeks, .41 to .69 at 1 year). The total *T* score was used in this study, with higher scores representing more symptoms of depression.
2. The *State Anxiety* subscale of the *State-Trait Anxiety Inventory for Children* (STAIC; Spielberger, 1983). This measure was completed by siblings to assess symptoms of anxiety that may be related to the cancer experience (Spielberger, 1983) and has adequate reliability (.82 for males and .87 for females). Higher scores indicated siblings experienced more symptoms of anxiety. The STAIC-Parent Form, previously adapted from Spielberger to be identical in form and content to the children's measure (Barrera, Chung, and Fleming, 2004a), was completed by parents to examine their perceptions of sibling anxiety symptoms. The revised measure had adequate face and content validity. The test-retest alpha coefficient was .65 (Barrera et al., 2004a).

3. The *Child Behavior Checklist* (CBCL; Achenbach, 1991). This measure, completed by parents, consists of 118 items that assess various emotional and behavioral problems in children, summarized in several subscales and behavioral categories. The total problems, internalizing behavior problems, externalizing behavior problems, somatic complaints syndrome[4], and anxious/depressed syndrome[4] *T* scores were considered in this study as parent reported measures of general internalizing and externalizing behavior problems, somatic problems, and symptoms of anxiety and depression. The internalizing behavior problems subscale consists of 31 items (e.g., would rather be alone than with others, stomach aches or cramps, feels worthless or inferior), the externalizing behavior problems subscale consists of 33 items (e.g., lying or cheating, argues a lot, gets in many fights), and the total problems scale includes all but two test items (i.e., asthma, allergy). The somatic complaints syndrome includes 9 items (e.g., feels dizzy, overtired). Lastly, the anxious/depressed syndrome includes 14 items (e.g., complains of loneliness, too fearful or anxious). Higher scores suggested siblings experienced more behavior or somatic problems. The test-retest reliabilities of the internalizing behavior, externalizing behavior, and total problems scales were .89, .93, and .93 respectively. The test-retest reliabilities of the somatic complaints and anxious/depressed syndromes were .95 and .86 respectively.

Predictor Measures

The following questionnaires, completed by siblings, provided measures of self-esteem, social support, and primary cognitive appraisal. These factors were hypothesized to directly and/or indirectly impact sibling psychological adjustment.

[4] Throughout this dissertation, somatic complaints syndrome and anxious/depressed syndrome scores will be referred to as somatic complaints and anxious/depressed scores.

1. The *Self Perception Profile for Children* (SPPC; Harter, 1985a) or *Adolescents* (SPPA; Harter, 1988). The global self-worth subscale of this measure was employed to assess sibling perceptions of general self-esteem. The global self-worth subscale includes six items scored on a four-point Likert scale, with higher scores suggesting more positive self-esteem. The internal consistencies on U.S. samples were .80-.89 (Harter, 1988).
2. The *Sibling Perception Questionnaire* (SPQ; Carpenter & Sahler, 1991). Sibling social support was measured with the communication subscale consisting of 5 items scored on a five-point Likert response format: "I can talk to my parents about my school work," "I can talk to other people my age about my brother/sister's illness," "I can talk to my parents about my bother/sister's illness," "I can talk to other adults (like my teachers) about my brother/sister's illness," and "When my brother/sister was diagnosed, my parents told me about it." Primary cognitive appraisal was assessed with the 6 item intrapersonal thoughts and feelings subscale, scored on a five-point Likert response format: "I feel mad about my brother/sister's illness," "I worry about my bother/sister's illness," "I still wonder why my brother/sister got sick," "I understand my parents have to spend more time with my sick brother/sister," "My bother/sister's illness makes me sad," "I wish there was something I could do to make my brother/sister feel better." As scoring of the social support and cognitive appraisal subscales was designed to yield low social support and cognitive appraisal scores that represented high perceived social support and positive cognitive appraisals, these subscales were scored so high scores represented high social support and positive cognitive appraisal. These subscales are widely used to examine the impact of chronic illnesses such as cancer on siblings (Barrera et al., 2004c; Guite, Lobato, Kao, & Plante, 2004; Havermans, & Eiser, 1994; Lenton, Stallard, Lewis, Mastroyannopoulou, 2001; Lobato, & Kao, 2002; Lobato, Kao, & Plante, 2005; Sidhu et

al., 2006; Sloper, & While, 1996; Taylor, Fuggle, & Charman, 2001). The intrapersonal thoughts and feelings subscale was selected to assess primary cognitive appraisal as it was employed to examine cognitive appraisal in siblings (e.g., Sloper & While, 1996) and it includes items that specifically assess the influence of the cancer experience on a sibling's well-being, thoughts, and feelings. The SPQ has adequate validity and the internal consistencies for the thoughts and feelings and communication subscales are .74 and .48 respectively (Carpenter & Sahler, 1991). Additionally, for the purpose of the current study, the internal consistency of the SPQ was tested twice, 6 weeks apart, with a sample of 13 siblings. The average alpha coefficients at time 1 and time 2 were .71 and .80 respectively.

The following demographic and disease characteristics of the child with cancer were also collected: sibling age at the time of the first assessment, sibling gender, and maternal education as an index of family SES. Education was stratified as: (a) grade 9-13 and (b) post-secondary education (i.e., minimum of one semester of college or university). Disease factors of the child with cancer included: diagnosis (1. leukemia/lymphoma/other blood disorders, 2. brain tumor, 3. other tumor) and days since diagnosis at the time of the first assessment.

Statistical Analysis

Preliminary Analyses

A number of preliminary statistical analyses were conducted to: (a) select and reduce the number of outcome variables to be included in the regression analyses and (b) describe the population (e.g., mean, standard deviation, spread, frequency, skewness). Seven measures of internalizing behavior problems were collected: parent reported total problems, internalizing behavior problems, anxious/depressed, somatic problems, and STAIC scores, and sibling reported STAIC and CDI scores. To reduce the number of internalizing behavior measures,

Pearson product-moment correlations with listwise deletion and mean substitution for missing data between these outcome measures were conducted. When measures were highly correlated, one measure of child reported anxiety, child reported depression, parent reported anxiety, and parent reported depression was selected.

Secondly, Pearson and Spearman correlations with listwise deletion and mean substitution for missing data were conducted on the continuous and categorical variables respectively to examine associations between the predictor and outcome variables. Correlations between the predictor variables were examined for evidence of multicollinearity.

Aim 1 Analyses

Thirdly, descriptive and summary statistics were calculated with listwise deletion and mean substitution for missing data to describe the population. Moreover, the skewness and frequencies of each outcome variable were examined to identify the distribution of psychological and somatic difficulties siblings experienced and address Aim 1. In particular, the number of siblings who scored in the borderline and/or clinically significant range on each outcome measure were identified. Sibling reported anxiety and depression *T* scores 1.5 standard deviations above normative sample means were considered clinically significant (Kovacs, 1992; Spielberger, 1983). Parent reported internalizing and externalizing behavior *T* scores between 60 and 63 and above 63 were considered borderline and clinically significant respectively (Achenbach 1991). Parent reported somatic *T* scores between 67 and 70 and above 70 were considered borderline and clinically significant respectively (Achenbach 1991). The number of siblings with borderline and/or clinically significant scores were subsequently compared to normative data presented by Kovacs, Spielberger, and Achenbach.

Multiple imputation.

To manage missing data and avoid problems such as loss of power due to a decrease in the number of cases, an inflated risk of a type II error, and changing the distribution of the variable by decreasing the variance that arises from techniques such as listwise deletion and mean substitution (Buhi, Goodson, & Neilands, 2008), multiple imputation was conducted on the incomplete data set prior to the regression analyses. Multiple imputation is a three step process that lessens the risk of lowering the variance of the sample and enables one to examine the variance due to the imputation process (Buhi et al., 2008). In Step 1, missing values are replaced with 2 or more imputed values to create multiple data sets in which the observed and intact values are maintained across data sets (Buhi et al., 2008). The imputations are expected to give reasonable predictions for the missing data (Schafer, 1999) that account for the relationships between the missing and observed data (Horton & Lipsitz, 2001). In Step 2, the imputed data sets are analyzed using standard statistical analyses and the parameter estimates and standard errors from each data set are saved. Finally, in Step 3, the estimates are combined to make a single data set. Missing data in the present study were imputed 20 times, resulting in 20 data sets. After statistical analyses revealed no significant differences between them, the data sets were combined to yield one final data set. The regression analyses described below were conducted on this data set.

Regression Analyses

Aim 2: Examine direct effects of sibling, family, and disease factors on sibling psychological and somatic difficulties.

The direct relationships between sibling, family, and disease factors and sibling psychological and somatic difficulties were initially examined to test hypotheses 2A (age predicts sibling psychological adjustment, with increased age associated with fewer externalizing behavior and somatic problems), 2B (gender predicts sibling psychological and somatic

difficulties, with male siblings demonstrating fewer somatic problems and internalizing behavior problems than female siblings), 2C (SES predicts sibling psychological adjustment, with higher SES associated with fewer symptoms of internalizing behaviors, externalizing behaviors, and somatic problems), 2D (disease factors predict sibling psychological outcomes, with diagnoses such as brain tumors and more recent diagnoses associated with more psychological and somatic difficulties), and 2E (sibling primary cognitive appraisal, self-esteem, and social support predict sibling psychological adjustment, with higher self-esteem and perceived social support and positive cognitive appraisal associated with fewer internalizing, externalizing, and somatic problems).

As the study is guided by the disability-stress-coping model (Wallender & Varni, 1992) and attempts to validate segments of the model, independent variables were entered into hierarchical regression analyses, according to the model. At Step 1, the covariates age, gender, and SES were entered. Age was entered as a continuous variable for each regression model. However, as stratifying age into latency aged (i.e., 7-12 years) and adolescent (i.e., 13-17 years) siblings resulted in significant age effects for internalizing behavior problems, age was also entered as a dichotomous variable with two levels (i.e., 0 = latency, 1 = adolescents) for the internalizing behavior regression model. The disease factors, diagnosis and days since diagnosis, were entered at Step 2. Diagnosis was entered as a dichotomous variable that was dummy coded (i.e., 0 = other tumor, leukemia, lymphoma, other blood disorder, 1 = brain tumor) as only brain tumor diagnoses were significantly associated with sibling adjustment. Self-esteem, social support, and primary cognitive appraisal were entered at Step 3.

Aim 3: Examine moderating effects of age.

Hierarchical regression analyses outlined by Baron and Kenny (1986) were conducted to test the moderating effects identified in hypotheses 3A (sibling age moderates the relationship

between sibling gender and adjustment) and 3B (sibling age moderates the relationship between both primary cognitive appraisal and social support and sibling adjustment). Prior to conducting the analyses, sibling age, cognitive appraisal, and social support scores were centered to facilitate interpretation and reduce multicollinearity between the predictor variables (Aiken & West, 1991). At Step 1, the relationship between the hypothesized moderator variable (i.e., age) and outcome variable (e.g., symptoms of depression) was entered. At Step 2, the relationship between an independent variable (e.g., gender) and the outcome variable was entered. At Step 3, the interaction between the independent variable and moderator variable was entered to examine whether the latter moderated the relationship between the independent variable and sibling adjustment. A moderator hypothesis was supported if the interaction (i.e., Step 3) was significant (Baron & Kenny, 1986). Significant main effects at Steps 1 and 2 were not required for significant moderation (Baron & Kenny, 1986). To display the nature of significant interactions, Aiken and West (1991) recommend simple slope regression lines for predictor (e.g., social support) and moderator (i.e., age) scores that fall one standard deviation above and below the mean are plotted. However, an examination of group endorsement revealed very few participants met these criteria and fell one standard deviation above or below the mean. Thus, to more accurately reflect group endorsement, simple slope regression lines for predictor and moderator scores that fell half a standard deviation above or below the mean were plotted. Procedures outlined by Aiken and West were subsequently used to determine if the simple slope regression lines were significantly different from zero and therefore, identify the nature of all significant moderations. Lastly, independent *t*-tests comparing adjustment scores for younger and older siblings at each predictor level (e.g., gender, social support, cognitive appraisal) were computed to investigate potential age differences for various predictors (e.g., low social support).

Aim 4: Examine mediating effects of cognitive appraisal.

Hierarchical regression analyses, as outlined by Baron and Kenny (1986), were conducted to address Aim 4 and explore whether primary cognitive appraisal mediates the relationship between sibling self-esteem and sibling adjustment (i.e., symptoms of anxiety and depression, internalizing and externalizing behavior problems, and somatic problems). At Step 1, sibling self-esteem scores were entered to identify whether self-esteem was significantly associated with sibling adjustment. At Step 2, sibling primary cognitive appraisal scores were entered to examine if cognitive appraisal was associated with sibling outcomes. At Step 3, the association between self-esteem and primary cognitive appraisal scores was entered to examine whether self-esteem scores predicted cognitive appraisal scores. At Step 4, self-esteem and primary cognitive appraisal scores were entered together to identify whether collectively, they predicted sibling adjustment. To demonstrate mediation, self-esteem should be less highly associated with sibling adjustment when cognitive appraisal is included as a predictor. The degree to which the association between self-esteem and an outcome variable was reduced (i.e., decrease in regression coefficients) from Step 1 to Step 4 was an indicator of the potency of the mediation (Baron & Kenny, 1986; Holmbeck, 1997). Post-hoc probing of significant mediational effects was conducted using Baron and Kenny's procedures and version of the Sobel (1982) equation.

Statistical Power and Type I and Type II Error Considerations

Cohen (1992) indicates four variables are involved in all statistical inferences: sample size, significance criterion (alpha), population effect size, and statistical power. Each variable influences the others (Cohen, 1992) and must be considered when attempting to minimize the risk of both Type I errors (i.e., rejecting the null hypothesis when it is true) and Type II errors (i.e., accepting the null hypothesis when it is false). Statistical power is the probability a test will correctly reject the null hypothesis, with increased power associated with a decreased risk of a

Type II error. Harris (1985) suggests to achieve adequate power in regression analyses, ten participants for every predictor variable are required. Thus, the current sample size of 108 participants was sufficient to obtain regression results with adequate power. However, LeCroy and Krysik (2007) argue effect size measures are important to interpreting statistical results as they provide different information than alpha levels. That is, effect sizes identify the strength of the relationship between two variables (e.g., predictor variable age and outcome variable anxiety symptoms) and therefore, the practical importance of the results (LeCroy & Krysik, 2007). Regression R^2 change values were considered as a measure of effect size (Cohen, 1988; LeCroy & Krysik, 2007) for the direct and moderation regression analyses, with R^2 values of .02, .15, and .35 representing small, medium, and large effect sizes respectively (Cohen, 1988).

