Parental Adjustment: An Examination of Caregivers of Pediatric Cancer Survivors

by

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Dissertation submitted in partial fulfillment of
the requirements for the degree of Doctor
of Philosophy in the Department of
Psychology and Neuroscience in the Graduate School
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<u>ABSTRACT</u>

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Abstract

Caregivers of survivors of pediatric cancer face ongoing social, emotional and financial challenges that may result in enduring illness- and caregiving-related distress. After patients complete treatment there are challenges that persist. Indeed, emerging physical and cognitive "late effects," resulting from the disease and treatment, require families to adapt to a new normal state that may require significant long-term follow-up and care by the survivor and their primary caregiver. This study compares the psychological adjustment of caregivers of pediatric cancer survivors with caregivers of healthy children. In addition, this study evaluates individual factors, such as family functioning and coping style, which may be associated with poorer adjustment among caregivers of pediatric cancer survivors. Caregivers of pediatric cancer survivors (n = 64) and caregivers of healthy children (n = 64) were recruited during regularly scheduled clinic visits to complete questionnaires including a demographic and illness questionnaire and measures of psychosocial functioning. A series of multivariate analyses of covariance were conducted to assess for differences in caregiver distress by group. Caregivers of survivors reported significantly more child-specific parenting stress and somatization than caregivers of healthy children. Hierarchical regression modeling revealed that *Escape-avoidance* coping and *Supportive* family functioning predicted 25-40% of the variance in parenting-related psychological adjustment among caregivers of survivors.

Caregivers of pediatric cancer survivors face unique challenges that contribute to ongoing distress, particularly related to the parenting role. This population may benefit from interventions aimed at reducing avoidance based coping and improving family functioning.

Dedication

This work is dedicated to my family: my parents, who foster in me a belief that I can face any challenge, my sisters, who generously share their humor and friendship, my children, who inspire me daily to be better, and my husband, a true partner, without whom none of this would be possible.

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the hopes that their experiences might someday help another family in need. Their

generosity continues to inspire me.

1. Introduction

Cure rates for childhood cancer now exceed 75%, up from 30% in 1960 (ACS, 2007). As the new population of pediatric cancer survivors and their families grows, there is increased need for psychosocial research on adjustment and its predictors. Indeed, because parents serve as the primary caregiver and decision maker for their children, parental adaptation to the long-term cancer experience impacts the adjustment and quality of life of the cancer survivor and the entire family system.

Parents of pediatric cancer survivors often face residual effects of the disease and its treatment, plus the risk of newly emerging late effects or disease recurrence, that require significant long-term follow-up and care. In addition to concerns about physical health, pediatric cancer survivors often experience late effects including cognitive and learning deficits. These concerns require vigilance and action on the part of the parent to identify and meet their child's unique academic and social needs (Hutchinson & Bonner, 2008). There is a growing body of literature supporting the conclusion that parental stress and distress continue well past the completion of treatment, which suggests parents struggle with adapting to the "new normal" and continue to have fears about the long-term health and wellbeing of their child (Hutchinson, Willard, Hardy, & Bonner, in press; Kazak et al., 2004; Van Dongen-Melman et al., 1995; Van Dongen-Melman, Van Zuuren, & Verhulst, 1998; Vrijmoet-Wiersma et al., 2008).

There is also evidence of variability in the adjustment of parents of pediatric

cancer survivors. While research suggests that caring for a survivor takes an emotional

and physical toll on parents (Hardy et al., 2008; Norberg, 2007), new findings suggest

that many parents are able to identify benefits and positive outcomes associated with

their child's disease (Kim, Schulz, & Carver, 2007; Stuber, 2006). Given this variability, it

is important to identify the factors associated with parental adjustment. Guided by the

Transactional Stress and Coping (TSC) model of parental adjustment to childhood

illness, family functioning and coping style have been identified as individual variables

that may mediate the adjustment process in parents of pediatric cancer survivors.

1.1 Sources of Stress

Childhood cancer has distinct and persistent psychosocial effects on parents of

survivors. Recent qualitative research has revealed numerous stressors for parents of

survivors, including stress related directly to the cancer, such as secondary illness,

disabilities, attentional deficits, and loss of normal life and activities, as well as parenting

stressors, such as ongoing feelings of grief, loss and uncertainty (Patterson, Holm, &

Gurney, 2004). Moreover, these stressors remain salient sources of distress and

considerable strains on family resources well after treatment is completed. Indeed,

research suggests that the treatment itself has little or no effect on reported problems;

however, when treatment leads to long-term sequelae in the child, parents experience

significant effects, including symptoms of depression and anxiety. This suggests that it is

not the acute stressors during active treatment, but the emergence of new disease related

stressors that impact parent adjustment (Van Dongen-Melman et al., 1995; Van Dongen-

Melman et al., 1998).

1.1.1 Physical and Cognitive Late Effects

Studies of pediatric cancer survivors have examined many salient potential

stressors with which caregivers must cope, including physical risks (e.g., secondary

cancer and endocrine issues), cognitive late effects, and impaired social functioning

(Nagarajan et al., 2003; Oeffinger & Hudson, 2004; Oeffinger et al., 2004; Zebrack et al.,

2004). Physical health consequences for cancer survivors vary as a result of diagnosis

and treatment intensity, and may include endocrine dysfunction, bladder or kidney

disease, cardiomyopathy, osteoporosis, infertility, secondary cancers, hearing loss, and

neurocognitive deficits (Oeffinger & Hudson, 2004; Oeffinger et al., 2004). Indeed, the

Childhood Cancer Survivor Study, a retrospective study of adult survivors of childhood

cancer, revealed that survivors are 3.3 times more likely than their siblings to have a

chronic medical condition, and 8.2 times more likely to have a severe or life-threatening

condition (Oeffinger et al., 2006).

Brain tumors and leukemia, specifically acute lymphocytic leukemia (ALL), have

been the focus of many studies of neurocognitive outcomes in cancer survivors because

they have the most profound effect on functioning directly and indirectly, as a result of treatment to the central nervous system (CNS) (Armstrong & Mulhern, 1999; Moore, 2005). Brain tumors and ALL are also the two most common forms of childhood cancer; children with malignant tumors of the brain and central nervous system account for approximately 20% of all childhood cancer diagnoses, while children with leukemia (cancer of the blood) comprise approximately 30% (ACS, 2007).

Children with ALL are at high risk for brain metastases; therefore, treatment often targets the CNS. Traditionally, CNS preventative therapy has included cranial radiation therapy (CRT) and intrathecal chemotherapy, usually with methotrexate. Due to the risks of CNS toxicity, treatment is now usually limited to intrathecal and systemic chemotherapy. However, use of intrathecal methotrexate still places children at an elevated risk for neurocognitive sequelae, including difficulties with attention, memory, and visual-motor integration (Butler & Mulhern, 2005). When neurocognitive impairments do occur, they have a significant impact on a child's success in school, self esteem, social relationships, and long-term quality of life.

Children with brain tumors are also at high risk for late effects. In the physical domain, survivors may experience hearing loss, vision loss, growth failure, seizure disorder, endocrine dysfunction, chronic fatigue and gross and fine motor impairments (Armstrong & Mulhern, 1999). In the cognitive domain, survivors experience deficits in

memory (Dennis et al., 1991), academic (Armstrong & Mulhern, 1999) and attentional functioning (S. J. Thompson et al., 2001). Rates of late effects vary as a function of gender, age at treatment, tumor type, tumor location and treatment type (Butler & Copeland, 2002; Moore, 2005; Mulhern, Kovnar, Kun, Crisco, & Williams, 1988). Although children with brain tumors are a heterogeneous group, they are a distinct and unique subgroup of survivors that are clearly placed at the greatest risk for severe late effects due to the disease and its treatments.

Ongoing social impairments in survivors of cancer, including social isolation, may also contribute to the difficult task of parenting a survivor (Patenaude & Kupst, 2005; Vannatta, Gartstein, Short, & Noll, 1998). In addition to frequent absences from school and social functions and physical symptoms (such as chronic fatigue), there may be a neurocognitive aspect to cancer survivors' social impairments. Indeed, children treated for brain tumors exhibit higher levels of social impairment, including deficits in facial expression recognition (Bonner et al., 2008). Moreover, children treated for brain tumors are at increased risk for acquiring nonverbal learning disabilities (Buono et al., 1998). Nonverbal learning disabilities are characterized by problems in social perceptions, social judgment and social interaction skills. These effects, which emerge over time, can negatively impact a child's quality of life in both the academic and social realm.

The physical and cognitive late effects exhibited in cancer survivors extend the impact of the disease indefinitely, posing additional and chronic parenting and caregiver stress. Recent research indicates that parents of learning or cognitively disabled children exhibit higher levels of distress than do parents of nondisabled children (Baker & McCal, 1995; Dyson, 1996; Fuller & Rankin, 1994). This stress may, in turn, impact the cognitive and social development of survivors. Indeed, it has recently been proposed that the relationship between family stress and survivors' neurodevelopmental sequelae is bi-directional (Peterson & Drotar, 2006).

1.1.2 Emotional Adjustment

In addition to coping with their child's physical, cognitive and social issues, parents must cope with their own emotions, which may include grief, loss, and uncertainty about the future. Van-Dongen-Melman and colleagues (1998) used qualitative techniques to gain insight into the emotional experiences of parents of childhood cancer survivors. There were several domains in which parents experienced a sense of loss. Nearly every parent reported a loss of positivity towards life and a sense that they would never again be the same relaxed person they had been prior to their child's diagnosis. Many parents also reported a heightened vulnerability to traumatic experiences that renders them less able to cope with stressors. A third loss was that of

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time and energy that was spent caring for their ill child who could have been spent differently had their child not been ill.

Late effects also contribute to parents' sense of loss with respect to the child. Parents in Von Dongen-Melman's sample described the loss of the child they had prior to the cancer; although their child was cured of cancer, he or she was not necessarily the same healthy child as before. Some of the children became mentally and/or physically handicapped as a result of their disease, contributing to the loss of the image and reality of a healthy child (Van Dongen-Melman et al., 1998). These reports are consistent with the cumulative findings that parents of children with disabilities and chronic illness suffer long-term periodic sadness (Burke, Hainsworth, Eakes, & Lindgren, 1992). Indeed, parents report "chronic sorrow" – prolonged feelings of loss, grief, disappointment and fear in response to continual losses associated with chronic illness. This sorrow is appropriate, considering the many challenges of parenting a chronically ill child, and does not meet criteria of clinical psychopathology. Unfortunately, it often goes unrecognized and untreated (Bonner et al., 2006).

Parenting a cancer survivor also requires coping with uncertainty about the future. Uncertainty, characterized by not knowing what caused the cancer, not knowing the child's prognosis and not knowing how late effects will impact their child's future, has been linked to psychological distress, characterized by anxiety, depression, cognitive

disturbances and feelings of helplessness (Stewart & Mishel, 2000). Moreover, a recent review of parental uncertainty and posttraumatic stress literatures also supports a linkage that may be important in developing interventions to support parents of children with serious illnesses (Santacroce, 2003).

