

**DEVELOPMENTAL AND FAMILY-LEVEL DETERMINANTS OF SIBLING  
ADJUSTMENT TO CHILDHOOD CANCER**

by

**Kristin A. Long**

A.B., Princeton University, 2003

M.S., University of Pittsburgh, 2008

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This dissertation was presented

by

Kristin A. Long

It was defended on

June 4, 2012

and approved by

Melissa Alderfer, PhD, Assistant Professor

Susan Campbell, PhD, Professor

Kirk Erickson, PhD, Assistant Professor

Linda Ewing, PhD, Assistant Professor

Robert Noll, PhD, Professor

Dissertation Advisor: Anna Marsland, PhD, Associate Professor

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Each year, 14,000 children and adolescents are diagnosed with cancer in the United States (Howlader et al., 2012). Significant biomedical advances have led to survival rates exceeding 80% (Jemal, et al., 2008). However, prolonged, complicated, and intensive treatment regimens often challenge and disrupt the entire family system, with effects extending to siblings. The present study examined the role of family risk factors in predicting distress among 209 siblings (ages 8-18) of children with cancer. Findings showed that greater sibling distress is independently associated with higher levels of sibling-reported problems with family functioning and parental psychological control, lower levels of sibling-reported maternal acceptance, and lower levels of paternal self-reported acceptance, with a similar trend for higher levels of parental posttraumatic stress. When examined as a function of sibling age, findings indicated that effects of sibling-reported maternal psychological control on sibling distress are stronger for older siblings, while effects of mother-reported problems with family roles on sibling distress are stronger for younger siblings. When the family risk factors were considered in combination, results supported a quadratic model in which the association between family risk and sibling distress was stronger at higher levels of cumulative family risk. Finally, hypotheses that a more positive family environment would buffer the negative influence of parent PTSS on sibling distress were largely unsupported. Together, findings support a family systems model of sibling

adjustment to a brother's or sister's cancer diagnosis in which elevated sibling distress is predicted by higher levels of family risk factors, alone and in combination.

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## 1.0 INTRODUCTION

Each year, 14,000 children and adolescents are diagnosed with cancer in the United States (Howlader et al., 2012). Significant biomedical advances over the past several decades have increased the average 5-year survival rate for these children to over 80% (Jemal, et al., 2008). However, improvements in cancer prognosis and survival have required prolonged, complicated, and intensive treatment regimens, which have the potential to challenge and disrupt the entire family system (Alderfer & Kazak, 2006).

Family caregivers consistently endorse high levels of strain as they confront a child's cancer and treatment (Vrijmoet-Wiersma et al., 2008), which may limit their ability to attend to the needs of other family members, including siblings. Existing literature suggests that the effects of childhood cancer on siblings are neither simple nor straightforward. Surprisingly, factors that may moderate sibling adjustment have received limited research attention (Alderfer, et al., 2010). Given the proximal influence of the family on child adjustment to chronic stressors (Bronfenbrenner, 1977), we have taken a developmentally sensitive, family systems approach to studying sibling adjustment to childhood cancer. Specifically, we have examined the influence of sibling age, parental cancer-related distress, family functioning, and parenting behaviors on siblings' adjustment to a brother's or sister's cancer diagnosis.

## **1.1 RESEARCH INVESTIGATING THE ADJUSTMENT OF SIBLINGS OF CHILDREN WITH CANCER**

### **1.1.1 Summary of the Extant Sibling Literature.**

The process of sibling adjustment to childhood cancer and the nature of siblings' psychosocial outcomes remain poorly understood (Alderfer & Noll, 2005). In general, qualitative work yields rich accounts of siblings' experiences, which are characterized by fear, loneliness, and a loss of security, but also by increased empathy and maturity (Alderfer, et al., 2010). The quantitative literature includes frequent mixed findings, but a recent meta-analysis confirmed that siblings of children with cancer are more likely to experience higher levels of internalizing and externalizing symptoms, along with lower levels of positive self-attributes, than siblings of healthy children (Vermaes, van Susante, & van Bakel, 2012). Further, siblings endorse high levels of negative emotion, low levels of positive emotion, poor quality of life, and moderate levels of cancer-related posttraumatic stress symptoms (PTSS) (Alderfer et al., 2010).

Although a considerable subset of siblings falls into the clinical ranges on measures of psychopathology symptoms, many siblings function well in the face of their brother's or sister's diagnosis (Alderfer et al., 2010). To date, the literature has not identified factors differentiating siblings who endorse ongoing difficulties from those who show resilience in the face of cancer-related stressors. With regard to demographic factors, a meta-analysis reported that siblings' internalizing or externalizing symptoms were not moderated by sibling gender, age, or birth order relative to the child with the chronic health condition (Vermaes et al., 2012). There is modest support for associations of greater sibling distress with more intrusive treatments for the



child with cancer and greater potential for life threat (Vermaes et al., 2012) and shorter time elapsed since diagnosis (Alderfer et al., 2010), but again, findings have been mixed.

Taken together, the literature on siblings of children with cancer suggests that this group experiences strong emotional reactions to their brother's or sister's cancer, which may result in heightened distress in a subset of siblings. The complex qualitative accounts reported in the literature, coupled with the mixed quantitative findings, suggest that sibling adjustment may be influenced by factors other than the cancer diagnosis per se, but risk and protective factors are not well addressed in the extant literature.

### **1.1.2 Critique of the Literature**

Despite dozens of research studies examining sibling adjustment to childhood cancer, there remains a surprisingly unsophisticated understanding of the experiences of this population (Alderfer, et al., 2010). Quantitative studies tend to employ cross-sectional designs and enroll small, heterogeneous samples which are collapsed across sibling demographic characteristics (e.g., age, race, gender) and illness variables (e.g., specific cancer diagnosis, treatment intensity, treatment status, time since diagnosis). Further, small sample sizes restrict power to detect small or medium effects and do not allow for the examination of potential moderators and mediators of sibling adjustment, including developmental factors. In addition, investigations of parents and families rely almost exclusively on mother report and fail to consider the role of fathers in sibling or patient adjustment.

On a broader level, both qualitative and quantitative studies generally are not grounded in existing theoretical models, nor do they integrate their findings into existing sibling research.

The result is that the extant sibling literature lacks coherence, which makes it difficult to integrate findings and draw firm conclusions. Given associations between child development and the family context (Sameroff, 1994), the limited attention to family factors in studies of sibling adjustment is a striking gap. Although preliminary evidence supports a model in which family factors moderate connections between parent and patient distress (Robinson, Gerhardt, Vannatta, & Noll, 2007), this has not been assessed in siblings. Indeed, examination of family-level risk and protective factors may partially explain inconsistencies in the extant sibling literature.

The present research addresses some of these limitations by (1) being grounded in principles of well-established theories, including developmental psychopathology and family systems; (2) employing a larger sample of siblings, thereby ensuring adequate power to examine developmental and contextual factors including sibling age, parent mental health, family functioning, and parenting, both individually and together as a constellation of family risk factors, (3) considering the roles of both mothers and fathers in sibling adjustment, and (4) limiting enrollment to families who are on active treatment and/or who are within 2-yr of the initial cancer diagnosis.

## **1.2 GUIDING THEORETICAL MODELS**

### **1.2.1 A Developmental Psychopathology Framework**

According to the developmental psychopathology framework, trajectories of child development are influenced by multiple internal and environmental factors interacting over time (Cummings,

Davies, & Campbell, 2000). A significant life event, such as childhood cancer, is neither necessary nor sufficient to predict present or future maladjustment or competence. Rather, a cancer diagnosis in oneself or a family member may have negative, positive, or negligible consequences depending on interactions among additional risk or protective factors such as family, peer, and school support; developmental competencies; temperament; and coping abilities (Garmezy, 1985).

This framework suggests that if an adverse event is experienced in the context of an otherwise healthy rearing environment, the affected child may have a low level of risk for poor emotional, behavioral, social, and academic outcomes, despite the presence of stress (Cummings, et al., 2000). Although a child in a risky situation has a higher *probability* of experiencing psychological difficulties (Cummings, et al., 2000), he or she can always proceed down (or return to) a path characterized by positive functioning (Sroufe, 1997).

A child who exhibits age-appropriate social, emotional, behavioral, and academic functioning despite serious threats to adaptation demonstrates resilience (Masten, 2001). While foundational work posited that resilience is an extraordinary outcome that is evident in only a minority of children facing adversity, recent conceptualizations suggest that resilience is relatively common. This may account for findings that many patients and their siblings function well in the face of childhood cancer (Alderfer et al., 2010; Noll & Kupst, 2007). Nonetheless, not all children display resilience in the face of threat. In this regard, it has been suggested that a child's risk for maladaptive outcomes increases when basic adaptational systems, such as parenting, are impaired prior to or as a result of the risk situation (Masten, 2001).

Risk and resilience reflect dynamic processes in response to a stressor, unfolding over time in the context of other risk and protective factors (Luthar, Doernberger, & Ziegler, 1993;

Rutter & Sroufe, 2000; Sroufe, 1997). Consistent with this process approach, an event may be a risk *indicator* but not necessarily a risk *mechanism* (Rutter, Pickles, Murray, & Eaves, 2001). For example, childhood cancer generally does not directly influence sibling development. Rather, the interplay of disease factors (e.g., treatment course, prognosis) unfolding within the family system (e.g., parent distress, family functioning, parenting) interact over time with characteristics of the sibling to influence developmental outcomes. Indeed, it appears that the cumulative number of risks, including both adverse events and environmental factors, may be a better predictor of future adjustment than the strength or severity of any one in particular (Sameroff, 2000). At present, risk and protective factors that may influence sibling adaptation to childhood cancer at the time of diagnosis or over time have received limited research attention.

### **1.2.2 Family Systems within the Social Ecology of Childhood**

Failure to consider factors likely to moderate psychosocial outcomes in siblings of children with cancer is inconsistent with developmental psychopathology and other theories of development that emphasize multiple contextual factors. For example, social ecological models (Bronfenbrenner, 1977) conceptualize child development as unfolding within various interacting social contexts including the family, school, community, and larger socio-cultural systems. These models suggest that the child is at the center of multiple nested social systems and that more proximal levels of influence (e.g., family characteristics) have a stronger impact on child development than more distal factors (e.g., community or cultural characteristics).

Siblings from more functional families are likely to display more positive adjustment outcomes. In this regard, the health of the family system is measured by the degree to which it

functions effectively as a whole, above and beyond the functioning of individual family members. For example, the McMaster Model of Family Functioning is based on the following tenets of systems theory: (1) all parts of a system are interrelated, (2) one family member's functioning cannot be understood separately from the larger family system, (3) family-level functioning cannot be understood by studying its component members and/or subsystems, and (4) family structure, organization, and transactional patterns influence individual members' behavior (Epstein, Bishop, & Levin, 1978; Miller, Ryan, Keitner, Bishop, & Epstein, 2000).

According to these assumptions, the McMaster Model focuses on six overlapping dimensions of family functioning: problem-solving, defined as a family's ability to successfully resolve challenges that threaten the family's integrity and/or functional capacity; communication, which refers to the clarity, directness, and effectiveness with which information is exchanged among family members; roles, which are the recurrent patterns of behavior by which individuals fulfill family functions; affective responsiveness, defined as the family's ability to respond to stimuli with the appropriate quality and quantity of emotions appropriate to the context; affective involvement, which refers to the degree to which the family values the activities and interests of individual members; and behavior control, which refers to a family's pattern of handling behavior in various situations (Epstein, et al., 1978; Miller, et al., 2000).

In the context of childhood cancer, some domains of family functioning shift to allow families to adapt to the needs of cancer- and treatment-related stressors (McCubbin, Balling, Possin, Frierdich, & Bryne, 2002). For example, it is well established that mothers' and fathers' roles shift such that mothers often assume more responsibility for the child with cancer while fathers may play a larger role in parenting other children in the home, in addition to maintaining previous occupational responsibilities (Chesler & Parry, 2001; Clarke, 2006; Kars, Duijnste,

Pool, van Delden, & Grypdonck, 2008; Martinson, et al., 1999; McGrath, Paton, & Huff, 2005; Reay, Bignold, Ball, & Cribb, 1998; Yeh, Lee, & Chen, 2000; Young, Dixon-Woods, Findlay, & Heney, 2002). These role shifts may decrease the amount of time parents are able to spend with siblings and the degree to which parents can monitor siblings' activities.

Other patterns of family interaction may not change as a function of the cancer diagnosis but are nonetheless likely to affect siblings' adjustment. For example, the family's problem-solving capacities may allow siblings to maintain their normal social and school-related activities in the face of the cancer and may influence the extent to which siblings assume responsibility for caretaking or household tasks. Strong family communication may facilitate open dialogue about the ill child's diagnosis, prognosis, and treatment regimen and the associated consequences for the family and siblings. In turn, adequate levels of affective responsiveness may contribute to parents' ability to model appropriate emotional reactions to childhood cancer, including fear, sadness, or hope. Overall, stable qualities of family functioning, coupled with the family's ability to adapt to cancer-related challenges, are likely to influence siblings' adjustment to cancer-related stressors and associated family changes.

### **1.3 PRESENT MODEL OF SIBLING ADJUSTMENT**

Consistent with the developmental psychopathology perspective, our overarching model of sibling adjustment considers multiple levels of interactive influences (Figure 1). After a child's cancer diagnosis, families report a series of day-to-day changes required to adapt to challenging treatment demands (Long & Marsland, 2011). A family's ability to make these adaptations, as

well as the effects of these changes on siblings' adjustment, are likely to be influenced by qualities of the family system and its members. Specifically, we believe that aspects of the sibling (e.g., developmental level, temperament, gender, emotion regulation abilities) interact with aspects of the family (e.g., parent mental health, parenting, family functioning) to contribute to siblings' cognitive and emotional reactions to the cancer. These reactions, in turn, are likely to influence the valence and intensity of siblings' adjustment outcomes. For example, sibling outcomes range from experiences of enhanced maturity, posttraumatic growth, and academic competence to decreased quality of life and symptoms of psychopathology (Alderfer, et al., 2010). Finally, it is likely that siblings' adjustment will also influence the family. Indeed, most of the relationships posited in our model are bidirectional in nature.

Although many contextual factors are likely to contribute to sibling adjustment to cancer, our research focuses on sibling age as a proxy for developmental level and on aspects of the family (Figures 2 & 3). With regard to age, older children and teenagers have developed more sophisticated methods of coping with cancer-related challenges, but they also are more likely to have the cognitive capabilities to understand the nuances of the diagnosis and prognosis and to appraise their brother's or sister's cancer as a threat. Other individual characteristics such as temperament and emotion regulation abilities are not considered in the present research. However, these and other factors are likely to influence siblings' reactions to their brother's or sister's cancer, via both main effects and interactions with family factors, and should be included in future research examining sibling adjustment.

Given that childhood cancer is considered a family-level stressor (Long & Marsland, 2001) and that the family is the most proximal external level of influence on children's development (Bronfenbrenner, 1977), we also consider the role of the family on sibling

adjustment. Although not directly examined in the current study, the effects of parent and family factors on child and adolescent outcomes may be partially explained by their effects on emotion regulation, which refers to internal and external processes involved in initiating, maintaining, and modulating the occurrence, intensity, and expression of emotions (Morris, Silk, Steinberg, Myers, & Robinson, 2007). In the context of childhood cancer, strong emotion regulation skills developed earlier in life may allow siblings to respond to stressful aspects of the cancer flexibly and effectively. Thus, the present research focuses on aspects of the family context that are likely to influence siblings' emotion regulation abilities, including parenting behaviors (acceptance and psychological control), the family context (general family functioning or specific domains including affective responsiveness and involvement, roles, communication, and problem-solving), and parental mental health (cancer-related distress).

It is expected that these overlapping family constructs will each account for some degree of the variance in sibling outcomes but may be more influential when considered as a group. For this reason, we have tested two models of the effects of combined family factors on sibling distress. First, we have tested the hypothesis that a more positive family environment (family functioning and/or parenting) may buffer the effects of greater parent PTSS on sibling distress (Figure 2). Second, we have calculated cumulative family risk scores for each sibling by dichotomizing each family risk variable into high (top 20% of poor parenting acceptance, psychological control, and general family functioning and at least one parent meeting criteria for posttraumatic stress disorder [PTSD]) or low levels of risk (bottom 80% of each family variable and neither parent meeting criteria for PTSD; Sameroff, Seifer, Barocas, Zax, & Greenspan, 1987; Belsky & Fearon, 2002). Dichotomized scores were summed to form a cumulative family risk index score, and the association of this score with sibling distress was evaluated (Figure 3).



Finally, we have examined the hypothesis that the effects of family variables on sibling distress is likely to vary as a function of sibling age. In the following sections, we review the literature related to each of the constructs included in our model.

### **1.3.1 Sibling Age / Developmental Stage**

Existing literature examining functioning of siblings of children with cancer frequently enrolled small samples of siblings spanning considerable age ranges. As a consequence, results are collapsed across siblings with different developmental competencies and needs, resulting in a literature that is largely insensitive to developmental factors (Alderfer, et al., 2010; Murray, 2000a, 2000b). This approach to data collection is inconsistent with principles of developmental psychology which suggest that the meaning of the illness and its impact on psychosocial functioning will vary as a function of cognitive, emotional, and social development. Accordingly, we propose one of the first studies to systematically examine sibling adjustment according to developmental stage, as assessed by age. Although findings are mixed, past work suggests that adolescent siblings may exhibit more difficulty adjusting to the cancer diagnosis than school-aged siblings (Alderfer, et al., 2010). Thus, we anticipate that adolescent siblings will show higher levels of distress than their school-aged counterparts.

Adolescence is characterized by attempts to regulate emotional states and behaviors according to long-term goals in a manner that is increasingly independent of adults (Steinberg, 2005). Emotional, cognitive, and behavioral processes mature at different rates during the course of adolescence, and difficulties coordinating these regulatory systems can lead to heightened emotional distress (Steinberg, 2005). Indeed, adolescents' increased susceptibility to distress

may be linked to functional and structural changes in areas of the brain involved with emotion regulation, with some aspects of emotional intensity and reactivity more closely related to pubertal maturation than to age (Steinberg, 1987). Although adolescence is a time of heightened emotionality, the majority of teens do not develop significant emotional, social, or behavioral difficulties (Steinberg & Morris, 2001). Rather, adolescent-onset problems may have their roots in earlier developmental influences, such as poor socialization of emotion regulation, which can lead to insufficient coping mechanisms in the face of normative adolescent stressors (Steinberg & Morris, 2001). In the case of childhood cancer, it is possible that adolescent siblings who have poorly coordinated regulatory systems may have more difficulty coping with normal emotional reactions to their brother's or sister's illness.

In addition to emotion regulation processes maturing during adolescence, neural changes from late childhood to middle adolescence are responsible for enhanced cognitive capacities that underlie abstract and hypothetical thinking (Steinberg, 2005). Accordingly, adolescent siblings are able to form a more sophisticated understanding of the meaning and potential consequences of a cancer diagnosis compared to their school-aged counterparts. Their primary appraisal of the event as a threat, particularly in the case of advanced cancer or relapse, may increase the perceived stressfulness of the event. Thoughts about the meaning and consequences of cancer, coupled with adolescents' developmental propensity to have difficulty coordinating emotional regulatory systems, may lead to heightened emotional reactions to their brother's or sister's cancer diagnosis.

Finally, emotional and cognitive changes characteristic of adolescence are accompanied by shifts in interpersonal relationships. Mild levels of parent-child conflict increase during early adolescence, along with declines in reported closeness and time spent together. The relationship

with parents becomes less contentious later in adolescence, and pubertal maturation is accompanied by more equality in parent-adolescent relationships (Steinberg & Morris, 2001). Similarly, sibling conflict in early adolescence gradually improves as siblings move into middle and later adolescence, with the quality of sibling relationships being influenced by the quality of parent-adolescent relationships (Brody, Stoneman, & McCoy, 1994). At the same time, adolescents spend more time with friends and less time with their families compared to school aged children, though the quality of parenting relationships influences adolescents' choice of peers and the quality of interactions with them (Larson & Richards, 1991).

Taken together, the literature on adolescent development suggests that this may be a particularly difficult time to confront stressors related to a brother's or sister's cancer diagnosis, as this developmental period is characterized by increasingly sophisticated cognitive skills, heightened emotional distress, striving for autonomy, and an associated shift in the focus of relationships from the family to the peer group. A family crisis may present the adolescent with a difficult choice. On one hand, the adolescent sibling may choose to identify with the family and therefore delay the normal developmental processes of individuation and shift of focus from the family to the peer group (Freeman, O'Dell, & Meola, 2000). On the other hand, the adolescent sibling who chooses to maintain his or her focus on peer relationships may experience guilt about being away from the family during the time of crisis (Quinn, 2004). There is little empirical work examining these possible trajectories that can follow a brother's or sister's cancer diagnosis. Further, developmental level is likely to interact with aspects of the family to influence sibling adjustment.

