

THE IMPACT OF FAMILY FUNCTIONING ON CHILDREN'S ADAPTATION
DURING A PARENT'S BONE MARROW TRANSPLANTATION

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DEDICATION

I would like to dedicate this work to families of cancer and BMT patients, to their caregivers, especially at Indiana University and St. Louis University Hospitals, and lastly, to my mother, who for years, diligently persevered and fostered our family through my father's illness.

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ABSTRACT

Mary L. Spath

THE IMPACT OF FAMILY FUNCTIONING ON CHILDREN'S ADAPTATION DURING A PARENT'S BONE MARROW TRANSPLANTATION

Bone marrow transplantation (BMT) is being used ever more widely for advanced and refractory malignancies. The family unit and individual members are profoundly affected by this treatment process. Few studies have examined the effect of parental BMT on the family, and there are no known studies that have investigated the impact of parental BMT on children. A descriptive design with longitudinal data from 61 children, ages 10-18, examined children's adaptation, characterized as emotional and behavioral response, during the acute phase of parental BMT. The study included 3 time points: pre-transplant, during parental hospitalization, and one month after transplantation. The Response to Stress Questionnaire, and subscales from the Child Health Questionnaire and Family Environment Scale were used to assess child, parent, and family variables associated with child adaptation. Child emotional and behavioral response significantly improved over the course of the parent's transplant, and significant changes in children's use of coping strategies at each time point were found. The model accounted for 27% to 46% of the explained variance in child behavioral response, and accounted for 41% of the explained variance in emotional response prior to the parent's BMT and one month after BMT. The model did not explain the variance of child emotional response, however, during the parent's hospitalization. Family structural change, family conflict, and disengagement coping were found to be the predominant variables significantly associated with more negative child behavioral response across the transplant trajectory.

Female child gender and increased use of disengagement coping before the parent's BMT, autologous BMT during the parent's hospitalization, and increased family structural change when the parent returned home one month later were significantly associated with more negative emotional response in children. Additional cross-sectional and longitudinal studies, using mixed methods, and include both parent and child data, are needed to substantiate the validity of findings. The data also suggest that significant variables in this model warrant further study to determine their association with one another and to refine the model to better explain children's adaptation.

Betsy L. Fife, PhD, RN

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CHAPTER 1

THE NATURE OF THE STUDY

Introduction

Children of parents with acute medical illnesses share in their parent's illness. The family, which generally provides the primary source of stability for children during illness, may also be the source of stress. The study of the adaptation of children to changes and disruptions caused by a parent's illness is in its early stages. This study will examine how family environmental variables are associated with the child's psychosocial adaptation to parental bone marrow transplant.

Bone marrow transplantation (BMT) is being used ever more widely for advanced and refractory malignancies; approximately 16,700 were conducted in 2006 alone (Center for International Blood and Marrow Transplant Research, 2006). Introduced in 1956, the use of this therapy has grown in the treatment of both hematologic and solid tumors. There are two types of transplants: allogeneic transplants are primarily used as the definitive treatment in hematologic malignancies (leukemias, anemias, and some lymphomas); and autologous or peripheral blood stem cell (PBSCT) transplants are used in multiple myeloma, testicular, neuroblastoma, lymphoma, and other solid tumors. Both types of transplantation are aggressive therapies in that high doses of ablative chemotherapy and/or radiation are given followed by infusion of bone marrow or stem cell rescue to offset the therapy's toxic effects. The dose-related intensity of this therapy causes toxicities to be higher than routine chemotherapy and radiation. Hospitalization, generally lasting two weeks up to several months due to extensive and life-threatening side effects, such as acute immunosuppression, results in a prolonged and tenuous

recovery period (Andryowski, 1994; Neitzert, Ritvo, & Dancey, 1998; Shivnan, Shelton, & Onners, 1996; Zittoun, Achard, & Ruszniewski, 1998). Prolonged hospitalization at transplant centers, often at a distance from the family's home, and changes within the family resulting from caregiving in the immediate home recovery period, cause separation and lack of continuity in the family. Adaptation of family members to these changes and illness demands may instigate significant emotional and behavioral stress related problems.

Effects of Acute Illness on the Family

Emphasis on the holistic nature of treatment and healing, which has included the family of the ill individual, has evolved over the last 20 years. The effects of illness in one family member have been found to affect and provoke changes in the entire family (Northouse, 2005; Weihs & Reiss, 1996; Zabora, 1992).

Parental illness has been found to pose unique problems. Studies have demonstrated that decreased ability of the ill parent to perform employment and domestic tasks, changes in family and individual member's activities, hospitalization, increased illness demands, and focus on the ill parent and his/her needs have instigated changes in daily family routines, task delegation, and role performance.

Researchers have found that parental illness can cause distress within the family unit as well as for individual family members. In an early study by Peters and Esses (1985) families with a parent with multiple sclerosis were compared to families with non-ill parents. Increased levels of family conflict and decreased levels of cohesion were found in the affected families. A series of studies on families with maternal breast cancer by Lewis and colleagues (1989, 1993, 1996) linked mothers' and fathers' depressed

mood to decreased marital adjustment and higher illness demands, which were associated with less effective family coping behavior and child dysfunction.

Indirect and subtle changes in parenting style in both parents due to the emotional and physical impact of the illness have been noted to have effects on the spouse, children, and the overall family unit (Armistead, Klein, & Forehand, 1995). Increased parental irritability, shouting, and expressions of stress were noted in families with a parent with multiple sclerosis (Rehm & Catanzaro, 1998). Decreased patience, during times of family conflict and when managing parenting demands, was noted by both parents. This was attributed to fatigue, a sense of the need to protect the ill spouse by the healthy spouse, and decreased tolerance for stress by the ill parent. Families' engagement in activities lessened. Families reported that more planning and less spontaneity were associated with activities, while adjustments were made to include the affected parent in more sedentary family activities. Many of these characteristics have been similarly reported by families living with parental cancer (Rehm & Catanzaro, 1998).

The psychological hope for success and the fear of failure that accompanies the radical nature of BMT can be overwhelming to patients and their families. Lack of future orientation, reluctance to plan ahead, postponed engagements, and delayed vacations were found in an early longitudinal study of BMT families (Zabora, 1992). Although only preliminary findings were provided in the literature from this study, it was clear that uncertainty regarding the outcome of the BMT resulted in families feeling as though they were suspended in the moment, and required them to put the future on hold.

Provision of both physical care and emotional support by family caregivers of BMT recipients is noted in the literature. Strains associated with the complexities and

acuity of care, the extended duration of recovery, the generally younger age of caregivers and families, and difficulties in reintegrating the ill family member back into the family unit are described (Eilers, 1997; Eldredge et al., 2006; Stetz, 1996).

Only a few studies have examined the family during bone marrow transplantation. In a longitudinal study of families pre- and post-autologous and allogeneic bone marrow transplant, Keogh (1998) identified a high level of psychological distress in family members at the pre-transplant and three month post-transplant timepoints, which was then resolved over the course of the next nine months. Molassiotis and colleagues (1997) found that strong family relationships were associated with psychosocial recovery and quality of life in both bone marrow and cancer patient groups. Not surprisingly, they also found that these groups were significantly different in terms of family cohesion, control, and conflict when compared with a control group of families without an ill member.

Effects of Parental Acute Illness on Children

Significant stressful life events, such as parental illness, may pose increased risk for psychological distress and disorder in children and adolescents (Grant & Compas, 1995). Serious parental illness was found to affect approximately 5-15% of children and teens in an early study of a cancer population (Worsham, Compas, & Ey, 1997). Similar findings (13-14%) were reported in a recent study conducted on adolescents of parents with a variety of medical illnesses (Pedersen & Revensen, 2005).

Over the past 20 years recognition of the impact of a parent's physical illness on children has been more widely explored. This is largely due to a more holistic view of illness and healing that has recognized the importance of the family. However, the study of children's response and adaptation to parental illness still remains in its early stages.

While overall findings provide definitive evidence of children's vulnerability, the extent and types of distress have been varied.

Most of the research regarding children of parents with illness has been focused upon three objectives: a) to identify the presence of psychosocial distress in children, b) to examine the types of psychosocial distress caused by the parent's illness, and c) to identify variables that may play a significant role in determining the child's response and adaptation. The goal of investigators is to target children most vulnerable to psychosocial morbidity so that effective interventions can be constructed. Significant and replicated findings on some individual child variables, such as adolescent age and female gender, are associated with increased child vulnerability to psychosocial distress. The quality of the family environment is generally recognized as an influential variable, however, specific characteristics, such as family cohesion and family conflict, and specific qualities of family relationships require more study.

Only one study could be found in the literature that specifically mentioned the effects of parental BMT on children. Examining the family's response to parental bone marrow transplantation, Zabora and colleagues (1992) published some preliminary data on their findings from a study of 160 family members. The authors did not report the number of children in this sample. A narrative summary of children's emotional response by age group was reported. No final results of this study were found in the literature. Therefore, despite the increased use of bone marrow transplantation and the growing study of children's response to parental illness, children's adaptation to parental bone marrow transplantation has been only minimally examined. Because BMT is a high-risk therapy used in acute malignancies and often used when front-line therapies have failed,

high levels of anxiety and uncertainty regarding the potential outcome of the therapy, the possibility of the parent's death, and the changes in the family are likely to have a substantial impact on the well-being of the children in these families.

Influence of the Family on Children's Adaptation

Within the domain of the social sciences, the family has been found to be a primary significant influence on children's development and in their response to stress (Grant & Compas, 1995; Berk, 1997; Bjorklund, 2000). The individual and collective interactions of family members with one another and the overall function of the family to encounter and manage stressful situations is known to be crucial to their emotional, behavioral, and social stability. Family functioning has been found to influence a child's ability to adapt to the changes incurred by a parent's illness (Huizinga et al., 2003; Watson et al., 2006; Woods & Lewis, 1995). In developing models for child adaptation to parental illness, family cohesion, family communication, family coping style, and family attachment style have been identified as integral components, although the roles of these variables as mediators and moderators of child adaptation are currently only hypothesized (Pedersen & Revensen, 2005), and remain unclear. The importance of examining family variables for the significance of their effect on children's adaptation to parental illness is essential in directing clinical services and supportive interventions and in identifying children and families at risk.

Theoretical Background

Systems theory has provided a basis not only for understanding the family as an interacting dynamic system, but has been used to conceptualize how individuals within the context of the family respond to illness (Rolland, 1987). Family systems models

acknowledge the inter-relatedness of the individual family member to the family unit, that is, the family is affected by and affects the individual member.

The proposed theoretical framework conceptualizes children's adaptation within the context of family adaptation, and conceptualizes the two as being interrelated. The trajectory is characterized by change and upheaval in the family as the child struggles to adapt. Minuchin's theory of family interaction (1987) served as a source theory for this model. Minuchin views the family as an interacting system that responds to internal and external changes by transforming itself to maintain function and continuity. This response is accomplished through the realignment of transactional patterns of individual family members during times of stress. Minuchin emphasizes that the ability of family members, including children, to mobilize and use alternative methods to maintain function influences both individual and family adaptation. Relational statements embedded within this theory that provide primary assumptions for this research are as follows:

- (a) individuals within a family do not resolve problems independently of one another;
- (b) support and resources available within the family influence adaptation to stress; and
- (c) perception is based upon an individual's estimation of his/her resources for confronting a stressor.

Appraisal, as understood within the framework of Lazarus' Stress and Coping theory (1966), is also an underlying theoretical construct in the examination of children's perception of and response to their parent's illness. Lazarus proposed that stress is the result of a transactional process between the person and environment that is affected by cognitive, affective, and coping variables (Lyon, 2000). The integration of psychosocial factors and cognitive ability contributes to the *individual* cognitive process from which a

person perceives and then appraises a situation. Because cognitive development is a progressive mental ability (Hockenberry, 2005), children's age and developmental maturation are influential in their cognitive processing abilities.

Although there are several models of family adaptation to illness in the literature, only two currently exist for children encountering parental illness (Armistead et al., 1995; Pedersen & Revenson, 2005). Both are limited by either their narrow focus or by their lack of testing. The model illustrated in Figure 1, page 22, was formulated to include variables derived from accepted psychological and sociological constructs related to the environment and population of this study; however, it includes only those contextual and interpersonal variables that are measured for this study.

The Proposed Study Model

The child's perception of the impact of the illness is highly influenced by the child's cognitive ability (Woods & Lewis, 1995; Lewis & Hammond, 1996) and by the family's ability to manage during the course of the parent's illness. In this model, the family provides the primary context in which the child constructs his/her view of the parent's illness. The family's interaction and the ability of the family to manage change brought about by the illness influence the child's interpretation of the illness. The family, which has traditionally been viewed as a primary source of stability for children, may also be a source of child stress. Separation due to prolonged or recurrent hospitalization, changes in family routine, interruptions that occur in family life, and family conflict due to parental illness are often sources of stress. The child's emotional and behavioral status and the stability of the family are reciprocally and mutually interrelated. Assured by this model, is the contribution of each family member, including the child, to family stability

and family adaptation. At the same time, the family system influences the adaptation of each member. In other words, not only does the level of family stability affect the child's ability to integrate and adapt to the crisis, but the child plays an important role in the overall integration and adaptation of the family.

Children's coping is a primary mediator affecting how the context of the situation affects the child's adaptation. Because coping is a process, coping behaviors used at the outset of the transplant trajectory may have a significant impact on the child's long-term coping and adaptation.

Statement of the Problem

There is a lack of research that examines family function and children's response during the course of a parent's BMT therapy. Only a few studies between 1992 and 2005 have examined family stress, family care giving needs, or the influence of the family unit on the psychosocial integrity of the family members of the transplant recipient (Foxall & Gaston-Johansson, 1996; Wilkins & Woodgate, 2007; Wochna, 1997; Zabora et al., 1992). None of them focused on the impact on the child from the child's point of view. Given the increasing use of BMT and gaps in the current literature, there is a need for research that identifies the points in the BMT trajectory when children are most vulnerable, variables indicating specific children who are most vulnerable, and those variables that have the greatest impact on child adaptation. These findings could then be used as the basis for developing interventions that positively influence their adaptation at this most stressful time.

Purpose of the Study

The purpose of this study is to increase understanding of how children adapt to the stress of acute parental bone marrow transplantation. Specifically, this study will examine the effect of the variables included in the proposal's conceptual model on the adaptation of children, 10-18 years of age. These variables will include: (a) contextual factors; (b) **the child's** perception of the impact of the parent's cancer/BMT within the context of the family (here referred to as "perceived illness impact"); and (c) **the child's** use of coping on the outcome, adaptation, which is defined as emotional and behavioral response. The proposed study will examine these variables during the acute phase of the BMT which includes three specific data points: pre-transplant, during the parent's BMT hospitalization, and one month after the parent's hospital discharge. The analysis will be based on *children's self-reported* responses to selected items from three standardized instruments. Sixty-one children will participate in the study. The results of the study will provide information and understanding on how children perceive parental illness and on the effect of family characteristics on child adaptation

Research Significance

This research is significant because it will add to the overall empirical knowledge about how children are affected by parental illness. This study will be of particular value because there are few longitudinal studies in the literature that have explored changes in children during parental illness over time. Identifying specific time points at which children are likely to be particularly vulnerable during their parent's treatment trajectory will be helpful in planning interventions and in targeting specific when intervention will be most efficacious. At this time, there are no known studies that have specifically

examined children's response to parental bone marrow transplant, a therapy which has been increasingly utilized in the past 20 years to treat acute malignancies. Research findings from this study will benefit families as they, along with the patient, cope with the anxiety and changes that are a part of bone marrow transplantation.

In the more recent literature, there is increased acknowledgment of the important role of the family during times of parental illness, which has been noted to be associated with child psychosocial outcomes (Harris & Zakowski, 2003; Huizinga et al., 2003; Watson et al., 2005). Researchers acknowledge that future theory and research efforts must incorporate the environment, as well as the development of the child, when studying children in the context of parental cancer if this work is to be most meaningful (Lewis, 2007). This study includes both these categories of variables. Measuring family variables that may influence the child's perception of the impact of the parent's illness and adaptation, *from the child's point of view*, will promote understanding of what these variables contribute to psychosocial outcomes. Further understanding of how contextual variables, specifically child demographic and parental illness characteristics, affect perception of illness impact in children of parental BMT recipients will also add to previous research findings of all children affected by acute parental illness. Most importantly, this knowledge is necessary for the assessment of child vulnerability and the planning of interventions that are effective.

Conceptual and Operational Definitions

The following conceptual and operational definitions for this study were drawn from sociological and psychological constructs and are presented to aid in the understanding and clarity of this study. The outcome variable for this study, adaptation,

will be is conceptually defined as child emotional and behavioral response. The independent variables include contextual parent and child variables, the child's perception of the illness impact, and coping. These are conceptually and operationally defined.

Child Adaptation

Adaptation is a broad term that has been conceptually viewed as both a process and an outcome (Roy, 1997). Within the context of illness, coping has been associated with the process of adaptation. Adaptation, viewed as an outcome, has most often been conceptualized as psychological adjustment and has been associated with affective and behavioral well-being, quality of life, and life satisfaction (Rodin, 2003; Houtzager et al., 2004; Molassiotis, 1997). Child adaptation is the central concept examined in this study and it is the outcome for the theoretical framework. Adaptation is conceptually defined as the ability of the child to maintain emotional and behavioral stability during the disruption that occurs as a consequence of a parent's BMT. The family comprises the overarching context within which child adaptation occurs.

Operationalization. Child adaptation will be operationalized as child emotional response and as child behavior. Two subscales of the Child Health Questionnaire (CHQ-CF-87) by Landgraf, Abetz, and Ware (1996) will be used to measure these dimensions of child adaptation. Each of these variables will be reported upon separately in the presentation of results.

Coping

Coping is a dynamic multidimensional process manifested by cognitive and behavioral responses to an appraised stressor (Lazarus & Folkman, 1984). Coping is

distinguished from response to stress in that it involves deliberate efforts to manage the stressor and/or efforts to manage one's emotional response to the stressor. According to Lazarus and Folkman, adaptation outcomes of the coping process involve dimensions of quality of life, such as emotional, social, and behavioral well-being. Types of coping responses have been characterized and categorized by numerous theorists (Compas et al., 2001; Lazarus & Folkman, 1984).

Perception of the illness impact which influences coping response will be understood within the context of Lazarus and Folkman's theory (1984) of cognitive appraisal. According to this theory, perception of an event implies that an individual interprets an experience in view of its effect on his/her well-being. A subsequent judgment or inference about the event, an appraisal, is then made within the context of the individual's perception of the consequences or potential harm (Folkman et al., 1986). This evaluation is the result of cognitive perceptions of the individual experiencing the event.

The child's cognitive ability to understand and interpret the situation is affected by developmental maturation and by environmental cues. Because children may be less able to interpret the disease-related (biomedical) aspects of a parent's illness because of cognitive limitations, reliance on contextual and family environmental factors are highly influential in how the illness is perceived and construed by the child.

Operationalization. Coping will be measured using three of the subscales of the Responses to Stress Questionnaire (Connor-Smith et al., 2000). Based on a theoretical model of stress and coping in children by Compas and colleagues (1999), these three subscales will differentiate the types of coping strategies used by children: primary

control engagement coping, secondary control engagement coping, and disengagement coping. Engagement coping refers to deliberate responses that are directed toward a stressor. Primary engagement coping strategies are directed at changing the stressor or one's response to the stressor, such as problem solving and emotional regulation; secondary engagement coping strategies are focused on adaptation to the stressor, such as acceptance and distraction. Disengagement coping strategies are conscious deliberate efforts to deny and avoid the stressor.

Perception of Illness Impact

Perception of the illness impact will be measured as two family dimensions: family structural change and family interaction. Each of these dimensions will be described and operationalized individually.

Family Structural Change

Family Structural Change refers to changes in day to day family routines, roles, and functioning that occur as a result of the parent's illness. These changes often result in overall stress and disruption in the life of the child.

Operationalization. This concept is operationalized by eight items drawn from two questionnaires, the Response to Stress Questionnaire (Connor-Smith, Compas, Wadsworth, Thomsen, & Salzmen, 2000) and the Child Health Questionnaire (CHQ-Child Form-87, Section #9, You and Your Family; Landgraf, Abetz, & Ware, 1996). These items will be combined to assess the child's perception of the illness in regard to changes in family roles, family functioning, and disruption in everyday family activities. Participants will rate the amount of individual stress generated by the perceived effects of these structural changes.

Family Interaction

Family Interaction is a concept that is multi-dimensional. In this study, family interaction will be conceptualized as family cohesion, family conflict, and family expressiveness. Family cohesion is a multi-faceted concept that describes the family's feeling of closeness, as well as the member's ability to support and relate to one another, and to mobilize effectively as a unit despite adversity. Family conflict is the amount of openly expressed discord among family members. Family expressiveness is the degree to which family members are supported by one another in the open communication of feelings and concerns. Recent findings suggest that these variables may be influential in family and child coping and adaptation to illness (Harris & Zakowski, 2003; Huizinga et al., 2005; Watson et al., 2006).

Operationalization. **Children's perception** of the family environment will be measured using the Family Cohesion, Family Conflict, and Family Expressiveness subscales of the Family Environment Scale (Moos & Moos, 2002). These subscales combined are referred to as the Family Relationship Index of the larger scale which has been psychometrically tested and used in other studies (Edwards & Clarke, 2004; Fuemmeler et al., 2003; Robinson et al., 2009). This index measures the social and environmental characteristics of the family climate as represented by family members' commitment, openness, and discord with one another that may be influential in children's adaptation.

Person Contextual Variables

The person contextual variables pertain to child and parent demographic factors and factors related to the parent's illness, which include symptom interference, length of stay, and type of transplant.

Child Contextual Variables

The context provides a pre-existing framework within which all other events and situations take place and the framework within which adaptation occurs. Child age, gender, and chronological placement in the family will be examined for their relationship to both the child's perception of the illness impact and the outcome variable, child adaptation. Previous studies on children's adaptation to parental cancer and other acute parental illnesses have reported that age and gender have been associated with children's responses (Compas et al., 1994; Heiney et al., 1997; Korneluk & Lee, 1998; Nelson & While, 2002; Watson et al., 2006). Chronological rank of child in the family has not been studied. Collectively, the literature suggests that the greater cognitive abilities and developmental capabilities of adolescents, when compared to younger children, may predispose older children to heightened distress.

Operationalization. Age was determined by the participant's chronological age in years. Gender was designated by the classification of participants into two groups, male and female.

Parent Contextual Variables

The recipient parent's gender and the type of transplant will be examined for their relationship to the child's perception of the illness impact and to the outcome variable, child adaptation. The association of these variables with perception of illness impact and

adaptation variables has been studied in the literature with varied findings (Osborn, 2007; Visser et al., 2004).

Variables related to the illness experience (length of hospital stay and symptom interference) of the parent will also be examined. These variables are hypothesized to affect the child's perception of the seriousness of the illness and to affect the overall stress of separation and disruption. Findings in the literature suggest that symptom interference, resulting in the parent's functional impairment and psychological distress, are associated with both family and child adaptation (Hoke, 2001; Howes et al., 1994; Lewis & Hammond, 1996; Nelson & While, 2002; Visser et al., 2004). Length of hospital stay has not been studied as a variable associated with child adaptation to parental illness.

Operationalization. Parent gender was classified into one of two groups, male or female. Type of parental transplant was classified as either autologous or allogeneic. Length of hospital stay was operationalized as the number of inpatient hospital days including the day of admission and discharge.

Symptom interference is defined as experienced sensations or changes in the body associated with a particular disease that hinder, obstruct, or impede one's normal daily function (www.wordnet.princeton.edu). Parent symptom interference was operationalized through a score derived from the BMT Symptom Checklist which indicated the extent that the patient perceived a symptom interfered with daily living (Fife, 2000).

Assumptions

The study was based on the following assumptions:

1. Psychosocial development is influential in a child's perception and appraisal of an event.
2. The family provides the primary contextual environment in which the child lives.
3. Children's emotional and behavioral responses are affected by environmental influences.
4. Children use coping strategies in dealing with stressors.
5. Child participants will respond honestly regarding their perceptions and feelings during the interview.

Limitations

1. The study data were taken from a convenience sample that does not include adequate representation of minority groups.
2. The study data were obtained from children and thus represent only the perceptions of children.
3. The study sample was obtained from participants at a single site and may limit the generalizability of the results to other types of settings.
4. Two of the tools used in the study were adapted from parent instruments and had limited psychometric testing done with children prior to the study.

Study Aims and Research Questions

The following study aims and corresponding research questions will be tested to address the previously described purpose of the study, as well as gaps found in the literature.

Aim 1: To examine the bivariate relationships among all variables in the proposed model at each time point. The strength of these relationships will then determine which variables will be included in the regression model with adaptation measured by behavioral response and emotional response, as the dependent outcomes.

Aim 2: To examine changes in the effect of all variables in the model, which include family structure and interaction, coping strategies used, and the level of adaptation across three time points in the BMT trajectory: (T1) pre-transplant and prior to hospitalization; (T2) approximately two weeks after infusion of bone marrow/stems cells during hospitalization for the BMT; and (T3) one month after the transplant.

Aim 3: To examine the effects of the independent variables in the model as factors that impact the child's adaptation to the stress of a parent's BMT as they are added hierarchically to the regression at each of the three time points. (The theoretical model is found in Figure 1, page 22).

Research question 3.1: What is the effect of children's coping strategies on adaptation, characterized as behavioral and emotional response?

Research question 3.2: What is the effect of the child's perception of the illness, including family structural change and family interaction, on adaptation controlling for coping strategies?

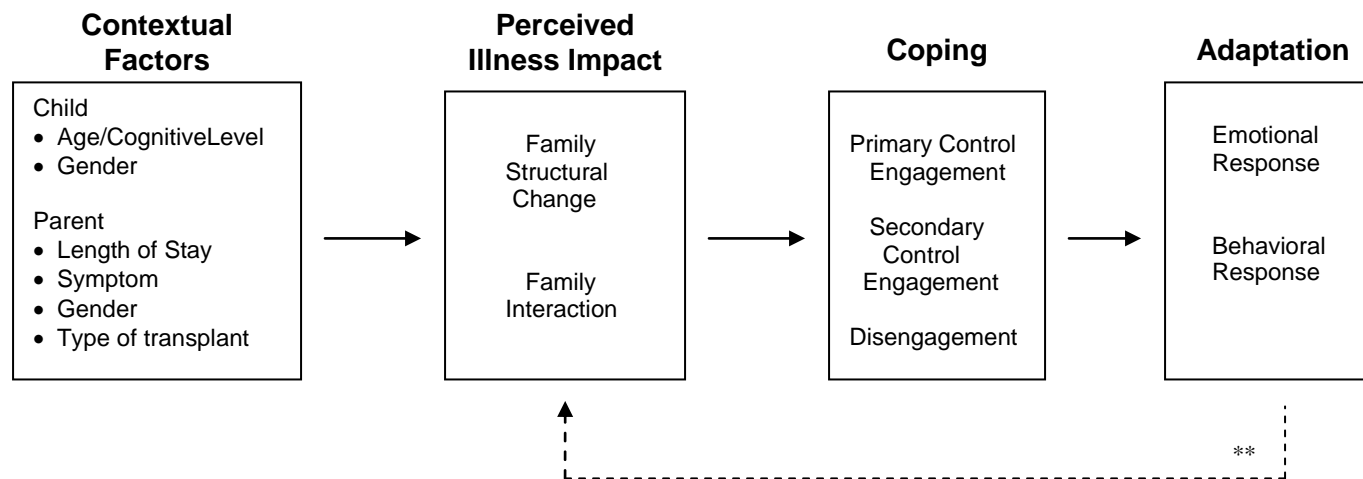
Research question 3.3: What is the effect of the child's age and gender on adaptation controlling for the child's perception of illness impact and coping strategies?

Research question 3.4: What is the effect of the recipient parent's gender, type of transplant, length of hospital stay, and symptom interference on adaptation controlling for the child's age and gender, the child's perception of illness impact, and coping strategies?

Summary

Children of parents undergoing bone marrow transplant for acute and recurrent malignancies are an important and understudied population. Research done on children of parents with other acute illnesses has shown that children share in illness-related psychosocial distress. The increased use of bone marrow transplant as a therapy and the use of BMT in younger patients who are in the child-rearing developmental stage of their lives, contributes to the need for research in this area. Because this population of vulnerable children has not been studied thus far based on findings in the literature, information about children during this type of family crisis will be significant and add important understanding to the adaptation of this population. The relationships among child factors, parent factors, perceived illness impact, and coping as they affect child adaptation to parental illness should be examined to identify children at risk and to design and target interventions that may be effective in managing secondary psychosocial morbidity. Chapter 1 has provided an overview of the problem, and includes discussion of the relationships among variables within the theoretical model, the purpose, research questions, and conceptual framework for this proposed study. Chapter 2 will provide an extensive discussion of the current literature related to the constructs of this study.

Figure 1. A Model of the Impact of the Family on Children's Adaptation to Parental Bone Marrow Transplantation



** The dashed line in the model from Adaptation to Perceived Illness Impact indicates that this path will not be included in the analysis, but implies the processual nature of coping and adaptation.

CHAPTER 2

REVIEW OF THE LITERATURE

This chapter will provide an overview of the theoretical constructs used in this study and a review of the empirical literature concerning children's adaptation to parental cancer and bone marrow transplant. A review of literature pertaining to those constructs in the theoretical model found in Figure 1, that together affect children's perceptions of the impact of the illness and the BMT on their lives, will be discussed. This will include children's developmental processing of illness, and variables pertaining to family interaction and family structural change, and coping strategies children use in their struggle to adapt. A review of the literature relative to these constructs will follow. The aims of this review are to (a) describe the concepts in the model, (b) review the state of knowledge regarding each concept, (c) critique the cited empirical literature, and (d) to identify the gaps in knowledge.

An Overview of the Literature on Family Systems

The integration of family systems theory into the study of children's adaptation to illness has been more recently noted, especially as the family has been incorporated as a variable of interest in the overall well-being of the patient. The shift from an individual to a holistic care paradigm, which includes the patient's family, has come about with the medical community's awareness and recognition of the importance of the patient's social network in health maintenance. The evolution of family systems theory since the 1950's has similarly been recognized and utilized by healthcare theorists and researchers (Cumsille & Eptstein, 1994; Eggenberger & Nelms, 2007; Pederson & Revenson, 2005; Rolland, 1987).

Family systems theory applies general systems theory to families. Family systems theory views the family as a hierarchical self-organized interactive system that regulates itself and its members' behaviors through rules, values, and codes (Cox & Paley, 1997). The importance of the family in adaptation to change was guided by the seminal work of Salvador Minuchin, who advocated a "structural family systems" theoretical framework (Minuchin, 1974). According to this theory, the individual is viewed within the natural social context of the family, which is composed of subsystems that define roles. The family system is altered when internal developmental changes in its members or external stressors necessitate adaptation. Transformation in the family structure occurs when families are confronted with stressors that require the family to adapt to demands placed upon them. In the case of illness in a family member, flexibility in family organization to support the individual and maintain family function is often necessary as children and parents reconfigure roles, communication, and patterns of living to accommodate a stressor (Minuchin, 1974). Adaptation is, then, enhanced by the family system's ability to assemble and activate alternative transactional patterns, or to restructure to maintain ongoing family continuity.

Family system theoretical frameworks have been widely used in studies which have examined the family's response to illness (Faulkner & Davey, 2002; Kotchik et al., 1996; Lewis, Ellison, & Woods, 1985). However, within the body of literature focusing specifically on children's response to parental illness, family systems theoretical frameworks have been less formally recognized. In many studies family system theory has been implied, or when mentioned in the text, it has been understated and under-defined. A review of the literature on children's adjustment to parental illness by

Korneluk and Lee in 1998, cited only three family system models used in these types of studies: Rolland's Family Systems Health Model (1987), Armistead's (and colleagues, 1995) Child Adjustment to Parental Illness Model, and Lewis' (and colleagues, 1993) Family Coping with Maternal Illness Model. Rolland's and Lewis' models are family focused models that include the individual, illness, and family systems variables. In both of these frameworks, illness demands relative to family burden and adaptation are strongly aligned with type of disease and time since diagnosis (Korneluk & Lee, 1998). These two variables have subsequently not been found to be significantly related to children's adaptation to parental illness (See section on demographic variables in this chapter). Although Armistead's model (1995) applies to children specifically, the model is limited in that it focuses on dysfunctional parenting and marital discord associated with parental illness and their impact on child maladaptation and dysfunction.

After 1998, an additional two models specific to children were developed and noted in the later literature. One model, The Family Ecology Framework (Pederson & Revenson, 2005), primarily based on stress and coping theories, incorporated family system variables to explain adolescent adaptation to parental illness. In this model, both individual and family system variables were proposed to mediate parental illness variables and the dependent variable, family and youth functioning. Family and external support systems (school and social support) were similarly combined with Lazarus and Folkman's stress and coping theory in a model for younger school aged children's adjustment to parental cancer (Nelson & While, 2002). These two models are in various stages of psychometric testing.

Overview of Stress and Coping Theory

Much of the early research on adaptation to illness has been guided by Lazarus and Folkman's stress and coping theory (1989). Instruments, models, and intervention tools have largely been based on the constructs of this theory (Compas et al., 2001; Laubmeier & Zakowski, 2004; Lewis, 2007; Weisz, McCabe, & Dennig, 1994; Wonghongkul et al., 2006). However, this discussion will be restricted as the theory pertains to children's stress and coping.

Lazarus (1966) proposed that stress is the result of a transactional process between the person and environment which is affected by cognitive, affective, and coping variables (Lyon, 2000). According to this theory, individual appraisal is an essential ongoing process that serves as a critical mediator of person-environment transactions and is defined as a judgment and inference in which data about an event is assimilated into a constellation of ideas and expectations, causing the person "to evaluate the relevance of the encounter to his or her well-being" (Folkman et al., 1986, p. 992). Appraisal is based on the cognitive assessment of the stressful event, on the assessment of one's abilities to cope, available resources, and on an evaluation of how one has been able to cope with this or similar stressors in the past (Lazarus, 1966). Therefore, appraisal, is dynamic, fluid, and changes during the duration of the stressful event. The significance of the Lazarus and Folkman model is that it is based on both the stimulus event and on the person, and recognizes that psychological factors affecting the person are influential. Although, they cite other authors' work in contending that a person's past experiences, needs, societal and cultural pressures, and conditioning are forces affecting how the

person interprets the stimulus, this theory is primarily one that is based on *individual* stress.

Coping is also viewed as an individual and dynamic process by Lazarus and Folkman (1984). Coping is a voluntary response to an appraisal that motivates an individual to purposeful efforts to manage a stressor or to manage one's emotional response to the stressor. Coping strategies are categorized as problem-focused or emotion-focused by Lazarus and Folkman, and are targeted at stress reduction by diminishing the source of stress or by efforts to manage one's emotional response.

Within the body of literature related to children's adaptation to parental illness, stress and coping theory has been the most dominant (Compas et al., 2001; Lewis, 2007). Lazarus and Folkman's theory has been used primarily to examine how children cope. Compas and colleagues (1994, 1996) used Lazarus and Folkman's stress and coping theory in two early studies of cancer patients, their spouses, and their children to examine the relationships of individual appraisals with the types of coping mechanisms (emotional versus problem-focused) used by family members. Children, adolescents, and young adults (n=134) were categorized by age. Low personal control and high external control related to their parent's cancer were found in all three age groups, and little problem-focused coping was used. Appraisals of higher stress, as defined by Lazarus and Folkman's theory, were related to higher psychosocial distress, which was associated with emotional-focused coping, primarily avoidance.

Terminology, elements, and constructs associated with Lazarus and Folkman's theory have been noted in studies of children responding to parental illness (Barnes et al., 1998; Pakenham & Bursnall, 2006; Pedersen & Revensen, 2005). Conceptualizations of

Lazarus and Folkman's terminology have also been modified for use and for more extensive study in children. Coping, for instance, has been further conceptualized and measured according to many of the refined models that more extensively delineate types of coping (Carver, Scheier, & Weintrub, 1989; Compas et al., 2001; Weisz, McCabe, & Dennig, 1994). In contrast, in a number of studies relative to the coping of children, Lazarus and Folkman's theory has not been formally identified when used, general stress theory has been implied, or no theoretical framework has been mentioned.