1.2 Impact of Stress on Caregivers

The intense caregiving demands elicited by parenting a cancer survivor often exceed that of parenting a healthy child. As such, parents of cancer survivors may be at risk for impaired physical and mental health. A growing body of literature supports the conclusion that some level of parental distress is ongoing, long after treatment is completed. While acute parental distress related to a child's cancer diagnosis may decrease over time, many parents continue to suffer from clinical levels of distress, even five years off treatment (Wijnberg-Williams, Kamps, Klip, & Hoekstra-Weebers, 2006). Indeed, in a recent study of caregivers of pediatric brain tumor patients, parents of off-treatment children reported similar levels of ongoing illness- and caregiving-related distress as parents of children still receiving treatment (Hutchinson et al., in press).

Kazak and colleagues' research on post traumatic stress symptoms (PTSS) and post traumatic stress disorder (PTSD) in parents of cancer survivors provides additional evidence of distress in this population (Alderfer, Cnaan, Annunziato, & Kazak, 2005; Brown, Madan-Swain, & Lambert, 2003; Kazak et al., 2004). PTSS are common in

families of childhood cancer survivors. Indeed, parents report more symptomology than former patients, with one study reporting that nearly 30% of mothers met diagnostic criteria for PTSD since their child's diagnosis and nearly 20% of families had at least one parent currently meeting criteria for PTSD (Kazak et al., 2004). Core symptoms of PTSD in cancer survivors and their families include avoidance of the illness experience (e.g. hospitals and clinics), intrusion of the cancer experience into daily activities and in response to minor illness, and hypervigilance. These symptoms persist for parents long after treatment ends (Kazak et al., 1997). The fact that parents report even greater symptomology than the patients themselves leads to the characterization of parents as "second order patients" (Lederber, 1998).

Parenting a cancer survivor may be a more prolonged caregiving relationship than parenting other chronically ill children. One study noted that more than 60% of young adult survivors continue to be accompanied to long-term follow-up visits by a parent, more than three times the rate observed for young adults attending a diabetes clinic (Ressler, Cash, McNeill, Joy, & Rosoff, 2003). A more recent study compared parents of adult survivors of pediatric cancer who continued to accompany their children to long-term follow-up clinic appointments and parents of current pediatric cancer patients on or within one year of active treatment (Hardy et al., 2008). The parents of adult survivors reported just as much anxiety and stress referable to their

child's illness as parents of children on active treatment. These results suggest that parents who continue to accompany their young adult child to clinic may remain psychologically vulnerable many years after the end of treatment and that the impact of having a child with a life-threatening illness may not diminish even years into the child's survivorship.

Such stress and its effect on caregiver health and wellness have been well documented in several other populations. For example, findings in the Alzheimer's literature indicate that the majority of family caregivers experience high levels of caregiving burden (68%) and exhibit depressive symptoms (62%) (Papastavrou, Kalokerinou, Papacostas, Tsangari, & Sourtzi, 2007). Similarly, caregivers of those who have suffered a traumatic brain injury also experience distress, as well as impairment in family functioning (Ergh et al., 2003; Ergh et al., 2002).

The burden of caring for a cancer survivor may be particularly taxing for mothers. Indeed, psychological exhaustion, or "burnout," was assessed in a sample of parents of brain tumor survivors. Mothers of brain tumor survivors reported significantly higher rates of burnout than mothers of healthy children; however, no significant relationship between burnout and parenting a brain tumor survivor was found for fathers (K. E. Robinson, Gerhardt, Vannatta, & Noll, 2007). This finding may be due to the much higher rates of mothers assuming the role of caregiver than fathers.

Studies of pediatric cancer populations have shown up to 77% of mothers consistently identify themselves as the primary caregiver of a pediatric cancer patient and/or survivor (Bonner et al., 2006).

Elevated levels of stress in parents are associated with poorer outcomes for both parent and child. Parental distress impacts children's quality of life, both directly and indirectly. Indeed, significant relationships were found between maternal depressive symptoms, anxiety, parenting stress and children's quality of life in a recent study of families of pediatric cancer patients (Roddenberry & Renk, 2008). Maternal distress is also associated with higher reported child emotional distress and higher child somatic distress following a cancer diagnosis (Steele, Dreyer, & Phipps, 2004).

Parenting stress may also contribute to poorer physical health outcomes for parents as well. Many studies examining the impact of acute and chronic stress on health have found that increased stress leads to a decrease in immune functioning and an increased risk of infectious disease in healthy individuals (Cohen, Doyle, & Skoner, 1999; Cohen, Tyrrell, & Smith, 1991; Cohen & Williamson, 1991; Herbert & Cohen, 1993; Naliboff et al., 1991). Other commonly reported physical effects on caregiver well-being include insufficient time for rest or exercise, perceived poor health, lowered immune function, and increased rates of mortality, depression and anxiety (Kiecolt-Glaser et al.,

1987; Schulz & Beach, 1999). These physical risks also have potential to negatively impact parenting behavior and child outcomes.

Few studies have specifically focused on health and wellness outcomes in parents of cancer survivors. One study followed parents of childhood cancer patients for a period of 18 months and found no improvement of parent physical and mental wellbeing over time (Svavarsdottir, 2005). However, one small Taiwanese study compared the physical health of parents of children in active treatment and parents of children off treatment (Chien et al., 2003). They found that while parents of children off treatment reported better overall health and wellbeing than parents of children in active treatment, their health was poorer than a normative sample.

Similar to other pediatric populations, there is significant variability in the adaptation and adjustment of parents (Van Dongen-Melman et al., 1998). Overall, the research on adaptation indicates that childhood chronic illness is a significant life stressor leading to increased risk for psychological adjustment problems (Goldberg, Morris, Simmons, Fowler, & et al., 1990; Kazak & Barakat, 1997; R. J. Thompson, Jr. & Gustafson, 1996). Despite significant sources of stress, results of research on parents of cancer survivors to date has yielded conflicting results, some indicating overall high rates of good adjustment (Frank, Brown, Blount, & Bunke, 2001) while others indicate high levels of persistent distress (Hutchinson et al., in press; Sloper, 2000; Van Dongen-

Melman et al., 1998; Wijnberg-Williams et al., 2006). Moreover, recent research has

revealed that some parents reporting finding benefits, such as personal growth, as a

result of their experiences with their child's cancer (Kim et al., 2007; Stuber, 2006).

Recognition of variability in psychological adjustment has prompted efforts to

distinguish the correlates of adjustment that identify individuals at high risk for

adjustment problems. There is some evidence that this variability is not linked directly

to child factors, such as diagnosis and treatment intensity. Therefore, it is crucial to

examine parental variables that may serve to mediate the stress and coping process.

1.3 Transactional Stress and Coping Model

The transactional stress and coping (TSC) model (Figure 1) proposed by

Thompson and Gustafson (1996) is a model for maternal adaptation to stress that views

chronic illness as a potential stressor to which the individual and family system

endeavor to adapt. The illness parameters in the TSC model reflect illness type and

severity; the demographic parameters in the model include gender, age and

socioeconomic status. The focus on the model, however, is on the child and maternal

adaptational processes that contribute over and above the contributions of illness and

demographic parameters.

Guided by Lazarus and Folkman's (1984) cognitive model of stress and coping,

Thompson and colleagues included coping methods (Folkman & Lazarus, 1988b) and

family functioning (Moos & Moos, 1981) as maternal adaptational processes in the basic

model. These processes were also selected based on their salience as potential

intervention targets. Coping methods in the TSC model are categorized as either

palliative, a combination of avoidance, wishful thinking and emotion-focused coping, or

adaptive, problem-focused coping aimed at altering the stressful transaction by

attempting to change the environment or self. Family functioning in the TSC model is

categorized by three dimensions: conflicted, controlling or supportive. Combined, these

mediational processes account for significant portions of variance (approximately 20%)

in maternal adjustment to childhood chronic illness over and above illness and

demographic contributions (R. J. Thompson, Jr. & Gustafson, 1996; R. J. Thompson, Jr.,

Gustafson, Gil, Kinney, & Spock, 1999). More specifically, persistently good maternal

adjustment is associated with less frequent use of palliative coping methods and higher

levels of family supportiveness. Notably, illness severity did not make a substantial

contribution to the variance in maternal adjustment.

1.3.1 Maternal Mediational Variables

Using the TSC model as a framework (R. J. Thompson, Jr. & Gustafson, 1996),

family functioning and coping style have been identified as key variables that may

mediate the psychosocial adjustment of caregivers of pediatric cancer survivors.

1.3.1.1 Family Functioning

Family functioning has been shown to moderate the relationship between parent

and child distress in families coping with childhood cancer (K. E. Robinson et al., 2007).

Family functioning in the TSC model is categorized by three dimensions: *Conflicted*

(characterized by high conflict, low cohesion and low organization), *Controlling*

(characterized by high control, high achievement orientation, and low independence)

and *Supportive* (characterized by high cohesion and high expressiveness). The utility of

these dimensions in predicting psychosocial outcomes in childhood chronic illness

populations has been established (Kronenberger & Thompson, 1990).

There is evidence to support the influence of these dimensions of family

functioning on adjustment in cancer survivors. One study of adolescent cancer survivors

(Rait, Ostroff, Smith, & Cella, 1992) examined the perceived family functioning of 88

adolescents who had successfully completed treatment for pediatric cancer and the

relationship between family functioning and post-treatment adjustment. Adolescent

cancer survivors reported lower levels of supportive family functioning than the

normative sample of healthy adolescents and their families. Supportive family

functioning and family adaptability were also strongly associated with better post-

treatment psychological adjustment for the adolescents.

This relationship between family functioning and adjustment is also apparent in parents of children treated for cancer. Streisand and colleagues (2003) examined the relationship between pediatric specific parenting stress and family functioning in a sample of 116 parents of children treated for cancer. After controlling for treatment status (on vs. off treatment), they found that parenting stress related to caring for a child with a medical illness was associated with poorer family functioning, reflected in difficulty in establishing effective behavioral control within the family. The total number of stressful situations encountered by parents is also associated with the level of affective involvement with the family, indicating that parents responding simultaneously to a large number of stressors may have diminished quantity and quality of emotional resources to provide to the entire family.

Attitudes and attributions regarding control within the family environment appear to be salient issues for families of survivors. Childhood cancer is associated with a loss of control in which parents have a very limited ability to influence events in the cancer experience. Attitudes towards control played an important role in the overall wellbeing of parents of cancer survivors in Van Dongen-Melman's (1998) qualitative study. Parents with an extreme orientation towards control, either internal control or external control, experienced more difficulty in adjustment. Van Dongen-Melman and colleagues hypothesized that parents with extreme internal orientation may be so

overcome with a sense of personal responsibility that they suffer from feelings of guilt, anxiety and depression. Parents with extreme external orientations blamed their environment (e.g. the health care system), leading to feelings of anger and resentment. Parents whose appraisals of control were more balanced seemed to be more accepting of reality and experienced less guilt.