### 1.3.2 Parent Mental Health

One aspect of the family that has the potential to influence sibling adjustment to childhood cancer is disturbances to parent mental health that either precede or result from the cancer diagnosis. In the general child development literature, parent mental health has been widely examined as a determinant of child adjustment (Downey & Coyne, 1990). In the context of childhood cancer, reviews of the literature examining parental reactions to their child's diagnosis show that, when compared with normative data, most mothers and fathers report heightened mean levels of distress throughout the first year after diagnosis (Pai, et al., 2007) with a subgroup of parents experiencing ongoing distress that can persist for years (Vrijmoet-Wiersma, et al., 2008). In general, levels of anxiety peak around the time of the cancer diagnosis and recede thereafter, with ongoing anxiety being a risk factor for the later development of cancer-related PTSD (Vrijmoet-Wiersma, et al., 2008). In contrast, reports of the time course of depressive symptoms are mixed. While some studies report that depression levels decline with time since cancer diagnosis, others report ongoing elevated symptomatology at multiple time points thereafter (Vrijmoet-Wiersma, et al., 2008).

In parents of children with cancer, symptoms of depression and anxiety can either precede or result from the cancer diagnosis. In contrast, PTSS and PTSD are specific to the cancer experience, and symptoms can persist for years after the end of cancer treatment. Given comorbidity with other mental health disorders, it has been suggested that development of PTSD following a child's cancer diagnosis may reflect an underlying predisposition to psychopathology in the parent rather than a normative reaction to traumatic events (Bruce, 2006). In a review of the literature, Bruce (2006) reported that the incidence of cancer-related

PTSD diagnoses in parents ranges from 6.2 to 25%, with lifetime prevalence rates ranging from 27 to 54%. In studies that did not use a standardized diagnostic tool, estimates of clinically-severe levels of PTSS in parents of childhood cancer patients range from 9.8 to 44%.

Given that PTSS and PTSD emerge in parents after their child's cancer diagnosis, this index of distress may be particularly useful for studying the impact of parental mental health on sibling adjustment to childhood cancer. In general, parents who endorse greater emotional strain at the time of diagnosis tend to continue to experience higher levels of distress, even after treatment ends. Demographic risk indicators for ongoing parent distress include less education, lower socioeconomic status (SES), and greater perceived financial strain (Vrijmoet-Wiersma, et al., 2008). In addition, female gender, perceived severity of cancer and treatment, family conflict, poor social support, and emotion-focused coping are associated with parental cancer-related PTSD, but objective medical information such as treatment modality or severity is not (Bruce, 2006). Taken together, the literature shows that heightened parental distress is a normative reaction to a child's cancer diagnosis. In some cases, the distress may persist for years after the end of treatment. The extant literature does not address whether maternal or paternal distress, alone or in combination with other individual or family-level factors, contributes to sibling adjustment.

### **1.3.3 Family Functioning**

While genetic similarity accounts for some of the connection between parent and child mental health, evidence suggests that psychosocial aspects of the family environment contribute as well (Hammen & Rudolph, 2003). Consistent with these findings, research is increasingly

incorporating aspects of the family system into models predicting child adjustment (Sameroff, 1994). Children's symptoms of depression and PTSS have been linked to lower levels of family cohesion, flexibility, and communication and to higher levels of conflict (Fletcher, 2003; Hammen & Rudolph, 2003) and to changes to family routines (Fiese, et al., 2002). With regard to childhood cancer, family risk factors can precede and/or result from a child's diagnosis. For example, preexisting dysfunctional patterns of family interaction may limit the family's ability to adapt in the face of cancer, while adequate levels of problem-solving abilities and communication skills can facilitate a family's ongoing adjustment to cancer-related stressors.

Childhood cancer can impact the family system at multiple levels, from individual family members' experiences of day-to-day changes in routines, roles, and responsibilities and related distress, to disrupted relationships between family members and functioning of the family system as a whole. A small body of work examining mean differences in family functioning variables between families facing cancer and control or normative samples suggests that most families do not shift their underlying patterns of functioning after a cancer diagnosis (Long & Marsland, 2011). Further, the majority of studies included in this recent family review reported no mean differences in family functioning compared to norms (Foley, Barakat, Herman-Liu, Radcliffe, & Molloy, 2000; Kronenberger, et al., 1998; Maurice-Stam, Oort, Last, & Grootenhuis, 2008; Streisand, Kazak, & Tercyak, 2003) or comparison groups (Brown, Madan-Swain, & Lambert, 2003; Kazak, et al., 1997; Sawyer, Antoniou, Toogood, & Rice, 1997; Sawyer, Antoniou, Toogood, Rice, & Baghurst, 2000; Sawyer, Streiner, Antoniou, Toogood, & Rice, 1998), and longitudinal studies have failed to show reliable changes in family functioning variables over time after the cancer diagnosis (Barrera, Atenafu, & Pinto, 2009; Sawyer, et al., 1997; Sawyer, et al., 2000; Sawyer, et al., 1998).

Although most families' underlying structure and functioning do not appear to change following a child's cancer diagnosis, findings from qualitative research underscore the degree to which families face disruptions in daily routines, family life, employment, and economic burden, as well as shifts in mothers' and fathers' roles and responsibilities in response to cancer and its treatment (Long & Marsland, 2011). In general, mothers assume the majority of caretaking responsibilities for the child with cancer, while fathers attend to household, work, and sibling care. This pattern of household management has been termed "tag-team parenting" (Mercer & Ritchie, 1997) and may be particularly important when considering sibling adjustment. Unlike children in families that are not facing childhood cancer, siblings of children with cancer may have a higher proportion of their day-to-day caretaking needs addressed by fathers. Thus, it is possible that fathers' mental health, parenting behaviors, and contributions to effective family management may have an equal or even greater degree of influence on sibling adjustment compared to that of mothers.

A small body of qualitative work suggests that families continue to adjust to changing demands over the treatment course (Woodgate & Degner, 2004). Unlike parent mental health which tends to show a linear improvement over time, the order with which family changes occur and the degree to which challenges are resolved vary widely across families. In general, positive family adaptation has been linked to higher levels of cohesiveness, open communication, flexibility, and extended family support (Long & Marsland, 2011).

Taken together, this body of work suggests that most families experience day-to-day changes in their roles and routines after their child is diagnosed with cancer, without necessarily shifting their underlying patterns of family functioning. Indeed, it has been suggested that healthy pre-diagnosis functioning may allow families to shift roles and routines appropriately to

accommodate the needs of the cancer and its treatment (Long & Marsland, 2011; McCubbin, et al., 2002). Therefore, preexisting patterns of family functioning are likely to interact with cancer-related stressors and parent mental health to influence the family environment in the context of childhood cancer.

#### **1.3.4 Parenting**

Another aspect of the family system that is hypothesized to impact sibling adjustment is parenting, which has been identified as a fundamental system of adaptation which, if impaired, contributes to maladaptive child and adolescent outcomes (Masten, 2001). In healthy families, positive parenting behaviors help to establish a supportive emotional climate and promote positive parent-child relationships. Specifically, parental nurturance and involvement leads children to be more receptive to parents' influence, and the combination of support and structure allows children to develop self-regulatory skills (Steinberg & Morris, 2001). Parenting effects differ across childhood and adolescent development, and effects of positive parenting build over time (Collins, Maccoby, Steinberg, Hetherington, & Bornstein, 2000). For example, processes set in motion by supportive, firm parenting in preadolescence foster the development of competence and psychological wellbeing through the teenage years (Steinberg & Morris, 2001).

During middle childhood, parent-child relationships are characterized by increased reciprocity as compared to earlier stages of development. Parents facilitate the processes through which children acquire skills related to emotion regulation, self-management, and responsibility, and they help their children navigate the process of expanding their social network to include relationships outside the family. Enhanced cognitive reasoning skills contribute to better social



understanding, including children's abilities to understand aspects of themselves and their relationships with people within and outside the family unit (for review, see Collins, Madsen, & Susman-Stillman, 2002). In middle childhood, beliefs about human biology are frequently unsophisticated and sometimes inaccurate (Morris, Taplin, & Gelman, 2000). This has implications in the context of childhood cancer, where parents may need to facilitate siblings' understanding of their brother's or sister's diagnosis.

As children move into adolescence, the parent-teen relationship is increasingly based on conversation, negotiation, and joint decision-making (Maccoby, 1984). These relationships also are characterized by increased conflict, which may serve an important development purpose by allowing parents and adolescents to realign roles and expectations (Steinberg & Morris, 2001). A meta-analysis by Laursen and colleagues (1998) showed that the frequency of parent-child conflict increases from childhood to early adolescence and declines gradually and linearly thereafter. Rates of conflict between parents and adolescents are moderated by other factors, including preexisting family (dis)harmony, communication, parenting style, attachment, and closeness (Laursen, et al., 1998).

Despite mild conflict with parents, family relationships maintain a central role during adolescence. Parent-child bonds established earlier in life continue during this time, with most adolescents reporting warm, supportive relationships with their parents (Collins & Laursen, 2004). The exception to this pattern is found in families who have a history of interpersonal difficulties prior to the child entering adolescence (Steinberg & Morris, 2001), which challenges the family's ability to adapt and maintain closeness during this developmental transition (Collins & Laursen, 2004).

Across adolescence, the benefits of supportive parenting may be particularly important in the context of stressful life events. As discussed above, adolescents exhibit higher levels of emotional lability than children or adults, and stressful life events can trigger heightened emotional distress (Steinberg & Morris, 2001). Furthermore, increasing levels of autonomy may make adolescents more likely to avoid parents rather than rely on them as a source of coping support during stressful periods (Collins & Laursen, 2004). In contrast, in families characterized by warm, responsive parenting, the stressor may introduce the opportunity for increased closeness (Morris, et al., 2007). For example, parents who exhibit high levels of emotion coaching may notice, label, and validate their adolescent child's emotions; engage in emotional problem solving; and support his or her coping efforts (Gottman, Katz, & Hooven, 1996).

In the case of childhood cancer, parents' physical and emotional availability to siblings during cancer treatment may be limited. However, the quality of the relationship between parents and siblings, established prior to the cancer diagnosis, may modulate siblings' reactions to cancer-related stressors. Despite decreased availability, some parents are likely to engage in emotion coaching to assist siblings in coping with emotional aspects of the cancer diagnosis, which may lead to even closer parent-child relationships, more sophisticated emotion regulation skills, and positive adjustment in the sibling. In other cases, poor parent-child relationships may make parents less likely to engage in emotion coaching and may make siblings less receptive to parental involvement, which may result in poorer sibling coping and increased probability of adjustment difficulties.

## **1.4 EMPIRICAL EVIDENCE SUPPORTING THE RELATIONSHIPS AMONG CONSTRUCTS IN THE PROPOSED MODEL**

### **1.4.1 Associations between Parent Mental Health & Family Functioning**

Consistent with a family systems framework (Sameroff, 1994), empirical work suggests that family functioning and parent mental health are related in the context of childhood cancer. Cross-sectional work with families of children with cancer reveals associations between family functioning and parental posttraumatic stress (Barakat, et al., 1997; Brown, et al., 2003; Kazak, et al., 1997), acute stress (Patino-Fernandez, et al., 2008), anxiety symptoms (Kazak, et al., 1997; Kazak, et al., 1998; Patino-Fernandez, et al., 2008), depression symptoms (Shapiro, Perez, & Warden, 1998) and general adjustment (Sloper, 1996). Specifically, higher parent distress in the context of childhood cancer is correlated with lower family supportiveness (Brown, et al., 2003; Fuemmeler, Brown, Williams, & Barredo, 2003), higher family conflict (Brown, et al., 2003; Patino-Fernandez, et al., 2008), lower family satisfaction (Barakat, et al., 1997; Kazak, et al., 1997), poorer communication (Kazak, et al., 1997), and lower family cohesion (Kazak, et al., 1997; Maurice-Stam, et al., 2008; Patino-Fernandez, et al., 2008). Similarly, a mixed-method study of 98 families (120 parents) 6-mos. post-diagnosis found significantly higher distress in families for whom the cancer resulted in negative work effects, financial strain, decreased family and marital closeness, and increased marital tension (Sloper, 1996).

In addition to cross-sectional work, longitudinal studies of families facing childhood cancer suggest that aspects of the family may influence the course of parent distress over time. For example, Sloper (2000) found that family cohesion at 6-mos. post-diagnosis predicted parent

distress concurrently and one year later in a sample of 99 families in the United Kingdom. A second longitudinal study focused on the experiences of caretakers around the time of their child's bone marrow transplant (BMT). Family cohesion and conflict, but not expressiveness, strongly predicted parent distress across the transplant process (1-wk pre-transplant to 6-mos. post-transplant) such that parents with the most supportive family environments reported the lowest levels of distress (Phipps, Dunavant, Lensing, & Rai, 2005).

In contrast, two studies showed no significant association between family functioning and parent distress in families facing cancer. In a cross-sectional study of 24 mothers preparing for their child's BMT, family environment was not significantly correlated with maternal distress. However, the authors speculated that this was due to insufficient power, citing trends for associations between family functioning and mother-reported depressive and obsessive-compulsive symptoms, with  $r$ 's ranging from .25 to .29 (Kronenberger, et al., 1998). Similarly, a longitudinal study revealed no significant relationship between baseline family functioning and depression scores 6-mos. later in a sample of 51 mothers of children with non-CNS cancers. However, when measured concurrently, depression scores were correlated with other indices of family life, including spousal assistance at baseline, 3-mos., and 6-mos., and family routines at 3-mos. ( $r$ 's =  $-.38$ ,  $-.48$ ,  $-.35$ , and  $-.29$ , respectively) (Manne, et al., 1996). Thus, the two studies reporting nonsignificant results are nonetheless consistent with the idea that aspects of family functioning and parent mental health are related in families of children with cancer, though the timing and direction of these effects remains unclear.

### **1.4.2 Parent Mental Health & Family Functioning as Predictors of Child Outcomes**

Although hypotheses linking parent mental health and family functioning to sibling adjustment to childhood cancer have not been tested, empirical work in developmental and pediatric psychology support these proposed links. The association between parent mental health and child adjustment is well established in the general developmental literature (Downey & Coyne, 1990) as well as in pediatric cancer populations (Jobe-Shields, et al., 2009; Mulhern, Fairclough, Smith, & Douglas, 1992; Robinson et al., 2007; Trask, et al., 2003). Despite significant correlations between mental health of children and adolescents with cancer and their parents, however, the presence of parental distress alone is not sufficient to predict child and adolescent distress (Taieb, Moro, Baubet, Revah-Levy, & Flament, 2003). Rather, parental distress likely interacts with other aspects of the rearing environment, such as family functioning or parenting, to predict child and adolescent outcomes.

In the context of childhood cancer, cross-sectional and prospective work reveals that positive aspects of family functioning are associated with fewer internalizing and externalizing symptoms in survivors of hypothalamic/chiasmatic brain tumors (Foley, et al., 2000) and in a sample with mixed cancer diagnoses when measured concurrently at 1, 6, and 9-mos. after diagnosis (Sawyer, et al., 1997; Sawyer, et al., 2000; Sawyer, et al., 1998). When considering specific domains of family functioning, better family relationship quality is associated with lower internalizing and externalizing scores in samples of mixed on- and off-treatment child and adolescent patients (Fuemmeler, et al., 2003; Morris, et al., 1997; Noojin, Causey, Gros, Bertolone, & Carter, 1999). Also, adolescent survivors are more likely to exhibit cancer-related PTSS or PTSD when their families have unhealthy levels of problem-solving skills, affective

responsiveness, and affective involvement (Alderfer, Navsaria, & Kazak, 2009), higher levels of chaotic functioning (Pelcovitz, et al., 1998), or lower family satisfaction (Kazak, et al., 1997). When assessed prospectively, family functioning within a month after diagnosis predicts patient behavioral problems 6-, 9- (Varni, Katz, Colegrove Jr., & Dolgin, 1996), and 24-months later (Sawyer, et al., 1997; Sawyer, et al., 2000; Sawyer, et al., 1998).

Together, the literature suggests that parental mental health, affective components of family relationships, and family problem-solving skills each individually influences adjustment of children and adolescents with cancer. One important question, then, is how these variables might combine to influence outcomes in pediatric cancer populations. Very little research examines this question. To date, one study has examined whether aspects of the family environment moderate relationships between parent and child/adolescent adjustment in the context of childhood cancer. In a cross-sectional analysis of 8- to 15-year-olds with cancer, family relationship quality moderated the association between father distress and child/adolescent anxiety such that children and adolescents in families with more positive interpersonal relationship quality showed lower anxiety in the presence of father anxiety (Robinson, et al., 2007). Another study investigated whether parental depression moderates the relationship of family cohesion and expressiveness with distress in 6- to 18-year-olds with cancer who were awaiting stem cell or bone marrow transplantation. When parents had low depression scores, greater family cohesion and expressiveness were associated with lower distress in children and adolescents with cancer. When parents had higher depression scores, cohesion and expressiveness did not predict child and adolescent distress (Jobe-Shields, et al., 2009). Thus, initial work examining moderation is promising but leaves many questions unanswered. The

contribution of parent mental health and family functioning to sibling adjustment, alone or in combination, has not been addressed empirically.

### **1.4.3 Parenting & Child Outcomes**

Links between parenting and child/adolescent adjustment are well established, but at the same time, appear to be multidimensional and complex. Dimensions of parenting that have received considerable research attention and are relevant for siblings of children with cancer include acceptance versus rejection and psychological control versus autonomy. In non-cancer populations, a meta-analysis reported that risk for anxiety symptoms is increased with higher levels of parental rejection and lower levels of autonomy-granting (McLeod, Weisz, & Wood, 2007). Similar results have been found with regard to other internalizing symptoms (Gray & Steinberg, 1999; Papp, Cummings, & Goeke-Morey, 2005; Rogers, Buchanan, & Winchell, 2003). These parenting dimensions also appear to be important for children and adolescents who experience ongoing stressors. For example, in adolescents with depressed mothers, low levels of parental psychological control and maternal overinvolvement, and high levels of maternal warmth, are associated with adolescent resilience (Brennan, Le Brocque, & Hammen, 2003).

Unlike parent mental health and family functioning, parenting has been examined infrequently in families facing childhood cancer. A small body of quantitative work focuses on parenting stress in the face of childhood cancer and yields mixed results. Although higher parenting stress was reported by fathers of children and adolescents on active treatment (Kazak & Barakat, 1997) and by mothers of children and adolescents with brain tumors 2- to 5-yr post-diagnosis (Radcliffe, Bennett, Kazak, Foley, & Phillips, 1996), primary caregiving parents of

children and adolescents with mixed treatment status reported levels of parenting stress consistent with those of standardization samples (Bonner, Hardy, Willard, & Hutchinson, 2007). In addition, a longitudinal study of 68 families showed different patterns of stability versus change in various parenting dimensions over the first 24-wk after diagnosis. While parenting consistency decreased from the 12-14 to 22-24 week measurements, parenting responsiveness, control, and nurturance remained unchanged over this time (Steele, Long, Reddy, Luhr, & Phipps, 2003).

Two main themes emerge from qualitative work on parenting among families of children with cancer: (1) parents perceive an intensification of their parenting role after the diagnosis, resulting in closer relationships and deeper bonds with the child with cancer, and (2) parents report overprotecting and spoiling the child with cancer and setting fewer limits for him or her (Long & Marsland, 2011). Findings suggesting increased overprotectiveness are also reported in quantitative investigations. For example, adolescent cancer survivors view their parents as significantly more overprotective than adolescent physical abuse survivors or healthy comparisons (Sloper, 1996), and parents of children on active cancer treatment report higher levels of overprotection and lower levels of discipline than controls, particularly within first year after diagnosis (Hillman, 1997). However, the *effects* of enhanced parent-child bonds or parental overprotectiveness have not been investigated in the pediatric cancer research.

The literature examining parenting in the context of childhood cancer generally fails to distinguish between parenting of the child/adolescent with cancer versus siblings. However, three descriptive reports address this issue. By and large, parents are more indulgent toward and less demanding of the child with cancer compared to siblings (van Dongen-Melman, Van Zuuren, & Verhulst, 1998), suggesting that some differential parenting emerges. Some parents



describe the child or adolescent with cancer as vulnerable and disadvantaged compared to siblings, which contributes to the persistence with which he/she remains the center of attention within the family (van Dongen-Melman, et al., 1998). However, other work shows variability in preferential treatment, citing an equal split between parents who report treating the child or adolescent with cancer similarly to other children versus those who indicate that the child with cancer needs extra attention and care (Quinn, 2004). Similarly, a descriptive, quantitative study in Taiwan found that 44% of parents report favoring patients over siblings, with the majority of parents placing fewer academic and household demands on the child/adolescent with cancer and affording him/her greater leniency as compared to siblings (Chao, Chen, Wang, Wu, & Yeh, 2003). The effects of variability in parenting on sibling adjustment, as well as the meanings that siblings ascribe to these relationship patterns, remain uninvestigated.

Taken together, the literature on parenting in the context of childhood cancer is small and focuses on parenting of the child with cancer, leaving questions about siblings unanswered. Furthermore, the pediatric cancer literature does not consider the effects of many parenting dimensions that have been shown to influence child and adolescent adjustment in the larger developmental psychopathology literature, including acceptance-rejection and psychological control-autonomy.

#### **1.4.4 Parenting in the Context of Traumatic Stress: Influence on Child Outcomes**

The literature examining parenting in the context of childhood cancer is small, but parenting has been investigated in the context of other stressors. For example, in children with a history of sexual abuse, parental psychological control predicts children's PTSD symptoms and warmth

predicts children's depression symptoms (Deblinger, Steer, & Lippmann, 1999). Similarly, hostile parenting is associated with internalizing and PTSD symptoms in children and adolescents who have experienced a variety of traumatic events (Valentino, Berkowitz, & Stover, 2010), while positive parenting is associated with fewer trauma symptoms in adolescents from families with a history domestic violence (Levendosky, Huth-Bocks, & Semel, 2002).

