Neurocognitive deficits and parental adjustment predict functional impairment in acute lymphoblastic leukemia and lymphoma: a pilot study

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NEUROCOGNITIVE DEFICITS AND PARENTAL ADJUSTMENT PREDICT FUNCTIONAL IMPAIRMENT IN ACUTE LYMPHOMBLASTIC LEUKEMIA AND LYMPHOMA: A PILOT STUDY

By

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ABSTRACT

Childhood cancer survivors are at risk for long-term neurocognitive and psychosocial morbidities. Research, however, has only recently examined the overall salience of these late effects and how they translate into functional impairment. The purpose of the current study was to characterize the frequency/severity of functional impairment as well as identify significant neurocognitive and psychosocial determinants of functional impairment. 50 English speaking child-parent dyads were enrolled in the study. Children were between the ages of four and nineteen years and were at least 2 years post diagnosis with leukemia/lymphoma. Participants were recruited through a pediatric oncology late effects clinic where parents completed psychosocial and functional impairment questionnaires while a brief neuropsychological exam was administered to children. Results found that 26% of participants were identified as demonstrating significant functional impairment. However, the one significant predictor of functional impairment was parental stress. While children demonstrated both neurocognitive deficits and functional impairments, neurocognitive
deficits did not predict these functional difficulties. Results instead favored psychosocial factors, such as parental stress, as a predictor of overall functional impairment.
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Chapter 1 Introduction

The past two decades have witnessed a significant increase in survival for children diagnosed with cancer. Leukemia and lymphoma are the most prevalent forms of childhood cancer, with 41% of cancer diagnoses in children ages 0-19 are either leukemia or lymphoma (Institute of Medicine, 2003). Although leukemia and lymphoma are the most prevalent forms of childhood cancer, both demonstrate a remarkable advancement in survival, as 80-85% of children diagnosed reach five years of survival (Institute of Medicine, 2003). This dramatic increase in survival rates is largely due to the use of more aggressive modern therapies such as chemotherapy, radiation, surgery, or combination of the three.

Unfortunately, aggressive modern treatments come with a cost, as survivors are at risk for long-term morbidity as a result of the disease and aggressive treatments used to combat it (Dickerman, 2007; Patenaude & Kupst, 2005). These adverse late effects become manifest in a variety of treatment-related physical effects, including, coronary artery disease, congestive heart failure, second cancers, renal failure or dialysis, and hearing loss (Dickerman, 2007). Oeffinger et al. (2006) reported that 73.4% of pediatric cancer survivors experienced some type of chronic health condition and 42% of survivors experienced a severe, disabling, or even life-threatening condition. Additionally, survivors are at risk for adverse psychological outcomes such as neurocognitive deficits (Krull et al., 2008), as well as social and emotional problems 2 years post-treatment and beyond (Patenaude & Kupst, 2005). Although adverse late effects are not exclusively limited to leukemia/lymphomas, due to the relatively high prevalence rates and the significant advances in treatment and survival rates, the proposed study will focus attention specifically on pediatric leukemia and lymphoma survivors and the adverse outcomes associated with survival.
Functional Impairment

Pediatric cancer survivors are at risk for developing a variety of psychosocial deficits. These problematic psychosocial outcomes are very heterogeneous, as they can range from minor social difficulties to severe emotional problems and can even qualify for a clinical disorder. The literature has identified social skills/competence, and mental/emotional health, as two major domains in which survivors can demonstrate problems (Hudson, Mertens, Yasui, Hobbie, & Chen, 2003; Ness et al., 2008; Schultz et al., 2007). Compared to healthy peers, survivors have been found to experience more behavior problems, less social competence, fewer friends, and a decreased peer activities (Schultz et al., 2007; Stam, Grootenhius, & Last, 2001). Furthermore, survivors have been found to demonstrate greater symptoms of global distress (Zeltzer et al., 2008), depression, anxiety (Schultz et al, 2007), identity problems, and posttraumatic stress symptoms (Stam et al., 2001). Although this research provides important information regarding the types of deficits experienced by pediatric cancer survivors, there is an important piece missing. Very little is known regarding the salience of these psychosocial deficits on day-to-day functioning.

Functional impairment represents an important emerging construct within pediatric health, as it provides important additional information beyond specific psychosocial deficits. However, the importance of functional impairment in the context of physical and psychological health has only recently emerged. In 1980 the World Health Organization published the International Classification of Impairments, Disabilities, and Handicaps (ICIDH), which for the first time highlighted the consequences of the disease in terms of overall functioning. In other words, this new classification system helped shift the focus from cause to impact. Palermo et al. (2008) defines functional impairment as the extent to which
children are unable to perform daily activities. Such activities can be physical, social, or personal. In this regard, the importance of functional impairment as a construct lies in its ability to identify areas that are particularly salient to the child’s life. These salient areas include the ability to attend school and play with friends (Palermo et al., 2008). Overall, functional impairment can be conceptualized as an inability to engage in routine day-to-day activities. Bird et al. (2005) identified three major domains in which day-to-day functioning may be disrupted. These domains include: interpersonal relations, school/work functioning, and self-care/self-fulfillment.

While it is clear that pediatric cancer survivors experience a variety of adverse cognitive and psychosocial outcomes (Patenaude & Kupst, 2005; Moore, 2005), it remains unclear how these adverse outcomes translate into an overall deficit in functional capacity. Although previously, functional impairment has been linked to children with TBI, ADHD, and other mental health problems (Fay et al., 2009; Huppert, Simpson, Nissenson, Liebowitz, & Foa, 2009; Lollar, 2008; Wille, 2008), it has yet to be examined in relation to the pediatric cancer experience. There is, however, some evidence suggesting that pediatric cancer survivors do indeed suffer functional deficits. Hudson et al., (2003) found that childhood cancer survivors were significantly more likely to demonstrate functional impairment, which was defined as needing help with personal care, routine daily rituals, and keeping and holding a job. Pediatric cancer survivors have also demonstrated difficulty functioning in school environments and getting and maintaining a job (Mitby et al., 2003; Pang et al., 2008).

There are three major advantages to examining functional impairment in pediatric cancer survivors. First is that information regarding multiple domains of deficits such as physical, social, and personal (Palermo et al., 2008) are included. This synthesis of
information is critically important, as cancer survivors demonstrate a wide range of deficits in academic, social, and emotional domains (Patenaude & Kupst, 2005). This type of assessment moves beyond assessing any single domain in isolation (Palermo et al., 2008) such as anxiety, depression, or social skills and provides information regarding how these deficits synthesize and impede day-to-day activities. In other words, functional impairment encompasses the wide range of psychosocial deficits that manifest in pediatric cancer survivors.

Second, measuring functional impairment is that it provides clinically relevant information. As stated above, survivors demonstrate a myriad of adverse psychosocial deficits. The identification of such deficits, however, does not explain much beyond the fact that they exist. This information is significantly limited as many questions still remain. For example, how significant are these deficits? Are they minimal or so severe that they dramatically interfere with the child’s ability to function? As such, functional impairment can indicate the severity of deficits and if they demand clinical attention.

A final advantage of assessing functional impairment is the ability to identify targets of treatments and interventions. For example, Bird et al., (2005) identified three major functional domains, which include: interpersonal relations, school/work functioning, and self-care/self-fulfillment. As such, this type of assessment is able to reveal where exactly functioning is disrupted and therefore where treatment interventions should be targeted. Determining the extent of functional impairment provides an opportunity for targeted psychological intervention in pediatric oncology.
Assessing Functional Impairment

Despite these advantages of functional impairment assessments, little empirical work has explored the construct of functional impairment in pediatric cancer survivors. The Karnofsky Performance Scale (KPS) remains the leading tool in assessing functional status in pediatric cancer survivors enrolled in clinical trials. The KPS, however, is significantly flawed as it is limited to physician-defined status and is a subjective assessment of performance status in which the physician assigns the child a percentage from 1-100, where scores are benchmarked to criteria (general category and specific criteria) that are ascertained in clinic follow-up exams. This score represents the child’s ability to perform normal daily activities and the extent to which assistance is needed (Schag, Heinrich, & Ganz, 1984). Higher scores are indicative of normal functioning while lower scores indicate severe disability. The KPS is anchored in more physical disabilities rather than social and emotional disability (Taylor, Olver, Sivanthan, Chi, & Purnell, 1999).

Due to the large degree of subjectivity inherent in this assessment, the KPS has questionable reliability and validity (Jones et al., 2009; Schag et al., 1984; Taylor et al., 1999). It is of limited value as an overall assessment of functionality because it fails to assess social, emotional, and cognitive functioning and instead focuses exclusively upon physical impairments (Grieco & Long, 1984; Schaafsma & Osoba, 1994). The usefulness of the KPS in assessing children in particular has also been questioned with the conclusion that a quality of life scale may be more beneficial when assessing children because it is more informative than a subjective assessment of functionality carried out by a physician (Milstein, Cohen, & Sinks, 1985).
The Brief Impairment Scale (BIS), developed by Bird et al. (2005) represents an alternative measure of functional impairment in pediatric populations. The BIS is a 23-item assessment that provides a global measure of impairment along three domains of functioning: interpersonal relations, school/work functioning, and self-care/self-fulfillment. The assessment, which is completed by parents, is intended to assess the degree to which their child struggles with various day-to-day activities and has been used with children ranging from 4-18 years. There is ample evidence indicating that parent report of child functionality is valid and reliable (Bird, 1999). The BIS has demonstrated internal consistency with alpha ranging from .81 to .88 as well as test-retest reliability. Convergent validity was demonstrated by significant correlations ($r = -0.53, 0.52, \text{ and } -0.52; p < .001$) between the BIS and an established measure of the same construct, the CGAS (Shaffer et al, 1983).