1.3.1.2 Coping

Coping methods in the TSC model are categorized as either palliative or adaptive (R. J. Thompson, Jr. & Gustafson, 1996). Palliative coping is defined by emotion-focused attempts to cope, such as avoidance or wishful thinking; adaptive coping is defined as problem-focused attempts to cope, such as attempting to change the environment or self. There is strong evidence to suggest that coping processes have significant relations to psychological symptoms, with problem-focused forms of coping, such as planful problem solving, being negatively correlated with symptoms, and emotion-focused coping, such as avoidance, being positively correlated with symptoms (Folkman & Lazarus, 1988a; Folkman, Lazarus, Gruen, & DeLongis, 1986).

In Van Dongen-Melman's qualitative study of parents of cancer survivors (1998), reported coping strategies were categorized as avoidance or confrontation. Avoidance strategies included drinking alcohol, smoking, distancing oneself, telling oneself not to think about it, focusing on other issues such as work and hobbies, striving towards

normality, and avoiding information about the disease and its long-term consequences. In contrast, confrontation strategies included seeking information about the disease, talking with other parents of children with cancer, talking with medical staff, seeking help and assistance, and accepting the disease and its late effects. The diversity in coping strategies illustrates the variance in the degree of openness of communication about personal feelings. It appears that a moderate ability to express and deal with emotions may be ideal, as parents described as highly emotionally involved (either anxious or angry) had children whose emotional functioning was below the normal range (Van Dongen-Melman et al., 1998).

Given the growing evidence of ongoing stressors associated with parenting cancer survivors, further research is necessary to better understand the mechanisms through which parents cope with the strain of parenting a cancer survivor and how parenting stress in this population influences both physical and mental health. PRIMARY AIM: To determine the unique and largely unrecognized impact of surviving childhood cancer on caregivers', specifically mothers', self-reported psychological adjustment.

Primary Hypothesis: It is hypothesized that mothers of cancer survivors will show poorer adjustment when compared to demographically similar peers.

SECONDARY AIM: To identify what factors are associated with poorer adjustment among parents of survivors (e.g., cognitive processes, treatment variables, demographic factors).

Secondary Hypothesis: It is hypothesized that individual factors, such as coping style and family functioning, will be strongly associated with psychological outcomes.

2. Method

The study objectives were assessed using a sample of 64 mothers of pediatric

cancer survivors followed by the Divisions of Pediatric Neuro-Oncology and Pediatric

Hematology and Oncology at Duke University Medical Center (DUMC) and 64 mothers

of healthy children recruited from the community. All participants were age 18 or older,

able to read and write English, and had at least one child under the age of 17.

Questionnaire data were collected by mail from each group; the survivor's medical data

was verified by chart review.

2.1 Procedures

2.1.1 Mothers of Pediatric Cancer Survivors

Eligible participants, defined as the primary maternal medical caregiver of

children age 17 or younger at least one year off treatment, were identified by their

child's attending physician (pediatric oncologist or pediatric neuropsychologist) and

either mailed a packet of questionnaires or approached by a research assistant at a

regularly scheduled clinic visit. Participants were offered a $10 gift card to a local

superstore as compensation for their participation. All participants were provided a

detailed cover letter from the child's attending oncology physician explaining the study

and a packet of questionnaires (as well as directions for completing them). Participants

approached at regularly scheduled clinic appointments were verbally provided

information about the study procedures and rationale. If interested in participating,

consent procedures were completed using methods approved by the DUMC

Institutional Review Board (IRB). All participants were asked to return their

questionnaire packet in the included postage-paid and addressed envelope within 3

weeks. Upon return of the completed packet, a gift card was mailed to the participant

along with a letter of appreciation. Illness and treatment-related information was

verified by chart review. Participation in the study took approximately sixty minutes.

2.1.2 Mothers of Healthy Children

Mothers in the control group were recruited from the community using several

IRB-approved mechanisms, including approaching potential participants in a local

pediatric practice and posting advertisements. Participants were offered a $10 gift card

to a local superstore as compensation for their participation. Mothers of children with a

chronic or serious medical or mental illness were excluded from participating. In the

pediatric practice, eligible mothers were approached by a research assistant at their

child's regularly scheduled well-child visit. The study and rationale were explained to

the mother and eligibility criteria were verified. If interesting in participating, they

completed consent procedures using methods approved by the DUMC IRB. The

participants were provided the packet of questionnaires (along with directions for

completing them) and asked to consider their parenting experience with their oldest

child under the age of 17 when responding. If the participant was not able to complete the questionnaire packet during her child's visit, she was provided with a postage-paid and addressed envelope in order to return her questionnaires.

Healthy control participants were also recruited via an IRB approved advertisement that was emailed, posted in local schools, and placed on Duke University Medical Center's clinical trials website (dukehealth.org). The advertisement provided the research assistant's contact phone number and email address. When mothers responded, the research assistant explained the study procedures and rationale and verified eligibility criteria. All eligible and interested mothers were mailed a packet containing a detailed cover letter from the research assistant explaining the study, two copies of the consent form and a packet of questionnaires (as well as directions for completing them). Participants were asked to return their questionnaire packet in the included postage-paid and addressed envelope within three weeks. Each participant was provided with the name and phone number of the Principal Investigator and project coordinator in case of questions or concerns. Upon return of the completed packet, a gift card was mailed to the participant along with a letter of appreciation.

2.2 Participants

2.2.1 Mothers of Pediatric Cancer Survivors

English-speaking mothers of survivors of pediatric cancer between the ages of one and seventeen who had been off all treatment and medically stable for at least one year were eligible for participation. An initial mailing was sent to approximately 60 survivors seen in our off-therapy clinic that met initial eligibility criteria; 24 (40%) returned their packets of questionnaires. In addition, a convenience sample of 74 mothers of survivors were approached in clinic and 70 (95%) agreed to participate in the study and were consented. Reasons given for not participating were lack of time (N = 3) and lack of interest (N = 1). Of the 70 mothers consented in clinic, 40 (57%) mothers returned completed packets. One mother withdrew after consenting due to a recurrence of her child's cancer. The overall response rate was 47.8% (64 out of 134).

The final sample of 64 mothers of survivors was 41.1 years of age (range 28 – 57; SD = 5.98); the survivors (57.8% male) were 11.5 years of age (range 3 – 17; SD = 3.13) and 6.1 years (range 1 – 17 years; SD = 4.00) from completion of treatment. Diagnoses were varied, but the majority of the sample included survivors of either a brain tumor (56.7%) or leukemia (21.9%). Consistent with the make-up of DUMC's patient population, the majority of the sample was Caucasian. See Tables 1 and 2 for all demographic and treatment-related information.

2.2.2 Mothers of Healthy Children

English-speaking mothers of healthy children between the ages of one and seventeen were eligible for participation. Of the 120 mothers to whom the study was described, consent was obtained from a total of 111 (92.5%) mothers, 77 (70%) of whom returned completed packets. The final sample of 64 mothers of survivors was selected using the child age as a criterion in an attempt to age-match the two groups. In the final sample of 64 participants, the mean age of the mothers was 36.6 years of age (range 26 – 52; SD = 3.54), and their children (47% male) were 9.4 years of age (range 4 – 17; SD = 3.62). See Table 1 for all demographic-related information.

2.3 Measures

The following measures were used. It took approximately 60 minutes for respondents to complete the measures.

2.3.1 Predictors

Demographic Questionnaire. The demographic questionnaire was used to gather information about the mothers (age, education level, marital status) and about their child (gender, age, grade). In addition, respondents were asked about their specific relationship to the child and to identify the primary caregiver of the child (themselves or someone else). Mothers of cancer survivors also provided details of their child's medical history, including diagnosis and treatment, on this questionnaire; this data was verified by chart review. Mothers in both groups completed this measure.

Intensity of Treatment Rating Scale -- Revised (ITR-2; Werba et al., 2007). This scale

classifies pediatric oncology diseases and treatments into four groups (1 – 4), from

minimally intensive to most intensive. It has been shown to have good validity and

reliability, and has been used as a predictor variable in studies of adjustment in

survivors of pediatric cancer survivors and their families (e.g. Barakat, Kazak, Gallagher,

Meeske, & Stuber, 2000). This scale was used to classify the intensity of treatment for the

survivor group only.

Ways of Coping (Folkman & Lazarus, 1988b). This is a 65-item measure designed

to identify the thoughts and actions an individual has used to cope with a specific

stressful encounter. It measures coping processes and strategies including confrontive

coping, distancing, self-controlling, seeking social support, accepting responsibility,

escape-avoidance, planful problem solving, and positive reappraisal. Individuals

respond to each item on a four-point Likert scale, indicating the frequency with which

each strategy is used, from 0 (Does not apply and/or not used) to 3 (Used a great deal).

Factor analysis delineated two broad subcategories of coping: palliative and adaptive

(Folkman & Lazarus, 1980). *Palliative* coping is the sum of item scores from the emotion-

focused factors, such as distancing and escape-avoidance. *Adaptive* coping is the sum of

the problem-focused factors, such as planful problem solving and positive reappraisal.

Mothers in both groups completed the Ways of Coping. The *Palliative* subscale, *Adaptive*

subscale and the *Palliative:Adaptive* ratio were used for this study.

Family Environment Scale (FES; Moos & Moos, 1981). The FES is a widely used 90-

item true/false self-report instrument designed to assess family functioning. The FES

provides 10 individual scales of family functioning, which cluster into three empirically

derived higher-order components: *Supportive, Conflicted* and *Controlling* (Kronenberger &

Thompson, 1990). Mothers in both groups completed the FES. The *Supportive, Conflicted*

and *Controlling* subscales were used for this study.

2.3.2 Outcomes

Parenting Stress Index (PSI; Abidin, 1995)*.* The PSI consists of 120 items that

quantify parenting-specific stress across parent and child specific domains. Each

question is rated by respondents on a 7-point Likert-type scale of 1 (strongly agrees) to 7

(strongly disagree). Mothers in both groups completed the PSI. The *Child Domain* and

the *Parent Domain* subscales were used for this study.

Brief Symptom Inventory (BSI; Derogatis & Spencer, 1982). The BSI is a 53-item self-

report inventory for adolescents and adults designed to reflect a broad array of

psychological symptom patterns. It includes 9 symptom dimensions (somatization,

obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic

anxiety, paranoid ideation, and psychotocism). Both groups of mothers completed the

BSI. The subscales of *Depression, Anxiety*, and *Somatization* were used for this study.

Parent Experience of Child Illness (PECI; Bonner et al., 2006). The PECI is a 25-item

parent-report measure of the parent's illness-specific adjustment to their child's serious

or chronic illness. It was initially validated in a study of 149 parents of children with

brain tumors. An initial factor structure yielded 4 factors: *Guilt and Worry*, *Unresolved*

Sorrow and Anger, *Long-term Uncertainty* and *Emotional Resources*. Only mothers of

survivors completed the PECI.

3. Results

This study was powered based on the primary aim. The sample of 128 participants (64 per group) was associated with a power of at least .80 to detect group differences at a .05 level, as calculated using the medium effect-sizes (d= .5).