In addition to main effects, aspects of parenting have been shown to moderate the impact of a trauma on adolescent mental health. For example, positive parenting predicted fewer adolescent trauma symptoms across families with domestic violence, but the effects were stronger in families with lower levels of violence compared to those with more pronounced violence (Levendosky, et al., 2002). Also, parental overprotectiveness was shown to augment the impact of a natural disaster on adolescent mental health such that adolescents who both experienced deleterious effects of a flood and whose parents exhibited higher levels of overprotection had higher levels of PTSD symptoms (Bokszczanin, 2008).

Although a small body of previous work has examined the independent roles of parent mental health and parenting in the prediction of child/adolescent adjustment to a stressful life event, the interaction of these two factors has received far less empirical attention. Furthermore, this research is complicated by the fact that parent mental health, parenting, child functioning, and the likelihood of experiencing negative life events are correlated (Webster-Stratton & Hammond, 1988). Nonetheless, there is a small literature reporting a protective role for parental warmth and psychological autonomy in stressful contexts such as maternal depression (e.g., Brennan, et al., 2003). This suggests that it is possible that positive parenting may buffer the potentially maladaptive effects of parent PTSS on siblings' adjustment in the context of childhood cancer-related stressors.

## **1.5 MOTHERS & FATHERS: DIFFERENCES IN PARENTING & MENTAL HEALTH**

In the context of childhood cancer, qualitative research has shown that siblings tend to spend more time with fathers, while mothers are invested more in the treatment of the child with cancer (Jones & Neil-Urban, 2003; McGrath, et al., 2005). This raises the possibility that sibling adjustment to childhood cancer may be more related to fathers' than mothers' mental health and parenting styles, and the literature shows differences between mothers and fathers in these domains.

Female gender is a risk factor for heightened parental distress following a child's cancer diagnosis (Bruce, 2006; Clarke, McCarthy, Downie, Ashley, & Anderson, 2009; Vrijmoet-Wiersma, et al., 2008). Consistent with a meta-analysis reporting higher levels of distress in mothers (Pai, et al., 2007), a systematic review of gender differences in parents of children with cancer revealed that approximately half of the 20 studies reviewed found higher levels of depression, anxiety, psychosomatic symptoms, and hopelessness in mothers than fathers (Clarke, et al., 2009). The remainder of studies reported no differences in parent distress according to parent gender. Null findings may reflect the timing of data collection, given evidence that gender differences are present during the earlier cancer treatment stages but decline thereafter (Dahlquist, Czyzewski, & Jones, 1996; Sloper, 2000). Parent coping style also appears to differ by gender, with mothers of children with cancer relying more on emotion-focused approaches and fathers utilizing more problem-focused strategies (Clarke, et al., 2009). It is unclear whether gender differences in parents' reactions to cancer are specific to the cancer experience or reflect population differences (Grigoriadis & Robinson, 2007).

Although not examined in the context of childhood cancer, the larger developmental literature reveals parenting differences between mothers and fathers. Most mothers and fathers within the same families display similar parenting styles by the time the child reaches adolescence (Steinberg & Morris, 2001), but there are differences in the nature of day-to-day interactions with their children. For example, adolescents spend more time with mothers and tend to have higher levels of emotional disclosure with their mothers than with their fathers (Collins & Laursen, 2004). Similarly, mothers report being more accepting and supportive of adolescents' emotional expressions and endorse significantly higher levels of emotion coaching than fathers (Stocker, Richmond, & Rhoades, 2007). In turn, parental emotion coaching is negatively correlated with adolescents' internalizing symptoms (Stocker, et al., 2007).

Emotion coaching may be particularly relevant for siblings of children with cancer, who show high levels of negative emotion in response to their brother's or sister's diagnosis (Alderfer, et al., 2010). Since siblings tend to spend more time with fathers, who report lower levels of emotional coaching and acceptance of negative emotions than mothers, it is possible that fathers' lower levels of responsiveness to negative emotion could lead to increased sibling distress. The role of fathers' mental health and parenting as a possible influence on sibling adjustment to childhood cancer has not been examined empirically.

## **1.6 INTEGRATION: CANCER, RISK, & SIBLING DISTRESS**

A childhood cancer diagnosis is a random, negative life event that interacts with aspects of individual family members and their environments to influence each family member's

adjustment. In the risk and resilience framework, the cancer diagnosis can be considered a risk indicator, and risk and resilience processes begin unfolding with day-to-day changes in a family's roles, routines, and organizational structure. Day-to-day family changes do not, in and of themselves, qualify as risk. Rather, these shifts appear to be normative and even adaptive responses to childhood cancer (McCubbin, et al., 2002). However, the family's ability to mobilize and shift effectively in the face of cancer is likely to be influenced by pre-existing patterns of family functioning and parenting; these contextual factors are also likely to influence sibling adjustment to their brother's or sister's cancer. Furthermore, these family-related risk or protective factors are likely to be more pronounced by the time siblings reach adolescence, as effects build over time. Thus, it is expected that the impact of parenting and family functioning variables on sibling adjustment will be more pronounced in adolescent siblings than in their school-aged counterparts.

Family factors likely to influence sibling adjustment include pre-diagnosis parenting acceptance and psychological control, the overall quality of family functioning, and specific domains of family functioning including roles, communication, problem-solving, and affective responsiveness. Families functioning well in these domains are likely to follow a path of flexible reorganization to adapt to the stressor. Challenges to parent mental health and parenting are expected to be temporary and improve with time after diagnosis. In addition, a family's sense of self-efficacy to confront cancer-related challenges will be enhanced with repeated successes (Woodgate & Degner, 2003). As members of these systems, siblings are likely to have well-developed emotion regulation skills and may appraise their situation more positively (e.g., "we are working together to confront my brother's cancer"). They are less likely to develop

symptoms of psychopathology and instead may show outcomes indicative of steeling or posttraumatic growth.

In families with less adaptive patterns of pre-cancer functioning, cancer-related changes may further the family down a path of maladjustment. Normative cancer-related changes to day-to-day family roles and routines can lead to further disorganization. These families are more likely to experience ongoing deficits in parenting and persistent patterns of poor mental health. Given bidirectional influences among individual family members, family subsystems, and the larger family system, it is likely that these factors will co-occur (Sameroff, 2000). Although each risk factor (e.g., parent posttraumatic stress, parenting rejection and psychological control, and poor family functioning) is likely to predict sibling adjustment, their effect on sibling distress may be more pronounced when considered as a group. This constellation of risk factors, combined with the presence of the cancer diagnosis, has the potential to challenge the integrity of the family unit, and siblings in these families may appraise their situation more harshly. In turn, these siblings are more likely to exhibit heightened distress in the context of their brother's or sister's cancer.

## **1.7 SPECIFIC AIMS**

Our overarching goal was to systematically examine sibling adjustment after a childhood cancer diagnosis in relation to developmental and family-level factors, including sibling age, parent mental health, family functioning, and parenting. This line of research is supported by the social ecology framework (Bronfenbrenner, 1977), which suggests that sibling adjustment is influenced

by individual characteristics and aspects of the larger family system and its members, especially parents. Specific aims and hypotheses are as follows:

1. Main Effects: Determine whether sibling age, parent cancer-related distress (PTSS), family functioning, and/or parenting behaviors predict sibling distress.

Hypothesis 1a: Adolescent siblings will show higher levels of distress compared to their school-aged counterparts.

Hypothesis 1b: Higher levels of parental PTSS will predict greater sibling distress.

Hypothesis 1c: Poorer family functioning, including lower general family adjustment, problems with family roles, poorer problem-solving and communication skills, and poorer affective responsiveness and involvement, will predict greater sibling distress.

Hypothesis 1d: Lower levels of parenting acceptance and higher levels of parenting psychological control will predict greater sibling distress.

2. Moderation Effects: Assess whether (1) family functioning and/or parenting moderates the relationship between parental PTSS and sibling adjustment, and (2) whether the impact of family and parent variables on sibling distress changes according to sibling age.

Hypothesis 2a: There will be an interaction between parental PTSS and family functioning in predicting sibling distress. In parents who endorse higher levels of PTSS, the negative impact on siblings will be buffered by a more positive family environment.

Hypothesis 2b (Exploratory): In parents who endorse higher levels of PTSS, the impact on sibling distress will be buffered by higher levels of acceptance and lower levels of psychological control.

Hypothesis 2c: The effect of parent and family variables on sibling distress will be greater for older siblings.

3. Combined Effects: Examine the association of sibling distress with the constellation of family-level risk factors (poorer general family functioning, parenting acceptance and psychological control, and parent PTSS) using the cumulative risk methodology.

Hypothesis 3a: Higher levels of cumulative family risk will predict greater sibling distress.

Hypothesis 3b: The relationship between cumulative family risk and sibling distress will be stronger at higher levels of family risk.

4. Gender Effects: Characterize the contribution of mother versus father reports of mental health, family functioning, and parenting on sibling adjustment.

Hypothesis 4 (Exploratory): Sibling distress will be more strongly related to father- than mother-reported PTSS, family functioning, and parenting.



## 2.0 METHODS

### 2.1 SAMPLE & PROCEDURE

The sample includes 210 families of children with cancer, including siblings ( $n = 210$ ), mothers ( $n = 187$ ), and fathers ( $n = 71$ ). Data were collected as part of two larger studies at The Children's Hospital of Philadelphia. Inclusion criteria were as follows: the child with cancer was receiving active treatment and/or was within 2 years of diagnosis and currently living; the sibling was between the ages of 8 and 18 years (Study 1) or 8 to 15 years (Study 2); and the family spoke English fluently. For the Study 1, either the mother or the father, but not both, were enrolled. For Study 2, both mothers and fathers in the same families were invited to participate.

In both studies, families were identified by tumor registry lists at The Children's Hospital of Philadelphia, screened for eligibility, and invited to participate by letter and follow-up phone call. The enrollment rates were 75% for Study 1 and 81% for Study 2. A home visit was scheduled with those families interested in participation. After procurement of assent and consent, siblings and their parents each completed a battery of psychosocial questionnaires. Parents were asked to report on their experience of posttraumatic stress, family functioning, and parenting behaviors. Siblings were asked to report on their own distress, family functioning, and parenting behaviors (see below). All eligible siblings interested in participation were enrolled.

For families with more than one enrolled sibling, the sibling closest in age to the child with cancer was selected to be included in analyses.

## 2.2 MEASURES

### 2.2.1 Posttraumatic Stress Diagnostic Scale

*Posttraumatic Stress Diagnostic Scale* (PDS) (Foa, 1995)– Completed by parents, this 49-item measure assesses the presence and severity of PTSD symptomatology and impairment and closely corresponds to the PTSD diagnostic criteria as outlined by the *Diagnostic and Statistical Manual of Mental Disorders-IV* (American Psychiatric Association, 1994). The measure yields a total severity score, three subscales (avoidance, reexperiencing, and arousal), and a functional impairment index. In addition, information about PTSD diagnostic status was derived by assessing whether each participant met diagnostic criteria as outlined in the DSM-IV: (1) presence of a traumatic event; (2) response involving intense fear, horror, or helplessness; (3) re-experiencing ( $\geq 1$  symptom); (4) avoidance ( $\geq 3$  symptoms); (5) arousal ( $\geq 2$  symptoms); (6) symptom duration  $\geq 1$ -mos., and (7) functional impairment.

The instructions were altered to ensure that parents reported symptoms related their experience of their child's cancer. Satisfactory internal consistency has been demonstrated in the validation sample ( $\alpha = .89$ ) (Foa, Cashman, Jaycox, & Perry, 1997) and in a sample of adult women with breast cancer ( $\alpha = .92$ ) (Brown, et al., 2007). In the present sample, internal consistency for the total score was calculated to be .92 for both mothers and fathers. The scale

also has been shown to have high test-retest reliability (coefficient = .83), and adequate concurrent and convergent validity with other PTSD scales (Foa, et al., 1997). This scale has been used in previous studies of parents of childhood cancer survivors (Fuemmeler, Mullins, & Marx, 2001; Glover & Poland, 2002).

### **2.2.2 Family Assessment Device**

*Family Assessment Device* (FAD) (Epstein, Baldwin, & Bishop, 1983) – Completed by both parents and siblings, this 60-item measure evaluates family functioning according to the McMaster model of family functioning. Along with a general functioning scale, the FAD has six subscales: problem-solving, communication, roles, affective responsiveness, affective involvement, and behavior control. Consistent with the McMaster Model of family functioning, which suggests that family domains are interrelated, the six subscales overlap to some degree (Miller, et al., 2000; Ridenour, Daley, & Reich, 1999). This measure is considered “well established” for chronic illness populations (Alderfer, et al., 2008). Psychometric acceptability has been demonstrated in families of children with cancer (Kabacoff, Miller, Bishop, Epstein, & Keiter, 1990). A cut score of 2 on the general functioning subscale was used to identify families with unhealthy patterns of functioning (Miller, Epstein, Bishop, & Keitner, 1985).

In the present sample, internal consistency was good for general family functioning (sibling: alpha = .84, mother: alpha = .84; father: alpha = .83). Internal consistency ranged from poor to acceptable across subscales: problem solving (sibling: alpha = .65, mother: alpha = .65; father: alpha = .63), communication (sibling: alpha = .49, mother: alpha = .70; father: alpha = .64), roles (sibling: alpha = .60, mother: alpha = .68; father: alpha = .76), affective

responsiveness (sibling:  $\alpha = .57$ , mother:  $\alpha = .67$ ; father:  $\alpha = .72$ ), affective involvement (sibling:  $\alpha = .63$ , mother:  $\alpha = .72$ ; father:  $\alpha = .73$ ), and behavioral control (sibling:  $\alpha = .57$ , mother:  $\alpha = .73$ ; father:  $\alpha = .73$ ). In general, internal consistency was higher for parent- than sibling-report.

### **2.2.3 Child Report of Parent Behaviors Inventory, Short Form**

*Child Report of Parent Behaviors Inventory, Short Form* (CRPBI – 30) (Schludermann & Schludermann, 1988). With versions for both children and parents, these 30-item scales measure perceptions of parenting behaviors. Items are rated on 3-point Likert scales, (1 = “Not like my parent/me” to 3 = “A lot like my parent/me”). Three subscales include Acceptance-Rejection (e.g., “My mother/father is a person who makes me feel better after talking over my worries with her/her”), Psychological Control – Autonomy (e.g., “She/he says if I really cared for her/him, I would not do things that cause her/him to worry”) and Firm Control (e.g., “She/he is very strict with me”); the Acceptance-Rejection and Psychological Control-Autonomy subscales were used in this study. In the present sample, internal consistency ranges from poor to excellent. Alpha values for the Acceptance subscale are .90-.91 for sibling report, .81 for mother report, and .86 for father report; alpha values for Psychological Control are .79-.80 for sibling report, .73 for mother report, and .57 for father report.

#### **2.2.4 Child Depression Inventory, Short Form**

*Child Depression Inventory – Short Form* (CDI-S) (Kovacs, 1992) – Completed by siblings, this 10-item self-report questionnaire assesses the frequency and severity of depressive symptoms. In the present study, the total score was used as a measure of sibling distress, and raw scores were converted to T-scores to characterize the percentage of sibling participants whose depression scores fell into the borderline (T-scores from 60-69) or clinical range (T-scores  $\geq 70$ ). This measure has relatively high levels of internal consistency, test-retest reliability, and predictive validity (Ialongo, Edelsohn, & Kellam, 2001; Mattison, Handford, Kales, & Goodman, 1990), along with adequate construct (Worchel., Rae, Olson, & Crowley, 1992) and discriminate validity (Carey, Faulisch, Greshman, Ruggiero, & Enyart, 1987). The CDI has been used previously with children with cancer (Mulhern, Fairclough, Douglas, & Smith, 1994) and their siblings (Barrera, Chung, Greenberg, & Fleming, 2002; Barrera, Fleming, & Khan, 2004; Chao, Chen, Wang, Wu, & Yeh, 2003; Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004; Packman, et al., 1997; Wellisch, Crater, Wiley, Belin, & Weinstein, 2006), and with children of adults with cancer (Compas, et al., 1994; Harris & Zakowski, 2003; Welch, Wadsworth, & Compas, 1996). In the present study, internal consistency was calculated to be .78.

#### **2.2.5 Revised Children’s Manifest Anxiety Scale**

*Revised Children’s Manifest Anxiety Scale* (RCMAS) (Reynolds & Richmond, 1985). Completed by the siblings, this 37-item self-report measure assesses anxiety. The total anxiety score was used as a measure of sibling distress, and raw scores were converted to T-scores to characterize

the percentage of sibling participants whose anxiety scores fell into the borderline (T-scores from 60-69) or clinical range (T-scores  $\geq 70$ ). Past research using this measure reported internal consistency values that exceed 0.80 (Gerard & Reynolds, 1991) and adequate test-retest reliability (Wisniewski, Mulick, Genshaft, & Coury, 1987). This measure has been validated for children / adolescents ages four to 19. The RCMAS has been used previously in research on siblings of children with cancer (Alderfer & Hodges, 2010; Alderfer, Labay, & Kazak, 2003; Kazak, et al., 2004; Packman, et al., 1997; Packman, Fine, et al., 2004; Packman, Gong, vanZutphen, Shaffer, & Crittenden, 2004). In the present sample, internal consistency was calculated to be .88.

### **2.2.6 Child PTSD Symptom Scale**

*Child PTSD Symptom Scale* (CPSS) (Foa, Johnson, Feeny, & Treadwell, 2001) – Completed by siblings, this 26-item measure is the child / adolescent version of the PDS (Foa, 1995). In addition to a total symptom severity scale, the CPSS assesses the presence and severity of three clusters of PTSD symptoms in children ages eight to 18: re-experiencing, avoidance, and increased arousal. Information about PTSD diagnostic status was derived by assessing whether each participant met diagnostic criteria as outlined in the DSM-IV: (1) presence of a traumatic event; (2) response involving intense fear, horror, or helplessness; (3) re-experiencing ( $\geq 1$  symptom); (4) avoidance ( $\geq 3$  symptoms); (5) arousal ( $\geq 2$  symptoms); (6) symptom duration  $\geq 1$ -mos., and (7) functional impairment. Finally, a cut score of 11 was used to identify siblings endorsing moderate to severe levels of PTSS, which is reported to have high sensitivity (95%) and specificity (96%) for predicting cases of child/adolescent PTSD (Foa et al., 2001).

Wording was altered to ensure that siblings' responses referred to their brother's or sister's cancer. Good internal consistency was reported in the validation sample ( $\alpha = .89$ ) (Foa, et al., 2001) and in a sample of children of women with breast cancer ( $\alpha = .73$ ) (Brown, et al., 2007). In the present sample, internal consistency was calculated to be .89. Adequate test-retest reliability ( $r = .84$ ) and convergent validity with other child PTSD measures ( $r = .80$ ,  $p < .001$ ) have been established (Foa, et al., 2001).

## 2.3 DATA ANALYSIS

### 2.3.1 Preliminary Analyses

The first set of preliminary analyses examined levels of skew and kurtosis among outcome variables. Skew and kurtosis values were considered adequate when the quotient of skew (or kurtosis) divided by the standard error of the skew (or kurtosis) was less than 3. When necessary, outcome variables were transformed to obtain acceptable distributions.

The next set of preliminary analyses aimed to examine overlapping variance among factors and reduce data. First, we examined correlations among the measures of sibling distress (depression, anxiety, and posttraumatic stress symptoms). An  $r$  value of 0.6 was used as the minimum criterion for combining measures to form a composite distress score, and a principal components factor analysis with varimax rotation was conducted to characterize how the three distress scores loaded onto a single composite distress scale. Next, the concordance between parent- and sibling-reported family functioning and parenting variables was examined using

Pearson correlations. Again, an  $r$  value of 0.6 was used as the criterion for combining parent- and sibling-reported family functioning and parenting variables.

Another set of preliminary analyses was carried out to determine the strength of the associations between sibling distress and the following possible covariates: time since diagnosis, sibling gender, number of children in the family, sibling birth order, race, socioeconomic status, and marital status. This was tested using Pearson correlations for continuous variables and independent samples  $t$ -tests for categorical variables. Factors that were significantly correlated with sibling distress were entered as covariates into all subsequent analyses.

Finally, descriptive information was provided with regard to mean scores, standard deviations, and the percentage of siblings who fall into the clinical ranges on each standardized distress measure. Information about the percentage of parents who meet criteria for PTSD and the number of families that fall into the “unhealthy” range on the measure of family functioning was also included. Because only 52 families have family functioning data from all three family members (siblings, mothers, and fathers), family mean scores were not calculated; rather, the percentage of families falling into the unhealthy range of family functioning was presented separately for sibling, mother, and father report.

### **2.3.2 Specific Aim 1: Main Effects**

To assess the hypothesis that higher levels of sibling distress are associated with older sibling age, lower levels of parenting acceptance, and higher levels of parental PTSS, parenting psychological control, and family functioning problems, Pearson correlations were calculated between each of these predictors and sibling distress. Then, a series of regression analyses was



carried out to assess the independent effects of each predictor variable on sibling distress. Covariates were entered into the first step of each regression equation, and each predictor variable (sibling age, parental PTSS, family functioning, and parenting) was entered into Step 2 of separate regressions predicting sibling distress.