While it is critically important to understand the degree to which pediatric cancer survivors experience functional impairments, it is equally important to understand the factors that are associated with such impairments. An understanding of associated factors will allow for a better ability to assess risk and determine where to target interventions. The following sections will provide a discussion of the possible factors that predict functional impairment

**Neurocognitive Predictors of Functional Impairment**

Neurocognitive impairment is one of the most common late effects experienced by pediatric cancer survivors with up to 40% manifesting some type of cognitive impairment (Krull et al., 2008). Research indicates that the cognitive impairments are incredibly heterogenous, as a very broad range or impairments has been identified by the literature. For example, survivors have demonstrated deficits in the domains of attention, executive functioning, memory, information processing speed, and visual-spatial skills (Moore, 2005;
Moore, Ater, & Copeland, 1992; Mulhern Wasserman, Fairclough & Ochs 1988). The heterogeneity of cognitive deficits, however, is not easily explained by the wide variety of cancer diagnoses. Childhood survivors of leukemia have demonstrated both general intellectual declines (Mulhern, Fairclough, & Ochs; 1991) and specific cognitive deficits in domains such as: attention, motor and perceptual timing, memory, and verbal ability (Anderson, Smibert, Ekert, & Godber, 1994; Christie, Leiper, Chessells, & Vargha-Khadem, 1995; Mahone, Prahme, Ruble, Motofsky, & Shwartz, 2007)

**General intellectual impairment.** Montour-Proulx et al. (2004) examined the cognitive functioning of a group of pediatric leukemia survivors and found that their scores on the Weschler Intelligence Scale for Children performance IQ were significantly lower than normative means. Another study also found pediatric cancer survivors demonstrated general IQ deficits when compared against healthy controls (Anderson et al., 1994). Mulhern et al. (1991) found evidence supporting a general cognitive deficit in pediatric leukemia survivors, as 22-30% of survivors exhibited a clinically significant deterioration in overall IQ values. As noted earlier, extant literature has identified numerous cognitive deficits experienced by pediatric cancer survivors. As such, there is some debate within the literature concerning whether survivors demonstrate a very general intellectual decline or whether they experience cognitive deficits in very specific domains.

**Specific cognitive impairment.** While some research seems to suggest a more general intellectual deficit, there is evidence that suggests pediatric leukemia survivors experience very specific cognitive deficits. The manifestation of specific cognitive deficits seems to be varied, as there is not a particular domain of impairment that is commonly experienced by pediatric cancer survivors. Mulhern et al. (1988) found evidence for visual-
spatial memory and verbal memory deficits, while Christie et al. (1995) found deficits in non-verbal ability and short-term memory. Others have found that adult survivors of childhood cancer demonstrated deficits on executive functioning tasks (Ness et al. 2008). Attention and verbal deficits have also been identified in studies (Anderson et al., 1994). There is evidence pediatric leukemia survivors may also be at risk for motor and perceptual timing deficits (Mahone et al., 2007).

Neuroimaging studies have sought to identify the substrate of neurocognitive deficits with findings suggesting that survivors may be experiencing structural abnormalities such as leukoencephalopathy intracerebral calcifications and white matter alterations (Dickerman 2007; Iuvone, Mariotti, Colosimo, Guzetta, Ruggiero, & Riccari 2002). Although not much is known regarding how these structural abnormalities specifically relate to functioning, it has been suggested that such structural abnormalities may be related to memory, attention, and IQ deficits. Iuvone et al. (2002) examined the relationship between structural abnormalities and cognitive outcomes in pediatric leukemia survivors and found support for such a hypothesis, as white matter abnormalities were associated with poorer performance on visual motor integration tasks in about 50% of the patients.

In general, the research indicates that many pediatric leukemia survivors are at risk for developing some form of cognitive deficit and it is projected that between 50 and 60% of survivors are at risk for developing some form of neurocognitive dysfunction (Nathan et al., 2007). However, the research indicates that such cognitive impairments are varied and wide-ranging. There is no common presentation of cognitive deficits as they can vary in nature, severity, and range. Additionally, current research does not provide much more information beyond this descriptive level. It is important, however, to understand what these deficits
mean on a functional level. For example, how much do neurocognitive deficits impact school related functioning? To what extent do attention and memory deficits influence a child’s ability to interact with friends and family? Do IQ deficits influence a child’s sense of self-worth, self-satisfaction? While research indicates that survivors demonstrate functional impairment as well as neurocognitive deficits, there is very little information concerning the linkage.

**Neurocognitive deficits and functional impairment.** There is reason to hypothesize a link between neurocognitive deficits and overall functional deficits. Many neurocognitive deficits experienced by pediatric cancer survivors are integral skills for school related tasks. For example, deficits in general intellectual ability, memory, non-verbal ability, and executive functioning, could potentially disrupt a child’s ability to function in an academic domain. Research has found that survivors demonstrated impairment in one of the three functional domains, identified by Bird and colleagues (2005) as school/work functioning. One study found that pediatric cancer survivors demonstrated greater difficulties with scholastic achievement and were significantly less likely to complete high school when compared to a group of healthy controls. They were also more likely to require special education services. (Mitby et al., 2003). Another study conducted by Pang and colleagues (2008) examined the degree to which cancer survivors found and maintained successful employment. Self-reported employment history in 10,399 childhood cancer survivors and 3,083 siblings were analyzed, indicating that survivors of childhood cancer were at higher risk for later employment difficulties and were more likely to be unemployed. Although no empirical work has yet explored the link between neurocognitive deficits and functional impairment in pediatric cancer survivors, there is strong reason to believe that they may be
related. More information on how neurocognitive deficits predict the functionality of pediatric cancer survivors will help inform decisions regarding the necessity of assessments for this particular population and where to target treatment interventions

**Parental Adjustment Predictors of Functional Impairment**

Parental adjustment is one factor that has recently received a significant amount of attention within pediatric health. A significant body of literature indicates that parents often have difficulty adjusting to their child’s illness. As such, they are at risk for experiencing high levels of stress, distress, uncertainty, depression, and anxiety (Mishel 1983; Pai, Drotar, Zebracki, Moore, & Youngstrom, 2006; Streisand, Rodrique, Houck, Graham-Pole, & Berland, 2000). In one study assessing the emotional well being of parents who had a child diagnosed with cancer, 13 of the 18 parents assessed suffered symptoms of anxiety outside the normal range (Hughes & Lieberman, 1990). Parents who experience a child diagnosed with cancer are also at risk for developing posttraumatic stress. Studies comparing parent’s personal experience of PTS symptoms with their children’s self-reported experience found parents reported more trauma-related stress and PTSD symptomatology than their child (Kazak, Alderfer, Rourke, Simms, Streisand, & Grossman, 2004). It may be the case that the parent is actually more aware of the life-threatening nature of the illness, and therefore more likely to experience their child’s cancer as a traumatic event.

A child’s cancer diagnosis is an undeniably distressing event for a parent. But what is pivotal concerning child outcomes is how the parent’s reaction to the illness influences the child. Overall, the literature indicates that parental adjustment plays an influential role in their child’s psychological adjustment and that parental maladjustment can lead to problematic child outcomes (Chaney et al., 1997; Davis et al., 2001; Thompson, Gil, Burback,
Keith, & Kinney, 1993). However, because functional impairment is a new construct within pediatric cancer populations, no empirical work has been done to determine what these adjustment difficulties mean in terms of overall functional capacity. As such, parental adjustment needs to be further explored in terms of its relationship to functional impairment.

There is, however, strong evidence that suggests parental adjustment influences child adjustment. For example, an extensive body of research indicates that children learn the attitudes and behaviors expressed by parents and internalize them as their own (Chaney et al., 1997; Mishel, 1983; Thompson et al., 1993; Varni, Katz, Colegrove, & Dolgin 1993). Therefore, parents who display high levels of stress, depression, and anxiety may have undue influence on their child’s experience of stress, depression, and anxiety. The relationship between parental adjustment and child adjustment seems to be evident in a variety of pediatric health populations. For example, a study examining the correlation between parental and child adjustment in children with insulin-dependent diabetes mellitus (IDDM) found that increased levels of fathers’ distress was highly associated with elevated levels of the child’s distress (Chaney et al., 1997). These results are highly suggestive of the influential nature of parental attitudes when dealing with childhood illnesses. This conclusion is further supported by Thompson and colleagues (1993), who found poor maternal adjustment was associated with poor coping methods and high levels of stress in children with sickle cell disease. Similar results were found for children diagnosed with diabetes and asthma as high levels of parenting stress were significantly predictive of the child reacting to their illness with high levels of uncertainty (Mullins et al., 2007). Such high levels of uncertainty in children are problematic, as uncertainty has been linked to poor psychological outcomes in pediatric populations (Mishel, 1983). Children seem to be very sensitive to their
parents’ responses such that parental reactions guide and inform the child’s own reactions. As such, it can be concluded that parental adjustment is a powerful indicator of a child’s own adjustment and that the parent’s emotional response often informs the child’s emotional response.