The complexity of finding suitable theoretical frameworks that are applicable to children's experience continues to be a recognized deficiency. Because the literature lacks specific models and theories to explain children's perceptions related to potentially threatening events, researchers have used adult models of stress and coping, despite a lack of testing for the adequacy of these frameworks (Berk, 1997; Compas et al., 1988; Ryan-Wenger et al., 1997). Lewis (2007) cautions researchers to consider new and more encompassing theoretical models when examining children's response to parental cancer, implying that stress and coping theory is not sufficient in itself. Application of singular models of stress and coping, like that of Lazarus and Folkman's, does not sufficiently incorporate the family environment, developmental influences, and the contextual social variables which impact children's response.

Literature Related to the Theoretical Model for this Study

The review of the literature will be comprised of research findings which support the four components of the proposed model for this study (See Figure 1, page 21.). The exogenous, Contextual Component, includes pre-existing individual child and ill parent variables. Endogenous evolving variables include the Perceived Impact of Illness

Component, family interaction and family structural change, and the Coping Component includes specific coping strategies. The outcome is indicated by the Adaptation Component which includes emotional and behavioral response.

Literature Review for Child Contextual Factors

Child age and child gender will be examined as child contextual factors. The section concerning child age is composed of a review of the findings which demonstrate children's age-related cognitive processing of illness, and a review of findings which examine children's age-related response to a parent's illness.

Child Age

Cognitive Developmental Processes related to Age/Maturation. Developmental components have been noted to greatly influence the child's perception of the illness experience and thus their response to illness. Experts agree that cognition is a developing and integrative mental process that influences the perception of events and objects in the environment (Keating, 2004). As children develop, they acquire increasingly extensive and sophisticated cognitive abilities to process information more efficiently, to reason abstractly, to think logically, and to perform intellectual functions (Hockenberry, 2005). Children's perception of illness, therefore, is influenced by their individual cognitive abilities and the range of processes which contribute to these abilities.

Piaget's work has been a prominent source theory used to explain developmental differences in how children cognitively perceive and appraise stressors (Jouriles et al., 2000; Lourenco, 2003; Skybo, 2005; Thies & Walsh, 1999). Application of Piaget's developmental theory to children's perception of illness has also guided clinicians in

anticipating children's developmental stressors and conflicts related to parental illness, and in proposing age appropriate interventions (Lewandowski, 1992).

While there is a call for the use of theoretical frameworks to undergird the study of children's cognitive processes in the perception of illness, there is debate regarding the extent to which any known framework may be applied. Many contemporary psychologists are less enthusiastic about promoting Piaget's framework and advocate that it not be applied as the universal template for cognitive and psychosocial development in children (Bjorklund, 2000). Rather, they propose that cognition in children may actually progress more rapidly and less uniformly than once thought and that children are able to understand cause and effect at earlier ages, both which have implications for how children understand and conceptualize situations involving serious illness.

There is an acknowledged lack of studies related to the evaluative processes of children despite recognition that perception and evaluation contribute to psychosocial adaptation to stressors (Hasan & Power, 2004; Schmidt et al., 2003). However, there is general agreement that cognitive abilities associated with age-related maturational development are critically influential in how the child is able to process and construct events.

Studies exploring *age-related* variations in children's knowledge and perception of illness have been helpful in differentiating how children comprehend illness. Kalish (1998) studied 22 preschoolers and primary aged children, ages 3.5 to 6.5 years, and found that children, even at these young ages, were able to perceive illness. Children made definite determinations regarding illness related outcomes (ie. all children in a room exposed to a sick visitor will get sick, without exception) and could make limited

associations about the cause of an illness. Children viewed illness as an event that affected all members of a group without exception and were unable to see variable outcomes that potentially could occur due to illness. While these findings are questionable in view of the small sample size and the range of age of participants in this study, the comparison of the findings of this young age group to the varied and less concrete responses found in older children indicates that developmental changes in cause-effect conceptualization are affected by cognitive maturation.

The ability of children to structure information and major concepts into schema was found to contribute to noted differences in the illness perception of 20 subjects in a study by Hergenrather and Rabinowitz (1991). Twenty children in each of three age groups, 6 - 7 year olds, 9 - 10 year olds, and 13 - 14 year olds, were given three distinct measures to test structural organization of knowledge regarding illness. Three sorting tasks were used to examine whether children used outward behavioral or inward symptoms to determine illness, to examine how children recognize illness, and to determine what participants understood about illness cause. To determine how children differentiate illness from wellness, children were given 21 illness-related pictures depicting cause, treatment, and symptoms and asked to sort the pictures into categories and then explain their rationale for selection. Illness recognition was tested by asking children to respond to 14 multiple choice scenario items that were read to them, and they were then asked to pick the answer that determined how the subject in the scenario knew he/she was sick. Each item used answers related to symptoms and treatment. Twelve multiple choice items, also read to the participant, were used to test the child's understanding of cause of illness. Children were asked to discern the cause of scenario-

based symptoms of particular illnesses as being treatment induced, as a consequence to negative behavior, as having a contagious cause, or as a result of their true cause.

Analysis of the data indicated that there were differences in these age groups: behavioral cues (going to the doctor, staying in bed, having your temperature taken) rather than symptom cues (vomiting, persistent cough) were used more by younger children, ages 6 - 7. Younger children associated illness with misbehavior, such as non-compliance with parental instruction. Middle-aged children, ages 9 - 10, were more likely to associate illness with illness-related concepts, such as single agent contagion, treatment, and symptoms than younger ones. Children in the older age group, 13 - 14, were found to associate the correct cause of illness with the scenario and were more likely to understand illness as related to multiple causes and not related to misbehavior. According to the investigators, older children, ages 13 and 14 years of age, thus organize their understanding of concepts, such as illness, based less on behavioral cues and more on knowledge about the specific illness. These findings offer support to the trajectory of ascending cognitive abilities of children in the accurate and more expanded interpretation of illness, and support the theory that even very young children are able to have a basic understanding of illness. Limitations of the study include the small size of the participant groups, and the relatively moderate to high socioeconomic status of parents of participants from a suburban church and a university faculty.

A later study of 83 well children, ages 4 - 12 years, divided into 4 age groups supported earlier findings regarding the increasing sophistication of children's conceptualization of illness and health with increasing age (Myant & Williams, 2005). Qualitative content analysis of children's responses to illness vignettes was done.

Accuracy and understanding of specific illnesses became increasingly refined as children aged. Although this study found that children in all age groups defined illness more by symptoms than by behaviors, researchers concluded that there were differing levels of understanding for each type of illness. The authors found that children's responses (within and across age groups) regarding recovery strategies, as well as other domains of knowledge about specific illnesses, were more accurate for common everyday illnesses (cold, chicken pox, and toothache) than for a less experienced illness type (asthma). This implies that experiential knowledge impacts understanding, and more importantly, that there may be variations in children's understanding of illness based on context and type of illness. The investigators provided no data as to the exposure of the children to a family member's illness.

There is a lack of research as to when and how children's cognitive maturation enables them to make a judgment regarding the potential threat of illness. Due to the limited research in this area, knowledge gained from other studies that have examined the child's perception of threat within the contexts of parental divorce (Skybo, 2005), violence (Sheets, Sandler, & West, 1996), and bullying (Hunter & Boyle, 2004) have provided findings to support a theory of children's increasing maturational cognitive ability to appraise a situation relative to negative and positive potential outcomes.

One study was found that tested children's cognitive ability to specifically discern threat related to illness (Thies & Walsh, 1999). Seventy-nine chronically ill children subdivided into 3 age groups (8 - 9, 11 - 12, and 14 - 16) were interviewed and asked to describe a situation that they found difficult due to their chronic illness. Children were then given a list of emotion words that might describe their feelings about the situation

and asked to quantify the intensity of that emotion (ie. sad, upset, angry). The children were then asked to explain the rationale for their selection and were also asked to discuss why the recalled situation was important to them. The purpose of this questioning was to understand the child's ability to know the "stakes" considered by the child in their appraisal. The children's responses were scored based on the cognitive processes underlying the children's perceived judgment of the stressor through a scoring system of cognitive appraisal developed by the author (The Developmental Scoring Categories of Cognitive Appraisal). This tool evaluated both "stakes" and "action" according to a hierarchical category system based on increasingly complex levels of understanding that was aligned with Piaget's cognitive development model. Inter-rater reliability was high for both parts of the instrument (.967 for Evaluation of Stakes, .811 for Evaluation of Action). According to the investigators, concrete thinking, as described in Piaget's operational stage, was characterized in the scoring system as a child's appraisal of the stressor in terms of the external interference it causes to the child's wants and desires. Pre-operational thinking about the "stakes" of a situation incorporated the views of others, including others opinions about the situation or view of them. The "stakes" of a situation by an older child in the operational (abstract) stage were scored in terms of the older child's internal understanding and assimilation of the stressor related to him/herself, especially in terms of his/her ability to consider "cause and effect."

From their findings, the investigators concluded that there was a trend from concrete to abstract reasoning in their evaluation of the stakes of the situation (appraisal) as children's development matured. There was likewise a trend from external to internal behavioral response to stress as children aged. Using one-way ANOVA, the investigators

found that all three age subgroups displayed responses consistent to those of Piaget's developmental model; that is, the 8 - 9 year old group had more concrete (external/outside interference) appraisals of illness related stress, the 11 - 12 year olds expressed early abstract cognitive responses that incorporated others' viewpoints and content from a more social context, and the 14 - 16 year olds perceived illness related stressors from a personal context which integrated their own viewpoint within a particular system of which they were a part. The authors emphasized that due to increasing cognitive development, thought processes become more integrated and the older child's ability to view stressors becomes more complex. Varied implications that the stressor has on the individual, others, relationships, and the future become increasingly apparent to the child with cognitive development (Thies & Walsh, 1999).

The child's appraisal of the seriousness of the illness. Two early studies by Compas and colleagues (1996, 1994) found that the only factor that was associated with psychosocial distress in children and adolescents of parents with cancer was children's perception of the parent's illness. More serious and stressful appraisal of the parent's cancer was positively correlated with the child's emotional distress. One hundred thirty four children of a parent with cancer, ages 6 - 32 years of age, from three cancer clinics, who lived with the ill parent or who had frequent contact with the parent during treatment, comprised the study sample. Of these children, 68% were between 6 and 18 years of age. The investigators found that children's perceptions of the seriousness of their parent's illness were generally accurate, because the acuity of their perceptions were positively correlated with the diagnostic disease stage ($p < 0.05$) and prognosis ($p < 0.05$). The investigators broke down the sample by age (preadolescent, adolescent, and

young adults) and found that age was not a significant factor in children's perceptions of seriousness nor in the perceptions of the stressfulness of the parent's cancer. They did not look at the relationship of gender with illness perception. A serious limitation of the study was that the instrument used a single four point Likert item to assess children's perceptions of parental illness and children's perception of the severity of the parent's cancer. This, therefore, may not clearly reflect the consistency in the three groups regarding appraisal nor adequately account for the potential differences in the maturational processes by which cognitive perception is determined.

Age-related findings regarding children's response to parental illness. Very early seminal work laid the groundwork for the exploration of differences in psychosocial distress in children of parents with cancer based on age. Lewis, Ellison, and Woods (1985) in a qualitative study of 126 families interviewed mothers with breast cancer, their partners, and children. No report was given regarding the number of children in the family sample. Differing themes, based on age, were present as children talked about their distress. Reactions of young children (7 - 9 years of age) were characterized by fear regarding the family and the future. Children between ten and 13 years of age were preoccupied with how the illness affected them and their daily lifestyle, and adolescents were distressed by the potential for the changed relationships between them, their parents, and others in the family (Lewis, Ellison, & Woods, 1985). Later studies corroborated and expanded upon these early findings. Erroneous ideas about the cause of illness were found to predispose younger children to magical thinking and magical cures to resolve underlying fearful perceptions (Christ et al., 1993; Lewandowski, 1992). Pre-adolescent

concerns regarding individual needs and changes in activities due to parental illness were found in a study by Armsden and Lewis (1993).

Adolescent children of ill parents have been found to be the most vulnerable age group in regard to problems of distress and adaptation (Armistead, Klein, & Forehand, 1995; Compas et al., 1994; Pedersen & Revenson, 2005). The early study by Compas and colleagues (1994) on children of parents with a variety of types of cancer, including breast cancer (32%), gynecologic cancer (21%), brain malignancies (12%), lung cancer (7%), hematologic malignancies (10%), and gastrointestinal, testicular melanoma, and other malignancies (18%), found that adolescents had significantly higher levels of anxiety and depressive symptoms than children of parent's with cancer ($p < .05$). However, in this same study sample, overall stress-response symptoms, such as avoidance and intrusiveness, as measured by the Impact of Event Scale (Horowitz, Wilner, & Alvarez, 1979), was less in adolescents than in younger children. The investigators theorized that developmental differences or inaccurate portrayal by children of themselves to the interviewers may have biased these findings.

Two other literature reviews examining the impact of parental cancer on children have corroborated findings regarding adolescents' higher incidence of psychosocial distress (Osborn, 2007; Visser et al., 2004). Each reviewer identified various studies (Osborn, $n = 8$; Visser, $n = 13$) in which age was examined for its association with emotional and behavioral problems. Self-report was the primary method used for obtaining data in adolescents; parental report was used for younger children. Both reviewers concluded from their analyses that adolescents tended to report more overall problems and more internalized problems, such as anxiety and depression, than younger

children. Self-reports indicated more problems, manifested by distress, than parent reports. Therefore, one cannot conclude adolescents necessarily experience more distress, as this difference may be a function of the method used for data collection.

Rationale for why adolescents may be at increased risk for psychosocial difficulties has been generally ascribed to their more sophisticated cognitive abilities, emotional sensitivity and empathy, and their internalization of emotion (Compas et al., 1996; Lewis, 1996). Adolescent developmental activities, including parent-child arguments and the need to seek out information, although found to be normal or generally helpful behaviors, were conversely distressing and anxiety-provoking when the adolescent was reluctant to engage in these activities or when obtained information was unclear or misinterpreted (Adams-Greenly et al., 1986; Lewis & Hammond, 1996). Grabiak and colleagues (2007) later reported in an analysis of the literature that adolescents' perceptions of their parent's cancer was additionally complicated by the lack of information adolescents received about the illness from both family and external supports. More recent research has demonstrated that other external environmental factors may be significant variables contributing to adolescents' vulnerability to distress (Huizinga et al., 2003; Watson et al., 2006). This will be further addressed in later sections of this literature review.

Child Gender

Up to this point, findings regarding child gender have suggested that girls, especially adolescent girls, may be most impacted by their parent's illness. Adolescent girls were found to be the most emotionally affected group especially when their mother was the ill parent (Compas et al., 1994; Grant & Compas, 1995; Howes et al., 1994;

Visser, 2005). These researchers theorized that adolescent girls may be the most vulnerable due to increased household responsibilities, gender identification with the affected parent (mother), and their heightened rumination may potentially exacerbate increased distress (Grant & Compas, 1995).

A pilot qualitative study of adolescent girls (n = 3, ages 17 - 21 years), using thematic analysis, reported that girls perceived their parent's cancer as a family event (Sears & Sheppard, 2004). The girls' priority became ensuring family function and individual family member's needs, including that of the ill parent. This perception influenced the pursuit and accomplishment of their own interests and activities, and was found to cause both conflict and personal satisfaction. The study used a volunteer sample, and it was limited by its small sample size and retrospective reports, but was helpful in pinpointing perceptions that quantitative measures may not access.

Gender was a significant variable in a study examining psychosocial distress and adolescent perception of the seriousness of their parent's cancer (Huizinga et al., 2005). Correlation analysis was used to evaluate adolescents' (n = 284, ages 11 - 23) perception of the seriousness of illness with self-reported stress response, as measured by the Impact of Event Scale (Horowitz, Wilner, & Alvarez, 1979) to determine heightened clinical levels of distress. Although the study sample was large, 64% of the parents were diagnosed with maternal gender-related cancer; the rest of the sample varied in type of malignancy. Daughters' reports of more serious perceptions of the parent's illness were correlated with reported higher levels of internal distress. Heightened levels of intrusion (r = .18, p = .024), avoidance (r = .22, p = .006) and total distress (r = .022, p = .006) were noted. Adolescent boys' perceptions of parental illness were not significantly

correlated with these same variables. The investigators only correlated perception with items on the IES Scale to assess post-traumatic symptoms at a clinical level, and did not correlate perception with emotional and behavioral functioning (reported on the Youth Self-Report). Analysis of this association would have added to the interpretation of the findings.

One study by Visser and colleagues (2005) found that in addition to adolescent children of parents with cancer, younger sons, ages 4 - 11 years, had more problems that were in the clinical range than a norm group as reported by parents. Ill parents reported significantly more internalizing problems, which include withdrawal, anxiety, and depression, in younger sons than in adolescent sons ($p = .023$). No rationale for these findings were discussed by the investigators, however, further study of boys in this age group may be warranted.

It is significant that a majority of studies have found that girls, especially adolescent girls, are the most vulnerable group of children of cancer patients with regard to distress (Nelson & While, 2002; Compas et al., 1994; Grant & Compas, 1995; Huizinga et al., 2004; Welch, Wadsworth, & Compas, 1996). Because a large number of studies have been conducted with children of mothers with breast cancer, this finding may be over-represented when generalized to children of parents with all types of cancer.

Literature Review for Parent Contextual Variables

Parent's Diagnosis and Prognosis

According to Pederson and Revenson (2005) the literature has primarily viewed parental illness in terms of *diagnostic type* or *severity*. Diagnostic type is based on medical labels associated with the illness and does not take into account the variations

that can exist between patients with the same diagnosis. Similarly, severity of illness is often based on characteristics of the illness which are not static, and which are contextually based; that is, severity of the illness can change, and the modifications caused by the illness may be quite different from family to family, and different over the course of time. Although, Rolland's model of family adaptation to illness (1987, 1994), based on descriptors to qualify illness (onset, course, outcome, degree of incapacitation, and predictability) takes into account the multidimensionality of illness, these elements have rarely been applied in totality to the examination of illness in regard to the psychosocial function of families and individuals. The abundance of cross-sectional, rather than longitudinal designs, has also limited the findings regarding the impact of the transient nature of illness related variables on the family unit and on individual family members' psychosocial functioning.

Findings relatively early in this body of research demonstrated that the parent's specific diagnosis was not a primary factor in child response to parental illness. Armistead, Klein and Forehand (1995) reviewed the literature through 1994 (n = 12), composed of six studies using controls and six studies using a within group design. The purpose of this review was two-fold: to determine if parental illness was associated with child functioning (between groups design), and to examine factors which may affect children's function (within groups design). Analysis of the six studies that used a within groups design to examine factors related to child psychosocial function revealed that dimensions of the parent's illness, as defined by Rolland (1987), were associated with children's distress. The dimensions include onset, course, outcome, and degree of incapacitation. Therefore, the reviewers concluded that the integration of various illness

dimensions could cause variations in child and family psychosocial response to a particular illness, and that even further variations could occur as these were combined with other child and parent factors. Based on the studies they examined, the reviewers specifically cited disrupted parenting resulting from parental illness as a key factor in children's psychosocial distress. A limitation of this review is that it was composed of a small number of studies, and that these findings may have been used to support the development of the authors' theoretical model.

Subsequent studies (Compas et al., 1996; Hoke, 2001; Watson et al., 2006) have also found that diagnostic labels associated with particular illnesses, such as stage of the disease and time since diagnosis, have not been significantly associated with children's psychosocial function. Based on the research findings of Armistead and colleagues (1995), later researchers have examined the association of the parent's illness characteristics with family function. They have concluded that illness characteristics mediated family relationships, rather than directly affecting child emotional health and adjustment (Brandt & Weinert, 1998; Huizinga et al., 2005) which appeared to place the child at higher risk.

An early small study did not find parent's illness prognosis to be a significant factor in 11 - 16 year old children's psychosocial function (Nelson et al., 1994); however, this finding is questionable in that the investigators did not discuss how illness prognosis was determined. Poorer patient prognosis was found to be associated with higher levels of self-reported distress in adolescents in a study by Compas and colleagues (1994, 1996). In this study, children's perception and appraisal of the parent's illness, rather than the objective diagnosis and prognosis of the disease, mediated their distress. In

subsequent studies, disease prognosis and type of cancer have consistently not been found to be significant in children's psychosocial adjustment and adaptation (Hoke, 2001; Howes et al., 1994; Watson, 2006).

Other illness related variables have been studied. Time since diagnosis of the parent's cancer has consistently not been associated with children's psychosocial functioning (Lewis et al., 1993; Huizinga et al., 2005; Visser et al., 2005; Watson et al., 2006). Treatment variables, specifically chemotherapy compared to surgical and radiation therapies, have generally not been found to be associated with children's psychosocial function (Hoke, 2001; Visser et al., 2005; Watson et al., 2006). However, recurrence of parental cancer was found to be associated with increased self-reported psychosocial distress in both adolescent boys and girls as reported by Huizinga (et al., 2005) and Visser (et al., 2005). When parental cancer recurred, higher levels of internalizing problems, as measured by the Child Behavior Checklist, were reported by adolescent boys, and higher levels of both internalizing problems and the total number of problems were reported by adolescent girls.

Parent Symptom Interference

According to Given and colleagues (1993) a patient's psychological distress is affected more by the patient's symptoms than by objective categorization of disease severity. Similarly, children's observations of their parent's symptoms and symptom-related behaviors were found to be influential in how the child perceives and interprets the severity, extent, and resolution of the parent's illness in early investigations (Compas et al., 1994; Christ et al., 1993). However, direct associations between parent symptom/

illness complications have not been consistently associated with children's psychosocial adaptation.

An early qualitative study of 87 children, ages 7 - 11 years, found that children were especially distraught when visible symptoms such as alopecia, vomiting, and weight loss occurred in the parent (Christ et al., 1993). Increased anxiety and fear was found in these children. Children were found to associate themselves with the cause of the parent's illness. Adolescents (n = 120) were found to make attempts at either advocating for the ill parent when symptoms were noted or detaching to avoid witnessing their parent's distress. The study was limited in that participants were dealing with parental terminal illness, which may weaken its generalizability to other children whose parents may have less apparent life-threatening malignancies.

Severity of side effects, such as nausea, diarrhea, hair loss, fatigue, and hospitalization for treatment related infection and bleeding were not found to be significant in a study of children (n = 35, ages 8 - 16 years) of mothers with breast cancer (Hoke, 2001). A comparison group of children whose mothers had benign breast biopsies was used. Both parental and child reports were obtained. An earlier study of children (n = 32, ages 3 - 18 years) of mothers with breast cancer reported contradictory findings (Howes et al., 1994). Serious complications related to the mother's treatment and illness accounted for less parent-reported child psychosocial behavioral distress. Further analysis of this study indicated that family adaptability as measured on the FACES III (Family Adaptability and Cohesion) scores correlated significantly with the presence of serious complications ($r = .47$, $p = .01$). In regression analysis, the relationship between serious complications and psychosocial function were sustained, even when maternal distress

was held constant. The authors concluded that family adaptability may mediate the relationship between a parent's serious complications and a lower level of psychosocial distress in the child; that is, adaptable families may achieve higher levels of flexibility and mobilization in meeting the demands caused by the complications. This study is significant in providing preliminary information regarding family variables that may be protective of children's psychosocial stability. Both studies were limited by small sample sizes, composed almost entirely of Caucasian subjects, and they utilized cross-sectional designs which limited generalizability across time.

Symptoms of emotional distress in the parent may also be indirectly associated with child adaptation. A study by Watson and colleagues (2006) examining various family variables in the adaptation of children of breast cancer patients found that maternal depression was a significant predictor in child psychosocial distress. The Beck Depression Inventory-II (Beck et al., 1996) was used to assess the maternal depressive state, the Child Behavior Checklist was used to evaluate children's psychosocial function, and the Child Health Questionnaire was used to measure child distress. In univariate analysis, higher maternal depression scores significantly predicted ($p = .0001$) internalizing problems, which include social withdrawal, somatic complaints, anxiety, and depression. Higher maternal depression significantly predicted total CBCL problems ($p=0.003$) in children, and was also found to be a significant predictor of heightened levels of distress warranting clinical intervention in children as measured by the CHQ ($p < .001$). In multivariate analysis of parental and family variables, maternal depression was the most significant factor in children's internalizing problems ($p = .002$), and the strongest predictor of mental health distress as measured by the CHQ ($p < .001$). These

findings were based on self-reports by both the mothers and their children which augmented the reliability of the findings. Depression in the well father was not found to be associated with psychosocial problems in the study sample. The sample size was substantial (n = 107 families). Only one child per family was targeted to participate to avoid duplication of results. The sample group was 83% Caucasian, and time since parent diagnosis was variable, from three to 36 months. A limitation of the study was its cross-sectional design, and the fact that only 56 (57%) of the adolescents were compliant in completing their self-reported questionnaires.

Additionally, a number of other studies have similarly reported an association between parents' manifestations of emotional distress and children's psychosocial problems. Three studies reported a positive association between self-reported distress in ill mothers (physical and emotional distress) and increased psychosocial problems in their children (Hoke, 2001; Howes et al., 1994; Lewis & Darby, 2003). Positive associations between self-reported adolescent psychosocial distress with poor parental coping style (Nelson & While, 2002) and parental depression (Watson et al., 2006) have added to the literature findings. Within the context of family systems' theory, it is assumed that the parents' perceptions and resulting responses influence the perception and responses of their children. The literature suggests that parental psychosocial variables may be potentially significant in how the child views the impact of the illness, and may therefore warrant further study.

Parent Gender

The influence of the ill parent's gender has been an inconsistent variable affecting child psychosocial adaptation. In two separate studies by Nelson and colleagues (1994;

2002) no significant differences were noted in children's psychosocial distress related to the gender of the parent who had cancer. A recent study by Visser and colleagues (2005) found that significantly more psychosocial problems were self-reported by adolescents and by spouses of the ill parent when the father was ill with cancer than when the mother was ill. Younger children, 4 - 11 years of age, were also reported to have more psychosocial problems by the parents when the father had cancer. Although these findings were based on a relatively large sample of 180 families comprised of 336 children, 114 younger children and 222 adolescents, 11 - 18 years of age, only 19% of these families had a father with cancer limiting the generalizability of these results.

There are some findings in the literature which suggest that heightened psychosocial distress may occur when the gender of the parent is the same as that of the child. Three early studies of mothers with cancer reported that daughters, when compared to sons, were noted to self-report the highest levels of emotional distress (Compas et al., 1994; Grant & Compas, 1995; Welch, Wadsworth, & Compas, 1996); although it is unclear if these findings were all derived from the same sample. In these studies, adolescents were more distressed when the illness affected the same gendered parent, that is, adolescent sons of ill fathers and adolescent daughters of ill mothers were found to have more stress response symptoms when compared to adolescent daughters of ill fathers and adolescent sons of ill mothers. This finding was not confirmed in a later study of 81 adolescents, ages 11 - 18 years of age (Rodrigue & Houck, 2001). No significant interaction between gender of the child and gender of the ill parent was noted.

Parent Hospitalization

No studies were found in the literature that examined the association of the ill parent's length of stay in the hospital with children's adaptation. Several qualitative studies have reported that avoidance, checking on the parent, and increased arguments and disagreements with other family members occurred especially when the parent was hospitalized (Christ et al., 1993; Issel, Ersek, & Lewis, 1990; Zahlis & Lewis, 1998).

Literature Review for Children's Perception of the Illness Impact

Serious parental illness is often particularly disruptive and presents numerous challenges to families. Factors associated with the parent's cancer, including the severity of the illness and the impact of the illness on the family, are interpreted by the child in day to day family interactions and in the overall climate of the family (Siegel et al., 1992; Howes et al., 1994). Findings from early studies provided a foundation for the bi-directional relationship between the individual and the family unit in adapting to parental illness. Family members' advocacy for one another (Lewis et al., 1985) and the adaptability of the family unit to illness-related changes were associated with individual member's perception of the illness and coping (Siegel, 1992; Howes et al., 1994). Characteristics of the family unit have been increasingly recognized as an influential contextual variable that profoundly influences the adaptation of individual family members (Lewis, 1996; Lewis & Hammond, 1996; Watson et al., 2006).

In the proposed model, perception of the illness impact is influenced by the family system variables, family interaction and family structural change. A review of the literature on these constructs, as defined in the model, follows.

Family Interaction

The interactions of individual members and their collective relationships to one another and to the family unit are influenced in significant ways by transitions and events that occur with each member. Various dimensions which contribute to the overall environment of a family have been recognized (Moos & Moos, 2002). This study will focus on family interaction which contributes to the overall family environment. Family interaction will be operationalized as family cohesion, family conflict, and family expressiveness. Literature that has focused on these three concepts will be examined in this section to provide a basis for the examination of this variable in the proposed study.

Family Cohesion

Within the context of children's psychosocial function, family cohesion has been associated with children's behavior at all stages of child development (Richmond & Stocker, 2006) and with adolescent depression (Cumsille & Epstein, 1994). Family cohesion has been found to be a buffer in child psychosocial distress in stressful family circumstances including divorce, socioeconomic hardship, and homosexuality (Dreman & Ronen-Eliav, 1997; Vandewater & Lansford, 2005; Willoughby, Malik, & Lindahl, 2006). Adaptation of both the family unit and children to a family member's illness has similarly been reported to be associated with family cohesion. Studies of family response to children's serious illness have shown that family cohesion was positively associated with adaptation (Horowitz & Kazak, 1990; Kazak & Barakat, 1997). In a study of children (n=41) during *pediatric* bone marrow transplantation, family cohesion was significantly related to improved resiliency during the acute phase of the transplant and positive adaptive outcomes in the ill child (Phipps & Mulhern, 1995). Investigators

studying a variety of types of parental illnesses have explored the association of family cohesion with stress and coping, and conversely explored the impact of parental illness as a strain and stressor on family cohesion (Dura & Beck, 1988; Kliewer & Lewis, 1998). Family cohesion has generally been found to be negatively associated with child distress and to be a protective factor in child adaptation to the parent's illness.

A Dutch pilot study of 15 children by Huizinga and colleagues (2003) explored family cohesion and adaptability in 7-18 year old children of parents with cancer. Semi-structured taped interviews and standardized questionnaires were used to gather data from both parents and from children. The Child Behavior Checklist (CBCL, Achenbach & Edelbrock, 1991) which measures emotional and behavioral problems along with child competency in social, school, and other activities was completed by parents. Additionally, children over 11 years of age completed the Youth Self-Report, a parallel instrument using child self-report. Semi-structured interviews, one with the ill parent and the partner, and the second with the children were conducted to collect information relevant to the family. The cohesion and adaptability subscales of the Dutch version (GDS) of the Family Adaptation and Cohesion Evaluation Scale (FACES) by Olson (et al., 1985) were used to assess these family variables. The reliability of this scale is high (Cronbach alpha = .87). Scores on the cohesion subscale ranged from low to extremely high. Low scores reflected disengaged families, moderate scores were categorized as separated families, and high scores reflected connected families. Extremely high levels of family cohesion represented enmeshed families, characterized by emotional connection or separation of family members with one another. Families with extremely high levels of cohesion were considered dysfunctional. The ill and well parents and seven children, 11

years and older, completed this questionnaire. The results indicated that extremely low family cohesion, or disengagement, was associated with increased prevalence of internalizing problems in children as reported by their parents ($r = .65$, $p = .031$). Other categories of assessed family cohesion were not significantly correlated with children's emotional or behavioral problems. Several limitations are noted in this study. The small sample size, and therefore the study's low statistical power, hinders drawing conclusions. Second, cultural and social differences in Dutch children regarding their perception of family cohesion may affect the generalizability of the findings. Third, this study was conducted well after the parent's cancer therapy was completed, two to 52 months after treatment, when most of the parents were considered to be without disease. The potential effects of history and maturation errors affecting internal validity should be considered when examining the findings, especially in regard to the study's generalizability to other populations at different points in the cancer illness trajectory. The study is significant in that qualitative interviews provide an enhanced overall understanding of family environmental influences that affect children's emotional and behavioral distress, and additionally provide information as to when children may be more vulnerable. Information obtained during the qualitative parent and child interviews indicated that parents retrospectively remembered observing more emotional and behavioral changes in their children during the acute phase of the illness, shortly after diagnosis. These changes included heightened anxiety regarding the fear of losing their parent, withdrawal, and behavior changes that primarily normalized within a few months to six months after diagnosis.

A larger British cross-sectional study found that family cohesion was a predictor of emotional and behavioral problems in children of mothers with breast cancer (Watson et al., 2006). Forty one children ages 6 - 10 years of age and 63 children ages 11 - 17 and their parents and a school teacher comprised the sample. Measurement of child emotional and behavioral health was assessed by parents using the Child Behavior Checklist for younger children and the Youth Self-Report form (children 11 - 17) and the Teacher Report form. The CBCL forms separate child problems into 3 categories: Internalizing problems, externalizing problems, which include aggressive behaviors, and total problems. The Child Health Questionnaire (CF-87) and the Parent Form (PF-98) were also used to measure child distress. The family cohesion subscale of the Family Environment Scale was used to assess family cohesion by both the parents (Cronbach alpha = .712) and the children ages 11-17 (Cronbach alpha = .769), therefore, only measurement of family cohesion by this age group was used in the analysis. Due to low recruitment and compliance rates, data from breast cancer partners and from teachers were not analyzed. The investigators found that lower family cohesion as reported by mothers ($p = .001$) and by children ($p = .01$) were associated with higher reports of externalizing problems. When multivariate analysis was done on significant variables, as reported by the mother, lower family cohesion was found to predict both internalizing ($p = .001$) and externalizing problems ($p = .001$) by children. The investigators concluded that child age, parent treatment, and time since diagnosis were not significant factors associated with psychosocial problems in children. This study adds to the findings regarding the association of family cohesion to child emotional and behavioral problems because of the consistency of the findings *between the mothers and children*. Further, it

encourages future studies to explore family cohesion in patients and children, and to explore the impact of this variable on emotional and behavioral problems in children ages 11 - 17.

A comparative study of children of cancer patients corroborated findings that lower levels of family cohesion were associated with increased adolescent psychosocial distress, and further clarified the significance of cohesion when other family variables were examined (Harris & Zakowski, 2003). Twenty seven adolescents of a parent with cancer, from 22 family units, were compared with a matched group of 23 adolescents of parents without cancer in this cross-sectional study. Distress was defined as depression, anxiety, degree of subjective impact as a result of the illness, and PTSD-like symptoms. The influence of family environment variables on adolescent distress was tested using 3 subscales (cohesion, expressiveness, and conflict) of the Family Environment Scale (Moos & Moos, 2002). These three subscales combined have been formally referred to as the Family Relationship Index. Unlike previous experimental studies examining distress in children of cancer patients (Capelli et al., 2005; Compas et al., 1996; Heiney et al., 1997; Visser et al., 2005) there were no significant differences found between the healthy parent adolescent group and the ill parent adolescent group in anxiety or depression using T-test analysis. Significant correlations between total family environment scores, as measured by the three subscales of the FES, indicated that a positive family environment was associated with decreased adolescent depressive symptoms and lower anxiety. This analysis was done only on the group of adolescents of cancer patients. Individual correlations on the three subscales with depressive symptoms indicated that family cohesion was significantly inversely correlated with adolescent depression ($r = -.47$,

$p < .01$), and was significantly inversely correlated with adolescent anxiety ($r = -.32$, $p < .03$). Because the investigators hypothesized that expressive communication mediated family cohesion and adolescent anxiety, multiple regression analysis was performed on the two combined groups. When expressiveness was controlled, there was no longer a significant relationship between family cohesion and adolescent anxiety. Expressiveness was therefore considered a mediator in this relationship. Harris and Zakowski did not analyze the role of expressiveness in the relationship of family cohesion and adolescent depression. This study was significant in that a comparative control group design was employed unlike many studies conducted up until that time. However, it is difficult to draw conclusions related to differences in the distress levels of the two groups, those with ill parents and those whose parents were well, because the sample size was small and the comparison group with well parents was asked to relate the questions to a generic stressor in their lives. The study was limited in its generalizability as the participants were primarily Caucasian. Second, the participants were not studied until a year or more after their parent's cancer diagnosis, which may have obscured rating the more profound impact of the illness on the child's psychological distress during the onset of the parent's cancer.