### **2.3.3 Specific Aim 2: Moderation Effects**

To assess whether family functioning and/or parenting moderates the relationship between parent PTSS and sibling distress, we used multiple regression. Step 1 included the covariates, Step 2 included parental PTSS and family functioning (or parenting) variables, and Step 3 included an interaction term between parent PTSS and family functioning (or parenting). When significant, post-hoc analyses were carried out to determine the nature of the interaction. Separate regression analyses were run for data provided by mothers and fathers.

To examine whether associations of parent PTSS, family functioning, or parenting with sibling distress are moderated by sibling age, we used multiple regression. As above, Step 1 included control variables, Step 2 included sibling age and family functioning (or parent mental health or parenting), and Step 3 included an interaction term between sibling age and family functioning (or parent mental health or parenting). Post-hoc analyses were carried out as necessary.

### **2.3.4 Specific Aim 3: Cumulative Risk**

To examine the utility of employing a cumulative risk framework to sibling distress, a regression analysis was carried out. First, a family cumulative risk index score was calculated for each sibling (Sameroff et al., 1987; Belsky & Fearon, 2002). The four family variables were dichotomized, as follows: parent PTSD was scored 1 if either mother's or father's self-reported PTSD symptoms met criteria for PTSD diagnosis and scored 0 if neither met PTSD criteria; sibling-reported problems with general family functioning was scored 1 if the score fell in the top 20% of the sample and scored 0 if it fell in the bottom 80%; sibling-reported parenting acceptance was averaged across mother and father data, and it was scored 1 if the average acceptance score fell in the bottom 20% and scored 0 if it fell in the top 80%; sibling-reported parenting psychological control also was averaged across mother and father data, and it was scored 1 if the average psychological control score fell in the top 20% and scored 0 if it fell in the bottom 80%. Then, the dichotomized variables were summed to compute the cumulative family risk score, ranging from 0-4. Regression analyses were carried out in which covariates were entered into Step 1, the linear family risk index score was entered into Step 2, and the quadratic family risk index score was entered into Step 3 of a regression equation predicting sibling distress.

### **2.3.5 Specific Aim 4: Effects of Parent Gender**

To assess the exploratory hypothesis that effects of parent PTSS and parenting on sibling distress would be larger for fathers than mothers, we first summarized the pattern of findings and the

strength of associations between sibling distress and mother- versus father-reported PTSS and parenting variables. When sibling distress was significantly associated with mothers' and fathers' reports of the same variable, both parent variables were entered into the same step of a regression equation predicting sibling distress. Then, the strength of the beta coefficients were statistically compared for mothers versus fathers using Fisher's z-tests.

## 2.4 POWER CONSIDERATIONS

Power estimates were carried out using G\*Power (Faul, Erdfelder, Buchner, & Lang, 2009). To test main effects using regression, power analyses were conducted using 5 predictors (the factor under examination plus four covariates). Our sample of mothers ( $n = 187$ ) yields power of .99 to detect a medium effect size ( $f^2 = 0.15$ ) at an alpha of .05. Our sample of fathers ( $n = 71$ ) yields power of .67 to detect a medium effect size at an alpha level of .05.

The power analysis for moderation analyses was based on an effect size reported in a study of family functioning as a moderator of the association between father and child anxiety in a sample of children with cancer (Robinson, et al., 2007). Based on this relatively small effect size ( $f^2 = .049$ ), with an alpha value of .05, using seven predictors (4 covariates plus parent PTSS, family functioning, and the interaction between them), our sample of mothers yields power of .55 to detect moderation effects, and our sample of fathers yields power of .20 to detect these effects. Therefore, we acknowledge that this study lacks sufficient power to examine moderation in our sample of fathers; these analyses are considered exploratory, and effect sizes were reported.

## **3.0 RESULTS**

### **3.1 SAMPLE CHARACTERISTICS**

Data were collected from 210 families. Parent/guardian data were provided by mothers and/or fathers in all but one family, in which grandparents were the respondents. This family was not included in analyses in order to maintain uniformity across respondent type, yielding a final sample of 209 families (209 siblings, 186 mothers, and 70 fathers). See Table 1 for a summary of demographic and illness information.

### **3.2 PRELIMINARY ANALYSES**

#### **3.2.1 Examination of Skew & Kurtosis**

Levels of kurtosis were acceptable for each outcome variable. Sibling-reported scores on measures of depression and posttraumatic stress symptoms were positively skewed. The PTSS distribution was normalized using logarithmic transformation, and the depression distribution was normalized by using the squared inverse of the original scores. Transformed PTSS and

depression scores were used in all analyses and in the formation of the composite distress variable (see below).

### **3.2.2 Data Reduction**

Multiple measures were used to assess sibling distress, and these data were examined for the purpose of reducing the number of outcome constructs. Correlations among sibling-reported symptoms of depression, anxiety, and posttraumatic stress ranged from 0.58-0.72 (Table 2) and therefore were standardized and combined to create an average distress score. For this purpose, a principal components factor analysis with varimax rotation of the three distress scales was conducted. Symptoms of depression (0.85), anxiety (0.91), and posttraumatic stress (0.88) loaded onto a single factor, which accounted for 77% of the variance. Hence, we created a distress scale by averaging the standardized scale scores and equally weighing each of the factors contributing to sibling distress in the calculation of the overall distress scale score.

### **3.2.3 Parent-Sibling Concordance**

The next set of descriptive analyses examined the concordance between sibling- and parent-reported parenting and family constructs. Associations between parent self-reported parenting and child-reported perceptions of parenting acceptance and psychological control scores ranged from  $r = .08$  to  $r = .35$ , depending on the respondent (mother versus father) and parenting subscale (Table 3). Similarly, correlations between mother- and father-reported FAD subscales ranged from  $r = 0.14$  to  $r = 0.54$ , and correlations between sibling and parent report of FAD

subscales ranged from  $r = 0.005$  to  $r = 0.31$  (see Table 4 for FAD subscale correlations between and within respondents). Therefore, mother-, father-, and sibling-reported family functioning data were examined separately.

### 3.2.4 Selecting Covariates

Based on the theoretical model and existing literature, eight potential covariates were examined for their relationship with outcome measures. Four of these variables (time since diagnosis, birth order relative to the child with cancer, presence of additional siblings in the family, and gender) were not significantly associated with the individual outcome measures or the composite distress measure and therefore were not included in subsequent analyses (Tables 2 and 5). Although sibling age was not significantly associated with the outcome measures, this factor was entered as a covariate in all analyses due to its conceptual importance in our developmental model of sibling adjustment.

The other three variables were associated with outcome measures, as follows: lower family income was associated with higher levels of sibling PTSS, anxiety, and overall distress; non-white race was associated with higher levels of sibling PTSS, depression, and overall distress; and siblings whose parents were unmarried had higher levels of PTSS, anxiety, and overall distress. When income, race, and marital status all were entered into the same step of a regression model predicting sibling distress, none of the coefficients were significant (Beta's  $\leq .11$ ,  $p$ 's  $\geq .11$ ), suggesting that the effect on sibling distress is due to the overlapping variance among these three factors. All subsequent analyses controlled for sibling age, family income, race, and marital status.

### 3.2.5 Clinical Picture

One quarter of the sample of siblings met criteria for PTSD, and 62% fell into the moderate to severe range of PTSS reactions (score  $\geq 11$  on the CPSS). In comparison to standardized T-score distributions of borderline (T-score = 60-69) and clinical (T-scores  $\geq 70$ ) levels of symptomatology, the majority of sibling participants in the present study did not endorse clinically significant levels of depression or anxiety (Table 6). With regard to anxiety, the percentage of siblings falling into the borderline range (14%) is similar to that reported in the normative population, but the percentage in the clinical range (5%) is two and a half times higher. The rate of depression scores in the borderline range (5%) is lower than the normative rate, but the rate of siblings endorsing clinical levels of depression (3%) is consistent with that reported in the normative population.

We also characterized rates of parent PTSD and family functioning. In this regard, 35% of mothers and 28% of fathers met criteria for PTSD. With regard to family functioning, 47% of siblings, 26% of mothers, and 38% of fathers endorsed unhealthy levels of family functioning problems on the general functioning subscale of the FAD.

### **3.3 SPECIFIC AIM 1: MAIN EFFECTS**

#### **3.3.1 Correlations**

Pearson correlations were calculated to test our hypotheses that higher levels of sibling distress would be associated with older sibling age, lower parental acceptance, higher parental psychological control, higher levels of parent cancer-related posttraumatic stress, and poorer family functioning. Correlation results show that higher levels of sibling distress are associated with lower levels of sibling-reported perceptions of parental acceptance and higher levels of sibling-reported parental psychological control (Table 3). Greater sibling distress also is associated with lower levels of father-reported parenting acceptance and higher levels of both mother and father self-reported posttraumatic stress. When measures of sibling distress are examined separately, results show that mother PTSS is associated with sibling depression symptoms while father PTSS is associated with sibling PTSS. With regard to family functioning variables, sibling distress is positively associated with sibling-reported problems in all domains of family functioning assessed, as well as with father-reported problems with family roles (Table 4). As discussed previously, sibling age is not directly associated with sibling distress.

#### **3.3.2 Regressions**

Next, a series of linear regressions were carried out to further assess our hypotheses that higher levels of sibling distress would be independently predicted by older sibling age, higher levels of parent PTSS, and poorer patterns of parenting and family functioning. Here, covariates (income,



race, marital status, and sibling age [except for in the sibling age analysis]) were entered into Step 1, and each predictor variable was entered into Step 2 of separate regressions predicting overall sibling distress.

The regression equation examining sibling age as a predictor of sibling-reported distress was not significant ( $\beta = -0.017, \Delta R^2 = 0.00, p = 0.81$ ), nor were those examining the effect of mother-reported PTSS ( $\beta = 0.13, \Delta R^2 = 0.017, p = 0.08$ ) or father-reported PTSS ( $\beta = 0.21, \Delta R^2 = 0.038, p = 0.11$ ) on sibling distress.

Another set of regression analyses examined the association of sibling distress with maternal (self- and sibling-reported) and paternal (self- and sibling-reported) parenting acceptance and psychological control. With regard to mothers, higher sibling distress was predicted by lower levels of child-reported maternal acceptance ( $\beta = -0.17, \Delta R^2 = 0.026, p = 0.02$ ) but not by mothers' self-reported acceptance ( $\beta = 0.035, \Delta R^2 = 0.001, p = 0.64$ ). Similarly, higher sibling distress also was predicted by higher levels of sibling-reported maternal psychological control ( $\beta = 0.21, \Delta R^2 = 0.059, p < 0.001$ ) but not by mothers' self-reported psychological control ( $\beta = 0.047, \Delta R^2 = 0.002, p = 0.55$ ). In reference to fathers, sibling distress was predicted by lower levels of father self-reported acceptance: ( $\beta = -0.25, \Delta R^2 = 0.061, p = 0.05$ ), but results for sibling-reported paternal acceptance were nonsignificant ( $\beta = -0.12, \Delta R^2 = 0.012, p = 0.12$ ). In contrast, greater sibling distress was predicted by higher levels of child-reported paternal psychological control ( $\beta = 0.21, \Delta R^2 = 0.039, p = 0.005$ ) but not by father self-reported psychological control ( $\beta = 0.022, \Delta R^2 = 0.00, p = 0.87$ ).

Next, the association of sibling distress with sibling-, mother-, and father-reported family functioning scales was examined. With regard to *family problem solving*, higher levels of sibling-reported distress were predicted by greater problem-solving difficulties within the family

as reported by the sibling ( $\beta = 0.22, \Delta R^2 = 0.047, p = 0.002$ ) but was not significantly associated with family problem solving as reported by mothers ( $\beta = -0.13, \Delta R^2 = 0.016, p = 0.08$ ) or fathers ( $\beta = -0.025, \Delta R^2 = 0.001, p = 0.85$ ). With regard to *family communication*, higher levels of sibling distress were predicted by more problems with family communication as reported by siblings ( $\beta = 0.28, \Delta R^2 = 0.080, p < 0.001$ ) but was not significantly associated with family problem solving as reported by mothers ( $\beta = -0.072, \Delta R^2 = 0.005, p = 0.33$ ) or fathers ( $\beta = -0.12, \Delta R^2 = 0.013, p = 0.37$ ). With regard to *family roles*, higher levels of sibling distress were predicted by more problems with family roles as reported by siblings ( $\beta = 0.33, \Delta R^2 = 0.12, p < 0.001$ ) and fathers ( $\beta = 0.26, \Delta R^2 = 0.060, p = 0.05$ ) but not by mothers ( $\beta = 0.028, \Delta R^2 = 0.001, p = 0.72$ ). With regard to *family affective involvement*, higher levels of sibling distress were associated with more problems with family affective involvement as reported by siblings ( $\beta = 0.34, \Delta R^2 = 0.11, p < 0.001$ ) but not by mothers ( $\beta = 0.057, \Delta R^2 = 0.003, p = 0.45$ ) or fathers ( $\beta = -0.042, \Delta R^2 = 0.002, p = 0.76$ ). With regard to *family affective responsiveness*, greater sibling distress was predicted by higher levels of problems with affective responsiveness within the family as reported by siblings ( $\beta = 0.17, \Delta R^2 = 0.027, p = 0.02$ ) but not by mothers ( $\beta = -0.12, \Delta R^2 = 0.014, p = 0.11$ ) or fathers ( $\beta = 0.003, \Delta R^2 = 0.00, p = 0.98$ ). Finally, with regard to *general family functioning*, higher levels of sibling distress were predicted by more problems in general family functioning as reported by siblings ( $\beta = 0.40, \Delta R^2 = 0.15, p < 0.001$ ) but not by mothers ( $\beta = 0.004, \Delta R^2 = 0.00, p = 0.96$ ) or fathers ( $\beta = 0.12, \Delta R^2 = 0.014, p = 0.36$ ).

### **3.3.3 Main Effects Summary**

Taken together, initial results suggest that parent PTSS marginally predicts the composite measure of sibling distress, but these associations no longer reached significance once covariates were taken into account. Further, the nature of bivariate associations between parent PTSS and sibling distress were different for mothers versus fathers. While higher levels of mother-reported PTSS was positively associated with sibling-reported depression, higher levels of father-reported PTSS was positively associated with sibling PTSS. With regard to parenting, sibling distress was predicted by higher levels of sibling-perceived mother and father psychological control, lower levels of sibling-perceived mother acceptance, and lower levels of father self-reported acceptance. With regard to family functioning, child perceptions of problems in each domain of family functioning assessed (problem solving, communication, roles, affective involvement and responsiveness, and general functioning) significantly predicted higher levels of sibling distress. Father-reported problems with family roles also predicted sibling distress. Finally, sibling age did not significantly predict sibling distress. Thus, our findings support the hypotheses that sibling distress is related to siblings' perceptions of problems in the family environment and sibling-reported parenting.

## **3.4 SPECIFIC AIM 2: MODERATION EFFECTS**

A series of multiple regression analyses was carried out to assess whether family functioning and/or parenting variables moderate the relationship between parent PTSS and sibling

adjustment. Covariates (income, race, marital status, and sibling age) were entered into Step 1, parent PTSS and each parenting / family functioning moderator was entered into Step 2, and the interaction of parent PTSS and the parenting / family moderator was entered into Step 3 of regressions predicting sibling distress. In this way, separate moderation analyses were carried out for sibling-, mother-, and father-reported parenting acceptance, psychological control, and family functioning (problem-solving, communication, roles, affective involvement, affective responsiveness, and general functioning).

### **3.4.1 Mother PTSS & Sibling Distress**

Findings showed a significant interaction between mother-reported posttraumatic stress and sibling-reported problems with general family functioning in predicting sibling-reported distress (interaction term:  $\beta = 0.14$ ,  $\Delta R^2 = 0.018$ ,  $p = 0.05$ ; Figure 4). Examination of simple slopes suggests that there is a marginally-significant positive association between mother PTSS and sibling distress at higher levels of sibling-reported problems with family functioning ( $p = .07$ ), while the association between mother PTSS and sibling distress is nonsignificant at lower levels of sibling-reported problems with general family functioning, after controlling for sibling age, family income, race, and marital status.

The relationship between mother PTSS and sibling distress also was moderated by mother-reported problems with family problem solving (interaction term:  $\beta = -0.17$ ,  $\Delta R^2 = 0.026$ ,  $p = 0.03$ ; Figure 5) but in the opposite direction to what was hypothesized. In other words, the positive relationship between mother PTSS and sibling distress was significant when mothers reported low levels of problems with family problem solving, but the association between sibling

and mother distress was nonsignificant when mothers reported more problems with family problem solving. Thus, siblings who endorsed the highest levels of distress were those whose mothers reported lower levels of problems with family problem-solving but higher levels of maternal PTSS.

The relationship between maternal PTSS and sibling distress was not significantly moderated by sibling-, mother-, or father-reported parenting acceptance or psychological control ( $\Delta R^2$ 's  $\leq .013$ ,  $p$ 's  $\geq .11$ ), or by sibling-, mother-, or father-reported general family functioning (except where noted above), family problem solving (except where noted above), communication, roles, affective responsiveness, or affective involvement ( $\Delta R^2$ 's  $\leq .018$ ,  $p$ 's  $\geq .06$ ).

### 3.4.2 Father PTSS & Sibling Distress

The relationship between father PTSS and sibling distress was moderated by father self-reported psychological control (interaction term:  $\beta = 0.32$ ,  $\Delta R^2 = 0.074$ ,  $p = 0.03$ ; Figure 6) such that the relationship between father PTSS and sibling distress is positive and significant at higher levels of father self-reported psychological control but not at lower levels of father-reported psychological control. In other words, siblings have the most distress when fathers endorse high levels of both PTSS and psychological control.

The relationship between paternal PTSS and sibling distress was not significantly moderated by sibling-, mother-, or father-reported parenting acceptance or by sibling- or mother-reported psychological control ( $\Delta R^2$ 's  $\leq .027$ ,  $p$ 's  $\geq .18$ ). Similarly, the relationship between paternal PTSS and sibling distress was not significantly moderated by sibling-, mother-, or

father-reported general family functioning, family problem solving, communication, roles, affective responsiveness, or affective involvement ( $\Delta R^2$ 's  $\leq .068$ ,  $p$ 's  $\geq .07$ ).

### **3.4.3 Family Functioning & Parenting Moderation Summary**

Findings suggest an interaction between maternal PTSS and sibling-reported problems with general family functioning in predicting sibling distress such that the impact of maternal PTSS on sibling distress is marginally significant only at higher levels of sibling-reported problems with general family functioning. Contrary to expectations, findings also suggest that the positive association between maternal PTSS and sibling distress is only evident at lower levels of mother-reported problems with family problem solving. Finally, there is an interaction between father-reported PTSS and father self-reported psychological control such that sibling distress is highest when levels of both paternal PTSS and self-reported psychological control are elevated. All other interactions between parent PTSS and parenting or family functioning variables were nonsignificant.

### **3.4.4 Moderation by Sibling Age**

A series of multiple regression analyses was carried out to assess the hypotheses that parent and family variables have a greater influence on the adjustment of adolescent than school-aged siblings. Covariates (income, race, and marital status) were entered into Step 1, the parenting / parent PTSS / family variable and age were entered into Step 2, and the interaction of the

parenting / parent PTSS / family variable and sibling age was entered into Step 3 of regressions predicting sibling distress.

With regard to parenting psychological control, sibling age moderated the relationships between sibling distress and sibling-reported mother psychological control such that the magnitude of this relationship is stronger for older siblings (interaction term:  $\beta = .15$ ,  $\Delta R^2 = 0.020$ ,  $p = 0.04$ ; Figure 7). Sibling age did not significantly moderate the relationships between sibling distress and sibling-reported paternal psychological control, maternal or paternal self-reported psychological control, or sibling-, mother-, or father-reported parenting acceptance ( $\Delta R^2$ 's  $\leq .016$ ,  $p$ 's  $\geq .11$ ). The effects of parent PTSS on sibling distress did not vary according to sibling age (maternal PTSS:  $\Delta R^2 = .018$ ,  $p = .07$ ; paternal PTSS:  $\Delta R^2 = .0048$ ,  $p = .58$ ).

With regard to family functioning, sibling age moderated the relationship between sibling distress and mother-reported problems with family roles such that the magnitude of this relationships was stronger for younger siblings (interaction term:  $\beta = -0.18$ ,  $\Delta R^2 = 0.030$ ,  $p = 0.02$ ; Figure 8). Sibling age did not significantly moderate the relationships between sibling distress and sibling-, mother-, or father-reported general family functioning, problem-solving, communication, roles (except where noted above), affective involvement, or affective responsiveness ( $\Delta R^2$ 's  $\leq .014$ ,  $p$ 's  $\geq .12$ ).

### 3.4.5 Age Moderation Summary

The relationship between sibling-perceived maternal psychological control and sibling distress was stronger for older siblings, while the effects of mother-reported problems with family roles on sibling-reported distress were stronger for younger siblings. Effects of parent PTSS on sibling

distress did not vary as a function of sibling age. All other interactions between sibling age and parent PTSS, parenting, or family functioning variables were nonsignificant.