Parental adjustment has also been found to influence behavioral responses as well as emotional responses in pediatric populations. For example, one study found that conflict within the parent-child dynamic resulted in lower adherence and metabolic control in children with IDDM (Miller-Johnson, Emery, Marvin, Clarke, Lovinger, & Martin, 1994). A different study found that parental warmth was strongly associated with better adherence in children with diabetes (Davis et al., 2001). In a yet another study, Eaton, Larson, Mengel, Campbell, Mengel, and Montague (1992) found that psychosocial variables such as anxiety, depression, and family processes were related to the management and control of diabetes in children and adolescents. The literature concerning parental influence on child outcomes in pediatric health populations is in overwhelming agreement. Parental attitudes and adjustment significantly influence the child’s health behavior and health outcomes (Davis et al., 2001; Forsyth, Horwitz, Leventhal, & Burger, 1996; Miller-Johnson et al., 1994).

Although, the literature is significantly limited, one study has found promising evidence in support of parental adjustment as a significant predictor of problematic functional outcomes in pediatric cancer survivors. This study explored the relationship between parental experiences of depression and stress and the child’s quality of life. Results found that parental anxiety and depression were indeed linked to the child’s quality of life, such that increases in parental anxiety and depression contributed to a decrease in the child’s quality of life (Roddenberry & Renk, 2007)
Defining parental adjustment. Mullins et al. (2007) operationalized parent adjustment into four specific components believed to be instrumental in later functional outcomes in children with chronic illness. Such factors include: parental stress, parental overprotection, perceived child vulnerability, and illness uncertainty. With the exception of parenting stress, these specific parental adjustment variables have not been studied extensively in pediatric cancer populations but have proven influential in the more general childhood chronic illness populations. Mullins et al. (2007), found preliminary evidence that suggests such parental adjustment categories are significantly associated with poor child outcomes.

Parental stress. Mothers of chronically ill children are at risk for both heightened stress and psychological distress (Streisand et al., 2000). This is important because parental stress is a major factor that predicts child distress (Chaney et al., 1997; Robinson, Gerhardt, Vannatta, & Knoll, 2007). Additionally, studies with chronically ill populations have documented the linkage between parental stress and distress on the child’s cognitive and social development (Kazak et al., 2005; Pai et al., 2006).

Parental overprotection. Parental overprotection has been defined as overindulgent, over solicitous, and overanxious parenting (Thomasgard, Wetz, Edelbrock & Shonkoff, 1995). It involves a specific pattern of parent behaviors where overprotective parents are described as highly vigilant, having difficulty with separation, exercising a high level of control, and discouraging independent behavior. Such levels of protection are considered to be excessive given the developmental level and actual abilities of the child. While a certain amount of protection by parents of pediatric cancer survivors may be adaptive, this behavior becomes problematic when it becomes excessive and continues over an extended amount of
time. Excessive amounts of overprotection have been linked with child adjustment
difficulties. For example, Holmbeck et al., (2002) found that among children diagnosed with
spina bifida, behavioral autonomy was significantly lacking in those children whose parents
demonstrated high levels of overprotection.

**Parent perceptions of child vulnerability.** In contrast to a specific pattern of
behaviors, parental perceptions of child vulnerability refer to parental attitudes or beliefs.
Such attitudes include conscious and unconscious perceptions of fear regarding their child’s
health and/or potential premature death (Thomasgard, 1998). Research indicates that parents
who had a child experience a life-threatening illness were significantly more likely to view
their child as vulnerable (Thomasgard & Metz, 1997). More importantly, increases in
behavior problems were observed in those children whose parents perceived them as highly
vulnerable in comparison to children who were not viewed as vulnerable by their parents
(Forsyth et al., 1996).

**Illness uncertainty.** Illness uncertainty surrounding the current status of the illness
has been identified as another parental adjustment factor that has previously been linked with
poor psychological outcomes in pediatric populations. Recent research has revealed that
parental experiences of illness uncertainty can have a significantly negative impact on the
psychological well being of the parents. Such studies have found that illness uncertainty in
parents significantly predicts higher levels of distress (Carpentier, Chaney, Mullins, &
Wagner, 2006; Steward & Mishel, 2000). Mishel (1983) has also suggested that parents who
cope effectively are more apt to provide positive and stable support for their child and that
factors such as illness uncertainty impede the parent’s ability to cope and therefore provide
effective support.
Illness uncertainty has also been found to significantly influence the psychological well-being of children. Studies have found evidence to support the association between uncertainty and depressive symptoms in children with illnesses such as diabetes and JRA (Hoff et al., 2002; White, Chaney, Mullins, & Wagner 2005). However, research has yet to delineate the relationship between parental uncertainty and problematic child outcomes. However, the previously established influential nature of the parent-adjustment, as articulated above, strongly suggests that parental illness uncertainty may increase a child’s risk for experiencing illness uncertainty and therefore increase the risk for poor adjustment and problematic functional outcomes.

**Summary of parental adjustment.** Parents managing their child’s treatment for leukemia and lymphoma have been shown to demonstrate high levels of stress, distress, overprotection, uncertainty, and are more apt to perceive their child as vulnerable (Mishel, 1983; Pai et al., 2006; Thomasgard et al., 1995). The relationship of such parental adjustment variables is strongly suspected to have an effect on the child’s functional impairment outcome, yet little research to date has explored this relationship (Varni et al., 1993; Mishel, 1983). Future research needs to examine this relationship further and delineate the degree to which parental adjustment contributes to functional difficulties.
Chapter 2 The Present Study

Overall, pediatric cancer survivors demonstrate neurocognitive deficits and are exposed to parental adjustment problems. Due to a general lack of research in this area, it remains less clear as to how these neurocognitive deficits and parental adjustment problems interfere with the child's overall functional capabilities. This type of knowledge, however, would help to identify pediatric cancer survivors at risk for functional deficits and help determine treatment targets.

As such, the first research question this study investigates is the degree to which pediatric cancer survivors demonstrate significant functional impairment. The second question addressed by the study is the relative predictive power of both neurocognitive deficits and the different components of parental adjustment on functional impairment. In summary, this study develops pilot data that tests a model of the neuropsychological and psychosocial factors associated with functional impairment in children who have been treated for leukemia/lymphoma. The two main objectives of this study were: 1) to characterize the frequency and severity of functional impairment in children treated for leukemia/lymphoma who are at least two years off treatment and 2) identify determinants of functional impairment in children treated for leukemia/lymphoma.
Chapter 3 Methods

Participants

Participants included 50 English speaking pediatric cancer survivors. Eligible individuals were children between the ages of 4 and 19, previously treated for leukemia or lymphoma, at least 2 years post-treatment. Individuals excluded from the study were those who, at the discretion of the investigator, did not have the ability to successfully complete the neurocognitive testing (e.g., those with moderate to profound mental retardation). Parents who were unable to complete questionnaires in English were also excluded from the study.

Procedure

Parents of potential participants were recruited and enrolled by a trained research assistant during the participant’s routine clinic appointment at the Young Enduring Survivors (YES) Clinic. Prior to their clinic visit, eligible participants were identified by clinic staff as eligible and introduced to the study via a letter that was mailed one week prior to their clinic appointment containing information regarding the study, as well as consent materials.

Candidate participants were invited to participate by a trained research assistant, who provided the parents and child with study information and obtained informed consent and assent for those who were interested. Participants were offered the option of completing the research activities at the clinic visit or at another scheduled time. The majority of participants (96%) preferred to complete that day, however, two families scheduled a time to return to the clinic to complete the study. The procedure began with a neurobehavioral exam, administered to the child by a trained psychometrist, in a private clinic room, requiring approximately 20-30 minutes of child time. At the same time, parent adjustment and functional impairment questionnaires were completed by the parent, requiring approximately
30-40 minute. Upon completion of the study participants received a $20 gift card as appreciation for their participation.

Measures

**Neurocognitive measures.** All children completed a brief screening battery of neuropsychological tests, administered by a trained psychometrist. Test selection was based on a preliminary study conducted by Krull et al. (2008). The study found that this brief neuropsychological battery demonstrated good test-retest reliability and accurately predicted global intellect, reading skills, and mathematics skills. Overall the screening battery was found to be both a practical and reliable method of identifying neurocognitive deficits. The battery took roughly 20-30 minutes and included the following measures (in order of administration): Beery Developmental Test of Visual-Motor Integration (Beery, 1997), Digit Span Test, (ages 4-5: the Kaufman Assessment Battery for Children, Second Edition (Kaufman & Kaufman, 2004); ages 6-17: the Wechsler Intelligence Scales for Children-Fourth Edition (Wechsler, 2003); ages 18-19: Wechsler Adult Intelligence Scales-Third Edition (Wechsler, 1997)), Trail Making Tests (Reitan, 1993), Purdue Pegboard Test (Tiffin, 1968), and the Verbal Fluency Test (COWAT; Benton, Hamsher, Sivan,1983).

**Parental psychosocial measures.** All parent participants completed brief psychosocial and functional impairment measures, administered by a trained research assistant either during the child’s regular clinic visit or during another time when the participant agreed to return. If both parents were available to fill out questionnaire’s priority was given to the mothers. The battery took roughly 30-45 minutes to complete and included the following measures.
The Parent Protection Scale (PPS; Thomasgard et al., 1995) was used to measure parental overprotection. The PPS is a 25-item self-report measure assessing protective parenting behaviors. Respondents rate each statement on a four-point scale ranging from 0 (“never”) to 3 (“always”) as to the degree to which the statement is descriptive of their behavior with their child. Items include such statements as: “I comfort my child immediately when he/she cries,” and “I let my child make his/her own decisions.” Higher scores represent greater levels of parental protective behaviors. Clinically significant overprotective behavior is represented by a score of 39 or greater (Thomasgard & Metz, 1997). Factor analysis of the PPS has yielded four subscales: supervision, separation problems, dependence, and control.