The significance criterion, alpha, represents the maximum risk of making a Type I error (Cohen, 1992). All preliminary and regression analyses were tested with an alpha level of .05. As twenty five regression analyses were conducted with an alpha level of .05, the expected number of false significant results was 1.25. Although a more conservative alpha value of .01 or a Bonferroni adjustment may have reduced the likelihood of a Type I error, the latter may have resulted in an overly conservative alpha level of .002 (i.e., .05/25 regression analyses) (Mundfrom, Perrett, Piccone, & Roozeboom, 2006). Such an adjustment may have greatly enhanced the likelihood of making a Type II error.

CHAPTER THREE

Results

Preliminary Analyses

Pearson correlations among the measures of internalizing behavior problems (i.e., parent reported total problems, internalizing behavior problems, anxious/depressed, and STAIC scores, and sibling reported STAIC and CDI scores) revealed the following highly significant correlations[5]: parent reported internalizing behavior problems scores and parent reported anxious/depressed scores (.85, $p < .001$), parent reported STAIC scores and parent reported internalizing behavior problems scores (.62, $p < .001$), and parent reported STAIC scores and parent reported anxious/depressed scores (.53, $p < .001$). Moreover, parent reported STAIC scores were highly correlated with sibling reported STAIC scores (.52, $p < .001$) and parent reported total behavior problems were highly correlated with parent reported internalizing behavior problems (.93, $p < .001$). Given these correlations, parent reported internalizing behavior problem scores were selected to represent parent perceptions of sibling internalizing behavior problems. This measure, in addition to sibling reported STAIC and CDI scores, were included in the remaining preliminary and regression analyses.

Table 2 presents results from the bivariate correlations between the eight predictor variables (i.e., age, gender, SES, diagnosis, days since diagnosis, self-esteem, social support, primary cognitive appraisal) and five outcome factors (i.e., sibling reported symptoms of anxiety and depression, parent reported internalizing and externalizing behavior problems, parent reported somatic problems). Whereas age, days since diagnosis, self-esteem, social support, and primary cognitive appraisal were entered as continuous variables, SES and diagnosis were entered as categorical variables. As indicated in Table 2, few predictor variables correlated with outcome factors. Sibling self-esteem, social support, and primary cognitive appraisal were

[5] As the preliminary analyses conducted with listwise deletion and mean substitution yielded comparable results, only the results with listwise deletion are presented.

associated with sibling reported symptoms of anxiety and depression and parent reported internalizing behavior problems, externalizing behavior problems, and/or somatic problems. Gender only correlated with parent reported somatic problems. Age, SES, diagnosis, and days since diagnosis did not correlate with any of the outcome variables. Although not presented in Table 2, the predictor variables were not highly correlated. As there was no evidence of multicollinearity, the eight predictor and five outcome variables were included in the regression analyses.

Table 2

Bivariate Correlations for Independent and Dependent Variables

	Dependent variables				
Independent variables	Sibling reported anxiety (STAIC)	Sibling reported depression (CDI)	Parent reported internalizing behavior problems (CBCL)	Parent reported externalizing behavior problems (CBCL)	Parent reported somatic problems (CBCL)
Age	.13	.11	-.12	.01	-.09
Gender	.01	.15	.10	-.01	.22*
SES	.00	.06	.01	.15	.01
Diagnosis	-.03	-.17	-.08	-.05	-.15
Days since diagnosis	-.01	.08	-.09	.02	-.01
Self-esteem	-.28**	-.44**	.08	-.16	.06
Social support	-.31**	-.32**	-.29**	-.29**	-.22*
Primary cognitive appraisal	-.39**	-.43**	-.03	-.02	-.11

*$p < .05$, **$p < .01$

Aim 1: Examine Sibling Psychological and Somatic Difficulties

The results of the descriptive analyses for the continuous predictor (i.e., age, days since diagnosis, self-esteem, social support, and primary cognitive appraisal) and outcome variables (i.e., symptoms of anxiety and depression, internalizing behavior problems, externalizing behavior problems, and somatic problems) are presented in Table 3. The predictor variables, age

and days since diagnosis, were positively skewed and self-esteem was slightly negatively skewed. Sibling CDI and STAIC scores and parent reported somatic problem scores were positively skewed. As these variables were not normally distributed, a log transformation of each variable was conducted and regression analyses were conducted on both untransformed and transformed data sets. As the results were similar, only results on the untransformed data are reported.

Table 3

Descriptive Statistics for Predictor and Outcome Variables

Measure	N	M	SD	Min	Max	Skew	SE
Age	108	10.4	2.5	7	17	.65	.23
Days since diagnosis	108	538	649	16	3800	3.10	.23
Self-esteem	89	3.4	.57	1.80	4.00	-.84	.26
Social support	104	17.63	4.53	7.00	25.00	-.35	.24
Primary cognitive appraisal	103	18.61	5.10	8.00	29.00	.01	.24
Sibling reported anxiety (STAIC)	103	30.93	7.86	17.00	57.00	1.01	.24
Sibling reported depression (CDI)	105	48.15	11.24	35.00	100.00	1.82	.24
Parent reported internalizing behaviors (CBCL)	103	52.95	11.50	31.00	79.00	.08	.24
Parent reported externalizing behaviors (CBCL)	103	49.94	10.47	30.00	79.00	.07	.24
Parent reported somatic problems (CBCL)	102	56.01	7.21	50.00	79.00	1.23	.24

The number of siblings with borderline and/or clinically significant outcome scores were examined to test Hypothesis 1 (siblings and parents report higher levels of symptoms of anxiety and depression, internalizing behavior problems, externalizing behavior problems, and somatic problems than the normative population). These results, in addition to normative data, are presented in Table 4. Based on normative data presented by Kovacs (1992) and Spielberger

(1983), 8% and 15% of siblings reported clinically significant levels of depression and anxiety respectively. Normative data presented by Kovacs and Spielberger suggests approximately 7% of children and adolescents in the normal population experience clinically significant levels of depression or anxiety. Thus, the results indicate siblings experienced normative levels of depression symptoms and higher rates of clinically significant anxiety symptoms than the normative population. Based on normative data presented by Achenbach (1991), 30% of parents reported their sons/daughters experienced borderline or clinical levels of internalizing behavior problems. Nineteen percent were clinically significant. Fewer parents reported borderline (4%) or clinical (11%) levels of externalizing behavior problems. Normative data (Achenbach, 1991) suggests 18% of children and adolescents are reported to experience borderline or clinically significant internalizing or externalizing behavior problems. Thus, whereas siblings were reported to experience higher rates of borderline and clinical levels of internalizing behavior problems than the normative population, their externalizing behavior scores were comparable to normative values. Although only 11% of parents reported borderline or clinically significant levels of somatic problems, 8% reported clinical levels. These levels are markedly higher than the 2% of children and adolescents in the normative population reported to experience borderline or clinically significant somatic problems (Achenbach, 1991).

Table 4

Frequency of Borderline and/or Clinically Significant Psychological and Somatic Problems

Outcome measure	% of siblings	% of normative population
Sibling reported anxiety (STAIC)	15	7
Sibling reported depression (CDI)	8	7
Parent reported internalizing behaviors (CBCL)	30	18
Parent reported externalizing behaviors (CBCL)	15	18
Parent reported somatic problems (CBCL)	11	2

Regression Analyses

Aim 2: Examine Direct Effects of Sibling, Family, and Disease Factors on Sibling Psychological and Somatic Difficulties

Sibling reported depression (CDI).

Results of the depression regression analysis are presented in Table 5. At Step 1, the predictors age, gender, and SES did not significantly predict symptoms of depression (R^2 = .03, F(3, 96) = 1.12, p > .05). At Step 2, brain tumor diagnosis and days since diagnosis did not significantly predict sibling reported depression scores (R^2 change = .03, F(2, 94) = 1.64, p > .05 respectively). At Step 3, self-esteem, social support, and primary cognitive appraisal were associated with symptoms of depression, after the effects of age, gender, SES, brain tumor diagnosis, and days since diagnosis were controlled for (R^2 change = .36, F(3, 91) = 18.64, p < .01). The addition of these variables had a large effect size (Cohen, 1988), accounting for 36% of the variance in depression scores. These results suggest siblings with higher self-esteem and social support and positive primary cognitive appraisal reported fewer symptoms of depression.

Table 5

Summary of Final Model of Analyses Regressing Predictor Variables on Sibling Reported Depression Scores (CDI)

	Predictor	R^2 change	*ß*	S.E. *ß*	*p*
Step 1		.03			
	Age		-.21	.38	.574
	Gender		-3.02	1.85	.107
	SES		-.92	2.20	.677
Step 2		.07			
	Brain tumor		3.47	2.95	.242
	Days since diagnosis		.00	.00	.125
Step 3		.36			
	Self-esteem		-8.01	1.91	.000
	Social support		-.55	.21	.011
	Primary cognitive appraisal		-.70	.19	.001

Sibling reported anxiety (STAIC).

Table 6 displays the regression results for sibling reported symptoms of anxiety. At Step 1, age, gender, and SES were not significantly related to sibling reported symptoms of anxiety (R^2 = .03, F(3, 96) = 1.09, $p > .05$). At Step 2, brain tumor diagnosis and days since diagnosis significantly predicted anxiety (R^2 change = .08, F(2, 94) = 4.38, $p < .05$), though with a small effect size (Cohen 1988), after the effects of age, gender, and SES were controlled for. These results indicate when children were diagnosed with brain tumors, as compared to leukemia, lymphoma, other blood disorders, and other tumors, siblings reported significantly more anxiety symptoms. When self-esteem, social support, and primary cognitive appraisal were entered at Step 3, the model was significant (R^2 change = .22, F(3, 91) = 10.20, $p < .01$), with a medium effect size (Cohen, 1988) and after controlling for the effects of sibling age, gender, SES, days since diagnosis, and brain tumor diagnosis. The addition of these variables accounted for 22% of the variance in anxiety scores. However, only primary cognitive appraisal and social support

were significantly related to sibling reported anxiety scores. These results suggest positive primary cognitive appraisal and high perceived social support were associated with fewer symptoms of anxiety, as reported by siblings.

Table 6

Summary of Final Model of Analyses Regressing Predictor Variables on Sibling Reported Anxiety Scores (STAIC)

	Predictor	R^2 change	*ß*	S.E. *ß*	*p*
Step 1		.03			
	Age		.32	.28	.260
	Gender		.21	1.39	.882
	SES		-1.82	1.65	.275
Step 2		.08			
	Brain tumor		5.17	2.22	.022
	Days since diagnosis		.00	.00	.200
Step 3		.22			
	Self-esteem		-1.59	1.44	.272
	Social support		-.50	.16	.003
	Primary cognitive appraisal		-.53	.15	.000

Parent reported internalizing behavior problems (CBCL).

At Steps 1 and 2, the covariates age, gender, SES, brain tumor diagnosis, and days since diagnosis did not predict parent reported internalizing behavior problems (R^2 = .03, F(3, 96) = .89, $p > .05$, R^2 change = .02, F(2, 94) = .99, $p > .05$). However, when age was dichotomized into younger (i.e., 7-12 years) and older (i.e., 13-17 years) siblings at Step 1, age, gender, and SES significantly predicted internalizing behaviors (R^2 = .08, F(3, 96) = 2.67, $p < .05$), with only age significantly associated (Table 7) and with a small effect size (Cohen, 1988). Descriptive analyses indicate parents reported siblings 7 to 12 years of age (M = 55.22, SD = 11.53) had more internalizing problems than siblings 13 to 17 years of age (M = 47.20, SD = 8.68). Brain tumor diagnosis and days since diagnosis did not predict internalizing problems after the effects

of gender, SES, and dichotomous age were controlled for (R^2 change = .02, F(2, 94) = .95, p > .05). At Step 3, self-esteem, social support, and primary cognitive appraisal significantly predicted internalizing problems (R^2 change = .07, F(3, 91) = 2.65, p < .05), when the effects of the covariates were controlled for. However, only perceptions of high social support were associated with low parent reported internalizing behavior problems (Table 7). Moreover, the addition of self-esteem, social support, and cognitive appraisal to the model had a small effect size (Cohen, 1988).

Table 7

Summary of Final Model of Analyses Regressing Predictor Variables on Parent Reported Internalizing Behavior Scores (CBCL)

	Predictor	R^2 change	*ß*	S.E. *ß*	*p*
Step 1		.08			
	Age		-7.87	3.02	.011
	Gender		-2.22	2.26	.329
	SES		-.21	2.68	.939
Step 2		.02			
	Brain tumor		1.95	3.59	.588
	Days since diagnosis		.00	.00	.297
Step 3		.07			
	Self-esteem		-.81	2.37	.734
	Social support		-.65	.26	.014
	Primary cognitive appraisal		-.15	.24	.520

Parent reported externalizing behavior problems (CBCL).

Table 8 presents results from the externalizing behavior problems regression analysis. Age, gender, SES, brain tumor diagnosis, and days since diagnosis were not related to externalizing behavior problems at Steps 1 and 2 (R^2 = .02, F(3, 96) = .57, p > .05, R^2 change = .02, F(2, 94) = 1.04, p > .05). At Step 3, self-esteem, social support, and primary cognitive appraisal significantly predicted parent reported externalizing problems (R^2 change = .12, F(3,

91) = 4.23, $p < .01$), adjusted for the effects of the covariates. However, only higher social support was significantly related to fewer externalizing behavior problems. The addition of self-esteem, social support, and cognitive appraisal had a small effect size (Cohen, 1988).

Table 8

Summary of Final Model of Analyses Regressing Predictor Variables on Parent Reported Externalizing Behavior Scores (CBCL)

	Predictor	R^2 change	*ß*	S.E. *ß*	*p*
Step 1		.02			
	Age		-.33	.42	.440
	Gender		-.39	2.08	.854
	SES		2.78	2.47	.263
Step 2		.02			
	Brain tumor		2.78	3.31	.403
	Days since diagnosis		.00	.00	.945
Step 3		.12			
	Self-esteem		-3.97	2.15	.068
	Social support		-.59	.24	.015
	Primary cognitive appraisal		.02	.22	.922

Parent reported somatic problems (CBCL).

