

**PATIENT AND FAMILY RESPONSES TO A CHILD UNDERGOING BMT
DURING TRANSITIONS IN CARE**

By

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ABSTRACT

Background

Pediatric blood and marrow transplantation (BMT) is an intense treatment reserved for life-threatening oncologic, hematologic, and immunologic illnesses of childhood. While BMT offers hope for cure, the intensity of the treatment and recovery period, and the associated burdens have been linked to sequelae such as decreased quality of life (QOL), emotional distress, and financial burdens. Even though pediatric BMT affects all family members, very little is known about the direct effects of BMT on the parents of BMT patients, and the resultant family impact. Self-management concepts have been identified by parents as important during their child's BMT and recovery period, but parent and family self-management has not been explored in the pediatric BMT population.

Purpose

The specific aims of this study are to 1) Examine the relationship of family context (complexity of condition and treatment, physical and social environment, and parent and family characteristics) to parent self-regulation, QOL, and general health at five time points during the first 12 months following BMT; 2) Examine the effect over time of complexity of condition and treatment, as measured by BMT complications, on parent impact, general health, and QOL, and 3) Explore the family self-management experience of the transition from acute to chronic care following pediatric BMT.

Method

This study used an explanatory sequential mixed methods design. The quantitative portion included a secondary analysis of an existing data set, and the qualitative portion

consisted of semi-structured family as group interviews. The qualitative portion of this study was guided by the human science phenomenology approach described by Van Manen. The Individual and Family Self-Management Theory guided this study.

Results

The quantitative analysis included 363 parents of pediatric BMT patients. The Parent Impact Scale of the Child Health Ratings Inventories (CHRIs) was found to have a single factor structure. In the first three months after BMT, Parent Impact was significantly worse in the presence of aGVHD \geq grade 2, worse organ toxicity, and systemic infection. From three months to one year, Parent Impact was significantly worse with the presence of systemic infection. A year after BMT, one-third of parents reported taking at least one day off per week from work/cutting back on usual activities.

Longitudinally, parent QOL was significantly better among partnered parents, Hispanic parents, parents with fewer children, parents with lower Parent Impact, and parents of children with better organ function. Parent General Health was significantly better among parents with higher educational attainment, parents with lower Parent Impact, parents of children with non-malignant diagnoses, and parents of children with worse organ toxicity.

Thirteen families participated in the family as group interviews including 11 mothers, 5 fathers, 4 BMT patients, and 4 siblings. Major themes included *Making a Plan*, *Taking Care of Basic Needs*, and *Managing Emotional Burdens*. Even though the circumstances of BMT were similar among different families, families described using vastly different self-management strategies to survive the BMT period.

Conclusions

This study provided important information about the self-management of parents and families during pediatric BMT. In the quantitative analysis parents reported taking time off work/cutting back on usual activities throughout the first year after BMT. In the qualitative interviews, parents described the stress of making work arrangements, and the resulting financial strain.

The findings that family characteristics, such as the number of siblings and partner status of the parents; as well as patient characteristics, such as infection and organ toxicity are related to parent QOL and general health provide further evidence that parent and child outcomes are linked. This information is also critical for the future formation of interventions designed to support families through pediatric BMT. Furthermore, knowing these factors may help clinicians to identify families most at risk for negative sequelae. Future research is needed to study the stability of qualitative concepts over time, and develop family-centered interventions designed to help families achieve their self-management goals during pediatric BMT.

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DEDICATIONS

To Christian –

I'm so thankful for your love and support

And to Dawn –

Thank you for being my friend. This is for you.

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Chapter 1: Introduction

Blood and Marrow Transplantation

Blood and Marrow Transplant (BMT – also called Hematopoietic Stem Cell Transplant, or HSCT) offers children with life-threatening illnesses, and their families, hope of long-term cure (Ho, Horne, & Szer, 2002). About 2,500 children age 0-20 in the U.S. undergo BMT each year (Pasquini, 2010) with 5-year survival rates ranging from 20% to 90%, depending on the type of disease and complications of treatment (Pasquini, 2010). Uniquely, BMT is characterized by an intensive phase of treatment over a period of 100 days or more where the child undergoes transplantation and acute inpatient hospitalization and then transitions to intensive outpatient and home-based monitoring in which the parent as caregiver plays a major role. For families whose homes are distant from the transplant center, temporary housing near the transplant center is often required. Parents have characterized this period of BMT and transition from acute to chronic care as “family, organizational, and financial chaos” (Larsen, Heilmann, Johansen, & Adamsen, 2012). Despite the strain of this treatment and recovery process, few studies have gone on to link the parent experience of BMT to the family experience of BMT. Moreover, the lack of literature on the family experience of pediatric cancer has been identified as a significant barrier to the development of family-focused interventions to promote family well-being during this highly distressing time (Hinds, Feetham, Patterson Kelly, & Nolan, 2012). The proposed study seeks to address these gaps using the Individual and Family Self-Management Theory (Ryan

& Sawin, 2009) to 1: examine parent responses to the child's BMT during the transition from acute to chronic care during the first year after BMT, then, 2: use risk and protective factors identified in this longitudinal quantitative analysis to further explore the experience of the transition from the family perspective. "Family as group" interviews will provide a family context of the experience of the transition. Including children in these interviews will give the opportunity to hear "the child's voice" in this experience (Reeve et al, 2013).

Therefore, the purpose of the proposed study is to examine the relationships among the context, process, and outcomes of parent and family self-management, and explore the experience of parent and family self-management in the first year after BMT, particularly during the transition period.

SPECIFIC AIMS AND HYPOTHESES OF RESEARCH PROPOSAL

The specific Aims of this proposal are:

Aim 1: *Examine the relationship of family context (complexity of condition and treatment, physical and social environment, and parent and family characteristics) to parent self-regulation, QOL, and general health over the first 12 months following BMT.*

Hypothesis 1: Parent impact, general health, and QOL will be rated worse with allogeneic transplant type, longer duration of illness, worse stage of illness, minority race or ethnic status, lower income, younger child age, older parent age, lower parental education level, and presence of siblings in the first year after BMT.

Aim 2: *Examine the effect over time of complexity of condition and treatment, as measured by BMT complications, on parent impact, general health, and QOL.*

Hypothesis 2.1: Parent impact, general health, and QOL will be rated worse for parents whose child develops early BMT complications in the first 3 months after BMT.

Hypothesis 2.2: Parent impact, general health, and QOL will be rated worse for parents whose child develops late BMT complications, 6 months or more after BMT.

Aim 3: *Explore the family self-management experience of the transition from acute to chronic care following pediatric BMT.*

BACKGROUND

Pediatric BMT patients, their parents, and their siblings have all reported adverse effects of BMT (Packman, Weber, Wallace, & Bugescu, 2010); some of the risks to BMT patients and their families include anxiety, depression, post-traumatic stress symptoms, decreased QOL, and decreased self-esteem (Packman, Weber, Wallace, & Bugescu, 2010). Furthermore, pediatric BMT patients and their families are vulnerable not only during the acute phase of the transplant, but BMT can cause long-term physical, emotional, psychosocial, and mental health vulnerability (Barrera, Atenafu, Doyle, Berlin-Romalis, & Hancock, 2012; Clarke, 2011; Drew, Goodenough, Maurice, Foreman, & Willis, 2005; Manne et al., 2003). The transition from acute care in and near the hospital to chronic care based from the family home has also been cited as a highly stressful period for patients and their parents (Mayer et al., 2010; Parsons et al., 2006; Terrin et al., 2013).