Criterion validity, using criterion-referenced clinical history as the basis for comparison, has been demonstrated to be acceptable (Thomasgard et al., 1995). Previous studies have demonstrated moderate to high internal reliability ($\alpha = .73$) and high test-retest reliability ($r = .86, p = .001$) (Thomasgard et al., 1995). Cronbach alpha for this sample was .61. The PPS has been used in studies with a sample of parents of chronically ill children and adolescents (Bourdeau, Mullins, Carpentier, Colletti, & Wolfe-Christensen, 2007; Mullins et al., 2007).

Parental perceptions of child vulnerability were assessed using the Child Vulnerability Scale (CVS; Forsyth et al., 1996). The CVS is an eight item self-report scale with a 4-point response scale ranging from 0 (“definitely false”) to 3 (“definitely true”); higher scores reflect greater perceived child vulnerability (0-24). Items include statements such as: “In general my child seems less healthy than other children,” and “I get concerned about circles under my child’s eyes.” The cutoff for clinically significant perceived vulnerability is recommended at 10 and the measure yields one overall summary score. Validity of the CVS has been demonstrated through studies comparing the scores of the CVS
to scores on the Child Behavior Checklist (CBCL). Previous studies have also demonstrated adequate internal reliability (Cronbach’s alpha) of .74 (Forsyth et al., 1996) and a correlation of $r = .84$ for test-retest reliability (Thomasgard & Metz, 1993). Cronbach alpha for this sample was .72. The CVS has previously been used on chronically ill child and adolescent populations (Bourdeau, Mullins, Carpentier, Colletti, & Wolfe-Christensen, 2007; Mullins et al., 2007).

Parenting stress was assessed using The Parenting Stress Index/Short Form (PSI/SF; Abidin, 1990). The PSI/SF is a 36-item parent self-report that produces a score on three subscales, including Parental Distress, Parent Child Dysfunctional Interactions, and Difficult Child, as well as an overall summary score. The assessment contains a 5-point response scale ranging from 1 (“strongly agree”) to 5 (“strongly disagree”). Items include statements such as: “I feel trapped by my responsibilities as a parent,” and “My child makes more demands on me than most children.” The manual indicates that a score of 90 is recommended as a clinical cutoff score. The validity for the short form is similar to that of the full-length PSI and has been established on a range of populations including parents of children with asthma and diabetes (Carson & Schauer, 1992; Wysocki, Huxtable, Linscheid, & Wayne, 1989). The PSI/SF is correlated with the full-length PSI instrument ($r = .94$) and two-week test-retest reliability of the full-length PSI with the PSI/SF is .95 (Abidin, 1990). Cronbach alpha for this sample was .90.

The Care of My Child with Cancer Scale (Wells et al., 2002) assessed the time and difficulty associated with providing care for a child previously diagnosed with cancer. Caring for a child with cancer necessitates a major restructuring of family life in order to deal with new caretaking responsibilities. As such, this measure was used to assess the demands of
illness related caregiving and caregiver burden. This is a 34-item parent report scale in which items assess the time and effort associated with caregiving tasks. Item responses are structured on 5-point Likert-response scale for both time (ranging from > 5 hours a week to none) and effort (ranging from “a great deal” to “none”). Items include statements such as: “Providing emotional support” and “Comforting your child through the pain of cancer and its treatment”. Parents are instructed to indicate both the amount of time and the amount of effort per week required to complete such caregiving tasks. This assessment has demonstrated construct validity (Wells et al., 2002) as well as internal consistency (alpha = .93), and strong test-retest reliability (r = .90). Cronbach alpha for this sample was .96.

The Uncertainty Management and Coping Skills Scale for Parents (UMCSS-P) is a 25-item self-report measure assessing parent’s acquisition of uncertainty management skills. The scale was adapted in part from the Self-Control Scale (Rosenbaum, 1990), Mishel’s scales for adult cancer (Mishel et al., 2002), and the Multidimensional Scale of Perceived Social Support (Zimet, Dalhem, Zimet, & Farley, 1988). The measure includes items assessing utilization of cognitive reframing and problem-solving strategies, communication with medical staff, and perception of support systems. The questionnaire includes items such as, “When I have something that makes me worry, I try to think how I can handle my worry.” Mishel and Germino (2002) have reported internal consistency for the cognitive reframing, problem-solving, and communication dimensions. Cronbach alpha for this sample was .77.

Functional Impairment was assessed with the Brief Impairment Scale (BIS; Bird et al., 2005). The BIS is a 23-item assessment that provides a global measure of impairment along three domains of functioning: interpersonal relations, school/work functioning, and self-care/self-fulfillment. The assessment, which is completed by parents, is intended to
assess the degree to which their child struggles with various activities. Responses are on a 4-point Likert scale ranging from 0 (“no problem”) to 3 (“serious problem”). The assessment is prefaced by the statement “In general, how much of a problem do you think your child has with”. It then includes item statements such as: “Getting involved in activities together with the rest of the family?” “Making friends”: and “Getting his/her schoolwork done on time?”.

Convergent validity was demonstrated by significant correlations ($r = -0.53, 0.52, \text{ and } -0.52; p < .001$) between the BIS and an established measure of the same construct, the CGAS (Shaffer et al, 1983). The BIS has internal consistency with alpha ranging from .81 to .88 as well as fair to substantial test-retest reliability. Cronbach alpha for this sample was .89.

**Scoring**

All raw neurocognitive data was transformed into standard scores or Z-scores, which were then used in the final analyses. Similarly, summary scores were calculated for the BIS (Bird et al., 2005), PSI (Abidin, 1990), PPS (Thomasgard et al., 1995), CVS (Forsyth et al., 1996), and CMCC (Wells et al., 2002), using the scoring instructions specified by the creators of each assessment. The UMCSS-P, however, uses items from a variety of different assessments and therefore does not contain a set of specified scoring criteria. The first twenty questions on the scale assess the use of coping strategies such as cognitive reframing and problem-solving skills. Responses are measured on a ten point Likert scale with higher numbers suggesting a stronger use of such coping strategies. The last four questions on the scale assess the degree to which reciprocal communication exists between parents and the medical staff. These questions are rated on a 5-point Likert scale with higher numbers reflecting better communication. Given the differences between these two sets of items, it was determined that the two subscales be scored independently. Summary scores for each
 subscale were calculated by first summing the number of valid items, then dividing by the number of valid items, and finally multiplying by the total number of items on the scale. This method was intended to protect against biased summary estimates that might result from incomplete or missing item responses.

**Data Analysis**

Descriptive statistics were calculated for all the neurocognitive data using the transformed Z-scores. A one-sample t test was used to compare results of neurocognitive testing to a normative population. Descriptive statistics and frequencies were also calculated from the summary scores of the parental adjustment questionnaires and BIS. An omnibus repeated measures MANOVA tested differences in impairment across the three domains of functioning (e.g. work/school, interpersonal, and self-care/self fulfillment) and paired sample t-tests were conducted as follow up tests.

A Principal components analysis (PCA) with varimax rotation was employed as a data reduction technique. The overall goal of the analysis was to consolidate both parental adjustment and neurocognitive measures and isolate independent constructs in both areas. An eigenvalue greater than one was used as the criterion for factor inclusion.

Simple bivariate correlations were computed for the BIS summary scores and the factor scores that emerged from the PCA. Simple bivariate correlations were also computed for BIS summary scores and the raw scores that emerged from neurocognitive testing and parental adjustment questionnaires. Multiple regression (using stepwise selection) examined predictors of BIS summary scores with both neurocognitive and parental adjustment data were entered as predictors of the BIS summary scores. Parental adjustment and
neurocognitive factors were entered in separate blocks, with neurocognitive factors entered as the first block.

The data from the BIS summary score was then recoded to reflect two separate groups: those who met criteria for clinically significant functional impairment and those who did not. A Logistic regression was used in which factor scores from both neurocognitive and parental adjustment data were entered as predictors. Finally, the two groups (i.e. clinical caseness vs. no caseness) were compared across all neurocognitive and parental adjustment factor scores to determine if significant differences existed.
Chapter 4 Results

Demographics

Mean age of the 50 child survivors was 12 years (SD = 2.6), and ranged from age 7-18. Forty-four percent (n=22) of the sample was female, while fifty-six percent (n=28) of the sample was male. In terms of ethnic distribution, 54 percent (n=27) were Hispanic, 32 percent (n=16) were White, 6 percent (n=3) were American Indian/Alaska Native, 6 percent (n=3) were Asian, and one participant was missing information regarding ethnicity. The ethnic distribution of this sample is somewhat different than what is found in studies. The Childhood Cancer Survivor Study (CCSS), which represents the largest and most extensively studied cohort of childhood and adolescent cancer survivors, demonstrates a much different ethnic distribution. In this sample, 87% of participants were white, 2% were black, 5% were Hispanic, 1% was Asian and 5% was characterized as “other” (Robison et al. 2002). Sixteen percent (n=8) of children in the sample were receiving special education services.