### 3.5 SPECIFIC AIM 3: CUMULATIVE RISK

The distribution of cumulative family risk scores was as follows: 35.9% of families ( $n = 75$ ) had a family risk score of 0, 37.8% of families ( $n = 79$ ) had a family risk score of 1, 16.3% of families ( $n = 34$ ) had a family risk score of 2, 7.7% of families ( $n = 16$ ) had a family risk score of 3, and 2.4% of families ( $n = 5$ ) had a family risk score of 4. Because of the small number of families with a risk score of 4, siblings with scores of 3 or 4 were combined for all analyses.

The effect of cumulative family risk on sibling distress was assessed with linear regression, in which the covariates (age, income, race, and parent marital status) were entered into Step 1, the family risk score was entered into Step 2, and the square of the family risk score was entered into Step 3. Results showed that the linear family risk score significantly predicted sibling distress ( $\beta = .31, \Delta R^2 = .093, p < .001$ ) when the quadratic term was not included in the model. When the quadratic term was entered into the model, the linear cumulative risk score was no longer significant ( $\beta = -.19, p = .33$ ) and the quadratic family risk score significantly predicted sibling distress ( $\beta = .53, \Delta R^2 = .033, p = .006$ ; Figure 9). This suggests that the strength of the association between cumulative family risk and sibling distress is stronger at higher levels of risk.



### 3.6 SPECIFIC AIM 4: MOTHER VERSUS FATHER EFFECTS

The final set of analyses examined the exploratory hypothesis that sibling adjustment would be more strongly related to father- than mother-reported PTSS and parenting. For this purpose, the pattern of findings and strength of associations between sibling distress and PTSS and parenting variables were examined for mothers and fathers.

With regard to parental cancer-related distress, both mother- and father-reported PTSS were significantly associated with sibling distress in bivariate analyses (Table 3). Follow-up analyses were performed to determine the relative contribution of mother versus father variables on sibling distress. When both mother- and father-reported PTSS were entered into the same step of the regression, the beta values were nonsignificant for both mother- and father-reported PTSS ( $\Delta R^2 = .047$ ,  $p = .32$ ;  $\beta_{\text{mother}} = 0.20$ ,  $p = 0.21$ ;  $\beta_{\text{father}} = 0.085$ ,  $p = 0.63$ ). This suggests that it is the shared variance between mother and father PTSS that accounts for the effect on sibling distress, rather than the unique effects of either parent's PTSS.

With regard to parenting, lower levels of father self-reported acceptance were significantly associated with greater sibling distress; the relationship between mother-reported acceptance and sibling distress was not significant. Sibling reports of parenting variables (acceptance and psychological control) were of similar magnitude for mothers and fathers and were both significantly associated with sibling distress. Follow-up analyses were performed to determine the relative contribution of mother versus father variables. When sibling-reported maternal and paternal psychological control were entered into the same step of a regression equation predicting sibling distress, the magnitude of the effect size appeared to be larger for mothers ( $\Delta R^2 = .061$ ,  $p = .002$ ;  $\beta_{\text{mother}} = 0.20$ ,  $p = 0.03$ ;  $\beta_{\text{father}} = 0.072$ ,  $p = 0.45$ ). However, when

coefficients were compared statistically, the difference between sibling-reported maternal and paternal psychological control was nonsignificant (Fisher's  $z = .91$ ,  $p = .18$ ). Similar results were found when sibling-reported mother and father parenting acceptance were entered into the same step of a regression equation ( $\Delta R^2 = .032$ ,  $p = .04$ ;  $\beta_{\text{mother}} = -0.15$ ,  $p = 0.05$ ;  $\beta_{\text{father}} = -0.070$ ,  $p = 0.38$ ). Again, the difference between the coefficients for sibling-reported maternal and paternal parenting acceptance was nonsignificant (Fisher's  $z = .55$ ,  $p = .29$ ). Together, these results suggest that the associations of sibling-reported parenting variables and sibling distress are similar for mothers and fathers.

#### 4.0 DISCUSSION

The current research employed a family systems framework to examine the role of family-related risk and protective factors on sibling adjustment to a brother's or sister's childhood cancer diagnosis. Given conceptual and empirical overlap, the three sibling-reported distress outcomes (symptoms of anxiety, depression, and posttraumatic stress) were combined to create one composite measure of sibling distress. Overall, findings suggested that siblings' perceptions of poorer family functioning and parenting were associated with higher scores on this composite distress measure. Although initial analyses also showed a positive association of sibling distress with mother and father PTSS, these associations did not withstand adjustment for sibling age, race, family income, and parents' marital status, which may have an important influence on both sibling and parent distress. Moreover, the relationship between sibling-perceived parent psychological control and sibling distress was stronger for older siblings, while the effects of mother-reported problems with family roles on sibling-reported distress were stronger for younger siblings. When family risk factors were considered together, there was only limited support for our hypothesis that a more positive family environment would buffer the negative effects of parent PTSS on sibling distress. However, the present findings offered preliminary support for a quadratic cumulative risk model in which higher levels of cumulative family risk were disproportionately associated with elevated sibling distress.

#### 4.1 CLINICAL PICTURE

Consistent with past research (Alderfer et al., 2010), the majority of sibling participants did not endorse clinically significant levels of depression or anxiety. With regard to anxiety, the percentage of siblings falling into the borderline range (14%) is similar to that reported in the normative population, but the percentage in the clinical range is two and a half times higher (5%). The percentage of siblings endorsing levels of depression symptoms in the borderline range (5%) is lower than normative rates, while the percentage in the clinical range (3%) is similar to that reported in the normative population. In contrast, 62% of siblings in the present study endorsed moderate to severe levels of PTSS, and one quarter met criteria for PTSD. These rates are higher than those reported in previous studies (Alderfer et al., 2003; Packman et al., 2004), which may reflect differences regarding longer time since diagnosis and off-treatment status. Rates of clinically-significant PTSS in the current sibling sample are higher than those reported in pediatric cancer survivors, estimated to be 4.7-21% (Bruce, 2006), and rates of PTSD diagnosis are considerably higher than the lifetime prevalence of PTSD in the United States, estimated to be 7-8% (Keane, Marshall, & Taft, 2006).

This clinical picture suggests that a subset of siblings may be at risk for anxiety-related reactions to a brother's or sister's cancer, particularly PTSD, and underscores the importance of examining sibling functioning using relevant outcome measures. In this regard, Alderfer and colleagues (2010) recommended that future work with siblings of children with cancer focus less on comparing rates of anxiety or depression between siblings of children with cancer and comparisons, and instead, focus more on identifying and characterizing relevant emotional

and/or developmental outcomes such as posttraumatic stress, quality of life, or siblings' course of social or emotional development over time.

Although the focus of our study is on sibling functioning, we also report on rates of parent PTSD and unhealthy family functioning. In our sample, 20 of the 71 fathers (28%) and 66 of the 187 mothers (35%) met criteria for PTSD, which is similar to previously reported rates of parental PTSD in the context of childhood cancer. In a review of the literature, Bruce (2006) reported that the incidence of parental cancer-related PTSD diagnoses ranges from 6.2 to 25%, with lifetime prevalence rates ranging from 27 to 54%. With regard to family functioning, 47% of siblings, 26% of mothers, and 38% of fathers endorsed clinical levels of family functioning problems. Findings that siblings report more family functioning problems than parents are consistent with previous findings comparing rates of unhealthy functioning as reported by patients versus parents (Alderfer et al., 2008).

## **4.2 MAIN EFFECTS**

### **4.2.1 Family Functioning**

Reports of family functioning varied widely according to respondent, with mother-father correlations ranging from 0.14 to 0.54 across family functioning subscales, and parent-sibling correlations ranging from 0.005 to 0.31 across subscales. These levels of parent concordance are similar to those reported in a review of family assessment measures that are frequently employed in studies of pediatric populations, which reported mother-father concordance levels ranging

from .24 to .53 across FAD subscales (Alderfer et al., 2008). To our knowledge, there are no published studies examining concordance between parents' and siblings' perceptions of family functioning problems.

In general, greater sibling distress was predicted by more sibling-reported family functioning problems across subscales, but not by mother- or father-reported family functioning problems. One exception is that higher levels of sibling distress were associated with more father-reported problems with family roles.

Links between family functioning and sibling adjustment have not been addressed previously in the context of cancer. However, similar findings have been reported in siblings of children with other childhood chronic health conditions. For example, a study of 65 African-American siblings of children with sickle cell disease found that parent-reported sibling internalizing and externalizing were positively correlated with parent-reported family support and expressiveness and were negatively correlated with family conflict (Gold, Treadwell, Weissman, & Vichinsky, 2008). This study operationalized "support" as the extent to which family members offer caring, affection, kindness and assistance, which is comparable to the affective responsiveness construct in the present study. Associations between family and sibling functioning also have been examined in the context of childhood disability. A study of siblings of children with disabilities (N = 49, age 7-16) showed that sibling adjustment, as assessed by the Strengths and Difficulties Questionnaire (Harter, 1985), was predicted by higher levels of mother-reported family routines, problem-solving, communication, and hardiness, which the authors defined as perceived control over managing stressful family situations (Giallo & Gavidia-Payne, 2006). Similarly, parent-reported social competence of siblings of children with

Down Syndrome (N = 41, age 7-18) was predicted by better mother-reported problem-solving communication (van Riper, 2000).

Taken together, the present findings have some similarity to past literature demonstrating the importance of family functioning in predicting sibling adjustment to childhood chronic health conditions and disabilities. However, there are some important differences. Unlike the present findings, previous work demonstrated links between *parent*-reported family functioning and sibling functioning. It is important to note that these past studies used parent measures of sibling functioning (Gold et al., 2008; van Riper, 2000) or employed a non-psychopathology measure of siblings' self-reported adjustment (Giallo & Gavidia-Payne, 2006). Thus, the significant associations between parent-reported family functioning and sibling adjustment reported in past studies may reflect respondent bias or may suggest that more nuanced measures of sibling functioning, rather than psychopathology symptom scales, are more relevant for this population.

In the context of cancer, the present findings are consistent with cross-sectional and prospective work showing that lower distress in children and adolescents with cancer is associated with positive aspects of family functioning including better family relationship quality and problem-solving skills and higher levels of affective involvement and responsiveness (Alderfer et al., 2009; Foley et al., 2000; Fuemmeler et al., 2003; Morris et al., 1997; Noojin et al., 1999; Sawyer et al., 1997, 1998, 2000). Unlike the present findings, however, previously reported effects on child functioning are similar across parent- and patient-reported family functioning. This may reflect differences in patients' versus siblings' perceptions of the family environment, or it may reflect objective differences in the quality of the family environment as experienced by siblings versus patients or parents during cancer treatment.

Current findings that sibling distress is predicted by self- but not parent-reported family functioning can be explained in multiple ways. One possibility is that the FAD measure taps into *perceptions* of family functioning as opposed to objective information, and in turn, siblings' own perceptions of the family may overlap with their subjective reports of distress to a greater extent than parental perceptions of family functioning. Similarly, more distressed siblings may perceive more problems with their family environment, which raises questions about respondent bias and direction of effect. Alternately, associations between sibling distress and family functioning difficulties may reflect a third variable, which may influence both the quality of the siblings' family environment and their own subjective experiences of distress.

It is also possible that well-documented disruptions in family life following a childhood cancer diagnosis may reduce the extent to which family members can accurately report on the nature and quality of *current* family functioning. This may be especially true for mothers, who often assume the role of medical caretaker for the child with cancer and spend a great deal of time in the hospital or clinic, and therefore may be less attuned to sibling or family functioning. Indeed, the finding that father-reported roles are associated with sibling distress may be particularly important in the context of childhood cancer, given the realignment of family roles and responsibilities that often places fathers in the role of primary sibling caretaker (Long & Marsland, 2011).

In addition to main effects, the current study found that sibling age moderates the relationship between higher levels of mother-reported problems with family roles and elevated sibling distress such that the magnitude of this relationship is stronger for younger siblings. This pattern of results may reflect children's preference for structure with regard to family roles and



routines, or their increased reliance on the family rather than the peer group, as compared to their adolescent sibling counterparts.

#### **4.2.2 Parenting**

The current findings showed that greater sibling distress was predicted by higher levels of sibling-reported maternal and paternal psychological control. This relationship was moderated by sibling age such that the strength of the positive association between sibling-reported mother psychological control and sibling distress was greater for older siblings. Higher levels of sibling distress also were associated with lower levels of sibling-reported maternal acceptance and self-reported paternal acceptance. Sibling age did not moderate relationships between parenting acceptance and sibling distress.

These findings are consistent with the body of work linking higher levels of parenting psychological control (or lower levels of autonomy-granting) to more internalizing symptoms in children and adolescents (e.g., Gray & Steinberg, 1999; McLeod et al., 2007; Papp et al., 2005; Rogers et al., 2003), including those with cancer (Morris et al., 1997). It is not surprising that the strength of this association changes as a function of age. Higher levels of parenting psychological control are at odds with adolescents' increasingly independent attempts to regulate their emotional states and behaviors (Steinberg, 2005), and this developmental mismatch may be a source of distress for adolescents. Alternately, it is also possible that adolescents who are experiencing higher levels of distress may perceive more parenting psychological control. As with associations between sibling distress and perceptions of family functioning discussed above,

it is also possible that siblings' subjective distress and perceptions of parenting may be related due to their shared association with a third variable.

Although our interpretation is limited by the cross-sectional nature of our analyses, it is possible that effects of parenting on sibling functioning are longstanding and reflect the cumulative effects of early childhood relationships building up over time. Siblings' perceptions of parents' general patterns of acceptance and psychological control may serve as the backdrop against which they adapt to cancer-related family stressors. For example, siblings' beliefs about parental relationships may influence their interpretations of and reactions to cancer-related parenting changes, such as well documented increases in differential treatment in favor of the child with cancer (Chao et al., 2003; Quinn, 2004; van Dongen-Melman et al., 1998). Siblings who perceive higher levels of parenting acceptance may be more likely to interpret the preferential treatment as fair and therefore adjust more positively (Kowal, Kramer, Krull, & Crick, 2002). Similarly, parents with higher levels of warmth and acceptance are more likely to engage in emotion coaching, which may contribute to siblings' abilities to cope with emotional aspects of the cancer diagnosis and build more sophisticated emotion regulation skills. In contrast, siblings with a history of interpersonal conflict with parents may have more difficulty adapting to cancer-related parenting changes. Longstanding poor parent-child relationships may make parents less likely to engage in emotion coaching and may make siblings less receptive to parental involvement, which may increase the likelihood of sibling distress in the context of their brother's or sister's illness.

### 4.2.3 Parent PTSS

Mother and father PTSS were significantly associated with each other ( $r = 0.33$ ), and both were significantly correlated with sibling distress in bivariate analyses ( $r$ 's = .15 and .23, respectively). When examining the individual sibling distress indices, higher levels of mother-reported PTSS were positively associated with sibling-reported depression, while higher levels of father-reported PTSS were positively associated with sibling PTSS. When covariates (sibling age and race, family income, and marital status) were taken into account, the association of parental PTSS with the combined measure of sibling distress was no longer significant.

Concordance between parent and sibling PTSS has not been examined previously in the context of childhood cancer. Our findings are consistent with some previous work suggesting that PTSS is more likely to be associated between a mother and a father than between a parent and a cancer survivor (Taieb et al., 2003). However, we use some caution when comparing the present findings to previous work in children or adolescents with cancer, since the pathways to PTSS reactions are likely to be different in sibling versus patient populations. It may be particularly difficult for parents with higher levels of PTSS to address the emotional and practical needs of siblings, which may be perceived as less urgent than those of the child with cancer and with whom they may spend less time while the child with cancer is on active treatment. Other differences between patients and siblings may include knowledge about the illness, treatment, and prognosis and accessibility to mental health professionals. In this regard, hospital staff often are routinely available to help patients process illness-related fears and to answer questions, but they generally are unavailable to siblings. Therefore, siblings may have less of an opportunity to talk about the cancer with either parents or hospital-based mental health

professionals, thereby limiting the degree to which they can process cancer-related emotions and clarify misinformation. This, in turn, may lead to siblings' increased risk for elevated PTSS.

### 4.3 INTERACTIONS

Findings showed that the relationship between father-reported PTSS and sibling distress was buffered by lower levels of father self-reported psychological control, with the interaction between father PTSS and father psychological control accounting for 7.4% of the variance in sibling functioning. In other words, the association between father and sibling distress is more pronounced at high levels of father-reported psychological control, and siblings who are high on both measures are the most likely to endorse elevated distress.

Second, the effect of mother PTSS on sibling distress differed as a function of sibling-reported problems with general family functioning such that the positive association between maternal PTSS and sibling distress was marginally significant only at higher levels of sibling-reported problems with general family functioning. These findings suggest that siblings are more likely to endorse higher levels of distress in the presence of *both* elevated maternal PTSS and perceptions of poorer family functioning. The effect size was smaller than that expected, with the interaction between maternal PTSS and family functioning accounting for 1.8% of the variance in sibling distress. Although a different domain of family functioning was assessed, the present results are similar to those reported in a cross-sectional study of 8- to 15-year-olds with cancer, where patients from families endorsing more positive interpersonal relationships showed lower

anxiety in the presence of father anxiety compared to patients in families with less positive interpersonal relationships (Robinson, et al., 2007).

Finally, the relationship between mother PTSS and sibling distress differed according to mother-reported problems with family problem solving. Contrary to expectations, the positive association between maternal PTSS and sibling distress was evident only at *low* levels of mother-reported problems with family problem-solving. Again, the effect size was small, with the interaction term accounting for less than 3% of the variance in sibling distress.

Taken together, the findings from the set of analyses examining interactions among parent and family variables in predicting sibling distress revealed fewer significant effects and smaller effect sizes than what was expected. Indeed, the three significant findings were accompanied by dozens of nonsignificant moderation analyses. Low power likely contributed to this pattern of null findings, particularly for fathers. However, the small effect sizes suggest that the strength of the moderation findings are relatively low, and it is possible that the few significant effects may reflect Type I error. This approach may have been too narrow by considering only two aspects of the family environment at any given time rather than assessing the role of context more broadly. Therefore, we also analyzed the data using the cumulative risk approach.

#### **4.4 CUMULATIVE RISK**

Given individual associations of sibling-reported parenting and family functioning variables with sibling distress, we also examined how these family factors influence sibling functioning in

combination. Our findings provide initial support for a cumulative family risk model of sibling adjustment. In these analyses, family risks included having one or both parents with a cancer-related PTSD diagnosis, scoring in the top 20% for sibling-reported problems in general family functioning, scoring in the top 20% for sibling-reported parenting psychological control, and scoring in the bottom 20% for sibling-reported acceptance. Results showed that siblings' self-reported distress remained near or below the mean when siblings reported zero, one, or two family risks. However, siblings who reported three or four family risks showed a disproportionate increase in self-reported distress symptoms, thereby suggesting that the presence of multiple family risks has a synergistic effect on sibling distress. In these families, it is possible that the stressors associated with a child's cancer diagnosis challenged the adaptational capabilities of the family unit, with effects of poor family adaptation extending to siblings.

Within the developmental psychopathology framework, the idea that child functioning is better predicted by a higher number of nonspecific risk factors, rather than any one particular risk factor, has its roots in the seminal "Isle of Wight" studies (Rutter, Tizard, Yule, Graham, & Whitmore, 1976) and later was expanded in the Rochester Longitudinal Study (e.g., Sameroff et al., 1987). Since then, the approach of calculating a multiple risk score by summing dichotomized risk variables has been applied widely, with higher risk scores associated with lower intelligence (Sameroff, Seifer, Barocas, Zax, & Greenspan, 1987) and the development of internalizing and externalizing problems (Forehand, Biggar, & Kotchick, 1998). This approach also has been applied to intervention research, with findings showing that children from families with higher levels of cumulative risk derive greater benefit from the Head Start intervention (Hubbs-Tait, Culp, Culp, & Miller, 2002). In pediatric psychology, higher levels of cumulative

risk were shown to predict burden among families of children with traumatic brain injuries (Josie et al., 2008), and higher scores on a cumulative risk index including cultural, socio-contextual, and asthma-specific risks were associated with increased asthma morbidity in urban children (Koinis-Mitchell et al., 2007).

Although a cumulative or multiple risk model has not been applied to siblings, previous work from our laboratory has applied a contextual threat model to examining adjustment among siblings of children with cancer (Long, Alderfer, Ewing, & Marsland, In Press). Broadly based on Brown and Harris's (1978) work, contextual threat was defined as an aggregate measure of objective stress surrounding the cancer experience. Rather than focusing on discreet predictors, contextual threat encompasses a set of circumstances that are considered as a whole given the likelihood that they act synergistically to predict functioning. Siblings who were experiencing more concurrent stressors in addition to the cancer, along with fewer resources to help them cope with these stressors, were assigned higher contextual threat ratings. Findings confirmed that higher contextual threat predicted greater sibling distress independent of the intensity of the ill child's treatment or demographic variables (Long et al., In Press). Despite methodological differences in the contextual threat and cumulative risk approaches, both sets of findings underscore the importance of considering the sibling experience in the context of additional risk and protective factors that may or may not be related to the cancer experience.