3.1 Primary Aim

3.1.1 Preliminary Analyses

Prior to hypothesis testing, a series of t-tests and chi-square analyses were performed to test for differences in the demographic variables of the two groups. The only differences to reach significance were maternal education level ($t(126)$ = 2.65, p < .01), maternal age ($t(126)$ = 4.54, p < .01) and child age ($t(126)$ = 3.19, p < .01) . Mothers of survivors were older and had fewer years of education than mothers of healthy children; the survivors were also older than the healthy children. Therefore, maternal age and education were controlled for in all subsequent analyses by entering them as continuous covariates in multivariate analyses of covariance (MANCOVA). Child age and maternal age were significantly correlated (r = .55, p < .01); thus, only maternal age was included.

In order to characterize the two groups, they were compared on 5 predictor variables of interest, the Family Environment Scale (FES) *Conflicted*, *Controlling* and *Supportive* subscales and the Ways of Coping (WAYS) *Palliative* and *Adaptive* coping subscales using two MANCOVAs, one for the FES subscales and one for the WAYS

subscales, including maternal age and maternal education as continuous covariates

(Table 3).

In regards to family functioning, mothers of survivors report significantly higher

scores on the *Controlling* ($F(3, 114) = 7.41$, $p < .05$) subscale of the FES. No significant

differences were reported on the *Conflicted* ($F (3, 114) = .052$, $p = NS$) or *Supportive* ($F(3,$

$114) = 1.76$, $p = NS$) domains. No significant differences were reported on the *Palliative*

($F(3, 114) = .10$, $p = NS$), *Adaptive* ($F(3, 114) = .00$, $p = NS$) or *Palliative:Adaptive* ratio ($F(3,$

$114) = .003$, $p = NS$) subscales of the Ways of Coping.

3.1.2 Hypothesis Testing

The primary aim hypothesized that mothers of survivors would exhibit poorer

psychological adjustment when compared to demographically similar peers. In order to

test the specific aim, the two groups were compared on five outcome indices of

psychological adjustment: the Parenting Stress Index (PSI) *Child Domain* and *Parent*

Domain and the Brief Symptom Inventory (BSI) *Anxiety*, *Depression* and *Somatization*

subscales. Two multivariate analyses of covariance (MANCOVA) were performed, one

for the PSI subscales and one for the BSI subscales (Table 3). As noted above, maternal

age and maternal education were included covariates along with group status.

Compared to mothers of healthy children, mothers of pediatric cancer survivors

reported significantly higher levels of parenting stress in the *Child Domain* ($F(2, 116) =$

4.00, $p < .05$). No differences were reported between the groups on parent-specific parenting stress ($F(2, 116)= 0.07$, $p = $ NS) (see Figure 2). A MANCOVA was conducted comparing the two groups on the subscales that comprise the *Child Domain* of the PSI, again controlling for maternal age and education (Table 4). *Child Demandingness* ($F(6, 112) = 7.33$, $p < .025$), *Child Mood* ($F(6, 112) = 3.95$, $p = .049$), and *Child Acceptability* ($F(6, 112) = 16.0$, $p < .05$) were all significantly higher in mothers of survivors as compared to mothers of healthy children.

On the Brief Symptom Inventory (BSI), mothers of cancer survivors reported significantly higher levels of *Somatization* ($F(3, 114) = 7.33$, $p < .017$) than mothers of healthy children. In addition, there was a trend towards significance on the *Anxiety* subscale ($F(3, 114) = 3.73$, $p = .056$), with mothers of survivors reporting more symptoms of anxiety than mothers of healthy children. Of note, both groups reported elevated levels of anxiety symptoms; 52 (85.0%) of mothers of survivors fell in the at-risk or clinically significant range (≥ 60), while 43 (72.9%) of mothers of healthy children fell in the at-risk or clinically significant range (≥ 60). No significant differences were reported on symptoms of depression between groups ($F(3, 114) = 2.41$, $p = $ NS) (see Figure 3).

The PECI was only completed by mothers of cancer survivors; therefore, no comparative analyses were conducted between groups. However, the mean PECI scores

reported are commensurate with the mean scores reported by mothers of off-treatment

pediatric cancer patients in previous research (Hutchinson et al., in press).

In sum, significant differences were found between mothers of pediatric cancer

survivors and mothers of healthy children. Specifically, mothers of survivors report

higher levels of parenting stress, particularly related to their perceptions of the child's

mood, demandingness and acceptability. In addition, mothers of survivors report more

symptoms of general distress, such as somatization and anxiety, than mothers of healthy

children, than mothers of healthy children.

3.2 Secondary Aim

3.2.1 Preliminary Analyses

3.2.1.1 Correlations

Prior to hypothesis testing, a correlation matrix of treatment-related predictors

(treatment intensity, diagnosis) was generated in order to assess for multicolinerarity (*r*

≥ .7) and to assess for the relationship between demographic and treatment variables

with outcome variables (Table 5). Notably, the treatment and demographic predictors

were not significantly correlated with the outcome measures, with two exceptions:

mother's education level and PECI *Emotional Resources* were positively correlated (*r* =

.28, *p* < .05) and child's age at diagnosis and BSI *Anxiety* were negatively correlated (*r* = -

.30, *p* < .05). In addition, a correlation matrix was generated to assess for the relationship

between outcome variables (Table 6). Of note, the four PECI subscales were strongly associated with the PSI *Child* and *Parent Domains*, but not the BSI subscales.

Further, two correlation matrices were generated to assess for the relationship between predictor variables, family functioning and coping, and outcome measures of psychological adjustment (Tables 7 and 8). Results revealed significant positive correlations between the Conflict subscale of the FES and the PSI *Child* ($r = .33$, $p < .05$) and *Parent* ($r = .43$, $p < .01$) *Domains*, as well as the BSI *Depression* ($r = .28$, $p < .05$), *Anxiety* ($r = .33$, $p < .05$) and *Somatization* ($r = .31$, $p < .05$) subscales. Similarly, the *Supportive* subscale of the FES was negatively correlated with the PSI *Child* ($r = -.40$, $p < .01$) and *Parent* ($r = -.47$, $p < .01$) *Domains*, as well as the BSI *Depression* ($r = -.32$, $p < .05$) and *Anxiety* ($r = -.28$, $p < .05$) subscales. As expected, the FES *Supportive* was significantly positively correlated with PECI *Emotional Resources* ($r = .28$, $p < .05$).

The PECI was also significantly correlated with the Ways of Coping. Specifically, *Palliative* coping was significantly positively correlated with *Guilt and Worry* ($r = .23$, $p < .05$) and *Unresolved Sorrow and Anger* ($r = .34$, $p < .01$). The *Palliative:Adaptive* coping ratio was also positively correlated *Unresolved Sorrow and Anger* ($r = .32$, $p < .05$) and *Long-term Uncertainty* ($r = .26$, $p < .05$), and negatively correlated with *Emotional Resources* ($r = -.29$, $p < .05$). Further, a correlation matrix was generated to look at specific coping mechanisms within the *Palliative* and *Adaptive* Subscales (Table 8). This matrix revealed significant

relationships between the *Escape-Avoidance* coping mechanism and PSI *Child* (r = .41, p < .01) and *Parent* (r = .27, p < .05) *Domains*, as well as all four subscales of the PECI: *Guilt and Worry* (r = .44, p < .01), *Unresolved Sorrow and Anger* (r = .47, p < .01), *Long-term Uncertainty* (r = .37, p < .01) and *Emotional Resources* (r = -.38, p < .01).

3.2.1.2 Brain Tumor vs. Other Cancer

Given the established relationship between diagnosis, treatment intensity and child outcomes, the participants were spilt into two groups for further comparison: mothers of brain tumor survivors (n = 34; 56.7%) and mothers of other cancer survivors (n = 30; 43.3%). The two groups differed significantly on time since completing treatment (t (62) = -2.01, p < .05); the brain tumor survivors were off-treatment for an average of 5.2 years (SD = 3.53) and the other cancer survivors were off-treatment for an average of 7.2 years (SD = 4.35). As expected, the two groups also differed significantly on treatment intensity (χ^2(3, N = 64) = 14.43, p < .01), with the brain tumor group reporting significantly more children receiving high intensity treatments (n = 27, 77%) than the other cancer group (n = 12, 41%).

A series of MANCOVAs were conducted entering diagnostic category (brain tumor or other cancer) and treatment intensity as fixed factors and controlling for time off treatment as a covariate (Table 9). For this analysis, treatment intensity was transformed into a dichotomous variable, with Low Intensity and Moderate Intensity

forming one group (Low; n = 25) and High Intensity and Very High intensity forming the other group (High; n = 39). On the BSI, there was a significant main effect for diagnostic category ($F(2, 53) = 4.28$, $p < .05$), such that the mothers of other cancer survivors ($M = 64.7$, $SD = 13.26$) reported significantly higher scores on the *Somatization* subscale than mothers of brain tumor survivors ($M = 55.9$, $SD = .9.60$). Notably, no other significant main effects were found for diagnostic category or treatment intensity on the PSI *Child* or *Parent Domains*, the BSI *Depression, Anxiety* and *Somatization* subscales or the PECI *Guilt and Worry, Unresolved Sorrow and Anger, Long-term Uncertainty* and *Emotional Resources* subscales.

However, there were two significant interaction effects for diagnostic category and treatment intensity (see Figures 3 and 4). Analyses revealed a significant interaction effect at the multivariate level on the PECI *Unresolved Sorrow and Anger* subscale ($F(2, 53) = 6.75$, $p < .05$). Specifically, mothers of other cancer survivors exhibited more *Unresolved Sorrow and Anger* then mothers of brain tumor survivors only when their children received high intensity treatment (see Figure 3). Analyses also revealed a significant interaction effect at the multivariate level on the PECI *Emotional Resources* subscale ($F(2, 53) = 4.52$, $p < .05$). Specifically, mothers of other cancer survivors exhibited less *Emotional Resources* than mothers of brain tumor survivors only when their children received high intensity treatment (see Figure 4).

3.2.2 Hypothesis Testing

In the second aim, it was hypothesized that individual variables, such as coping and family functioning, would be strongly associated with psychological outcomes of parents of survivors. In order to examine possible predictors of psychological adjustment in mothers of cancer survivors, multiple hierarchical linear regression analyses were conducted with the five psychological adjustment outcomes from the primary aim (PSI and BSI subscales*)*, along with the four PECI subscales (Tables 10-18). Given that this study is currently powered based on the primary aim, power to test the secondary aim is limited. Therefore, the total number of predictors entered into any model was limited to six. In Step 1, demographic, illness and/or treatment variables that have been previously shown to influence the dependent variable were entered using forced entry. In Step 2, variables of interest to the current study predicted to influence the dependent variable were entered, also using forced entry.