Hierarchical regression results for somatic problems are presented in Table 9. At Step 1, the covariates age, gender, and SES did not predict somatic problems ($R^2 = .06$, F(3, 96) = 1.91, $p > .05$). However, gender made a significant contribution, with a small effect size (Cohen, 1988). Descriptive results indicated parents reported fewer somatic problems in male siblings than female siblings (M females = 57.08, SD = 7.65; M males = 54.73, SD = 6.10). At Step 2, brain tumor diagnosis and days since diagnosis did not significantly predict somatic problems (R^2 change = .01, F(2, 94) = .41, $p > .05$). At Step 3, self-esteem, social support, and primary cognitive appraisal did not significantly predict somatic problems (R^2 change = .07, F(3, 91) = 2.53, $p > .05$). However, higher perceived social support was significantly associated with fewer

parent reported somatic problems. Moreover, adding self-esteem, social support, and cognitive appraisal had a small effect size (Cohen, 1988).

Table 9

Summary of Final Model of Analyses Regressing Predictor Variables on Parent Reported Somatic Scores (CBCL)

	Predictor	R^2 change	*β*	S.E. *β*	*p*
Step 1		.06			
	Age		-.25	.28	.369
	Gender		-3.26	1.37	.020
	SES		.76	1.63	.640
Step 2		.01			
	Brain tumor		-1.34	2.18	.541
	Days since diagnosis		.00	.00	.345
Step 3		.07			
	Self-esteem		1.35	1.42	.343
	Social support		-.37	.16	.021
	Primary cognitive appraisal		-.22	.14	.120

In summary, the regression analyses indicate the predictor variables entered into the regression models had small to large effect sizes, with between 14% and 42% of the variance in outcome scores accounted for by the predictor variables. By examining the direct effect of sibling, family, and disease factors on sibling psychological and somatic difficulties, several hypotheses were confirmed. The sibling, family, and/or disease factors significantly associated with fewer sibling and parent reported psychological and/or somatic problems are presented in Figure 5. The hierarchical regression results provide some evidence to suggest age and gender account for variation in sibling psychological adjustment, with male siblings reported to have fewer somatic problems than female siblings. Moreover, younger siblings (i.e., 7 to 12 years) were reported to have more internalizing problems than older siblings (i.e., 13 to 17 years). Sibling SES was not significantly associated with sibling outcomes. The hypothesis that disease

factors would predict sibling adjustment was partially supported. Having a brother or sister diagnosed with a brain tumor was associated with more symptoms of anxiety. None of the other outcomes, however, were related to disease factors.

The hypothesis that sibling self-esteem, social support, and primary cognitive appraisal were significantly related to sibling psychological and somatic difficulties was supported as these variables were significantly associated with sibling reported symptoms of depression. Social support and primary cognitive appraisal were significantly associated with sibling reported anxiety symptoms. Only sibling social support predicted parent reported internalizing behavior problems, externalizing behavior problems, and somatic problems. Collectively, the current results indicate siblings who reported higher self-esteem were less likely to experience symptoms of depression than siblings who reported lower self-esteem. Additionally, siblings who reported higher perceived social support were less likely to report symptoms of anxiety and depression. Moreover, the parents of these siblings were less likely to report internalizing and externalizing behavior problems and somatic problems than those whose children reported lower perceived social support. Lastly, siblings who reported positive primary cognitive appraisals, as compared to siblings who reported negative primary cognitive appraisals, were less likely to report symptoms of anxiety and depression.

Aim 3: Examine Moderating Effects of Age

The third aim of the study was to examine the moderating effects of age. Age was predicted to moderate the relationship between sibling gender, primary cognitive appraisal, and social support and sibling psychological and somatic difficulties. For each hierarchical regression analysis, the relationship between the proposed moderator variable (i.e., age) and sibling outcome (e.g., depression symptoms) was entered at Step 1, the relationship between an independent variable (e.g., gender) and the outcome was entered at Step 2, and the interaction

between the predictor variables was entered at Step 3. Results, presented below, revealed significant age x gender, age x social support, and age x cognitive appraisal moderations. Regression results for non-significant moderations are presented in Appendix A.

Age x gender.

Age moderated the relationship between gender and symptoms of depression (R^2 change = .04, F(1, 104) = 4.25, $p < .05$), after the direct effects of age and gender were controlled for. The effect size was small (Cohen, 1988). As a follow-up, sibling gender scores and sibling age scores that fell half a standard deviation above and below the mean were plotted to display the nature of this interaction (Figure 6). Testing of simple slopes (Aiken & West, 1991) revealed the simple slope of younger siblings was not significantly different from zero ($p > .05$), but the slope of the line representing older siblings was significantly different from zero ($p < .05$). Thus, for older siblings, gender predicted a change in depression scores, with male siblings reporting fewer symptoms of depression than female siblings. In contrast, gender did not predict a change in sibling reported depression scores for younger siblings. Post-hoc independent t-tests comparing younger and older female siblings and younger and older male siblings indicated both younger and older female siblings and younger and older male siblings did not report significantly different depression scores ($t(43) = -1.59$, $p > .05$ and $t(35) = 1.0$, $p > .05$, respectively).

Age x social support.

Age was also found to moderate the relationship between social support and depression scores, with a small effect size (Cohen, 1988), after the direct effects of age and social support was controlled for (R^2 change = .04, F(1, 104) = 4.39, $p < .05$) (Figure 7). Testing of simple slopes (Aiken & West, 1991) indicated the simple slope representing younger siblings ($p < .01$), but not older siblings ($p > .05$), was significantly different from zero. Thus, social support predicted a change in depression scores for younger, but not older siblings, with younger siblings

with high social support reporting fewer symptoms of depression than those with low social support. These results do not support the hypothesis that high perceived social support would be associated with fewer adjustment difficulties in older, but not younger, siblings. Independent *t*-tests revealed younger siblings with low perceived social support did not have significantly different depression scores than older siblings with low social support ($t(27) = -1.67$, $p > .05$). Similarly, no significant difference in depression scores was found for younger and older siblings with high perceived social support ($t(28) = .92$, $p > .05$).

Age x cognitive appraisal.

Lastly, age moderated the relationship between cognitive appraisal and symptoms of depression, after the effects of age and cognitive appraisal was controlled for (R^2 change = .03, $F(1, 104) = 4.42$, $p < .05$) (Figure 8). Age also moderated the relationship between primary cognitive appraisal and both internalizing behavior problems (R^2 change = .04, $F(1, 104) = 4.42$, $p < .05$) (Figure 9) and somatic problems (R^2 change = .06, $F(1, 104) = 6.21$, $p < .05$) (Figure 10). The effect size for each interaction was small (Cohen, 1988). Testing of the simple slopes (Aiken & West, 1991) revealed the slope representing younger siblings ($p < .05$), but not older siblings ($p > .05$), was significantly different from zero for somatic problems. Thus, primary cognitive appraisal predicted a change in somatic scores for younger, but not older siblings, with younger siblings with positive primary cognitive appraisals experiencing fewer somatic difficulties than those with negative cognitive appraisals (Figure 10). Testing of the simple slopes revealed the slopes representing both younger and older siblings was significantly different from zero for symptoms of depression ($p < .01$) (Figure 8). Thus, although primary cognitive appraisal predicted a change in depression scores for younger and older siblings, it had a greater influence for younger siblings. Lastly, although the internalizing behavior problems regression results revealed a significant age by primary cognitive appraisal interaction, the slopes

representing younger and older siblings were not significantly different from zero ($p > .05$) (Figure 9). The significant internalizing behavior problems regression interaction, therefore, may be a Type I error and reflect adequate power of the sample size, rather than a true interaction. These results do not support the hypothesis that positive cognitive appraisal would be associated with fewer adjustment difficulties in older, but not younger, siblings.

Follow-up *t*-tests revealed few differences between younger and older siblings with negative or positive cognitive appraisals. Specifically, younger and older siblings with negative cognitive appraisals had depression, internalizing behavior, and somatic scores that were not significantly different ($t(27) = -1.99$, $p > .05$, $t(27) = -1.47$, $p > .05$, and $t(27) = -1.41$, $p > .05$ respectively). Similarly, the depression and internalizing behavior scores for younger and older siblings with positive appraisals were not significantly different ($t(21) = -.01$, $p > .05$, $t(21) = -1.85$, $p > .05$, respectively). In contrast, although not apparent in Figure 10, follow-up *t*-tests indicated the somatic scores of younger and older siblings with positive cognitive appraisals were significantly different from one another ($t(21) = 2.69$, $p < .05$), with older siblings (M = 52.70, SD = 4.38) experiencing fewer somatic problems than younger siblings (M = 59.18, SD = 6.97).

Aim 4: Examine Mediating Effects of Primary Cognitive Appraisal

Lastly, hierarchical regression analyses were conducted to explore whether primary cognitive appraisal mediated the relationship between self-esteem and sibling psychological and somatic difficulties. The mediation model was not confirmed for parent reported internalizing behavior, externalizing behavior, and somatic problems as significant associations were not found at each step of the regression model outlined by Baron and Kenny (1986) (see Appendix A for regression results). However, the mediation model was partially confirmed for sibling reported anxiety and depression scores (Figures 11 and 12). At Step 1, self-esteem was

significantly associated with anxiety (ß = -4.11, S.E. ß = 1.34, $p < .01$) and depression (ß = -10.45, S.E. ß = 1.74, $p < .01$) scores. Results of Step 2 confirmed primary cognitive appraisal was significantly associated with symptoms of anxiety (ß = -.55, S.E. ß = .14, $p < .01$) and depression (ß = -.88, S.E. ß = .20, $p < .01$). At Step 3, self-esteem was significantly associated with primary cognitive appraisal (ß = 2.70, S.E. ß = .87, $p < .01$). When self-esteem and primary cognitive appraisal were entered at Step 4 to predict anxiety, the effect of self-esteem on anxiety was reduced to ß = -2.86, S.E. ß = 1.35 $p < .05$. When self-esteem and cognitive appraisal were entered at Step 4 to predict depression, the effect of self-esteem on depression was also reduced to ß = -8.82, S.E. ß = 1.75 $p < .01$. As the contribution of self-esteem to depression and anxiety remained significant, primary cognitive appraisal was confirmed to partially mediate the relationship between self-esteem and anxiety and depression scores. Post-hoc probing confirmed the significance of the partial mediation effects for sibling depression (Sobel Test = -2.18, $p = .03$) and anxiety (Sobel Test = -2.18, $p = .03$).

CHAPTER FOUR

Discussion

Aim 1: Examine Sibling Psychological and Somatic Difficulties

The initial aim of the present study was to examine some of the psychological and somatic difficulties siblings experience. Results indicate, in general, parents as a group reported normally distributed internalizing and externalizing behavior problems, suggesting siblings experience behavior patterns typical of children the same age. However, parents reported proportionally more siblings of children with cancer experience borderline and clinically significant internalizing behavior problems (30%) and somatic problems (11%), based on normative values (internalizing = 18%; somatic = 2%; Achenbach, 1991). Additionally, more siblings reported clinically significant symptoms of anxiety (15%) than expected based on normative values (7%; Spielberger, 1983). These results support previous research (Barrera et al., 2004c; Carpenter & Sahler, 1991; Cohen et al., 1994; Heffernan & Zanelli, 1997; Houtzager et al., 2005; Powazek et al. 1980; Sahler et al., 1994; Zeltzer et al., 1996) and indicate siblings who were referred for, and subsequently attended, intervention groups to address parental concerns experience higher levels of psychological and somatic problems than the normative population.

Theoretical models provide insight into the present findings. According to Lazarus & Folkman (1984) and the disability-stress-coping model (Wallander & Varni, 1992), cancer experiences result in stress when siblings view their experiences and situations as taxing their ability to cope and threatening their well-being. Specifically, siblings may perceive disrupted routines, parental absences, emotional distress, physical pain, and uncertainty as stressful and threatening. These events can result in siblings feeling stress, tension, and/or nervousness that translate into symptoms of anxiety (Hamama et al., 2000). As Wallander and Varni suggest, perceived stress may be the primary risk factor for psychological adjustment difficulties including anxiety. Although the cancer experience was not likely the sole contributor to

psychological and somatic difficulties, the cancer experience, combined with other life stressors encountered by some siblings, likely led to more reports of anxiety symptoms than population norms.

The finding that parents reported more internalizing behavior and somatic problems than expected in the normal population is consistent with previous research. Such reports may reflect parental distress and anxiety because stress and demands associated with the cancer experience could alter parental perceptions of sibling behavior (Cohen et al., 1994), resulting in high levels of parent reported internalizing and somatic problems. However, internalizing behavior and somatic problems in siblings may truly reflect sibling internal distress and anxiety (Houtzager et al., 2004). In addition, regarding somatic problems, Houtzager and colleagues suggest siblings can recognize parents readily respond to physical symptoms in children with cancer. As such, siblings may complain of physical symptoms in attempt to gain attention and care.

Parent reports suggest siblings do not experience more externalizing behaviors than the normative population. These findings conflict with the majority of more recent research that employed standardized and non-standardized questionnaires and found siblings and parents report high levels of externalizing behavior problems (Cohen et al., 1994; Heffernan & Zanelli, 1997; Sloper & While, 1996). Although parents reported siblings exhibit normative externalizing behavior problems, it is possible the stress and demands of the cancer experience are risk factors for externalizing behavior problems and/or siblings exhibit externalizing behaviors as a coping response to gain parental attention and care. However, as many parents are frequently consumed with caring for the child with cancer and absent from the home, parents may not observe, and therefore report, the behavior (Barrera et al., 2004a). Additionally, some siblings may exhibit externalizing behavior problems in environments outside the home (e.g., school) but inhibit the

behaviors at home, likely to avoid exacerbating parental distress. In turn, siblings may manifest stress associated with cancer in other ways, including internalizing and somatic problems.

To date, few researchers have reported on depressive symptomatology in siblings. Rather, researchers frequently include, and therefore assess, symptoms of depression with general internalizing behavior problem measures such as the CBCL (Achenbach, 1991). Consequently, depressive symptoms are rarely differentiated from other internalizing difficulties and reported. Given limited knowledge pertaining to depressive symptomatology in siblings, the finding that very few siblings reported clinical levels of depressive symptoms and 30% of parents reported siblings exhibited clinically significant internalizing behavior problems is significant. The current findings suggest the stressors and challenges associated with cancer and cancer treatment are not risk factors for depressive symptomatology in siblings.

In summary, results of the present study indicate siblings are not equally affected by the cancer experience. Whereas the majority of siblings adjusted well and experienced no major psychopathology, some siblings reported clinically significant symptoms of anxiety and were reported by parents to have clinically significant internalizing behavior and somatic problems. Thus, the cancer experience appears to be a risk factor for self reported symptoms of anxiety and parent reported internalizing and somatic difficulties. By examining the sibling, family, and disease factors significantly related to psychological and somatic outcomes, the study provides some insight into why sibling adjustment varies and the mechanisms by which the cancer experience affects sibling adjustment.