#### 4.5 PARENT GENDER

An exploratory aim involved characterizing the effects of maternal versus paternal PTSS and parenting on sibling distress. In general, effects of parent-reported variables on sibling functioning were similar between mothers and fathers. Both mother- and father-reported PTSS were positively associated with the combined measure of sibling distress in bivariate analyses, but these results did not withstand adjustment for covariates. The effect sizes appeared larger for paternal than for maternal PTSS across both bivariate and regression analyses. When mother and father PTSS were considered together, neither uniquely contributed to the variance in sibling distress, likely due to the shared variance within parent pairs and between parent PTSS and demographic factors. Thus, our results suggest that both mother and father PTSS contribute to sibling distress, but not uniquely. With regard to parenting, fathers' self-reported acceptance appeared to be more strongly related to sibling distress than mothers' self-reported acceptance, but the effect sizes of sibling-reported parental acceptance and psychological control were similar between mothers and for fathers. Together, findings suggest that effect sizes generally were similar for mothers and fathers.

Our results differ from findings previously reported in the literature examining associations between parent and child depression, which suggests that maternal and paternal depression contribute independently to a child's risk for various types of psychopathology (for review, see Goodman, 2007). This is not surprising, given that our measure of parent mental health assesses cancer-related PTSS and not a depression diagnosis. Indeed, it is likely that our findings reflect overlap in parents' perceptions of stress surrounding the cancer experience,



which may influence siblings' functioning directly or indirectly through effects on the marital or parenting relationships or the family climate.

In the context of childhood cancer, the majority of work examining concordance between parent and patient distress has enrolled primarily or exclusively mothers (Jobe-Shields et al., 2009; Mulhern et al., 1992; Trask et al., 2003). The one study which examined mothers and fathers separately did not find main effects for positive associations of patient distress with either mother or father distress (Robinson et al., 2007). However, as discussed previously, it is difficult to generalize findings to siblings given qualitative reports that siblings tend to spend more time with fathers while mothers are invested in treatment of child with cancer (Jones & Neil-Urban, 2003; McGrath, et al., 2005) as well as differences in the meaning ascribed to the "parent" role when a child has a potentially life-threatening illness.

We interpret findings comparing maternal and paternal influences on sibling functioning cautiously. First, it is not clear whether or not our sample of fathers is representative of the population of fathers of children with cancer. The first phase of data collection enrolled one parent per family, and therefore, these fathers were likely to have a caretaking role in their families. Although the second phase of data collection attempted to enroll both parents whenever possible, this approach was limited by practical factors such as family structure (e.g., unmarried, separated, divorced) or work demands. Thus, our sample of fathers is likely to come from more traditional family contexts and play a larger role in childrearing responsibilities than those fathers who did not participate. Second, the sample of fathers is considerably smaller than the sample of mothers ( $N$ 's = 71 and 187, respectively), which provides decreased power for analyses involving father-reported variables.

#### 4.6 DEMOGRAPHIC & ILLNESS-RELATED FACTORS

During preliminary analyses evaluating potential covariates, we examined demographic and illness-related variables that might influence sibling adjustment to a brother's or sister's cancer diagnosis. Family income, race, and marital status each were associated with sibling distress. When entered together into a regression model predicting sibling distress, the coefficients were nonsignificant for all three of these demographic factors, therefore suggesting that the association with sibling distress was due to overlapping variance. These variables have received surprisingly limited attention in the sibling literature, with a recent systematic review reporting that none of the 37 quantitative studies included information on race, ethnicity, or socioeconomic status (Alderfer et al., 2010). However, these variables have been studied more extensively in the developmental psychopathology literature.

By and large, family income has been shown to have an inverse relationship with levels of childhood psychopathology when assessed alone or as part of composite indices of socioeconomic status (e.g., Beesdo, Knappe, & Pine, 2009; Grant, Compas, Stuhlmacher, Thurm, McMahon, & Halpert, 2003). Results for race are less straightforward. Consistent with the current findings, large-scale epidemiological studies and meta-analyses suggest that race does not independently account for differences in rates of pediatric diagnoses including depression or PTSD (Roberts, Roberts, & Chen, 1997; Shannon, Lonigan, Finch, & Taylor, 1994). However, race is highly correlated with other risk factors such as income and neighborhood disadvantage and has been linked to higher levels of cumulative stress (Chyu & Upchurch, 2011). In the context of childhood disability, a small body of work has demonstrated that 8- to 15-year-old Latino siblings of children with intellectual disabilities have higher rates of internalizing

symptoms and negative emotions and worse personal adjustment than matched controls, independent of income (Lobato et al., 2011).

The current findings also showed that siblings of unmarried parents endorsed higher levels of distress. Although single parenthood has been associated with academic and behavioral problems in youth, more recent work has accounted for the fact that single parenthood is confounded with lower income and African American race. For example, work by Dunifon and Kowaleski-Jones (2002) demonstrated differences in outcomes across racial groups such that single parenthood was not a risk factor for lower math scores or delinquency in African American children. It is not clear if the same mechanisms account for associations between marital status and distress in siblings of children with chronic illnesses. For example, documented decreases in the amount of time spent with mothers in single- versus married-parent families (21 versus 31 hours per week, respectively; Sandberg & Hofferth, 2001) may be particularly relevant for siblings of children with cancer, given the time demands of coordinating a child's cancer treatment. In general, the small body of descriptive work addressing differences among married and unmarried parents of children with cancer suggests that single parents endorse lower levels of support and perceive greater burden regarding medical treatment, financial security, employment demands, and family management compared to their married counterparts (Long & Marsland, 2011).

Sibling age, birth order relative to the child with cancer, presence of additional siblings in the family, and gender were not significantly associated with the individual outcome measures or the composite measure of sibling distress. In general, the existing studies of sibling functioning that have considered the role of demographic factors have yielded inconsistent findings (Alderfer et al., 2010). Our results agree with findings of a recent meta-analysis which reported that sibling

distress does not vary by sibling age, gender, or birth order relative to the child with the chronic health condition (Vermaes et al., 2012). Given the theoretical importance of sibling age and developmental level on sibling adjustment to a brother's or sister's chronic illness, the absence of main effects for sibling age as a predictor of sibling distress in the present sample was surprising. However, the role of parenting psychological control and problems with family roles varied as a function of sibling age, underscoring the importance of attending to sibling development when examining contextual factors.

Finally, time since diagnosis was not associated with sibling distress. The extant literature examining time since diagnosis has reported mixed results (Alderfer et al., 2010), with some studies finding that relationships between time since diagnosis and sibling functioning were nonsignificant (e.g., Houtzager, Grootenhuis, Hoekstra-Weebers, Caron, & Last, 2003).

#### **4.7 STRENGTHS & LIMITATIONS**

Grounded in the social ecology, family systems, and developmental psychopathology frameworks, this research is a first step toward testing more sophisticated models of sibling adjustment that include multiple levels of influence acting reciprocally over time. By examining risk and protective factors likely to influence sibling adjustment, we go beyond the existing literature which focuses on identifying mean differences in psychopathology symptoms between siblings and controls. To our knowledge, this is the first study to examine whether sibling distress is influenced by parent mental health, parenting, or family functioning, alone or in

combination. Further, it is the first to apply a cumulative risk framework to siblings of children with a chronic health condition.

This work also extends the literature by considering the role of father-reported mental health, family functioning, and parenting on sibling outcomes. Lack of father data is a pervasive problem in the fields of developmental and pediatric psychology (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005), and fathers may be particularly relevant for siblings in light of qualitative findings that fathers assume increased caretaking responsibilities for siblings while mothers spend time at the hospital caring for the children with cancer (Chesler & Parry, 2001; Clarke, 2006; Kars, et al., 2008; Martinson, et al., 1999; McGrath, et al., 2005; Reay, et al., 1998; Yeh, et al., 2000; Young, et al., 2002).

From a methodological standpoint, our sample of siblings was larger than those employed in past studies, thereby enabling us to address preliminary questions regarding the ways in which developmental and contextual factors interact to influence sibling adjustment. A systematic recruitment strategy ensured that all eligible families were invited to participate, and efforts to contact eligible families via multiple methods (mail and telephone) contributed to high response rates (Phase 1 = 75%, Phase 2 = 81%). Data collection within participants' homes allowed us to assess sibling functioning in a context that is more typical than the hospital setting, which siblings may associate with cancer-related stressors. Finally, multiple informants provided information regarding family and parenting factors, which gave us the opportunity to assess inter-rater agreement as well as to examine the relationship of parent- versus sibling-perceived parenting and family functioning variables with sibling distress.

Despite these strengths, this research also has conceptual and methodological limitations. Most fundamentally, the cross-sectional design limits the extent to which we can determine

direction of effects or assess changes in sibling functioning over time. As a result, questions of timing, direction, and continuity remain unaddressed. Similarly, we do not have data on family, parent, or sibling functioning prior to the cancer diagnosis, which limits our ability to disentangle effects directly or indirectly related to the cancer experience from those that reflect more typical developmental processes. Also, our multi-factorial approach to conceptualization, coupled with the availability of data from multiple sources for each family, resulted in a large number of analyses. Therefore, it is possible that some of the findings reported here may reflect Type I error.

With regard to the sample, sibling participants span a large age range (8-18), over which developmental competencies and siblings' roles within the family are likely to differ. Our sample of fathers is relatively small, which limits our power to detect small or medium sized effects. This is particularly true for the moderation analyses, which had low power to detect findings related to father-reported constructs (power = .20) and modest power to detect changes related to mother-reported constructs (power = .55). Also, we do not have demographic or baseline functioning data on families who chose not to participate in this research, and therefore, we do not know whether our sample is representative of the larger population of families receiving treatment for childhood cancer. However, the large number of Caucasian families and the relatively high income and education levels raises questions about the extent to which this sample is representative. Potential implications of a non-representative sample include the possibility of not adequately considering economic or cultural differences in how families adapt to a childhood chronic illness.

With regard to treatment-related variables, our sample includes families currently on active treatment and/or within two years of diagnosis, but we do not have data on treatment

status for each family. The qualitative literature suggests that families realign to meet the needs of cancer treatment, and therefore, it is possible that the role of parent and family factors on siblings may differ systematically based on whether or not the child with cancer is receiving active treatment. This was not addressed in the current work.

This research has conceptual limitations as well. On a basic level, research involving families is limited by the lack of a precise definition of “family” that is relevant for all participants. For example, many cultures consider extended family and non-blood-related “relatives” in conceptual and practical definitions of family. Therefore, research findings based on the nuclear family may not generalize across cultures. Similarly, the idea of family functioning is a broad, multi-factorial concept with multiple overlapping domains. The extant research is difficult to integrate due to the lack of precise definitions for various aspects of family functioning as well as the lack of a generally agreed upon nomenclature for family functioning terms. This ambiguity raises questions about whether or not the present study is addressing the “right” measure of the “right” construct. Further, one of the more consistent findings from the pediatric literature is that high levels of family conflict are associated with elevated distress in individual family members. However, the measure of family functioning employed in this study does not include a conflict subscale.

The developmental psychopathology literature emphasizes the role of multiple individual and contextual influences on child development and adjustment (Boyce, et al., 1998). Although the present work focuses on several promising contextual factors, we acknowledge that there are additional influences on sibling adjustment to cancer that are not considered. For example, the bidirectional, interactive processes linking parenting and child temperament have been well established (Collins, et al., 2000). Asking questions about the role of parenting in sibling

adjustment, without concurrently considering sibling temperament, is likely to underestimate the variance accounted for by parenting. Similarly, it is difficult to adequately assess sibling adjustment to cancer-related stressors without also considering their emotion regulation abilities.

From a social ecology standpoint, parenting and family functioning are likely to interact with other contexts of child development to impact sibling adjustment. For example, parents play a significant role in determining the quality and extent of peer influence through effects on friend selection and behavioral monitoring (Collins, et al., 2000), and this process may be disrupted by a childhood cancer diagnosis. Also, siblings' support from schools may be particularly important when the family is less available emotionally or physically during intense treatment periods. Thus, the exclusion of other individual and contextual factors that are likely to interact with family factors is a limitation of this work.

#### **4.8 FUTURE DIRECTIONS**

The present findings represent a first step in characterizing the association of family risk factors and sibling distress. Our understanding of these associations could be enhanced by studying sibling and family factors longitudinally, which would allow us to make stronger conclusions about the direction of effects and to better understand the role of developmental processes in sibling adjustment to cancer-related stressors. A prospective design would inform trajectories of sibling functioning after a brother's or sister's cancer diagnosis. Although anecdotal reports suggest that the cancer experience influences siblings' life choices and self-concept into



adulthood, there are no empirical data to support or refute these reports or to characterize the percentage of siblings following different pathways of adjustment.

To build on the current findings, future work also could consider the role of treatment status when examining the family environment and parent-sibling relationships. Given qualitative reports suggesting that intensive treatment periods are associated with increased family disruption, it is possible that family effects on siblings could be more pronounced during these times. Similarly, the marked discrepancies between parents' and siblings' reports of family functioning variables in the present study suggest that we were measuring family members' perceptions of how the family functions as a unit. Although this is valuable in its own right, it does not allow us to form conclusions about the role of more objective patterns of family functioning. Therefore, future work may benefit from teasing apart the influence of objective versus subjective information about family functioning, which would inform both etiology and targets of intervention (e.g., a cognitive approach to modifying siblings' beliefs about the family versus a family systems approach to encourage more functional patterns of interaction).

Our understanding of at-risk siblings would be enhanced by expanding the cumulative risk model to include other potential risk factors for sibling adjustment difficulties, such as limited peer support or heightened neighborhood risk. Along these lines, future work should aim to recruit a more heterogeneous sample with regard to race and socioeconomic status, in whom the nature or strength of the relationships identified in the present work may differ. For example, non-Caucasian families and those with lower socioeconomic status have higher levels of cumulative risk (Koinis-Mitchell et al., 2008), suggesting that siblings from these groups may endorse elevated distress in the context of their brother's or sister's diagnosis. Further, a sibling's cultural background is likely to influence patterns of family roles and relationships, which in turn

may change the meaning ascribed to having an ill brother or sister and result in greater sibling caretaking responsibilities. Considerable differences in how “family” and “illness” are conceptualized across cultures underscore the importance of testing our models and future interventions across different groups.

As mentioned above, future work should examine pathways to sibling adjustment in order to identify at-risk siblings and inform the development of culturally-sensitive interventions. Future research should test the hypothesis that siblings who have higher levels of family risk would benefit more from intervention. Given the considerable difficulties regarding feasibility of sibling participation in interventions, it also will be important to identify barriers and to develop and test creative approaches to implementing interventions that are both consistent with a cumulative family risk model and sensitive to the many stressors faced by families of children with cancer, including significant amounts of time spent out of the home or physically apart from the sibling.

#### **4.9 CLINICAL IMPLICATIONS**

Our findings lay the groundwork for understanding the role of the family in different trajectories of sibling adjustment to a brother’s or sister’s cancer diagnosis. This is in line with our larger goal of being able to identify at-risk siblings and design sibling- or family-focused interventions that are sensitive to the unique needs of families who are confronting the considerable emotional and practical challenges associated with childhood cancer. More specifically, the present findings underscore the importance of screening families for both the quantity and severity of

family risk factors, including poor family functioning and high levels of parenting psychological control. Family-focused interventions should be recommended to families who score highly on multiple indices of risk. At present, there are no empirically grounded interventions that focus on sibling adjustment. Although the present findings suggest that multiple family risks are a promising marker of the need for intervention, more research is needed to elucidate the mechanisms linking cumulative family stress to sibling distress and to identify appropriate targets of treatment. This could include improving family problem-solving, communication, affective regulation, or parenting skills, or providing siblings with coping skills training. In addition, it will be important to tailor interventions to siblings' developmental level.

We acknowledge that delivery of services to families and siblings presents considerable practical challenges, including the absence of siblings from the hospital setting and the absence of parents from the home setting. As a first step, it will be important to educate parents and medical professionals about the effects of childhood cancer on siblings, including the elevated rates of PTSS and PTSD in this population, and to help them problem-solve ways of overcoming barriers to providing siblings with mental health evaluations or treatment services.

#### **4.10 CONCLUSIONS**

Taken together, the literature on siblings of children with cancer suggests that this group experiences strong emotional reactions to a brother's or sister's cancer diagnosis, which may place them at higher risk for ongoing elevations in distress. Frequent mixed findings suggest that sibling adjustment may be influenced by factors other than the cancer diagnosis per se, but this is

not adequately addressed in the extant literature. The developmental psychopathology and social ecology frameworks indicate that models of sibling adjustment should consider the family system in which the sibling is developing. Indeed, the present findings suggest that siblings' perceptions of poorer family functioning and parenting are associated with higher self-reported distress, with the importance of some factors (e.g., maternal psychological control) increasing as siblings move into adolescence. Further, our findings are consistent with a cumulative model of family risk, in which sibling distress increases disproportionately in the presence of multiple family risks. Although the cross-sectional nature of this work limits our ability to determine direction of effect or to elucidate changes over time, we believe that this is an important first step in investigating the relationship between developmental and family-level factors and sibling adjustment to childhood cancer.

Table 1: Demographic & Illness Data

<b>Sibling:</b>	
Sibling Age (Years)	<i>M(SD)</i> = 12.52(2.67); <i>Range</i> = 8.08 to 18.00
Sibling Gender	54.8% Female (n = 114)
Sibling Relative Birth Order	35.3% Younger than the Child with Cancer (n = 73)
<b>Family:</b>	
Number of People in the Household	<i>M(SD)</i> = 5.04(1.34); <i>Range</i> = 3 to 13
Mother Age (Years)	<i>M(SD)</i> = 40.88(5.62); <i>Range</i> = 24.00 to 56.00
Father Age (Years)	<i>M(SD)</i> = 43.77(5.92); <i>Range</i> = 26.00 to 63.00
Age of Child with Cancer (Years)	<i>M(SD)</i> = 10.85(5.41); <i>Range</i> = 0.83 to 25.08
Gender of Child with Cancer	46.6% Female (n = 97)
Additional Children in Home	39.9% Have ≥ 3 children in home (n = 83)
Hispanic or Latino Ethnicity (Children)	4.8% Hispanic/Latino (n = 10)
Racial Background (Children)	84.5% White (n = 174) 13.5% Black / African-American / mixed race (n = 28) 2.0% Unknown or Other (n = 4)
Parent (Respondent) Highest Level of Education	22.7% Some High School / High School Graduate (n = 47) 24.2% Some College (n = 50) 7.2% Graduate of 2-yr College (n = 15) 26.6% Graduate of 4-yr College (n = 55) 19.3% Graduate / Professional School (n = 40)
Parent (Respondent) Marital Status	84.2% Married / Partnered (n = 175) 7.7% Never Married (n = 16) 7.7% Separated / Divorced (n = 16) 0.5% Widowed (n = 1)
Parent (Respondent) Employment Status	37.7% Full-time (n = 78) 22.2% Part-time (n = 46) 40.1% Not Employed (n = 83)
Family Income	8.0% <\$25,000 (n=16) 13.9% \$25,000 - \$49,999 (n = 28) 37.3% \$50,000 - \$99,999 (n = 75) 40.7% ≥ \$100,000 (n = 82)
<b>Illness:</b>	
Time Since Diagnosis (Months)	<i>M(SD)</i> = 17.48(7.72); <i>Range</i> = 1.00 to 38.00
Cancer Diagnosis Category	31.7% leukemia (n = 66) 13.9% lymphoma (n = 29) 39.4% solid tumor (n = 82) 13.0% brain tumor (n = 27) 1.9% other diagnosis (n = 4)
Treatment Category (Note: Can have more than one)	65.2% Port / Broviac (n = 135) 58.7% Biopsy (n = 122) 36.4% Resection (n = 75) 83.1% Chemotherapy (n = 172) 38.2% Radiation (n = 79) 5.3% Stem Cell Transplant (n = 11) 6.3% Bone Marrow Transplant (n = 13)

Table 2: Pearson correlations between sibling distress & proposed continuous covariates

	Income	Age	TimeDx	Distress	Depression	Anxiety	PTSS
Family Income	1.00	.08	.02	<b>-.18*</b>	-.08	<b>-.17*</b>	<b>-.20**</b>
Sibling Age		1.00	-.04	-.02	.10	-.08	-.08
Time Since Diagnosis			1.00	-.09	-.11	-.08	-.03
Distress (Composite)				1.00	<b>.85**</b>	<b>.90**</b>	<b>.88**</b>
Depression (Transformed)					1.00	<b>.65**</b>	<b>.58**</b>
Anxiety						1.00	<b>.72**</b>
PTSS (Transformed)							1.00

*\*p<.05*

*\*\*p<.01*

Table 3: Pearson correlations (r) among sibling distress, parenting, & parent PTSS variables