The regression model for the *Child Domain* of the PSI revealed that demographic variables accounted for a significant, though modest, portion of the variance in parenting stress scores ($\Delta R^2 = .13$). In particular, marital status ($\beta = -.31$, $p < .05$) reached statistical significance as a predictor of parenting stress. However, a much larger portion of the variance ($\Delta R^2 = .27$) was accounted for by supportive family functioning ($\beta = -.43$, $p < .01$) and the use of escape-avoidance coping mechanisms ($\beta = .34$, $p < .01$). The overall

model ($F(2, 46) = 5.07$, $p < .001$) accounted for approximately 40% of the total overall variance in child specific parenting stress. The regression model for the *Parent Domain* of the PSI revealed a similar pattern. Specifically, marital status ($\beta = -.31$, $p < .05$) and supportive family functioning ($\beta = -.43$, $p < .01$) accounted for a statistically significant portions of the variance in parent-specific parenting stress, with the overall model ($F(2, 46) = 3.98$, $p < .01$) predicting 34% of the total variance.

Regression models for the BSI *Depression*, *Anxiety* and *Somatization* subscales failed to reach significance, as did the specific demographic, family functioning and coping predictor variables within the models.

The hierarchical regression model for the four PECI subscales was conducted using three illness and treatment variables as predictors in Step 1: child's age at diagnosis, time off-treatment and treatment intensity. Across all four subscales, these factors accounted for a small proportion of the variance in parent adjustment; however, no individual illness or treatment predictor reached statistical significance. The model also included three individual predictors hypothesized to influence parental adjustment: *Escape-Avoidance* coping, *Child Demandingness* (a subscale of the PSI *Child Domain*), and the *Supportive* subscale from the FES.

For the three PECI distress scales (*Guilt and Worry*, *Unresolved Sorrow and Anger* and *Long-term Uncertainty*), the overall model reached statistical significance.

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Specifically, the model accounted for 41% of the overall variance in *Guilt and Worry* (F (3, 47) = 5.33, $p < .01$), 41% of the overall variance in *Unresolved Sorrow and Anger* (F (3, 47) = 5.33, $p < .01$), and 33% of the overall variance in *Long-term Uncertainty* (F (3, 47) = 3.90, $p < .01$). *Escape-Avoidance* and *Child Demandingness* reached statistical significance as individual predictors. For the fourth PECI subscale, *Emotional Resources*, the overall model was also statistically significant (F (3, 47) = 3.26, $p < .01$), accounting for 29% of the overall variance in *Emotional Resources*. The only individual predictor to reach statistical significance in this model was *Child Demandingness* ($\beta = -.38$, $p < .01$).

In sum, hierarchical regression modeling revealed escape-avoidance coping and family supportiveness to be important individual variables in predicting parenting-related psychological adjustment in mothers of pediatric cancer survivors.

4. Discussion

The current study assessed the psychological adjustment of mothers of pediatric cancer survivors. As hypothesized, parents of survivors reported significantly higher levels of parenting stress and general psychological distress than mothers of healthy children. In addition, emotion-focused coping and supportive family functioning predicted a significant portion of the variance in parental adjustment, over and above demographic, illness or treatment variables. It also appears that caregivers of "other" (non-brain tumor) cancer survivors that receive high-intensity treatment are at the highest risk for adjustment difficulties. These findings serve to document the ongoing needs of these caregivers and identify salient targets for intervention aimed at reducing ongoing distress.

4.1 Evidence of Ongoing Distress

4.1.1 Parenting Stress

In regards to parenting stress, significant differences between groups were largely in the child domain of stress. That is, individual factors related to the child, as opposed to the parent, contribute to the difference in overall parenting stress between groups. High scores in the child domain may be associated with children who display qualities that make it difficult for parents to fulfill their parenting role; indeed, the child domain is typically more elevated than the parent domain in families of disabled

children (Abidin, 1995). In the current study, parent perceptions regarding three key child attributes were identified: *Acceptability*, *Mood*, and *Demandingness*.

Elevated scores on the *Acceptability* subscale are produced when the child possesses physical, intellectual and emotional characteristics that do not match the expectations the parent had for their child. Given the higher rates of physical, cognitive and learning disabilities that emerge as late effects in pediatric cancer survivors, it is to be expected that parents of survivors will face new and challenging qualities of their child that were not anticipated prior to their diagnosis. Indeed, when faced with the long-term consequences of cancer, parents often grieve for the loss of the child they expected to have.

High scores on the *Mood* subscale are associated with children whose affective functioning shows evidence of dysfunction; these children are more likely to be unhappy, depressed, and to frequently cry. Similarly, child *Demandingness* is elevated when the parent experiences the child as placing many demands on him or her. Pediatric cancer survivors often place additional caregiving demands on their parents as a result of their ongoing medical and cognitive issues that require long-term follow-up care and close vigilance. This is consistent with findings that parents of off-treatment pediatric cancer patients report similar levels of caregiving burden as parents of on-treatment patients (Hutchinson et al., in press).

4.1.2 General Distress

Mothers of survivors reported significantly more *Somatization*, defined as distress resulting from somatic symptoms. Consistent with findings in other illness groups (Lawoko, Soares, Lawoko, & Soares, 2006), this finding suggests that parents serving as caregivers may be more likely to express their emotional state through physical symptoms. Given the high demands on caregivers' time and energy, it is also likely that their self-care (e.g. getting adequate rest, nutrition and exercise) is sub-optimal, which can also contribute to somatic complaints. Although mothers of survivors reported more symptoms of depression and anxiety, these differences failed to reach statistical significance. Taken together, these results support our hypothesis that the range of distress symptoms experienced by these caregivers may not be captured adequately by conventional measures of distress. Rather, functioning of these caregivers is probably best assessed using a combination of general psychiatric measures and those designed specifically to evaluate illness-related psychosocial functioning.

4.1.3 Diagnosis and Treatment Intensity

Neither diagnosis nor treatment intensity were independently predictive of parental outcomes. However, there was a significant interaction between diagnosis (brain tumor vs. other cancer) and treatment intensity. As such, it appears that parents of children with other (non-brain tumor) diagnoses that receive high intensity treatment

are at highest risk for long-term adjustment difficulties, such as *Unresolved Sorrow and*

Anger. This is likely due to the expectations parents develop upon receiving their child's

diagnosis. When children receive a diagnosis of a brain tumor, they are given the

expectation of physical and cognitive late effects, and therefore may be better prepared

for the post-treatment reality. Whereas with other cancers, such as leukemia, parents are

generally given very high survival rates and less information regarding possible late

effects; thus, they have higher expectations for a good outcome for their child. These

parents may be less prepared, and experience more adjustment difficulties, when late

effects do emerge.

4.2 Intervention Targets

Result of multiple regression analyses indicate that demographic, illness and

treatment variables predict only a modest portion (2 – 13%) of the variance in parental

adjustment across measures. Rather, individual variables, including family functioning

and coping mechanisms, predicted a much larger portion (25 – 39%) of the variance in

parental adjustment on measures of parenting and illness specific distress. These

findings are consistent with past research with other illness groups, similarly guided by

the transactional stress and coping model. These results identify family functioning and

coping style as key targets for interventions with caregivers.

4.2.1 Coping

Based on past research guided by the Transactional Stress and Coping model, it was hypothesized that *Palliative* (emotion-focused) coping, and a higher ratio of *Palliative* coping to *Adaptive* (problem-focused) coping would be associated with poorer outcomes. Correlation analyses supported this hypothesis; *Palliative* coping was associated with higher levels of *Guilt and Worry* and *Unresolved Sorrow and Anger*, while a higher *Palliative:Adaptive* ratio was associated with higher levels of *Guilt and Worry*, *Long-term Uncertainty* and fewer *Emotional Resources*.

The results of the current study further identify *Escape-avoidance* coping to be associated with poorer outcomes, particularly those related to parenting and illness-related distress. *Escape-avoidance* coping includes wishful thinking, hoping for a miracle, or avoiding thoughts and emotions related to the stressor by eating, drinking, sleeping or smoking. This coping mechanism predicted a statistically significant portion of the variance in parenting stress, *Unresolved Sorrow and Anger*, *Guilt and Worry*, and *Long-term Uncertainty*. As such, emotion-focused coping, particularly *Escape-avoidance* coping, is likely to be a salient target for intervention for parents of pediatric cancer survivors. Interventions reducing the use emotion-focused coping are likely to result in less parenting stress, guilt and worry, sorrow and anger, and long-term uncertainty.

4.2.2 Family Functioning

It was hypothesized that family functioning would be associated with parental adjustment. The results of current study indicate that higher levels of family conflict and lower levels of family support are associated with increased levels of parenting stress and general distress, including depression, anxiety and somatization. Moreover, family supportiveness predicted a statistically significant portion of the variance in the child and parent domains of parenting stress. Thus, interventions aimed at improving family supportiveness and decreasing family conflict are likely to result in less family stress and fewer symptoms of general distress. More specifically, interventions should target increasing family cohesion and expressiveness, while reducing family conflict.

While the *Controlling* subscale of family functioning was not significantly associated with parental adjustment outcomes, it was reported to be significantly higher by mothers of pediatric cancer survivors as compared to mothers of healthy children. Thus, families of survivors are more likely to be characterized by high control, high achievement orientation, and low independence. It is not clear from these results whether high levels of control are adaptive or maladaptive for families of survivors. It is likely that children's physical or cognitive late effects may require more boundaries and monitoring by parents. However, over-control or over-involvement may disrupt the process of healthy development in children and adolescents.

As a consequence of the uncertainty and anxiety about the recurrence of the disease or the appearance of emerging late effects, parents of survivors tend to perceive their child as a vulnerable child, both in their physical condition and psychosocial development. Many parents attempt to compensate for the difficult events during treatment and protect their child from potentially upsetting or stressful events. Indeed, many parents of survivors report placing more physical limitations on their child, while setting less psychological demands (Van Dongen-Melman et al., 1998). This is in contrast to normal development, in which physical limitations are reduced with increasing age, whereas psychological demands increase. Families able to set demands for their children, thus establishing a more controlling family environment, appeared to be the most balanced and had better child outcomes than families unable or unwilling to do so (Van Dongen-Melman et al., 1998).

Parents of cancer survivors often lack the typical transition of increased autonomy that occurs when a child reaches young adulthood. Indeed, many parents maintain an extended period of active parenting that may include accompanying their child to medical appointments, providing therapy at home, remedial teaching, psychosocial support or extended financial support (Ressler et al., 2003; Van Dongen-Melman et al., 1998). This extended caregiving may interfere with the long-term goals and expectations for the future of both parents and children. One study of identity in

adolescent survivors of childhood cancer (Madan-Swain et al., 2000) found a greater frequency of survivors in the "foreclosed" identity status than their healthy peers. The foreclosure state of identity formation occurs when one prematurely commits to certain goals, values and beliefs, often internalizing the values of significant adults in their lives. Healthy adolescent identity formation requires individuation, a process in which one differentiates self from parent without totally disconnecting from the family. Reaching identity achievement status requires a period of questioning or reflection that may be particularly challenging for cancer survivors living in a very controlling family environment.

Notably, neither family functioning nor coping mechanisms predicted any significant portion of the variance in general distress (depression, anxiety or somatization). These are much broader constructs, likely to be impacted by a wider variety of factors than measures of parenting and illness specific distress. Moreover, measures of general distress may lack sensitivity to detect the specific distress experienced by parents of survivors, thus lack variability associated with individual factors such as coping and family functioning.