Not all studies, however, have found a significant association between child adaptation and family cohesion. Nelson and While (2002), examining a variety of factors that were thought to potentially impact child adjustment and adaptation, found that family cohesion was not significant. The study was conducted one year after a parent's diagnosis of cancer. Eighty British children, ages 8 - 16 years, from five major cancer centers participated. Mixed methods were used. The investigators used the expressiveness and

cohesion subscales of the Family Environment Scale (Moos & Moos, 2002) to look at the association between these variables and child behavioral adjustment. Child behavioral adjustment was reported by the mother's and the teacher's completion of companion forms of the Rutter Scale (Rutter, Tizard, & Whitmore, 1970), and with self-reported child anxiety as measured by the Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1997). Reliability for the RCMAS was high (Cronbach alpha = .89), however, there was no reported reliability for the Rutter Scale. This study was critically limited in that the cohesion and expressiveness subscale data came from the parents and not the children, thus the child's perception of these two variables was not taken into account. The sample was relatively small; it was comprised primarily of children of breast cancer mothers (63%), and did not provide clear results of the analysis by age categories.

Researchers examining adaptation must clearly differentiate between the quality of individual family relationships and cohesion within the family as a unit, and recognize the association of these two variables. According to Richmond and Stocker (2006), family cohesion is a variable independent of individual dyadic relationships within the family; that is, it contributes its own variance after controlling for dyadic relationships. In a recent study examining associations between family cohesion and adolescent siblings' behavior, family cohesion was a separate variable positively associated with adolescent behavior, although the investigators theorized that stressful family dyadic relationships may disrupt family functioning and deter family cohesion. Within this context, a brief overview of the literature pertaining to research findings on family dyadic relationships as associated with child adaptation will be described.

Dyadic family relationships (marital, child and ill parent, child and well parent) have similarly been explored in the literature as single individual variables impacting child psychosocial adaptation to parental illness. Mixed findings on the effect of marital adjustment and marital satisfaction have been associated with the overall function of families and children adapting to cancer (Lewis & Darby, 2003; Lewis, Hammond, & Woods, 1993; Phipps & Mulhern, 1995). More consistent evidence in both cancer and non-cancer populations has indicated a positive association between the quality of the parent/child relationship and child adaptation (Lewis, 1996; Woods & Lewis, 1995; Steele et al., 1997). Similarly, the pre-diagnosis relationship of children and their parents was found to be associated with the adaptation of children to terminal parental cancer by Christ and colleagues (1993, 1994). However, family member's dyadic interpersonal relationships have not been studied for their association with family cohesion within the context of parental illness.

A cross-sectional secondary descriptive analysis of 174 families of parents with multiple sclerosis was instrumental in dissecting the association of family relational patterns to child adaptation (Brandt & Weinert, 1998). The study sample consisted of 96 boys and 78 girls ranging from 7 - 17 years of age, no further demographic data were provided. The investigators examined a wide range of independent variables categorized as parental, illness, and family for their contribution to child function and adaptation. The Child Behavior Checklist (Achenbach & Edelbrock, 1991) was used to measure child psychosocial stability and function. Family adaptability and cohesion were examined using the FACES II Scale (Olsen, 1985). Family cohesion was defined as relational sharing and closeness; family adaptability was defined as the flexibility of the family to

alter relationships to changes in the family environment. Discriminant analyses were used to identify factors that delineated two groups of children, those at risk for psychosocial problems and those not at risk. The ill parent's functional status, or physical disability, and illness related behaviors, or daily psychosocial functioning, *were not found* to be significant. More importantly, factors that were found to correlate most significantly were financial status and family relational patterns, specifically the ability to alter family relationships due to external pressure and the level of marital agreement. In the first step of the discriminant analysis, family cohesion was significantly correlated with child adaptation, however, it was not significant in the second step. In their analysis, Brandt and Weinert concluded that parent illness characteristics, including disability and psychosocial functioning, may mediate family relationships, rather than directly affect child psychosocial function and adaptation. Family adaptability was not tested for its association with family cohesion, but it seems likely that the two factors are related. Limitations of the study included the use of only one instrument, The Child Behavior Checklist, to measure the dependent variable. Second, data on child behavior was reported by the non-ill parent and did not include the child's own self-report. The study was also limited by the lack of availability of child data which would have significantly contributed to the overall findings, and by the predictive inability of the design to accurately identify at-risk children from the limited significant discriminator variables that were found.

Within the context of parental illness and child risk for maladaptive adaptation, further studies are needed to explore and perhaps confirm the relationship of parental illness factors to dyadic relational patterns, and how this impacts family cohesion. These

studies provide a basis for further study to formulate an explanatory model for the association of dyadic relationship variables with family cohesion, and to reinforce the theory that various dyadic relationships among family members affect the family's systemic cohesion, function, and potential adaptation.

Family Conflict

Little has been written on family conflict as an independent variable associated with adaptation to illness. Rather, conflict has been viewed along with family cohesion and family communication within the dimensions of family relationships that impact distress during illness. Mixed findings related to the prevalence of conflict in families with an ill parent were noted in early studies (Hirsch et al., 1985; Peters & Esses, 1985), and provided a basis for further study of this variable.

Harris and Zakowski (2003) used the Family Environment Scale (Moos & Moos, 2002) to examine family variables associated with distress of adolescents of parents with cancer (See above description of study in the Family Cohesion section). In their study, cohesion, expressiveness and conflict were correlated individually with anxiety and depression in adolescents, and then combined and correlated with adolescent anxiety (Revised Child Manifest Anxiety Scale, 1985) and depression (Child Depression Index, 1992) scores. Family conflict scores were significantly positively correlated with adolescent depression scores ($r = .48, p < .01$). Conflict was not correlated significantly with adolescent anxiety ($r = .25, p = .09$). When cohesion, expressiveness, and conflict were combined as a total score to measure family relationships within the context of the family environment, (as suggested by the authors of the FES), there was a significant correlation of the total score with both adolescent depression scores ($r = -.59, p < .05$) and

with adolescent anxiety scores ($r = -0.54, p < .05$). Limitations of the study included its relatively small sample size and one year post-diagnosis data collection. The study provided preliminary findings on family conflict as it relates to adolescent distress, and adds to the findings of the importance of the family environment when confronting and adapting to illness.

Family Expressiveness

Family expressiveness (communication) has been found to be one of the single most validated family-based predictors of children's response and adaptation to parental cancer (Osborn, 2007). However, how this variable functions within the array of other family variables (ie. cohesion, conflict, organization, etc.) has been far less studied. The importance of further study regarding expressiveness/communication is crucial because it is a potentially viable area where relevant clinical intervention could be targeted.

Parental introspection, described as communication reflecting on the family's management of problems or challenges during parental cancer, was one dimension of family expressiveness studied in an early study by Lewis and colleagues (1993). Eighty young families of breast cancer mothers, with children between the ages of 6 and 12 years of age, were studied in an effort to construct a model of family functioning. The shared reflections of family members on the daily reorganization of tasks, routines, and activities and on their reevaluation of goals were explored for their relationship to the quality of psychosocial functioning in the child and in the ongoing function of the household. The introspection subscale of an adapted version of F-COPES Scale (McCubbin et al., 1982) was used to measure these behaviors. Child psychosocial functioning was measured by the peer relations subscale of the (FPRQ) Family Peer

Relationship Questionnaire (Ellison, 1983). Other scales were used to assess family functioning (FACES II total score) and to measure interaction between family members (togetherness subscale of FPRQ). Scales were completed by the mother and her partner. Path analysis was used to examine relationships among the variables. Results indicated that increased family reflective communication and interaction regarding management of the illness within the family significantly predicted adaptive child psychosocial behavior, better household functioning, and a higher level of quality of exchange between the non-ill parent and the child. The investigators proposed that parental reflection and feedback regarding daily activities, changes in family roles, and problem-solving about the illness would reinforce the ongoing function and coping of the family, which in turn, would stabilize the child. Although the study was limited by its sample characteristics with high socioeconomic standing, early stage cancer, and children in the primary and middle school age group, it provided a foundation for the exploration of relationships among family variables and their impact on child psychosocial response.

Spira and Kenmore (2000) in a qualitative study found that the use of communication by adolescent girls of mothers with breast cancer was an important strategy in decreasing adolescents' anxiety. The girls reported that communication was helpful in dissuading their fears and in promoting better understanding of the illness. Connectedness to their mother was also a benefit of open communication reported in the participant interviews. Girls said they felt betrayed and isolated when parents did not talk about the mother's illness, and conveyed that conversation about the illness encouraged them to seek out information which was helpful in coping.

Expressiveness was also found to be significantly correlated with adolescent distress in the study by Harris and Zakowski (2003). (See above section on Family Cohesion for more detailed information about this study). In this study, expressiveness was inversely correlated with depression ($r = -.29, p = .05$) and with anxiety ($r = -.35, p = .01$). As previously stated, the researchers found statistical evidence to support their hypothesis that expressiveness mediated the relationship of family cohesion and adolescent anxiety, and concluded that adolescents who can express their thoughts and feelings in the family may have less distress. The authors proposed that expressiveness may stimulate cognitive processing of stressors that may in turn support the psychological well-being of both adolescents of parents with cancer, as well as adolescents who face other stressors.

A study of children of breast cancer mothers found an association between family communication and affective responsiveness, defined as the ability of family members to express affection to one another, with adolescents' reports of externalizing problems (Watson et al., 2006). The Child Behavior Checklist (Achenbach, 1991) was used to assess the outcome variables of emotional and behavioral problems; the McMaster Family Assessment Device (Epstein et al., 1983) was the primary instrument used to assess family function variables. Cronbach alphas for the FAD were reported to range from .616 to .890. Using univariate regression analysis, lower levels of communication ($p = .001$) and lower levels of affective responsiveness ($p = .007$) were associated with increased externalizing of behavioral problems by 57 adolescent reports (Watson et al., 2006). Lower communication levels were also associated with total problems on the CBCL ($p = .007$). Family communication and affective responsiveness variables were not

significant when mothers were the informants ($n = 105$), however, there was overlap between mothers and adolescents in other family variables examined. A limitation to the study's generalizability is that its population was composed of breast cancer patients in the early stage of the disease.

A few studies have focused specifically on parent-child communication and expressiveness as it affects child psychosocial function, and these have primarily been done with adolescent populations. Houck and colleagues (2007) examined the relationship of parent-adolescent communication and adolescent psychosocial performance in a study of 38 teens, ages 12 - 17, of parents with chronic life-threatening diseases (cancer, liver, kidney and heart disease). The Parent-Adolescent Communication Scale (Olson, 1985) composed of two subscales, "openness" and "problems", were used to assess quality and satisfaction of communication as well as adolescent-parent communication. Several scales were used to assess adolescent psychosocial distress including the Child Behavior Checklist (Achenbach, 1991), the Reynolds Adolescent Depression Scale (Reynolds, 1987), the Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1997), and the Impact of Event Scale (Horowitz, Wilner, & Alvarez, 1979). The investigators found adolescents who reported higher levels of the quality of communication with their well parent had significantly decreased anxiety, behavioral, and posttraumatic stress symptoms. Based on a post-hoc analysis of data by gender, problematic communication between the adolescent and the father, whether he was ill or healthy, was associated with adolescent symptoms of depression. The study was limited by its small sample size, and by the fact that only adolescent reports of perceived communication were obtained.

Parent-adolescent communication in families of cancer patients was the focus of a recent cross-sectional study by Huizinga and colleagues (2005) which examined the quality of communication patterns for their association with adolescent anxiety and stress response symptoms. Two hundred twelve adolescents (11 - 18 years of age, mean age 15.1 years) completed a self-report questionnaire, the Parent-Adolescent Communication Scale (See Houck study above). Two subscales of the Impact of Event Scale, intrusion and avoidance (Horowitz, Wilner, & Alvarez, 1979), were used to measure psychosocial distress in adolescents. Adolescents of mothers with cancer were found to communicate less openly when compared to a control group of adolescents with non-ill mothers; no differences were noted in the quality of communication between the adolescent group with ill fathers and the group with non-ill fathers. Daughters who reported more open communication with the non-ill parent had fewer stress response symptoms. Both sons and daughters who reported more problems in communication with the ill or non-ill parent had more stress response symptoms. Females were primarily affected by intrusion and avoidance symptoms. Intrusive thoughts, feelings, and behavior were predicted most by problem communication with the healthy parent; avoidance was predicted most by problem communication with the ill parent. Investigators concluded that problem communication between adolescents and parents had a more negative effect than lack of open communication with respect to avoidance and intrusion symptoms. Because the study was cross-sectional, analysis of changes in parent-adolescent communication patterns and their effect over time were not obtained. The use of a control group of adolescents with healthy parents and the use of reference norms for comparison of communication differences was helpful in determining the extent to which children of

parental cancer patients differed. The findings were limited in that only two dimensions of adolescent distress were measured, avoidance and intrusion, and these were used by the investigators to reflect post-traumatic stress syndrome. Findings, therefore, may underestimate the effects of parent-adolescent communication on more global symptoms of children's distress.

Family Structural Change

Family structural change refers to changes within the family that occur as a result of the parent's illness. Illness and treatment can interfere with family member's performance of their customary roles, which ultimately affects the function of the family system (Eilers, 1997; Rolland, 1994). The ability of the family system to be flexible, to alter patterns of family functioning, and adapt to changes resulting from the parent's illness impacts each member, including the child. In the following section, literature examining changes in the roles and tasks of family members and changes in routines and activities in the family due to parental illness will be examined for their association with child adaptation.

Changes in Family Roles and Tasks of Family Members

Early investigations examining the effects of serious illness on the family identified role restructuring as a stressor to the family (Anthony, 1970; Olsen, 1970). When family members have to take on new or additional duties and assume roles of the ill family member during his/her absence and treatment, family conflict may result. In an early study examining the impact of breast cancer on the family, alterations in roles were noted to be a complex adjustment for many families because it entailed the ongoing reconfiguration of roles over the course of the mother's illness (Lewis, Ellison, & Woods,

1985). Although restructuring implies that others “fill in”, it does not ensure that gaps in parenting, task completion, role performance, and role overload will not result.

Two early studies provided initial information on adolescents’ distress as related to changing roles and assuming extra family responsibilities due to parental cancer (Christ et al., 1993; Christ, Siegel, & Sperber, 1994). Qualitative methods were used in both studies. The investigators found that child developmental characteristics were related to the differing perspectives of younger (n = 87) and older children (n = 120) about their increased responsibilities and roles. Younger children, ages 7 - 11, were resentful of their older siblings, ages 11 - 17, who took over a parental role or responsibility. Adolescents viewed added household responsibilities and caretaking for other siblings and the parent to be stressful. The researchers reported that additional tasks and role responsibilities conflicted with age-related developmental needs, stating that adolescents found it more difficult to acquire independence relative to the parent and the home, and had less time for social activities.

A subsequent landmark study by Grant and Compas (1995) examined 55 adolescents (ages 11 - 18) of parents with various types of cancers in a mixed methods cross-sectional design. Adolescent girls, especially those with ill mothers, experienced increased anxiety and emotional distress, when compared to boys. Girls associated their heightened stress to increased household chores and role strain related to tasks and functions normally attributed to mothers. A subscale reflecting family responsibilities and role strain of the Adolescent Perceived Events Scale (APES; Compas, Davis, Forsythe, & Wagner, 1987) was used to pinpoint specific role changes and associated stress, such as doing household chores, changes in privileges or responsibilities, younger sibling care,

and expectations by parents. Girls also reported, however, that engaging in these tasks was also helpful in distracting them from their parent's cancer. Because the study was cross-sectional, and differences in types of coping mechanisms between girls and boys were examined, increased rumination in girls compared to increased distraction in boys, it was difficult to account for the girls' emotional distress over time and whether this was attributed to increased responsibilities or to girls' use of ruminative coping.

These findings were supported in a similar study of adolescent daughters who self-reported heightened anxiety related to their expanded role, especially during transitional times during their mother's illness trajectory (Spira & Kennemore, 2000). Transitional periods, when roles were being changed, and then relinquished, were found to be vulnerable periods of heightened emotional distress. During interviews adolescent daughters reported that they derived personal satisfaction in their ability to help, and when this was no longer needed, they felt that they had been displaced. A majority of the girls (ages 12 - 17) were less concerned about the tasks resulting from assuming some of the mother's role functions than about the effect that the role changes would have on their individual relationship with their mother. They reported worry and fear that the pre-existing relationship they had with their mother before her illness would be altered.

A qualitative cross-sectional retrospective pilot study of children aged 11 to 16 years was the only study which reported that adolescent boys had more difficulty than girls related to taking on extra household responsibilities, although both girls and boys acknowledged an increase in domestic tasks and caretaking for siblings during the parent's diagnosis and treatment (Nelson & While, 1994). Adolescent boys, when compared to girls, reported less time spent engaging in sports, hobbies, and in social

activities with friends. The boys who reported decreased social activities reported that their mothers had cancer and that many had younger siblings at home. A sub-group of boys were noted to have anxiety levels above the mean for the entire sample. Significant characteristics reported by this group included that the parent's illness had affected their schoolwork ($p < .01$), the amount of time for sports ($p < .02$), and leisure time for friends ($p < .01$). They also reported they did not feel able to discuss the illness with either parent ($p < .01$). Later studies did not support the findings that boys have increased anxiety when compared to girls, although these studies did not specifically examine these variables related to anxiety (Huizinga et al., 2005; Welch, Wadsworth, & Compas, 1996). The study by Nelson and While (1994) is limited by its small sample size ($n = 24$), and interviews that were conducted with a diverse group of adolescents. They varied extensively by the length of time since their parent's diagnosis (2 - 6 years), as well as by the age when they were reporting their experiences (ages 7 - 16 years of age). These findings, however, provide a clearer understanding of the rationale of why children may find extra household responsibilities disturbing, that is, not the tasks themselves but the disruption they cause in their social life. Further, it suggests that boys should not be overlooked when examining role changes and increased responsibilities when targeting interventions.

Two recent studies have provided support for the association of role changes due to parental illness with child distress during adaptation. A pilot study of adolescents of mothers with breast cancer, using three structured focus group sessions, was done by Davey and colleagues (2005) to elicit information for planning a future intervention. Ten adolescents, ages 13 - 18 years, stated their lives had been complicated with additional

burdensome roles and household responsibilities due to their mother's illness. Protecting the ill mother and looking out for her physical and emotional well-being was reported. The focus group was composed of five males and five females and was racially heterogeneous. This study was important because the investigators set up safeguards to limit threats that may impair research validity. These included shorter periods of time since parent diagnosis to ensure more accurate child reporting, a diverse sample, and children whose mothers were not in the terminal stage of the disease. Second, the study was clinically relevant for designing an intervention. Characteristics of the study that limit generalizability were the small sample size, it was retrospective, and used purposive sampling.

A significant association ($p \leq .01$) between changes in role allocation, as measured by the Family Assessment Device (Epstein et al., 1983) and increased child internal, external, and total problem scores on the Child Behavior Checklist was noted in the second recent study of women with breast cancer (Watson et al., 2006). Role allocation was found to be a significant predictor of internalizing problems in children ($p = .01$). Family functioning, as measured by a subscale of the Family Assessment Device and reported by both mothers and children, was found to be predictive of externalizing problems in children ($p \leq .003$). Watson's work has been considered to be the most comprehensive examination of family influences on children of parental cancer (Osborn, 2007). The sample size was substantial; 63 adolescents responded to the FAD questionnaires and a high level of congruence between adolescents and mothers was noted in their evaluation of family function. The Family Assessment Device is a highly reliable scale that measures seven areas: problem solving, communication, role

allocation, affective responsiveness, affective involvement, and behavior control. It has been used to distinguish between dysfunctional and functional families, it has been used in a variety of contexts, and it has good psychometric properties. Alpha coefficients in this study ranged from .616 - .890.

Changes in Routine and Family Activities

Although changes in the routines of family life, including changes in day to day family activities, have been less frequently reported in the literature, it is surmised that these changes cause disruption (Armsden & Lewis, 1994; Davey, Askew, & Godette., 2003; Issel, Ersek, & Lewis, 1990). Increased illness demands impact the family's ability to function resulting in role changes, separation, shifting of available resources to support the ill parent, and financial strain due to lost earnings and medical expenses. Activities, that previously characterized the family's sense of identity, such as family vacations, celebrations, and day to day family gatherings, undergo changes that affect how the child perceives the consistency and stability of the family environment. Financial constraints may impact the family's ability to afford previous activities, hobbies, and outings even when the ill parent has regained their health and can return to previous roles.

Studies examining the ill parent's and the spouse's concerns about family function during the cancer trajectory have noted that both the ill parent and the spouse desire to keep the family's environment as "normal" as possible (Hilton, 2000; Houldin, 2007). During qualitative semi-structured interviews with male partners of breast cancer patients, men reported that maintaining previous routines and activities with children presented an ongoing challenge, and they gauged their own ability to cope with the partner's illness by maintaining family day to day routines (Hilton, 2000). According to

Zabora and colleagues (1992), patients who underwent BMT and their families reported that at six months post-transplant they began to feel a sense of normalcy, as usual routines began to return. However, participants also reported that while daily family function was stabilizing, ongoing fatigue due to caregiving demands and heightened stress related to financial constraints began to emerge.

Like the male partner, children also view the continuation of routines and daily activities as a reflection of the family's and their own ability to manage. Qualitative interviews conducted during a pilot study revealed that both parents and children perceived the continuation of normal routines of daily life to be protective in dealing with the parent's cancer (Huizinga et al., 2003). However, maintaining normal routines was difficult due to illness demands and lack of domestic support that prohibited ongoing everyday activities and customary practices. Family members experienced increase stress and turmoil when routines were disrupted, and were found to experience added strain and conflict when external support became intrusive (Hilton, 2000). Zabora and colleagues (1992) found that disruption in planning for future family activities was a source of stress in the early stages of parental bone marrow transplantation.

Literature Review for Children's Coping

Children use coping strategies as deliberate efforts to offset stress and adjust to situations. These efforts are central to the process of adaptation. Coping strategies vary and are hypothesized to be influenced by age differences, individual temperament, consequences, family, peers, and well being (Nurmi, 2004), although many of the findings on these variables are preliminary or inconsistent. For example, research on the effect of age and developmental maturation on the types of coping strategies that children

and adolescents use is inconclusive, although most studies indicate that emotion-focused coping increases with age (Nurmi, 2004). Differing classifications of types of coping strategies and inconsistencies in the measurement of coping have complicated conclusive findings in children's use of coping (Compas et al., 2001).

Within the context of parental illness, children's coping strategies are focused on decreasing the impact of the illness on the family, on the affected parent, and on themselves. Parental coping styles, descriptive variables about the child, and family functioning variables which may influence the child's coping strategies have been previously discussed. The following is a summation of literature focused on specific coping strategies used by children during a parent's cancer trajectory.

A recent review of the literature by Grabiak and colleagues (2007) provides an overview of the ways that adolescents, ages 10 - 18, cope with a parent's cancer. Based on Lazarus' delineation of types of coping (1980), the reviewers conclude that children use both problem-focused and emotion-focused coping. Seeking information about the parent's illness, taking on more household tasks, and nurturing other siblings exemplified problem-focused coping (Nelson, Sloper, & While, 1996; Sears & Sheppard, 2004). Emotion-focused strategies, such as humor and expressive writing (Davey et al., 2003; Spira & Kennemore, 2000), faith, hope, and distraction (Davey et al., 2003), and ruminative coping (Grant & Compas, 1995) are reported. More emotion-focused coping and avoidance behaviors were found in adolescents compared to preadolescents due to higher levels of distress (Compas et al., 1996; Weisz, McCabe, & Dennig, 1994). These researchers attributed this finding to older children's more refined ability to have insight into the complexities of illness, and their inability to control illness. Therefore, a shift in

focus to sustaining and fortifying themselves and their families, rather than a focus on changing the situation, is characteristic of children as they age. Adolescents tend to recognize that the illness is more affected by sources outside of themselves and therefore they use more emotion-focused coping behaviors.

Compas and colleagues (1994), using a large sample of children (n=134), found that children perceived very little control over their parent's cancer. This finding was noted across age groups of children (preadolescent, adolescents, and young adults) who were closely involved with the parent during illness and treatment. Children perceived parental illness as beyond their control, and saw others, such as healthcare personnel, the well parent, and adult family members, as having more effect on their parent's illness. Compas and colleagues used this conclusion as evidence to support their theory that emotion-focused coping, rather than problem-focused coping, is used by children. Therefore, the study is limited by the narrow application of these findings to support their conclusions regarding coping.

Children report that social support is helpful in the coping process. An early study by Issel, Ersek, and Lewis (1990) revealed that both family members and friends were important. Children, 13 - 20 years of age, compared to younger children ages 6 - 12, reported twice as often that friends and non-family adults helped them cope. The study was limited by its sample of children of breast cancer patients who were primarily Caucasian, and from middle to high socioeconomic status. Family, friends, and the school environment provided a sense of normalcy and distraction to adolescents of mothers with breast cancer (Kristjanson, Chalmers, & Woodgate, 2004). Thirty-one adolescents reported that the continuation of schoolwork, school activities, and being able to count on

friends for support during stressful times aided their coping. Adolescents stated that being able to express themselves to healthcare providers who would listen, without being critical of their views, was a helpful coping strategy. Support groups and structured information sessions were helpful resources in adolescent coping (Davey, 2005).

Disengagement or passive strategies such as avoidance, distancing, and wishful thinking during times of stress have been recognized by psychologists and child development experts as a distinct subtype of coping used by children (Compas et al., 2001), although these strategies have been found to be significantly associated with poorer psychological adaptation and higher levels of child distress (Coles, Leech, & Parkenham, 2007; Compas et al., 1996; Kotchick et al., 1996). In an early study examining children of mothers with breast cancer, approximately 20% of older children, ages 13 - 20 years, deliberately tried to avoid dealing with their mother's illness through the use of disengagement strategies (Issel, Ersek, & Lewis, 1992). More avoidance responses in adolescent girls, compared to adolescent boys, were noted by Huizinga and colleagues (2005), in a study of 220 adolescents of parents with cancer.

Developmental level, gender, racial, and culture differences are important considerations when examining children's coping strategies during parental illness. Davey and colleagues (2005) examined coping in children of parents with cancer and found that boys, Caucasian and African-American, and African-American girls used more introspection, withdrawal coping strategies, and engagement in routine activities than Caucasian girls who used expressive engagement coping strategies. African-American adolescents tended to rely more on their faith and religious practices, attendance at church and praying, as coping strategies than Caucasian adolescents.

In the examination of coping in children, developmental considerations must be considered because they influence the child's perception of the illness and therefore the ways in which the child copes. Even in children, coping needs to be looked at as an ongoing process which is consistently evaluated and refined, rather than as the use of single strategies that occur in isolation (Compas, Connor-Smith, Saltzman, Thompsen, & Wadsworth, 2000; Schmidt, Petersen, & Bullinger, 2003). Currently, an examination of children's coping with parental cancer as an ongoing process has been only minimally studied and reported in the literature.

Literature Review for Adaptation

Within the context of illness, especially chronic illness, adaptation has been widely used to describe both the process of response to illness as well as the outcome of this process. The meanings have often been used interchangeably, and there is a failure by investigators to conceptually define this concept so that the research focus can be clearly understood. In studies on cancer, the term adaptation has been associated with individual and family response to both the biologic and the extenuating psychosocial changes that occur. Stress, coping, quality of life, affective and behavioral function, resilience, social support, and distress are all concepts that have been associated with adaptation and sometimes used interchangeably (Rodin, 2003; Houtzager et al., 2004; Molassiotis, 1997).

A two-tiered family system stress framework for chronic illness, The Family Adjustment and Adaptation Response Model (Patterson, 1988; Patterson & Garwick, 1994), is helpful in providing conceptual clarity of the two terms, adjustment and adaptation. In this framework, adjustment, the first phase of the model, is characterized as

the initial time in which the individual or family attempts to meet illness demands with their own existing resources and abilities. Crisis occurs when individual or family resources wane and become overwhelmed in meeting the demands, causing a lack of balance within the system. This state of crisis compels the individual/family to make modifications through the development of new coping strategies and resources, reduction of the stressor demands, and/or by reconstructing one's view and meaning of the stressor. The authors contend that "the adjustment-crisis-adaptation process" is cyclical over time in response to the changing nature of the stressor. It is implied that adaptation is a product of modification and coping so that the illness can be integrated within the changing individual and/or family system.

In the body of literature examining children's response to parental illness, child psychosocial function has been used as an indicator of adaptation. This has resulted in the use of instruments which measure constructs of psychosocial function in children. However, conceptual clarification by investigators to define adaptation, which would help to explain the use of these instruments, has been sorely lacking in the literature.

Children's Adaptation to Parental Illness

A host of studies have looked at the effects of illness on the collective family and individual family member's stress and function (Kim et al., 2006; Korneluk & Lee, 1998; Kotchick et al., 1996; Malassiotis et al., 1997). While a majority of these studies have focused upon the effects of illness on the caretaker or spouse, there has also been an increasing awareness and effort to examine the effects of parental illness on children.

Empirical studies on children adapting to parental illness have been conducted on a wide array of populations. Children of parents with chronic illnesses such as diabetes,

severe headaches, and irritable bowel syndrome and children of parents with debilitating illnesses such as hemophilia, HIV, and multiple sclerosis (Kotchik et al., 1996; Forehand et al., 1998; Woodring et al., 2005; Brandt & Weinert, 1998) have been studied. Children of parents with cancer are a primary population that has been described and reported on in the general literature on parental illness (Romer et al., 2002).

The literature provides a broad range of studies which have specifically evaluated how children respond to a parent's physical illness (Armsden & Lewis, 1994; Forehand, Steele, Armistead, Morse, Simon, & Clark, 1998; Morgan, Sanford, & Johnson, 1992; Peters & Esses, 1988; Romer et al., 2002; Welch, Wadsworth, & Compas, 1996). Findings of these studies have focused on ascertaining if children, in fact, do have psychosocial problems related to parental illness, and in identifying factors which may influence psychosocial distress. To date, findings regarding whether children have psychosocial distress related to parental physical illness are diverse and at times contradictory, in terms of the degree of impact the parental illness has on children. Studies vary in their reports regarding which illness, parent, and child variables have been found to affect children's psychosocial morbidity.

In an early review of the literature, conducted during the infancy of research on this topic, investigators were unable to find enough empirical evidence to conclude that children have psychosocial problems as a result of the parent's illness (Armistead et al., 1995). The reviewers' conclusion was primarily based on their examination of a small number of studies up to that time (n=6) which had used either a control sample or normative data for comparison. A later review of the literature which examined 28 studies concluded that while children are distressed by their parent's illness, many were

not in the clinical range to warrant professional intervention (Korneluk & Lee, 1998). Based on data from multiple informants (ill parents, well partner, and children), the reviewers concluded that these findings did not discount that emotional and behavioral problems result from parent illness. They found that a significantly greater number of children did *self-report* clinically relevant symptoms. The investigators advocated future study of family system variables which support positive family functioning to promote effective child coping. The review was limited in that it was composed of studies of children whose parents had either illnesses associated with social stigma or illnesses with poor prognoses, such as children of fathers with hemophilia (Steele et al., 1997); children of mothers with AIDS (Forehand et al., 1998); and children of cancer patients (Birenbaum et al., 1999; Compas et al., 1994; Osborn, 2007).

A later literature review by Romer, Barkmann, and Schulte-Markwort (2002) examined a broad spectrum of literature and then drew conclusions on child psychosocial morbidity. Both qualitative (n = 9) and quantitative studies (n = 30) were included. This review was extensive in that it also included 5 previous reviews, three theoretical articles, and five case studies. Although the review was published in 2002, it contained only literature up until 1998. From their review of empirical studies, the reviewers concluded that children of seriously ill parents (ie. cancer, AIDS, renal failure) had more symptoms than controls, symptoms that they internalized such as anxiety and depression. Several factors were found to significantly affect the degree to which children experienced emotional stress and psychosocial maladaptation including sex of the ill parent, gender and age of the child, and the child's perception of the illness. Forty percent of the empirical studies that were reviewed pertained to children of cancer patients, which may

bias the overall conclusions. Methodological problems found by the reviewers were that studies lacked theoretical frameworks, they were primarily cross-sectional and therefore did not examine variables across time, and they were generally composed of small samples.

Children's Adaptation to Parental Cancer

Only one study has examined children's response to parental bone marrow transplantation (Zabora et al., 1992). The focus of this study was to explore long-term psychosocial issues which affect families after transplant, and was not focused specifically on children of parents undergoing transplant. Therefore, findings on children were minimal. Study time points did not focus on the acute transplant period; data were collected pre-transplant, at 3 months, 6 months, and then every 6 months up until 2 years post-transplant. One hundred sixty "significant others" comprised the study sample and no further description of the participants was given. Children, grouped into 3 categories by age, were examined for their psychosocial response to their parent's post-transplant experience. The number of child participants was not reported. It was unclear who reported the data, the parent or child. Children under the age of five years were observed to have grief and withdrawal reactions, children over the age of five up to adolescence (not defined by age by the author) were found to be confused by the illness, and adolescents were reported to experience stressors related to shifts in roles and responsibilities. Only preliminary findings were published. No further findings on this study have been found in the literature. No other studies pertaining to children of parents undergoing bone marrow transplantation are noted in the current literature. *Because the literature on this population is minimal, findings from research on children's adaptation*

to parental cancer will be examined. Although BMT for the treatment of cancer is a unique situation characterized by high levels of fear and uncertainty related to the imminently life-threatening nature of the procedure, and although it differs from some situations involving cancers that are chronic, the overall illness/treatment characteristics and resulting life changes for families and children enduring parental cancer best serves this literature review.

Research has generated consistent evidence that children of parents with cancer, like children of parents with other types of illness, may experience both emotional and behavioral distress (Compas et al., 1994; Siegel et al., 1992). Consistent with research related to generic parental illness, research in children of parents with cancer initially focused on establishing the extent of psychosocial problems found in this population. Definitive conclusions regarding the extent of emotional and behavioral distress are complicated by methodological differences in studies. However, most researchers, when looking at the aggregate of findings believe that a small, but significant, population of children do have psychosocial distress, and that a significant group may warrant clinical intervention (Osborn, 2007; Romer et al., 2002; Visser et al., 2004). Identification of factors which predispose children to psychosocial vulnerability continues to be a primary focus of research. Moreover, variables that may enhance adaptation are now being studied.

In two early studies, children of parents with breast cancer were not found to have significantly increased emotional and behavioral problems. Comparison normative data and a comparison group of children of parents with diabetes and non-malignant breast disease were used in a study which examined 13 children's (m = 10.8 yrs.) behavioral

adjustment and self-esteem (Armsden & Lewis, 1994). Child behavior assessment was based upon data collected from the mother using the Louisville Behavior Checklist (Miller, 1980) and upon the report of a nurse-observer using the Zeitlin Coping Inventory (1978). In a similar study of 32 children of breast cancer mothers (m = 12 yrs.), behavior was assessed by mothers using the Child Behavior Checklist (Howes et al., 1994). Normative data were used for comparison. In both studies, child behavioral problems were found to be lower in the children whose mothers had breast cancer than in the comparison groups. No differences in total problems, as noted on the Louisville Behavior Checklist, nor on the Child Behavior Checklist, were found. Both studies were limited by their small sample size, primarily single-rater reporting by the mother, and on measurement parameters. A limitation of these findings is that the combined score (TPS), rather than the individual scores for internalizing (TIS) and externalizing (TES) symptoms, may not have provided as sensitive nor as complete results when compared to each measured individually.

However, a later study by Nelson and While (2002) did not corroborate these early findings related to the incidence of psychosocial problems. Nelson and While (2002) reported in a study of 80 children, ages 8 - 16 years, that the prevalence of psychosocial adjustment disorders was consistent with the prevalence rate of 25 - 39% found in studies prior to 1991 (Northouse, 1984; Pederson & Valanis, 1988; Roy, 1990). A recent Dutch study by Huizinga and colleagues (2005) found that in adolescents and young adults (n = 284), 21% of the sons and 35% of the daughters of cancer patients self-reported clinically significant stress response symptoms when compared to norm group values. The larger sample size and the heterogeneous sample of adolescents strengthened

the merit of this latter study in assessing the incidence of adolescent psychosocial morbidity.