	S-Dep	S-Anx	S-PTSS	SM-Acc	SM-PsyC	SF-Acc	SF-PsyC	M-Acc	M-PsyC	F-Acc	F-PsyC	M-PTSS	F-PTSS
S-Age	.10	-.08	-.08	<b>-.21**</b>	-.07	<b>-.19**</b>	<b>-.14**</b>	<b>-.16*</b>	-.06	-.01	-.05	.08	<b>-.23<sup>t</sup></b>
S-Distress	<b>.85**</b>	<b>.90**</b>	<b>.88**</b>	<b>-.19**</b>	<b>.27**</b>	<b>-.17*</b>	<b>.24**</b>	-.02	.10	<b>-.23<sup>+</sup></b>	-.03	<b>.15*</b>	<b>.23<sup>+</sup></b>
S-Depression	1.00	<b>.64**</b>	<b>.58**</b>	<b>-.30**</b>	<b>.18**</b>	<b>-.25**</b>	<b>.12<sup>+</sup></b>	<b>-.12<sup>+</sup></b>	.07	<b>-.24*</b>	.00	<b>.17*</b>	.12
S-Anxiety		1.00	<b>.72**</b>	-.09	<b>.24**</b>	<b>-.15*</b>	<b>.25**</b>	.05	.06	-.19	-.03	.09	<b>.20<sup>+</sup></b>
S-PTSS			1.00	-.11	<b>.28**</b>	-.04	<b>.27**</b>	.02	<b>.12<sup>+</sup></b>	-.14	-.06	<b>.14<sup>+</sup></b>	<b>.28*</b>
SM-Acc				1.00	<b>-.30**</b>	<b>.37**</b>	-.07	<b>.35**</b>	<b>-.20**</b>	.10	.01	-.11	-.03
SM-PsyC					1.00	-.03	<b>.67**</b>	-.05	<b>.24**</b>	-.11	.12	.05	.08
SF-Acc						1.00	<b>-.14*</b>	<b>.18*</b>	-.06	<b>.23<sup>+</sup></b>	-.18	-.07	-.03
SF-PsyCon							1.00	.08	<b>.16*</b>	-.11	.08	.04	.04
M-Acc								1.00	<b>-.16*</b>	.21	.00	-.08	-.04
M-PsyC									1.00	<b>-.23<sup>+</sup></b>	<b>.31*</b>	<b>.19*</b>	.19
F-Acc										1.00	<b>-.33**</b>	-.11	.05
F-PsyC											1.00	.01	.12
M-PTSS												1.00	<b>.33*</b>
F-PTSS													1.00

\*  $p < .05$ , \*\*  $p < .01$ , <sup>t</sup>  $p \leq .10$

Distr = Composite Sibling Distress

Reporter: S = Sibling, SM = Sibling Report on Mother, SF = Sibling Report on Father, M = Mother, F = Father

Parenting: Acc = Acceptance, PsyC = Psychological Control

Table 4: Pearson correlations (r) among sibling distress & family functioning problem variables

	S-PS	S-C	S-R	S-AR	S-AI	S-GF	M-PS	M-C	M-R	M-AR	M-AI	M-GF	F-PS	F-C	F-R	F-AR	F-AI	F-GF
S-Age	<b>.16*</b>	-.01	-.02	-.04	-.05	.09	<b>.23**</b>	<b>.14<sup>t</sup></b>	<b>.14<sup>t</sup></b>	.08	.06	<b>.19**</b>	-.04	-.06	-.05	.00	-.01	-.10
Distr	<b>.22**</b>	<b>.29**</b>	<b>.39**</b>	<b>.21**</b>	<b>.36**</b>	<b>.43**</b>	-.11	-.06	.09	-.10	.08	.02	-.04	-.12	<b>.25*</b>	-.03	-.05	.11
S-PS	1.00	<b>.52**</b>	<b>.43**</b>	<b>.47**</b>	<b>.25**</b>	<b>.61**</b>	<b>.20**</b>	<b>.19**</b>	<b>.27**</b>	<b>.27**</b>	<b>.27**</b>	<b>.26**</b>	.19	<b>.22<sup>t</sup></b>	<b>.23<sup>t</sup></b>	.14	.18	<b>.25*</b>
S-C		1.00	<b>.45**</b>	<b>.55**</b>	<b>.25**</b>	<b>.60**</b>	<b>.15*</b>	<b>.24**</b>	<b>.25**</b>	<b>.26**</b>	<b>.20**</b>	<b>.30**</b>	.16	.12	.11	.18	.06	.15
S-R			1.00	<b>.42**</b>	<b>.51**</b>	<b>.61**</b>	<b>.15*</b>	<b>.16*</b>	<b>.31**</b>	<b>.20**</b>	.12	<b>.21**</b>	.07	.10	<b>.28*</b>	.19	.18	<b>.24*</b>
S-AR				1.00	<b>.40**</b>	<b>.65**</b>	<b>.22**</b>	<b>.19**</b>	<b>.22**</b>	<b>.28**</b>	<b>.19*</b>	<b>.25**</b>	.17	.16	.24	<b>.25*</b>	.10	<b>.23<sup>t</sup></b>
S-AI					1.00	<b>.53**</b>	.08	.01	<b>.16*</b>	.11	<b>.14<sup>t</sup></b>	.08	.16	.02	.11	.08	.01	.17
S-GF						1.00	<b>.19**</b>	<b>.25**</b>	<b>.29**</b>	<b>.27**</b>	<b>.29**</b>	<b>.30**</b>	.15	.14	<b>.28*</b>	.12	.18	<b>.25*</b>
M-PS							1.00	<b>.65**</b>	<b>.48**</b>	<b>.56**</b>	<b>.41**</b>	<b>.71**</b>	<b>.25<sup>t</sup></b>	.19	<b>.26<sup>t</sup></b>	<b>.26<sup>t</sup></b>	<b>.25<sup>t</sup></b>	<b>.29*</b>
M-C								1.00	<b>.44**</b>	<b>.69**</b>	<b>.55**</b>	<b>.74**</b>	<b>.31*</b>	<b>.24<sup>t</sup></b>	<b>.26<sup>t</sup></b>	<b>.38**</b>	<b>.36**</b>	<b>.40**</b>
M-R									1.00	<b>.44**</b>	<b>.55**</b>	<b>.61**</b>	<b>.25<sup>t</sup></b>	<b>.33*</b>	<b>.54**</b>	<b>.23<sup>t</sup></b>	<b>.29*</b>	<b>.52**</b>
M-AR										1.00	<b>.50**</b>	<b>.70**</b>	<b>.29*</b>	<b>.31*</b>	<b>.31*</b>	<b>.50**</b>	<b>.30*</b>	<b>.40**</b>
M-AI											1.00	<b>.59**</b>	<b>.30*</b>	.17	.22	.23	<b>.29*</b>	<b>.35*</b>
M-GF												1.00	<b>.44**</b>	<b>.35*</b>	<b>.40**</b>	<b>.46**</b>	<b>.32*</b>	<b>.49**</b>
F-PS													1.00	<b>.64**</b>	<b>.43**</b>	<b>.47**</b>	<b>.42**</b>	<b>.68**</b>
F-C														1.00	<b>.55**</b>	<b>.43**</b>	<b>.47**</b>	<b>.74**</b>
F-R															1.00	<b>.27*</b>	<b>.36**</b>	<b>.75**</b>
F-AR																1.00	<b>.44**</b>	<b>.40**</b>
F-AI																	1.00	<b>.64**</b>
F-GF																		1.00

*p* < .05, \*\* *p* < .01, <sup>+</sup> *p* ≤ .10

Distr = Composite Sibling Distress; Reporter: S = Sibling, M = Mother, F = Father

Family Functioning Problems: PS = Problem Solving, C = Communication, R = Roles, AR = Affective Responsiveness, AI = Affective Involvement, GF = General Functioning



Table 5: T-tests comparing sibling-reported distress according to proposed categorical covariates

Grouping Variable	Distress $M_1$	Distress $M_2$	t-value	p-value
<b>Race<sup>a</sup></b>	<b><math>M_{\text{white}} = -0.081</math></b>	<b><math>M_{\text{non-white}} = 0.40</math></b>	<b>-2.89</b>	<b>.004**</b>
<b>Marital Status<sup>b</sup></b>	<b><math>M_{\text{married}} = -0.064</math></b>	<b><math>M_{\text{unmarried}} = 0.29</math></b>	<b>-2.12</b>	<b>.04*</b>
Additional Siblings	$M_{\text{add'l sibs}} = 0.0061$	$M_{\text{no add'l sibs}} = -0.018$	0.19	.85
Sibling Gender	$M_{\text{female}} = 0.057$	$M_{\text{male}} = -0.085$	1.16	.25
Birth Order Relative to the Child With Cancer	$M_{\text{older}} = -0.0046$	$M_{\text{younger}} = 0.015$	0.15	.88

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

<sup>a</sup> Race was dichotomized into white (n = 172, 84%) and non-white (n = 32, 16%)

<sup>b</sup> Marital status was dichotomized into married (married or remarried, n = 175) or unmarried (never married, separated, divorced, widowed, n = 33)

Table 6: Descriptive statistics for measures of sibling distress, parent PTSD, & family functioning

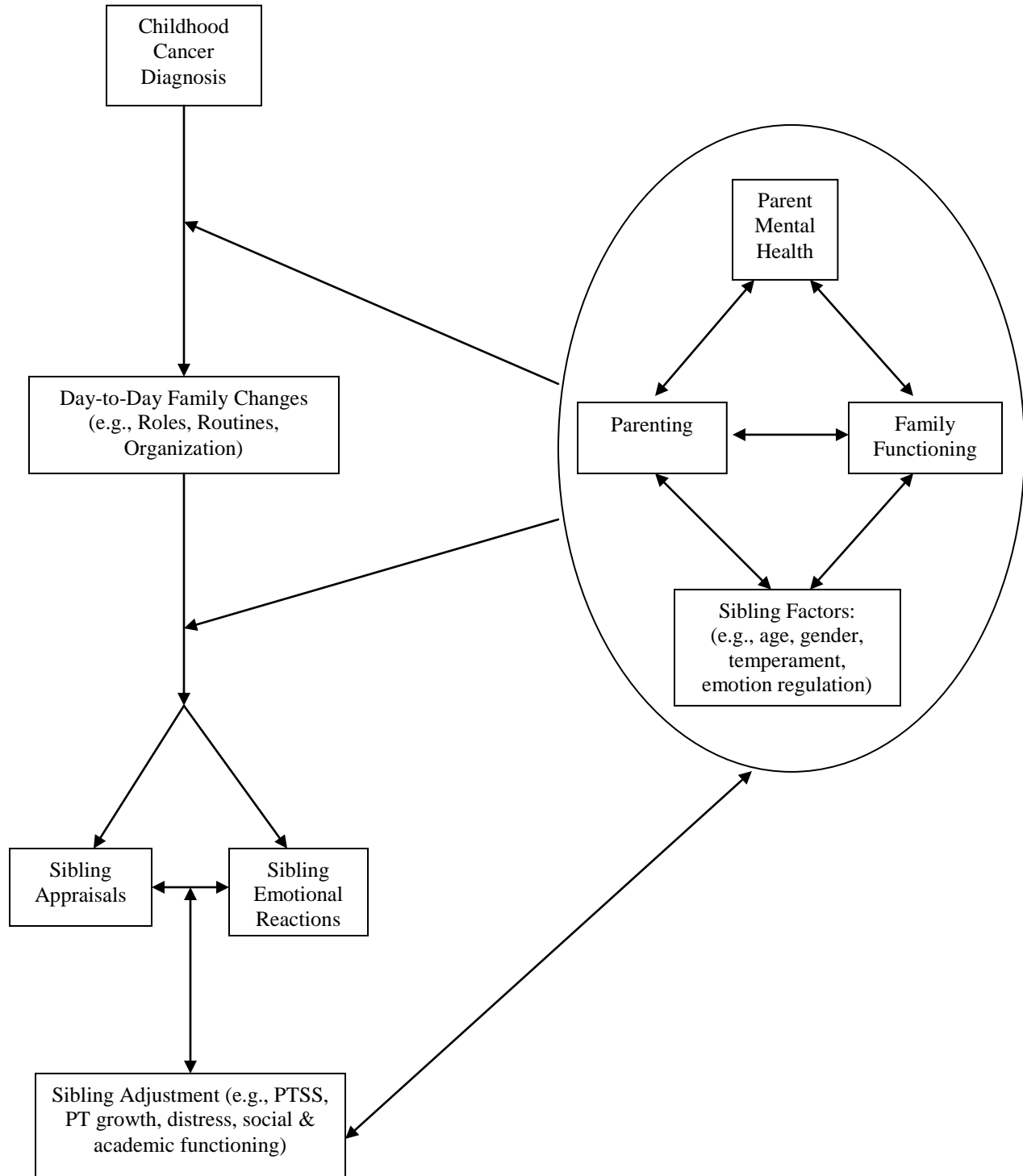
Outcome	Minimum	Maximum	Mean	Std. Dev.	Clinical Range
Sibling Depression Total Score (CDI)	0	15	2.64	2.89	Borderline: <sup>a</sup> N = 11 (5.3%) Clinical: <sup>b</sup> N = 7 (3.3%)
Sibling Anxiety Total Score (RCMAS)	0	27	11.40	6.50	Borderline: <sup>a</sup> N = 29 (13.9%) Clinical: <sup>b</sup> N = 10 (4.8%)
Sibling Posttraumatic Stress Total Score (CPSS)	0	50	15.13	9.77	Meets Diagnostic Criteria: N = 52 (25%) Moderate/Severe Reactions: <sup>c</sup> N = 130 (62.2%)
Sibling Distress Composite Score	-1.89	2.34	-0.0078	0.88	N / A
Parent Posttraumatic Stress Total Score (PDS)	Mother: 0 Father: 0	Mother: 46 Father: 44	Mother: 13.17 Father: 11.83	Mother: 9.92 Father: 10.34	Meets Diagnostic Criteria: Mother: N = 66 (34.9%) Father: N = 20 (27.8%)
Family Functioning (FAD)	Sib: 1.00 Mother: 1.00 Father: 1.08	Sib: 4.00 Mother: 2.67 Father: 3.08	Sib: 1.95 Mother: 1.72 Father: 1.84	Sib: 0.47 Mother: 0.40 Father: 0.41	Rates of Unhealthy Functioning: <sup>d</sup> Sib: N = 96 (47.3%) Mother: N = 49 (26.2%) Father: N = 27 (38.0%)

<sup>a</sup> Borderline range includes T-scores between 60 and 69; 16% of scores in the standardized T-score distribution fall into this range

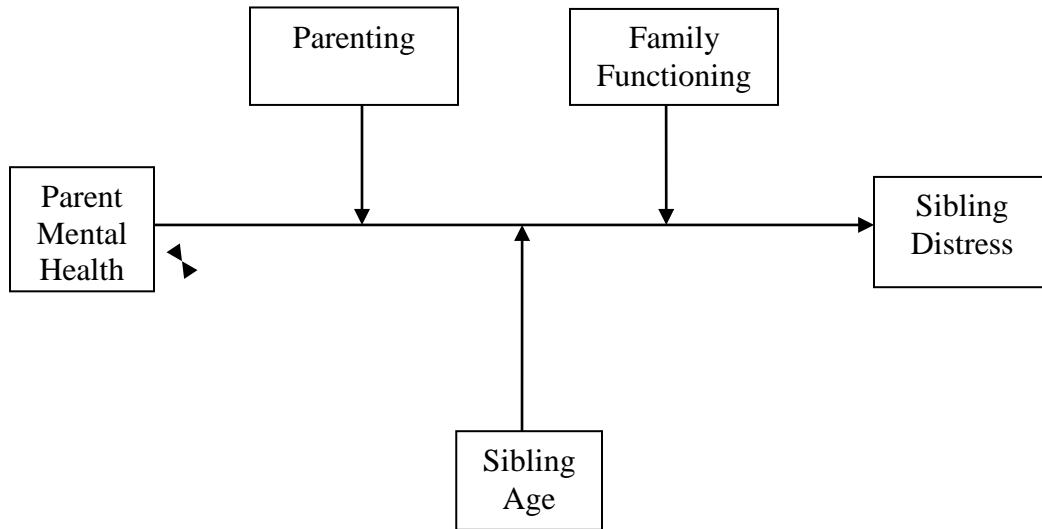
<sup>b</sup> Clinical range includes T-scores  $\geq 70$ ; 2% of scores in the standardized T-score distribution fall into this range

<sup>c</sup> Moderate to severe reactions correspond to CPSS raw scores  $\geq 11$

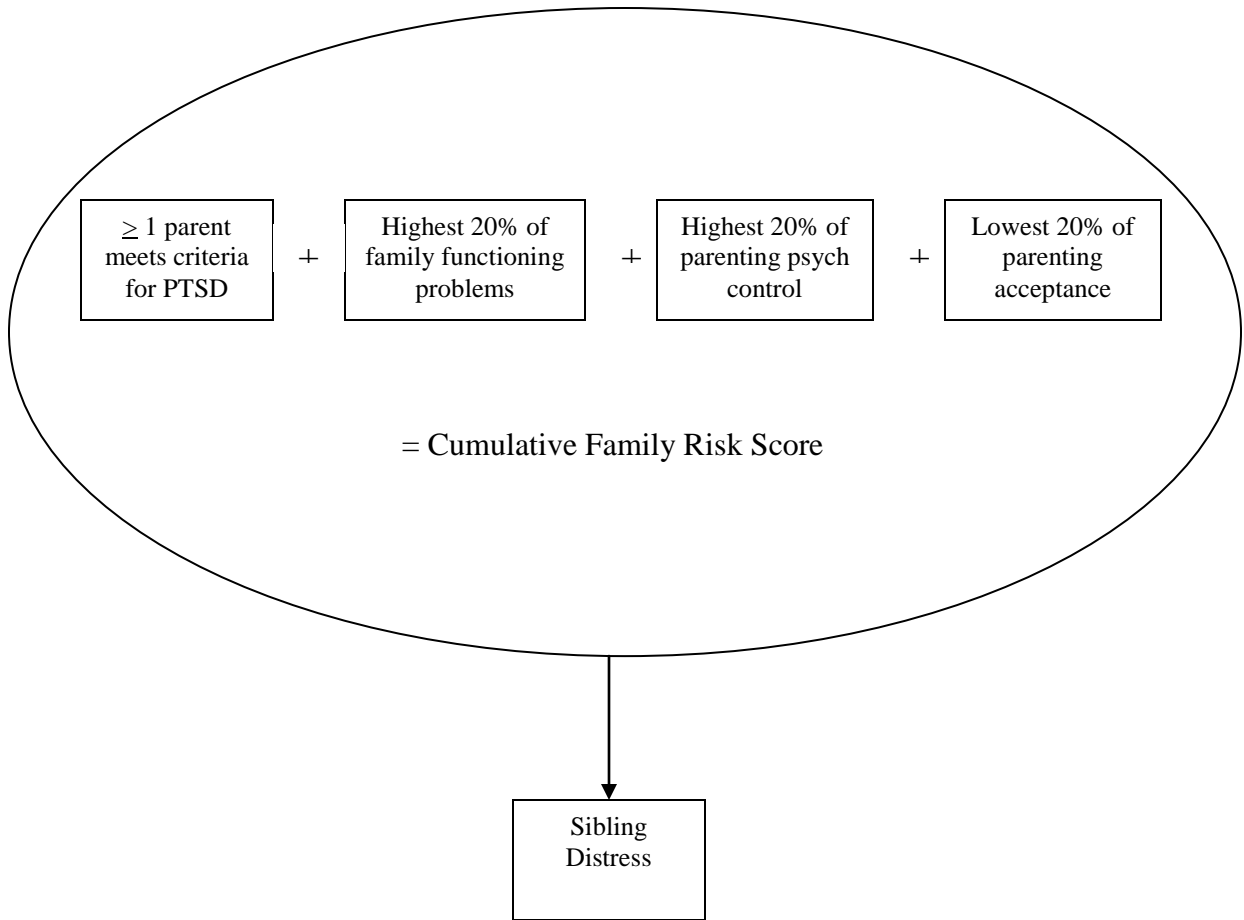
<sup>d</sup> Unhealthy levels of family functioning are indicated by scores of  $\geq 2$  on the FAD



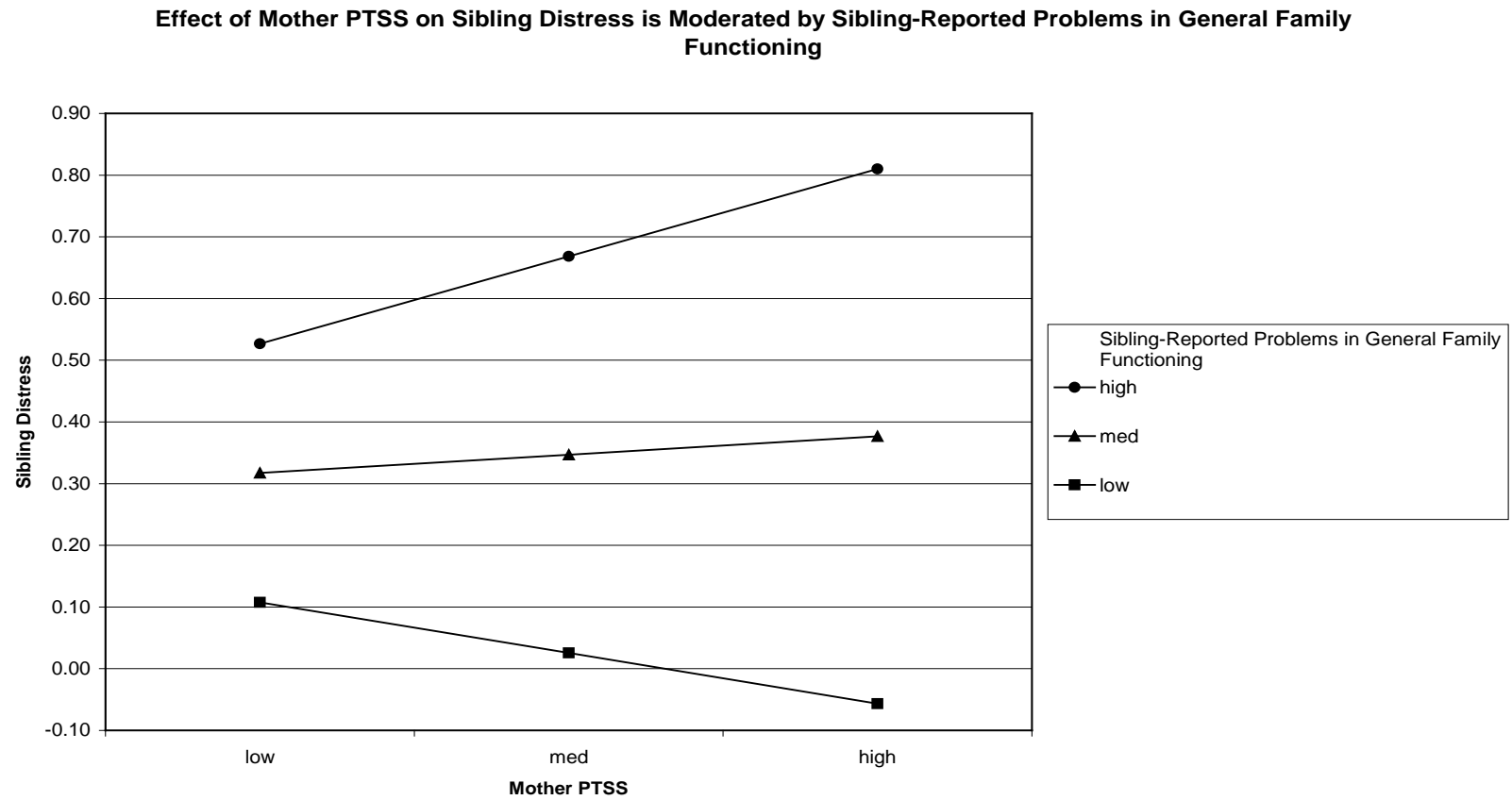
**Figure 1:** Cancer-related family changes, along with family-level and individual risk and protective factors, influence siblings' adjustment to a brother's or sister's cancer diagnosis