In terms of the marital status of the parents, 56 percent (n=27) were married, 14% (n=7) were not married but living with their significant other. 18% (n=9) were single, and 10% were divorced (n=5). The number of additional adults in the household ranged from 0-4 (M=1, SD=.76). The number of additional children in the household ranged from 0-6 (M=2.4, SD=1.1). Ten percent (n=5) of parents did not complete high school, 22% (n=5) completed high school, 14% (n=7) completed at least one year of college, 8% (n=4) had an Associates Degree, 22% (n=11) had a Bachelors degree, 14% (n=7) had received additional education beyond a Bachelor’s degree, and 6% (n=3) had received some type of professional/vocational training.
In terms of family income, 16% \((n=8)\) had a gross annual income below 10,000 dollars, 16% \((n=8)\) had an income between 10,000 and 30,000, 14% \((n=7)\) had an income between 30,000 and 50,000, 18% \((n=9)\) had an income between 50,000 and 70,000, 8% \((n=4)\) had an income between 70,000 and 90,000, and 24% \((n=12)\) had an income greater than 90,000.

**Neurocognitive Deficits**

Results from the one-sample t-test revealed that in this sample of pediatric cancer survivors, performance on neurocognitive testing was significantly below the normative population. Results are summarized in Table 1. These findings are consistent with previous research indicating that pediatric cancer survivors demonstrate deficits across domains of neurocognitive functioning.

**Parental Adjustment**

The scores on the PPS were normally distributed. Total scores ranged from 19-48 \((M=33.4, SD=5.9)\). Based on a clinical cutoff score of 39 (Thomasgard, 1995), 30% of parents in this sample evidenced clinically significant protective behaviors. This is similar to previous findings regarding overprotective behaviors as measured by the Parent Protection Scale (Thomasgard, 1998).
Table 1. One-Sample T Test Comparing Neurocognitive Performance to Normative Scores.

<table>
<thead>
<tr>
<th>Test</th>
<th>Sample Mean</th>
<th>Normative Mean</th>
<th>t</th>
<th>Significance</th>
</tr>
</thead>
<tbody>
<tr>
<td>Beery VMI</td>
<td>89.5</td>
<td>100</td>
<td>-4.488</td>
<td>.000</td>
</tr>
<tr>
<td>Digit Span</td>
<td>8.63</td>
<td>10</td>
<td>-2.900</td>
<td>.006</td>
</tr>
<tr>
<td>Trails A</td>
<td>-0.289</td>
<td>0</td>
<td>-1.749</td>
<td>.087</td>
</tr>
<tr>
<td>Trails B</td>
<td>-0.96</td>
<td>0</td>
<td>-2.731</td>
<td>.009</td>
</tr>
<tr>
<td>Purdue (Right Hand)</td>
<td>-0.70</td>
<td>0</td>
<td>-6.198</td>
<td>.000</td>
</tr>
<tr>
<td>Purdue (Left Hand)</td>
<td>-0.92</td>
<td>0</td>
<td>-5.881</td>
<td>.000</td>
</tr>
<tr>
<td>Purdue (Both Hands)</td>
<td>-0.95</td>
<td>0</td>
<td>-6.099</td>
<td>.000</td>
</tr>
<tr>
<td>Verbal Fluency (FAS)</td>
<td>-1.11</td>
<td>0</td>
<td>-4.646</td>
<td>.000</td>
</tr>
</tbody>
</table>

The scores on the CVS were normally distributed. Total scores ranged from 0-13 (M=4, SD=3). Based on a clinical cutoff score of ten (Forsyth et al., 1996), only 6% of children were perceived as vulnerable, which is only slightly lower than previous findings with children experiencing chronic illness (Forsyth et al., 1996; Thomasgard, 1998).

The scores on the PSI-SF were normally distributed. Total scores ranged from 39-112 (M=64.5, SD=17.3). This distribution of scores was actually lower than normative populations (M = 71.96, SD = 15.4; Abidin, 1990). Based on an empirical cutoff of 90 (Abidin, 1990), only 8% of the sample evidenced clinically significant levels of stress. These
rates are actually much lower in comparison to rates of parental stress found in a similar population of cancer survivors (Winjinberg-William, Kamps, Klip, & Hoekstra-Weber, 2006).

The scores on the CMCC were not normally distributed and scores were heavily concentrated at the top end of the scale (i.e. 70% of scores fell above the mean), suggesting a rather minimal care giving demand. Scores ranged from 22-75 (M= 63.3, SD= 12.6).

The scores on the UMCSS-P coping skills subtest were not normally distributed. Scores ranged from 31-200 (M=151.1, SD=34.4). However, over 90% of the sample (n=43) scored within the top end of the scale, between 110-200. This distribution of responses suggests that most parents implemented positive coping strategies. The communication subscale scores ranged from four 4-20 (M=17.5, SD=3). Again scores were more heavily concentrated within the top end of the scale, thus suggesting that most parents were able to maintain good communication with health providers.

**Functional Impairment**

The distribution of total scores for the BIS ranged from 0-33 (M=9.6, SD=7.6). Based on an empirical cutoff of 14 (Bird et al., 2005), 26% of the sample demonstrated clinically significant functional impairment. Omnibus repeated measures MANOVA was used to test if there were differences in impairment across the three different domains of functioning (e.g. work/school, interpersonal, and self-care/self fulfillment). Results were significant, $F(2, 48)=3.847, p < .05$. Follow up tests revealed less reported impairment in the interpersonal domain (M = 2.5, SD = 2.6), than the school/work domain (M = 3.8, SD = 4.4), $t(49) = -2.297, p = .026$. There was also less reported impairment in the interpersonal domain in comparison to the self-care/self-fulfillment domain (M= 3.2, SD = 2.2), $t (49) = -2.117, p =
.039. Taken together, these results suggest that pediatric cancer survivors demonstrate similar levels of impairment in both the school and self-care/self-fulfillment domain, yet overall seem to experience less impairment in interpersonal functioning.

An omnibus one-way ANOVA was used to compare functional impairment rates across ethnicity. Comparisons were made across Whites (M=7.3), Hispanics (M=9.9), and Other (M=14.3). These differences were not significant, p > .05.

**Factor Analysis**

A two-factor solution emerged from the analysis of the neurocognitive data, accounting for 60% of the total variance. Factor loadings ranged from 0.49 to 0.86. The first factor represented general cognitive ability, as it included verbal fluency skills, processing speed, working memory, and executive functioning skills. The second factor represented motor control, as it included tasks measuring fine motor control and visual-motor integration skills.

Summary scores from the parental adjustment measures, which included the PPS, CVS, CMCC, UMCSS-P, and PSI/SF, were all included in the principal components analysis. A three-factor solution emerged, which accounted for 69% of the variance. Factor loadings ranged from 0.6 to 0.9. The first factor represented parental stress and included all subscales from the PSI-SF. The second factor represented parental attitudes and perceptions surrounding their child’s illness and included coping styles and perceptions of child vulnerability. The final factor represented parental involvement/care and included overprotective behaviors, communication with health providers, and the effort and time involved in child-care.

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Predicting Functional Impairment

Correlation coefficients were computed among the three BIS subscale scores, the total BIS summary score, and the five new factor scores: general cognitive ability, motor control, parental stress, attitude/perceptions surrounding illness, and care/involvement. The purpose was to determine how neurocognitive deficits and parental adjustment related to functional impairment. Results are presented in Table 2. These results suggest that neurocognitive deficits are not strongly related to functional impairment in pediatric ALL survivors. Parental stress, however, one of the three parental adjustment factors, does seem to be significantly related to functional impairment, such that greater levels of stress are related to greater levels of impairment.

Table 2. Bivariate Correlations Among Functional Impairment, and Neurocognitive and Parental Adjustment Factors Scores.

<table>
<thead>
<tr>
<th></th>
<th>Interpersonal Impairment</th>
<th>School/Work Impairment</th>
<th>Self-Care/Satisfaction Impairment</th>
<th>Total Impairment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Motor Control</td>
<td>-0.095</td>
<td>0.125</td>
<td>-0.105</td>
<td>0.010</td>
</tr>
<tr>
<td>General Cognitive Ability</td>
<td>-0.209</td>
<td>-0.186</td>
<td>-0.180</td>
<td>-0.235</td>
</tr>
<tr>
<td>Parental Stress</td>
<td>0.320*</td>
<td>0.361*</td>
<td>0.282</td>
<td>0.408**</td>
</tr>
<tr>
<td>Perceptions and Attitudes</td>
<td>0.086</td>
<td>0.042</td>
<td>-0.218</td>
<td>-0.010</td>
</tr>
<tr>
<td>Care/Involvement</td>
<td>-0.147</td>
<td>-0.039</td>
<td>-0.260</td>
<td>-0.150</td>
</tr>
</tbody>
</table>

* Correlation is significant at p<0.05 level (2-tailed)
* Correlation is significant at p<0.01 level (2-tailed)
Additional correlation coefficients were computed among the four BIS summary scores and the raw scores obtained from all the neurocognitive measures and parental questionnaires. This analysis served as a reliability check, as the goal was to obtain consistent results across both correlation analyses. Consistent results would help ensure that no significant information was lost as a result of using the factors scores as opposed to the raw scores. Results were consistent with the previous analysis, though there were some minor differences. Among these differences was a significant correlation between the PPS and self-care/satisfaction impairment, a significant correlation between the Beery VMI (visual motor integration) and self-care/satisfaction impairment, as well as the Purdue right hand. However, these correlations were small and were non-significant after a Bonferroni adjustment was applied. Overall, results revealed no significant relationships between neurocognitive functioning, parental adjustment, and functional impairment, with the exception of parental stress. These results were consistent with the previous analysis and suggest that the use of the factor scores in further analyses was appropriate. The complete correlation matrix is presented in Table 3.