Family in the Context of BMT

Family, defined as a self-identified group of two or more members (Eggenberger & Nelms, 2007) is described as the, “primary unit for health” and as

“the most important context within which health and illness occur and illness is resolved” (Craft-Rosenberg & Pehler, 2011). Including the family context in healthcare research has been cited as an important and necessary addition to the literature (Hinds, Feetham, Patterson Kelly & Nolan, 2012; Olson, 2011; Packman et al., 2010). Furthermore, although it is known that individual and family contextual characteristics, such as patient and parent age, parent education and employment status, family income, and number of siblings, can be risk and protective factors that have the potential to influence patient and family outcomes in BMT (Barrera et al., 2012; Hovén, von Essen, & Norberg, 2013; Jobe-Shields et al., 2009; Manne et al., 2002; Phipps, Dunavant, Lensing, & Rai, 2004, 2005; Rini et al., b; Zois, 2011), little is known about the family experience of BMT (Barrera & Atenafu, 2008; Clarke, Eiser, & Skinner, 2008). Two recent systematic reviews (Clarke et al., 2008; Packman et al., 2010) stressed that understanding key risk and protective factors would be an important addition to pediatric BMT science. The primary reasons these factors have not been studied are small heterogeneous samples and resultant lack of statistical power (Clarke et al., 2008). Because parent and child outcomes have been shown to be linked in many studies, learning how to support all members of the family will be an important step to expanding the scientific basis for supporting families through BMT process (Barrera, Boyd-Pringle, Sumbler, & Saunders, 2000; Jobe-Shields et al., 2009; Phipps et al., 2005; Terrin et al., 2013).

Conceptual Framework

The Individual and Family Self-Management Theory (IFSM)

The Individual and Family Self- Management Theory (IFSMT) is a descriptive middle range systems theory, based on the multidimensionality of the individual and family (Ryan & Sawin, 2009). In this model, self-management is described as a process by which individuals and families achieve health-related outcomes (Ryan & Sawin, 2009). The theory suggests three conceptual levels: context, process, and outcomes (Ryan & Sawin, 2009). Factors in the contextual dimension influence both individual and family engagement in the process of self-management, and directly influence outcomes (Ryan & Sawin, 2009).

Context

Complexity of Condition and Treatment

BMT was first developed in the 1950's and 1960's to treat hematologic and immunologic disorders (e.g., aplastic anemia and severe combined immunodeficiency disorder), but because so little was known about human histocompatibility antigens, only identical twins could be used as donors. As scientific discovery about transplant biology and therapeutic advances (e.g., development of immunosuppressants, use of fractionated total body irradiation), the application of BMT expanded greatly to include patients with better risk malignancies. From the mid 1980's to early 1990's, an unrelated donor registry was established. Potential donors underwent initial typing and were "on call" if a

matched patient were identified. Today, while most of the children and adolescents who undergo BMT have an oncologic diagnosis, as treatment toxicity has decreased and overall survival improved, BMT is now considered an option for a growing number of non-malignant hematologic and metabolic diseases. In 2010, about 25% of all BMT recipients between the age of 0 and 20 had a non-oncologic diagnosis (Pasquini, 2010).

There are two broad categories of BMT: allogeneic (meaning the graft cells are from someone other than the patient), and autologous (meaning the graft cells are from the patient). Allogeneic BMT is higher risk than autologous BMT because receiving “foreign” cells (that is, not self) puts patients at risk for graft-vs-host disease (GVHD), an immunologic response of the transplanted graft cells to the indigenous tissues of the patient. GVHD is graded from Grade 1-4, and may affect the tissues of the gut, lung, or skin (Faraci et al., 2012). It can occur in the acute stage of BMT (months 0-3 after transplant), or in the chronic stage (greater than 3 months post BMT). Because of the serious nature of GVHD, allogeneic BMT recipients are given immunosuppressant therapy following transplant to decrease the risk of GVHD. Immunosuppressants are typically weaned 6-12 months after BMT, if there are no signs of GVHD, but may be given longer until GVHD symptoms subside.

Allogeneic grafts are matched as closely as possible to the patient’s human leukocyte antigen (HLA) type as possible to decrease the risk of GVHD (Copelan, 2006). Because of this, relatives, primarily HLA matched siblings, are preferred donors. If no appropriately matched relative is available, then other graft sources may be used including matched unrelated donors (MUD), and umbilical cord blood.

For many years, minority BMT recipients were at a disadvantage because the BMT registry of possible unrelated donors is primarily made up of Caucasian individuals, which are less likely to be an HLA match with minority BMT recipients (Oevermann & Handgretinger, 2012). However, recent medical advancements have made it possible to use parents' (who, by definition are a 50% HLA match) and other relatives' bone marrow for the graft or unrelated (and partially mismatched) umbilical cords, which have expanded treatment options for all patients, but particularly for minority recipients (Copelan, 2006).

The BMT course begins with a typically inpatient conditioning treatment consisting of chemotherapy with/without radiation, designed to eliminate residual disease and immunosuppress the patient to more readily "accept" the donor cells (Copelan, 2006). Conditioning regimens for BMT vary by disease type and stage, and type of transplant. Myeloablative conditioning, often used in children, includes about a week of chemotherapy with/without total body irradiation. This conditioning treatment places the patient at high risk for infection and end-organ toxicity (Copelan, 2006) due to its intensity.

Once the patient is clinically stable, and the transplanted cells have engrafted over a period of 3-6 weeks, the patient and caretaking parent are discharged either to their home (if in close proximity to the transplant center) or to a temporary housing facility close to the hospital so the patient can access transplant care emergently when needed. The caretaking parent is responsible to administer medications, perform central line care, monitor the patient closely for signs of infection and/or GVHD, and take the patient to frequent outpatient clinic visits. No

matter the age of the patient, an adult must be with the patient 24 hours a day, as complications can arise swiftly and progress rapidly.

The patient and caretaking parent typically remain in close proximity and contact with the transplant center until approximately 100 days post-transplant. After 100 days, patients and caretaking parent can return to the family home if the patient is clinically stable. If the transplant center is not the patient's primary healthcare facility, care is usually transferred back to the primary facility between 3-12 months after BMT, depending on the patient's condition and need for ongoing specialized care. The probability of 5-year survival for BMT ranges from 20% -90% depending on the indication for transplant, stage of disease, type of transplant, and complications, but mean overall survival is about 70% (Pasquini, 2010).

Characteristics of the Family and Environment

There is evidence that family and environmental factors affect BMT and psychosocial outcomes. Phipps, Dunavant, Lensing & Rei (2002), found type of transplant affected coping; patients undergoing MUD transplants had the greatest distress, followed by matched sibling and autologous transplants. Barrera & Atenafu (2008) found that two years after transplant, siblings of BMT survivors had more internalizing problems (anxiety, depression, and social withdrawal) than the survivors did. Similarly, Packman, Gong, VanZutphen, Schaffer & Crittendem (2004) found one third of siblings of BMT survivors reported moderate to severe posttraumatic stress. Furthermore, donor siblings reported significantly higher anxiety and significantly lower self-esteem than non-donor siblings (Packman, 1999).

Family finances also cause parents, particularly fathers (Rodrigue et al., 1996), a great deal of distress during BMT. Parents of BMT patients report greater distress related to finances than parents of pediatric oncology patients who did not undergo BMT, and lower socioeconomic status (SES) significantly increases parent distress (Clarke, 2011; Phipps et al., 2004). This is particularly salient because the study in which Phipps et al. (2004) found that lower SES is linked with parent distress was conducted at a transplant center that provided housing and food for the resident parent at no cost. Concerning patients, socioeconomic status (SES) did not affect patient distress while hospitalized, but in the post hospitalization phase lower SES was associated with greater patient distress (Phipps et al., 2002). Financial concerns do not abate at the end of BMT; a sample of parents with a child 2-16 years post BMT reported durable financial difficulties. One possible reason this financial vulnerability persists could be that BMT patients were significantly more likely than non-BMT oncology patients to require specialized medical consultations as part of long-term care (Clarke, 2011; Vrijmoet-Wiersma et al., 2010).