Biernbaum and colleagues (1999) added to the findings regarding the incidence rate of psychosocial problems in an adolescent population and provided more extensive information regarding emotional and behavioral manifestations of these problems. Sixty-six children of parents with cancer between the ages of 6 and 18 were divided into two groups by age, those that were 6 - 10 years of age and those 11 - 18 years of age, to account for variables related to developmental level. Although the study did not report using a specific theoretical framework, family systems theory was implied. The use of multiple raters of child adjustment (the adolescent, the ill parent, and the well parent) and the use of compatible forms of the Child Behavior Checklist, (the parent and youth forms), were included to augment the reliability of the findings. Behavioral manifestations of internalizing emotional distress were measured by the Social Withdrawal, Anxiety/Depression, and Somatic Complaints subscales, while externalizing manifestations of emotional distress were measured using the Delinquent Behavior and Aggressive Behavior subscales. Total problems, the sum of all scores on each scale of the CBCL, were compared to normative data. Both ill parents and adolescents reported higher levels of internalizing problems than that noted in normative data. Adolescents self-reported significantly higher on both the internalizing and behavioral problem scores than the norm group ($p = .05$). Ill parent reports and adolescent self-reports demonstrated a significantly higher proportion (14%) of adolescents with problems above the clinical cut-off level than normative data, as rated by two of three raters.

Two very recent studies lend support to these findings which suggest increased distress in children of parents with cancer (Capelli et al., 2005; Visser et al., 2005). Both studies used a comparative design using control groups. Fifty-five female adolescents matched with children from a healthy parent group reported significantly more emotional distress, primarily manifested by worry, about their mother's breast cancer than the control group (Cappelli et al., 2005). This finding was based on the adolescents' fear and concern regarding their own genetic risk for the illness. A larger study (n = 186 families, 336 children) with a more heterogeneous sample which included parents with various types of cancer, was done by Visser and colleagues (2005). They looked at both young children's (age 4 - 11 years) and adolescents' (age 12 - 18 years) emotional and behavioral function as compared to norm groups. Young sons, as reported by the ill parent, and adolescent daughters, as self-reported and as reported by the ill parent, had significantly increased emotional problems when compared to their respective norm groups, and scores which were above clinical cut-off scores for intervention. Although significant emotional distress was found, it is likely that these results may underestimate the incidence of emotional distress in these groups. A high number of eligible families (57%) declined study participation after being approached, and many cited fear of their own or their children's potential development of emotional distress due to study participation.

A thematic review of the literature by Pedersen and Revenson (2005) corroborated the increased incidence of emotional distress in adolescents. The review included examination of studies conducted on children of parents with several life-threatening illness diagnoses, primarily cancer. Findings from the literature regarding

psychosocial well-being were categorized according to psychological distress, problem behavior, and positive well-being, characterized by self-esteem and personal growth. Mixed findings or insufficient findings related to problem behaviors and positive well-being were noted in adolescents, and thus no definitive conclusions on these two outcomes were found. However, findings related to psychological distress indicated that adolescents experienced more negative emotions that are directed inward and subsequently affect functioning, such as depression, anxiety, and somatization (Grant & Compas, 1995; Lester, Stein, & Bursch, 2003; Mikail & von Baeyer, 1990).

Inconsistent findings over the last ten years and methodological issues have made the interpretation of findings regarding children's emotional and behavioral response to parental cancer quite difficult. Small sample sizes which decreased statistical power, samples consisting primarily of adolescents whose parents had breast cancer, and the use of comparison groups that were not representative were noted in a recent review of ten quantitative studies which used a comparison group, normative data, or both (Osborn, 2007). Inconsistencies between parent and child reports regarding children's distress has also been problematic in assuring the accuracy of reported data and in clearly understanding children's response. Informant agreement between children and parents has been considered moderate (Birenbaum, 1999; Visser et al., 2004) to poor (Osborn, 2007). Children's and adolescents' self-reports of emotional and behavioral distress have been higher than parent's ratings (Armsden & Lewis, 1994; Welch et al., 1996). To complicate data interpretation further, ill-parent physical and psychosocial distress has also been found to be positively associated with parent ratings of their children's distress. Higher levels of total problems in adolescents (Hoke, 2001), higher levels of internalizing

problems in children (Heiney et al., 1997), and higher parental reports of problems in their children were reported when both parents were depressed (Lewis & Darby, 2003). Fathers were found to have more benign evaluations of their children's emotional and behavioral functioning when the mother was the ill parent (Visser et al., 2005), while mothers and adolescents were congruent in their ratings.

Qualitative studies have been noted to reveal higher levels and broader dimensions of psychosocial distress when compared to studies which have used quantitative methods (Huizinga et al., 2003; Visser et al., 2004). Christ and colleagues (1993, 1994) in two early qualitative studies found that children and adolescents of terminally ill cancer patients had heightened levels of psychosocial distress. Emotional distress was described in 87 younger children, aged 7 - 11 years. Fear regarding the parent's symptoms and potential death, guilt, quality of sibling relationships, and disrupted school and social function were found in this group. Adolescents, ages 11 - 17 years of age, n=120, were found by Christ (1994) to display increased emotional distress, including emotional detachment, conflict regarding their availability to the ill parent, and pain related to their ability to anticipate future losses. The relatively large number of participants in these two qualitative studies heightened the credibility of these findings.

Currently, there is less evidence to suggest that children of a parent with cancer experience more behavioral problems when compared to normative groups of school-age and adolescent children. However, this finding is somewhat preliminary in that it is based on quantitative studies (Birenbaum et al., 1999; Capelli, Verma, & Korneluk, 2005; Visser et al., 2005) which may not capture the subtleties of behavioral distress. Withdrawal, increased physical closeness with the ill parent, increased attention to and

checking on the ill parent, and taking on tasks to help the parent were found in school aged children in qualitative studies (Issel, Ersek, & Lewis, 1990; Zahlis & Lewis, 1998). Findings of increased adolescent behavioral problems in the literature have been reported to be associated with individual parental variables, such as parental depressed mood (Lewis & Darby, 2003) and increased parental illness demands (Lewis, 1996) and they were also reported to be associated with other family related variables, including lack of expressiveness and cohesion (Watson et al., 2006).

Conclusion

The effects of parental illness on the child cannot be studied apart from contextual and family influences which impact the child's response. In this review, an examination of studies which address these variables in the proposed model was presented. Findings in the literature support the inclusion of the variables in the proposed theoretical model which includes contextual factors, family structural and interaction variables, coping, and a variable unique to children, cognitive development.

Although children's response to parental acute illness has been studied, the research has been primarily recent, and therefore is in its early stages. However, studies of children whose parents have acute illnesses and studies of children of cancer patients indicate that a significant group of children do have psychosocial distress, and that cognitive development must be considered in the study of child response and adaptation. Thusfar, although diverse findings have been noted, there appear to be consistent findings which suggest that adolescents, especially girls, are at heightened risk for emotional distress; and that adolescents tend to have more emotional distress, while younger children manifest more behavioral distress. Parental illness variables, specifically medical

variables including diagnostic type of illness and treatment, have not significantly been associated with child adaptation. Diverse study findings regarding the association of other variables to one another and to the outcome variable, adaptation, have yielded primarily inconclusive results.

Gaps in the literature are numerous. Generally, replication of previous studies is needed to confirm preliminary findings. Further study of contextual variables is needed to investigate the association of these variables to one another and to further determine their relevance in predicting child adaptation. Because recent consistent findings in the literature indicate that family variables, such as expressiveness and cohesion, are significantly associated with child adaptation to parental illness, further study of these variables is crucial in clarifying their role. Examination of children's adaptation to parental life-threatening illness during the acute phase of treatment is sparse. At this time, few interventions to assist and support children have been reported in the literature, and those that have been reported often lack credible empirical methods to test the efficacy of the intervention (Spath, 2007). Most importantly, there is only one study in the literature which has reported preliminary findings, which consisted of a narrative synthesis rather than analyzed data, on the psychosocial response of children to a parent's bone marrow transplant. Additionally, it seems unlikely that further results from this study will be made known (Zabora et al., 1992).

A number of limitations exist within the current research. Theoretical frameworks to support research studies are sparse in the literature, and when noted, are often limited by use of a single framework that does not integrate theoretical components that must be considered in children. Studies of children's responses to parental cancer

have generally had small sample sizes, often less than 50 participants, and have been conducted predominantly with children of mothers with breast cancer, thus limiting the scope of this research. Few longitudinal studies have been conducted to examine how children respond and adapt to the parent's illness over time, thereby limiting an understanding of children's long term response and adaptation, and further limiting understanding of dynamic causal relationships among variables. Inconsistent child and parent reports, samples composed of various age groups, and minimal use of comparative normative data make analysis difficult.

In conclusion, no published studies have focused on children of bone marrow transplant patients using the variables in this hypothesized model. This study will add to the current literature on children adapting to the acute phase of parental illness, and provide a longitudinal study during this highly stressful period in the trajectory from which to examine adaptation over time. The study will include the examination of family variables that have been found in other studies to be potentially significant in child psychosocial function, and will examine the child's use and effect of specific coping strategies on adaptation. As bone marrow transplant continues to be used more widely to treat young child-rearing parents, this study will fill a gap in the nonexistent research literature concerning children dealing with the acute phase of parental illness. It will also provide a basis for the development of future interventions for this distressed population.

CHAPTER 3

RESEARCH DESIGN AND METHODOLOGY

Chapter 3 describes the research methods for the study. Information about the study design, sampling and recruitment of participants, setting, human subject protection, instruments, data collection procedures, and data analysis will be included in this section.

Study Design

This is a prospective longitudinal study of children whose parents were undergoing an autologous or allogeneic bone marrow transplant as treatment for a malignancy. The study was conducted at Indiana University Simon Cancer Center and includes families and children of in-patient transplant recipients. Data were gathered as part of a larger study funded by the American Cancer Society entitled “Family Adaptation to Bone Marrow Transplant”, which involved over 200 patients and families. The principal investigator in this study, Dr. Betsy Fife, granted permission for the use of data for the purposes of this research. Child data obtained from the first three time points, or the acute phase of the parent’s transplant, were used to examine children’s adaptation early in the treatment trajectory. These time points were (T1) pre-transplant and prior to hospitalization; (T2) approximately two weeks into the hospitalization for the BMT; and (T3) one month after discharge.

Sample

The sample included 61 children, 10 - 18 years of age. This age range was chosen for two reasons. First, cognitive ability to discern threat and appraise stressful situations by age 10 has been noted in the literature (Compas, Malcarne, & Fondcaro, 1988; Ryan-Wenger, Sharrer, & Wynd, 1997; Schmidt, Petersen, & Bullinger, 2003). Second,

instruments used in this study have been tested on children in this age range. The proposed sample for this study was derived from a convenience sample of families who were recruited for the larger source study, “Family adaptation to Bone Marrow Transplant.” All available families coming to the Indiana University Simon Cancer Center for BMT were approached about their interest in participation.

Sample Size

The final sample size was influenced by the sample recruited for the larger study. Data from a sample of 61 child participants were included in this study. The statistical package, nQuery Advisor (Statistical Solutions, 2006), was used to calculate the power analysis for the individual aims. For Aim 1, a two-sided correlation test with a .05 significance level could detect a true population correlation as small as .355 with 80% power. For Aim 2, a single-group repeated measures ANOVA with a .05 significance level has 80% power to detect an effect size (variance in the means divided by the common variance times 1 minus the correlation) of .054. For Aim 3, a two-sided partial t test with .05 significance level from *a multiple linear* regression model, adjusted for 5 covariates, has 82% power to detect a .10 increase in R^2 when a 6th covariate is added to the model, assuming the 5 initial covariates explained 20% of the variance. This calculation was done assuming that six covariates would be used in the final model. The use of a maximum of six covariates is *based on the limited sample size*, and because there are steps in the model which justify the use of six. (Note: I also examined a model with all 13 covariates as depicted in Figure 1 on page 21. This was done cautiously, that is, results were only interpreted after verifying that standard errors were not inflated, so that over-fitting did not affect the stability of the coefficients. However, a power analysis was

also calculated for adding a 13th covariate with 12 covariates in the model, using the same significance level (.05) and the same number of subjects (n = 60). This analysis indicated 82% power.

Inclusion Criteria

The inclusion criteria are as follows:

1. The child was 10 years of age or older, but no older than 18 years of age at the onset of the study.
2. A parent was having a bone marrow transplant for the first time.
3. The child was living in the home during the time and up to one month after parental transplantation.
4. The child had access to a working phone.
5. The child was able to speak English.
6. The child was willing to complete phone questionnaire interviews.
7. The child had the ability to understand and verbalize responses to the questions that were asked.

Exclusion Criteria

Children were excluded from the study if the family unit changed during data collection, such that the child was no longer living in the home of the parent who was the BMT recipient.

Instrumentation

Adaptation

Subscales from the Child Health Questionnaire (CHQ-Child Form-87, Subscales five and six) were used to obtain data on children's adaptation, which had been

operationalized as emotional response and behavioral response. The subscales assessed behavioral and emotional well-being: subscale five assessed behavior and subscale six assessed emotional/mental health. The questionnaire proposed for this study is divided into two self-report parts given by the child: (a) 18 questions described specific behaviors over the previous 2-week period using 5-point rating scales which ranged from “never” to “very often” (Appendix A), and (b) 16 items asked children about their feelings and moods on 5-point rating scales which ranged from “none of the time” to “all of the time” over the previous two week period (Appendix A). The two subscales constituted 34 data items in the domains necessary for this study. Reliability for these two subscales has been tested in four populations: children with ADHD, children with cystic fibrosis, children with end stage renal disease, and non-ill children in a large school based sample. Internal consistency reliability coefficients for subscale five (behavior) ranged from .80 - .85; reliability for subscale six (mental health) ranged from .82 - .86. Reliability, indicated by coefficient alpha, was determined by this investigator on this scale, utilizing the data obtained for this research, prior to the start of the analysis.

Scoring for these subscales was done according to the guidelines recommended by Landgraf, Abetz, and Ware in the CHQ Manual (1996). Values for each item were summed and then divided by the number of items in each subscale to obtain a score which reflected that dimension of emotional and behavioral adaptation.

The Child Health Questionnaire is a tool used to measure the physical and psychosocial well-being of children. The questionnaire has several versions which include the original and comprehensive parent responder form (CHQ-PF97), two additional shorter parent forms (CHQ-PF50 and CHQ-PF28), and a version for child

responders (CHQ-CF87). The Child Health Questionnaire was developed in 1990 as an assessment instrument of the general functional status and well-being of children ages 5 and older that could be applied to diverse populations of children. Core concepts underlying the instrument include physical mobility, behavior, mental health, general health, social functioning, school functioning, family functioning, symptoms, conditions, self esteem, pain, risk behaviors, social support, parental impact, sick days, and coping (Landgraf, Abetz, & Ware, CHQ Manual, 1996). These core concepts are measured in both parent and child forms of the instrument. The CHQ Child Form-87 has 12 subscales measuring these core concepts; an additional two subscales pertinent to parental impact comprise the CHQ Parent Form-87. The Child Health Questionnaire Form-87 was designed for children 10 years of age and older. It is tailored for telephone interviewing directly with the child responder.

Reliability and validity have been well established. Internal consistency reliability on the CHQ Child Form-87 was calculated on a school based sample ($n = 263$) and on three samples of children with clinical diagnoses: ADHD ($n = 55$), end stage renal disease ($n = 20$), and cystic fibrosis ($n = 30$). All of the subscales, except the General Health Subscale, had consistently high reliability estimates (.70) in each of the four tested groups. This was based on the accepted standards of reliability calculation for group analysis (Nunnally & Bernstein, 1994), whereby a minimum alpha coefficient of greater than or equal to .70 is considered acceptable. High reliabilities were reported by Waters and colleagues (2001) in a large cross-sectional sample of school aged subjects ($n = 2361$, ages 12-18) in Australia on an 80 item English form derived from the CHQ Child Form-87. Cronbach's alpha coefficients for the subscales were reported as $r > .7$, inter-

item correlations on 90% of the items were $r > .4$. The scale has also been translated into a Dutch version. Instrument reliability for that version was reported as above the acceptable standard (Raaijmakers et al., 2002).

Coping

Three subscales from the Response to Stress Questionnaire (Connor-Smith et al., 2000), developed for use with adolescents, were used to measure three dimensions of coping: primary control engagement coping, secondary control engagement coping, and disengagement coping (Appendix A). Four versions of the instrument are available which are tailored to specific types of stressors, such as social stress, family conflict, economic strain, and pain. Items are consistent in the four versions. A modified Social Stress version was used in this study. The version is available for use in adolescents with demonstrated reliability and validity.

The Primary Control Engagement Coping subscale consists of items on problem solving, emotional regulation, and emotional expression. Secondary Control Engagement Coping items include acceptance, distraction, cognitive restructuring, and positive thinking. Denial, avoidance, and wishful thinking items comprise the Disengagement Coping subscale. Reliability indicated by Cronbach's alpha was 0.82 for the Primary Engagement Control Coping subscale, .80 for the Secondary Control Engagement Coping subscale, and .73 for the Disengagement Coping subscale in a sample of 437 adolescents. Cronbach's alphas for these same subscales in the other three versions of the instrument were similar. Test-retest reliability data on 101 adolescents taking the Social Stress version of the RSQ at a 1 - 2 week interval after taking the first test were reported as .81

for the primary engagement coping subscale, .74 for the secondary engagement coping subscale, and .69 for the disengagement coping subscale.

Adequate convergent and discriminant validity coefficients were found for the Primary and Secondary Control Engagement Subscales and the Disengagement Coping Subscale of the Response to Stress Questionnaire with similar subscales on the COPE (Carver, Scheier, & Weintrub, 1989). The COPE is a widely used instrument to measure coping strategies. Primary control engagement was correlated with seven subscales on the COPE scale; secondary control engagement was correlated with two subscales on the COPE scale; and disengagement coping was correlated with five subscales on the COPE scale.

The Response to Stress Questionnaire (Connor-Smith et al., 2000) is a scale developed for adolescents, ages 11 - 18 years of age, to measure a broad range of coping and involuntary stress responses. In the initial items of this questionnaire, subjects are asked to identify aspects of an identified stressor which they perceive as problematic, and then rate the degree of their stress. The rest of the questionnaire contains numerous items which assess the child's coping strategies based on the problems identified in the initial items. The scale is versatile and is based on a bio-psychosocial theoretical framework. Psychometric testing of the scale was done on 3 samples, each sample facing a different stressor (peer, family conflict and economic strain, and recurrent pain). Internal consistency reliabilities (Cronbach's alphas) for the five factors in the scale ranged from .73 to .89 in the peer stressor group, .84 to .92 for family conflict and .80 to .92 for economic strain stressors in the second sample, and .67 to .88 for the recurrent strain stressor in the third sample. Test-retest reliability was good (.69 - .81). Strong convergent

and discriminant validity with similar constructs on the COPE scale (Carver et al., 1989) were noted. Two of the samples were tested for correlations between parent and adolescent reports on coping. Significant correlations for most of the coping subscales were found.

Perceived Illness Impact

The Perceived Illness Impact was measured by children's perceptions of how the parent's illness had affected them, their family members, and the ongoing functioning of the family. Two sub-variables, family structural change and family interaction, were measured.

Family Structural Change

Four items from the Responses to Stress Questionnaire (Connor-Smith et al., 2000) and four items from the Child Health Questionnaire, CHQ-Child Form-87, Section #9, You and Your Family (Landgraf, Abetz, & Ware, 1996) were used to measure family structural change. For the purposes of this study, this instrument, consisting of eight items, was called "The Family Structural Change Questionnaire" (Appendix A). The focus of this measure is to assess the child's perception of personal stress and family disruption resulting from changes in daily family routines, roles, and functioning.

The level of child personal stress resulting from the parent's transplant is measured by the four items taken from the Responses to Stress Questionnaire (Connor-Smith et al., 2000). A detailed description of the Responses to Stress Questionnaire was previously given in the section of this chapter entitled "Coping". The four items used in the Family Structural Change Questionnaire have been adapted for use in the context of parental bone marrow transplantation. The child was asked to rate how stressful specific

items are for them that refer to family changes and taking on added responsibilities. These items are: difficulty getting schoolwork done, inability to socialize with friends, money problems, and increased chores around the house. A 4-point rating scale ranging from “not at all” to “a lot” is used to assess the child’s appraisal of personal stress associated with each of the items.

Disruption in the family’s daily routines and activities resulting from the parent’s transplant was measured by the four items taken from the Family Activities subscale of the Child Health Questionnaire, Child Form-87 (Landgraf, Abetz, & Ware, 1996). Frequency of family disruption due to the parent’s illness was measured by asking the child to rate four items: a) limitations in the types of activities they could do as a family; b) limitations in the ability of the family to “pick up and go” on a moment’s notice, interruptions in everyday family household activities; and cancellation of family plans. A 5-point rating scale ranging from “never” to “very often” was used to assess the frequency of family disruption as reported by the child.

Scoring for the Family Structural Change Scale was done in the following manner: “almost never” and “never” responses from the four items taken from the Child Health Questionnaire (Landgraf, Abetz, & Ware, 1996) was collapsed into one answer so that the four responses can be aligned with those derived from the four-point item responses taken from the Response to Stress Questionnaire (Connor-Smith et al., 2000), as described above. The values for the eight items were summed and then divided by eight to calculate a total score. Scores ranged from 1 - 4 with a higher score indicating a higher level of family change and disruption caused by the parent’s illness. The Family Structural Change Scale was factor analyzed and reliability testing was done on these

items using Cronbach's alpha prior to the start of the analyses. The specific content of the final measure was determined based on the results of the factor analysis. A detailed description of the source instrument, the CHQ-Child Form-87, has been presented earlier in the adaptation section of this chapter.

Family Interaction

For the purposes of this study, three subscales from the Family Environment Scale (Moos & Moos, 1994) were used to measure family interaction. The Family Environment Scale evaluates the social and environmental characteristics of the family system, and identifies areas of change that may be influential in children's responses. The scale in entirety consists of nine subscales. Children's perception of family interaction will be measured using the Family Cohesion, Family Conflict, and Family Expressiveness subscales (Appendix A) which constitute the Relationship Index. The Family Environment Scale has three forms: the Real Form, the Ideal Form, and the Expectations Form. For the purposes of this study, the subscales from the Real Form were used. These measure perceptions of family members with respect to how they relate to one another given the current family environment.

The instrument is written at a sixth grade reading level and is appropriate for children, approximately 11 years of age and up, and for adults. I am also including children 10 years of age as the questionnaire is being administered by interview. The entire instrument consists of 45 self-report true-false items; each subscale contains 9 items. Correlations among the subscales have ranged from $-.53$ to $.45$, demonstrating that each subscale measures distinct descriptors of the family environment (Buros Institute of Mental Measurement, 1992).

Two specific populations were used in the developmental testing of the Family Environment Scale: 1,432 normal and 788 troubled families. Families from all areas of the country were included and were heterogeneous in terms of characteristics, specifically, age of adults, structure, race, family stage, number of children, and nuclear vs. those including extended family members. Troubled families consisted of those confronting alcohol abuse, depression, legal problems, and medical crises. Internal consistency was reported for each of the nine subscales based on a sample of 1,067 participants. Cronbach's alpha was used to indicate reliability and was reported to be .78 for the Cohesion Subscale, .75 for the Conflict Subscale, and .69 for the Expressiveness Subscale. In two other samples of families, those with a member who was depressed ($n = 904$), and those living with alcohol abuse ($n = 356$), internal consistencies on these three subscales were congruent with those originally reported by Moos & Moos, 2002. Among the ten subscales, cohesion is the most highly correlated with the others, particularly with expressiveness ($r = .4$) and with conflict ($r = -.44$) in a sample of 1,468 adults. Subscale inter-correlations among 621 adolescents were higher between cohesion and conflict ($r = -.53$) and less between cohesion and expressiveness ($r = .32$). These indicate that cohesion is associated with other aspects of the family environment. Test-retest reliability was calculated at a two month interval on 47 individuals. Test-retest reliability was .86 for Cohesion, .73 for Expressiveness, and .85 for Conflict subscales. Adolescent one week test-retest reliability was reported as good in an early study (Gehring & Feldman, 1988) for cohesion and control. In psychometric testing, youth were found to differ in that they reported slightly lower overall scores on cohesion and expressiveness, and slightly higher scores on conflict than parents.

Content validity for the Family Environment Scale was substantiated by the authors' report of their process of item selection according to their initial definitions of the scale's constructs, such as cohesion and conflict. Construct and discriminant validity of the scale's constructs were compared with similar scales assessing perceptions of the family and family function. Moos and Moos (2002) reported on other studies which found high correlations between family relationship as measured by cohesion in the FES with the Family Assessment Device (FAD) and the Family Adaptability and Cohesion Evaluation Scales (FACES II and III), and with family function subscales (family problems, family disorder, and family attachment) on the Minnesota Multiphasic Personality Inventory (MMPI). Discriminant validity in the subscales was also reported by Moos and Moos (2002) with other instruments measuring different aspects of the constructs (Blinn, 1988; Holahan & Moos, 1983; Russell, 1980). Moos and Moos (2002) reported expert validity testing of the scale by correlating its predictability with the predictability of professional staff and therapists' ratings of family cohesion and conflict using other means of assessment. The Family Environment Scale has been used in over 150 research studies examining diverse populations (Moos & Moos, 1996).

The cohesion, expressiveness, and conflict subscales each consist of nine items with a dichotomous response format (True/False). Moos and Moos (2002) reported that multi-point response formats can and have been used, although they recommend the simpler dichotomous format. To score each subscale, the sum of responses given as identified on a scoring key is calculated. This raw score is then compared to a standard score based on the normative data, and can be plotted graphically. Higher scores for each subscale indicate increased perception of the construct. The authors report that the FES

has been translated and adapted for use in many non-English formats and has been used in long term studies.

Demographic Factors

The child demographic variables that were explored are age and gender. Parent demographic variables included the gender of the ill parent and type of transplant. Child and parent demographic factors were obtained from an investigator-developed data sheet on the family. This information was obtained at the beginning of the larger family study. Length of parent hospitalization was obtained from the inpatient hospital record.

Parent Factors

Parent Symptom Interference Subscale

This Symptom Interference Subscale of the BMT Symptom Checklist (Fife, 2000) consists of 16 potential symptoms including nausea, vomiting, fever, fatigue, pain, mucositis, and psychosocial symptoms of isolation, loneliness, and boredom (Appendix A.). A 5-point rating scale is used to rate each symptom (“not at all”, “a little”, “a fair amount”, “much”, and “very much”). The ill parent was asked to rate the degree to which each symptom interfered with his/her daily life. This was obtained from the recipient parent at each of the three time points. The total score was used to quantify the level of symptom interference. Length of parent stay in the hospital was collected from an audit of the medical record.

Procedures

Procedure for Sample Recruitment and Enrollment

Subject recruitment to the study was secondarily obtained through recruitment of the entire family. All potential BMT patients and their families were informed of the

study at a weekly BMT Educational Seminar that is offered for all prospective patients and their families who are undergoing this therapy. All aspects of the BMT program are discussed in this meeting. During this session, a member of the research team provided general information about the study to interested families. At that point, or prior to hospital admission, any interested potential participants filled out a form stating they would like more information about the study and gave permission for the research team to contact them, in accordance with HIPAA guidelines. One of the researchers then contacted the family in person or by phone to review the study and obtain consent. If parents consented to participate, all children living with them between the ages of 10 and 18 were invited to participate. Parents gave written consent and the children provided assent.

Procedure for Data Collection

Child interviews were conducted by phone because the majority of the families lived at a considerable distance from the transplant center. Parents and children were not given a copy of the interview questionnaire prior to the interview to prevent parent preparatory coaching and to ensure that the items are direct responses from the children. However, children were given a color coded answer sheet to assist them in responding to the items as they were being asked by the interviewer.

An introductory letter and picture of the interviewer was sent to the child participant following recruitment. Prior to the telephone interview, the interviewer called the child, introduced herself, verified the child's assent to participate in the study, and set up a time for the initial telephone interview. These measures were implemented to help the child feel more at ease with the interviewer when the telephone interviews began.

Telephone interviews, using the same instruments, were done at the three time points of the study: T1, within one week prior to hospitalization; T2, at two weeks after the start of hospitalization; and T3, at one month after patient discharge from hospital. All interviews were conducted by the doctoral candidate. Subjects were informed at the start of each telephone interview that the interview may be broken into more than one session if necessary to decrease subject burden. I, as the interviewer, stopped the interview and completed it at a later time if I perceived that the subject was having difficulty or did not want to be in the interview process.

Procedure for Data Security

Pencil and paper interview data in this study are stored in a locked file cabinet maintained solely for the purposes of this study. All information stored on computers is password-encrypted. Paper and electronic files that contain identifying information are not related to participant interview data. The computer used for data input and storage is protected with both software and hardware firewalls.

Data were collected through telephone interviews that lasted approximately 35-40 minutes. Two possible methods of recording data from the child interviews have been used. An indirect method whereby telephone data was recorded using paper and pencil and then later entered into a university protected secure server was necessary early in the study. The second method was to directly enter telephone data as they were collected into a study computer by the interviewer. In both cases, data from the computer were downloaded and compiled by the Division of Biostatistics of the Indiana University School of Medicine in Indianapolis. No names are associated with the entered data, and codes are assigned to insure confidentiality. Access to the data is limited to authorized

study personnel only and available through a secure website. To maintain accuracy, a research assistant verified that data collected on hard copy telephone interview sheets (method 1) corresponded to the computer data. The paper and pencil forms were necessary early in the study as the data files had not yet been set up.

Protection of Human Subjects

Human subjects were recruited for this proposed research through the larger family study conducted by Dr. Fife. In keeping with the policies of the Health Insurance Portability and Accountability Act (HIPAA) and Indiana University, all research was reviewed by the Institutional Review Board (IRB) and the Scientific Review Committee (SRC) of the Indiana University Simon Cancer Center.

The consent process for the children was established in accordance with a procedure approved by the IRB. Each child participating in the study signed an assent to assure that she/he chose to participate and was not being coerced. The parent or guardian's consent was obtained first.

The consent included an explanation of the nature, purpose, procedures, risks and safeguards in understandable language for both the parent and child populations. The voluntary nature of participation and the right to withdraw without penalty were explained to both the parent and the child. The child was told that he/she may refuse to answer any of the questions that they feel are too uncomfortable to talk about. I, as the interviewer, stopped the interview if the child indicated "feeling tired".

Adequacy of Protection Against Risks

I conducted all telephone interviews with each child. During the initial phone interview, I verified the child's assent to participate in the study and reviewed with the

child the contents of the assent form including voluntary participation, confidentiality, elective termination of participation, and potential risks and benefits. At the beginning of each subsequent interview, I asked the child for his/her verbal assent for participation in the interview session and inquired if the child had any questions and/or need for clarification about his/her participation.

This research involved slight emotional and psychological risk. Precautions were taken to minimize these risks by: a) thoroughly explaining to the child in developmentally appropriate language the purpose of the study, b) emphasizing that participation is voluntary, c) allowing the child to stop at any time during the interview, d) using a single well-trained interviewer with a master's degree in psychiatric nursing, and e) coding data for confidentiality. These measures were employed at each data collection session. Because this study involves minors and because of the sensitive nature of the subject and questionnaire items, a protocol that outlines a procedure for the management of any untoward emotional effects from the child's participation was created. Any informational data from children that is of concern to the researcher about the child's well-being was disclosed to the parent, with the pre-interview assent of the child. Available psychology consultants and a protocol for proceeding in this type of instance were available through the larger family study.

Data Analysis

The data analysis plan included data cleaning, assessment of the reliability of the instruments, and a description of the sample and instruments. Data were collected and downloaded into a secure web server at the Indiana University School of Medicine Division of Biostatistics. Questionnaires were coded and cleaned by researchers and

statisticians. Members of the research team assessed the data for missing responses, errors when transcribing data from paper to computer in the early stage of the study, miscoding of items, and outliers. Types and rates of errors were documented and reported to the research team for decision making regarding possible editing, correction, and data imputation (Van den Broeck et al., 2005). Prior to beginning data analyses, final measures were determined using factor analyses and reliability testing was done on each instrument. A data set was converted and compiled in SPSS-PC for Windows, version 15.0 (SPSS, Inc., 2007) for this study by statisticians prior to their release to this investigator. The data file included responses to all individual items; scoring was done according to the instrument's manual for use. All scores were computed and analyzed using SPSS 16.0. Data will be analyzed using descriptive and inferential statistics. Analyses were done according to the specific aims presented below.

Preliminary Analysis

Analysis began with the objective determination of the reliability of the measures. Coefficient alpha was calculated as a measure of internal consistency and reliability on all multiple-item scales. In addition, all scales were factor analyzed using varimax rotation to assess the dimensionality of the scale and construct validity. For example, it was expected that there would be at least two dimensions indicating family structure—one relative to interaction within the family unit and the other pertaining to structural change and its effects on the child's personal life. In addition, all scales were evaluated for missing data. Because multiple item scales and subscales were used to test variables in the model, and because the scales tended to be fairly unidimensional with 'replaceable'

items, when computing scale scores, a person-specific mean of non-missing items was substituted for missing items if 50% or fewer of the scale's items were missing. When more than 50% of the items were missing on a particular scale, the subject's data on that scale was excluded for use in the analysis.

Statistical Analysis of Aims

Each of the following specific aims and corresponding research questions were analyzed using appropriate statistical methods as noted below. Using the theoretical model in Figure 1, page 21, as a basis, the primary purpose of this analysis was to determine the strength of the factors most important in affecting children's adaptation during the acute phase of a parent's BMT.

Descriptive statistics were performed on each variable in the model at each of the three critical time points during the acute phase of the BMT trajectory: (T1) pre-transplant and prior to hospitalization; (T2) approximately two weeks after infusion of bone marrow or stem cells during the parent's hospitalization for the BMT; and (T3) one month after the transplant. Descriptive statistics, which included frequencies, stem and leaf plots, central tendencies, and variability were completed for all variables. Moreover, all variables were examined for outliers, and appropriate strategies were instituted to correct for problems that were identified.

Aim 1: To examine the bivariate relationships among all variables in the proposed model at each time point. The strength of these relationships then determined which variables were considered in the regression models with the dependent variable, adaptation, measured by behavioral response and emotional response.

Each of the independent variables was examined separately for their association with one another and with the dependent variables. This was carried out by calculation of a correlation matrix that included all independent and dependent variables, and it was done at each of the three time points. A p - value $\geq .20$ for bivariate Pearson correlation (r) with the variables indicating adaptation was considered sufficiently significant to include the variable in the multiple regression models.

Aim 2: To examine changes in the effects of all variables in the model, which include family structure and interaction, coping strategies used, and the level of adaptation across three time points in the BMT trajectory: (T1) pre-transplant and prior to hospitalization; (T2) approximately two weeks after infusion of bone marrow/stems cells during hospitalization for the BMT; and (T3) one month after the transplant.

Repeated measures ANOVA models that examine overall time effect comparing each time point to one another, were used. Time was treated as a class (i.e., categorical) variable in the repeated measures ANOVA models so that the model will not have to restrictively assume the trend over time is linear. Emotional distress and behavioral distress were modeled separately as the dependent outcomes, while analyzing the impact of each independent variable.

Aim 3: To examine the effects of the independent variables in the model as factors that impact the child's adaptation to the stress of a parent's BMT as they are added hierarchically to the regression at each of the three time points.

Research question 3.1: What is the effect of children's coping strategies on adaptation?

Research question 3.2: What is the effect of the child's perception of the illness, including family structural change and family interaction, on adaptation controlling for coping strategies?

Research question 3.3: What is the effect of the child's age and gender on adaptation controlling for perception of illness impact and coping strategies?

Research question 3.4: What is the effect of the recipient parent's gender, type of transplant, length of hospital stay, and symptom interference on adaptation controlling for coping strategies, the child's perception of the illness, and the child's age and gender?

Hierarchical regression analysis, a multiple regression procedure whereby variables were entered in successive stages into the regression based on the theoretical model found in Figure 1, page 21, was implemented. This was done to examine the strength of relationships hypothesized for the variables in the model while controlling for the effects of other variables. Separate regression models were analyzed for each of the two dependent variables that provided the measure of adaptation. The significance of the contribution of each group of variables was indicated by the degree of change in R^2 . The hierarchical addition of the variables began with coping strategies that were added as a group. This was followed by the family variables serving as measures of the perceived illness impact, then by those contextual variables found to be significantly associated with dependent variables in the correlation matrix. A significance level of $p \leq .05$ was used to determine statistical significance.

CHAPTER 4

RESULTS

This chapter details the results of data analyses guided by the conceptual model presented in Chapter 1 and by the outline of the data analyses as described in Chapter 3. Results of the preliminary analyses that include data screening and reliabilities of measures are reported initially, followed by the results of the analyses specific to each aim and hypothesis. SPSS 15.0 was used for all statistical procedures in this study.