Aim 2: Examine Direct Effects of Sibling, Family, and Disease Factors on Sibling Psychological and Somatic Difficulties

Guided by the disability-stress-coping model (Wallander & Varni, 1992), the second aim of the study was to examine the direct effect of sibling (i.e., age, gender, self-esteem, primary

cognitive appraisal), family (i.e., SES, social support), and disease (i.e., diagnosis, days since diagnosis) factors on sibling adjustment. Several factors, described below and presented in Figure 5, were significantly associated with sibling psychological and somatic difficulties. Moreover, there were small to large effect sizes between predictor variables (e.g., age, brain tumor diagnosis, self-esteem, social support, and primary cognitive appraisal) and the adjustment measures, with up to 42% of the variance in adjustment scores accounted for by the predictor variables. Thus, there were meaningful and practical relationships between the predictor and outcome variables, particularly between self-esteem, social support, and cognitive appraisal and sibling adjustment.

SES and Days Since Diagnosis

Socioeconomic status and days since diagnosis were hypothesized to predict sibling adjustment, with siblings whose mothers had more education experiencing fewer adjustment problems and fewer days since diagnosis associated with more psychological and somatic problems. These hypotheses were largely unsupported. Regression analyses indicated family SES and days since diagnosis were not associated with sibling and parent reported psychological and somatic problems, suggesting siblings who were from families with higher SES were not more likely to experience fewer psychological and somatic problems. Moreover, siblings whose brothers or sisters were diagnosed more recently were not more likely to experience psychological and somatic problems. These results are partially inconsistent with Wallander and Varni's (1992) disability-stress-coping model and some previous reports (e.g., Bendor, 1990; Cohen et al., 1994; Gogan & Slavin, 1981; Hamama et al., 2000; Houtzager et al., 2004; Sahler et al., 1994; Sargent et al., 1995; Schuler et al., 1985; Sloper & While, 1996).

A lack of direct association between family SES and sibling outcomes can be explained, in part, by the sensitivity of the SES measure, maternal education. Parental education is

frequently used as a measure of SES in health research as it is accepted to be a multidimensional construct comprised of diverse socioeconomic factors that may directly and/or indirectly influence individual outcomes (Braveman et al., 2005). However, the cancer experience frequently leads to considerable financial and social demands. For example, mothers frequently reduce or terminate paid employment to care for the child with cancer, thus placing financial demands on the family and reducing the contact she has with the child's siblings. Moreover, many families are forced to pay for prescriptions, travel, and accommodations, thereby leaving them financially unstable and emotionally stressed. Maternal education, therefore, may not accurately reflect current economic and social circumstances of families living with cancer. Thus, maternal education as a SES measure may have been unable to detect the potential influences of SES on sibling adjustment.

A lack of association between days since diagnosis and psychological and somatic difficulties may reflect a population of siblings who was referred for intervention over psychological and behavioral concerns. Although the siblings had lived with pediatric cancer for varying lengths of time and some research suggests adjustment difficulties decrease with increased time since diagnosis (Cohen et al., 1994; Hamam et al., 2000; Houtzager et al., 2004), it is possible most sibling participants were referred for intervention because they were struggling to cope and were exhibiting psychological and somatic difficulties that parents found concerning. As siblings and parents likely identified such adjustment difficulties on the current study's outcome measures, sibling psychological and somatic difficulties were not associated with increased time since diagnosis. Moreover, the results support some previous findings (e.g., Alderfer et al., 2003; Barrera & Atenafu, 2008; Lown et al., 2008) and suggest siblings may experience some psychological difficulties many months and years after diagnosis. Such

difficulties may reflect continued stressors and uncertainties related to extended treatments, cancer relapse, late or long-term side-effects, and so on.

Diagnosis

Diagnosis of the child with cancer was hypothesized to be associated with sibling outcomes. In particular, diagnoses such as brain tumors that may have more devastating effects on the child with cancer were expected to be associated with more psychological and somatic problems in siblings. Results of the present study indicate brain tumor diagnoses were associated with sibling reported symptoms of anxiety, with a moderate effect size. Siblings whose brothers or sisters were diagnosed with a brain tumor reported more anxiety symptoms than siblings whose brothers or sisters were diagnosed with other tumors, leukemia, lymphoma, and other blood disorders. Thus, brain tumor diagnoses in the child with cancer also appear to impact siblings and may be considered a risk factor for sibling reported symptoms of anxiety. Although severity and prognosis cannot be fully deduced from diagnosis, siblings whose brothers or sisters are diagnosed with brain tumors are required to cope with substantial uncertainty about the survival of their brothers or sisters, pervasive late-effects, and possible behavior changes (Houtzager, Grootenhuis, & Last, 2001). Moreover, siblings witness emotional suffering in their parents. These experiences appear to be related to more sibling reported symptoms of anxiety.

Age

Age was predicted to influence sibling adjustment, with increased age associated with fewer externalizing behavior and somatic problems. This hypothesis was not supported. However, siblings 13 to 17 years of age were reported to experience fewer internalizing behavior problems than siblings 7 to 12 years. This finding supports research that utilized interviews and standardized questionnaires completed by siblings (e.g., Hamama et al., 2000; Schuler et al., 1985). Whereas the frequency of parent and sibling reported internalizing behavior problems

such as symptoms of anxiety and depression, anger, sadness, worry, and withdrawal have been previously documented (Carpenter & Sahler, 1991; Chesler et al., 1992; Cohen et al., 1994; Gogan & Slavin, 1981; Martinson et al., 1990; Schuler et al., 1985; Williams et al., 2009), few researchers have found age differences using parent reported standardized measures.

Older siblings may be buffered from the negative effects of the cancer experience because they have the cognitive maturity to accurately understand the illness and treatment, more developed emotional and social skills to respond to and cope with stress, and more independence that results in enhanced support from activities and individuals including peers outside the home (Hamama et al., 2000; Sargent et al., 1995). Additionally, parents may have less understanding of the emotional states of adolescents because adolescents may reach out to peers for support rather than share concerns with parents. In contrast, younger siblings may be more dependent on the family for support and therefore, are more affected by the absence of parents and other family members (Barbarin et al., 1995; Sargent et al., 1995). The current results suggest stressors and demands associated with the cancer experience lead to more internalizing problems in the younger sibling population.

Gender

Gender was hypothesized to be associated with psychological and somatic problems, with male siblings less likely to experience somatic problems and internalizing behavior problems, including symptoms of anxiety and depression, than female siblings. This hypothesis was partially supported as gender was associated with parent reported somatic problems, with males experiencing fewer somatic problems than females. These results provide new insight into the adjustment of male and female siblings. Compared to male siblings, female siblings may have more household responsibilities, be more involved with the illness, and experience more restrictions and disruptions in their daily lives (Houtzager et al., 2004). As female siblings may

seek and rely more on social support (Thompson & Gustafson, 1996), these experiences may lead to more stress and, in part, account for more somatic complaints in female siblings. However, as Houtzager and colleagues (2004) suggest, siblings may recognize physical symptoms in the child with cancer gain the attention of parents. Female siblings, therefore, may be more likely to try and secure parental attention and care through somatic complaints.

In contrast, gender was not associated with sibling reported symptoms of anxiety and depression and parent reported internalizing behavior problems. These findings contradict previous research (e.g., Alderfer et al., 2003; Barrera et al., 2004c; Hamama et al., 2000) and suggest females do not experience more symptoms of anxiety and depression and parent reported internalizing problems than male siblings. Although these results may be an accurate reflection of sibling adjustment, it is possible female siblings exhibited general internalizing problems but parents may not have been attuned to, or present to observe, such problems.

Social Support

Sibling social support was predicted to be associated with sibling psychological and somatic difficulties. Social support was associated with each sibling outcome, with more perceived social support associated with fewer sibling reported symptoms of anxiety and depression. Higher perceived social support was also associated with fewer parent reported internalizing and externalizing behavior problems and somatic problems. Furthermore, social support, in conjunction with self-esteem and cognitive appraisal, had small to large effect sizes when predicting sibling adjustment, indicating some of the results must be considered with caution. Nonetheless, these findings support previous research (Barrera et al., 2004c; Carpenter & Sahler, 1991; Varni & Katz, 1997; Varni et al., 1994; Williams et al., 2002) and suggest perceived social support is a protective factor against psychological adjustment difficulties and somatic problems, regardless of the informant. These results also parallel those from research

conducted with adolescents in the normal population, with enhanced social support associated with fewer psychological difficulties including fewer symptoms of depression (Kaltiala-Heino, Rimpela, Rantanen, & Laippala, 2001; Newman, Newman, Griffen, O'Connor, & Spas, 2007).

Bisschop, Kriegsman, Beekman, and Deeg (2004) report individuals with chronic illnesses such as cancer have experiences that result in unique needs and expectations of their social network. These researchers postulate when such individuals receive social support from family and friends, their psychological well-being is enhanced as the support satisfies a need to feel consistency and belonging. However, Bisschop and colleagues also suggest individuals with chronic illnesses encounter stressors, demands, and experiences that can limit their ability to utilize or maintain social networks (Bisschop et al., 2004). Siblings, therefore, may experience stress and demands that lead to increased need for social support, but may be limited in their ability to access social support as parents may be physically and/or emotionally unavailable. Furthermore, changes to school and daily life routines can reduce the contact siblings have with friends and change the contact they have with family members. Siblings, therefore, may be protected against emotional distress if they not only maintain social connections, but believe their social networks are accessible and supportive. That is, if siblings perceive and receive high social support, they may believe they have the resources and ability to cope with the cancer experience. In turn, they experience less stress and psychological and somatic difficulties.

Primary Cognitive Appraisal

Primary cognitive appraisal was hypothesized to be associated with sibling and parent reported outcomes, with positive appraisal predicting fewer sibling and parent reported psychological and somatic difficulties. Results indicated positive primary cognitive appraisal was predictive of fewer sibling reported symptoms of anxiety and depression. Moreover, cognitive appraisal, with self-esteem and/or social support, had medium to large effect sizes

when predicting these psychological problems. Thus, positive primary cognitive appraisal appears to be a protective factor against symptoms of anxiety and depression. This finding is significant as cognitive appraisal is suggested to be a key factor in coping with chronic illnesses such as cancer (Franks & Roesch, 2006; Folkman, Lazarus, Dunkel-Schetter, DeLongis, & Gruen, 1986b; Hamama-Raz & Solomon, 2006; Jenkins & Pargament, 1988; Juth et al., 2008; Lazarus & Folkman, 1984; Samson & Siam, 2008; Wallander & Varni, 1992; Wallander & Varni, 1998). Moreover, some research has examined the impact of cognitive appraisal on children and adults living with chronic illnesses and found positive cognitive appraisal is related to coping and associated with better psychological adjustment and less emotional distress (Folkman et al., 1986b; Hamama-Raz & Solomon, 2006; Ireys, Gross, Werthamer-Larsson & Kolodner, 1994; Jenkins & Pargament, 1988). The impact of cognitive appraisal on sibling psychological adjustment, however, has rarely been investigated.

The cognitive appraisal measure examined primary cognitive appraisal and how siblings interpreted and evaluated the cancer experience and the extent to which they perceived it as a threat (Franks & Roesch, 2006; Juth et al., 2008; Samson & Siam, 2008). Primary cognitive appraisal is viewed as central to adjustment as it precedes, and therefore influences, later processes including secondary cognitive appraisal (i.e., the evaluation of one's coping resources and available options to manage stressful encounters) and finally coping (i.e., an individual's cognitive and/or behavioral efforts to manage the demands of stress and experiences that are assessed as taxing or exceeding his/her resources) (Folkman et al., 1986b; Juth et al., 2008; Samson & Siam, 2008). Thus, the present findings suggest when siblings have positive primary cognitive appraisals and perceive the cancer experience as non-threatening, they experience less stress and therefore, fewer symptoms of anxiety and depression than siblings with negative cognitive appraisals. Contrarily, siblings with negative primary cognitive appraisals appear more

likely to view the cancer experience as a threat to their well-being and be at increased risk for symptoms of anxiety and depression. Although primary cognitive appraisal appears predictive of sibling psychological and somatic difficulties, the present study cannot account for the relationship between primary cognitive appraisals, secondary cognitive appraisals, coping strategies, and sibling psychological and somatic outcomes. Nonetheless, the study indicates primary cognitive appraisal plays an important role in sibling psychological adjustment. These findings have research and clinical implications that are explored below.

Self-esteem

Lastly, self-esteem was hypothesized to be associated with sibling psychological adjustment, with higher self-esteem associated with fewer sibling reported symptoms of anxiety and depression and parent reported internalizing and externalizing behavior problems and somatic complaints. This hypothesis was partially supported as self-esteem was predictive of symptoms of depression, with siblings reporting high self-esteem less likely to experience symptoms of depression. Moreover, self-esteem, when combined with social support and cognitive appraisal, had a large effect on the association with symptoms of depression. Although there is little empirical research into the influence of self-esteem on the adjustment of siblings of children with cancer, some have found self-esteem is related to psychological adjustment in individuals with and without chronic illnesses (see Bisschop et al., 2004; Harter, 1989; Ireys et al., 1994). Thus, the present finding that high self-esteem may serve a protective function against sibling symptoms of depression supports previous research and provides significant and novel knowledge to the area of sibling adjustment.

Self-esteem is the self-perception of one's skills, abilities, and personal qualities that guide and motivate cognitive processes and behaviors (Juth et al., 2008) such as coping behaviors. Therefore, high self-esteem is the positive evaluation of one's skills, abilities, and

characteristics that serve to assist him/her in coping with uncertainties and protect him/her against psychological difficulties. Thus, as suggested by Schneiderman, Ironson, and Siegel (2005), siblings with high self-esteem may have positive personal perceptions and evaluations that serve as coping resources to manage stressors such as those encountered during the cancer experience. Moreover, siblings with high self-esteem may perceive the cancer experience as non-threatening to their well-being. In turn, high self-esteem may minimize or prevent stress and protect siblings from symptoms of depression.

In describing the relationship between low self-esteem and psychological outcomes in individuals with chronic illness, Juth and colleagues (2008) propose low self-esteem may foster a continuous cycle of negative, and occasionally, depressed affect that impacts numerous areas of their functioning including appraisals of the illness and self-care. Consequently, siblings who reported symptoms of depression may have viewed the cancer experience as a threat to their well-being and believed they lack the resources and ability to cope with the challenges and demands of the cancer experience.