The healthy functioning of the family unit is linked to the child's adjustment to BMT (Jobe-Shields et al., 2009), yet few studies have examined BMT from the perspective of the family unit even though the Institute of Medicine, American Academy of Pediatrics, and oncology researchers have called for the inclusion of the family in healthcare research (Hinds, Feetham, Patterson Kelly & Nolan, 2012; Craft-Rosenberg & Pehler, 2011; EL, 2003; Olson, 2011). Obtaining the perspectives of multiple family members, and examining the impact of BMT on the family unit is increasingly complex in light of the changing composition of families in America.

Step-parents, cohabitating partners, same sex partners, step-siblings, and caretaking grandparents are a few examples of the changing nature of the family in America, and very little is known about the impact of pediatric BMT in these contexts (Kelly & Ganong, 2011; Olson, 2011; Patterson Kelly & Ganong, 2011)

The studies that have examined family factors have found that family cohesion, family expressiveness, and family adaptation are related to BMT patient distress (Barrera & Atenafu, 2008; Jobe-Shields et al., 2009). Furthermore, supportive family environments may serve as protective when families are faced with the acute stress of pediatric BMT (Manne et al., 2002).

The Process of Self-Management

Self-management has been described as, “the process of engaging in specific behaviors [that] enhance a person’s ability to manage and chronic illness” (Ryan & Sawin, 2009 p. 217). Very little is known about the processes individuals and families use to practice self-management through the BMT trajectory. One way self-management can be measured is through self-regulation, or the ability to control emotional, behavioral, or cognitive processes (Ryan & Sawin, 2009). Self-regulation has been correlated with adult BMT patients’ QOL (Solberg Nes, Ehlers, Patten & Gastineau 2013), but this has not been studied in the pediatric BMT population. This is important because parental caregivers of BMT patients have identified self-regulation concepts, including maintaining the family and taking care of themselves, as important topics for pediatric BMT researchers to address (Mayer et al., 2010). A recent study (Pennarola et al., 2012) investigated a related concept, parent activation, and found that parents’ self-activation about his/her own health

management is related to parental activation on behalf of the child. Understanding the processes parents and families undergo to practice self-management, including self-regulation, particularly in the pediatric population, has been identified as a critical need in nursing research (Ryan & Sawin, 2009).

Quality of Life and General Health Outcomes

Studies of pediatric BMT patients have shown patients' QOL tends to be lowest during the acute phase of BMT (particularly during the inpatient hospitalization), and then most patients show improvement with a return to pre-transplant or near pre-transplant levels within 6-12 months following BMT (Barrera, Atenafu, & Hancock, 2009; Clarke et al., 2008; Oberg, 2012; Parsons et al., 2006). Additionally, older pediatric patients, particularly adolescents and young adults reported poorer QOL than school-age patients (Felder-Puig, 2006).

Barrera & Atenafu (2008) also found that two years after BMT, siblings had statistically higher physical functioning scores than their siblings who underwent BMT, but psychosocial functioning was not significantly different between patients and siblings. This finding is most likely due to the intense physical nature of BMT, and suggests that siblings and patients are both affected by BMT psychosocially (Barrera & Atenafu, 2008).

There is conflicting evidence regarding parents' QOL following BMT. Some studies have found the QOL trajectory of parents is similar to patients, i.e., QOL is the worst in the inpatient phase of BMT, and then slowly returns to pre-transplant levels (Phipps et al., 2004). However, Terrin et al. (Terrin et al., 2013) found recovery of parent emotional functioning is more nuanced, and is moderated by

BMT complications such as GVHD and systemic infection. Other factors that are related to better parental QOL include: having a child with fewer behavior problems, being two years post BMT, and being an older mother (Barrera et al., 2012; Clarke, 2011; Terrin et al., 2013). Worse child health, a history of psychiatric illness, having a child who was conditioned for BMT with radiation, and having a female child undergo BMT were all related to poorer parent emotional functioning and psychological QOL (Barrera et al., 2012; Clarke, 2011; Terrin et al., 2013).

Despite the finding that parent QOL generally improves over time, Clarke et al. (2011) found that in a sample of mothers whose children underwent BMT 2-16 years previously, mothers of BMT patients had significantly lower mental well-being than mothers of pediatric oncology patients who did not undergo BMT. Similarly, Jalmsell et al. (Jalmsell, Onelöv, Steineck, Henter, & Kreicbergs, 2011) found that, compared to bereaved parents of pediatric oncology patients who did not undergo BMT, bereaved parents of BMT patients reported significantly lower QOL 4-9 years after the child's death.

Similarly to QOL, parental distress typically peaks in the acute phase of the transplant process, and slowly returns to baseline over time; however, there is evidence of a more nuanced recovery pattern, with a spike in distress around 6 weeks post transplant (Phipps et al., 2004, 2005; Jobe-Shields et al., 2009). Parent distress was increased with younger patient age; child-reported distress; previous experiences of child treatment-related distress; and child behavior problems (Jobe-Shields et al., 2009; Phipps et al., 2004, 2005). Furthermore, parent depressive symptoms moderated the relationship between family cohesion and child distress

as well as family expressiveness and child distress, meaning parental depressive symptoms could overcome the positive effects of family cohesion and family expressiveness, resulting in greater child distress (Phipps et al., 2004).

Mothers' physical and emotional functioning has been studied more thoroughly than fathers' and other caregivers' because mothers are typically the resident parent, and therefore are more likely to quit their jobs, relocate with the patient, and be directly exposed to the daily stressors of BMT (Manne et al., 2002, 2003; Rini et al., 2004). Mothers' fears were significantly related to their distress, and mothers exhibited a high level of distress and depressive symptoms prior to BMT when the child was first hospitalized (DuHamel, 2004; Manne et al., 2003, 2004; Streisand, Rodrigue, Houck, Graham-Pole, & Berlant, 2000). Even though mothers' fears and distress tend to abate over time, eighteen months after BMT 10-15% of mothers met diagnostic criteria for a depressive disorder; nearly 12% met diagnosis criteria for post-traumatic stress disorder (PTSD); and over half of mothers had post-traumatic stress symptoms (PTSS) (Manne et al., 2003, 2004). Intrinsic factors, such as optimism, acceptance, and humor were associated with increased benefit finding, and decreased depressive symptoms (Manne et al., 2003; Rini et al., 2004). Extrinsic factors, such as the child's transplant risk, the child's QOL, family income, spousal support, and social support from family and friends significantly impacted mothers' mental and physical health (Barrera et al., 2000; Manne et al., 2002; Rini et al., 2004).

Even though a number of prospective longitudinal studies have indicated that patients' and parents' physical and mental health is similar to pre-transplant

levels within 3-6 months following BMT, retrospective studies indicate pediatric BMT has long-lasting effects on physical and emotional health (Clarke, 2011; Drew et al., 2005; Vrijmoet-Wiersma et al., 2010). Pediatric BMT patients and their parents reported more late effects and lower QOL than pediatric oncology patients who did not undergo BMT (Clarke, 2011). Furthermore, compared to parents of healthy children, parents of children who underwent BMT 10 years previously were significantly more likely to view their children as vulnerable (Vrijmoet-Wiersma et al., 2010).