Data Cleaning Procedures

The data set for this study was obtained in an SPSS format from the principal investigator, Dr. Betsy Fife. Data were taken from the larger study, Family Adaptation to Bone Marrow Transplant. Data were initially checked for accuracy by Dr. Fife's research team. Prior to the start of data analyses for this study, out-of-range values, means, and standard deviations were evaluated using univariate statistical procedures. Inspection and frequency analysis of data from the three time points, resulted in finding less than 4% of missing data for any variable. Data from the symptom interference measure obtained from the BMT recipient in each family contained the most missing data, although this was below 4%. One child participant had more than 50% missing data on the Family Structural Change Questionnaire at Time 1 (T1), so this participant's data for that variable was excluded for that time point. All other missing data were randomly scattered throughout the data set. In these cases, person-specific means calculated from the available data were inserted for the missing values.

Univariate outliers were identified by examination of frequency distributions and by inspection of boxplots and stem-and-leaf plots for independent and dependent

variables at each timepoint. Identified outliers were primarily singular for each variable, with the exception of Family Environment Cohesion Subscale (See Table 1).

Analyses involving comparisons of Time 1 (T1), Time 2 (T2), and Time 3 (T3) data represent child sample sizes of N = 61 at T1, N = 59 at T2, and N = 57 at T3. Parent sample size at T1 was 36, T2 = 34, and T3 = 30, as indicated by the parent's submission of the symptom interference data.

Table 1.

Univariate Outliers*

Variable	Number of Outliers	Case Identified
Family Structural Change T1	1	#2
Family Structural Change T2	1	#58
Family Structural Change T3	1	#56
Primary Engagement Cop T2	1	#25
Primary Engagement Coping T3	1	#10
Secondary Engagement Coping T1	1	#19
Secondary Engagement Coping T2	1	#6
Disengagement T2	1	#5
Emotional Response T3	2	#56, #5
Symptom Interference T1	1	#6
Cohesion T1	7	#9,26,7,61, 32-45,60
Cohesion T2	3	#31, 43-58
Cohesion T3	2	#56, 35

*Variables not listed had no identified outliers.

Assumptions of Multivariate Analysis

Multivariate outliers are an unusual combination of variable scores that may distort the results of multiple regression analysis (Mertler & Vanatta, 2005). Assessment for critical multivariate outliers in the data set was done through the calculation of Mahalanobis distance as recommended by Tabachnick and Fidell (2007). Based on this calculation, none of the cases in the data set were identified as critical outliers ($p < .001$, $df = 10$, $X^2 > 29.588$) at any of the three time points when the subscales of the Response to Stress Questionnaire (RSQ) were entered separately. When Mahalanobis distance ($p < .001$, $df = 8$, $X^2 > 26.125$) was calculated on the total coping score, that is, the sum of the three subscale scores, no critical multivariate outliers were found.

Normality was evaluated by inspection of histograms, assessment for skewness and kurtosis, and the Kilmogorov-Smirnov (K - S) test on each variable at each time point. A significance level of $p < .001$ on the K - S test was used to assess significant deviation from normality (Mertler & Vannatta, 2005). Non-normal distributions for the Family Structural Change Questionnaire (FSCQ) at T3, the disengagement subscale of the RSQ at T2, the cohesion subscale of the FES at all 3 time points, and the conflict subscale of the FES at T1 and T3 were significant using the K - S test. Inspection of FSCQ (T3) and disengagement (T2) distributions on histograms did not appear skewed, suggesting that the skewness was minor and that the K - S test had high enough power to detect small deviations from normality. Distributions on family conflict at T1 and T3 were only slightly skewed by visual inspection of the histograms, and statistical analysis for skewness and kurtosis were within the acceptable range for normality (Mertler & Vannatta, 2003). Examination of histograms, Q - Q plots which indicate deviations in

observed scores from the straight line, and kurtosis and skewness statistics were congruent with the significant findings of non-normality for family cohesion distributions. However, outliers were noted in the data at each of the time points, and although not found to be extreme outliers, may account for the findings of slight non-normality. More importantly, it is expected that data in this study would be slightly skewed based on the unpredictability of children's responses to the extreme stress of the parent's illness and its effect on the family, and also due to cognitive, emotional, and social limitations which may influence extreme responses that may be statistically represented as outliers.

At each of the three time points, homogeneity of variance and covariance and multivariate normality were assessed by inspection of plots showing the standardized residuals or errors (ZRESID) against the standardized predicted values of the dependent variables (ZPRED), and by inspection of partial plots. Shapes and residual configurations indicated that multivariate normality was met at each time point.

Reliabilities of Measures

Internal consistency reliabilities for measures used in this study were calculated using Cronbach's coefficient alpha. Variables, measurement instruments, number of items, and alphas for T1 are presented in Table 2, page 115. Reliability was adequate for all of the measures except for the Expressiveness subscale of the Family Environment Scale, however, the subscale was retained because it measured a factor vital to the child's adaptation.

Internal consistency reliabilities for the two outcome measures characterizing adaptation were .81 (behavioral response) and .88 (emotional response). The item to total

correlations for these two outcome variables ranged from .168 to .678 and from .272 to .711, respectively.

Symptom interference was assessed with the BMT Symptom Checklist (Fife et al., 2000) and asked the recipient to rate the degree of interference with daily life caused by individual symptoms pertaining to BMT. Cronbach's coefficient alpha for interference caused by the symptoms of BMT was calculated at .80 for the Time 1 patient sample, which was similar to that reported by Dyer (2008) in the same population. In the original study for which the scale was developed (Fife et al., 2000), reliability was reported only on the frequency scale items which were used in the study, and not on the "interference" subscale. In this study, item to total correlations were strong except for the item, interference from "nosebleeds", ($r = .05$). This item was retained in the scale, however, because bleeding related to bone marrow suppression and resulting thrombocytopenia during BMT is a well documented symptom.

Three coping subscales from the Response to Stress Questionnaire (Connor-Smith et al., 2000) were used to measure children's coping strategies. Reliabilities for the coping subscales were performed and are listed in Table 2. Item-to-total correlations for the primary engagement coping subscale ranged from .25 to .68 which is considered acceptable (Nunnally & Bernstein, 1994). Internal consistency reliability for the secondary engagement coping subscale of the measure was .81; item-to-item correlations varied; two items had low item to item summary statistics below .3 (Ferketich, 1991). Item-to-total correlations ranged from .25 to .71. Coefficient alpha was not increased with the removal of any one item. With respect to the disengagement coping subscale, Cronbach's alpha internal consistency was .75, and item to total correlations ranged from

.27 to .65. Item-to-item correlations indicated that two items (#1 and #27) had slightly lower than the suggested .3 item-to-item summary statistic (Ferketich, 1991). Cronbach's alpha was found to decrease if any of the items were deleted from the subscale, therefore, all items were retained.

Factor Analysis of Selected Scales

Because of the small sample size, factor analyses were not run for most of the measurement instruments. However, for informational purposes, factor analysis was performed on the Expressiveness subscale of the Family Environment Scale due to its low internal consistency. Factor analysis was also run on the Family Structural Change Scale, a new instrument composed of items from two other established scales, to examine its structure. Principal components factor analysis (initial communalities were specified as 1.0) with varimax rotation was performed on the items in each of the two instruments to summarize patterns of correlations among items in each measure to augment and clarify interpretation (Tabachnik & Fidell, 2001). Examination of scree plots was also done. Item loadings were assessed for their conceptual relevance to the component.

As previously stated, reliability on the expressiveness subscale was low at the initial time point ($T1 = .45$). Reliability was rerun on the data at time points 2 and 3 and resulted in consistently low Cronbach alpha statistics ($T2 = .47$, $T3 = .41$). The reliability of the expressiveness subscale as completed by children was compared with the alpha coefficients for both the caregiver (.61) and the BMT recipient (.63). The subscale's reliability was not strong for adults either and suggests that the subscale has poor to marginal reliability in this population of ill parents and family members. Item-to-item correlations were examined and many item-to-item correlations were below .3 indicating

minimal relationships among items on the scale, which then resulted in decreased measurement of the central components of expressiveness (Ferketich, 1991). Two items had low item to total correlations, (.017 and .036) respectively, however, if deleted would not significantly increase the internal consistency of the scale (alpha increase to .513). Factor analysis with varimax rotation was performed and four factors above an eigenvalue of 1.0 were extracted explaining 63% of the variance. Visual inspection of the scree plot was congruent with the statistical findings. When the factor analysis was rerun deleting one of the items with low item to total correlations, four factors remained and the variance increased by only 1%. When the factor analysis was rerun again with both of these items deleted, three factors resulted and the variance dropped to 55%. The two items were retained because eliminating them from the subscale was not found to significantly increase statistical reliability or variance, and because their theoretical contribution to the overall subscale was determined to be important.

Three components with Eigenvalues greater than 1.0, comprising 64% of the variance, were initially noted in factor analysis performed on The Family Structural Change Scale. This scale was comprised of 8 items taken from two instruments and adapted for this study population. Factor loadings on each component were strong—all were above .54. Examination of the scree plot indicated that the use of either 2 or 3 components was plausible. Factor analysis was rerun restricting the components to two. In the analysis using two components, variance dropped to 49%, and one item's factor loading ("being unable to do things I usually enjoy doing with my friends") dropped to below .3 on both components. Examination of the inter-item correlation matrix revealed that this item correlated marginally with half of the other items in the scale. The item-to-

total scale correlation was .234 for this item, however, deletion of the item would not increase Cronbach's alpha. Based on these statistics and the theoretical importance of this item to the child's assessment of changes due to the parent's BMT, the item was retained in the scale. Therefore, three components were recognized in the factor analysis.

Table 2

Internal Consistency Reliability of Measures at Time 1

Variable	Measure	Number of Items	Cronbach's Alpha
Behavioral Response	Child Health Questionnaire (Subscale 5)	18	.81
Emotional Response	Child Health Questionnaire (Subscale 6)	16	.88
Coping	Response to Stress Questionnaire	29	
	Primary Engagement subscale	9	.77
	Secondary Engagement subscale	11	.81
	Disengagement subscale	9	.75
Perceived Illness Impact	Family Environment Scale Cohesion subscale	9	.75
	Conflict subscale	9	.72
	Expressiveness subscale	9	.45
Family Structural Change	Family Structural Change Scale	8	.68
Parent Symptom Inteference	BMT Symptom Checklist Symptom Interference subscale	16	.80

Descriptive Statistics for Demographic and Major Study Variables

Child and parent demographic and contextual data were collected for this study. Child demographic data included the age and gender of the child. Parent demographic data included the BMT recipient's gender and the type of transplant, and illness variables which included the level of the recipient's symptom interference at each time point, and the recipient's length of hospital stay.

Description of the Sample

A subset of transplant recipient families who had children between the ages of 10 and 18 were taken from a larger BMT study. All families reported that the parents were married, and that the non-ill spouse was the primary caregiver. One family was composed of an adolescent child who, since early childhood, was being raised by her two grandparents. Fifty-six percent of the ill parents reported attending or graduating from college, 58.8% reported that they were employed, and 25% reported being disabled. The remaining patients were unemployed, retired, or homemakers. Mean family size was 4 ($M = 4.26$). The maximum number of family members was 7 for the sample. Two-thirds of the families reported annual family income below \$79,999; half of the sample reported income below \$59,999. The sample consisted of primarily Caucasian families (See Table 3, page 120).

Frequencies of demographic data for the sample are found in Table 3, page 120. Children's ages ranged from 10 to 18 years with a mean age of 13.95 years. Over 60% were between 10 and 14 years of age. The sample included approximately 10% more boys than girls. To obtain a better view of the sample, children were divided into two age categories, 10 to 14 years of age and 15 to 18 years of age. Almost twice as many boys as

girls comprised the older group of children (15 boys and 8 girls). Eighteen boys and 20 girls comprised the younger age group. Approximately forty percent of the families, or 25 families, had more than one child in the study. Nineteen girls had a father undergoing transplant; nine girls had a mother undergoing transplant. Twenty boys had fathers undergoing transplant, and fifteen boys had a mother having a BMT. None of the children had parent recipients who died during the three time points of this study. A majority (89%) of the children who reported race were Caucasian. The high number of Caucasian children may be related to socioeconomic factors, such as financial resources, medical care accessibility, and religious-cultural influences, which make BMT more common in this group of patients.

The parent recipient group consisted of 36 participants, 20 were male and 16 were female. The mean age of the ill parent was 46.6 (31.4 - 58.1) years of age. The majority of ill parents had autologous (72.2%) versus allogeneic bone marrow transplants. Eighteen of the 20 male BMT recipients (90%) had autologous transplants; 8 of the 16 female parents (50%) had autologous BMT's. Length of stay in the hospital for the recipient varied widely from 10 to 52 days, with a mean stay of a little more than three weeks. Almost half of the parents were readmitted for subsequent hospitalizations related to transplant complications. Readmission length of stay was not a study variable included in the analyses due to the preliminary and exploratory nature of this study, the lack of previous findings regarding the effect of intermittent hospitalization on child adaptation to parent illness, and the wide variability in readmission occurrence and timing.

Assessment of the interference in personal function caused by the illness symptoms was collected from 31 of the 36 transplant recipients at Time 1. Five parents

did not report information or reported incomplete information on symptom interference. A wide range of symptom interference was noted among those parents reporting symptom interference even at this early time point, indicating that recipients were not asymptomatic initially and perceived their illness to be interfering at least minimally with their function. Missing data were not imputed due to the high variability of symptom interference noted among reporting parent recipients, and because a substantial number of participating parents did not report symptom interference across the three time points (14.5% at T1, 11.7% at T2, and 29.4% at T3). This variable is examined further in later analyses.

Preliminary Testing for Non-independence in the Study Sample

An assumption of multiple regression and ANOVA analyses is independence, which assumes that the data from each participant is unrelated to data collected from other study participants. Assuming independence in participants who may have some degree of non-independence, may lead to underestimated standard errors and therefore liberal p-values, although the point estimates, such as the value of the regression coefficients, are generally unbiased as a result of this violation of assumption of independence. As previously stated, child data was collected from 25 of 61 families who had more than one child in the family, therefore, kinship linkage could result in non-independence due to their exposure to the same causal factors (Kenny, Kashy, & Cook, 2006). To test for non-independence in the responses from these children, intra-class correlation analysis was conducted on the two outcome variables at each time point. The intraclass correlations were equal to .28, -.06, and .07 for emotional response at Time 1, Time 2, and Time 3, respectively, and equal to .28, .38, and .34 for behavioral response at

Time 1, Time 2, and Time 3, respectively, based on data from 17 child sibling pairs (oldest and next oldest child). All of these correlations were not significantly different from zero at the .05 alpha level. Therefore, there is not enough evidence in the present data to indicate that the within-family correlations are different from zero in the population for these two outcomes. However, because several correlations were positive in the .20 - .40 range, especially for the behavioral response, it is possible that with a larger sample size, and increased power, a significant positive intraclass correlation might be detected. Therefore, one limitation of this study is that the p-values reported here may be slightly lower (i.e. liberal) compared to the p-values that would have been observed using a method, such as mixed linear modeling, that accounts for the within-family correlation in the 20 families that contributed more than one child to the data of 61 children analyzed in this study.

Table 3

Sample characteristics at Time 1

	<i>n</i>	Frequency (%)	Mean (SD)	Median	Range (Possible)
Child Demographics					
<u>Age</u>	61		13.95 (2.47)	14.00/11.00	8(10-18)
(10-14 years of age)	38	(62.3)	12.29 (1.31)	12.00/11.00	4(10-14)
(15-18 years of age)	23	(37.3)	16.70(1.06)	17.00/17.00	4(15-18)
<u>Gender</u>	61				
Female	28	(45.9)			
(10-14 years of age)	20				
(15-18 years of age)	8				
Male	33	(54.1)			
(10-14 years of age)	18				
(15-18 years of age)	15				
<u>Race</u>					
Caucasian	48	(89)			
African American	2	(3.7)			
Hispanic	1	(1.85)			
Native American	1	(1.85)			
Blended	2	(3.7)			
Unknown/not reported	7				
Parent Demographics					
<u>Gender</u>	36				
Female	16	(44.4)			
Male	20	(55.6)			
Type of Transplant	36				
Allogeneic	10	(27.8)			
Autologous	26	(72.2)			
Days of Initial Hospitalization	36		22.06 (8.4)	21.00	42 (10 – 52)
1st Readmit		15 (41.7)	18.53 (29.75)	10.0	121 (2 -123)
2 nd Readmit		8 (10.61)	8.62	4.5	8.5 (2-10.5)
3 rd Readmit		2 (5.6)	8.0	8.0	8.0 (4-12.0)
4 th Readmit		1 (2.9)	4.0	4.0	0
Symptom Interference Score	32		30.2	29	16-80

Descriptive Statistics on Study Variables

Table 5, page 126, provides the means and standard deviations for each of the major independent variables at each time point. Further analysis of significant change over time for each of these variables will be examined in Aim 2.

An initial examination of children’s adaptation, as indicated by child emotional and behavioral response, was done by comparing means at each time point with published normative data. Mean scores of child emotional response, measured with the Mental Health subscale of the Child Health Questionnaire (CHQ CF-87), were less positive prior to and during parental transplant than child norms (Landgraf, Abetz, & Ware, 1996; Waters, Salmon, Wake, Wright, & Hesketh, 2001). Child emotional response did not differ from published norms when the parent returned home (T3). Mean scores of child behavioral response, measured by Subscale 5.1 of the CHQ CF-87, were lower than published norms at T1 and T3, however, during the parent’s hospitalization for BMT (T2), children’s behavioral response was slightly when compared to published norms (Table 4).

Table 4

CHQ Mean Scale Scores for Outcome Variables compared with Norms

Variable	CHQ Measure	Norm	BMT T1	BMT T2	BMT T3
Emotional Response	Mental Health (CHQ-CF 87, Subscale 6.0)	74.9 (16.3)	68.44 (14.2)	68.2 (12.1)	74.0 (13.8)
Behavioral Response	Getting Along (CHQ-CF 87, Subscale 5.1)	77.51 (13.8)	73.72 (13.1)	78.76 (13.9)	74.0 (10.3)

At each time point, one-way Anovas were performed in a preliminary analysis to evaluate whether there were significant differences on study variables by demographic or condition groups. Tables 6 through 8, pages 125 - 127, contain the results of this analysis. Significant results are discussed following the tables. Means and standard deviations of those demographic groups with significant differences on study variables are listed in Table 9, page 131.

Child Gender

There was a significant difference in the emotional adaptation between boy ($n = 33$) and girl ($n=28$) participants at time 1 ($F(1,61) = 10.910, p \leq .01$). Boys tended to have better emotional adaptation knowing their parent was preparing to have a BMT. During the parent's hospitalization and one month after, there was no significant difference by gender in the two groups. In managing stress during their ill parent's hospitalization, girls' report of greater use of primary engagement coping compared to boys; this difference was marginally significant ($F(1,61) = .633, p < .054$).

Child Age

For the purposes of this analysis, child age was divided into two categories to determine if significant differences existed in study variables. This was done for two reasons: a) child development experts agree that variations in child cognitive and functional development occur by stage and are influenced by individual, ecological, and experiential factors (Bjorkland, 2005), and b) to be consistent with numerous other studies examining children in the literature which have categorized children by age groups for analysis (Osborn, 2007). Children ages 10 through 14 years old comprised one group ($n = 38$), and children ages 15 through 18 years of age comprised the older group

(n = 23). Age groups were significantly different in their use of primary engagement coping strategies while their parent was hospitalized at T2 ($F(1,59) = 6.255, p \leq .016$). Older children tended to use primary engagement coping strategies, such as problem solving and expressing and controlling emotions, more frequently than younger children.

Because there is wide developmental variability in the two age categories described above, subsequent analyses were performed to more closely examine the sample for age-related differences. The sample was divided into four age groups: ages 10 and 11, ages 12 and 13, ages 14 - 16, and ages 17 and 18, and ANOVAs were rerun, using the Tukey-Kramer post-hoc method to test for pair-wise comparisons with more than three groups. Based on this analysis, significant differences by age group were found before the parent was hospitalized for the transplant (T1). Children ages 14 - 16 perceived significantly higher levels of family conflict than both the youngest and oldest age groups. However, when a Tukey-Kramer post hoc test was done, these differences were not significant. Based on ANOVA analysis children, ages 17 and 18 years of age, used significantly less disengagement coping than the three younger age groups at T1. A Tukey-Kramer post hoc test indicated that disengagement coping was significantly different between 12 and 13 year olds and children 17 and 18 years old. Children in the older age group used significantly less disengagement coping children in the 12 - 13 age group. The use of disengagement coping by children did not significantly differ by age during the parent's hospitalization and recovery.

Parent Gender

Significant differences between recipient gender and transplant type and between recipient gender and length of hospitalization were found in the sample. Males were

predominant in the autologous group of transplant recipients (18 male autologous vs. 2 male allogeneic). Equal numbers of female patients were found in the autologous and allogeneic groups (8 autologous vs. 8 allogeneic). Additionally, there was a significant difference in the length of hospital stay by gender ($F(1,34) = 4.192, p \leq .05$). Female BMT recipients stayed longer than male recipients by almost 5 days.

There were also significant differences in the coping strategies used by children related to the recipient parent's gender. At T1, prior to transplantation, primary coping strategies were used more when the recipient parent was the father ($F(1,61) = 4.212, p \leq .05$). During the parent's hospitalization for the BMT (T2) and at T3, when the patient was at home, significant differences were found in children's use of secondary engagement coping strategies by recipient parent gender ($F(1,59) = 5.299, p \leq .05$). When the recipient parent was the father, children reported the use of more secondary coping strategies focused upon adaptation to the situation, which included positive thinking, cognitive restructuring, acceptance, and distraction.

Table 5

Means and Standard Deviations for Major Study Variables

Variable	Measure	Time 1 (n=61)		Time 2 (n=59)		Time 3 (n=57)	
		Mean	SD	Mean	SD	Mean	SD
Behavioral Response	Child Health Questionnaire (Subscale 5)	69.56	8.86	72.93	9.82	73.85	7.64
Emotional Response	Child Health Questionnaire (Subscale 6)	59.48	8.67	59.65	7.72	63.40	8.86
Coping	Total Coping	70.10	11.33	66.33	11.48	65.98	12.48
	Primary Engagement SS	22.16	4.96	21.47	4.99	21.23	5.16
	Secondary Engagement SS	29.99	6.05	23.76	4.69	27.72	5.65
	Disengagement SS	17.95	5.01	21.40	5.85	17.03	4.66
Family Structural Change	Family Structural Change Scale	13.97	4.17	14.15	5.05	12.63	4.21
Perceived Illness Impact	FES						
	Cohesion SS	16.11	1.57	16.23	1.92	16.03	2.28
	Expressiveness SS	13.19	1.75	12.95	1.86	13.42	1.8
	Conflict SS	11.81	2.18	11.44	2.3	11.52	2.17
Symptom Interference	BMT Symptom Checklist	(n=31) 29.68	9.03	(n=32) 38.63	9.84	(n=29) 30.34	9.19

NOTES:

Higher score indicates more positive behavioral adaptation, more positive emotional adaptation, greater use of total and particular coping strategies, greater perceived family change, and greater perceived family cohesion, expressiveness, and conflict.

Table 6

One-Way Anova Results for Demographic and Continuous Variables – Time 1

Major Variables	Child gender	Child Age	Parent Gender	Type of Transplant
	F-value	F-value	F-value	F-value
Behavioral Response	1.252 (1,59)	2.780 (1,59)	.341 (1,59)	2.233 (1,59)
Emotional Response	10.910 (1,59)**	1.826 (1,59)	2.713 (1,59)	2.596 (1,59)
Primary coping	.633(1,59)†	2.483 (1,59)	4.212 (1,59)*	.544 (1,59)
Secondary Coping	.369 (1,59)	.025(1,59)	.241 (1,59)	.287 (1,59)
Disengagement Coping	2.479 (1,59)	3.099 (1,59)	.204 (1,59)	3.890 (1,59)†
Family structural Change	2.502 (1,59)	.368 (1,59)	3.095 (1,57)	10.868 (1,59)**
Family cohesion	.723 (1,59)	.194 (1,59)	.647 (1,59)	.273 (1,59)
Family expressiveness	2.597 (1,59)	.005 (1,59)	2.190 (1,59)	.884 (1,59)
Family conflict	.223 (1,59)	.010 (1,59)	.003 (1,59)	.658 (1,59)

Numbers in parentheses indicate degrees of freedom (between, within)

*** $p \leq .001$

** $p \leq .01$

* $p \leq .05$

† $p = .055$

Table 7.

One-Way Anova Results for Demographic and Continuous Variables – Time 2

Major Variables	Child gender	Child Age	Parent Gender	Type of Transplant
	F-value	F-value	F-value	F-value
Behavioral Response	.505(1,57)	.523(1,57)	.000(1,57)	.832(1,57)
Emotional Response	3.116(1,57)	.092(1,57)	.240(1,57)	4.708(1,57)*
Primary coping	3.917(1,57)	6.255(1,57)*	3.401(1,57)	2.028(1,57)
Secondary Coping	1.438(1,57)	.388(1,57)	5.299(1,57)*	.001(1,57)
Disengagement Coping	1.432(1,57)	.339(1,57)	1.239(1,57)	3.123(1,57)
Family structural Change	1.288(1,57)	.060(1,57)	.007(1,57)	2.107(1,57)
Family cohesion	.359(1,57)	3.504(1,57)	.123(1,57)	.296(1,57)
Family expressiveness	.037(1,57)	.353(1,57)	.100(1,57)	.080(1,57)
Family conflict	.051(1,57)	.140(1,57)	.253(1,57)	4.233(1,57)*

Numbers in parentheses indicate degrees of freedom (between, within)

*** $p \leq .001$

** $p \leq .01$

* $p \leq .05$

Table 8

One-Way Anova Results for Demographic and Continuous Variables – Time 3

Major Variables				
	Child gender	Child Age	Parent Gender	Type of Transplant
	F-value	F-value	F-value	F-value
Behavioral Response	.376(1,55)	.413(1,55)	.988(1,55)	.323(1,55)
Emotional Response	3.116(1,55)	1.513(1,55)	.993(1,55)	.311(1,55)
Primary coping	.577(1,55)	.740(1,55)	1.626(1,55)	.014(1,55)
Secondary Coping	.045(1,55)	.035(1,55)	4.378(1,55)*	.008(1,55)
Disengagement Coping	.732(1,55)	.262(1,55)	1.773(1,55)	.325(1,55)
Family structural Change	1.140(1,55)	.442(1,55)	.009(1,55)	2.345(1,55)
Family cohesion	.477(1,55)	.323(1,55)	1.223(1,55)	.175(1,55)
Family expressiveness	1.880(1,55)	.365(1,55)	.970(1,55)	1.792(1,55)
Family conflict	1.391(1,55)	1.000(1,55)	.030(1,55)	2.920(1,55)

Numbers in parentheses indicate degrees of freedom (between, within)

*** $p \leq .001$

** $p \leq .01$

* $p \leq .05$

Transplant Type

Because the variable, parent length of hospital stay, did not change over the transplant trajectory, an initial ANOVA was run to determine if there was a significant difference in the length of hospital stay between autologous and allogeneic transplant recipients. A significant difference in the length of the parent's hospitalization by the type of transplant was found ($F(1,34) = 35.426, p \leq .001$). Clients who had allogeneic transplants stayed in the hospital longer ($M = 31.6$ days) than those receiving autologous transplants ($M = 18.38$ days). This was not a surprising finding in that it is well known that an allogeneic transplant is a more complex procedure and involves a longer period of immunosuppression and a period of engraftment.

Autologous and allogeneic transplant groups differed marginally at T1 in their reports of symptom interference (Table 9); autologous recipients tended to report higher levels of symptom interference prior to transplant than allogeneic recipients ($F(1,29) = 3.984, p = .055$). At T2 and at T3, there were no significant differences in the level of symptom interference between the two groups. The statistics reflect the symptom interference of parents who reported these data; imputed data were not used.

Compared to children of parents who were to undergo allogeneic transplantation, children of autologous BMT recipients reported significantly higher levels of family structural change at time 1 ($F(1,59) = 10.868, p < .01$), and higher levels of family conflict ($F(1,57) = 4.233, p \leq .05$) at T2 during their parent's hospitalization (Tables 6 and 7). Greater use of disengagement coping strategies by children of autologous BMT recipients when compared to children of allogeneic transplant recipients was marginally significant at T1 while anticipating their parent's transplant ($F(1,59) = 3.890, p < .054$). Possible reasons for the relationships discussed above will be discussed in Chapter 5.

Interaction Effects

Two way ANOVA analyses were run to test for interaction effects of categorical variables on the independent and dependent variables in the model. Previous studies have found significant parent gender by child gender, child gender by child age, and child gender by treatment interaction effects on children's psychobehavioral functioning when a parent has cancer (Compas et al., 1994; Huizinga et al., 2005; Visser et al., 2005). In testing for interactions, only one interaction effect was found to be marginally significant: a two way interaction of child gender by child age group on perceived family structural change ($F = 3.323, p = .072$) at the time of the parent's hospitalization (T2). There was no

main effect for child gender ($F = .198, p = .658$) and child age group ($F = .415, p = .522$) on perceived family structural change at T2. However, younger male children, ages 10-14, perceived higher levels of family structural change ($M = 14.86$) than females ($M = 12.94$) at T2. Females, who were in the older age group, 15 - 18 years old, perceived higher levels of family structural change ($M = 16.38$) at T2 than males (13.21). The interaction accounted for 5.7% of the variance.

Table 9

Means and Standard Deviations for Demographic Groups on Variables in the Theoretical Model with Significant Differences

Major Study Variable	Time (T)	Demographic Variable			
		Male		Female	
		M	SD	M	SD
Emotional Response**	T1	Child Gender			
		Male		Female	
		62.602	7.709	55.785	8.407
Primary Coping*	T2	Child Age			
		Ages 10-14		Ages 15-18	
		20.297	4.818	23.455	4.896
Primary Coping (p ≤ .054)	T1	Parent Gender			
		Male		Female	
		23.222	4.917	20.640	4.707
Secondary Coping*	T2	24.884	4.794	22.125	4.089
	T3	28.970	6.205	25.869	4.192
Emotional Response**	T2	Type of Transplant			
		Allogeneic		Autologous	
		63.267	6.053	58.412	7.893
Family Structural Change**	T1	Type of Transplant			
		Allogeneic		Autologous	
		11.133	2.875	14.932	4.122
Disengagement Coping (p ≤ .053)	T1	Type of Transplant			
		Allogeneic		Autologous	
		15.875	4.688	18.688	4.971
Symptom Interference (p ≤ .055)	T1	Type of Transplant			
		Allogeneic		Autologous	
		25.333	7.141	31.863	8.659
Family Conflict*	T2	Type of Transplant			
		Allogeneic		Autologous	
		10.418	1.750	11.794	2.372

** p ≤ .01

* p ≤ .05

NOTE:

Higher score indicates more positive behavioral adaptation, more positive emotional adaptation, greater use of total and particular coping strategies, greater perceived family change, and greater perceived family cohesion, expressiveness, and conflict.

Aims and Research Questions

This section presents the results of analyses completed to address each of the study aims and research questions. Tables are included to display findings from the output of statistical tests that were used to address each aim.

Aim 1

Aim 1: To examine the bivariate relationships among all variables in the proposed model at each time point; see Figure 1, page 21 for the theoretical model. The strength of these relationships will then determine which variables will be included in the regression models with adaptation measured by behavioral response and emotional response, as the dependent outcomes.

Pearson correlation coefficients were run to evaluate the relationships among continuous variables in the model; these findings were then used to determine those variables that had an influence on the outcomes, emotional and behavioral adaptation. Those variables that were significantly correlated at .20 or higher with the dependent/outcome variables were then included in hierarchical multiple regression analyses based on the theoretical model.

Categorical variables

According to Field (2005), a point biserial correlation is used to determine relationships between variables when one variable is dichotomous and the other variable is continuous. Categorical variables, which include child gender, parent gender, and type of transplant (allogeneic versus autologous) were coded numerically as 0 or 1. Point biserial correlations were calculated to assess the presence of a significant relationship between the dichotomous variables and each of the outcome variables, emotional

response and behavioral response. The sign of the correlation coefficient is dependent on the way the dichotomous variable was categorically coded. Male gender was coded as “0” and female gender was coded as “1” for both child and parent gender. Type of parent transplant was coded as “0” for allogeneic; autologous transplant was coded as “1”.

Correlation statistics for the three categorical variables with the dependent variables are presented in the table of correlations (Table 10). Behavioral response was not associated with any of the three categorical variables at any of the three time points. At Time 1, female child gender was moderately associated with more negative emotional response. A significant association between more negative child emotional response during the parent’s hospitalization (T2) was found in children of autologous transplant recipients.

Table 10

Point-biserial correlations between categorical variables and dependent variables

	<u>Time 1</u>		<u>Time 2</u>		<u>Time 3</u>	
	Emotional Response	Behavioral Response	Emotional Response	Behavioral Response	Emotional Response	Behavioral Response
Child Gender (female)	-.39**	.14	.02	.09	.23	.12
Parent Gender (female)	.21	.08	.16	.00	.13	.13
Type of Transplant (auto)	-.21	.19	-.28*	.12	.08	.08

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

Table 11

Point biserial correlations of child gender with perception of illness and coping variables

	Time 1		Time 2		Time 3	
Child Gender (female)	FamStrucChg	-.20	FamStrucChg	-.07	FamStrucChg	-.14
	Cohesion	-.01	Cohesion	.14	Cohesion	-.09
	Expressiveness	-.20	Expressiveness	.03	Expressiveness	-.18
	Conflict	.06	Conflict	-.03	Conflict	.16
	Primary Coping	.10	Primary Coping	.25	Primary Coping	.10
	Second Coping	.08	Second Coping	-.16	Second Coping	.03
	Disengagement	.20	Disengagement	.16	Disengagement	.12
	Total Coping	.17	Total Coping	.25	Total Coping	.10

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

Table 12

Point biserial correlations of transplant type with perception of illness and coping variables

	Time 1		Time 2		Time 3	
Transplant Type (autologous)	FamStrucChg	.40**	FamStrucChg	-.19	FamStrucChg	.23
	Cohesion	.07	Cohesion	.06	Cohesion	.05
	Expressiveness	-.12	Expressiveness	-.04	Expressiveness	-.18
	Conflict	.11	Conflict	.26*	Conflict	.23
	Primary Coping	.10	Primary Coping	.18	Primary Coping	.02
	Second Coping	-.07	Second Coping	.00	Second Coping	-.01
	Disengagement	.25	Disengagement	.23	Disengagement	.08
	Total Coping	.11	Total Coping	.20	Total Coping	.03

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

Table 13

Point biserial correlations of parent gender with perception of illness and coping variables

	Time 1		Time 2		Time 3	
Parent Gender (female)	FamStrucChg	.23	FamStrucChg	.01	FamStrucChg	-.01
	Cohesion	-.10	Cohesion	-.00	Cohesion	.10
	Expressiveness	-.19	Expressiveness	.04	Expressiveness	-.13
	Conflict	-.02	Conflict	-.07	Conflict	-.02
	Primary Coping	-.26*	Primary Coping	-.24	Primary Coping	-.17
	Second Coping	-.06	Second Coping	-.29*	Second Coping	-.27*
	Disengagement	-.06	Disengagement	-.16	Disengagement	-.18
	Total Coping	-.17	Total Coping	-.29*	Total Coping	-.26

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

Tables 11 - 13 show the correlations between the three categorical variables and the continuous independent variables at each time point. Point biserial correlations were generally not significant. Transplant type was associated with two of the family variables, family structural change at T1 ($r = .40, p < .01$) and family conflict at T2 ($r = .26, p < .05$), suggesting that autologous transplant recipient families may be more vulnerable early on in the transplant trajectory. A significant association between female parent gender and decreased use of coping strategies was found across the three time points. The two types of coping that were significantly decreased when the mother was the recipient, primary (problem solving, emotional regulation, and emotional expression) and secondary (acceptance, distraction, cognitive restructuring, and positive thinking), are those strategies that are generally considered more favorable in managing a stressor. Across the three time points, a consistent and small significant association ($r = -.26$ to $-.29, p < .05$) between decreased use of these coping strategies and the mother as the BMT recipient occurred.