Aim 3: Examine Moderating Effects of Age

To further elucidate the manner by which the cancer experience influences sibling adjustment and the factors to be targeted through intervention support, the study examined whether sibling psychological and somatic difficulties were moderated by age. That is, the study sought to investigate whether there were specific ages (i.e., younger versus older) under which gender, level of primary cognitive appraisal, and level of social support exerted an effect on sibling psychological adjustment. Although significant moderations were found, the effect sizes were small. Thus, the results must be considered with caution.

Age x Gender

Age was predicted to moderate the relationship between gender and sibling psychological and somatic problems, with gender differentiating sibling outcomes when siblings were older but not younger. This hypothesis was partially confirmed; female siblings reported significantly more symptoms of depression than male siblings when they were older, but not younger (Figure 6). These results parallel those of Sahler and colleagues (1994), as well as those from normative populations (Hankin & Abramson, 1999; Nolen-Hoeksema, 2001; Piccinelli & Wilkinson, 2000). Adolescence is a particularly vulnerable time for female siblings (Bearman Miller & La Greca, 2005) and the present findings suggest older female siblings, as compared to male and younger female siblings, may be at increased risk for symptoms of depression. Eccles, Barber, Jozefowicz, Malenchuk, and Vida (1999) report adolescence is a period of significant life and physical changes and as females experience these changes concurrently and males experience them sequentially, adolescent females can be predisposed to more life stress than adolescent males. Because developmental changes and stressors require adolescent females to utilize coping resources (Eccles et al., 1999), resources for managing demands and stressors associated with the cancer experience may be limited and adolescent female siblings may lack the resources required to cope and adjust. Furthermore, older female siblings may be increasingly restricted from engaging in routine and social activities as they assume more household and caregiving responsibilities (Houtzager et al., 2004; Sahler et al., 2004). Thus, developmental changes, increased responsibilities, reduced social contact, and routine changes can place significant stressors and demands on older female siblings that tax their coping resources and lead to adjustment difficulties, including symptoms of depression (Bearman Miller & La Greca, 2005; Houtzager et al., 2004).

Age x Social Support

Age was also predicted to moderate the relationship between social support and sibling outcomes. Current results indicate social support influenced sibling reported symptoms of depression when siblings were younger but not older (Figure 7). That is, younger siblings who reported high social support reported fewer symptoms of depression than younger siblings with low social support. Symptoms of depression reported by older siblings did not vary significantly with the level of social support. Thus, high perceived social support appears to be more protective against symptoms of depression for younger siblings, as compared to older siblings.

Patistea and colleagues (2000) indicate there is considerable risk for communication changes between parents and siblings when children are diagnosed with cancer. Furthermore, although younger siblings, as compared to older siblings, may not be as threatened and affected by cancer diagnoses and treatment because they are less able to understand complex situations related to the disease and treatment (Barbarin et al., 1995; Hamama et al., 2000), they appear more affected by the absence of parents and other family members (Barbarin et al., 1995). Thus, when younger siblings believe individuals such as their parents provide attention and care and, in turn, feel highly supported, they may perceive fewer threats and experience fewer psychological problems. In contrast, older siblings can more fully understand the implications of the cancer diagnosis, prognosis, and treatment on the family and child with cancer (Gogan & Slavin, 1981; Murray, 2000). Moreover, hospital routines and increased caregiving responsibilities can lead to more disruption in the home and social lives of older siblings. As the cancer experience may present older siblings with greater responsibilities, challenges, and disruptions than younger siblings, high perceived social support may not provide older siblings with sufficient resources to manage the stressors they encounter and protect themselves against psychological problems. Thus, perceptions of social support do not appear to influence the psychological adjustment of

older siblings. These results have important implications for intervention support, particularly for younger siblings.

Age x Cognitive Appraisal

Lastly, primary cognitive appraisal was predicted to moderate the relationship between age and sibling psychological and somatic difficulties. Analyses conducted to evaluate the nature of significant regression analyses revealed age moderated the relationship between primary cognitive appraisal and both symptoms of depression and somatic problems. More specifically, primary cognitive appraisal influenced somatic problems for younger but not older siblings, with younger siblings with positive appraisals less likely to experience somatic problems than younger siblings with negative appraisals (Figure 10). Primary cognitive appraisal did not appear to influence the somatic problems of older siblings. Additionally, although primary cognitive appraisal influenced symptoms of depression for both younger and older siblings, it appeared to have more influence on younger siblings (Figure 8). These results indicate positive primary cognitive appraisal may serve more of a protective function against psychological difficulties for younger siblings. That is, when younger siblings more accurately understand the implications of cancer and view it as an experience that is not a threat to their well-being, they may be better protected against psychological problems. Moreover, although younger siblings may typically lack the cognitive and affective capacity to accurately understand such changes (Bendor, 1990), when siblings have positive primary cognitive appraisals, they may acquire knowledge and understanding that supports them in minimizing stress and preventing psychological difficulties.

Summary

In summary, current results suggest age moderated the relationship between the following predictor and outcome variables: sibling gender and symptoms of depression; perceived social support and symptoms of depression; and primary cognitive appraisal and symptoms of

depression and somatic problems. These findings highlight the prominent role of age, and therefore development, on sibling psychological adjustment. Particularly, high perceived social support and positive cognitive appraisals may provide younger siblings with resources and understanding that reduces the stress they perceive and the psychological difficulties they experience. The moderating influence of sibling age, and thus developmental level, on sibling psychological adjustment has important implications for the development of sibling intervention groups.

Aim 4: Examine Mediating Effects of Primary Cognitive Appraisal

Wallander and Varni's (1992) disability-stress-coping model claims factors such as cognitive appraisal directly and indirectly influence the psychological adjustment of siblings. Although results of the current study support previous findings (Sloper & While, 1996) and suggest primary cognitive appraisal predicts sibling reported symptoms of anxiety and depression, no researchers have investigated whether cognitive appraisal also mediates sibling psychological adjustment. The current study, therefore, sought to examine whether cognitive appraisal mediated the relationship between self-esteem and sibling adjustment. Specifically, the study investigated whether cognitive appraisal accounted for the relationship between self-esteem and sibling adjustment. Results of the investigation suggest primary cognitive appraisal partially accounted for, and thus partially mediated, the relation between self-esteem and symptoms of anxiety and depression. Thus, although self-esteem appears to play an important role in sibling adjustment, with siblings with high self-esteem less likely to experience symptoms of anxiety and depression, cognitive appraisal appears to be a factor or mechanism through which self-esteem influences sibling adjustment. That is, siblings with high self-esteem appear to develop positive cognitive appraisals and it is these positive evaluations that protect siblings against symptoms of anxiety and depression.

Self-esteem encompasses the attitudes, feelings, and/or evaluations one has about his/her abilities, skills, and overall qualities (Greenwald, Bellezza, & Banaji, 1988; Juth et al., 2008). Further, such self-perceptions may extend to other life domains by influencing the attitudes, thoughts, and beliefs one has about an illness and therefore, his/her appraisal processes and coping resources (Bisschop et al., 2004; Juth et al., 2008). As proposed by Juth and colleagues about individuals with chronic illnesses, siblings with low self-esteem may develop a continuous cycle of negative affect, views, and beliefs that leads them to negatively appraise cancer and perceive it as having a negative impact. Siblings with low self-esteem, therefore, may view cancer as a threatening and stressful experience. These negative perceptions, defined as negative primary cognitive appraisals, may, in part, be the source of psychological difficulties and partially explain the association between self-esteem and poor psychological adjustment. In contrast, as siblings with high self-esteem likely view their abilities, situations, and experiences in a more positive and hopeful manner, they may perceive the cancer experience as less threatening. Positive perceptions about the cancer experience, and thus positive primary cognitive appraisal, may protect siblings from experiencing psychological difficulties such as symptoms of anxiety and depression. Specifically, siblings with high self-esteem likely assess themselves positively and these beliefs and evaluations lend to more optimistic thoughts and beliefs about the illness (Bisschop et al., 2004), possibly resulting in less stress and psychological difficulties. As self-esteem is established to include beliefs about ones abilities, sibling self-esteem may also be associated with secondary cognitive appraisal and the perception of whether one has the coping resources and abilities to cope with the cancer experience and prevent adjustment difficulties (Juth et al., 2008). Although investigating the relationship between self-esteem, secondary cognitive appraisal, and sibling outcomes was beyond the scope

of the present investigation, the potential relationship provides important direction for future research and clinical intervention.

Clinical and Theoretical Implications

Guided by the disability-stress-coping model (Wallander & Varni, 1992), the study examined the present-day psychological adjustment of siblings to pediatric cancer and the influence of sibling, family, and disease factors on sibling psychological and somatic difficulties to guide the development and implementation of intervention groups and validate aspects of Wallander and Varni's model. Results indicate although most siblings adjust well and do not experience major psychological disturbance and adjustment difficulties, a small population of siblings experience more symptoms of anxiety, internalizing behavior problems, and somatic complaints than the normative population and may benefit from intervention support. Additionally, results support aspects of Wallander and Varni's model. Most notably, perceived social support, self-esteem, and cognitive appraisal appear to significantly influence sibling psychological and somatic difficulties, with small to large effect sizes. Moreover, age or developmental stage moderates the protective influence of gender, social support, and cognitive appraisal on sibling adjustment and primary cognitive appraisal partially mediates the relationship between self-esteem and sibling symptoms of anxiety and depression. The study also found age, gender, and diagnosis influence some psychological outcomes. These results demonstrate the utility of Wallander and Varni's model in guiding adjustment research and they have important implications for the development and implementation of sibling intervention groups. However, as the direct and indirect associations between the predictors and adjustment outcomes had primarily small effect sizes, there are factors and associations related to psychological and somatic outcomes that are unaccounted for.

Sibling anxiety is purported to stem, in part, from uncertainties about treatment and prognosis (Houtzager et al., 1999; McGrath, 2001; Patistea et al., 2000) and thoughts and feelings associated with limited or inaccurate knowledge about cancer. For example, siblings may experience anxiety because they appraise cancer as a threat (Hamama et al., 2000), worry about getting cancer (Gogan & Slavin, 1981), and feel guilty and angry (Houtzager et al., 2001). Thus, to reduce sibling anxiety, interventions must target sibling knowledge of the disease, including etiology and treatment (Bendor, 1990; Gogan & Slavin, 1981; Houtzager et al., 2001). In fact, research indicates when sibling knowledge and understanding is enhanced, siblings experience fewer psychological difficulties including anxiety, fear, and behavioral problems (Barrera et al., 2004a; Carpenter, Sahler, & Davis, 1990; Dolgin, Somer, Zaidel, & Zaizov, 1997; Evans, Stevens, Cushway, & Houghton, 1992; Houtzager et al., 2001; Lobato & Kao, 2002; Roeyers & Mycke, 1995; Sahler & Carpenter, 1989; Sidhu et al., 2006; Williams et al., 2003). Educating siblings on various aspects of cancer, cancer treatment, and prognosis not only clarifies misunderstandings, but supports siblings in understanding changes in family relations and emotional reactions of family members (Barera et al., 2004a; Houtzager et al., 2004). Houtzager and colleagues (2004) suggest by enhancing their knowledge about cancer, siblings may experience enhanced feelings of control and feel more secure and thus, less anxious. Education, therefore, provides a foundation for any psychological intervention addressing anxiety and somatic complaints and will help siblings minimize anxiety and remain optimistic and hopeful (Houtzager et al., 2004; Lobato & Kao, 2002).

Results of the present study imply siblings with high social support, high self-esteem, and/or positive primary cognitive appraisals experience fewer psychological and somatic difficulties, particularly when younger. Given the particularly prominent role of social support in protecting siblings from adjustment problems, optimizing perceptions of social support must be a

primary aim of intervention groups. Enhanced perceptions of social support can be attained in numerous manners. Firstly, group treatment provides a safe environment where siblings can build nurturing friendships and process, share, and address thoughts and feelings pertaining to the cancer experience, and therefore, support one another (Barrera et al., 2004a). Furthermore, by developing meaningful relationships with similar peers, siblings can sense they belong to a cohesive group that offers social support to its members. Houtzager and colleagues (2001) posit by sharing experiences and emotions, sibling anxiety can be reduced. Furthermore, throughout treatment, siblings can be encouraged to explore the importance of feeling supported, who they can turn to for support, and actions they can take to acquire the support they require. Although activities such as role plays and brainstorming may enhance sibling understanding of the need for support and how to secure it, some siblings will require specific support in exploring factors that deter or prevent them from seeking support. As current results suggest younger siblings with high perceived social support are more likely to experience fewer psychological difficulties than younger siblings with low perceived social support, it is especially important group interventions strive to enhance social support in younger siblings.

Researchers (e.g., Barrera et al., 2004a; Cassidy, 2000; Evans et al., 1992; Gold et al., 2008; Hamama et al., 2000) suggest intervention groups must focus on developing coping skills. That is, siblings should be encouraged to explore strategies that will support them in managing and thus alleviating stress and uncertainty. In fact, Barrera and colleagues found siblings who attended group intervention focusing, in part, on coping with issues pertaining to cancer and family and peer relations, had reduced symptoms of anxiety and depression following intervention. Researchers, however, have yet to examine coping strategies (e.g., emotion-focused and problem-focused coping strategies) that might serve to effectively reduce sibling stress and anxiety and thus, psychological and somatic difficulties. Although a discussion pertaining to the

focus of coping strategies in intervention groups is a next step of sibling psychological adjustment research, current results highlight the importance of the initial coping phase to adjustment: primary cognitive appraisal. For siblings, primary cognitive appraisal is the evaluation of whether the cancer experience threatens their well-being. Siblings with positive primary cognitive appraisals view their experiences as less stressful and threatening than siblings with negative primary cognitive appraisals. Thus, educating siblings about cancer may provide siblings with accurate knowledge that leads them to view the cancer experience as less threatening. Additionally, education may enhance siblings' sense of control (Houtzager et al., 2001) and lead siblings to have positive secondary appraisals and believe they have the resources and abilities to manage cancer-related stressors and demands. Consequently, enhancing sibling knowledge and social support through group intervention may serve a protective function against adjustment difficulties by targeting the development of positive primary and secondary cognitive appraisals.

As high self-esteem is associated with positive cognitive appraisals, interventions should also strive to enhance self-esteem in siblings. By developing nurturing friendships and group coherence and providing siblings with opportunities to process and share how cancer affected their lives and explore associated thoughts and feelings, sibling experiences can be normalized. Thus, although enhancing self-esteem cannot be the primary focus of intervention groups, safe environments in which siblings develop meaningful and supportive friendships and share similar experiences may enhance feelings of normalcy, acceptance, and thus self-esteem. In turn, siblings may develop positive cognitive appraisals and be protected against psychological problems.