There is limited information on parents' distant outcomes after a child's BMT. One study found five years post BMT, mothers were significantly more likely than population norms to report general stress, and mothers' stress levels five years after BMT were similar to those of parents whose child had just completed traditional cancer therapy. Bereaved parents of pediatric BMT patients also reported significantly poorer psychological and physical health than bereaved parents of pediatric oncology patients who did not undergo BMT, and death in hospital emerged as a significant risk factor for parental anxiety and stress (Drew et al., 2005; Jalmsell et al., 2011). Bereaved fathers were particularly prone to anxiety; compared to fathers of pediatric oncology patients who did not undergo BMT, anxiety was twice as prevalent in bereaved fathers of BMT patients (, 2011).

Very little is known about siblings' experience during the BMT course. Wilkins & Woodgate (2007) conducted qualitative interviews with siblings and found that the most common theme was "an interruption in family life," indicating

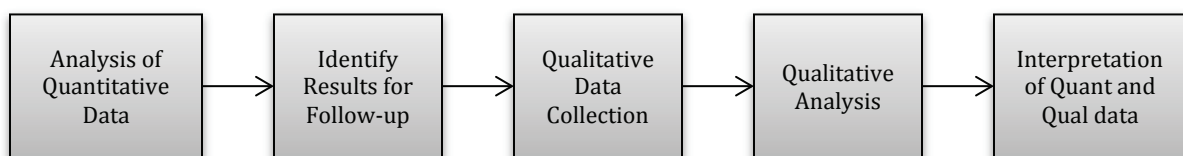
that siblings felt family life was no longer “normal.” Siblings also expressed a great deal of concern for their sick sibling (Wilkins and Woodgate, 2007).

RESEARCH AND DESIGN METHODS

Description of Study

The proposed study uses an explanatory sequential mixed methods design (see Figure 1) (Creswell & Plano Clark, 2011), which will use existing data from the Journeys to Recovery (JTR) study (funded by the American Cancer Society, RSG PB02-186-01-PBP, S. Parsons, PI) and HSCT-Comprehensive Health Enhancement Support System (HSCT-CHESS™) study (funded by NCI, R01 CA119196, S. Parsons, PI). Following quantitative data analysis, family-as-group interviews (Eggenberger & Nelms, 2007) will be conducted to explore the family experience of self-management following BMT during the transition from acute to chronic care, specifically focusing on the process of self-management. This mixed methods design was chosen to provide multiple perspectives and a more complete understanding of parent and family responses to a child undergoing BMT during transitions in care (Creswell, Classen, Piano Clark, & Klegg Smith, 2011)

Figure 1: Explanatory Sequential Mixed Methods Design



Theoretical Framework

The IFSMT was used to guide the specific aims and hypotheses of the proposed study (Ryan & Sawin, 2009). Relevant contextual, self-management, and outcome variables were retained in the model for the proposed study (See Figure 2).

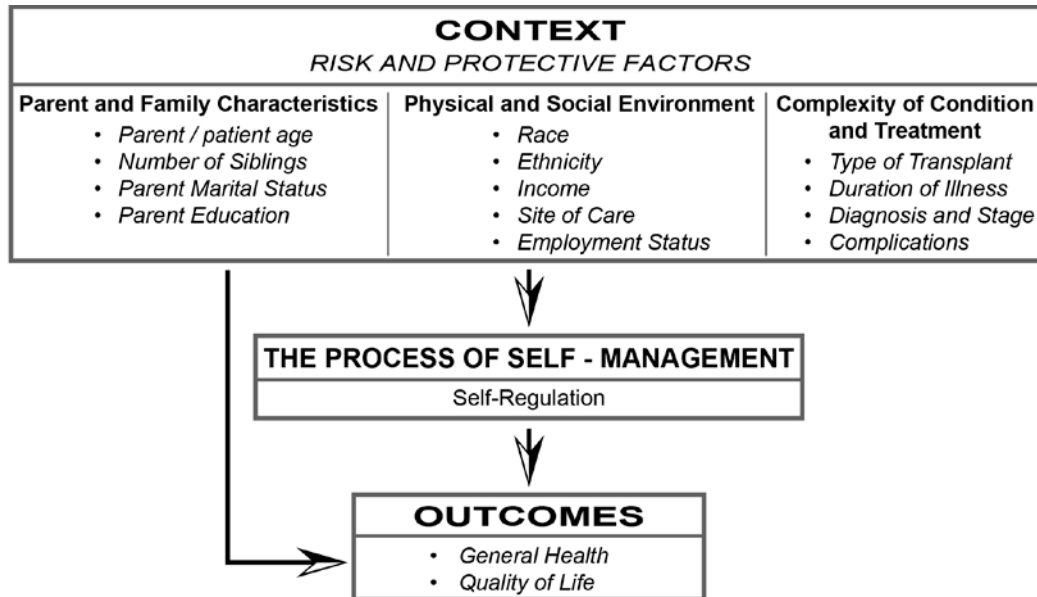


Figure 2: Theoretical Framework as conceptualized for this study

Quantitative Data

Quantitative data was drawn from a merged dataset of two completed studies: JTR and HSCT-CHESS™ (Mayer et al., 2010; Rodday, Terrin, & Parsons, 2013). Together, these studies were conducted longitudinally at eight pediatric BMT centers across the United States. Data were collected at five clinically important time points: baseline (hospital admission), 45 days (approximate hospital discharge), 3 months (approximate transition from hospital housing to the family home), 6 months (start of chronic phase), and 12 months. Inclusion criteria for the source studies were: pediatric BMT patient (age 2 months to 18 years) and caregiving parent, a working knowledge of English, and having a parent or legal guardian who

could consent on behalf of the child. Together, the two studies enrolled 363 patient and parent dyads of which the parents are included in the proposed study. All five time points of parent data from the 363 participants of the merged datasets will be included in this secondary analysis. Parent data was chosen for this proposal because self-regulation was not measured from the child's perspective in the source studies.

Description of the Sample

Quantitative data were collected in the source studies using the demographic, parent General Health, parent QOL, and Parent Impact domains of the Child Health Rating Inventories (CHRI). The CHRI has been validated in the pediatric BMT population (Parsons, Barlow, Levy, Supran & Kaplan, 1999; Parsons et al., 2005, 2006).

Parent and Family Characteristics: Individual and Family Characteristics were measured using the CHRI demographic questionnaire. Demographic variables were collected at the beginning of the transplant. Variables include patient and parent age, number of siblings, parent marital status, and parent educational level. Patient and parent age were collected as continuous variables. Number of siblings was collected as a discrete variable, and included all siblings living at home, including step or half siblings. Years of parent education were collected discretely, but any education attained after a bachelor's degree was categorized as "more than 17 years." Parent marital status was measured categorically, and responses included "married or living with a partner"; "divorced, separated, or widowed"; "never married"; or "other."

Physical and Social Environment: These items were measured using the CHRIs demographic questionnaire. The demographic questionnaire was administered at the beginning of the transplant. In the proposed study, Physical and Social Environment variables included race, ethnicity, and family income. Racial categories were based on the US Census with an additional option of “prefer not to answer” or “does not apply to me.” Race was coded as 1= White, 2= Black, 3=Asian, 4=Pacific Islander, 5=American Indian, 6=Mixed race, 7=No answer, 8=Categories don’t apply to me. Ethnicity was used to measure if the parent identified as Hispanic (yes/no). Income was coded as a categorical variable with 5 categories ranging from less than \$20,000 to \$80,000 and above.

Complexity of Condition and Treatment: disease severity is conceptualized by type of transplant, duration of illness, causal diagnosis for transplant, and BMT complications (infection, GVHD, organ toxicity). Trained research staff collected disease severity measures from the medical record prior to BMT, and BMT complications were recorded at the time of hospital discharge, 45 days, and 3, 6, and 12 months. Response options for type of transplant included autologous, allogeneic – related, and allogeneic – unrelated. Duration of illness was measured in months of illness prior to BMT. Early BMT complications (up to 3 months) included acute GVHD, infection, and Bearman overall toxicity (Bearman et al., 1988). Late BMT complications (6 months and beyond) included chronic GVHD and infection. In all cases standard classification scales were used to describe the clinical events.