Continuous Variables

Continuous variables in the conceptual model were screened for their associations with one another and for consideration in multiple regressions. According to Tabachnick and Fidell (2007), multiple regressions best reveal significant relationships among variables when the independent variables are highly correlated with the dependent variable. They permit evaluation of the relationship between each independent variable with the dependent variable while controlling for the effects of each other independent variable in the model. Correlations between the independent continuous variables and the dependent variable, adaptation, were also examined for inclusion in the multiple

regression analysis. Separate correlations of independent variables with the dependent adaptation variables, emotional response and behavioral response, were performed.

Tables 13 - 15, pages 142 - 144, present the results of the Pearson r correlations among all continuous independent and dependent variables in the model. Correlations were in the expected direction.

Correlation of Independent Variables with Child Behavioral Response

Illness Impact

Most of the perceived illness impact variables, as measured by the child's perception of family structural change and family interaction, were significantly correlated with child behavioral response. Family cohesion and family conflict were moderately correlated with child behavioral response throughout the transplant trajectory (See Tables 14 - 16, pages 140 - 142). A higher level of family cohesion was associated with more positive child behavioral response at all three time points (T1 = .48; T2 = .36; T3 = .41, $p < .01$). Higher levels of family conflict were consistently associated with more negative child behavioral response (T1 = -.57; T2 = -.56, T3 = -.41, $p < .01$). Positive child behavioral response was significantly correlated with higher levels of family expressiveness prior to transplant (.36, $p < .01$) and during the parent's hospitalization (.36, $p < .01$), however, there was no significant association between the two when the parent returned home (T3). A steady and moderate significant association between greater change in the family, measured as family structural change, and negative child behavioral response was noted throughout the transplant trajectory (T1 = -.49; T2 = -.42; T3 = -.54; $p < .01$).

Coping

Significant negative correlations between disengagement coping and child behavioral response were also found at all three time points of the transplant trajectory (See Tables 14 - 16, pages 140 - 142). Increased use of disengagement strategies by children to distance themselves from the parent's transplant was consistently related to more negative child behavioral response. This association was strongest at the first two time points ($-0.56, p < .01$) and slightly lower, but significant, when the parent returned home at T3 ($-0.36, p < .01$). Primary coping, which includes problem solving, emotional regulation, and emotional expression strategies, and secondary coping, which includes acceptance, distraction, cognitive restructuring, and positive thinking strategies were not significantly associated with child behavioral response at any of the three time points. The total coping score, a cumulative score of all three coping subscales, was not significantly correlated with behavioral adaptation prior to (T1) nor after the parent's hospitalization (T3). The use of increased levels of cumulative coping strategies by children with more negative behavioral adaptation ($r = -0.31, p < .05$) was found at T2, during the parent's hospitalization.

Correlations of Independent Variables with Child Emotional Response

As previously stated, none of the continuous contextual variables, which include child age, parent's length of hospital stay, and parent symptom interference, were significantly correlated with child emotional response at any of the three study time points. These correlations are given in Tables 14 - 16, pages 140 - 142.

Illness Impact

Significant relationships were found between the family environment variables, which include cohesion, conflict, and expressiveness, with child emotional response. A significant positive relationship between family cohesion and emotional response was found at T1 ($r = .29, p < .05$) and at T3 ($r = .29, p < .05$), indicating that higher levels of family cohesion were associated with more positive child emotional response. Family expressiveness and family conflict were significantly correlated with child emotional response at only one of the study time points. Higher levels of expressiveness in the family were associated with more positive child emotional response prior to the parent's transplant ($r = .30, p < .05$); higher levels of family conflict were associated with more negative child emotional response when the parent returned home after the transplant ($r = -.28, p < .05$).

Negative correlations between family structural change and emotional response were strongly significant at T1 ($r = -.52, p < .01$) and at T3 ($r = -.62, p < .01$), indicating that children's perceptions of higher levels of family change were associated with more negative emotional response. None of these variables (cohesion, expressiveness, conflict, family structural change) were significantly correlated with child emotional response at T2, during the parent's hospitalization. Across the transplant trajectory, there were more significant correlations between the family variables and child behavioral response when compared to the number of significant correlations between these same variables and child emotional response.

Coping

A significant moderate negative correlation between disengagement coping and child emotional response was found at both T1 and T3 (T1 and T3: $r = -.55$, $p < .01$), indicating that increased use of disengagement was associated with more negative child emotional response. Primary engagement coping ($r = -.30$, $p < .05$) and total coping ($r = -.38$, $p < .01$), were significantly negatively correlated with child emotional response at T3, one month after the transplant hospitalization (Table 16, page 142).

Table 14. Bivariate Correlations of Continuous Variables in the Model--Time 1

	BMT Stay	Child Age	Symptom Interfere	Cohesion	Express	Conflict	FSC	RSS Total	RSS Primary	RSS Second	RSS Diseng	Emotion Response
Child Age	-.04											
Symptom Interfere	-.21	-.12										
Cohesion	-.07	-.02	-.01									
Expressive	.15	.07	-.05	.23								
Conflict	.03	-.03	.19	-.39**	-.41*							
FSC	-.38**	.02	.23	-.17	-.28*	.20						
RSS Total	-.05	.01	-.02	-.11	.11	.20	.10					
RSS Primary	-.07	.26*	-.17	-.12	.18	.01	.11	.78**				
RSS Second	.09	.04	-.25	.01	.27*	.05	-.26*	.81**	.56**			
RSS Diseng	-.15	-.31*	.40**	-.37**	-.22	.39**	.44**	.52**	.55**	.06		
Emotion Response	.22	.23	-.21	.29*	.30*	-.24	-.52**	-.11	.02	.22	-.55**	
Behavior Response	.20	.20	-.13	.48**	.36**	-.57**	-.49**	-.17	-.17	.10	-.56**	.64**

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

Table 15. Bivariate Correlations of Continuous Variables in the Model--Time 2

	BMT Stay	Child Age	Symptom Interfere	Cohesion	Express	Conflict	FSC	RSS Total	RSS Primary	RSS Second	RSS Diseng	Emotion Response
Child Age	-.04											
Symptom Interfere	-.21	-.12										
Cohesion	-.03	-.13	-.03									
Expressive	.16	.16	-.14	.22								
Conflict	-.19	-.04	.23	-.58**	-.30*							
FSC	-.25	.07	.07	-.37**	-.31*	.17						
RSS Total	-.13	.18	.00	-.14	.01	.27*	.39**					
RSS Primary	-.12	.30*	-.13	-.10	.17	.05	.27*	.78**				
RSS Second	.08	.15	-.15	.11	.13	.02	-.09	.70**	.43**			
RSS Diseng	-.24	-.03	.21	-.28*	-.22	.46**	.62**	.75**	.35**	.22		
Emotion Response	.14	-.01	-.06	.16	.23	-.10	-.23	-.04	.12	.01	-.16	
Behavior Response	.23	-.14	-.20	.36**	.36**	-.56**	-.42**	-.31*	-.07	.03	-.56**	.24

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

Table 16. Bivariate Correlations of Continuous Variables in the Model--Time 3

	BMT Stay	Child Age	Symptom Cohesion Interfere	Expressive Conflict	FSC	RSS Total	RSS Primary	RSS Second	RSS Diseng	Emotion Response		
Child Age	-.04											
Symptom Interfere	.05	-.13										
Cohesion	.01	-.04	.08									
Expressive	.19	.26	.10	.38**								
Conflict	.05	-.20	-.34*	-.49**	-.29*							
FSC	-.01	-.05	.12	-.42**	-.17	.26*						
RSS Total	.21	.13	.10	.01	.32*	.11	.32*					
RSS Primary	.24	.22	.09	.02	.29*	.03	.28*	.83**				
RSS Second	.14	.15	.01	.14	.38*	.01	.05	.84**	.56**			
RSS Diseng	.14	-.07	.14	-.23	.08	.26	.49**	.74**	.45**	.41**		
Emotion Response	-.04	.06	.03	.29*	.12	-.28*	-.62**	-.38**	-.30*	-.12	-.55**	
Behavior Response	.06	-.07	.14	.41*	.11	-.41**	-.54**	-.25	-.18	-.09	-.36**	.72**

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

Correlations among Independent Variables

Correlations between continuous independent variables were examined.

Significant correlations were found for the demographic interval variables, child age, parent length of stay in the hospital, and parent's symptom interference (See Tables 14 - 16, pages 140 - 142). Child age was positively correlated with primary engagement coping at the first two time points, which were prior to and during the parent's hospitalization (T1: $r = .26$, $p < .05$; T2: $r = .30$, $p < .05$), indicating greater use of this type of coping in older children versus younger children. Child age was negatively correlated with disengagement coping at Time 1 prior to transplant, indicating that greater use of disengagement coping was associated with younger children ($r = .31$, $p < .05$). Length of parent's hospital stay was significant and negatively correlated with Family Structural Change at T1 ($r = -.38$, $p < .01$), when children were uncertain about how long their parent would be hospitalized. Parent's symptom interference was associated with child coping and with children's perception of the family's response to the parent illness. A moderate positive correlation ($r = .40$, $p < .01$) between parent symptom interference and child disengagement coping at T1 was found, indicating an association between higher levels of child disengagement coping and heightened parental symptom interference prior to the parent's hospitalization. A moderate negative correlation ($r = -.34$, $p < .05$) between parent symptom interference and family conflict was found after the parent returned home post-transplant (T3), indicating that higher levels of symptom interference in the ill parent were associated with lower levels of family conflict.

Several significant correlations among the family interaction variables, cohesion, expressiveness, conflict, and family structural change were found at all three time points. An expected negative correlation between family cohesion and family conflict was found at all three time points, ranging from $r = -.39$ to $-.58$, $p < .01$, demonstrating a consistent inverse relationship between the two variables. Similarly, a consistent significant negative correlation between family expressiveness and family conflict was also found at each time point; correlations ranged from $r = -.29$ ($p < .05$ at T3) to $r = -.41$ ($p < .01$ at T1). Higher levels of family cohesion were associated with higher levels of family expressiveness after the parent returned home post-transplantation, $r = .38$, $p < .01$.

Family interaction variables were also correlated with different dimensions of coping. Significant negative correlations between family cohesion and disengagement coping (T1: $r = -.37$, $p < .01$; T2: $r = -.28$, $p < .01$) and significant positive correlations between family conflict and disengagement coping (T1: $r = .39$, $p < .01$; T2: $r = .46$, $p < .01$) were predominant at the initial two time points. However, post hospitalization (T3), only family expressiveness correlated significantly with the coping variables, that is, increased levels of family expressiveness were associated with increased levels of primary ($r = .29$, $p < .05$) and secondary coping ($r = .38$, $p < .05$). None of the family interaction variables significantly correlated with disengagement coping (See Table 15, page 143). Higher levels of family expressiveness were also found to be positively associated with child secondary coping prior to transplant (T1).

At each time point, the child's perception of change in the family, as measured by the Family Structural Change Questionnaire, was significantly correlated with one or more of the family interaction variables, cohesion, expressiveness, and conflict (See

Tables 14 - 16, pages 140 - 142). A consistently strong association between higher levels of family structural change and greater use of disengagement coping by children was highly significant at all three time points (T1: $r = .44, p < .01$; T2: $r = .62, p < .01$; and T3: $r = .49, p < .01$). Greater use of cumulative coping strategies, operationalized as total coping, was associated with children's heightened perceptions of family structural change. This was found during the parent's hospitalization (T2: $r = .39, p < .01$) and when the ill parent returned home (T3: $r = .32, p < .01$), however, this association was not significant prior to the parent's hospitalization.

Aim 2

Aim 2: To examine changes in the effects of all variables in the model, which include family structure and interaction, coping strategies used, and the level of adaptation across three time points in the BMT trajectory: (T1) pre-transplant and prior to hospitalization; (T2) approximately two weeks after the infusion of bone marrow/stem cells during hospitalization for the BMT; and (T3) one month after the transplant.

Repeated measures ANOVA models were used to examine the overall time effect comparing each time point to one another. Time was treated as a class (i.e., categorical) variable in the repeated measures ANOVA models so that there was no restrictive assumption that the trend over time was linear. Emotional distress and behavioral distress were modeled separately.

Repeated Measures to Examine Changes over Time

Prior to running repeated measures ANOVA on the independent and dependent variables, the assumption of sphericity had to be met. The assumption of sphericity is an additional assumption of repeated measures analyses; when the assumption is not met,

the F ratio statistic is not considered reliable. Sphericity is a term used to indicate that the equality of variances of the differences in scores between time points is not different (Field, 2005). To determine that the sphericity assumption was met, Mauchley's test was performed through an SPSS analysis on each of the univariate repeated measures analyses. The Mauchley's test statistic is significant ($<.05$) when there are significant differences in variance differences, indicating that the assumption was not met. When the sphericity assumption was not met, the Greenhouse-Geisser correction was used as a conservative adjustment for low sample sizes to produce a reliable F statistic. Preliminary analysis of the change over time for each variable was done (Tables 17 and 17a, pages 150 and 151). The protected version of Fisher's least significant difference test (LSD), which involves testing each of the pair-wise time differences only if the overall time effect is significant, was used to control Type 1 error. This approach appropriately controls the family-wise Type 1 error when the number of pair-wise differences is less than four.

Analysis for Change over Time for Each Variable using Repeated Measures

Table 16 indicates changes over time for both independent and dependent variables. The assumption of sphericity was met on all of the variables except parent symptom interference. Mauchley's test statistic was calculated at $p=.02$ for this variable, which, therefore, violated the sphericity assumption ($\chi^2(2df) = 15.48, p < .05$). Degrees of freedom were corrected using Greenhouse-Geisser estimates of sphericity ($\epsilon = .75$). Further examination of within-subjects effects with this correction indicated that the parent's symptom interference did change over the three time points, $F(1.56, 39.12) = 14.47, p = .023$. The high F statistic indicates that time had a strong effect on symptom

interference. Contrast comparisons between time points indicated that symptom interference was markedly different between T1 at pre-transplant and T2, during hospitalization ($F = 49.243, p < .001$). Symptom interference differed significantly from T2 to T3 (post-hospitalization) when it declined sharply ($F = 16.605, p < .05$). These findings were expected based on the effects of the treatment regimen (chemotherapy, radiation therapy, and subsequent immunosuppression) during transplantation that result in symptoms that occur immediately after treatment, such as nausea and vomiting, and those occurring a number of days and continuing weeks after treatment, such as fatigue, hair loss, and symptoms of infection, resulting from bone marrow suppression.

Results of repeated measures calculations comparing change in independent and dependent variables indicated that family interaction, as measured by the variables family cohesion, family expressiveness, and family conflict, were consistent and did not significantly change over time (Table 17a, page 151). However, the results indicate a main effect of time on family structural change, $F(2,106) = 3.921, p < .05$. No significant difference in the level of family structural change occurred between the pre-transplant (T1) and hospitalization (T2). However, pair-wise contrasts between T3 (post hospitalization) and these initial two time points were significant (T1 - T3: $F = 5.126, P < .05$ and T2 - T3: $F = 6.197, p < .05$), suggesting that increasing family change was consistently perceived by children prior to and during the parent's hospitalization, and then significantly lessened when the parent returned home.

Children's use of overall coping strategies, comprised of the total score of the three types of coping, was significantly different over time ($F(2,110) = 5.589, p = .01$). Use of combined coping strategies by children was highest prior to the parent's transplant

as indicated by the consistent downward decline in the change in means direction. Children's use of total coping strategies was significantly decreased at the subsequent two study time points (T1 - T2: $F = 6.148$, $p < .05$; T1 - T3: $F = 9.642$, $p < .01$) when compared to pre-transplant. Specific types of coping were examined; a significant main effect for time on two of the coping subtypes, secondary (acceptance, distraction, cognitive restructuring, and positive thinking) and disengagement (denial, avoidance, and wishful thinking) coping was found, each of the main effects dramatic as indicated by high F ratios ($F = 36.048$ and $F = 29.833$, respectively), indicating time had a strong effect on the change in these variables over time. Contrasts revealed that use of secondary coping strategies by children was significant and differed markedly from T1 to T2 ($F = 82.597$, $p < .001$), and then significantly increased from time T2 to T3 ($F = 26.968$, $p < .001$). A significant difference in the use of secondary coping strategies from T1 to T3 was noted. Based on contrast comparisons, time had a marked effect ($F = 50.879$, $p < .001$) on children's use of disengagement coping strategies as there was a marked decrease in use of disengagement by children from T2 ($M = 21.578$) to T3 ($M = 16.839$). Interestingly, children's use of disengagement coping strategies was significantly different from pre-transplant (T1) to hospitalization (T2) ($F = 27.481$, $p < .001$), indicating an increased use of this coping strategy while the parent was hospitalized. No significant differences were found in children's use of primary engagement coping, which includes problem solving, emotional regulation, and emotional expression, over the three time points in the study.

The effect of time on adaptation was examined using separate repeated measures ANOVAS to evaluate children's emotional and behavioral response to the parent's

transplant. A significant main effect of time on emotional response was found ($F(1,55) = 8.7, p < .001$). Follow-up contrast tests (Table 17, page 150) revealed that emotional response significantly changed from T1 ($M = 59.071$) to T3 ($M = 63.518$) as indicated by an F-ratio of 17.379, $p < .001$, and again from T2 ($M = 59.538$) to T3 ($M = 63.518$) as indicated by an F-ratio of 11.467, $p < .01$. Child emotional response from pre-transplant (T1) to hospitalization (T2) did not significantly increase and stayed consistent during between these time points. Similarly, behavioral response was also affected by time ($F(1, 55) = 9.141, p < .001$). Behavioral response significantly differed from T1, the pre-transplant period, ($M = 69.271$) to T2 ($M = 72.607$), as indicated by an F-ratio of 7.589, $p < .05$, suggesting an increase in more positive behavioral adaptation over these time points. A significant positive change in child behavioral response occurred when comparing the child's response at the pre-transplant period (T1) to the period when the parent returned home post-transplant ($M = 73.856$). No significant difference in children's behavioral response was found between T2, the parent's hospitalization, ($M = 72.607$) and time 3, post-hospitalization ($M = 73.856$).

Based on these results both child behavioral response and child emotional response consistently improved over the course of the parent's transplant. A greater change in more positive behavioral response in children was seen between the pre-transplant to hospitalization time points, with no significant change from the parent's hospitalization to their return home. Conversely, a significant and more positive change in children's emotional response occurred at the later two periods of the acute transplant trajectory.

Table 17

Repeated Measures Analyses of Variances Indicating Change Across Time: Outcome Variables and Coping Strategies

Variable	N	Means	F	Within subjects effects (p-value)
Behavioral Response	56		9.141	.000
Time 1 vs. Time 2		69.271 vs. 72.607	7.589	.008
Time 2 vs. Time 3		72.607 vs. 73.856	1.411	.240
Time 1 vs. Time 3		69.271 vs. 73.856	18.84	.000
Emotional Response	56		8.7	.000
Time 1 vs. Time 2		59.071 vs. 59.538	.136	.713
Time 2 vs. Time 3		59.538 vs. 63.518	11.467	.001
Time 1 vs. Time 3		59.071 vs. 63.518	17.349	.000
Total Coping	56		5.589	.005
Time 1 vs. Time 2		70.520 vs. 67.077	6.148	.016
Time 2 vs. Time 3		67.077 vs. 65.589	.916	.343
Time 1 vs. Time 3		70.520 vs. 65.589	9.642	.003
Primary Engagement Coping	56		1.801	.170
Time 1 vs. Time 2		22.286 vs. 21.089	.800	.375
Time 2 vs. Time 3		21.089 vs. 21.732	1.165	.285
Time 1 vs. Time 3		22.286 vs. 21.732	3.314	.082
Secondary Engagement Coping	56		36.048	.000
Time 1 vs. Time 2		30.181 vs. 23.767	82.597	.000
Time 2 vs. Time 3		23.767 vs. 27.661	26.968	.000
Time 1 vs. Time 3		30.181 vs. 27.661	9.372	.003
Disengagement Coping	56		29.833	.000
Time 1 vs. Time 2		18.054 vs. 21.578	27.481	.000
Time 2 vs. Time 3		21.578 vs. 16.839	50.879	.000
Time 1 vs. Time 3		18.054 vs. 16.839	4.535	.038

** Indicates that Mauchley's test is significant; therefore, the assumption of sphericity is violated. The Greenhouse-Geisser calculation was used.

Note: Higher mean scores for behavioral and emotional response indicate better response. Higher scores on other variables indicate higher levels.

Table 17a

Repeated Measures Analyses of Variances Indicating Change Across Time: Family Interaction Variables and Parent Symptom Interference

Variable	N	Means	F	Within subjects effects (p-value)
Family Cohesion	56		.098	.906
Time 1 vs. Time 2		16.107 vs. 16.173	.080	.779
Time 2 vs. Time 3		16.173 vs. 16.053	.186	.668
Time 1 vs. Time 3		16.107 vs. 16.053	.033	.856
Family Conflict	55		.791	.456
Time 1 vs. Time 2		11.800 vs. 11.526	1.044	.311
Time 2 vs. Time 3		11.526 vs. 11.491	.016	.898
Time 1 vs. Time 3		11.800 vs. 11.491	1.379	.245
Family Expressiveness	56		1.883	.157
Time 1 vs. Time 2		13.172 vs. 12.910	1.206	.277
Time 2 vs. Time 3		12.910 vs. 13.388	3.241	.077
Time 1 vs. Time 3		13.172 vs. 13.388	.846	.362
Family Structural change	54		3.921	.023
Time 1 vs. Time 2		13.926 vs. 13.963	.005	.945
Time 2 vs. Time 3		13.963 vs. 12.574	6.197	.016
Time 1 vs. Time 3		13.926 vs. 12.574	5.126	.028
Parent Symptom Interference**	26		14.474	.000
Time 1 vs. Time 2		30.308 vs. 39.025	49.243	.000
Time 2 vs. Time 3		39.025 vs. 31.500	16.605	.000
Time 1 vs. Time 3		30.308 vs. 31.500	.330	.571

** Indicates that Mauchley's test is significant; therefore, the assumption of sphericity is violated. The Greenhouse-Geisser calculation was used.

Note: Higher mean scores for behavioral and emotional response indicate better response. Higher scores on other variables indicate higher levels.

Aim 3

Aim 3: To examine the effects of the independent variables in the theoretical model as factors that impact the child's adaptation to the stress of a parent's BMT, as they are added hierarchically to the regression at each of the three time points.

Multiple regression analyses were run to answer the three questions below at each of the three time points for each of the dependent variables, behavioral and emotional response. The independent variables are those included in the model, page 21, Figure 1. These *preliminary* findings will be discussed in response to each of the three research questions. A brief description of these initial regression results will follow each aim. The final regression analyses will be described more extensively in the later section of this chapter.

Preliminary Findings

Hierarchical addition of the variables began with coping strategies added to the regression equation first. They were then followed by the entry of the family variables serving as measures of the perceived illness impact, and then by the contextual variables in the final equation representing the full model. A significance level of $p < .05$ was used to determine the statistical significance for the changes in R^2 as groups of variables were added to the regression model. Tables 18 - 23, pages 158 - 163, show the semi-partial correlation coefficients for independent variables in the model at each of the three time points. They indicate the impact of each independent variable in the model on the outcome variables, emotional response and behavioral response, controlling for the effect of each other independent variable.

Research question 3.1: What is the effect of children's coping strategies on adaptation, characterized as behavioral and emotional response?

Coping strategies were not found to be significant variables in children's *behavioral* response across the three time points as subsequent variables were added to the equation, although combined coping efforts did account for 9.4% to 27.4% of the total variance in the three models across the time points of the study (See Tables 18 - 20, pages 158 - 160). Disengagement coping was found to be initially significant in the regression at T1 ($r_{sp} = -.480, p \leq .01$) and T2 ($r_{sp} = -.593, p \leq .001$), but became nonsignificant when the perception of illness variables were added.

Coping strategies accounted for up to 36.6% of the total variance in children's *emotional* response as explained by the regression model across the three time points (Tables 21 - 23, pages 161 - 163). The use of secondary engagement coping strategies were initially significant and associated with more positive emotional response at T1, however, significance was lost as the perception of illness variables were added to the regression equation (Table 21). However, a more positive emotional response by children was significantly associated with children's use of less disengagement coping at T1 ($r_{sp} = -.520, p \leq .001$), which sustained its significance as other variables were added to the regression equation ($r_{sp} = -.248, p \leq .05$). At T3, less disengagement coping was also associated with more positive emotional response ($r_{sp} = -.434, p \leq .01$), however, its significance did not persist as other variables were added to the regression model.

Research question 3.2: What is the effect of the child's perception of the illness, including family structural change and family interaction, on adaptation controlling for coping strategies?

The contribution of the child perception of the illness variables to the explained variance for child *behavioral* response is indicated by the change in R^2 when they are added to coping strategies in the second step of the hierarchical regression models (Tables 19 - 20, pages 159 - 160). At the three time points the F ratios were as follows: T1: F-ratio = 5.920, $p = .001$; T2: F-ratio = 4.768, $p = .003$; T3: F-ratio = 4.140, $p = .008$. Family conflict and structural change in the family were both significant at T1 and T2, however, they did not continue to be significant at T3. At both T1 ($r_{sp} = -.308$, $p \leq .01$) and T2 ($r_{sp} = -.255$, $p \leq .05$) higher levels of family conflict and higher levels of structural change in the family were associated with more negative behavioral response in children.

The addition of the perception of illness variables to the coping variables in the regression equation for child *emotional* response significantly added to the total variance of the model at T1 (F-ratio = 2.881, $p = .034$) and T3 (F-ratio = 3.457, $p = .019$). Family structural change was the predominant independent variable contributing to the child's emotional response at these time points. Children's perception of a higher level of family structural change was consistently associated with poorer emotional response at T1 ($r_{sp} = -.274$, $p \leq .05$) and T3 ($r_{sp} = -.298$, $p \leq .05$), when controlling for the coping variables and the other perception of illness variables (Tables 21 and 23, pages 161 and 163). At T3, family structural change maintained its significance when controlling for the addition of child and parent variables to the regression equation. However, family structural change became an insignificant contributor to children's emotional response with the entry of the contextual variables (Equation 4) to the regression model at T1. The correlation between family structural change and the type of parental transplant was significant ($-.40$, $p < .01$;

Table 12, page 134) in correlation analysis which may have accounted for its loss of significance in the model.

In conclusion, addition of variables representing perceived illness impact (family structural change, cohesion, expressiveness and conflict) contributed to the explained variance in adaptation, as indicated by child emotional and behavioral response, with the exception of emotional response at T2. Family structural change and family conflict were the two primary significant variables contributing to the variance. When controlling for coping strategies, children's perceptions of higher levels of family structural change significantly contributed to a less positive child behavioral response at T1, and to a less positive child emotional response at T1 and T3. Higher levels of family conflict significantly contributed to less positive behavioral response at T1 and T2, controlling for coping strategies and other perception of illness variables.

Research question 3.3: What is the effect of the child's age and gender on adaptation controlling for the child's perception of illness impact and coping strategies?

Child gender and child age were not found to be significant contributors to child *behavioral* response at any of the three time points when entered into the regression equations (See Tables 18 - 20, pages 158 - 160). However, at T3 with the addition of the child variables, the semi-partial correlation coefficient value of family conflict became significant ($r_{sp} = -.282, p \leq .05$). Neither of the child variables were found to be significantly correlated with family conflict at this time (Table 11, page 134 and Table 16, page 142).

Entry of these variables into the regression equation at T1 and T3 decreased the total variance accounted for by the model, although this was not significant as calculated

by an insignificant F-ratio statistic for the change at both time points (T1: F-ratio = .916, $p = .41$; T3: F-ratio = .63, $p = .54$). However, at T2 the total variance accounted for by the model increased from 45.9 to 51.9 percent, which resulted in a significant change in the F-ratio (F-ratio = 3.480, $p < .05$).

Children's *emotional* response was not significantly associated with either child gender or child age at any of the three time points (Tables 21 - 23, pages 161 - 163). The entry of child age and child gender variables to the equation did not account for a significant increase in total variance at T1 nor at T3 (T1: F-ratio change = 1.167, $p = .32$; T3: F-ratio change = .416, $p = .66$).

Therefore, child age and child gender were found to minimally contribute to the explained variance in behavioral response controlling for the previously entered variables. A significant increase in total explained variance by the model for child behavioral response occurred at T2 with the addition of the child variables.

Research question 3.4: What is the effect of the recipient parent's gender, type of transplant, length of hospital stay, and symptom interference on adaptation controlling for the child's age and gender, the child's perception of illness impact, and coping strategies?

The parental contextual variables, recipient parent gender, transplant type, length of hospital stay, and symptom interference did not contribute significantly to the explained variance in children's *behavioral response* at time points 1 and 2. When these variables were added to the regression they decreased the total variance, although this change was not significant as calculated by the F-ratio (T1: F-ratio = .643, $p = .59$; T2: F-ratio = .440, $p = .78$). However, at T3 the addition of the parental variables accounted for a marginally significant increase of more than 12% in the model's total variance (F-ratio

= 2.657, $p = .055$). At T3, when the parent was home from the hospital recuperating from the transplant, recipient gender and transplant type both contributed significantly to the regression equation explaining children's behavioral response. When the mother was the transplant recipient, child behavioral adaptation was more positive. Children whose parents had an autologous BMT were found to have more positive behavioral adaptation than those children whose parents had an allogeneic transplant.

The addition of the parental contextual variables to the regression equations for children's *emotional* response had little effect on the variance accounted for by the model. At T1, there was an increase in variance of 1% and at T3 there was a decrease in total variance of 2%. Both of these accounted for an insignificant change in the model's total variance (T1: F-ratio = 1.242, $p = .31$; T3: F-ratio = .780, $p = .55$). Transplant type was the only parental contextual variable significantly associated with children's emotional response, although this was at T2 when the R^2 , or explained variance for the total model, was not significant. At the time of the parent's hospitalization, parental autologous transplant, when compared to parental allogeneic transplant, was significantly associated with a more negative emotional response in children (Table 22, page 162).

The addition of the parental contextual variables to the model contributed little to the total variance of the model with the exception of explaining child behavioral response at T3. Recipient gender (mother) and transplant type (autologous) were the only parent contextual variables that were significantly associated with child adaptation, when controlling for other variables in the model.

TABLE 18 Regression with Model Variables: Time 1 Behavioral Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Contextual Factors (n=49)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.004	.058	.012	.055
Secondary Engagement Coping	.153	.011	.027	.032
Disengagement Coping	-.480**	-.157	-.084	-.143
Family Structural Change		-.250*	-.263*	-.206*
Cohesion		.133	.154	.107
Expressiveness		.025	.024	.013
Conflict		-.308**	-.305**	-.324**
Child Gender (female)			.024	.012
Child Age			.144	.126
Recipient Gender (female)				.083
Transplant Type (autologous)				.056
Symptom Interference				.118
BMT Stay				.075
R²	.226**	.462***	.459***	.447**

*** p ≤ .001 ** p ≤ .01 * p ≤ .05
Standard errors are in parentheses

TABLE 19 Regression with Model Variables: Time 2 Behavioral Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Contextual Factors (n=48)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	.082	.018	.039	.046
Secondary Engagement Coping	.081	-.022	.024	.047
Disengagement Coping	-.593***	-.094	-.138	-.103
Family Structural Change		-.081	-.057	-.080
Cohesion		.068	.025	.024
Expressiveness		.176	.203	.183
Conflict		-.255*	-.264*	-.260*
Child Gender (female)			.101	.101
Child Age			-.199	-.187
Recipient Gender (female)				.126
Transplant Type (autologous)				.049
Symptom Interference				.065
BMT Stay				.019
R²	.274**	.459***	.519***	.489***

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 20 Regression with Model Variables: Time 3 Behavioral Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Contextual Factors (n=40)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.166	-.099	-.080	-.188
Secondary Engagement Coping	.163	.012	.035	.093
Disengagement Coping	.240	.016	-.010	.107
Family Structural Change		-.160	-.164	-.214
Cohesion		.211	.164	.146
Expressiveness		-.090	-.015	.063
Conflict		-.251	-.282*	-.323*
Child Gender (female)			.049	.109
Child Age			-.142	-.137
Recipient Gender (female)				.315*
Transplant Type (autologous)				.273*
Symptom Interference				-.067
BMT Stay				.114
Adjusted R²	.094	.329*	.313*	.437*

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 21 Regression with Model Variables: Time 1 Emotional Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Contextual Factors (n=49)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.065	.022	.018	.060
Secondary Engagement Coping	.247*	.094	.107	.103
Disengagement Coping	-.520***	-.375*	-.234*	-.248*
Family Structural Change		-.274*	-.253*	-.194
Cohesion		.104	.093	.061
Expressiveness		.096	.063	.072
Conflict		-.040	-.046	-.064
Child Gender (female)			-.150	-.130
Child Age			.034	.019
Recipient Gender (female)				.155
Transplant Type (autologous)				.117
Symptom Interference				.045
BMT Stay				.107
Adjusted R²	.366***	.457***	.461***	.474**

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 22 Regression with Model Variables: Time 2 Emotional Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Contextual Factors (n=48)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	.165	.227	.217	.292*
Secondary Engagement Coping	.062	-.070	.064	-.138
Disengagement Coping	-.273	-.126	-.129	-.113
Family Structural Change		-.122	-.119	-.141
Cohesion		.194	.181	.300*
Expressiveness		.064	.066	.127
Conflict		.173	.168	-.304*
Child Gender (female)			.000	.147
Child Age			-.027	.042
Recipient Gender (female)				.167
Transplant Type (autologous)				-.365*
Symptom Interference				.196
BMT Stay				-.224
R²	.019	.032	-.018	.177

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 23 Regression with Model Variables: Time 3 Emotional Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Contextual Factors (n=40)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.182	-.077	-.063	-.123
Secondary Engagement Coping	.258	.067	.082	.108
Disengagement Coping	-.434**	-.189	-.204	-.130
Family Structural Change		-.298*	-.300*	-.332*
Cohesion		-.007	-.034	-.039
Expressiveness		.110	-.142	.163
Conflict		-.144	-.168	-.182
Child Gender (female)			.035	.057
Child Age			-.102	-.092
Recipient Gender (female)				.152
Transplant Type (autologous)				.146
Symptom Interference				.017
BMT Stay				.089
Adjusted R²	.342***	.483***	.464**	.448**

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

Final Regression Analyses

Regressions were rerun without the inclusion of the independent variable, recipient symptom interference. This was done because a) there was a significant amount of missing data from the recipient parent regarding his/her symptoms which caused the number in the sample to be lower for the preceding regressions, and b) because recipient symptom interference was not found to be significant in explaining the variance of child emotional or behavioral response across the time points of the study. Eliminating symptom interference from the regression analysis increased the number of examined cases from 49 to 59 at T1, from 48 to 59 cases at T2, and from 40 to 56 cases at T3. Imputation of the mean symptom interference score was not done because of the variability of scores by participants at each time point, and because of the individual subjective evaluation of how a participant may assess interference in daily life activities caused by symptoms. According to Tabachnick and Fidell (2001, p. 65), “deletion of a variable is acceptable as long as that variable is not critical to the analysis.”

When symptom interference was excluded from regressions on children’s *behavioral* response at T2 (adj. $r^2 = 43.1$) and T3 (adj. $r^2 = 30.8$), the total variance accounted for by the model declined. However, at T1, when symptom interference was excluded from the model, the explained variance increased by 3%. The total explained variance in *emotional* response across time points decreased when symptom interference was excluded from the model by 5% at T1, by 9% at T2, and by 13% at T3, although at this time point the variance explained by the model was not significant. Inclusion of recipient symptom interference in the regression models may therefore have contributed to the inaccuracy of the total explained variance of the models because the regressions were based on smaller sample sizes.

When regressions were rerun without the variable symptom interference (not shown), the significance of individual variables was relatively the same at each time point when compared to the regressions run when symptom interference was included in the data set. The only contextual variable which became significant in the regressions when symptom interference was not included was child gender, which became significant in the regression for emotional response at T1 ($p = .022$). Child age and BMT length of stay were not significant, and therefore, were not included in the final regression equations.

Final Regression Variables

Two contextual variables that were significant in the initial regressions and the regressions run when symptom interference was removed from the data set were used in the final regressions. These variables were: child gender and type of transplant. These variables were added to the regressions together in the third step of the regression. Parent gender was added because it was a significant variable in the preliminary regressions when symptom interference was included. Because it was not likely that this variable would be significant, it was entered in the regression equation last. Tables 24 through 29 show the final regression analysis as seen on pages 174 - 180. Separate regressions were run for each of the dependent variables indicating adaptation-- behavioral and emotional response.