Additional correlations were conducted between BIS scores and demographic variables in order to determine if Functional Impairment was related to specific demographic factors such as education, income, and number of people in household. Overall, the results suggest that both level of parent education as well annual income are negatively related to functional impairment. Lower levels of education are related to higher levels of functional impairment. This relationship pattern is also evident for annual income. Results are presented in Table 4.
Table 3. *Bivariate Correlations Among Functional Impairment and Neurocognitive and Parental Adjustment Raw Scores.*

<table>
<thead>
<tr>
<th></th>
<th>Interpersonal Impairment</th>
<th>School/Work Impairment</th>
<th>Self-Care/Satisfaction Impairment</th>
<th>Total Impairment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Beery VMI</td>
<td>-.246</td>
<td>-.039</td>
<td>-.321*</td>
<td>-.202</td>
</tr>
<tr>
<td>Digit Span</td>
<td>-.179</td>
<td>-.068</td>
<td>-.024</td>
<td>-.111</td>
</tr>
<tr>
<td>Trails A</td>
<td>-.244</td>
<td>-.080</td>
<td>-.135</td>
<td>-.168</td>
</tr>
<tr>
<td>Trails B</td>
<td>.055</td>
<td>-.084</td>
<td>-.073</td>
<td>-.054</td>
</tr>
<tr>
<td>Purdue (Right Hand)</td>
<td>.099</td>
<td>.313*</td>
<td>.185</td>
<td>.277</td>
</tr>
<tr>
<td>Purdue (Left Hand)</td>
<td>-.208</td>
<td>-.080</td>
<td>-.187</td>
<td>-.177</td>
</tr>
<tr>
<td>Purdue (Both Hands)</td>
<td>-.163</td>
<td>.021</td>
<td>-.229</td>
<td>-.113</td>
</tr>
<tr>
<td>Verbal Fluency (FAS)</td>
<td>-.186</td>
<td>-.111</td>
<td>-.123</td>
<td>-.164</td>
</tr>
<tr>
<td>Parent Protection</td>
<td>-.184</td>
<td>-.041</td>
<td>-.364*</td>
<td>-.192</td>
</tr>
<tr>
<td>Child Vulnerability</td>
<td>.173</td>
<td>.122</td>
<td>-.028</td>
<td>.124</td>
</tr>
<tr>
<td>Vulnerability Scale</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Scale</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Table 3 (cont.)

<table>
<thead>
<tr>
<th></th>
<th>Interpersonal Impairment</th>
<th>School/Work Impairment</th>
<th>Self-Care/Satisfaction Impairment</th>
<th>Total Impairment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parental Stress Index</td>
<td>.297*</td>
<td>.361*</td>
<td>.226</td>
<td>.385**</td>
</tr>
<tr>
<td>Care of My Child With Cancer</td>
<td>.031</td>
<td>.020</td>
<td>.029</td>
<td>.033</td>
</tr>
<tr>
<td>Coping</td>
<td>.137</td>
<td>.027</td>
<td>-.103</td>
<td>.031</td>
</tr>
<tr>
<td>Communication with Health Professionals</td>
<td>.031</td>
<td>.069</td>
<td>-.012</td>
<td>.050</td>
</tr>
</tbody>
</table>

* Correlation is significant at p< 0.05 level (2-tailed)
** Correlation is significant at p< 0.01 level (2-tailed)

Table 4. Bivariate Correlations Between BIS Summary Scores and Demographic Variables.

<table>
<thead>
<tr>
<th></th>
<th>Interpersonal Impairment</th>
<th>School/Work Impairment</th>
<th>Self-Care/Fulfillment Impairment</th>
<th>Total Impairment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Education</td>
<td>-.266</td>
<td>-.241</td>
<td>-.183</td>
<td>-.290*</td>
</tr>
<tr>
<td>Annual Income</td>
<td>-.161</td>
<td>-.299*</td>
<td>-.237</td>
<td>-.306*</td>
</tr>
<tr>
<td>Adults in Household</td>
<td>-.022</td>
<td>.020</td>
<td>.035</td>
<td>.016</td>
</tr>
<tr>
<td>Children in Household</td>
<td>-.019</td>
<td>-.104</td>
<td>.084</td>
<td>-.048</td>
</tr>
</tbody>
</table>

* Correlation is significant at p< 0.05 level (2-tailed)
** Correlation is significant at p< 0.01 level (2-tailed)
Multiple regression evaluated how the new factor scores predicted overall functional impairment. The predictors were two neurocognitive factor scores and three parental adjustment factor scores and the criterion variable was the BIS total score. Neurocognitive and parental adjustment scores were entered stepwise, in two different blocks, with neurocognitive factors entered as the first block. The results of the analysis indicated that parental stress accounted for a significant amount of variance in overall functional impairment $R^2 = .166, F(1, 45) = 8.987, p = .004, B = .408, p = .004$. Therefore parental stress accounted for 19% of the variance in overall functional impairment.

Additional regression analyses were conducted in order to determine how neurocognitive and parental adjustment factors predicted impairment in the three specific domains of functioning (i.e. school/work, interpersonal, self care/self satisfaction). Results were similar to the initial regression analysis. Parental stress was on the only factor that accounted for a significant amount of variance in school/work impairment, $R^2 = .13, F(1, 45) = 6.730, p = .013, B = .361, p = .013$. Parental stress also accounted for a significant amount of variance in interpersonal impairment, $R^2 = .102, F(1, 45) = 5.123, p = .028, B = .320, p = .028$. Interestingly, however, parental stress was not a significant predictor of functional impairment in the self-care/self-satisfaction domain and no variables were retained in the model. These results suggest that self-care/satisfaction impairment may fundamentally differ from other domains of impairment in terms of predictive factors.

The BIS summary scores were then recoded to reflect two separate groups: those who met criteria for clinically significant functional impairment versus those who did not meet criteria. This dichotomous outcome variable was used in a logistic regression to determine if either neurocognitive factor scores and/or parental adjustment factor scores were significant
predictors of clinical caseness. Neurocognitive factors were forced into the first block (enter method) and parental adjustment factors were entered in the second block, also using the enter method. Results were consistent with the prior multiple regression analyses. A model containing only neurocognitive factor scores did not significantly predict whether criterion was met for clinical functional impairment. This was true of a model containing both neurocognitive and parental adjustment factors. These results are presented in Table 5.

Individual analyses of predictors revealed parental stress to be a significant predictor of clinical caseness. These results are presented in Table 6. Goodness-of-fit analyses revealed that while a model including all neurocognitive and parental adjustment factors was 91% successful at classifying non-caseness, the model was only 30% successful at identifying clinical caseness. These results are consistent with previous analyses and suggest that with the exception of parental stress, parental adjustment and neurocognitive deficits do not significantly predict functional impairment in pediatric cancer survivors.

<table>
<thead>
<tr>
<th>Test</th>
<th>$\chi^2$</th>
<th>df</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Block 1 (Neurocognitive Factors):</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Overall Model</td>
<td>1.701</td>
<td>2</td>
<td>.427</td>
</tr>
<tr>
<td>Block 2 (Parental Factors ):</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Block</td>
<td>8.241</td>
<td>3</td>
<td>.041</td>
</tr>
<tr>
<td>Overall Model</td>
<td>9.941</td>
<td>5</td>
<td>.077</td>
</tr>
</tbody>
</table>

Table 5. Omnibus Logistic Regression Analysis of Clinical Functional Impairment.
### Table 6. Logistic Regression Analysis of Individual Predictors of Clinical Functional Impairment.

<table>
<thead>
<tr>
<th>Predictor</th>
<th>B</th>
<th>$\chi^2$</th>
<th>df</th>
<th>p</th>
<th>$\hat{e}^B$ (Odds Ratio)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Motor Control</td>
<td>-.425</td>
<td>1.05</td>
<td>1</td>
<td>.306</td>
<td>.654</td>
</tr>
<tr>
<td>General Cognitive Ability</td>
<td>.346</td>
<td>.780</td>
<td>1</td>
<td>.377</td>
<td>1.41</td>
</tr>
<tr>
<td>Parental Stress</td>
<td>.994</td>
<td>6.14</td>
<td>1</td>
<td>.013</td>
<td>2.70</td>
</tr>
<tr>
<td>Perceptions and Attitudes</td>
<td>-.266</td>
<td>.488</td>
<td>1</td>
<td>.485</td>
<td>.766</td>
</tr>
<tr>
<td>Care/Involvement</td>
<td>-.168</td>
<td>.224</td>
<td>1</td>
<td>.636</td>
<td>.846</td>
</tr>
</tbody>
</table>

The recoded BIS summary scores reflecting the two categories of clinical caseness versus no caseness were then used to divide the sample into two separate groups. MANOVA was then conducted comparing these two groups across all summary scores for parental adjustment and neurocognitive deficits. Results of the omnibus MANOVA was not significant, $F(14, 32) = 1.1, p = .368$. Follow tests revealed that parental stress was the only variable that was significantly different across the two groups, $F(1, 45) = 6.9, p = .012$. Unfortunately, this value was no longer significant after applying the Bonferroni adjustment. These results are consistent with results presented above and suggest that pediatric cancer survivors who experience clinically significant functional impairment are no different than those without functional impairment in terms of parental adjustment and neurocognitive deficits. However, there may be minor differences in parental stress levels.
Chapter 5 Discussion

Functional Impairment

Previous research has revealed that pediatric cancer survivors are at risk for a variety of late effects following their diagnosis and treatment. These late effects include physical malfunctions, chronic health conditions, neurocognitive deficits, and social emotional problems (Dickerman, 2007; Oeffinger et al., 2006). Unfortunately, however, these late effects have mostly been studied in isolation, independent of one another. How these late effects converge and impact day-to-day living is currently unknown. This current study introduced a novel approach to the study of late effects by measuring impairments across broad functional domains.