Self-Regulation: In the proposed study, self-regulation will be measured with the Parent Impact scale the CHRIs; in this study, parents’ emotional functioning, missed

time from work, planning for help, and attention to the parents' own needs will be a proxy for self-regulation. This is a new scale that measures the emotional and practical processes of parents that has never been reported before. Both parent and family self-regulation will be explored further in the qualitative interviews. The Parent Impact module was administered at baseline, 45 days, 3 months, 6 months, and 12 months.

Dissertation Organization

This dissertation consists of five chapters. Chapter one provides a description of study background, an overview of the study, and a description of the purpose and aims of the dissertation; the conceptual framework; and important concepts.

Chapter two (manuscript one) is a paper examining the psychometric properties of the Parent Impact Scale, and includes some preliminary analyses of the Parent Impact Scale. This manuscript was published in *Health and Quality of Life Outcomes* in 2015.

Chapter three (manuscript two) is based on the quantitative findings from Aims 1 and 2. This manuscript explores the outcomes of parent General Health and QOL.

Chapter four (manuscript three) reports the findings from the qualitative family-as-group interviews.

Chapter five presents: (1) a concise summary of the dissertation findings reported in Chapters three and four; (2) study strengths and limitations; (3) and study implications.

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Chapter 2: Manuscript One

The Impact of Pediatric BMT on parents: introduction to the Parent Impact Scale

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Abstract

Background

Parents often experience stress-related complications when their child requires a blood and marrow transplant (BMT). Previous studies have described the emotional toll BMT places on parents for many during the acute phase of care and within the context of clinical complications. In this paper we introduce the Parent Impact Scale (PARImpact), designed to capture the physical and emotional challenges of the child's health on the parent throughout the first year following BMT. The primary aim of this paper is to examine psychometric properties of PARImpact, and the secondary aim is to explore factors associated with PARImpact scores for further hypothesis generation.

Methods

Data for this analysis were drawn from a merged dataset of two longitudinal studies following BMT. Accompanying parents ($n = 363$) of children undergoing BMT were surveyed up to six times from pre-BMT baseline to one year after their child's BMT with a battery of measures. For this analysis, pre-BMT baseline responses to PARImpact were used to examine the factor structure with Principal Component Analysis (PCA) and Exploratory Factor Analysis (EFA). Construct validity was assessed, and unadjusted and adjusted multivariable regression was used to examine relationships between PARImpact and BMT clinical variables.

Results

PCA and EFA revealed a one-factor solution with acceptable item loading; Cronbach's α was 0.83 at baseline. Hypothesized differences in known groups were detected for BMT complications with significantly higher PARImpact scores for those with vs. without each complication. In the adjusted multivariable regression models, acute graft versus host disease ($b = 5.3$; $p = 0.03$), end organ toxicity ($b = 5.9$; $p < 0.01$), and systemic infection ($b = 9.1$; $p < 0.01$) were associated with significantly higher mean PARImpact scores in the first 3 months following transplant. After the first 3 months to 1 year post BMT, systemic infection was associated with increased mean PARImpact scores ($b = 19.2$; $p < 0.01$).

Conclusions

These initial results suggest that the PARImpact scale is a valid and reliable measure. Our finding that clinical complications increase the impact of BMT on the caretaking parent indicates the need for BMT healthcare professionals to identify these events and help parents navigate the BMT course. Clinical application of the PARImpact scale should be considered to identify high-risk families and provide targeted interventions to augment care.

Keywords

Blood and Marrow Transplantation, Caregiving, Stress, Parent Impact

Introduction/Background

Introduction

Blood and marrow transplant (BMT) offers children with life-threatening illness, and their families, hope of durable cure when no other curative treatment is available [1]. However, parents of children undergoing BMT often experience a great deal of stress [2], and are at risk for short- and long-term psychosocial sequelae, such as depression, anxiety, post-traumatic stress symptoms, and decreased quality of life (QOL) [3-6]. While some prospective studies have shown that parental well-being is most affected during the acute phase of the BMT and then improves with time, there is evidence that the pattern is more nuanced, and is related to other variables, such as previous emotional history and the clinical complications of the BMT [5,7-10]. Furthermore, retrospective studies show some bereaved and non-bereaved parents continue to experience distress for years after the BMT [11-13].

Enhancing our understanding of the parent, family, and BMT characteristics that are associated with increased parental impact may help clinicians identify parents most at risk, and lead to the development of interventions to support parents through the BMT trajectory. In one study of 49 parents/guardians of BMT recipients, 81% of parents/guardians reported they felt proceeding to BMT was their only choice, given its life-saving potential [14]. Because of the high stakes and demands of the treatment, support for these potentially vulnerable parent caregivers is paramount.

Previous research has documented that parental distress [9] and diminished parent emotional functioning are associated with BMT [7]. What has not been described is the direct effect of the child's emotional and physical condition on parents, and their ability to manage other obligations, such as work. In this paper we introduce the Parent Impact Scale (PARImpact), a scale within the Child Health Ratings Inventories (CHRI), [15] designed to capture the physical and emotional impact of the child's health condition on the parent. In this context, 'impact' is considered in the negative sense, conferring challenges to the parent. The primary aim of this analysis is to examine the psychometric properties of the PARImpact as a "stand-alone" scale of the impact of children's BMTs on parents, and parents' ability to meet their own needs. A secondary aim is to explore factors associated with PARImpact scores for further hypothesis generation.

Methods

Data for this analysis were drawn from a merged dataset of two completed dyadic longitudinal health-related quality of life (HRQL) studies comprised of children undergoing BMT and their accompanying parent (Journeys to Recovery (JTR) and HSCT-CHESSTM, described elsewhere [16-18]). Data were collected at eight pediatric BMT centers across the United States from 2003–2011 at clinically relevant time points: pre-BMT baseline, 45 days (represents the end of the inpatient hospitalization period), 3 months (end of the acute BMT period), 6 months, 9 months (HSCT-CHESSTM only), and one year after transplant (designed to capture the late complications and recovery period). Both source studies, including data from the PARImpact Scale described in this paper, were approved by the Tufts Medical Center Institutional Review Board (IRB) and the

IRB at each research center. IRB approval was also obtained to combine the data for this analysis.

Survey data in this study were drawn from the CHRIs-General, a generic HRQL measure with child, adolescent, and parent versions. The CHRIs contains scales to measure HRQL and related constructs: HRQL scales measure emotional, physical, and role functioning, which together represent the multi-dimensional construct of HRQL [19], while PARimpact and global QOL measure related constructs. The separate global QOL scale consists of nine items in which the respondent rates their overall quality of life in the areas of physical health, emotional health, and social health [15]. The CHRIs scales have been extensively validated within the pediatric BMT population [6,15].

Study Sample

Accompanying parents of pediatric patients aged 2 months to 18 years who were scheduled to undergo BMT at study sites were consecutively recruited. Dyadic participation of the parent and child was required. Inclusion criteria for parents were: ability to speak/understand English, at least 18 years old, parent or legal guardian of the BMT patient, and ability to provide consent to participate both for him or herself as well as for the BMT patient. While only parent data were included in this analysis, age-eligible child participants also provided assent/consent for his or her participation. Overall, 62% of eligible dyads consented to participate. Common reasons for non-participation included child refusal (27%), parent refusal (44%), and medical reasons (3%). Participants did not differ from non-participants based on child age, gender, or race/ethnicity.