4.3 Limitations

While the current study serves to document the ongoing needs and identify key intervention targets for caregivers of pediatric cancer survivors, it does have limitations.

First, the group of survivors is very diverse in regards to age, time off treatment, diagnosis and treatment. Although this diversity is generally accepted in research with pediatric cancer survivors in order reach sufficient sample sizes, it introduces significant variability into our analyses that may limit power to analyze relationships between variables. In addition, the survivor group and healthy group differed significantly on several demographic variables, including maternal education, maternal age and child age. While these factors were controlled for statistically, an ideal study would have demographically matched samples.

All data in this study (with the exception of illness and treatment data, which was verified by medical chart review) was self-reported by the participants. Thus, it is expected that there may be some bias in their responses, including correlations between measures. However, of the three outcome measures used in this study, only the PSI and the PECI were significantly related. The BSI was not significantly correlated with either the PSI or the PECI, which provides evidence for sufficient method variance.

This study was designed to be cross sectional; as such, it does not provide data regarding the trajectory of distress over time on an individual level. Indeed, to understand the *process* of stress and coping in caregivers will require a longitudinal design. In addition, the current study lacks specific data regarding children's level of functioning, such as IQ or adaptive functioning. Given the robust relationship between

treatment intensity and late effects, treatment intensity served as a proxy for late effects; however, each child develops differently. Therefore, future studies will also need to include information regarding survivors' medical and cognitive late effects, which likely impact caregiver distress.

4.4 Future Directions

4.4.1 Benefit Finding

There is a growing body of literature exploring the role of positive emotions in the coping process (Folkman, 2008). Specifically, the constructs of "posttraumatic growth" and "benefit finding" have recently received attention in the cancer survivorship literature (Barakat, Alderfer, & Kazak, 2006; Kim et al., 2007; Stuber, 2006). To further explore the relationship between benefit finding and parental adjustment, pilot qualitative data was collected from participants in the current study. Participants responded to the question, "Have there been any benefits, or positive outcomes, as a result of your experience having a child with cancer? If so, how would you describe them?" in written form. Efforts are currently underway to code the qualitative data according to themes within the domains of social support (e.g. the cancer community, closer family), personal growth (e.g. reprioritized values, increased faith) and child-specific benefits (e.g. child is stronger, improved parent-child bond). Once coded, the

data will be analyzed utilizing a mixed methods approach to assess relationships

between themes and quantitative measures of parental adjustment (BSI, PECI and PSI).

4.4.2 Longitudinal Methodology

There are no prospective, longitudinal studies to date examining parental stress

and coping as a process over the illness trajectory. These data are critical to developing

interventions for parents at key points in the cancer process. Very preliminary analyses

using the cross-sectional data from the current study suggests that parental distress

peaks in the 6 – 10 years post-treatment time frame. Clinically, parents have been

observed to experience a "honeymoon period" immediately post-treatment, associated

with relatively lower levels of distress. We hypothesize that caregiver stress may

increase again once late effects, such as declines in cognitive abilities, begin to emerge

and demonstrate an impact on learning. To fully understand the trajectory of distress

over time on an individual level will require a longitudinal design with a comprehensive

set of psychological and illness-specific measures administered at frequent intervals

over the course of the illness.

4.4.3 Caregiver Intervention

Currently, no interventions exist for parents of long-term cancer survivors. The

results of the current study clearly document that parental distress is ongoing. Thus, a

critical next step in this line of research is developing interventions to improve parents'

ability to cope with the demands placed on them as caregivers. Successful interventions would ultimately improve the quality of life for both the caregiver and the survivor. Given the role of family functioning and escape avoidance coping in predicting parental outcomes, these two constructs appear to be salient targets for intervention. Escape-avoidance coping may be best addressed using acceptance and commitment therapy (ACT), which targets experiential avoidance. Recently, the principals of ACT (i.e., mindfulness and acceptance) have been successfully applied in medical settings (P. Robinson, Gregg, Dahl, & Lundgren, 2005), with pediatric pain populations (Wicksell & Greco, 2008), and in parents of children diagnosed with autism (Blackledge & Hayes, 2006). Therefore, there is evidence to support a pilot study applying ACT with pediatric cancer survivors and their parents.

4.5 Conclusion

In summary, this work provides evidence that childhood cancer has a distinct and persistent late psychosocial impact on parents of survivors. Moreover, parenting and illness specific distress in this population may not be adequately captured on traditional measures of psychopathology. Guided by the transactional stress and coping model, this study identifies family functioning and coping mechanisms as individual factors that predict a significant portion of the variability in parental outcomes.

Interventions designed to address these factors may be beneficial to both the survivors and their families.

Parents of survivors face considerable stressors. They must provide emotional support to their children, while simultaneously processing their own fears and emotions. They must also face the uncertainty of newly emerging neurocognitive late effects as a result of their children's treatment and act as advocates for their children to ensure they are receiving adequate services in the educational system. Currently, little is being done to assist these parents in coping with the unique set of stressors associated with cancer experience. Targeting those variables that are known to predict positive adaptation to the stress of parenting a cancer survivor will serve to benefit caregivers and survivors alike. Indeed, while parenting stress is clearly detrimental for caregivers, it also impacts the survivors themselves. Parents of survivors continue to take an active parenting role even as survivors enter young adulthood; their behavior will have an ongoing influence over their children's development. The increasing number of survivors provides a unique opportunity to develop and implement interventions to provide families with a sense of hope and meaning from an otherwise difficult experience.

Appendix A: Tables

Table 1: Demographic Information by Group

	Survivor		Healthy	
	$M \pm SD$	N (%)	$M \pm SD$	N (%)
Mother Factors				
Age (years)*	41.1 ± 5.98		36.6 ± 5.33	
Race				
Caucasian		60 (93.8)		53 (82.8)
African-American		3 (4.7)		8 (12.5)
Asian-American		1 (1.6)		3 (4.7)
Education (years)*	15.4 ± 1.77		16.3 ± 1.83	
High School		8 (12.5)		2 (3.1)
College		46 (71.9)		37 (57.8)
Graduate School		10 (15.6)		25 (39.1)
Marital Status				
Married/Partnered		53 (82.8)		52 (81.3)
Single/Divorced/Widowed		11 (17.2)		12 (18.7)
Child Factors				
Age (years)*	11.5 ± 3.75		9.4 ± 3.62	
Gender				
Male		37 (57.8)		34 (53.1)
Female		27 (42.2)		30 (46.9)
Race				
Caucasian		57 (89.1)		49 (76.6)
African-American		3 (4.7)		9 (14.1)
Asian-American		2 (3.1)		4 (6.3)
Biracial		2 (3.1)		2 (3.2)

Note. The two groups differed significantly on mother education, mother age and child age; * $p < .05$.

Table 2: Illness and Treatment Data for Survivor Group

	$M \pm SD$	N (%)
Age at diagnosis	4.0 ± 3.17	
Months since diagnosis	72.7 ± 48.07	
Diagnosis		
Brain Tumor Total		34 (56.7)
Ependymoma		8 (23.5)
Astrocytoma		8 (23.5)
Optic Glioma		7 (20.5)
Medulloblastoma		6 (17.6)
Other		5 (14.7)
Other Cancer Total		30 (43.3)
ALL		14 (46.7)
Neuroblastoma		6 (20.0)
AML		2 (6.7)
Retinoblastoma		2 6.7)
Other		6 (20.0)
Treatment Intensity		
Least Intensive (Surgery only)		2 (3.1)
Moderately Intensive (1 Treatment modality)		23 (35.9)
Very Intensive (>1 Treatment modality)		32 (50.0)
Most Intensive (Relapse protocol, BMT)		7 (10.9)

Table 3: Questionnaire Means and Standard Deviations by Group

	Survivors $M \pm SD$	Healthy $M \pm SD$	F
Outcome Measures			
PSI			
Child Domain	109.8 ± 29.09	100.3 ± 26.06	4.00*
Parent Domain	128.2 ± 25.03	124.7 ± 28.84	.710
BSI			
Depression	59.7 ± 11.32	55.9 ± 11.61	2.41
Anxiety	62.1 ± 10.80	58.3 ± 13.02	3.73
Somatization	60.3 ± 12.29	52.8 ± 12.70	7.33**
PECI			
Guilt & Worry	1.51 ± .682		
Unres. Sorrow/Anger	1.23 ± .789		
LT Uncertainty	1.55 ± .892		
Emo. Resources	2.85 ± .697		
Predictor Variables			
FES			
Conflicted	-62.6 ± 31.37	-61.9 ± 28.96	.052
Controlling	120.3 ± 23.44	107.3 ± 30.36	7.41**
Supportive	260.6 ± 47.65	276.0 ± 39.65	1.76
Ways			
Palliative	24.9 ± 11.51	24.3 ± 11.67	.102
Adaptive	31.7 ± 10.86	32.3 ± 13.02	.000
Pall:Adapt	0.83 ± 0.39	0.81 ± 0.37	.003

 * $p < .05$; ** p < .01

PSI, Parenting Stress Index; BSI, Brief Symptom Inventory; FES, Family Environment Scale; Ways, Ways of Coping; PECI, Parent Experience of Child Illness; The scores for the BSI are *T*-scores with $M = 50$ and $SD = 10$. All others scores are means and standard deviations for the sample.

Table 4: PSI Child Domain Subscales: Means and Standard Deviations by Group

	Survivors M ± SD	Healthy M ± SD	F
PSI Child Domain			
Distractibility/ Hyperactivity	23.3 ± 5.26	23.2 ± 5.46	.170
Adaptability	25.8 ± 6.87	24.7 ± 7.17	1.37
Reinforces Parent	12.0 ± 6.55	10.8 ± 4.92	.125
Demandingness	21.8 ± 6.16	19.1 ± 6.38	7.33**
Mood	11.9 ± 5.00	10.4 ± 4.10	3.95*
Acceptability	16.2 ± 5.51	12.2 ± 4.10	16.0**

* $p < .05$; ** p < .01

PSI, Parenting Stress Index; Scores are means and standard deviations for the sample.