Based on the distribution of scores on the BIS, the results suggest that pediatric cancer survivors demonstrate a range of functional deficits in school/work, interpersonal relations, and self-care/self-fulfillment. Bird and colleagues (2005) have recommended a score of 14 or higher to be used as a cutoff score indicative of clinically significant functional impairment. However, the authors have also suggested that a score of seven or higher be used for screening purposes and may be indicative of some problems with functional impairment. Based on these numbers, the rates of functional impairment in this sample were substantial, as 26% demonstrated clinically elevated scores on a measure of global functional impairment while over half the sample evidenced some aspects of functional impairment (i.e. participants scored higher than a cutoff score of seven). Although the Brief Impairment Scale is a newer measure and thus the comparison of scores across different populations is limited, the rates in this sample are sizeable and suggest that functional impairment is a problem for pediatric
cancer survivors. For pediatric cancer survivors the tasks of day-to-day living are not accomplished with ease.

These results fit nicely within the larger research of pediatric late effects. Late effects that result from the cancer experience are considerable and manifest as physical impairments, psychosocial problems and cognitive deficits (Dickerman, 2007; Krull et al., 2008; Oeffinger et al., 2006; Patenaude & Kupst, 2005). Given this broad manifestation of problems, it is no surprise that these late effects converge to disrupt day-to-day living.

**Neurocognitive Deficits**

Previous research has specifically highlighted two common problematic outcomes or late effects, which result from the pediatric cancer experience (Patuende & Kupst, 2005; Moore, 2005). The first identified outcome is neurocognitive deficits. More specifically, childhood cancer survivors have been found to demonstrate deficits across domains of attention, executive functioning, processing speed, memory, verbal comprehension visuo-spatial skills, and visuo motor functioning (Buizer et al., 2009; Campbell et al., 2007). Consistent with previous research, this sample of pediatric cancer survivors demonstrated significant neurocognitive deficits, as evidenced by their below-normative levels of performance on the neurocognitive measures. Contrary to expectations, however, there was no apparent relationship between these distinct neurocognitive deficits and overall functional impairment. These results suggest that while pediatric cancer survivors do continue to experience deficits across neurocognitive domains, these deficits do not adversely impact broad functioning.
Parental Adjustment

The second problematic consequence of the cancer experience is parental adjustment. More specifically, parents managing their child’s treatment for leukemia and lymphoma have been shown to demonstrate high levels of stress, distress, overprotection, uncertainty, and are more apt to perceive their child as vulnerable (Mishel, 1983; Pai et al., 2006; Thomasgard et al., 1995). However, results were mixed in terms of the congruence between parental adjustment difficulties within this sample and previous findings. While the current study found similar levels of overprotection, this particular sample of parents demonstrated lower levels of stress, perceived vulnerability, and uncertainty.

Of particular note, was the sizeable difference in reported parental stress when compared to previous findings. In this sample, parents of cancer survivors generally reported less stress. This is inconsistent with previous research, as results generally tend to favor an increase in stress (Kazak & Barakat, 1997; Vrijmoet-Wiersma, van Klink, & Kolk et al., 2008). However, most of the research has approached parental stress from a post-traumatic stress framework and suggests that the initial traumatic event of the cancer diagnosis facilitates a post-traumatic stress response. According to Barakat and Kazak (1997), the traumatic response can manifest in an acute phase (i.e. response to diagnosis and treatment) but can continue into a chronic phase. This PTS response often consists of symptoms such as: avoidance, hyperarousal, and intrusive thoughts (Norberg & Green, 2007). The chronic phase is thought to be the result of subsequent chronic stressors such continued treatment, medical late effects, and threat of recurrence and PTS rates have been found in 10% of mothers of cancer survivors with a mean survival rate of five years (Barak & Kazak, 1997). PTS symptoms, however, were not measured in the current sample, as the focus was more on
chronic stressors of daily living associated with cancer survivorship. As such, comparing the current findings of parental stress rates to previous findings of posttraumatic stress may be less useful and informative. Additionally, interpretation of the current results should proceed with caution when placing them within the context of PTS research.

Norberg and Green (2007) have suggested that parental stress is somewhat different than the initial PTS response. While PTS symptoms originate as a result of a traumatic event and involve intense symptoms of avoidance, hyperarousal, and intrusive thoughts; general parental stress, as was measured in the current study, persists long after treatment termination and remission status and involves ongoing daily stressors such as threat of relapse, general recovery, late effects, and reintegration back into daily life. Studies that have examined rates of parental stress and distress, rather than post-traumatic stress, have found clinically elevated rates in 12% of parents, in a sample of children up to fourteen years post-diagnosis (Boman, Lindahl, & Bjork, 2003). The results of the current study fall below this rate, with only 8% of the sample reporting clinically elevated stress levels. These observed lower levels of stress may be a function of time. Previous research has found that parental stress and distress seem to decrease over time (Boman et al., 2003; Wijnberg-Williams et al., 2006). For some participants in the current study, over ten years had passed since treatment termination. The observed low level of stress notwithstanding, parental stress was significantly, though minimally, related to functional impairment. As anticipated, the relationship was such that high levels of parental stress were predictive of higher levels of functional impairment. These results suggest that while most parents experience relatively low levels of stress, there is a small subset of parents who do experience higher levels of stress. These high levels of stress seem to be associated, though somewhat minimally, with functional impairment in the child.
Because functional impairment is a relatively new construct within the field of pediatric health, there is currently a dearth of research that explores the association between parental stress and functional impairment. One study by Ingerski and colleagues (2010) found a significant relationship between parent reported traumatic stress symptoms and reported functional impairment in children with chronic illnesses. The generalizability of the results, however, should be interpreted with caution, as it has been previously suggested that posttraumatic stress is fundamentally different than the stress and distress that accompanies survivorship. Nonetheless, these results point to a relationship between parental maladjustment and functional impairment. Additional research is limited and as such, future research aiming to replicate these findings and extend them beyond posttraumatic stress symptoms is warranted.

While Functional Impairment remains largely explored, there is a growing body of research, however, that has explored a related construct, Quality of Life (QOL). QOL is similar to the construct of Functional Impairment in that it attempts to move beyond the measurement of isolated problems in order to capture a compressive and holistic picture of the child’s life. The World Health Organization (1948) defines QOL as a multidimensional construct encompassing physical, mental, and social well-being. Palermo and colleagues (2008) have suggested that while functional impairment is a distinct construct from QOL, they do share similarities as they both assess more global capabilities such as participation in daily activities, rather than specific deficits. The authors also note that much fewer measures of functional impairment have been developed and as such there is less research on functional impairment as a whole.
There is a moderate amount of research that suggests a negative relationship between parental psychopathology (e.g. stress, anxiety, depression) and child QOL such that increases in parental psychopathology relate to decreases in child QOL (Kazak & Barakat, 1997; Roddenberry & Renk, 2008; Vance, Morse, & Eiser, 2001). Additionally, there are studies that have specifically examined the relationship between parental stress and specific problematic child outcomes. For example, in children with arthritis, parental stress predicted child anxiety and pain (Anthony, Bromberg, Gil, & Schanberg, 2011). In summary, despite limited research, the results of the current study are consistent with previous research examining the relationship between parental stress, posttraumatic stress, functional impairment, and QOL.

Although parental stress was predictive of global impairment as well as the specific domains of interpersonal relations and school/work, of note, was the lack of association between parental stress and the domain of self-care/self-fulfillment. This result, coupled with the significant differences across the three specific functional domains suggests that the functional domains of school/work, interpersonal functioning, and self-care/self-fulfillment are fundamentally different from one another both in severity and associated factors.

The findings of the current study, however, assume that the Brief Impairment Scale is accurately assessing all relevant aspects of Functional Impairment, which may not be the case. The BIS was not created as a diagnostic instrument, but was created as a screening instrument with the intention of providing supplemental information regarding functional impairment (Bird et al. 2005). As such, the BIS measures very broad aspects of functional impairment and may miss specific nuances of functional impairment. For example, functional impairment within the school/work domain is assessed through questions about
grades/performance and absences. This instrument does not afford additional depth, as other aspects of functioning are missed such as: difficulty, time and effort, study habits, feelings of competence, enjoyment, and school related anxiety. Therefore, it may be the case that the BIS screens for functional impairment far too broadly and is not sensitive enough to fully assess the degree to which functional impairment manifests within this population. As such, the null results may be due to this insensitivity rather than a lack of relationship. ROC analysis revealed a wide range of scores that might be appropriate to set as the clinical cutoff (Bird et al., 2005). The authors state that a clinical cutoff score of 14 might lack sensitivity and therefore suggest lowering the clinical cutoff to seven for screening purposes, sacrificing specificity for higher sensitivity. Future research should explore the relationship between neurocognitive deficits and functional impairment in greater depth by using a global measure of impairment that assesses components of global impairment in more depth and detail. Additionally, 16 percent of the children in the sample were receiving special education services. Functional impairment within a special education setting may be perceived differently than in a normal classroom setting. As such it is unclear how the BIS assesses functional impairment within this specific subpopulation of children.