A total of 423 parents were enrolled in the JTR and HSCT-CHESS™ studies; 60 parents of children less than 5 years old were excluded from this analysis because in the JTR study parents of children in this age group did not complete the CHRIs, which left 363 parents in this analysis.

Measures

Parent Impact Scale

The PARImpact was developed as a separate scale within the parent-proxy version of the CHRIs-General to measure the response of the parent to the child's physical and emotional health needs. The scale consists of four items with five response options on a frequency-based Likert-type scale ranging from "none of the time" to "all of the time" (see Table 1 for PARImpact questions). The scale scores range from 0–100 with higher scores indicating greater parent impact. Starting 45 days after transplant ("day 45" the second data collection point), three additional open-response items were administered alongside the PARImpact scale items to ascertain the actions that parents took in the previous seven days as a result of their child's BMT. These included the number of missed work days in the past week due to the child's health, missed work days due to the parent's health, and the number of days someone came to help the family in the past week. While these items were not designed to be part of the scored PARImpact Scale, they are a related set of items that complement the PARImpact scale. Among these three items we focused on missed workdays due to the child's health for this analysis.

Table 1 PARImpact Items and Scores

Pre-BMT Baseline Scores (Cronbach's $\alpha = 0.83$)				
	n	Mean	Std.	
		Dev.	Floor	Ceiling
Summary Score*	363	51.41	24.65	2 4
Item Summaries				
Child's physical health causes suffering	363	60.54	28.32	6 19
Child's physical health limits time for parental needs	363	50.96	31.02	15 12
Child's emotional health causes suffering	363	51.24	31.35	13 15
Child's emotional health limits time for parental needs	363	42.91	30.70	20 8

*PARImpact is scored from 0–100 with higher scores indicating greater impact.

Parent Emotional Functioning

The Parent Emotional Functioning scale within the parent-proxy CHRIs General consists of seven questions measuring parent mood, anxiety, and distress [5,6,20]. The response options for each item utilize a five-point Likert-type scale. Scale scores range from 0–100 with higher scores denoting better emotional functioning. In the JTR study, the Parent Emotional Functioning scale demonstrated strong prediction of DSM-IV Axis 1 disorders, based on area under the receiver operating characteristic curve [21] for predicting a threshold or sub-threshold disorder from related modules of the Structured Clinical Interview for DSM-IV Axis 1 disorders (c-statistic = 0.75) [20].

Parent Global QOL

The parent version of the Global QOL scale of the CHRIs consists of nine questions designed to capture the multidimensionality of QOL as physical, mental, and social well-being. Responses are measured with a Likert-type scale with five response options ranging from “poor” to “excellent.” The scale is scored from 0–100, with higher scores indicating higher QOL [16].

Parent General Health

The General Health item in the CHRIs is a single summary item of parents’ general health appraisal. Parents were asked, “Overall, how would you rate your health”; response options include a five-point Likert-type scale, which is scored on a 0–100 scale with higher scores indicating better health. This item has been used extensively in clinical practice and research [22] and has been found to be associated multi-item scales of general health and other markers of disease and clinical outcomes [23].

Clinical Variables

Clinical data were collected at all assessment time points by trained study staff, using standardized data collection instruments. All clinical data were reviewed by the study PI (SP) for completeness and consistency. Pre-BMT baseline information included time since diagnosis (months), disease category (malignant or non-malignant), and transplant type (related allogeneic, unrelated allogeneic, or autologous). In follow up, specific clinical outcomes variables used in this analysis included early and late BMT complications, defined below.

Early BMT Complications

We assessed early BMT complications with standardized grading scales that assess both presence or absence and severity of the following: acute graft versus host disease (aGVHD) [24]; end organ toxicity, based on the Bearman Toxicity Scale; [25] and systemic infection, based on the National Cancer Institute's Common Toxicity Criteria for AE, v. 3.0 [26]. Each of the early BMT complications was dichotomized as follows: grade 2 or higher aGVHD; Bearman Toxicity Score maximum ratings of "intermediate" or "poor" within the first 3 months post-transplant; and grade 3 or higher infection, indicating systemic and/or life-threatening infection.

Late BMT Complications

Late BMT complications were defined as the extent of chronic graft versus host disease (cGVHD) [27,28] or a systemic infection that occurred after the 3-month data observation [26]. The Bearman scale was not designed for use beyond the 3-month mark post BMT.

Demographic Variables

Parents' demographic data, including age, race/ethnicity, education, marital status, household income, work status (e.g., full-time, part-time), and child's insurance were collected from parent participants at pre-BMT baseline. Parents also reported their child's race/ethnicity and sex.

Statistical Analysis

Demographic and clinical characteristics were described for the combined study sample using medians (interquartile range [IQR]), means (standard deviations [SD]), frequencies, and percentages at pre-BMT baseline. Parents' missed work days due to the child's health were also summarized, and Spearman's Rank was used to compare this open-response item to the PARImpact scale score from day 45 to 1 year post BMT.

Psychometric Properties of the PARImpact Scale

Principal component analysis (PCA) was used to determine the unidimensionality of the PARImpact scale, based on data collected at pre-BMT baseline. A scree plot was used to retain components with an Eigen value greater than 1.00. Exploratory factor analysis (EFA) was completed to examine factor loadings and uniqueness. Factor loadings >0.4 were considered acceptable [29].

Pre-BMT baseline raw scores, means, SDs, ceiling and floor effects, and percent missing were calculated for each item within the PARImpact. Cronbach's alpha [30] was calculated to estimate the internal consistency of the scale at pre-BMT baseline. For exploratory scale development, the minimum acceptable Cronbach's alpha is 0.7, but for established scales the minimum is 0.8 [31].

Validation of the PARImpact Scale

To assess convergent validity of the PARImpact within the pediatric BMT sample, Pearson correlations were calculated between the PARImpact and other scales of the

CHRIIs General, including Parent Emotional Functioning, Parent Global QOL, and Parent General Health. Correlations between 0 and .30 were classified as weak, .30-.60 as moderate, and > .60 as strong [32]. We hypothesized that parent impact would be strongly correlated to parents' emotional functioning, moderately correlated with Global QOL, and weakly correlated with parents' general health. The correlations were expected to have negative valence due to the directionality of the scales (e.g. higher scores for emotional functioning suggest better functioning, while higher scores for PARImpact suggest more negative impact).

Expected variation of PARImpact scores by known groups was explored using clinically important subgroups, such as early complications, and later complications. For known group comparisons, two separate binary variables were created: (1) Early Complications, which included aGVHD \geq grade 2 and/or systemic infection and/or "intermediate" or "poor" end organ toxicity; and (2) Late Complications, which included cGVHD and/or systemic infection.

Regression Analysis

We built two models for regression analysis: an early model and a late model. The early model included data from pre-BMT baseline, day 45, and 3 months. The late model included data after 3 months through 12 months post BMT. In both models, unadjusted longitudinal regressions were completed with demographic and clinical variables using residual maximum likelihood (REML). Timing of repeated assessments was calculated as the number of days since BMT. Based on a likelihood ratio test (data not shown), time was treated as continuous rather than categorical. Variables with an estimated coefficient

with $p \leq 0.2$ were included in the adjusted multivariable regression model. In the adjusted multivariable longitudinal regressions, variables with $p > 0.1$ were then removed from the model.

Auto regressive, unstructured, and compound symmetry correlation structures were compared using Akaike information criterion (AIC). To address the possibility that PARimpact scores may have been missing not at random (MNAR) over time, we stratified the final model by the extent and causes of missing data, defining strata as follows: (1) those with missing data due to a medical reason (e.g. child too sick) and (2) those with complete data or those with missing data not due to a medical reason (e.g., logistical reasons, such as transportation or work-related issues). The stratified models (called pattern mixture models, PMM) [33] assume the data are missing at random (MAR) within strata. We compared the stratified to the unstratified model using the likelihood ratio test to assess for the presence of MNAR. SAS version 9.3 was used for all statistical analyses; alpha was set at 0.05.