Houtzager and colleagues (2004) note researchers and clinicians require knowledge about who the at-risk siblings are and how to identify them. Although the current study found some

younger siblings, female siblings, and siblings with low perceived social support, low self-esteem, and negative primary cognitive appraisals may experience more psychological and somatic difficulties, these factors are very difficult for medical and support staff to perceive and measure. Furthermore, although current findings suggest factors such as age, gender, and diagnosis influence sibling adjustment, the results conflict with some previous results. However, as current and previous findings suggest some siblings may have enduring concerns and distress and can experience psychological difficulties months and years after diagnosis (Alderfer et al., 2003; Barrera & Attenafu, 2008; Houtzager et al., 2004; Lown et al., 2008; Van Dongen-Melman et al., 1995), it is important that psychological and somatic symptoms be addressed as soon as they arise. Thus, to optimize the likelihood siblings experiencing psychological difficulties are referred for intervention, medical and support staff may find it most advantageous to conduct initial screenings and interview parents on sibling behavior problems that may be indicative of adjustment difficulties (e.g., acting out, withdrawal, changes in academic performance, etc.). Secondly, staff may find it useful to educate parents and family members. In fact, researchers (e.g., Bendor, 1990; Gold et al., 2008; Hamama et al., 2000; Houtzager et al., 2005) and clinicians advocate family-centered interventions. For families living with pediatric cancer, such interventions support each family member and focus on family coping, social support, expressiveness, and decreased family conflict through individual, family, and/or group therapy (Gold et al., 2008).

Many researchers (e.g., Bendor, 1990; Evans et al., 1992; Gold et al., 2008; Hamama et al., 2000; Houtzager et al., 2005) highlight the importance of including parents in intervention work. Parent education and support may be particularly important for younger siblings as current findings suggest enhanced social support and positive cognitive appraisals may protect younger siblings against psychological difficulties. Distress in younger siblings is reported to be

associated with feeling less loved and important (Bendor, 1990) and guilty and/or responsible for the cancer (Murray, 2000). Additionally, younger siblings seem to be more affected by absent parents (Barbarin et al., 1995) and less knowledgeable about the disease and future as parents can limit the details they provide (Gogan & Slavin, 1981). By including parents in treatment, clinicians are able to provide parents with support and direction in parenting both the sibling(s) and the child with cancer. Specifically, parents can be encouraged to provide siblings with factual knowledge pertaining to diagnosis, treatment, and prognosis in a developmentally appropriate manner (Bendor, 1990; Hamama et al., 2000; Houtzager et al., 2005). Moreover, parents may benefit from strategies to reduce conflict and enhance emotional support for their children through increased conversations about thoughts and feelings related to cancer. By enhancing communication, support, and knowledge about cancer, parents can support siblings in developing positive cognitive appraisals and decrease negative emotional responses (Hamama et al., 2000). Lastly, parents can be supported in understanding the stressors and demands each family member encounters and the associated risks (e.g., symptoms of anxiety). Although parents can be encouraged to adopt parenting practices and strategies that support each child and minimize adjustment difficulties, parents should be educated about sibling intervention and when treatment should be pursued. Such practices will likely enhance sibling adjustment and/or the likelihood that siblings struggling to manage the stressors and demands of the cancer experience are referred for intervention support.

Limitations and Directions for Future Research

Despite numerous strengths, there are limitations to this study. As the participants were siblings whose brothers or sisters were treated in one hospital in a large metropolitan city, the results may not represent the adjustment of siblings from areas and hospitals with different medical and psychological care. Additionally, the siblings were referred for an intervention study

based on parental concerns of emotional and behavioral difficulties. Thus, the study may overestimate the number of siblings who experience symptoms of anxiety, internalizing behavior problems, and somatic complaints. The results, therefore, may not generalize to the broader population of siblings of children with cancer. However, as participation in intervention groups is affected by family resources (e.g., transportation, financial resources), sibling characteristics (e.g., willingness to attend), and parental factors (e.g., distress, attunement of parent to sibling), the current population may not represent siblings with more adjustment difficulties, but rather, a population of siblings who was identified as needing support and able to commit to the requirements of attending eight intervention sessions.

There are also limitations related to the study's informants and instruments. Firstly, parent and sibling reports may not fully and accurately reflect the psychological and somatic difficulties siblings experienced. Whereas siblings may deny or fail to perceive and report negative effects (Barrera et al., 2004a; Fife et al., 1987; Sharpe & Rossiter, 2002), parents may over report behavior problems because stress associated with the cancer experience alters their mood and perceptions and makes sibling behavior more burdensome (Cohen et al., 1994). Alternatively, parents may not accurately report psychological difficulties because they focus on caring for the child with cancer (Barrera et al., 2004a; Houtzager et al., 1999), they use denial as a coping mechanism (Fife et al., 1987), and/or they only observe siblings within home and family settings (Taylor et al., 2001). Thus, siblings may experience more psychological difficulties than reported.

Secondly, although multiple informants were utilized (i.e., siblings and parents), the results were somewhat inconsistent (e.g., predictors of parent reported internalizing behavior problems and sibling reported symptoms of anxiety). These discrepancies likely relate to the design and sensitivity of the measures that assessed symptoms of anxiety, symptoms of

depression, and internalizing behavior problems. The STAIC (Spielberger, 1983) and CDI (Kovacs, 1992) are self-report measures that assess internal states and symptoms of anxiety and depression, respectively, in children and adolescents. As the internalizing behavior problems subscale of the CBCL (Achenbach, 1991) assesses symptoms, affect, and behaviors pertaining to a host of psychological difficulties including anxiety and depression, this measure was likely unable to identify and discriminate between psychological problems. That is, as the internalizing behavior subscale of the CBCL includes an array of parent reported symptoms and information, this measure may lack the construct and discriminate validity required to detect specific psychopathologies (Aschenbrand, Angelosante, & Kendall, 2005; Jensen et al., 1996). As such, results pertaining to sibling reported symptoms of anxiety and depression and parent reported internalizing behavior problems were somewhat inconsistent. However, a comparison of the relationships between parent reported internalizing behavior problems and sibling reported symptoms of depression and anxiety revealed moderate correlations (.24, $p < .05$ for symptoms of depression, .22, $p < .05$ for symptoms of anxiety), indicating parents may be reliable reporters of sibling emotional states.

Thirdly, sibling psychological and somatic problems were assessed with instruments designed to diagnosis major psychological disturbances. As the measures may have had limited sensitivity in detecting mild adjustment difficulties, siblings may present with concerns and distress that result in altered psychological functioning or decreased quality of life (Houtzager et al., 2004; Van Dongen-Melman et al., 1995). These limitations suggest researchers utilize measures that identify discrete changes in functioning and are specific to the experiences of siblings whose brothers or sisters have cancer or other chronic illnesses. For example, research can utilize quality of life measures such as the PedsQL (Varni, Seid, & Rode, 1999), a measure designed to assess health-related quality of life in children and adolescents with acute and

chronic health conditions. Although designed to assess how children and adolescents diagnosed with chronic diseases perceive their illnesses to affect their social, physical, and mental functioning, the PedsQL (Varni et al., 1999) may prove to be effective in detecting significant difficulties faced by siblings. Moreover, to further address the aforementioned limitations, research must continue to investigate the psychological adjustment of siblings using multiple informants including siblings, mothers, fathers, healthcare providers, teachers, and so on.

Fourthly, the use of the SPQ (Carpenter & Sahler, 1991) to assess perceptions of social support and primary cognitive appraisal may also be considered a limitation to this study. Despite adequate psychometric properties, the social support subscale may not fully assess and account for the efficacy and accessibility of the multiple facets of social support. Additionally, this was the first known study to assess primary cognitive appraisal with the intrapersonal thoughts and feelings subscale of the SPQ (Carpenter & Sahler, 1991). Although the items of this subscale appear to assess primary cognitive appraisal and how siblings perceive the impact of the cancer experience on their well-being, the content validity of the scale and its ability to accurately measure the numerous aspects of primary cognitive appraisal is not well established. Moreover, as this subscale assesses, in part, sibling feelings (e.g., "I feel mad about my brother/sister's illness," "I worry about my brother/sister's illness," "My brother/sister's illness makes me sad"), the subscale appears to assess constructs and factors also examined by dependent measures (e.g., STAIC, Spielberger, 1983) and therefore, sibling outcomes. Consequently, the intrapersonal thoughts and feelings subscale may be used to both identify a construct (i.e., primary cognitive appraisal) that predicts sibling adjustment and serve as an outcome factor that evaluates the impact of the cancer experience on siblings. Thus, findings pertaining to sibling social support and primary cognitive appraisal may not fully reflect the extent to which social support and primary cognitive appraisal serve protective functions against

psychological and somatic difficulties. Researchers, therefore, must continue to evaluate the validity and reliability of the SPQ (Carpenter & Sahler, 1991) as a unique measure of the effects of cancer on siblings. In particular, an evaluation of the construct validity of the communication subscale, as a measure of social support for siblings, using measures including Harter's (1985b) Social Support Scale for Children and the intrapersonal thoughts and feelings subscale, as a measure of primary cognitive appraisal, using measures such as Kessler's (1998) Cognitive Appraisal of Health Scale, is warranted. Both the Harter and Kessler scales have well documented psychometric properties.

The present study examined the associations between primary cognitive appraisal and sibling adjustment. Although primary cognitive appraisal appears to both directly and indirectly influence sibling psychological and somatic difficulties, knowledge about the role of primary cognitive appraisal in subsequent stages of coping is largely unknown. Research examining cognitive appraisal, coping, and adjustment in individuals with and without chronic illnesses indicates there are significant associations between appraisals, coping strategies (e.g., avoidant behaviors), and adjustment difficulties (e.g., internalizing behavior problems) (Folkman et al., 1986b; Franks & Roesch, 2006; Gold et al., 2008). Thus, researchers must continue to examine how primary and secondary cognitive appraisals are affected when siblings encounter pediatric cancer and whether appraisals influence coping strategies and thus, adjustment. Such research will have important implications for sibling intervention programs.

Lastly, the present study examined how specific sibling, family, and disease factors identified by Wallander and Varni (1992) directly and/or indirectly influence sibling adjustment. Although social support, self-esteem, and/or primary cognitive appraisal account for a significant proportion of variance in sibling psychological and/or somatic difficulties, a large proportion of variance is unaccounted for. That is, the study does not account for all of the factors that predict

differences in sibling adjustment and should be targeted through intervention support. However, as the current study indicates research can identify factors that influence sibling adjustment and can be targeted through intervention, future researchers can utilize models such as the disability-stress-coping model (Wallander & Varni, 1992) and examine factors associated with sibling adjustment. Research will be most valuable if it examines factors amendable to intervention and provides both theoretical and clinical implications. Thus, although there is more to understand about the psychological adjustment of siblings and the effectiveness of intervention, the current study provides important insight and direction to optimize the adjustment and functioning of siblings whose brothers or sisters have pediatric cancer.

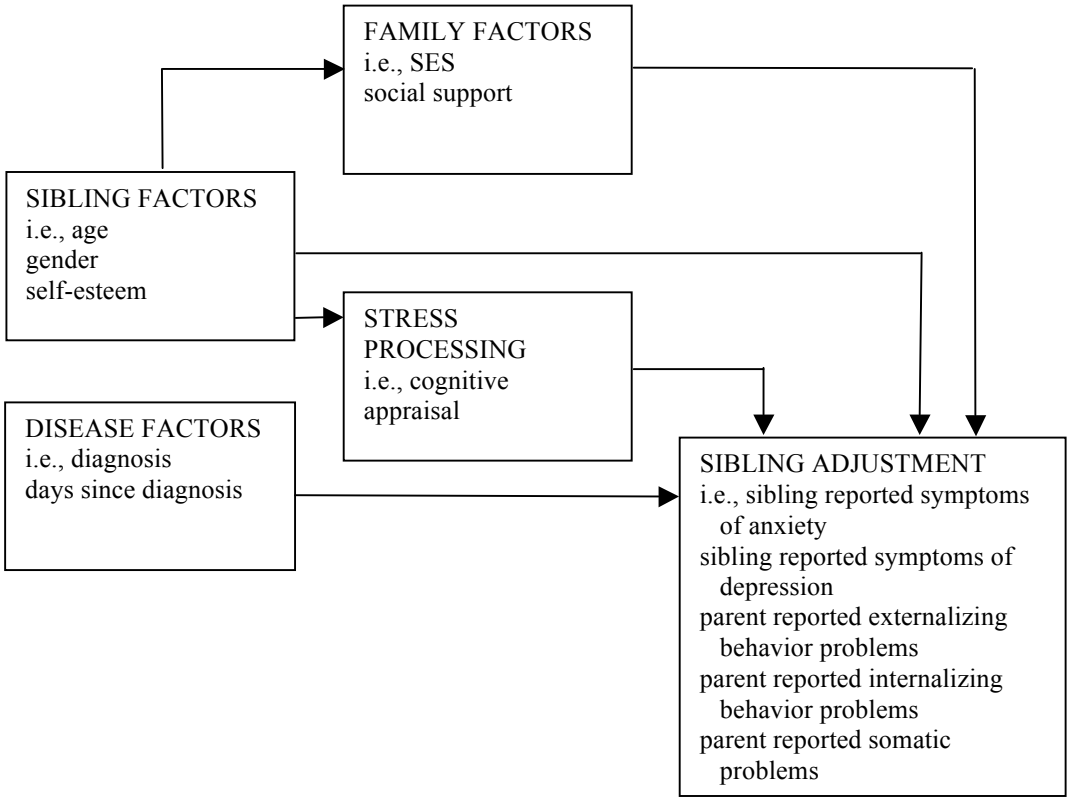

Figure 1. Adapted model of Wallander and Varni's (1992) disability-stress-coping model (Barrera et al., 2003; Barrera et al., 2004b; Barrera et al., 2007).