Table 5: Correlations between Demographic and Illness Factors and Outcome Measures within Survivor Group

	Maternal age	Maternal education	Child age	Child age at diagnosis	Months off treatment	Treatment Intensity
PSI Child Domain	-.168	-.103	.079	.040	.026	.182
PSI Parent Domain	.024	-.062	-.029	-.066	.027	.182
BSI Depression	.091	.007	.102	-.205	.152	-.051
BSI Anxiety	-.087	-.048	-.040	-.301[*]	.158	-.058
BSI Somatization	.134	-.076	.157	-.152	.147	-.078
PECI Guilt/Worry	-.074	.059	.100	-.014	.073	.073
PECI Sorr/Anger	-.062	-.128	.107	-.031	.052	.246
PECI LT Uncertainty	-.021	-.064	.182	-.241	-.046	.139
PECI Emotional Resources	.082	.279[*]	-.134	.052	-.149	-.059

Pearson Correlations, N = 64; * $p < .05$, ** $p < .01$, two tailed

Table 6: Correlations between Outcome Variables within the Survivor Group

	PECI				BSI		
	Guilt/ Worry	Sorr/ Anger	LT Uncer	Emo Res	Dep	Anx	Somat
PSI Child	.402**	.431**	.403*	-.491**	.239	.264*	.034
PSI Parent	.399**	.483**	.378**	-.391**	.399*	.400**	.313*
BSI Dep	.235	.252	.141	-.181			
BSI Anxiety	.240	.295*	.042	-.340**			
BSI Somat	.201	.234	.018	-.123			

Pearson Correlations, N = 64; * $p < .05$, ** $p < .01$, two tailed

Table 7: Correlations between Predictor and Outcome Variables within the Survivor Group

	PSI		BSI			PECI			
	Child	Parent	Dep	Anx	Somat	Guilt/ Worr	Sorr/ Anger	LT Uncer	Emo Res
FES Conflict	.332*	.432**	.281*	.329*	.311*	.168	.157	.086	-.162
FES Control	.230	.088	.023	.021	-.052	-.066	-.253	-.200	.167
FES Support	-.400**	-.472**	-.320*	-.280*	-.210	-.116	-.162	-.111	.275*
Ways: Palliative	.214	.157	.053	.058	.048	.232*	.337**	.243	-.192
Ways: Adaptive	.015	-.083	-.060	-.128	.041	-.046	-.108	-.077	.165
Ways: Pall:Adapt	.119	.170	.233	.087	.074	.249	.322*	.264*	-.288*

Pearson Correlations, N = 64; * $p < .05$, ** $p < .01$, two tailed

Table 8: Correlations between Coping Mechanisms and Outcome Variables within the Survivor Group

	PSI		BSI			PECI			
	Child	Parent	Dep	Anx	Somat	Guilt /Wor	Sor/ Anger	LT Uncer	Emo Res
Palliative									
Distancing	-.050	-.118	-.070	-.060	-.109	.002	-.004	-.023	.123
Self-Controlling	.142	.203	.124	.012	.042	.290*	.291*	.190	-.136
Accepting Respons.	.114	.084	-.033	.032	-.051	.188	.200	.153	-.127
Escape-Avoidance	.414**	.265*	.217	.101	.130	.440**	.473**	.368**	-.380**
Adaptive									
Confront. Coping	.053	.056	-.146	.105	-.025	.129	.080	-.008	.042
Seek Social Support	-.055	-.101	-.065	-.080	.069	-.022	-.106	-.053	.130
Planful Problem Solving	.111	-.130	-.083	.013	-.055	-.176	-.098	-.147	.067
Positive Reapprais.	.111	-.130	.008	-.175	.063	-.026	-.111	.005	.150

Pearson Correlations, N = 64; * $p < .05$, ** $p < .01$, two tailed.

Table 9: Questionnaire Means and Standard Deviations by Diagnostic Category

	Brain Tumor	Other Cancer	
	$M \pm SD$	$M \pm SD$	F
Outcome Measures			
PSI			
Child Domain	110.0 ± 24.53	110.4 ± 33.87	0.016
Parent Domain	125.4 ± 21.73	131.4 ± 28.58	0.234
BSI			
Depression	57.2 ± 11.94	62.7 ± 10.37	1.378
Anxiety	61.4 ± 10.99	62.7 ± 10.85	0.008
Somatization	55.9 ± 13.26	64.7 ± 9.60	4.280[*]
PECI			
Guilt & Worry	1.38 ± .663	1.63 ± 696	1.616
Unres. Sorrow/Anger	1.19 ± .756	1.23 ± .733	0.032
LT Uncertainty	1.58 ± .949	1.52 ± .859	0.018
Emo. Resources	2.82 ± .681	2.88 ± .587	1.327

* $p < .05$; ** $p < .01$

PSI, Parenting Stress Index; BSI, Brief Symptom Inventory; PECI, Parent Experience of Child Illness

The scores for the BSI are T-scores with $M = 50$ and $SD = 10$. All others scores are means and standard deviations for the sample.

Table 10: Hierarchical Regression Analyses Predicting PSI Child Domain from Demographic and Individual Risk Factors

	Child Domain					
	B	*SE B*	Std β[a]	*t*	*F*	ΔR^2
Step 1					1.71	.13
Maternal Age	-.59	.710	-.11	-.82		
Maternal Education	-30	2.38	-.02	-.13		
Marital Status	-27.3	12.61	-.31	-2.16*		
Job Status	-7.8	8.54	-.12	-.92		
Step 2					5.07**	.27
Escape-Avoidance Coping	2.2	.78	.340	2.81**		
FES Supportive	-.26	.09	-.430	-3.01**		

a: Regression weights at entry into the model

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

Table 11: Hierarchical Regression Analyses Predicting PSI Parent Domain from Demographic and Individual Risk Factors

	Parent Domain					
	B	$SE\ B$	Std β[a]	t	F	ΔR^2
Step 1					1.25	.09
Maternal Age	-.23	.617	.052	-.82		
Maternal Education	.62	2.38	.045	-.13		
Marital Status	-23.0	10.96	-.310	-2.16*		
Job Status	-3.4	7.43	-.063	-.92		
Step 2					3.98**	.25
Escape-Avoidance Coping	1.25	.70	.340	1.80		
FES Supportive	-.26	.08	-.430	-3.34**		

a: Regression weights at entry into the model

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

Table 12: Hierarchical Regression Analyses Predicting BSI Depression from Demographic and Individual Risk Factors

	Depression					
	B	*SE B*	Std β[a]	*t*	*F*	Δ*R²*
Step 1					.94	.07
Maternal Age	.14	.273	.07	.50		
Maternal Education	.03	.913	.01	.03		
Marital Status	-3.44	4.840	-.11	-.71		
Job Status						
Step 2					1.67	.11
Escape-Avoidance Coping	.47	.339	.195	1.38		
FES Supportive	-.07	.038	-.269	-1.72		

a: Regression weights at entry into the model

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

Table 13: Hierarchical Regression Analyses Predicting BSI Anxiety from Demographic and Individual Risk Factors

	Anxiety					
	B	$SE\ B$	Std β^a	t	F	ΔR^2
Step 1					.78	.06
Maternal Age	-.10	.271	-.05	-.37		
Maternal Education	.08	.908	.01	.09		
Marital Status	-6.28	4.812	-.20	-1.30		
Job Status	3.21	3.260	.14	.98		
Step 2					.81	.03
Escape-Avoidance Coping	.13	.352	.06	.38		
FES Supportive	-.05	.040	-.19	-1.17		

a: Regression weights at entry into the model

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

Table 14: Hierarchical Regression Analyses Predicting BSI Somatization from Demographic and Individual Risk Factors

	Somatization					
	B	*SE B*	Std β[a]	*t*	*F*	ΔR^2
Step 1					1.05	.08
Maternal Age	.23	.297	.11	.76		
Maternal Education	-.20	.994	-.03	-.21		
Marital Status	-5.98	5.27	-.17	-1.13		
Job Status	4.74	3.569	.19	1.32		
Step 2					1.10	.05
Escape-Avoidance Coping	.55	.383	.21	1.43		
FES Supportive	-.01	.043	-.051	-.31		

a: Regression weights at entry into the model

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

Table 15: Hierarchical Regression Analyses Predicting PECI Guilt and Worry from Illness, Treatment and Individual Risk Factors

	Guilt and Worry					
	B	SE B	Std β[a]	t	F	ΔR^2
Step 1					.29	.02
Child age at Diagnosis	-.00	.034	-.02	-.10		
Time off-treatment	.00	.002	.09	.59		
Treatment Intensity	.16	.211	.11	.78		
Step 2					5.33**	.39
Escape-Avoidance Coping	.04	.019	.29	2.29*		
Child Demandingness	.06	.015	.50	3.89**		
FES Supportive	.00	.002	.13	1.04		

a: Regression weights at entry into the model

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

Table 16: Hierarchical Regression Analyses Predicting PECI Unresolved Sorrow and Anger from Illness, Treatment and Individual Risk Factors

	Unresolved Sorrow and Anger					
	B	SE B	Std β[a]	t	F	ΔR^2
Step 1					.97	.06
Child age at Diagnosis	-.01	.036	-.03	-.21		
Time off-treatment	.00	.002	.104	.67		
Treatment Intensity	.36	.220	.24	1.65		
Step 2					5.29**	.35
Escape-Avoidance Coping	.05	.020	.34	2.54*		
Child Demandingness	.05	.016	.42	3.22**		
FES Supportive	.00	.002	.10	.78		

a: Regression weights at entry into the model

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

Table 17: Hierarchical Regression Analyses Predicting PECI Long-term Uncertainty from Illness, Treatment and Individual Risk Factors

	Long-term Uncertainty					
	B	SE B	Std β[a]	t	F	ΔR^2
Step 1					1.44	.08
Child age at Diagnosis	.07	.044	.26	1.69		
Time off-treatment	.00	.003	.10	.64		
Treatment Intensity	.21	.267	.11	.78		
Step 2					3.90**	.25
Escape-Avoidance Coping	.06	.027	.29	2.11*		
Child Demandingness	.05	.021	.36	2.62*		
FES Supportive	.00	.003	.08	.64		

a: Regression weights at entry into the model

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

Table 18: Hierarchical Regression Analyses Predicting PECI Emotional Resources from Illness, Treatment and Individual Risk Factors

	Emotional Resources					
	B	*SE B*	Std β[a]	*t*	*F*	ΔR^2
Step 1					.77	.02
Child age at Diagnosis	.00	.030	-.02	-.11		
Time off-treatment	.00	.002	-.16	-1.01		
Treatment Intensity	-.06	.018	-.05	-.31		
Step 2					3.26**	.27
Escape-Avoidance Coping	-.03	.018	-.19	-1.35		
Child Demandingness	-.04	.014	-.38	-2.71**		
FES Supportive	.00	.002	.092	.69		