Results

Study Sample

Pre-BMT baseline demographic and clinical characteristics are shown in Table 2. The majority of the sample was female and Caucasian with at least some college education. Most parents reported being married or living with a partner (80%), and reported having at least one additional child with the BMT recipient (84%).

Table 2 Participant Characteristics

	Mean		Standard Dev.	
Child Age	9.6		5.1	
Parent Age	38.7		7.5	
Length of illness (months)	11 (Median)		5, 37 (Q1, Q3)	
	Patient		Parent	
	n	Percent	n	Percent
Sex				
Female	169	47	301	83
Male	194	53	62	17
Race				
White	291	81	277	77
Black	24	7	21	6
Asian	11	3	13	4
Other	32	9	47	13
Hispanic Ethnicity				
Yes	63	18	64	18
No	297	83	297	82
	n		Percent	
Parent Marital Status				
Married/Living together	292		80	
Divorced/Separated/Widowed	49		14	
Never Married	18		5	
Other	4		1	
Family Income				
<\$20,000	31		9	
\$20,000-\$39,999	89		25	
\$40,000-\$79,000	114		32	
\$80,000 +	123		34	
Child's Insurance				
Private	235		65	
Public	121		33	
None/Unknown	7		2	
Parent Education				
< High School	21		6	
High School Graduate	91		25	
Some College	108		30	
College Graduate +	143		39	
Number of Siblings				
0	60		17	
1	134		37	
2 or more	169		47	
BMT Type				
Autologous	82		23	
Allogeneic – related	89		25	
Allogeneic – unrelated	192		53	
BMT Source				
Bone Marrow	200		55	
Peripheral blood	126		35	
Umbilical Cord Blood	35		10	
Other/Combined	2		1	
Death of Child within 12 months				
Yes	63		17	
No	300		83	

Disease Relapse within 12 months		
Yes	48	13
No	315	87

PARimpact Scores

Table 1 displays PARimpact scale and item means, SDs, and floor and ceiling effects at pre-BMT baseline. For each item, the full range of possible responses from 0–100 was utilized. Variability across the scale’s items was similar (SD 28.3-31.4). Responses at the floor and ceiling were <5% for the summary score, and ≤20% for the individual items; there were no missing data at pre-BMT baseline.

Table 3 displays open responses to the item, “In the past week, how many days have you missed work or cut down on usual activities due to this child’s health?” At 45 days after BMT, 68% of parents reported missing work or cutting down usual activities at least one day in the previous week. At 3 months this value was 61%, and at one year, it was 33%. This item was moderately correlated with the PARimpact scale score, with Spearman’s Rank correlation values ranging from 0.28-0.57 across time measurements.

Table 3 Parent self-reported days of missed work/decreased usual activity

Days of missed work/decreased usual activity due to this child’s health	Day 45 n(%)	3 months n(%)	6 months n(%)	12 months n(%)
0	93 (32)	111 (40)	140 (55)	156 (68)
1-3	53 (18)	70 (25)	61 (24)	44 (20)

Psychometric Properties

PCA and EFA

PCA indicated a single component with an Eigen value of 2.6; all other Eigen values were less than 1.0. This component, “PARimpact,” explained 66% of the variation in the PARimpact scale. In the EFA, factor loadings ranged from 0.60 to 0.88, and uniqueness

was <0.20. At pre-BMT baseline Cronbach's alpha was 0.83, and at follow-up time points Cronbach's alpha ranged from 0.84 – 0.90.

Construct Validity

The PARImpact and Parent Emotional Functioning scales were strongly correlated (–0.57 to –0.67) across time periods. The Pearson correlation for PARImpact and Global QOL ranged from –0.49 to –0.63 across time periods. General Health, which was hypothesized to be the least conceptually similar to PARImpact, was weakly or moderately correlated with PARImpact; Pearson's correlation ranged from –0.18 to –0.45 across time periods (see Table 4 for correlation coefficients at all time measurements).

Table 4 Correlation Coefficients for PARImpact, Global QOL, and General Health

	Pre-BMT	Day 45	3 months	6 months	1 year
Parent Emotional Functioning	-0.57	-0.66	-0.60	-0.67	-0.66
Parent Global QOL	-0.49	-0.50	-0.57	-0.58	-0.63
Parent General Health	-0.18	-0.25	-0.37	-0.31	-0.45

Discriminant Validity: Results of Known Groups Comparisons

Among parents completing the 3-month assessment, 71 (37%) BMT patients had early BMT complications. Parents of these children had a PARImpact score that was an average of 11.1 points higher than parents whose children did not experience early complications ($t = 3.75$; $p < 0.01$). Among parents completing the 12-month assessment, 86 (37%) experienced late BMT complications; parents whose children experienced late BMT complications averaged 6.8 points higher ($t = 1.91$; $p = 0.06$) than parents whose children did not.

Regression Analysis

Unadjusted Analysis

In the early model, none of the demographic variables met the criteria to be included in the adjusted model. Among the early complication clinical variables, aGVHD ($p < 0.01$), Bearman Toxicity Score ($p < 0.01$), and systemic infection ($p < 0.01$) met the criteria to be included in the adjusted model.

In the late model, parent sex ($p = 0.05$) was the only demographic variables to be included in the adjusted model. Among the late complication clinical variables, systemic infection ($p < 0.01$) was the only variable to be included in the adjusted model.

Adjusted Multivariable Analysis

Based on AIC, a compound symmetry correlation structure was selected for the final model. Results of the likelihood ratio test comparing the PMM to the repeated measures model indicated the presence of MNAR (Early: $\chi^2(5) = 12.7$, $p = 0.03$; Late: $\chi^2(3) = 16.7$, $p = < 0.01$), therefore PMM estimates were used in the final models. Estimated coefficients from the final models are displayed in Table 5.

Table 5 Estimated Coefficients for adjusted multivariable pattern mixture models

	Estimate	SE	p-value
Early (start of BMT – 3 months)			
Time	−0.07	0.02	<0.01
aGVHD \geq grade 2	5.31	2.48	0.03
Bearman Toxicity	5.91	1.97	<0.01
Systemic Infection	9.09	2.09	<0.01
Late (after 3 months - 1 year)			
Time	−0.02	0.01	0.02
Systemic Infection	19.18	3.14	<0.01

In the Early model, parents of children who experienced \geq grade 2 aGVHD had a mean PARImpact score that was 5 points higher than parents of children who did not ($p = 0.03$). Furthermore, PARImpact scores averaged nearly 6 points higher among parents of children who had Bearman Toxicity Score of “intermediate” or “poor” ($p < 0.01$); and scores averaged 9 points higher among parents of children who experienced a systemic infection in the first 3 months following BMT ($p < 0.01$).

In the Late model, parent sex did not meet the criteria to be retained in the final model ($p = 0.15$). Among parents of children who experienced a systemic infection, mean PARImpact scores were 19 points higher than parents of children who did not experience a systemic infection ($p < 0.01$).

Discussion

This study introduced the Parent Impact Scale, a four-item scale of the CHRIs General parent-proxy version. A scree plot and Eigen values supported a single factor solution, and PCA indicated a single factor explained more than 60% of variation. Factor loadings and uniqueness were acceptable, and the PARImpact scale demonstrated strong coefficient of internal consistency reliability ($\alpha = 0.83$).