Research question 3.1: What is the effect of children's coping strategies on adaptation, characterized as behavioral and emotional response?

Coping strategies accounted for 8.4 to 31.7% of the total variance of child *behavioral* response in Equation 1 of the model across the three time points, and was significant at T1 and T2 (Tables 24 - 26, pages 174 - 176). For behavioral response in this

set of regressions, there was an increase in total variance of 5% from T1 to T2, and from T2 to T3 there was a drop of 23%.

In the final regression equations, disengagement coping was the only coping strategy (T1: $r_{sp} = -.534$, $p \leq .01$, T2: $r_{sp} = -.593$, $p \leq .001$, T3: $r_{sp} = -.311$, $p \leq .05$) that was significantly associated with behavioral response. At T1 and T3, disengagement coping became insignificant when the perception of illness variables were added, and two of the perception of illness variables, family structural change and family conflict, were significant. However, at T2, disengagement coping retained its significance when these perception of illness variables and child and parent variables were added. In previous correlation analysis (Tables 14 - 15, pages 140 - 141), disengagement coping was found to be correlated significantly with three of the four perception of illness variables, family structural change (T1: $r = .44$, T2: $r = .61$), family conflict (T1: $r = .39$, T2: $r = .46$), and family cohesion (T1: $r = -.37$, T2: $r = -.28$). The moderate to strong significant correlations of disengagement coping with these variables may account for the loss of disengagement's significance in the model when these variables were added.

Coping accounted for 2% to 32% of the model's total explained variance in *emotional* response across the time points. At T2, the variance explained by the model was insignificant. Disengagement coping was a significant contributor to the explained variance in emotional response only at T1, and retained its significance at T1, pre-transplant, upon the addition of other variables across the model to the regression equation (Table 27, page 177). At T3, the post-hospitalization period, disengagement coping lost its significance when the perceived illness impact variables were added, and the variable, family structural change, became significant ($r_{sp} = -.298$, $p \leq .05$).

In summary, disengagement coping was associated with both behavioral and emotional response in children when not controlling for the effects of the other independent variables, except at T2 when it was not significantly associated with child emotional response. However, when the other independent variables were added to the regression incrementally according to the model, disengagement coping retained a significant impact throughout all steps of the regression analysis at T1 with emotional response as the dependent variable ($r_{sp} = -.271, p \leq .01$) and at T2 with behavioral response as the dependent variable ($r_{sp} = -.215, p \leq .05$).

Research question 3.2: What is the effect of the child's perception of the illness, including family structural change and family interaction, on adaptation controlling for coping strategies?

Examination of the correlation matrix for moderate to high inter-correlations among the family variables, family structural change, family cohesion, family conflict, and family expressiveness, was initially done to screen for multicollinearity, as well as for the strength of the association of these variables with the independent outcome variables. Bivariate correlations were all below .42. Statistical methods to assess for multicollinearity were done. The variance inflation factor (VIF), which is an indicator of the linear relationship of one predictor variable to another, and the tolerance statistic which is the reciprocal statistic, $1/VIF$, were used to assess multicollinearity. The VIF statistic was below 10, and the tolerance statistic was greater than .2 for each predictor variable indicating there were no problems with multicollinearity (Bowerman & O'Connell, 1990; Menard, 1995). Therefore, hierarchical regressions were run which

included each of these variables in the analyses to test their individual contributions to the explained variance in the dependent variables in the regression equations.

The contribution of the child perception of illness variables to the explained variance in *behavioral* response, while controlling for the effects of coping strategies was significant (Tables 24 - 26, pages 174 - 176). At T1, an increase from 26.7% to 49.1% in total variance with the addition of the perception of illness variables was significant (F-ratio = 7.040, $p = .000$); at T2, an increase of total variance from 31.7% to 40.5% (F-ratio = 3.028, $p = .026$) was significant; and at T3, an increase in total variance from 8.4% to 27.4% (F-ratio = 6.557, $p = .004$) was significant. The significant change in total variance accounted for by this set of variables indicates the important contribution of these variables to the model.

Family conflict and family structural change were the primary variables which contributed significantly to explaining the variance in child behavioral response across the three time points. Specifically, at T1 and T2, family conflict contributed significantly to behavioral response ($r_{sp} = -.274$, $p \leq .01$, $r_{sp} = -.255$, $p \leq .05$, respectively) controlling for the coping variables, and then retained its significance with the subsequent addition of parent and child variables to the regression equation (Tables 24 - 25, pages 174 - 175). Family structural change was also a significant contributor to behavioral response pre-transplant ($r_{sp} = -.226$, $p \leq .05$), and when the parent returned home ($r_{sp} = -.274$, $p \leq .01$), and retained its significance with the addition of subsequent child and parent variables to the model.

The addition of the child perception of illness variables explaining child *emotional* response at T1 increased the total variance by only 3%, which was not a

significant change (F ratio = 1.658, $p = .074$). At T2, the variance explained by the model became non-significant and the total variance declined to less than 1%. However, at T3 a large significant increase from 29% to 42% in the model's total variance for emotional response occurred with the addition of the perception of illness variables (F -ratio = 3.906, $p = .008$), suggesting the importance of these variables to the overall fit of the model once the parent returned home.

Of the four variables characterizing children's perception of the illness, family structural change was the only one associated with emotional response. At both T1 and T3, higher levels of family structural change were significantly associated with less positive *emotional* response in children when controlling for coping and other perception of illness variables (T1: $r_{sp} = -.223$, $p \leq .05$; T3: $r_{sp} = -.356$, $p \leq .01$). Family structural change became insignificant when the next set of variables, child gender and transplant type, were entered into the regression equation at T1, however, at T3 its significance was retained when controlling for other variables. The effect of the addition of the perception of illness variables to the regression equation had a significant effect on disengagement coping. At T1, the significance of the semi-partial correlation coefficient decreased from $-.524$, $p \leq .001$ to $-.296$, $p \leq .01$, despite retaining its significance when controlling for all other variables in the regression equation (Table 27, page 177). At T3, the addition of the perception of illness variables to the regression equation caused disengagement coping, which had previously been significant, to lose its significance (Table 28, page 181).

In summary, the addition of the perception of illness variables (family structural change, cohesion, expressiveness and conflict) contributed significantly to the explained variance of the regression equation at all of the time points for behavioral response,

however, for emotional response this was true only at T3. Family structural change was consistently significant in its association with child behavioral response at all three time points, and with emotional response at T1 and T3 (See Tables 24 - 29, pages 174 - 179). Family conflict was also significantly associated with behavioral response at T1 and T2, however, it was not significant at T3. It was also not associated with children's emotional response at any time point.

Research question 3.3: What is the effect of the child's age and gender on adaptation controlling for child's perception of illness impact and coping strategies?

Child age was not included in the final regression analysis as previously explained in the introduction to this section on page 165, because it was not significant in the preliminary regressions. The addition of child gender and the parent's transplant type to the regression equation explaining child *behavioral* response did not result in significant changes in the total explained variance at any of the three time points (T1: F-ratio = .014, ns; T2: F-ratio = 1.117, ns; T3: F-ratio = .000, ns).

At T1, the addition of child gender and transplant type to the regression equation significantly added to the total variance explaining child *emotional* response (F-ratio = 3.38, $p = .042$) when variance increased from 35.2% to 40.7%. Change in total variance was not significant at the other two time points (T2: F-ratio = 2.711, ns; T3: F-ratio = .038, ns). Female child gender was a significant variable ($r_{sp} = -.263$, $p \leq .05$) associated with a less positive emotional response prior to transplant (T1); autologous transplant type was significantly associated ($r_{sp} = -.294$, $p \leq .05$) with a less positive emotional response during the parent's hospitalization (T2). With the addition of these two variables at T1, the semi-partial correlation of family structural change decreased its association

with emotional response and became nonsignificant. Semi-correlation coefficient values for other variables in the regression equation were not significantly changed.

In conclusion, the addition of child gender and transplant type did not significantly affect the total explained variance in emotional response accounted for by the model, with the exception of T1 when it explained an additional 5.5% of the total variance of child emotional response. Female child gender (T1) and autologous transplant (T2) were significantly associated with child emotional response, but they were not associated with child behavioral response. Both of these variables retained their significance with the addition of the final variable, recipient gender, to the regression equation.

Research question 3.4: What is the effect of the recipient parent's gender, type of transplant, length of hospital stay, and symptom interference on adaptation controlling for child's age and gender, the child's perception of illness impact, and coping strategies?

The variables, length of hospital stay and symptom interference, were not included in the final regression analysis as explained in the introduction to this section on page 167. Type of transplant was included in Research question 3.3 due to its entry into the equations at the third step of the regression (see explanation on page 165). Therefore, recipient gender will be discussed relative to its effect on adaptation controlling for the effects of all other variables in the final model.

The addition of recipient gender to the regression equations did not significantly change the variance accounted for by the model at any of the time points for either child emotional response or for child behavioral response. The F-ratio change statistic for child behavioral response when adding recipient gender to the regression equations at each of

the three time points are as follows: T1 = .243, ns; T2 = .000, ns; and T3 = 2.645, ns. The F-change ratios for emotional response when adding recipient gender to the regression equations are as follows: T1 = 1.769, ns; T2 = .309, ns; and T3 = 1.361, ns. Inclusion of the variable, recipient gender, made little impact on the explained variance as indicated by either no change or an insignificant change in the semi-partial correlation coefficients of the significant variables (family conflict, disengagement coping, and family structural change) associated with child adaptation, as characterized by both behavioral and emotional response.

Overall, the final regressions indicated that the revised model, which excluded the nonsignificant contextual child and parent variables (child age, parent gender, symptom interference, and parent length of hospital stay), accounted for 27 to 46% of the explained variance in child behavioral response, and accounted for 5.6% to 41% of the explained variance in child emotional response. The model was not significant for child emotional response during parental hospitalization, T2. This was consistent with the initial regressions when the symptom interference variable was included, except for a decrease in the total variance accounted for by the final model (27%) when compared to the preliminary model (43.7%) on child behavioral response at T3 (Tables 20 and 26, pages 160 and 176). Variables consistently contributing to child behavioral response across time points were family structural change, family conflict, and disengagement coping. Higher levels of change in the family, higher levels of family conflict, and higher levels of disengagement coping were predominant factors associated with poorer behavioral response in children. Significant variables associated with emotional response in children over the three time points were not consistent and varied by time point. At T1, female

child gender and use of more disengagement strategies were associated with less positive child emotional response. At T3, when the parent was at home, higher levels of family structural change were associated with less positive emotional response. During the parent's hospitalization, T2, a less positive emotional response in children was associated with the parent having an autologous BMT.

TABLE 24 Final Regression for Time 1 Behavioral Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Significant Contextual Factors (n=59)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	.038	.057	.057	.065
Secondary Engagement Coping	.101	.014	.016	.012
Disengagement Coping	-.534**	-.175	-.167	-.171
Family Structural Change		-.226*	-.210*	-.204*
Cohesion		.174	.171	.172
Expressiveness		.031	.027	.040
Conflict		-.274**	-.275**	.266**
Child Gender (female)			.010	.002
Transplant Type (autologous)			.012	.006
Recipient Gender (female)				.047
R²	.267**	.491***	.470***	.462**

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 25 Final Regression Analyses for Time 2 Behavioral Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Significant Contextual Factors (n=59)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	.078	.042	.012	.012
Secondary Engagement Coping	.116	.052	.048	.047
Disengagement Coping	-.586***	-.208**	-.215*	-.215*
Family Structural Change		-.084*	-.095	-.094
Cohesion		.011	.011	.011
Expressiveness		.089	.085	.084
Conflict		-.237*	-.229*	-.229*
Child Gender (female)			.132	.131
Transplant Type (autologous)			.066	.061
Recipient Gender (female)				.001
R²	.317**	.405***	.408***	.416***

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 26 Final Regression Analyses for Time 3 Behavioral Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Significant Contextual Factors (n=56)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.056	-.037	-.034	-.045
Secondary Engagement Coping	.084	-.039	-.039	-.013
Disengagement Coping	-.311*	-.011	-.012	.021
Family Structural Change		-.284*	-.284*	-.309*
Cohesion		.109	.100	.115
Expressiveness		-.040	-.034	-.010
Conflict		.214	-.216	.223
Child Gender (female)			.001	.056
Transplant Type (autologous)			.033	.110
Recipient Gender (female)				.187
R²	.084	.274*	.244*	.270*

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 27 Final Regression Analyses for Time 1 Emotional Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Significant Contextual Factors (n=59)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.087	-.037	-.027	-.003
Secondary Engagement Coping	.261	.132	.165	.153
Disengagement Coping	-.524***	-.296**	-.259*	-.271**
Family Structural Change		-.223*	-.175	-.162
Cohesion		.096	.082	.087
Expressiveness		.073	.025	.062
Conflict		.037	.011	.025
Child Gender (female)			-.263*	-.222*
Transplant Type (autologous)			-.015	.035
Recipient Gender (female)				.133
R²	.321***	.352***	.407***	.416***

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 28 Final Regression Analyses for Time 2 Emotional Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Significant Contextual Factors (n=59)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	.193	.197	.235	.239
Secondary Engagement Coping	-.032	-.105	-.135	-.116
Disengagement Coping	-.237	-.067	-.076	-.073
Family Structural Change		-.112	-.072	-.079
Cohesion		.081	.139	.140
Expressiveness		.113	.131	.121
Conflict		.046	.131	.126
Child Gender (female)			.039	.049
Transplant Type (autologous)			-.294*	-.243*
Recipient Gender (female)				.071
R²	.023	.007	.070	.056

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

TABLE 29 Final Regression Analyses for Time 3 Emotional Response

Semi-partial Correlation Coefficients on Coping Strategies, Family Structural Change, Family Interaction, and Child and Recipient Significant Contextual Factors (n=56)

	Equation 1	Equation 2	Equation 3	Equation 4
Primary Engagement Coping	-.121	-.088	-.070	-.077
Secondary Engagement Coping	.164	.027	.031	.047
Disengagement Coping	-.475***	-.186	-.192	-.168
Family Structural Change		-.356**	-.352**	.366**
Cohesion		-.035	-.041	-.031
Expressiveness		.066	.053	.068
Conflict		-.074	-.075	-.080
Child Gender (female)			-.093	-.053
Transplant Type (autologous)			.055	.101
Recipient Gender (female)				.120
R²	.290***	.420***	.409***	.413***

*** p ≤ .001 ** p ≤ .01 * p ≤ .05

CHAPTER 5

DISCUSSION

This chapter begins with background information on the bone marrow transplant trajectory to enhance the discussion of the findings. Discussion of the empirical findings by study aims is then provided, along with a discussion of the theoretical, research, and clinical significance of the results. The chapter concludes with the limitations of the study and recommendations for future research.

Background

The use of a longitudinal design in this study requires that findings be examined within the context of events occurring at each of the individual time points and in the overall trajectory of the BMT. Therefore, it is important to be aware of the contextual characteristics of each time point. Below is a brief description of each time point with an emphasis on contextual influences which may aid in the interpretation of the findings.

It is important in this discussion to reiterate that prior to bone marrow transplant, families are well aware of the ill parent's diagnosis and are likely to know that the prognosis may not necessarily include a cure. The ill parent and the family have generally experienced the effects of chemotherapy to treat the malignancy, although the duration of the experience is variable, based on diagnosis and response to treatment. Because BMT is primarily used in clients who have been previously treated for a malignancy and have either not responded to standard therapy or who have had the malignancy recur, it is often considered unique from standard chemotherapy treatment in that it offers, for many, a final hope for cure.

An intense period of preparation for transplant occurs prior to the treatment at Time 1. Before hospitalization, the patient undergoes a series of medical tests, chemically induced immunosuppression to prepare the stem cells for harvest, and multiple days of having stem cells collected at the hospital, or, for allogeneic recipients, the testing and waiting that accompanies searching for a matched donor. Additionally, anticipation for the transplant is heightened by home preparations for the ill parent's absence, medical insurance approval for the transplant, and the knowledge of the acute and challenging nature of the treatment. Prospective transplant recipients are often in daily contact with the transplant center during the two weeks prior to transplant. Children in the family are aware of these events and witness the heightened activity and accompanying anxiety in the household, as the family anticipates and prepares for the treatment.

During the parent's hospitalization for transplant (T2), the ill parent is separated from the family. The well-parent is much less available to the children due to being at the hospital with the ill parent or maintaining family and parental responsibilities. Children may or may not see the ill parent during hospitalization; this depends upon the parent's condition, the distance of the family from the hospital, and other factors which may interfere. This is also a period when the parent is undergoing treatment and then recuperating from the immunosuppressive complications that may occur from treatment, which can make visitation with the family less than optimal.

Time 3, one month after the BMT, is a time of transition for the family. The family has made preparation for the parent to return home and is now attempting to re-group and assimilate the parent back into the family. The ill parent is often very fatigued, physically fragile, requires care by family members, remains prone to infection, and often

requires the care of a visiting nurse. Re-hospitalization for complications often occurs. Maintaining or refining family changes to accommodate the rehabilitation of the ill-parent continues. Financial burden and uncertainty regarding the success of the BMT as a “cure” surfaces and may provide further stress. This period, too, is one of waiting for the ill parent to regain function, and frustration can occur when this does not happen as quickly as the family or parent anticipates. Some ill parents are unable to return to work or find that they may need to change their occupation due to physical limitations.

Discussion of the Empirical Findings by Aim

Aim 1: To examine the bivariate relationships among all variables in the proposed model at each time point. The strength of these relationships will then determine which variables will be considered for inclusion in the regression models with adaptation measured by behavioral response and emotional response, as the dependent outcomes.

Categorical variables based on ANOVA

Based on ANOVA, the categorical independent variables, child gender, parent gender, transplant type, and child age by category, were not associated with behavioral response. In previous studies, associations of these variables with behavioral problems in children of parents with cancer have been minimal (Osborn, 2007). Significant associations, however, were found between some of the categorical study variables and children’s *emotional response* to parental transplantation.

Two of the categorical variables, child gender and transplant type, were associated with children’s emotional response. The emotional response of girls was more negative than boys at time 1, prior to the parent’s transplant. This is consistent with

conclusions derived from previous research findings in children of parents with cancer. Overall, studies have shown that girls, especially adolescent girls, have been found to have higher levels of emotional distress when compared to boys (Compas et al., 1994; Nelson & While, 2002; Visser et al., 2005).

Findings from studies on children's response to parental cancer indicate that specific medical variables, such as diagnosis and treatment type, are not associated with child psychosocial function (Hoke, 2001; Visser et al., 2005; Watson et al., 2006). However, in this study, the parent's type of transplant was significantly associated with children's emotional response during parental hospitalization (T2). Children of autologous transplant recipients had significantly more negative emotional response than children of parents undergoing an allogeneic transplant. This finding may be attributed to both methodological and theoretical differences. Almost all studies on children's response to parental cancer have been cross-sectional, not examining the longitudinal changes in emotional and behavioral response, as was done in this study, nor distinguishing the influence of hospitalization and parental separation that may be significant for these findings. Second, based on the initial ANOVA and correlation analyses, children of autologous transplant recipients, reported significantly higher levels of family structural change prior to the parent's BMT which may have contributed to the heightened level of family conflict and the overall level of child emotional distress while the parent was hospitalized (T2).

It is noteworthy, also, that children of parents undergoing allogeneic BMT had a more positive emotional response than children of autologous recipients during hospitalization. This was an unexpected finding in that allogeneic transplantation is

generally agreed upon to be a more complex treatment with higher risk of patient mortality and complications, and a longer duration of hospital stay. It could be surmised that allogeneic transplantation may result in less emotional distress in children because it is often done shortly after diagnosis or recurrence of a hematologic malignancy, while autologous transplant is often done after a longer period of the time, and follows ineffective, conventional treatment. Failure of previous treatment has been found to heighten distress in adolescents of mothers with breast cancer (Huizinga et al., 2004). Higher levels of emotional distress in children of autologous transplant recipients, when compared to children of allogeneic transplant recipients, may therefore be due to more pre-transplant exposure to parent treatment and symptoms. Higher levels of symptom interference reported by autologous recipient parents prior to transplantation support this finding. Second, higher levels of family conflict reported by children of autologous transplant recipients, when compared to children of allogeneic recipients, and the use of increased disengagement coping by these children prior to their parent's transplant (T1), suggests that there may be heightened emotional distress in these families.

Surprisingly, significant differences in adaptation were not found between younger and older children in regard to emotional and behavioral response. Many previous studies have found that adolescents were the most vulnerable group of children (Armistead, Klein, & Forehand, 1995; Compas et al., 1994; Pedersen & Revenson, 2005). However, lack of a clear distinction between age groups and inconsistency in the use of age groups among researchers has been noted. Therefore, it is difficult to compare these results to those of other studies.

Bivariate associations among variables

Continuous contextual variables

The significant association between the continuous contextual variables, child age and parent's symptom interference, and other independent variables in the model were primarily in the expected direction. Prior to and during parental BMT, the use of primary engagement coping, which includes problem solving, emotional regulation, and emotional expression strategies, became more frequent as children's age increased. This was consistent with findings in the literature associating children's increased use of purposeful active strategies to manage parental illness with age (Compas et al., 1996; Sears & Sheppard, 2004), and is plausible, given the increasing cognitive ability of children as they mature to discern the severity and implications of the parent's illness and therefore to effectively respond.

Higher levels of symptom interference were associated with an increased use of child disengagement coping prior to transplant. Although there were no data upon which to measure the duration of children's exposure to parental symptom interference prior to transplant and exposure to the anxiety of a cancer diagnoses and failed treatments, it could be surmised that children who have been exposed to their parent's symptoms and resulting inability to perform usual activities may be using disengagement strategies as a way to manage their stress. This could be supported by the finding that children of autologous transplant recipients, whose parents often have undergone extensive previous chemotherapy, were found to have used a higher level of disengagement coping at T1. In contrast, when the parent returned home from the BMT, increased parent symptom interference was associated with a lower level of conflict in the family, suggesting that

the family unit may have attempted to support the ill parent by keeping the environment tranquil.

Perception of illness variables

Significant relationships between several of the key independent study variables and adaptation were found. Over the course of the BMT transplant trajectory, children's perception of illness, as measured by family structural change, family cohesion, family conflict, and family expressiveness, was moderately to strongly associated with child adaptation. All of these variables were significantly associated with child *behavioral* response at each time point. Children's emotional response was associated with individual perception of illness variables at varying times during the transplant trajectory, and will be discussed below. Consistent with prior research findings, aspects of a positive family environment, which include the family's structure, cohesion, and communication, were found to be associated with better child adaptation (Harris & Zakowski, 2003; Huizinga et al., 2003; Watson et al., 2006).

Children's perception of a high level of change in the family due to the parent's illness was strongly associated with more negative emotional and behavioral responses. In other studies, the impact of role changes, illness demands, and disruption of routines due to a family member's illness have been associated with increased psychosocial distress in children (Davey et al., 2005; Hilton, 2000; Watson et al., 2006) as indicated by bivariate correlations. Prior to transplant, greater perceived family change by children of autologous transplant recipients was associated with their parents' self-report of heightened symptom interference. At that same time period, these children reported greater use of disengagement coping than children of allogeneic transplant recipients. The

association of family structural change, symptom interference, and disengagement coping with one another and their effect on more negative emotional adaptation in children of autologous BMT recipients prior to their parent's transplant is warranted and is an area for future exploration. This finding also points to the importance of a longitudinal design.

At all time points, higher levels of family cohesion were associated with more positive child behavioral adaptation. Positive emotional adaptation was associated with higher levels of family cohesion prior to transplant and during the parent's recovery. These findings were consistent with those of numerous previous studies examining both child and family members' adaptation to parental and sibling cancer (Horowitz & Kazak, 1990; Huizinga et al., 2003; Kazak & Barakat, 1997). Watson (2006), in a recent study using a similar sample in terms of size and age-range, reported that poorer family cohesion predicted internalizing and externalizing distress when the mother had breast cancer. Harris and Zakowski (2003) found that a lower level of family cohesion was associated with symptoms of depression and anxiety in adolescent children of parents with cancer. Therefore, in planning an intervention, an important consideration may be the inclusion of a component to support and reinforce family cohesion during the BMT trajectory.

Children's perceptions of higher levels of family conflict were consistently associated with more negative child behavioral response at all time points. A higher level of family conflict was also associated with more negative emotional response when the parent was at home recovering after the transplant. This finding is consistent with the only other study in the literature examining family conflict and parental cancer, a cross-

sectional study which found that adolescent depression was associated with higher levels of family conflict (Harris & Zakowski, 2003).

Significant associations of higher levels of perceived family conflict with decreased family cohesion and with greater family structural change were found once the parent was hospitalized (T2) and continued when the parent was recovering at home. Associations among these variables did not occur prior to the parent's transplant, and it may be that prior to transplant, their effects were not fully realized. Perhaps, once the parent was hospitalized and treatment started, family changes became more apparent and noticed by the child, resulting in the child's changed perception of the family environment. It may also be that family cohesion was impaired as a result of the increased changes and demands on the family, and family conflict heightened. Further exploration of the relationships among these variables is warranted.

Higher levels of family expressiveness were associated with more positive child behavioral response at all time points and with more positive emotional response prior to the parent's transplant. These findings were consistent with those of Harris and Zakowski (2003) who also used the Family Environment Scale to assess family expressiveness. They found that family expressiveness was associated with decreased anxiety and depression in adolescents of parents with cancer. Other studies have primarily examined children's one-to-one communication with parents and to a lesser extent the family's communication, and found an association between higher levels of expressiveness and less psychobehavioral distress in children (Huizinga et al., 2005; Watson et al., 2006).

Coping variables

With the exception of the association of primary engagement coping strategies and child behavioral response at T3, primary and secondary engagement coping strategies were not associated with child adaptation. However, disengagement coping was significantly associated with one or both child outcome variables, emotional and behavioral response, across the BMT trajectory. It is noteworthy that disengagement coping was associated with more negative behavioral adaptation at each time point, and with more negative emotional adaptation of the child prior to transplant and when the parent returned home. While disengagement coping has been associated with more negative adolescent emotional and behavioral adaptation to a variety of stressors, including parental illness (Compas et al., 2001), the consistent and strong relationship of this association during parental transplantation is a new finding which warrants the need for further examination in view of intervention planning.

Perception of higher levels of family change was significantly associated with children's increased use of disengagement coping strategies by children. Findings from a previous study that investigated the association of children's perception of their parent's cancer and children's use of coping strategies found that children primarily perceived their parent's illness as being influenced by their parents and the medical staff, and believed that they had little personal influence on its course (Compas et al., 1996). This perception was associated with use of fewer engagement coping strategies, such as problem solving, and increased use of disengagement strategies, including avoidance by children. However, numerous significant family changes may also signal family instability that may instigate disengagement strategies. In view of the lack of normalcy in

the family due to numerous day-to-day changes and the uncertainty of the future, disengagement strategies focused on maintaining personal and routine activities in an attempt to preserve normalcy may become the focus of the child's effort. It is not merely that children want to insulate themselves from the uncertainty posed by the situation, but that they actively seek out ways to maintain normalcy in their everyday lives.

With regard to engagement coping, differences in the use of different types of engagement coping strategies were associated with child age group, the parent's gender, and when they were used during the course of the parent's transplant. Throughout the transplant trajectory, the use of more primary and secondary engagement coping strategies was noted when the ill parent was the father. The association of children's use of more coping when the father was ill has not been found in previous studies, perhaps because almost half of the published studies have examined families facing maternal cancer (Osborn, 2006), and few have examined coping strategies. Mixed findings have not conclusively associated parent gender with psychosocial distress, although a recent study (Visser et al., 2005) reported that children had higher levels of psychosocial problems adapting to parental cancer when the father was ill. These findings indirectly support the findings of this study which implies that children may use increased coping measures when the father is the ill parent due to heightened emotional distress. In the context of BMT, longer hospitalization necessitated by the transplant treatment, may have influenced children to become more engaged in actively coping in supporting the care-giving mother, who may be viewed by children as in need of more assistance and emotional support than fathers who are caregivers.

In this study, older children, ages 15 - 18, when compared to younger children, ages 10-14, used more primary engagement coping strategies targeted at altering the events impacted by their parent's transplant or controlling their emotional response during their parent's hospitalization. This finding was consistent with findings of previous studies that older aged adolescents used more coping strategies to purposefully change the stressful situation and regulate their emotional response as compared to younger children (Compas et al., 1996; Weisz et al., 1994). Increased adolescent cognitive ability, resulting from developmental maturation, along with the increased ability to appraise a stressor and exert control has been associated with this finding.

A higher level of family expressiveness was associated with children's increased use of engagement coping strategies and with more positive emotional adaptation when the parent returned home after the transplant. This may reflect heightened attempts by the family and its individual members to communicate in solving problems, altering daily tasks, and in planning family matters to accommodate the recuperation of the ill parent. These associations may have implications for future potential intervention.

Summary

Findings from these analyses indicate that two groups of children may have heightened vulnerability to maladaptation at specific time points in the BMT trajectory. Girls, when compared to boys, had more negative emotional response prior to their parent's BMT. Second, children of autologous transplant recipients, when compared to children of allogeneic transplant recipients, may be more vulnerable to adaptation difficulties. Findings from this study indicate that these children perceived their families as having higher levels of conflict and undergoing greater family change than children of

parents receiving allogeneic transplants. Increased levels of symptom interference reported by autologous transplant recipients may also add to their children's distress and impact adaptation.

Results from the initial aim of this analysis indicated that there were significant associations in children's emotional and behavioral response with how the child perceived the parent's illness. Higher perception of change in the family's functioning as a result of the parent's transplant was positively associated with less positive emotional and behavioral adaptation in children at all time points. Higher levels of family conflict were associated with more negative child behavioral adaptation at all time points, and with more negative emotional adaptation when the parent returned home post-transplant. Disengagement coping was consistently moderately to strongly associated with more negative child behavioral response throughout the parent's transplant trajectory. Although the perception of illness variables and disengagement coping were the variables most consistently associated with emotional and behavioral response, significant bivariate associations between the perception of illness variables, coping strategies used by children, and the interruption caused by parent symptoms were found at different time points.

Only one of the study variables was significantly associated with children's emotional response during the parent's hospitalization (T2). More negative emotional adaptation at that time was associated with being a child of a parent undergoing autologous transplantation. Children's emotional adaptation was not related to the contextual variables, the family variables, nor the coping variables in the model. The lack of significant associations leads one to consider what other family, coping, or personal

influences, outside of those in this study, may be associated with how the child was emotionally responding. The parent's hospitalization is a unique time point because the child is physically separated from the parent and is primarily relying on information s/he obtains from others to make an appraisal of the ill parent's condition. Family variables not tested in this study, personal attributes of the child such as self-esteem, resilience, and the perception of external support may be other constructs which should be explored in the future for their influence on child emotional response while the parent is hospitalized for the transplant.

Aim 2: To examine changes in the effects of all variables in the model, which include family structure and interaction, coping strategies, and the level of adaptation across three time points in the BMT trajectory: (T1) pre-transplant and prior to hospitalization; (T2) approximately two weeks after infusion of bone marrow/stems cells during hospitalization for the BMT; and (T3) one month after the transplant.

Changes in the emotional and behavioral function of children occurred over the course of the acute phase of the transplant included in this study. Changes in children's emotional and behavioral response from time 1, before their parent's hospitalization, when compared to time 3, one month after transplant, were substantial as indicated by the large F-ratios. Findings indicated that children had more positive function, as demonstrated by increased levels of emotional and behavioral adaptation, post-transplant when the parent had returned home, and when family changes, while still present, had lessened. This is an expected finding in that the return of the ill parent to the home, and his/her presence, despite altered functioning, represented a partial return to "normalcy" and therefore, more positive emotional and behavioral responses. Previous studies have

associated a sense of “normalcy” in the family with family function, adaptation, and stability (Hilton, 2001; Houldin, 2007; Huizinga et al., 2003; McPherson, 2003).

Children’s behavior significantly improved during the parent’s hospitalization (T2) and when the ill parent was experiencing his/her highest level of reported symptom interference. This may have resulted from children’s heightened awareness of the negative effects of the treatment on the ill parent and on other family members, causing children to consciously regulate their behavior to prevent further disruption and stress during an already stressful time. One child, age 15, whose father was the ill parent, mentioned to the researcher that he was “trying not to get my mom upset—I’m trying to protect her.” Conversely, better behavior may also be attributed to children’s detachment from the situation, as indicated by significantly greater use of disengagement coping at this time than prior to transplant.

Children’s emotional adaptation was slower to change when compared to their behavioral adaptation. Emotional adaptation was consistent at T2 with that prior to transplant. A more positive change in children’s emotional state did not occur until after the transplant, between T2 and T3, when the imminent danger of the BMT may have lessened and an expectation of parent survival and family stability was renewed. When the parent returned home, children commented that they were “less scared now than before”. However, several children indicated they were still “worried the cancer will come back” or worried because the ill parent was not functioning as they had anticipated.

Stress levels related to children’s perception of greater change in family interaction were similar pre-transplant to levels during the parent’s hospitalization. This is an expected finding given changes that occur in preparing for the parent’s absence and

the additional duties and roles taken on by others. A significant decline in stress related to family structural change was noted after transplant when the parent returned home. This does not imply that changes in the family did not occur when the parent was at home. It is recognized that the intensity of the treatment results in slow recuperation and often limits the function of the ill parent. Children's perception of family change as less stressful at this time may be due to the child's view of the family as back to normal due to the presence of the ill parent in the home, rather than to the functional ability of the ill parent. The findings reinforce that pre-transplant intervention relative to expectations may be effective in helping families adjust and cope with both anticipated and unanticipated changes.

Children's perception of family interaction (cohesion, conflict, and expressiveness) did not significantly change over the course of the study time points. This was interesting in that children perceived significant change in their individual day to day living situation related to their parent's illness, as measured by the Family Structural Change Scale (FSCS). Despite the recognition of change in both the family and their own personal function within the family, as measured by the Response to Stress Questionnaire, their view of family interaction across the BMT trajectory remained relatively stable. A possible explanation for the incongruence of these findings may be that children did not associate changes in family interaction with stress related to family change. This may be too advanced an association for younger children to make. It may also be that children perceived the patterns in family life to be relatively stable over time even during the midst of change and crisis. Further research to study children's perceptions and their possible impact on children's appraisal of the threat of the parent's

illness and its subsequent effect on child resilience and coping, is needed. Second, the Family Environment Scale, used to measure family cohesion, conflict, and expressiveness must be considered. The scale uses a “True/False” response to items, which may not be as sensitive to detecting subtle variations in these variables thereby influencing the results of the analysis.

Types of coping used by children changed significantly over the course of the study. Children’s effort to actively change the situation or alter their response to it (primary engagement coping) did not vary significantly over the study. This may be due to their awareness throughout the study trajectory that their ability to impact the parent’s illness was beyond their capability. Disengagement coping changed significantly from one time point to another. A substantial increase from T1 to T2, and a substantial decrease from T2 to T3 indicated that use of disengagement coping peaked during the parent’s hospitalization, when the parent’s symptom interference and family change were also at their highest levels. At that same time, children’s efforts to accept the situation (secondary engagement coping) were at the lowest. An association between the increased use of disengagement coping and the decreased use of secondary engagement coping strategies was not found statistically significant in this study. However, increased use of disengagement coping strategies, such as wishful thinking, avoidance, and denial, may result from the infrequent or lack of interaction the child has with the ill parent due to hospitalization. Second, the increased level of parent symptom interference during hospitalization may have fostered further uncertainty about the parent’s outcome, thus reinforcing the child’s use of disengagement coping. The decreased use of secondary engagement coping methods which include positive thinking, cognitive restructuring, and

acceptance may be suppressed by the child's uncertainty and fear, and further complicated by the child's inability to cognitively understand the situation. The child's need for an alternate way to maintain psychosocial stability may influence the higher use of disengagement coping at this time. What is more important theoretically, however, is that coping strategies changed from time point to time point. This finding supports results of a longitudinal examination of children's coping with parental cancer by Helseth and Ulfasaet (2003), which concluded that children's coping strategies contextually vacillated, and that children employ various strategies to "come in and out" of a situation in an effort to adapt to change.