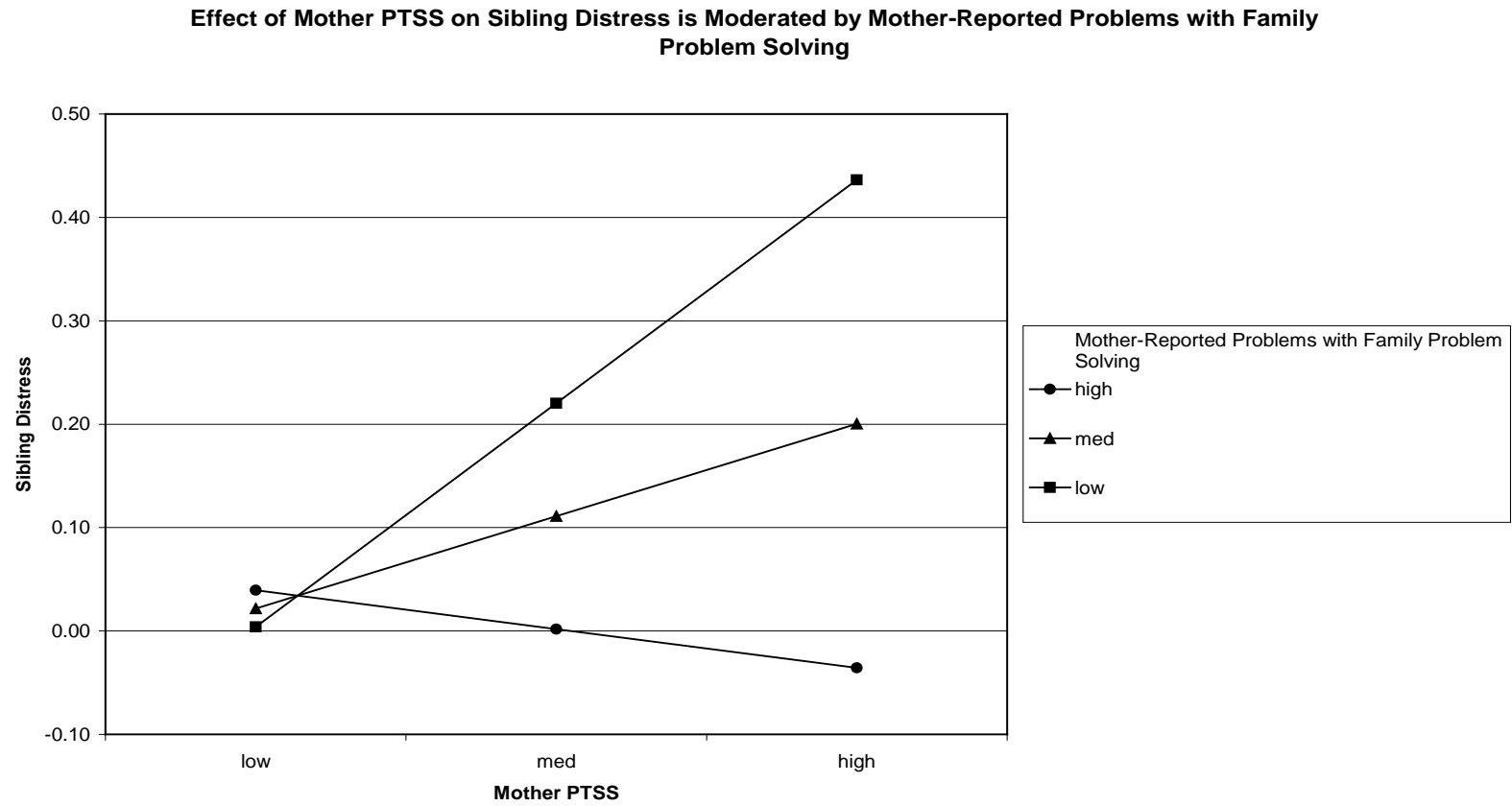
**Figure 2:** Parenting, family functioning, and sibling age as possible moderators of the association between parent PTSS and sibling distress



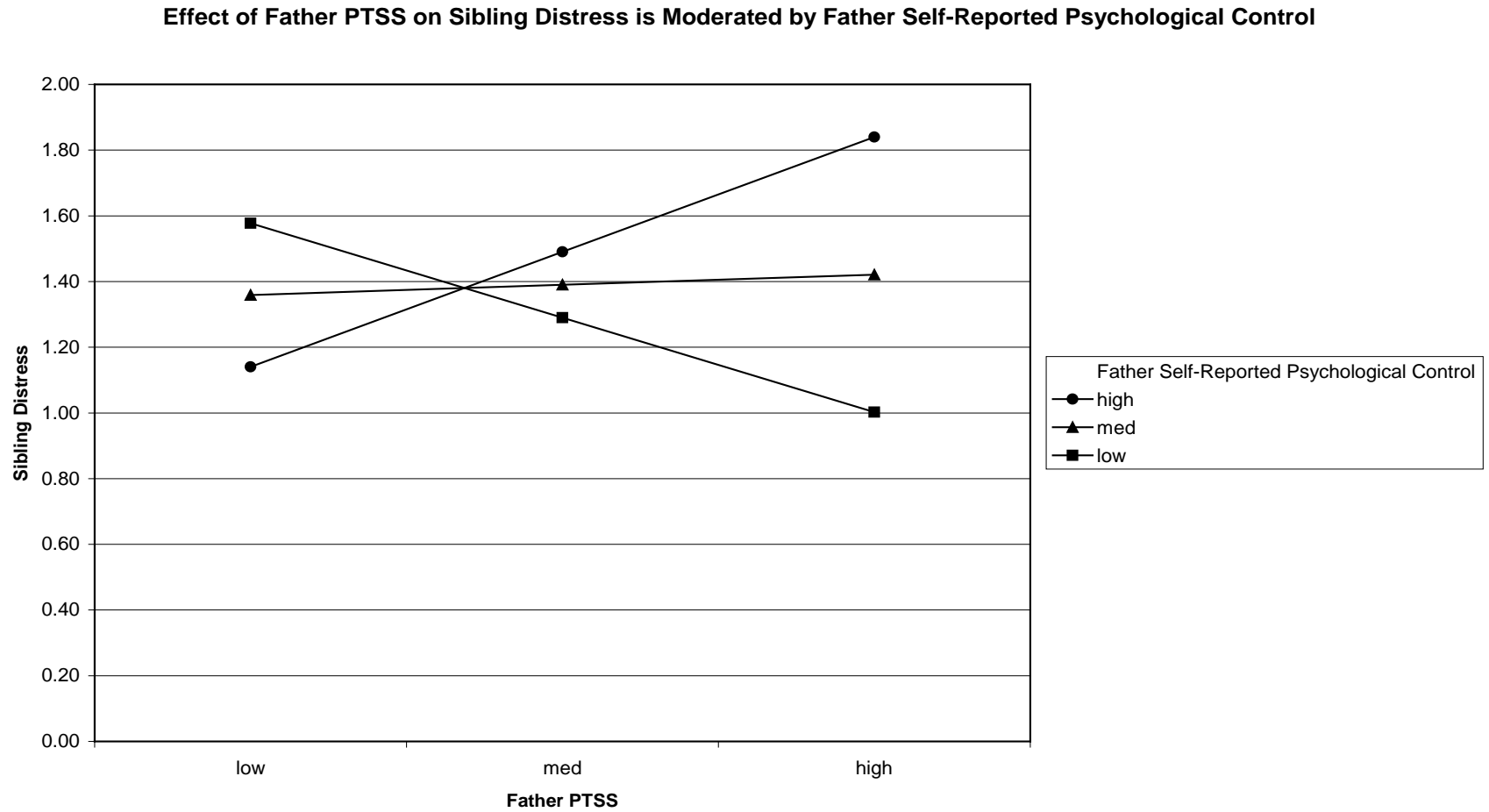
**Figure 3:** The cumulative family risk score is calculated by summing the dichotomized risk scores from each family predictor and is hypothesized to predict sibling distress



**Figure 4:** Effect of mother PTSS on sibling distress is marginally significant at higher levels of family functioning problems (simple slope=0.014,  $t(185)=1.84$ ,  $p=.07$ ) but not at mean (simple slope=0.003,  $t(185)=0.46$ ,  $p=.64$ ) or lower levels (simple slope=-0.008,  $t(185)=-0.90$ ,  $p=.39$ )

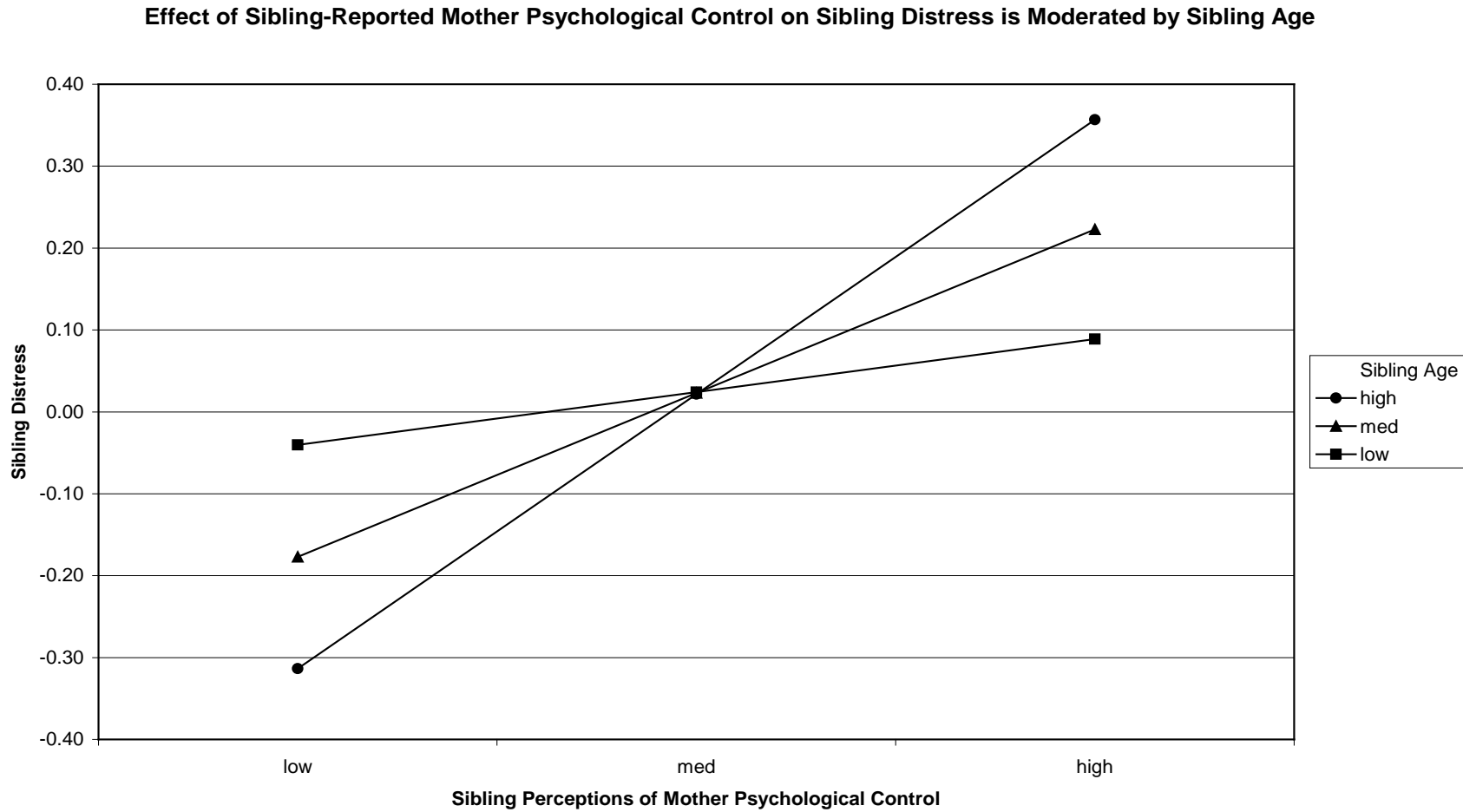


**Figure 5:** Effect of mother PTSS on sibling distress is significant at lower levels of problems with family problem solving (simple slope=0.022,  $t(185)=2.69$ ,  $p=.008$ ), but not at mean (simple slope=0.009,  $t(185)=1.34$ ,  $p=.18$ ) or higher levels (simple slope=-0.004,  $t(185)=-0.41$ ,  $p=.69$ )

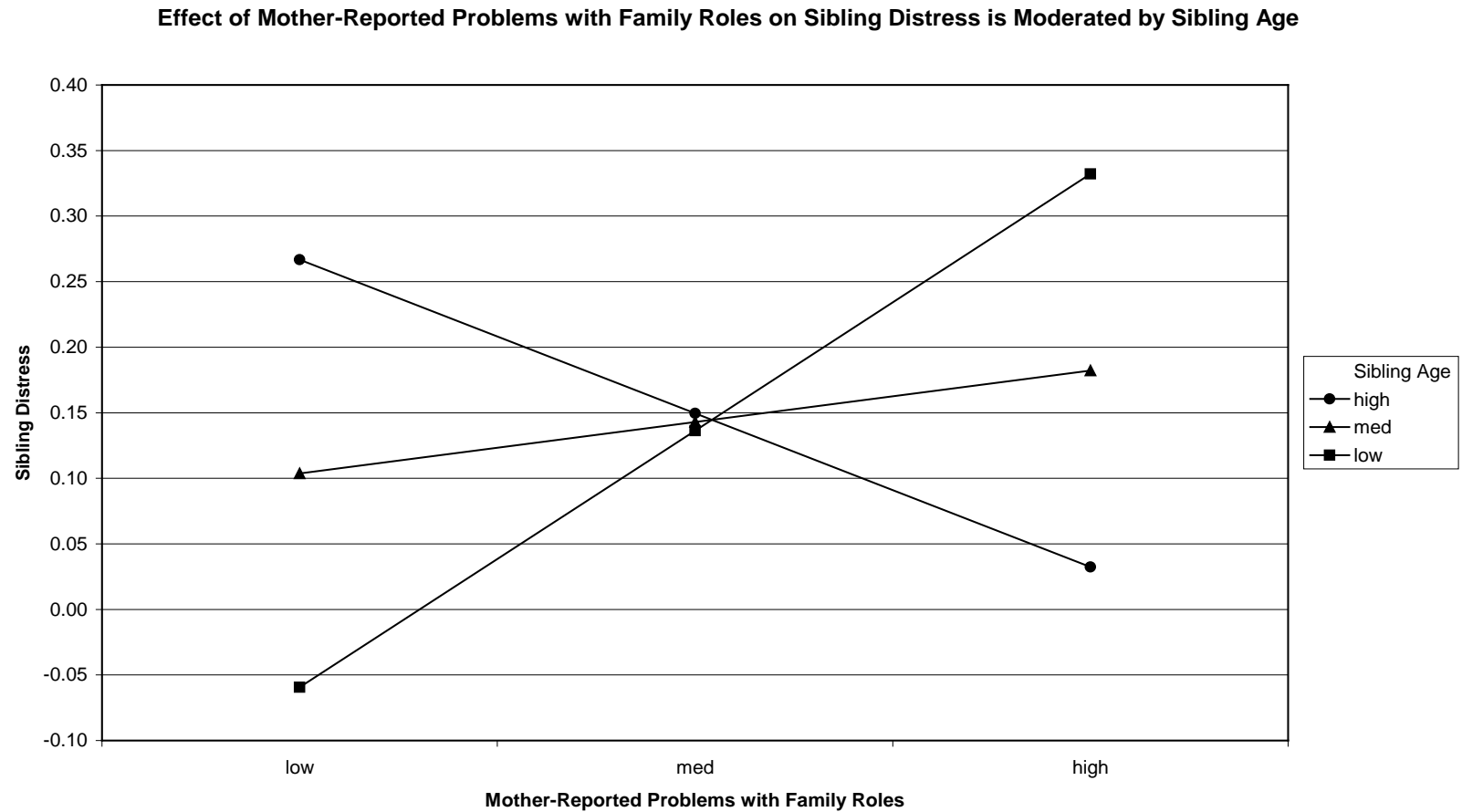


**Figure 6:** Effect of father PTSS on sibling distress is significant at higher levels of father self-reported psychological control (simple slope=0.034,  $t(66)=2.45$ ,  $p=.02$ ) but not at mean (simple slope=0.003,  $t(66)=0.25$ ,  $p=.80$ ) or lower levels (simple slope=-0.028,  $t(66)=-1.32$ ,  $p=.19$ )



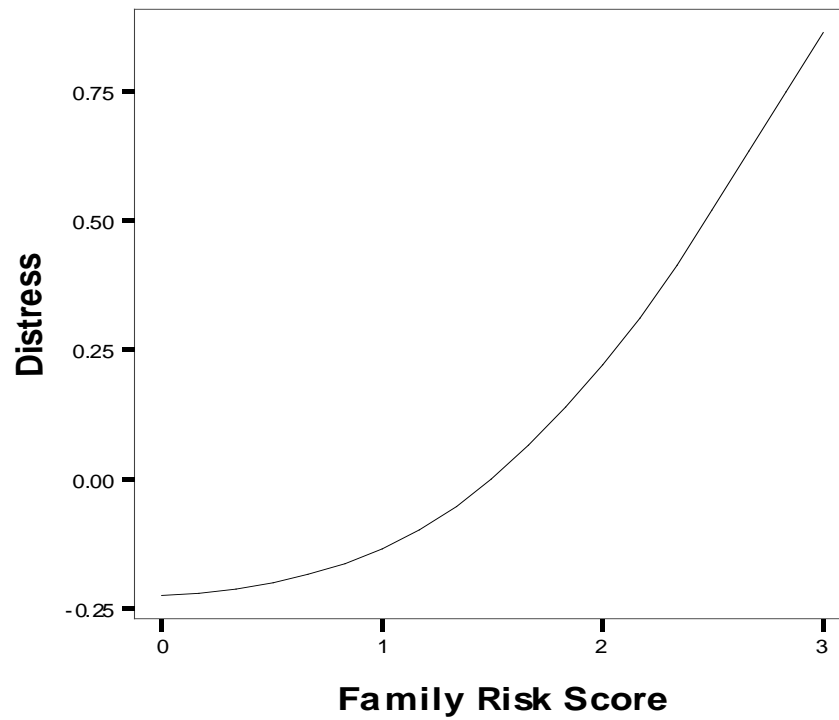


**Figure 7:** The relationship between sibling perceptions of maternal psychological control and sibling distress is significant at older (age 15, simple slope=0.079,  $t(203)=4.52$ ,  $p<.001$ ) and mean (age 12.5, simple slope=0.047,  $t(203)=3.34$ ,  $p<.001$ ) but not younger sibling age (age 10, simple slope=0.015,  $t(203)=0.74$ ,  $p=.46$ )



**Figure 8:** The relationship between mother-reported problems with family roles and sibling distress is significant for younger (age 10, simple slope=0.055,  $t(184)=2.06$ ,  $p=.04$ ) but not mean (age 12.5, simple slope=0.011,  $t(184)=0.60$ ,  $p=.55$ ) or older sibling age (age 15, simple slope=-0.033,  $t(184)=-1.36$ ,  $p=.18$ )

### Sibling Distress According to Number of Family Risks



**Figure 9:** The strength of the positive association between sibling distress and cumulative family risk is greater at higher levels of risk

*Note: a score of “3” reflects 3 or 4 family risks*

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