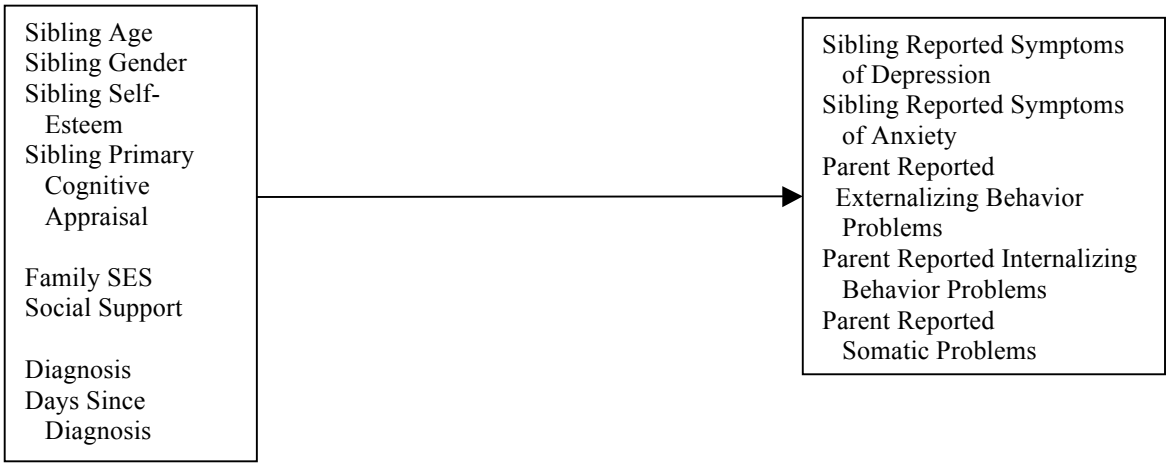

Figure 2. Pictorial representation of the direct relationship between sibling, family, and disease factors on sibling psychological adjustment.

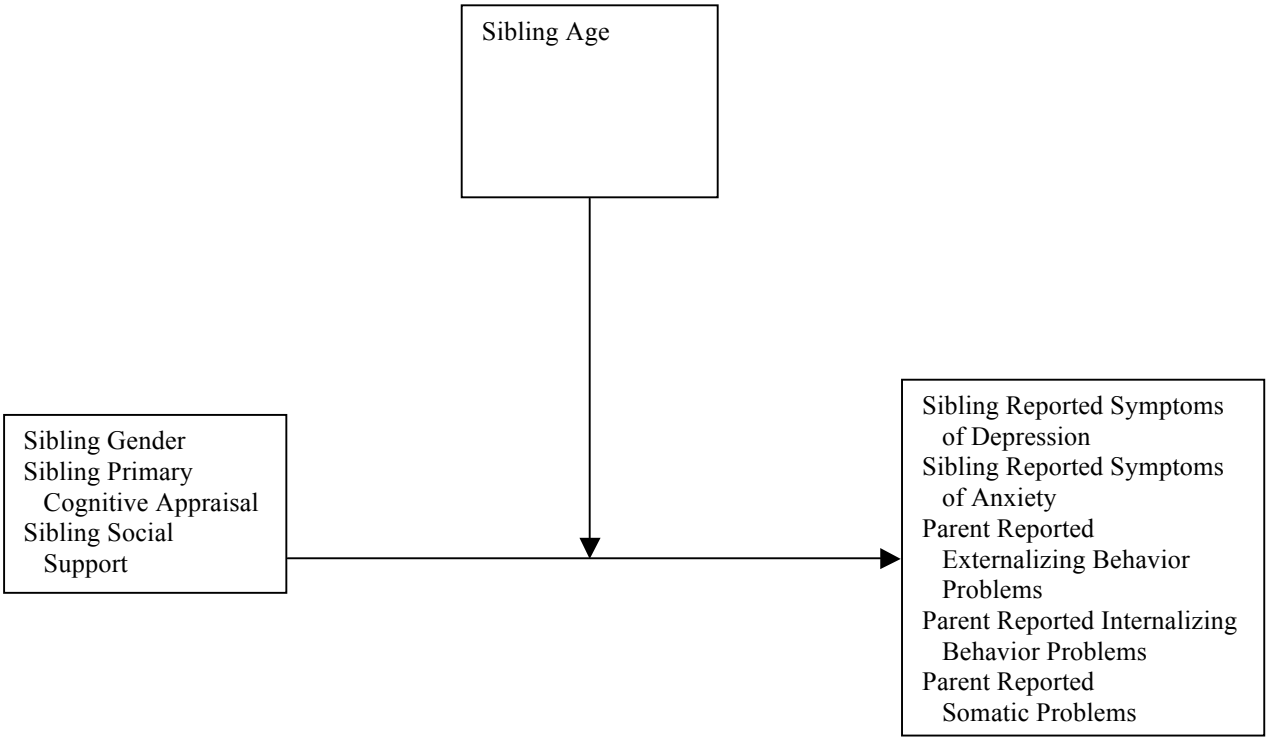

Figure 3. Pictorial representation of the moderating impact of sibling age on the relationship between sibling gender, primary cognitive appraisal, and social support and psychological adjustment.

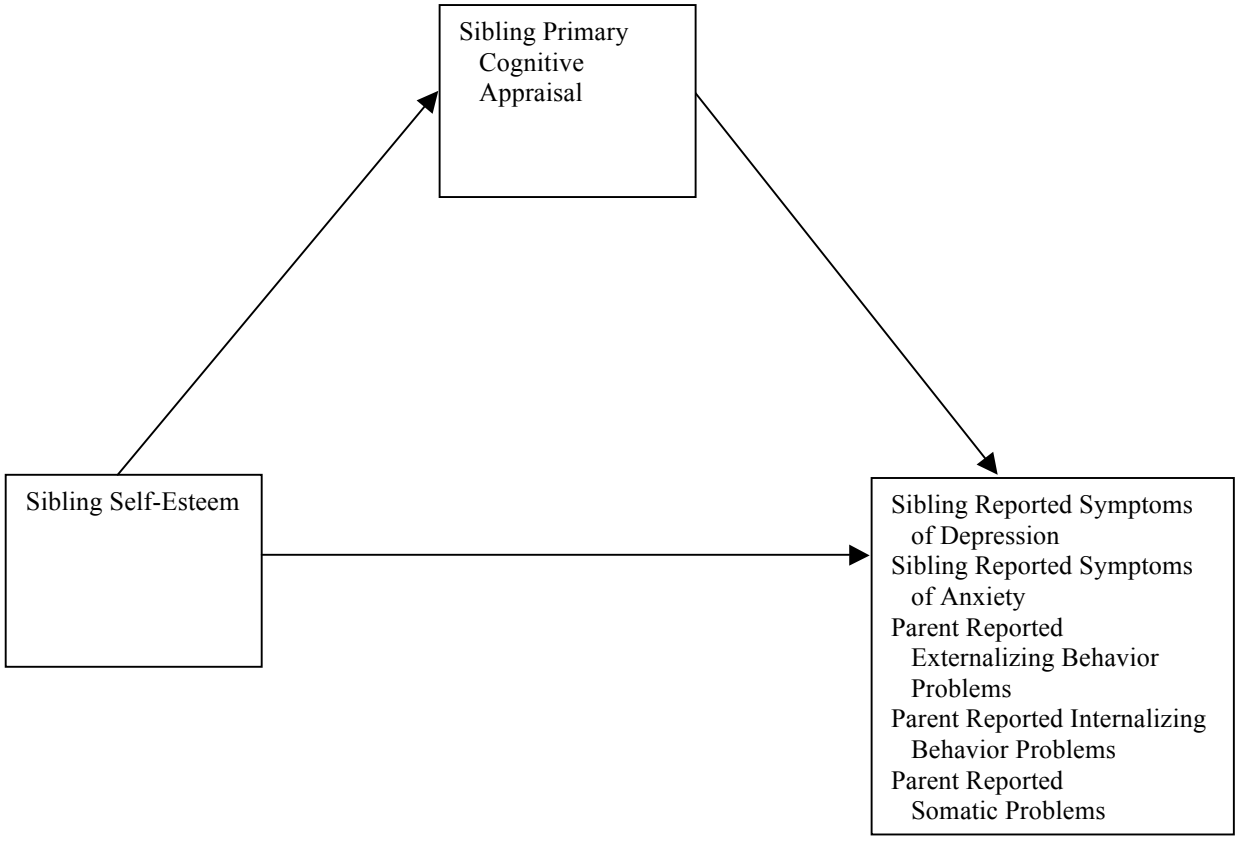

Figure 4. Pictorial representation of the mediating effect of sibling primary cognitive appraisal on the relationship between sibling self-esteem and sibling adjustment.

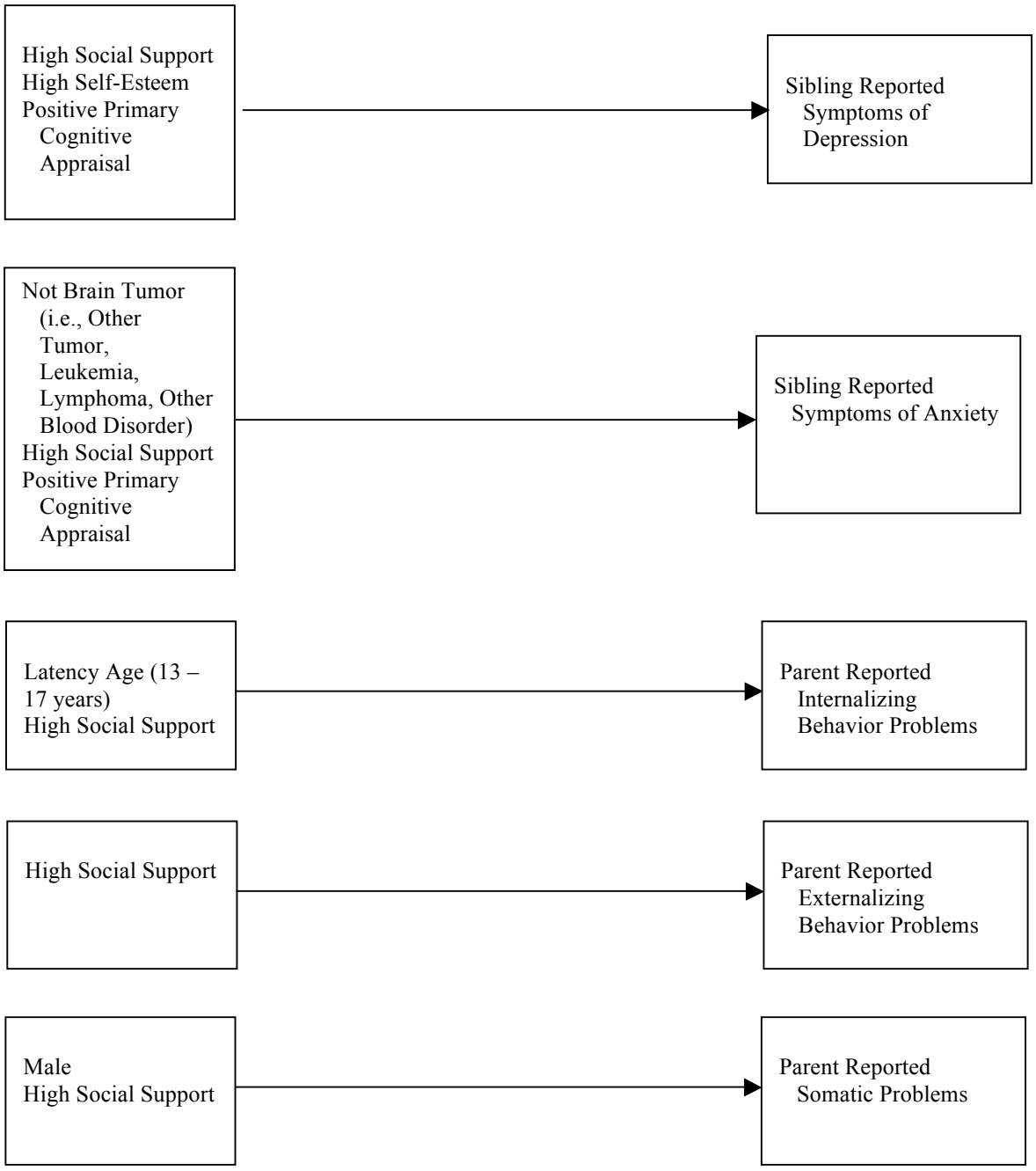

Figure 5. Summary of findings: Pictorial representation of the significant direct effects of sibling, family, and disease factors on each adjustment outcome. Boxes on the left identify protective factors.

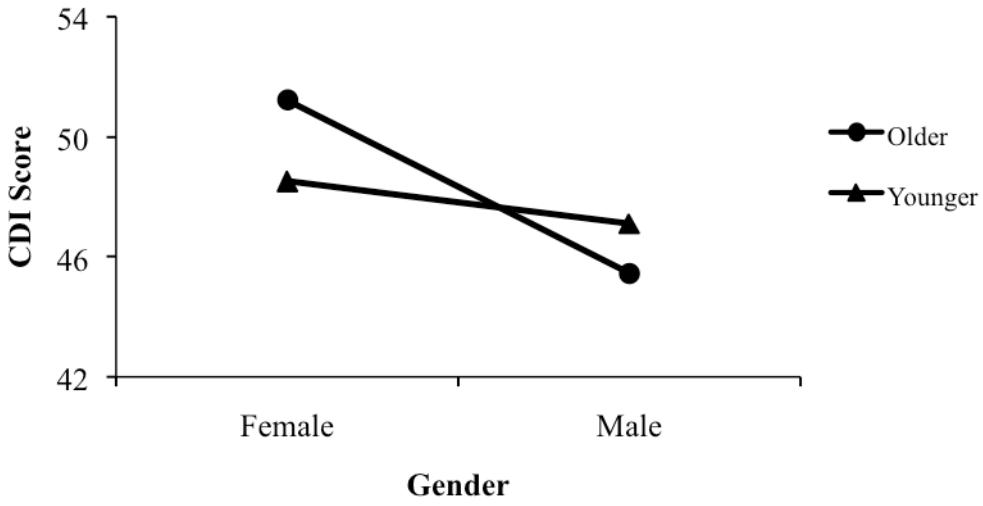

Figure 6. Age x gender moderation for CDI scores.

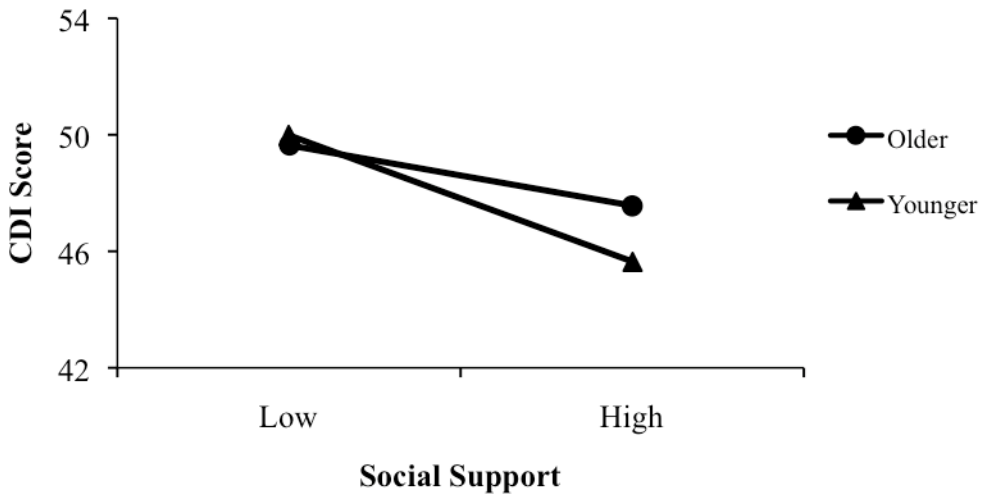

Figure 7. Age x social support moderation for CDI scores.