a: Regression weights at entry into the model

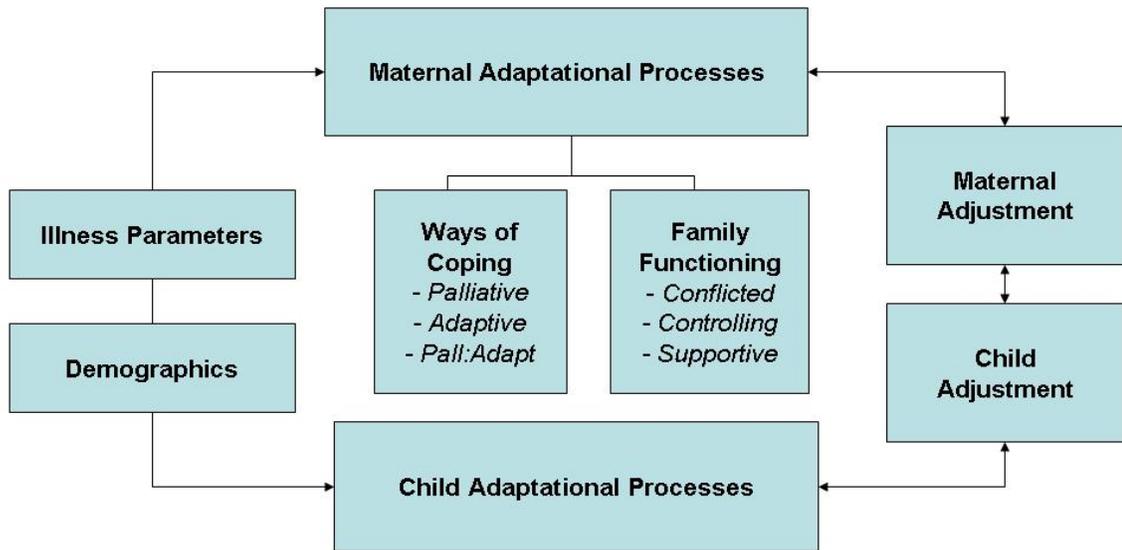* *p* < .05, ** *p* < .01

Appendix B: Figures

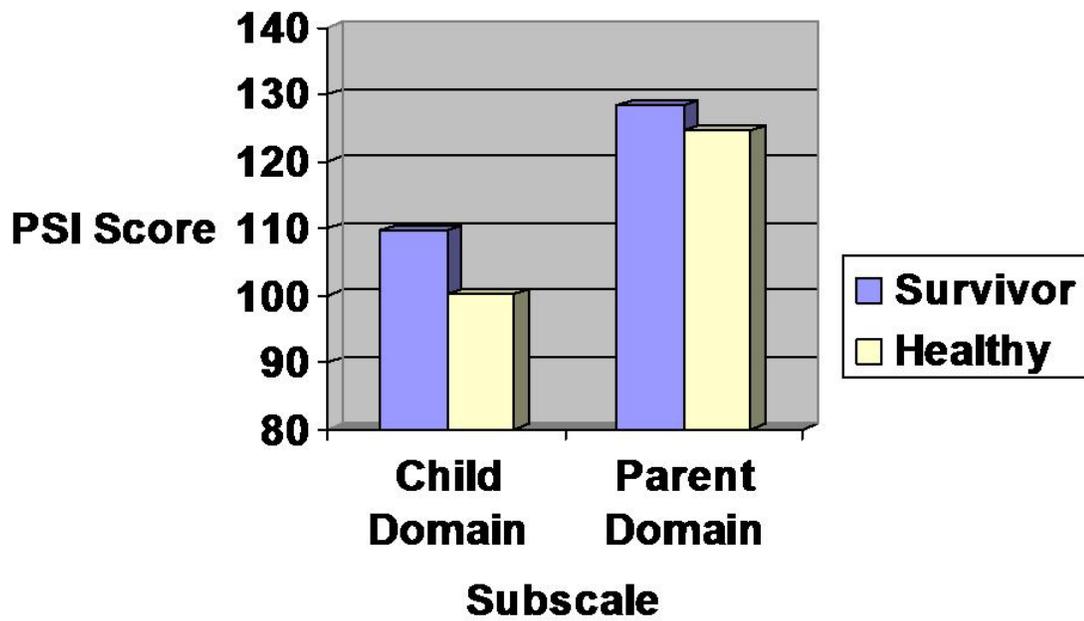**Figure 1: Adapted Transactional Stress and Coping Model**
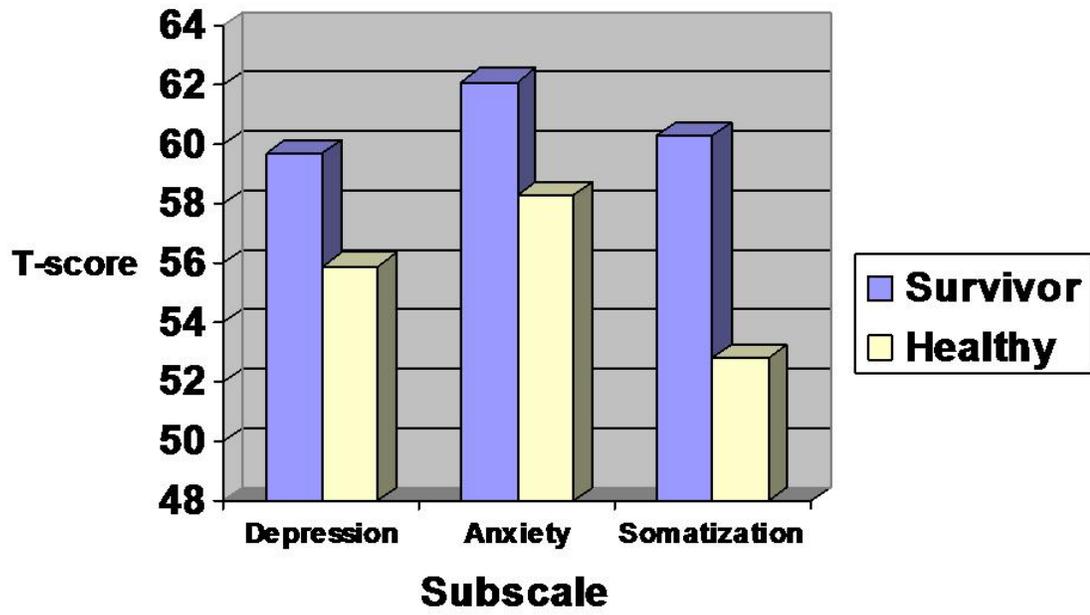
Figure 2: PSI Means by Group
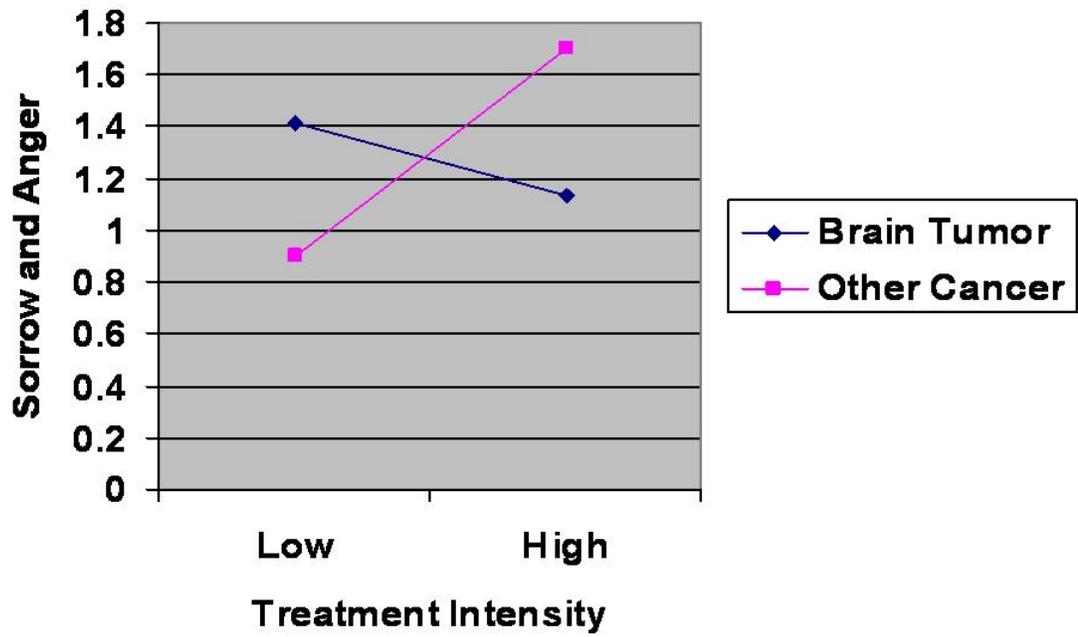
Figure 3: BSI Means by Group

Figure 4: Interaction Effects for Diagnostic Category and Treatment Intensity on PECI Unresolved Sorrow and Anger
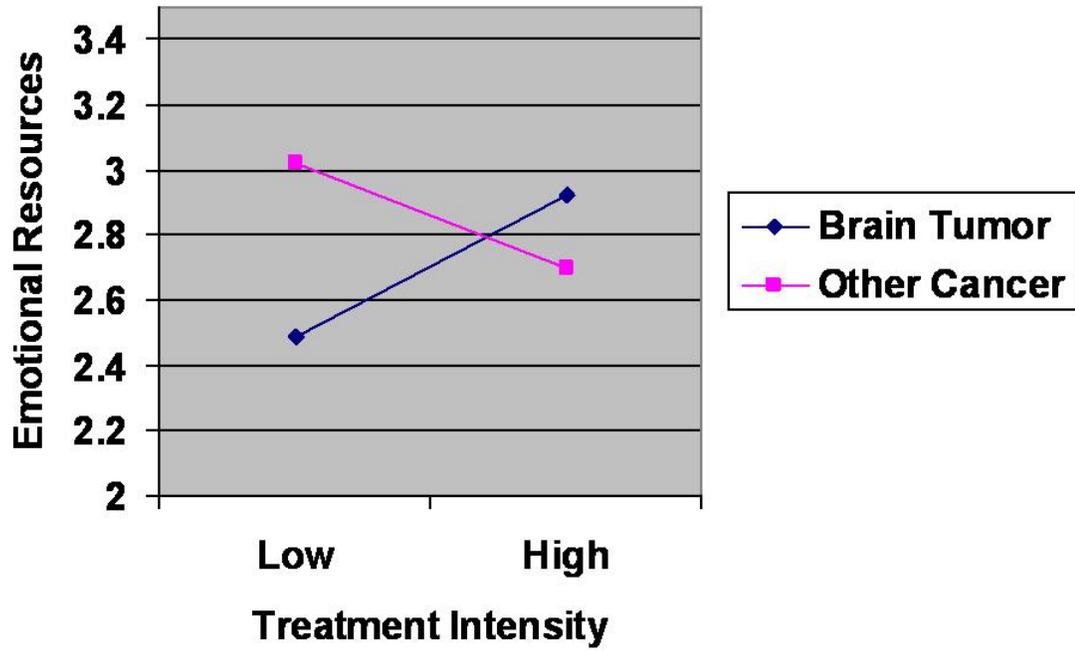
Figure 5: Interaction Effects for Diagnostic Category and Treatment Intensity on PECI Emotional Resources

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Biography

Katherine Conlon Hutchinson was born in Pittsburgh, Pennsylvania in 1975. In 1997, she earned a Bachelor of Arts degree in Psychology and Pre-Professional Studies (Pre-medicine) from the University of Notre Dame. Upon completing her degree, Katherine moved to Chicago and was worked as an information technology consultant, first for Deloitte Consulting and then for CapGemini. In 2002, Katherine enrolled in Duke University's doctoral program in psychology, where she pursued her interests in child clinical and pediatric psychology under the mentorship of Melanie J Bonner, Ph.D. While enrolled at Duke, Katherine earned her Master of Arts degree in Psychology in 2006. She was awarded multiple Conference Travel Awards and a Summer Research Grant for her dissertation project. Katherine was accepted into Duke University Medical Center's Psychology Internship Program in 2008. Katherine is a student member of the Society of Pediatric Psychology, American Psychological Association and Association of Behavioral and Cognitive Therapies.