**Implications**

First and foremost, the results of this study suggests that late effects do not exist in isolated domains but aggregate to impact broad functioning. As such, the assessment and evaluation of more broad functional impairment is necessary in this population. However, the significant differences across the specific functional domains as well as the differential predictive ability of parental stress suggest that global assessment of functioning is not sufficient and it is important to assess individual domains of functioning as well.
Additionally, increased parental stress can be used as a marker to help identify those individuals at heightened risk for functional impairment.

In addition to careful evaluation and assessment, the results suggest that some type of intervention may be needed to address this clinically significant functional impairment. Although the domains of school/work, interpersonal relations, and self-care/self-fulfillment are broader categories of impairment that still offer concrete targets for interventions, current interventions for pediatric cancer survivors are still lacking. Many interventions target very focused constructs such as specific cognitive rehabilitation. Unfortunately, however, how beneficial these interventions are for the individual in terms of broader day-to-day functioning is difficult to determine. For example, cognitive remediation interventions, which are intended to promote cognitive functioning, have only evidenced small to moderate positive results (Butler et al., 2008). Although Hardy, Willard, and Bonner (2011) found positive improvements in working memory and attention, as a result of a computer based cognitive remediation program, there is no research addressing the practical relevance of these improvements. For example, do improvements in working memory aid in academic performance or in peer relationships? The current study suggests that neurocognitive deficits do not map onto disruptions in day-to-day life and therefore interventions that attempt to target such deficits might lack practical relevance. However, as mentioned earlier, it could also be that the BIS were not a sensitive enough to pick up on the association.

The results of the current study suggest the need for future research to measure improvements in academic functioning, peer relationships and other broad functional domains in order to fully evaluate the efficacy of an intervention. For example, school reintegration interventions do focus on the specific domain of academic functioning. This
particular intervention is concerned with overall adjustment and the ease at which the child reintegrates back into the school setting. This represents a successful intervention that targets a broad functional domain that has practical relevance for a child’s day-to-day functioning (Prevatt, Heffer, & Lowe, 2000). The current study offers three additional specific and concrete targets for interventions, which have practical relevance.

Another implication of the current study is the possibility of an intervention targeted at the parents. The current study clearly supports a small positive relationship between parental stress and negative child outcomes. These results suggest that the parents themselves might also be targeted for interventions. Currently there is no research exploring how interventions targeted at the parents might improve child outcomes. However, there is research to suggest that parents are amenable to interventions intended to improve their own adjustment. Kazak et al. (2004), found positive results for a family directed treatment that was able to reduce intrusive thoughts in fathers of pediatric cancer survivors. Positive results were also found in the direction of decreased anxiety and posttraumatic stress for an intervention directed at caregivers of newly diagnosed children (Kazak et al., 2005). Results such as these only address the potential benefits to the caregiver and not the child. However, due to the bidirectional nature of adjustment difficulties within the parent-child relationship (Mishel, 1983; Varni et al., 1993), child improvement should also be considered when assessing the efficacy of interventions targeted at the parent to ameliorate stress.

Another implication favors a change in the research trend regarding neurocognitive deficits among pediatric cancer survivors. Research exploring the characteristics of neurocognitive deficits in pediatric cancer survivors has been undertaken with extreme vigor. The assumption underlying this proliferation of research is that the manifestation of
neurocognitive deficits has far reaching effects on other domains of the child’s life. The results of this study run contrary to that assumption and instead suggest that the presence of neurocognitive deficits is not particularly salient when it comes to overall functional capabilities. It is possible that pediatric cancer survivors have found a way to compensate for their neurocognitive deficits so that they don’t encroach upon other aspects of life. As such, it may be unnecessary to continue exploring neurocognitive deficits in this population with the previous vigor. However, there is currently no additional evidence that supports this interpretation.

Limitations and Directions for Future Research

This study represents a rather novel approach to the study of late effects in pediatric cancer survivors. As such, there were many limitations to the study and many directions in which future research may proceed. First, although this population demonstrated substantial rates of functional impairment, no control group was used in order to compare these rates. As such, there is no way to determine whether the rates of functional impairment in this population are significantly different than what would be found in a normal population. Other studies measuring isolated domains of functional impairment such as school/work functioning have found pediatric cancer survivors to demonstrate more impairment than age matched controls (Mitby et al., 2003; Pang et al., 2008). Additionally, pediatric cancer survivors also seem to experience lower QOL (Boman, 2007). However, future research should use aged matched controls in order to accurately capture how functional impairment affects pediatric cancer survivors across a variety of functional domains, in comparison to healthy children.
In a similar vein, this study was limited to a specific cancer diagnose of leukemia or lymphoma and excluded other cancers such as tumors and CNS cancers. As such, results regarding functional impairment can only be applied to leukemia and lymphoma and cannot be generalized to all cancers. Future research should then explore profiles of functional impairment across all cancer diagnoses.

Another limitation of the study was that parents were self-reporting their own personal adjustment as well as reporting on their child’s functional capabilities. Inherent in this design is the potential for biased reporting. For example, parents experiencing higher levels of stress may have a predisposition to perceive things with a negative bias. According to the “depression-distortion” hypothesis, parental depression has been strongly associated with a tendency to over report child behavior problems (Garstein, Bridgett, Dishion, & Kaufman, 2009). This perceptional distortion may also be true of stress and as such may be responsible for the relationship between parental stress and the problematic child outcomes reported in this study. In this way, parents who report relatively higher stress may be more prone to report functional impairment regardless of whether or not their child is truly experiencing functional impairment. Additionally, parents may not be fully aware of their child’s experiences and therefore may be ill equipped to report on them. For example, previous research shows a low level of concordance between parent and child report on measures of Quality of Life (Levi & Drotar, 1999). Future studies should employ additional informant measures of functional impairment such as child report, teacher report, and observation.

Another limiting factor of the study was the small sample size and the associated consequences. Due to the volume of patients seen at the clinic where data was collected, it
was projected that within a year the sample size would reach between fifty and sixty. Therefore, in order to maintain adequate power, the number of measures used to predict functional impairment was limited. However, it is likely that these measures did not adequately predict functional impairment. Although parental stress did account for some of the variance in functional impairment, over 83% of the variance in functional impairment remains unaccounted for. Therefore future research should work to better identify the factors associated with functional impairment. A large body of research supports a positive relationship between parental psychopathology and negative child outcomes in the form of both internalizing and externalizing behavior problems (Middleton, Scott, & Renk, 2009). Based on these findings, additional parental factors that might predict child functional impairment include: parental depression, anxiety, and posttraumatic stress symptoms (Davis, Parra, & Phipps, 2010; Roddenberry & Renk 2001; Vance et al., 2001). The results of the current study also suggest further exploration of demographic variables such as level of education and annual income.

Another limitation to the current study was the directional ambiguity of the relationship between parental stress and functional impairment. It was unclear as to whether parental stress contributed to an increase in functional impairment, vice versa, or both. Unfortunately, causality remained outside the scope of the current study. There remains limited research regarding the directionality of parental adjustment and problematic child outcomes, as most research has been correlational in nature. One hallmark study conducted by Davis and colleagues (2010) attempted to circumvent the directional ambiguity by examining the mediating role of child anger regulation within the relationship of parental posttraumatic stress symptoms and problematic child outcomes. The results of this study
found that poor anger regulation on behalf of the child accounted for the significant relationship between parental stress and problematic child outcomes. The authors suggest that the meditational relationship can be understood in terms of an emotion socialization framework. Because of their own PTS symptoms, parents may be less able to model and teach appropriate emotion regulation skills. Thus, children of parents who experience PTS symptoms demonstrate more regulation difficulties, which in turn, drive overall behavioral problems. This explanation favors of a causal relationship between parental posttraumatic stress and negative child outcomes. The generalizability of the results, however, again should be interpreted with caution, as it has been previously suggested that posttraumatic stress is somewhat different than the stress and distress that accompanies survivorship.

Unfortunately, however, the current study did not assess posttraumatic stress symptoms but instead assessed general parental stress and as such, this causal explanation may not apply to the current study. Additionally, no meditational analyses were conducted as a part of the current study. As such, there is not enough information within the current study to indicate a directional relationship and the interpretation of causality is only speculative. Future research should attempt to delineate a directional relationship between parental factors and child functional impairment outcomes. Rodenberry and Renk (2007) suggest possible mediating variables such as strength of the parent-child relationship, family communication, and family functioning.

**Summary**

In sum, previous research regarding the late effects of pediatric cancer survivors has been rather limited in scope, as neurocognitive and parental maladjustment late effects have only been examined in isolation and not in relation to overall functional impairment. This
study addressed the question of broad functional impairment and found that a considerable amount of pediatric cancer survivors do experience impairment in school/work, interpersonal relations, and self-care/self-fulfillment. Results indicated that neurocognitive and parental maladjustment did not significantly predict functional impairment, with the exception of parental stress. However, this study is only a pilot study and more research needs to be done to further explore characteristics of functional impairment as well as contributing factors within this population.
References