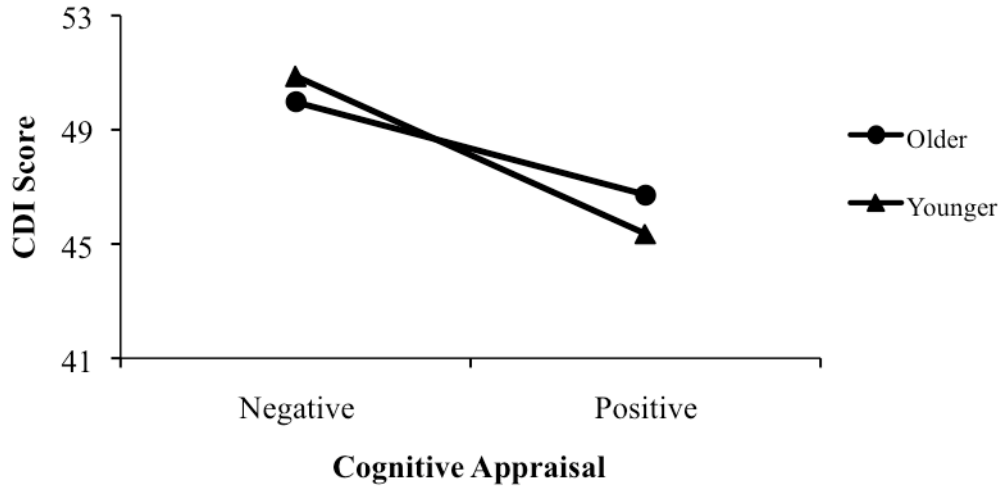

Figure 8. Age x primary cognitive appraisal moderation for CDI scores.

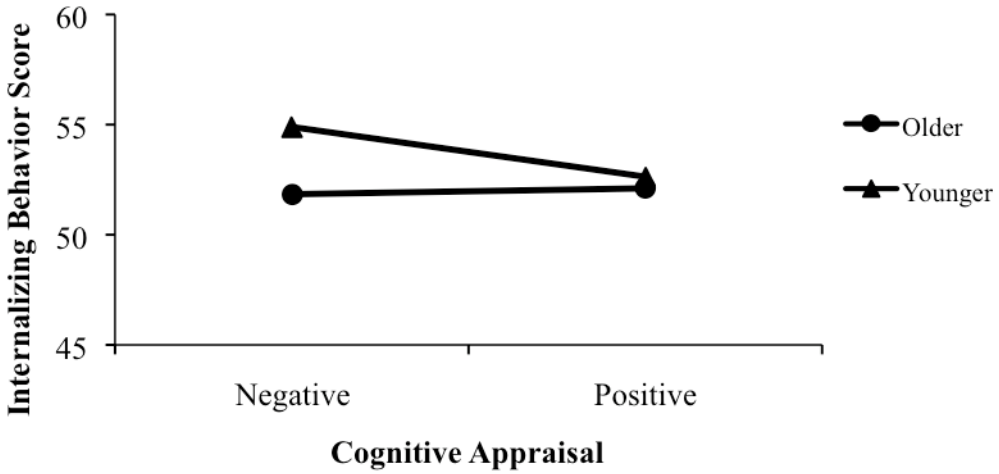

Figure 9. Age x primary cognitive appraisal moderation for internalizing behavior problems scores.

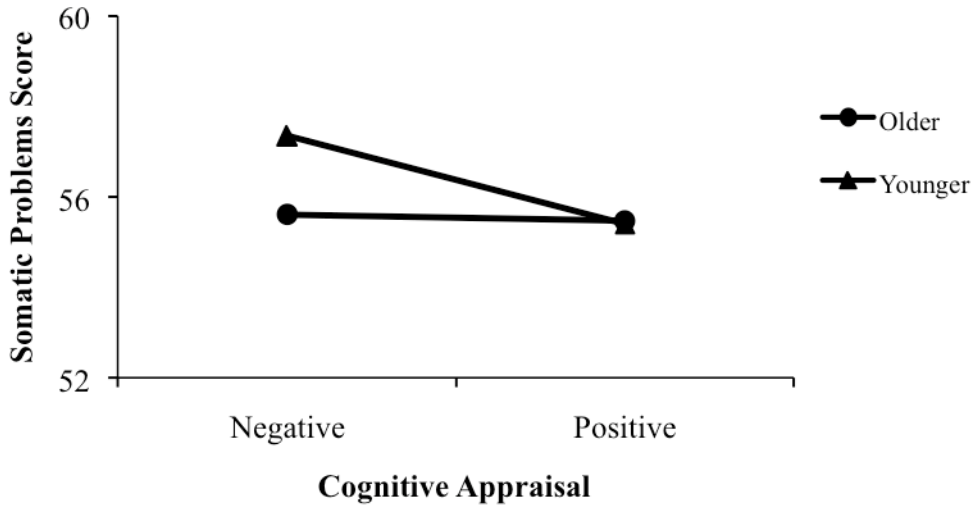

Figure 10. Age x primary cognitive appraisal moderation for somatic problems scores.

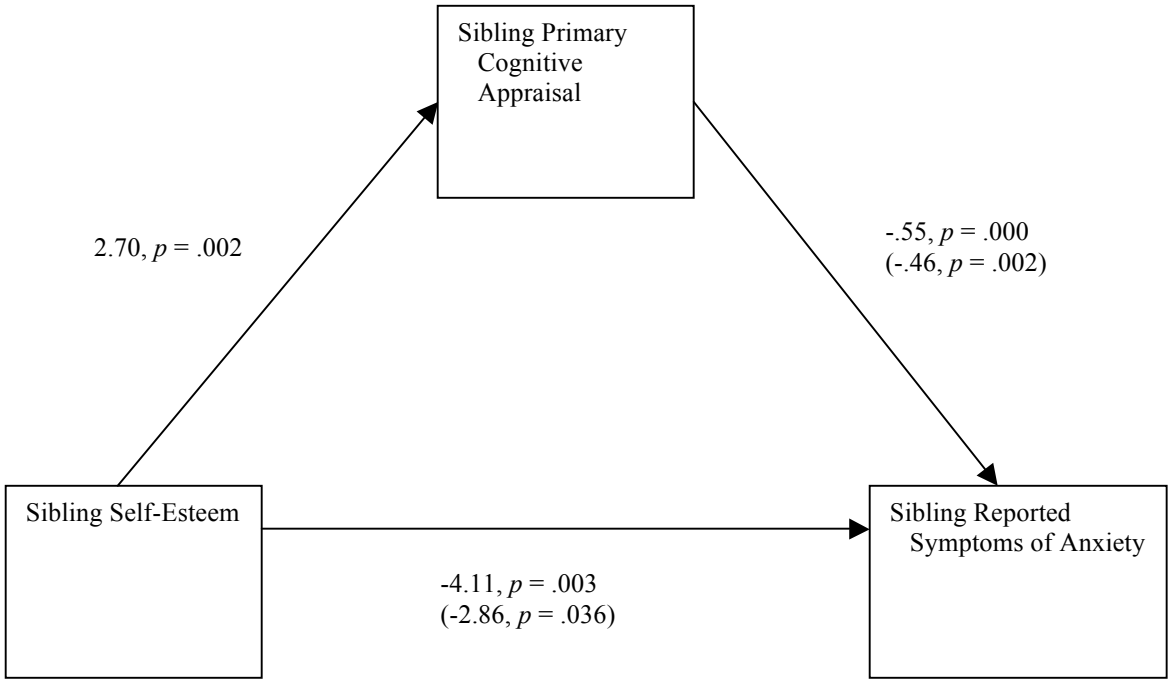

Figure 11. Mediational model for associations between sibling self-esteem and symptoms of anxiety as mediated by sibling primary cognitive appraisal. Values on paths are path coefficients (unstandardized *ß*s). Path coefficients outside parentheses are zero-order correlations. Path coefficients in parentheses are unstandardized partial regression coefficients from equations that include the other variable with a direct effect on the criterion.

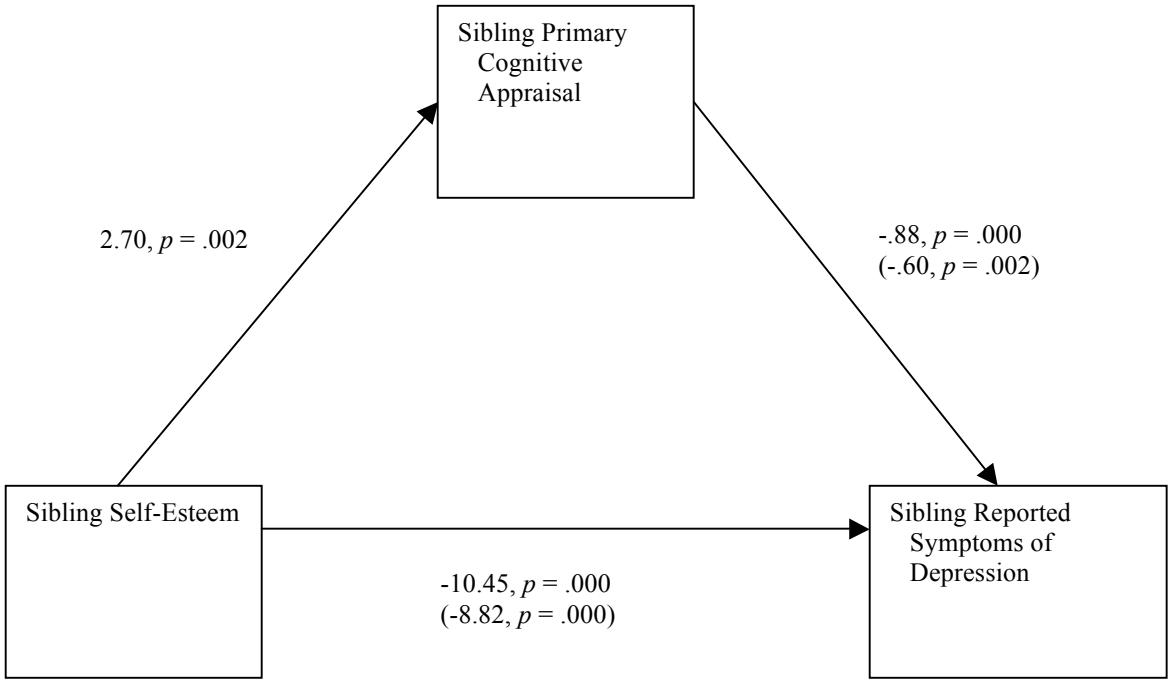

Figure 12. Mediational model for associations between sibling self-esteem and symptoms of depression as mediated by sibling primary cognitive appraisal. Values on paths are path coefficients (unstandardized *ß*s). Path coefficients outside parentheses are zero-order correlations. Path coefficients in parentheses are unstandardized partial regression coefficients from equations that include the other variable with a direct effect on the criterion.

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APPENDIX A

Non-Significant Moderations

Age x Gender for Symptoms of Anxiety

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	.78	.41	.058
Step 2	Gender	-.60	1.48	.685
Step 3	Age x Gender	-.74	.59	.208

Age x Social Support for Symptoms of Anxiety

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	.43	.28	.126
Step 2	Social Support	-.55	.16	.001
Step 3	Age x Social Support	.06	.07	.389

Age x Cognitive Appraisal for Symptoms of Anxiety

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.37	.28	.184
Step 2	Cognitive Appraisal	-.55	.14	.000
Step 3	Age x Cognitive Appraisal	.05	.06	.430

Age x Gender for Internalizing Behavior Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.59	.60	.322
Step 2	Gender	-1.95	2.18	.374
Step 3	Age x Gender	-.13	.86	.881

Age x Social Support for Internalizing Behavior Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.53	.41	.203
Step 2	Social Support	-.75	.24	.002
Step 3	Age x Social Support	.04	.10	.717

Age x Gender for Externalizing Behavior Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	.32	.55	.560
Step 2	Gender	-.35	2.00	.860
Step 3	Age x Gender	-.70	.79	.378

Age x Social Support for Externalizing Behavior Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.04	.38	.916
Step 2	Social Support	-.70	.22	.002
Step 3	Age x Social Support	-.02	.09	.809

Age x Cognitive Appraisal for Externalizing Behavior Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.14	.40	.733
Step 2	Cognitive Appraisal	-.13	.20	.513
Step 3	Age x Cognitive Appraisal	.13	.09	.128

Age x Gender for Somatic Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.13	.37	.727
Step 2	Gender	-2.38	1.35	.081
Step 3	Age x Gender	-.16	.53	.766

Age x Social Support for Somatic Problems

	Predictor	*ß*	S.E. *ß*	*p*
Step 1	Age	-.18	.26	.489
Step 2	Social Support	-.34	.15	.028
Step 3	Age x Social Support	.06	.06	.376

Non-Significant Mediations

Internalizing Behavior Problems

	Variable	*ß*	S.E. *ß*	*p*
Step 1	Self-Esteem	-1.00	2.05	.627
Step 2	Cognitive Appraisal	-.19	.22	.379
Step 3	Self-Esteem predicts Cognitive Appraisal	2.70	.87	.002
Step 4	Self-Esteem	-.52	2.15	.810
	Cognitive Appraisal	-.18	.23	.442

Externalizing Behavior Problems

	Variable	*ß*	S.E. *ß*	*p*
Step 1	Self-Esteem	-4.67	1.81	.011
Step 2	Cognitive Appraisal	-.13	.20	.524
Step 3	Self-Esteem predicts Cognitive Appraisal	2.70	.87	.002
Step 4	Self-Esteem	-4.72	1.90	.015
	Cognitive Appraisal	.02	.20	.926

Somatic Problems

	Variable	*ß*	S.E. *ß*	*p*
Step 1	Self-Esteem	.10	1.28	.939
Step 2	Cognitive Appraisal	-.21	.14	.134
Step 3	Self-Esteem predicts Cognitive Appraisal	2.70	.87	.002
Step 4	Self-Esteem	.71	1.33	.592
	Cognitive Appraisal	-.23	.14	.113