Summary

Based on the results of the analysis, the time prior to the parent's hospitalization may be the most difficult time for children when both emotional and behavioral distress are highest. Anticipation of the parent's hospitalization and treatment is increased at this time, as plans and arrangements are being made to maintain family routines, child care, and individual family members' needs. This may be an optimal time for an intervention to reduce fears, provide information, and encourage the use of more constructive engagement coping strategies during the transplant trajectory which have been associated with better psychological adjustment in children (Compas et al., 2000). Second, intervention for the family at this time may prepare families and children to cope more effectively with the stress and family change during the parent's transplant and recovery periods.

Aim 3: To examine the effects of the independent variables in the model as factors that impact the child's adaptation to the stress of a parent's BMT as they are

added hierarchically to the regression at each of the three time points. (The final regression analyses, Tables 23 - 28, will be used as a basis for this discussion of findings.)

The complexities of examining children's coping with stress and the small number of studies examining coping in children with parental cancer have provided limited findings (Compas et al., 1996; Huizinga et al., 2005a; Issel, Ersek, & Lewis, 1992). In this study, disengagement coping was the primary type of coping that significantly contributed to child adaptation at all time points. Further, use of disengagement coping was associated with negative outcomes for the child. Prior to the parent's BMT, use of more disengagement coping was significantly associated with higher levels of emotional distress. During the parent's hospitalization, higher levels of the use of disengagement coping by children were consistently associated with more negative behavioral response as each group of variables was added to the regression model. Use of disengagement coping during the parent's hospitalization was very high when compared to the pre-transplant and post-transplant periods, as per repeated measures analysis, and was associated with more negative child behavioral adaptation as indicated by the regression. It may have been much easier for the child to disengage from the situation at this time due to physical distance from the parent. At the two other time points, behavioral response at T1 and T3, and emotional response at T3, disengagement coping was initially significant and then became nonsignificant with the addition of the perception of illness variables into the regression. The strong positive association of disengagement coping with the child's perception of the illness variables, especially with family structural change and family conflict, may have contributed to the lack of

sustained significance of disengagement coping in accounting for the variance in child adaptation. Future exploration of the association of these variables may be helpful in understanding their relationship and the effects they exert on one another and the outcome variables.

The child's perception of the illness within the context of the family environment was significant in regressions with adaptation as the outcome. Considering the perception of illness variables, higher levels of family conflict and family structural change were the most significant independent variables associated with behavioral response and with emotional response across the transplant trajectory, with the exception of emotional response at T2. Prior to transplant, children's heightened perceptions of family structural change and family conflict due to the additional disruption within the family in preparation for the BMT was understandable, and provides contextual support for children's increased psychosocial distress at this time point. The association of disruption and discord in the family with changes in the functioning of the family would be expected, although this has only marginally been tested in prior research (Nelson, Sloper, Charlton, & While, 1994; Huizinga et al., 2003; Watson et al., 2006). Children's perception of family structural change was not significantly associated with child emotional and behavioral adaptation while the parent was hospitalized (T2). The impact of family changes on adaptation may have lessened as the family and child were more confident in their ability to function during the parent's absence. Higher perceived levels of family conflict, which was significantly associated with more negative adaptation pre-transplant, did not significantly increase or decrease from the pre-transplant period, but continued to remain significantly associated with more negative behavioral adaptation

during the parent's hospitalization. The ongoing influence of family conflict during the parent's hospitalization was not surprising, especially if the conflict interfered with the support of family members for one another, or if it added to the overall disruption in the household.

Stress related to children's perceptions of change in the family was the only variable significantly associated with child adaptation when the parent returned home after the transplant. Family conflict was not significantly associated with adaptation, nor the use of disengagement coping by children. During this period, the family transitions back to having the parent at home, recovering previous structure and roles, while implementing further changes to accommodate the rehabilitation of the ill-parent. Although the amount of family structural change was significantly less than during the parent's hospitalization and at pre-transplant, as indicated by repeated measures analysis, higher levels of family structural change were significantly associated with both negative emotional and behavioral adaptation. It is expected that children who perceive stressful family changes related to the parent's return home would be more likely to be behaviorally and emotionally upset.

Family cohesion and family expressiveness were not significantly associated with adaptation in the regression analysis, although both were positively associated with emotional and behavioral adaptation in the preliminary correlation analysis. As in this study, a significant association between family cohesion and expressiveness and child adaptation was found by Harris and Zakowski (2003), however, in further analysis they found that family expressiveness mediated the association of family cohesion and child anxiety. Family cohesion has also found been found to be associated with behavioral

response in children of breast cancer patients (Watson et al., 2006) in multivariate analysis. In this study, the low level of reliability of the measure, as indicated by alpha 0.45, may contribute to the lack of significance of family expressiveness. A different instrument may be more sensitive to obtaining results that reflect findings which concur with those of other studies. It is problematic that presently there is a dearth of instruments for children with strong reliability and validity that measure expressiveness in the family. Two previous studies of child adaptation to parental cancer have reported family cohesion to be significantly associated with child emotional response and behavioral response (Harris & Zakowski, 2003; Watson et al., 2006).

The contextual child and parent variables contributed least to the variance explained by the model. Female child gender contributed to emotional adaptation only prior to transplant (T1), significantly associated with more negative emotional adaptation for girls. Previous studies in children of parents with cancer have also found girls to be more emotionally distressed (Nelson & While, 2002; Visser et al., 2005; Watson et al., 2006). Child age was not significantly associated with child adaptation in this study, as has been found in several previous studies that have found older adolescent children have more emotional distress related to parental cancer (Compas et al., 1994; Heiney et al., 1997). In this study, the sample of older *adolescent* children may have been too small to detect significant age differences.

In terms of the parental variables, few were found to be significantly associated with children's adaptation. Parent's symptom interference and the length of their initial hospitalization were not significant variables at any time point. Findings from previous studies examining an association between parental symptoms and children's psychosocial

adaptation have been inconsistent. Younger children seem to be more distressed by physical symptoms in the parent (Christ, 1993), and more negative psychobehavioral adaptation in adolescents has been associated with parental anxiety and depressed mood (Lewis & Darby, 2003; Watson et al., 2006). The lack of significance of parental symptom interference on children's adaptation in this study may be attributed to several factors. Children in this study were not as young as those studies which reported child distress associated with exposure to parents' physical symptoms. Second, in this study parental fatigue, loneliness, boredom, pain, and isolation were assessed and reflect only aspects of the parent's mood and anxiety; a definitive assessment of anxiety and depressed mood was not included in this analysis. Third, unlike previous studies that report symptoms by frequency and severity, parental symptoms in this study were viewed in the context of how they affected the daily functioning of the ill parent (interference), which may not accurately represent other characteristics of symptoms that children perceived. Fourth, during BMT hospitalization (T2), when the ill parent's symptom interference was highest, children had little exposure to the parent.

Given that children of autologous transplant recipients had significantly higher levels of negative emotional response prior to the parents' transplant (T1) and had more negative emotional response and perceived higher levels of family conflict than children of allogeneic transplant recipients at T2, may indicate the need for heightened assessment and potential intervention targeted at this group. As previously stated, this was an unexpected finding and suggests that health care practitioners cannot assume families and children of autologous recipients are less affected by the stress of BMT. This finding may provide insight to health care practitioners who view allogeneic recipients and their

families as being more vulnerable to psychosocial distress than autologous recipients and their families, due to the longer trajectory and increased mortality and complications associated with allogeneic transplant.

Theoretical Implications

Based on the findings of this study, there is preliminary support for the theoretical model proposed in Chapter 1. Significant associations were found between several of the independent variables and the dependent variables, emotional and behavioral adaptation. At each time point, the child's perception of the illness, operationalized as family cohesion, family expressiveness, family conflict, and family structural change were significantly associated with both emotional and behavioral adaptation. Family conflict and family structural change were two of the three significant variables that were most frequently associated with adaptation in regression analyses. These findings provide evidence for the importance of the family component in the child's perception of the overall impact of the illness on their personal life. More importantly, they reflect the theoretical basis of the study, which calls attention to the interrelatedness of the family and its members during times of stress, and views children's adaptation within the context of the family.

The contribution of coping to the theoretical model is marginal as indicated by the findings of this study. The significance of disengagement coping and primary engagement coping strategies at various time points in the model provides additional support for considering coping as a processual variable integral to children's adaptation to stress. Future work in understanding how disengagement coping and family variables influence one another will be necessary in building a more complete model. Because

knowledge about coping in children is limited, these findings will add to the theoretical basis of understanding how children cope. Second, based on the findings of these analyses, coping in children is a protective and fluid process, as described by Helseth and Ulfsaet (2003), in which children intermittently use strategies to either face the parent's illness or to displace it to maintain a sense of stability. This has implications for how coping may be viewed in future theoretical frameworks applied to children's adaptation to parental illness.

Future study of significant relationships among variables in the model is needed if we are to more clearly understand the adaptation of children within families experiencing the crisis of serious illness. Exploring the addition of other variables to the model that may increase the explanatory power of the model and result in a more complete understanding of children's adaptation is needed. The findings of these analyses will most likely warrant future revisions of the model and model testing to strengthen its' theoretical applicability. There is also a need to explore why the theoretical model did not significantly explain children's emotional adaptation during the parent's hospitalization (T2). Children's comments, such as "I was worried—he was swollen everywhere" and having "trouble falling asleep at night...thinking about things" indicated that emotional upset was present. The measure used to assess children's emotional response may have lacked sensitivity for emotions that may be more prevalent at this time point when the parent was hospitalized and undergoing the transplant. A qualitative study at this time point (T2) to explore factors which may contribute to children's emotional stability would be helpful and may clarify specific contextual variables that are unique during the parent's hospitalization.

Research Implications

Other than a family study which provided a preliminary narrative synthesis, rather than analyzed results on children of parents undergoing BMT (Zabora et al., 1992), this study is unique in that it is the only study to provide measurable findings on the psychosocial adaptation of children during parental BMT. Previous studies of children responding to parental illness and parental cancer have primarily been cross-sectional. While they have provided important findings, they have not afforded examination of relationships over time and they have been less likely to provide evidence of significant relationships that promote increased understanding of children's adaptation (Pedhazur & Schmelkin, 1991). Although longitudinal designs are hindered by attrition and cost (Babbie, 2004; Polit & Beck, 2008), these barriers did not pose significant problems for this study in that the study was "nested" in a larger funded family study and attrition was minimal. Based on the longitudinal findings of this study, an intervention to promote children's adaptation prior to parental transplantation would be likely to contribute to minimizing secondary psychosocial morbidity.

Replication of this study in larger samples at other transplant sites which differ in characteristics from a university-based facility are needed to verify these results. The continuation of longitudinal studies that use mixed methods, qualitative and quantitative, and that include multiple reporters, parent and child, are recommended to substantiate the validity of findings. In addition, significant variables in this model could be further studied for their association with one another and for refining a more accurate and inclusive model that may better explain children's adaptation. Researchers can use the

findings of this study as a basis for future descriptive research comparing differences in adaptation of this population to children of parents with cancer or other serious illnesses.

Clinical Implications

Findings from this study suggest that children's adaptation to parental BMT may be supported by intervention early in the transplant trajectory. Findings suggest that children report more negative emotional and behavioral adaptation pre-transplant than during the transplant period and following the transplant. Anticipatory fear for the parent's well-being and cure, and anxiety related to the unknown circumstances that surround transplant may be sources of children's distress. In their comments to the researcher at the pre-transplant data collection, children expressed concern about "whether this will work." From the findings, it was relatively clear that children felt the parent's transplant and the events surrounding it were externally controlled, that they were sensitive to and reported stress related to changes and disruption in the family, and that they viewed the family unit's interaction as unchanged during the transplant trajectory. Based on the findings of the study that family change, family conflict, and child disengagement coping contributed significantly to child emotional and behavioral adaptation, it may be efficacious to help children through efforts focused on the family that include specific age-appropriate interventions directed specifically toward children. It can't be assumed that families who have been exposed to parental cancer prior to coming to BMT are coping well based on previous intervention, or because they have well-tested or refined coping mechanisms. In fact, children from families that come to transplant having experienced family conflict and turmoil due to the parent's illness may be especially vulnerable. Assessment of family characteristics prior to transplant would

be useful so that potentially vulnerable children and families could be targeted for preventive interventions.

Previous intervention studies which have measured the effects of intervention on children of parents with acute illness are few, although many intervention clinical programs have been implemented (Spath, 2007). Studies which have measured psychosocial functioning in children have demonstrated that interventions are significant in reducing psychosocial distress. Currently, in children of parents with cancer there is an increased recognition that intervention work is in its infancy, and the best way to deliver interventions to children is still being discussed and needs to be tested (Lewis, 2007). Embedding an intervention for children within a family intervention is proposed to be efficacious in making parents aware of children's distress, and in facilitating both child and family communication and coping within the context of the family (Bugge, Helseth, & Derbyshire, 2008).

Parents in this study confided their concern for their children's psychosocial well-being. Parents were not directly questioned, but at times would comment prior to and/or after their child's telephone interviews. A lack of communication with the child or an inability to discern "how the child is doing" was the predominant concern expressed by the parent. An intervention designed for the pre-transplant period that is family focused, and that also includes specific interventions for children, may be an important preventative measure.

Although expressiveness was not found to be a significant variable in the regression analyses of this study, it should be considered when planning an intervention for several reasons. First, other studies examining children's adaptation to parental cancer

have found communication and expressiveness to be significant factors in reducing psychosocial distress (Harris & Zakowski, 2003; Huizinga et al., 2005; Watson et al., 2006). Second, based on the correlation analysis for this study, higher levels of family expressiveness were significantly associated with decreased family structural change and decreased family conflict, which were both found to be significant variables contributing to child emotional and behavioral response across the transplant trajectory. Third, and most importantly, family expressiveness may be a modifiable characteristic, therefore, an intervention to encourage, educate, and facilitate communication and expressiveness among family members, including children and parents, may be helpful in promoting more positive adaptation through this intense period.

In conclusion, this study indicates that an intervention which begins prior to parental transplant, and that uses a method of screening children, as well as families, for their vulnerability to emotional and behavioral distress may be efficacious. Children who perceive their family as already experiencing high levels of conflict and increased stress related to family change, or those who previously have used disengagement as a predominant way of coping, could be targeted for perhaps more intense intervention. Because these variables were significantly associated over the three time points with adaptation, an intervention that is ongoing across the BMT trajectory is optimal.

Limitations

Several limitations influence the outcomes of the study and its generalizability. Two primary limitations exist. The study design was not based on an experimental design, and therefore, it cannot be assumed that the independent variables in the regression models have a “causal” effect on the dependent variables which characterize

child adaptation, that is child behavioral and emotional response. The use of a non-random sample and the inability to manipulate independent variables in this study does not meet the criteria of testing for causation as occurs in an experimental design. Also, there are admittedly potential bi-directional relationships of some of the variables with one another, such as family conflict and child behavioral response. Second, the sample was a limitation of the study in that it was a non-random sample from one site, which was composed of primarily Caucasian children, a majority between the ages of 10 and 14 years of age, and which contained a disparate number of boys over girls in the older age group (15 - 18 years of age). Second, although intra-class correlation testing was done to determine whether the assumption of independence of children's responses was violated when there was more than one child participant in a family, and although the intra-class correlations were not significantly different from zero, the violation of the assumption cannot be definitively ruled out due to the small sample size, and the limited power of this analysis, as previously discussed in the Analysis section. A study done at multiple sites in various geographical areas, and which used only one child from each family, may have allowed findings to be more generalizable to the larger population. The sample in this study may therefore not accurately represent children of parents undergoing BMT at other sites.

The instruments used in this study were not developed for the specific context of this study, but were adapted for use in this population. Two of the instruments, the Family Environment Scale (FES) and the Child Health Questionnaire (CHQ-87), have been used in numerous studies of children, including studies of children of parents with cancer (Harris & Zakowski, 2003; Nelson & While, 2002; Watson et al., 2006). The

Response to Stress Questionnaire (RSQ) has been used less frequently in previous studies. The RSQ has been used to examine adolescent coping in response to economic stress and family conflict, physical illness, interpersonal stress, and parental depression with Cronbach alphas ranging from .72 to .89 for the three subscales (Wadsworth et al., 2005; Schrier & Chen, 2008; Jaser et al., 2005; Wadsworth et al., 2004). Although this scale has good reliability in children, it has been used to examine children's coping with parental illness in only one study, and has not been used to examine coping in children of parents with cancer or who are undergoing BMT. To adapt the scale to this population, questions were framed to assess coping strategies used within the context of the parent's transplant. The second measure, the Family Structural Change Scale, was composed of a subset of preliminary items from the RSQ and items from the CHQ-87 that addressed family function. Items were also adapted to assess the child's perception of illness-related family change and the stress the child experiences as a result. This instrument is limited in that it had not been used in previous studies

Second, low reliability on the Expressiveness Subscale of the Family Environment Scale threatens internal consistency. Findings from previous studies suggest that family expressiveness and communication may be important contributing factors to child adaptation, and further study of these variables is advocated (Osborn, 2007). The results from this study, however, must be interpreted in light of instrument limitations.

Because participants need to understand questions in order to respond to the instrument items, cognitive deficiencies due to developmental differences must be considered as a possible limitation that may have been a threat to the accuracy of the data. This was controlled by using one interviewer over the course of the study who

assessed children during the interview for lack of understanding and intervened when needed to ensure that the child comprehended the questions. However, it cannot be assumed that these methods totally controlled for misinterpretation of the items and the accuracy of children's responses.

Potential threats to internal validity were parent attrition and repeated testing. Lack of report of symptom interference by some parents at all time points was significant, and attrition may have threatened the validity of the conclusions on this variable (Shadish, Cook, & Campbell, 2002). At T1, 14% of the parent sample did not report symptom interference, at T2, 11% did not report symptom interference, and at T3, 28% of the parent sample did not report symptom interference. Imputation of data for these subjects was not done because of the potential variability of their responses. Therefore, insufficient participant data on this variable may threaten statistical power leading to a Type II error, and thus inaccurate findings.

Testing is a potential threat to the internal validity of the study due to the repeated testing of the same population at three time points, the first two time points, in some clients, may have been as close as 2 weeks apart. However, this limitation could not be controlled due to the sequence of the transplant trajectory, and the importance of obtaining data at critical time points throughout the transplant trajectory.

Study Strengths

The primary strength of this study is that it is the first study examining children's response to parental BMT, which is a highly challenging treatment with significant ramifications for the family unit as well as for the individual member. The conceptual model is based on well-known and time-honored theories of family systems and stress

and coping, and takes into account child development. A strength of this study is the use of a strong observational study design (longitudinal) which is likely to have fewer potential biases than found in other designs, such as a retrospective design which suffers for example from recall bias. Additionally, a low level of attrition, the scant amount of missing data from child participants, and having only one researcher collect all the child data, which eliminates variability due to the interviewer, support the reliability of the findings. The use of a strong observational study design (longitudinal) which is likely to have fewer potential biases than found in other designs, such as recall bias which occurs in retrospective design, is a strength of this study. Additionally, a low level of attrition, the scant amount of missing data from child participants, and having only one researcher collect all the child data, which eliminates variability due to the interviewer, support the reliability of the findings. The strength of the findings related to children's emotional and behavioral adaptation and the family, contextual, and coping variables influencing adaptation provide information for future research and clinical interventions.

Conclusions and Recommendations

The primary purpose of this study was to increase understanding of how children adapt to the stress of parental bone marrow transplantation based on a conceptual model that incorporated context, perception of illness, and coping strategies. Significant associations among the variables in the model with emotional and behavioral adaptation were found. Differences based on child gender, parent gender, child age, and transplant type were found in several steps of the analyses. Based on the regression analysis, the key variables most strongly associated with adaptation were family structural change, family conflict, and disengagement coping. Increased levels of these factors were significantly

associated with more negative child adaptation. Further testing of the association of family structural change, family conflict, and disengagement coping in children is recommended so that the relationships of these independent variables to one another can be determined. This may necessitate revision of the model.

Children's adaptation consistently improved over the course of the parent's transplant trajectory. The child's perception of the family environment, however, remained consistent and did not change significantly over the course of the transplant, despite significant changes in the family which produced stress. Children's use of different coping strategies varied over the transplant trajectory, with secondary coping and disengagement coping showing the greatest change from pre-transplant to their parent's hospitalization. These findings suggest that the processual nature of coping in children facing parental BMT is an important factor to consider in this population and may provide information on how children cope with other stressors.

With the exception of children's emotional response during their parent's hospitalization, the model accounted for approximately 40% of the variance in children's adaptation across the BMT trajectory. Specifically, regression analyses indicated that the model did not explain children's emotional adaptation at the time of the parent's hospitalization. This will require further investigation, and may be a starting point for future model refinement.

The findings suggest that an intervention for children that is embedded in a family intervention may be optimal. Intervention may include screening children prior to transplant for their vulnerability to emotional and behavioral distress based on the findings of this study. Although bone marrow transplantation has become more widely

used for acute malignancies, it continues to be considered an extremely intensive and life-threatening therapy that affects both patients and families. This study heightens our awareness and increases our knowledge regarding the experience of children as their parents undergo BMT. Based on this analysis, interventions can be created and implemented to support children during this critical time.

APPENDIX A: INSTRUMENTS

Child Behavioral Response Scale

These questions ask you about how you are doing. Certain questions may sound alike but each one is different. Some questions ask about problems you may not have. That's great, but it's important for us to know. Please answer each question. There are not right or wrong answers. If you are unsure how to answer a question, give the best answer you can. All of your answers will be kept private.

During the past 2 weeks, how often did each of the following statements describe you?

1. How often did you act too young for your age?
Very often Fairly often Sometimes Almost never Never
2. How often did you argue with others?
Very often Fairly often Sometimes Almost never Never
3. How often did you have a hard time paying attention?
Very often Fairly often Sometimes Almost never Never
4. How often did you not do what your parent asked you to do?
Very often Fairly often Sometimes Almost never Never
5. How often did you want to be alone?
Very often Fairly often Sometimes Almost never Never
6. How often did you lie or cheat?
Very often Fairly often Sometimes Almost never Never
7. How often did you have a hard time getting others to like you?
Very often Fairly often Sometimes Almost never Never
8. How often did you feel clumsy?
Very often Fairly often Sometimes Almost never Never
9. How often did you run away from home?
Very often Fairly often Sometimes Almost never Never
10. How often did you have speech problems, for example, like stuttering?
Very often Fairly often Sometimes Almost never Never
11. How often did you steal things from your home?
Very often Fairly often Sometimes Almost never Never

12. How often did you steal things from outside your home?
Very often Fairly often Sometimes Almost never Never
13. How often did you act mean or moody if you did not get what you wanted?
Very often Fairly often Sometimes Almost never Never
14. How often did you get really mad when you did not get what you wanted?
Very often Fairly often Sometimes Almost never Never
15. How often did you find it hard to be with others?
Very often Fairly often Sometimes Almost never Never
16. How often did you have trouble getting along with others?
Very often Fairly often Sometimes Almost never Never
17. How often did you feel like things were out of control?
Very often Fairly often Sometimes Almost never Never
18. How often did you talk with one of your parents about your sick parent's illness?
Very often Fairly often Sometimes Almost never Never

Items 1-16 taken from the Child Health Questionnaire CF-87 (Landgraf and Ware, 1966)
Behavior Subscale, section 5.1, "Getting Along"

Child Emotional Response Scale

I am going to read you a list of questions about children's moods and feelings and ask you how much of the time they are true for you. I will ask you to tell me whether each question is true for you: all the time, most of the time, some of the time, a little of the time, or none of the time. There are no right or wrong answers. If you are unsure how to answer a question, give the best answer you can. All of your answers will be kept private.

During the past 2 weeks, how much of the time did you?

1. Feel sad?
All of the time Most of the time Some of the time A little of the time None of the time
2. Feel like crying?
All of the time Most of the time Some of the time A little of the time None of the time
3. Feel afraid or scared?
All of the time Most of the time Some of the time A little of the time None of the time
4. How much of the time did you worry about things over the past two weeks?
All of the time Most of the time Some of the time A little of the time None of the time
5. How often did you feel lonely?
All of the time Most of the time Some of the time A little of the time None of the time
6. How often did you feel unhappy?
All of the time Most of the time Some of the time A little of the time None of the time
7. How often did you feel nervous?
All of the time Most of the time Some of the time A little of the time None of the time
8. How often did you feel bothered or upset?
All of the time Most of the time Some of the time A little of the time None of the time

9. During the past two weeks, how much of the time did you feel happy?
All of the time Most of the time Some of the time A little of the time None of the time
10. How much of the time did you feel cheerful?
All of the time Most of the time Some of the time A little of the time None of the time
11. How much of the time did you enjoy the things you usually like to do?
All of the time Most of the time Some of the time A little of the time None of the time
12. How often did you have fun?
All of the time Most of the time Some of the time A little of the time None of the time
13. How often did you feel jittery or restless?
All of the time Most of the time Some of the time A little of the time None of the time
14. How much of the time did you have trouble sleeping during the past two weeks?
All of the time Most of the time Some of the time A little of the time None of the time
15. How often did you have headaches?
All of the time Most of the time Some of the time A little of the time None of the time
16. How often did you like yourself?
All of the time Most of the time Some of the time A little of the time None of the time
17. How often did you feel like things were out of control?
All of the time Most of the time Some of the time A little of the time None of the time
18. How often did you talk with one of your parents about your sick parent's illness?
All of the time Most of the time Some of the time A little of the time None of the time

Items 1-18 taken from the Child Health Questionnaire CF-87 (Landgraf and Ware, 1966) Mental Health subscale, section 6.0, "General Well Being"

Family Structural Change Questionnaire

First 4 questions taken from the Child Health Questionnaire, 1996, Landgraf & Ware, Section 9

During the past 2 weeks, how often has the health of someone in your family:

1. Limited the types of activities you could do as a family?
a. very often b. fairly often c. sometimes d. almost never e. never
2. Interrupted various everyday family activities (eating meals, watching TV)?
a. very often b. fairly often c. sometimes d. almost never e. never
3. Limited your ability as a family to “pick up and go” on a moment’s notice?
a. very often b. fairly often c. sometimes d. almost never e. never
4. Caused your family to cancel or change plans (personal or work) at the last minute?
a. very often b. fairly often c. sometimes d. almost never e. never

The next 4 questions modified from The Responses to Stress Questionnaire, Social Stress Version by Connor-Smith, Compas, Wadsworth, Thomsen, & Salzman, 2000

Having a parent go through a bone marrow transplant makes for some very tough times for everyone in the family. So that we can find out how things have been going for you, tell me how much each of the following things have been a problem for you since you knew your parent was going to have the BMT.

5. Feeling like money problems are a big worry for your family
How stressful or how much of a hassle is this?
1) Not at all 2) A little 3) Somewhat 4) Very
6. Difficulty getting my schoolwork done.....
How stressful or how much of a hassle is this?
1) Not at all 2) A little 3) Somewhat 4) Very
7. Being unable to do things I enjoy with my friends.....
How stressful or how much of a hassle is this?
1) Not at all 2) A little 3) Somewhat 4) Very

8. Having too many tasks to do around the house.....
How stressful or how much of a hassle is this?

1) Not at all

2) A little

3) Somewhat

4) Very

Family Cohesion Subscale

Please decide which of these statements are true about your family and which are false, and then circle that word below the statement. Circle true if you think the statement is true or mostly true, and circle false if you think it is false or mostly false about your family. We would like to know what your family seems like to you at this time.

1. Family members really help and support one another.
True False

2. We often seem to be killing time at home.
True False

3. We put a lot of energy into what we do at home.
True False

4. There is a feeling of togetherness in our family.
True False

5. We rarely volunteer when something has to be done at home.
True False

6. Family members really back each other up.
True False

7. There is very little group spirit in our family.
True False

8. We really get along well with each other.
True False

9. There is plenty of time and attention for everyone in our family.
True False

Subscale taken from the Family Environment Scale (Moos & Moos, 2002)

Family Conflict Subscale

Please decide which of these statements are true about your family and which are false, and then circle that word below the statement. Circle true if you think the statement is true or mostly true, and circle false if you think it is false or mostly false about your family. We would like to know what your family seems like to you at this time.

1. We fight a lot in our family.
True False
2. Family members rarely become openly angry.
True False
3. Family members sometimes get so angry that they throw things.
True False
4. Family members hardly ever lose their tempers.
True False
5. Family members often criticize each other.
True False
6. Family members sometimes hit each other.
True False
7. If there's a disagreement in our family, we try hard to smooth things over and keep the peace.
True False
8. Family members often try to one-up or out-do each other.
True False
9. In our family, we believe you don't ever get anywhere by raising your voice.
True False

Subscale taken from the Family Environment Scale (Moos & Moos, 2002)

Family Expressiveness Subscale

Please decide which of these statements are true about your family and which are false, and then circle that word below the statement. Circle true if you think the statement is true or mostly true, and circle false if you think it is false or mostly false about your family. We would like to know what your family seems like to you at this time.

1. Family members often keep their feelings to themselves.
True False
2. We say anything we want to around the home.
True False
3. It's hard to "blow off steam" at home without upsetting somebody.
True False
4. We tell each other about our personal problems.
True False
5. If we feel like doing something on the spur of the moment we often just pick up and go.
True False
6. Someone usually gets upset if you complain in our family.
True False
7. Money and paying bills is openly talked about in our family.
True False
8. We are usually careful about what we say to each other.
True False
9. There are a lot of spontaneous discussions in our family.
True False

Subscale taken from the Family Environment Scale (Moos & Moos, 2002)

Children's Coping Questionnaire

Initially, children were asked to respond to a list of items pertaining to their feelings and behaviors related to their parent's BMT, and asked to identify those which were a problem for them in the two weeks prior to data collection at each time point. These items included: feeling scared, being afraid to ask questions regarding what was going on with their ill parent, tension and conflict at home, changes in their daily routine at home, money problems in the family, increased tasks and chores, their uncertainty regarding the parent's transplant, inability to complete their schoolwork, less time with friends, and changes in the family related to the parent's illness. Children were then asked to respond to the following items which pertain to coping. The following questionnaire was then administered to children:

INTRODUCTION

This is a list of things people sometimes do, think, or feel when they have problems that cause stress. Everybody deals with problems in their own way—some people do a lot of things on this list and others do a few things. For each of the items below, please tell me how much of the time you do or feel these things.

1. I try not to feel anything.
Not at all A little Some A lot
2. I try to think of different ways to change the problem or fix the situation.
Not at all A little Some A lot
3. I wish that I were stronger or smarter so that things would be different.
Not at all A little Some A lot
4. I let someone or something know how I feel.
Not at all A little Some A lot
5. I decide I'm okay the way I am, even though I am not perfect.
Not at all A little Some A lot
6. When I'm around other people, I act like these problems never happened.
Not at all A little Some A lot
7. I deal with the problem by wishing it would just go away, that everything would work itself out.
Not at all A little Some A lot

8. I realize that I have to live with things the way they are.
 Not at all A little Some A lot
9. I try not to think about it, I try to forget all about it.
 Not at all A little Some A lot
10. I ask other people for help or for ideas about how to make the problem better.
 Not at all A little Some A lot
11. I tell myself that I can get through this.
 Not at all A little Some A lot
12. I let my feelings out.
 Not at all A little Some A lot
13. I get help from other people when I'm trying to figure out how to deal with my feelings.
 Not at all A little Some A lot
14. I wish that someone would just come and get me and my family out of this mess.
 Not at all A little Some A lot
15. I do things to try and fix the problem or take action to change things.
 Not at all A little Some A lot
16. I try to stay away from people and things that make me feel upset or remind me of the problem.
 Not at all A little Some A lot
17. I just take things as they are, I go with the flow.
 Not at all A little Some A lot
18. I think about happy things to take my mind off the problem or how I'm feeling.
 Not at all A little Some A lot
19. I get sympathy, understanding, or support from others.
 Not at all A little Some A lot
20. I tell myself that things could be worse.
 Not at all A little Some A lot

21. I tell myself that the problems don't matter, that they aren't a big deal.
 Not at all A little Some A lot
22. I think about the things that I'm learning from this situation, or something good that might come from it.
 Not at all A little Some A lot
23. When something goes wrong, I say to myself, "This isn't real."
 Not at all A little Some A lot
24. When I get upset by these problems, I do things to calm myself down.
 Not at all A little Some A lot
25. I keep my feelings under control when I have to, then let them out when they won't make things worse.
 Not at all A little Some A lot
26. I tell myself that everything will be alright.
 Not at all A little Some A lot
27. I think of ways to laugh about all this so it won't seem so bad.
 Not at all A little Some A lot
28. I imagine something really fun or exciting happening in my life.
 Not at all A little Some A lot
29. I try to believe all this never happened.
 Not at all A little Some A lot

Items 1-29 taken from the Response to Stress Questionnaire (Connor-Smith, Compas, Wadsworth, Thomson, & Salzman, 2000).

Patient Symptom Interference Subscale

Directions:

There are several problems that people may experience while undergoing a bone marrow transplant. Circle the response that best describes how much each of the symptoms listed below has interfered with your life or bothered you during the past two weeks.

1. Nausea?

Not at all A little A fair amount Much Very much

2. Vomiting?

Not at all A little A fair amount Much Very much

3. Mouth soreness?

Not at all A little A fair amount Much Very much

4. Difficulty swallowing?

Not at all A little A fair amount Much Very much

5. Diarrhea?

Not at all A little A fair amount Much Very much

6. Decreased appetite?

Not at all A little A fair amount Much Very much

7. Fever?

Not at all A little A fair amount Much Very much

8. Boredom?

Not at all A little A fair amount Much Very much

9. Tiredness?

Not at all A little A fair amount Much Very much

10. Headache?

Not at all	A little	A fair amount	Much	Very much
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11. Nosebleed?

Not at all	A little	A fair amount	Much	Very much
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12. Difficulty sleeping?

Not at all	A little	A fair amount	Much	Very much
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13. Pain?

Not at all	A little	A fair amount	Much	Very much
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14. Troubled by isolation?

Not at all	A little	A fair amount	Much	Very much
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15. Hair loss

Not at all	A little	A fair amount	Much	Very much
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16. Feelings of loneliness

Not at all	A little	A fair amount	Much	Very much
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Subscale taken from the BMT Symptom Checklist (Fife, 2000)

APPENDIX B: OVERVIEW OF FINDINGS

Overview of Findings

Adaptation

General findings:

- 1) Children's adaptation improved over the course of the BMT trajectory.
- 2) Children's emotional response was slower to improve when compared to behavioral response during the BMT trajectory.
- 3) Children had the poorest emotional and behavioral response prior to the parent's transplant (T1)

Behavioral response:

Less positive child behavioral response associated with:

Prior to parental BMT: Higher levels of family structural change

Higher levels of family conflict

During parental hospitalization: Higher levels of family conflict

Increased use of disengagement coping

1 month post BMT: Higher levels of family structural change

Emotional response:

Less positive child emotional response was associated with:

Prior to parental BMT: Female child gender

Increased use of disengagement coping strategies

During parental hospitalization: A parent having an autologous BMT

1 month post BMT: Increased levels of family structural change

Coping

- 1) Disengagement coping was the only coping strategy significantly associated with child adaptation (See emotional and behavioral response above)
- 2) Children's use of disengagement coping increased significantly from T1 (pre-transplant) to T2, peaked during their parent's hospitalization (T2), and decreased significantly when their parent was at home one month post-transplant (T3). The heightened use of disengagement coping at T2 occurred when family structural change and parent symptomatology were also at their highest levels.
- 3) Children's use of secondary coping strategies focused on adaptation to the stressful situation (acceptance, cognitive restructuring, positive thinking, distraction) was highest prior to their parent's BMT, significantly and dramatically decreased during their parent's hospitalization (T2), and then significantly increased when the parent returned home (T3).

Perception of Illness

- 1) Family conflict and family structural change were the only two Perception of Illness variables significantly associated with child adaptation (See emotional and behavioral response, above).
- 2) Children's perception of family interaction (cohesion, conflict, expressiveness) did not change significantly over the transplant trajectory.
- 3) Children's perception of family structural change was highest when the parent returned home one month after the BMT when compared to the other time points, and significantly had increased since their parent's hospitalization.

Contextual Variables

- 1) Child age, parent gender, parent symptom interference, and length of parent hospitalization were not associated with child adaptation. The type of transplant and child gender were associated with child emotional response (See emotional response, above).

Findings Related to the Model

- 1) The model accounted for approximately 40% of the variance in children's adaptation across the BMT trajectory, except for children's emotional adaptation during their parent's hospitalization. Therefore, the model was preliminarily supported by the findings.

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