The PARImpact also demonstrated hypothesized convergent and known groups validity. Specifically, the scale was strongly associated with global QOL and parent emotional functioning, but not with general health scales. In known group comparisons higher PARImpact scores were noted among parents whose children experienced early and late BMT complications ($p < 0.01$ and $p = 0.06$, respectively).

The additional open-response item of the PARImpact, assessed in follow-up time points, reveals that a high percentage of parents did miss work and/or cut down on usual activities during the year following BMT. This study is among the first to describe parents' work patterns and/or decrease of usual activities post transplant. By 3 months post BMT, more than 2/3 of parents took at least one day off of work and/or cut down on usual activities during the week prior to the assessment *due to the child's health*; by one year post BMT, nearly 1/3 of parents had taken off at least one day of work – a finding that reflects the high role disruption and associated economic burden that BMT places on families. This is consistent with other studies that have found more than one year after BMT, families of children who had BMT experience a greater financial impact than families of children who underwent treatment for leukemia, but did not require BMT [13]; and that 5–10 years after BMT, families of children who underwent BMT reported more financial strain than families of children with oncologic diagnoses who did not undergo BMT [11].

PMMs, which accounted for MNAR, showed mean PARImpact scores were significantly increased with clinical BMT complications, such as moderate to severe GVHD, “intermediate” or “poor” end organ toxicity, and systemic infection, which is consistent with other findings that parent emotional functioning declined with BMT clinical complications [5]. Systemic infection was associated with significantly higher mean PARImpact scores in the both the early and late models (9 points and 19 points higher respectively), which may be an indication of the distress and disruption that can be associated with a life-threatening complication like systemic infection at any point in the recovery trajectory. A more complete understanding of the durability of parent

vulnerability after BMT could lead to the development of interventions to decrease parent impact during and after BMT. In a retrospective study more than one year after BMT, parents indicated that education related to taking care of themselves would be helpful to parents during the BMT process [34].

Strengths and Limitations

This study represents eight years of research; it is strengthened by the power of the large sample. Although the majority of the sample was White/Non-Hispanic (70%), as is typical of studies in this clinical population, the study team was able to recruit more than 27% Non-White or Hispanic participants. Study participants also had high educational attainment and income. The longitudinal design strengthens study findings, as does the completeness of clinical outcomes data even when patient-reported outcomes were missed. Nevertheless, as would be expected in a longitudinal study in a critically ill population, some study participants were lost to follow-up, principally due to the child's death or relapse. Rigorous study procedures were used to mitigate loss to follow-up for non-medical reasons, and PMMs were used to account for data MNAR.

Another important limitation of the current study is that all of the survey measures used to assess the convergent validity of the PARImpact were self-reported by parent participants. This can result in shared variance, which may inflate measures of construct validity. Related data on missed work were also self-reported. However, we used clinical variables to examine known group validity, which were not parent reported. Finally, it is possible that there are other variables that contribute to parent impact during a child's BMT that were not included in the PARImpact scale.

Conclusion

The promising psychometric properties of the PARImpact scale indicate that parent impact is a unidimensional construct with clinical relevance. Our findings that early and late BMT complications, such as systemic infection, significantly increase the impact of BMT on the caretaking parent indicate the need for BMT healthcare professionals to identify these events and help parents navigate the BMT course. At minimum, BMT healthcare professionals should be mindful of the additional burden on the parent that complications bring, and proactively link parents to resources to help them cope with the added impact. Further study is needed to test the validity of the PARImpact scale in other populations of caretaking parents of children with chronic conditions.

Abbreviations

AIC, Akaike information criteria; BMT, Blood and marrow transplantation; CHRIs, Child Health Rating Inventories; EFA, Exploratory factor analysis; aGVHD, Acute graft versus host disease; cGVHD, Chronic graft versus host disease; HRQL, Health related quality of life; JTR, Journeys to Recovery; MAR, Missing at random; MNAR, Missing not at random; PARImpact, Parent Impact Scale; PCA, Principal component analysis; PMM, Pattern mixture models; QOL, Quality of Life; REML, Residual maximum likelihood.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

KH completed a portion of the statistical analysis and was responsible for drafting the manuscript. AMR was responsible for the majority of the statistical analysis, providing statistical expertise, and suggesting edits to the manuscript. MN helped to guide the concepts for the analysis and suggested edits to the manuscript. KB, MJK, SKP, KS, LH, CR, LS, SD, ECG, RN, and GC provided expert opinion and suggested edits. SP mentored the team, guided the conceptual development of the manuscript, and suggested edits. All authors read and approved the final manuscript.

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Chapter 3: Manuscript Two: Parents' General Health and QOL in the First Year Following Their Child's BMT

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INTRODUCTION

Pediatric blood and marrow transplant (BMT) is among the most intensive treatments currently available to children suffering from oncologic, immunologic, and hematologic illnesses. BMT provides hope of durable cure, but also can be the source of tremendous stress and worry, and carries the risk of life-threatening complications (Phipps, Dunavant, Lensing, & Rai, 2004). In addition to the physical demands of BMT, the child is required to live about a month in the hospital and about two additional months near the hospital before returning to the family home, which means parents need to be with the child undergoing BMT, care for any other siblings, manage the family home, and continue working to provide financially for the family (Mayer et al., 2010). Balancing family responsibilities in addition to caring for a seriously ill child can be challenging to parents, and may strain their emotional and physical resources (Mayer et al., 2010).

The literature has shown that parents' quality of life (QOL) is lowest at the start of BMT and increases as the child's recovery process ensues (Phipps et al., 2004). However, parents' QOL is influenced by factors such as child health, child behavior, and parent age; and recovery after a child's BMT may be influenced by other factors such as BMT complications (Barrera, Atenafu, Doyle, Berlin-Romalis, & Hancock, 2012); (Terrin et al., 2013); (Heinze et al., 2015). Additionally, even though physical and emotional health outcomes are frequently linked (Pinquart & Sörensen, 2007), very little is known about parents' physical health during and after their child undergoes BMT.

Further research is needed to understand factors related to how parents and families respond to the distressing experience of having a child undergo BMT.

The Individual and Family Self-Management Theory

Family, which is defined as two or more individuals who self-identify as family (Eggenberger, 2007), has been described as the primary unit for health and the most important context in which health and illness occur and in which illness is resolved (Craft-Rosenberg & Pehler, 2011). Obtaining a better understanding of family in the oncology setting has been cited as an important research goal (Hinds, Feetham, Kelly, & Nolan, 2012).

The Individual and Family Self Management Theory (IFSMT) is a descriptive mid-range systems theory with three main components: context, process, and outcomes (Ryan & Sawin, 2009). This analysis focuses on the context, including demographic and clinical variables, and the outcomes, including parent QOL and parent general health. One self-regulation variable, parent impact (Heinze et al., 2015), was also included as a covariate.

The theory of Individual and Family Self-Management (Ryan & Sawin, 2009) may provide insight into how parents and families manage these health challenges.

Therefore, the purpose of this study was to generate knowledge about variables related to the QOL and general health of parents whose child has undergone BMT during transitions from acute to chronic care. These findings could inform the development of interventions targeted to improve parents' health and help pediatric BMT clinicians better care for parents of BMT patients.

METHODS

Data for this analysis were taken from a merged dataset of two completed dyadic longitudinal health-related quality of life (HRQL) studies comprised of children undergoing BMT and their accompanying parent (Journeys to Recovery (JTR) and HSCT-CHESS™, described elsewhere (Kelly, Pennarola, Rodday, Parsons, & Journeys to Recovery Study, HSCT-CHESS™ Study, 2012). Data were collected at eight pediatric BMT centers across the United States from 2003–2011 at clinically relevant time points: baseline (pre-BMT), 45 days (represents the end of the inpatient hospitalization period), three months (end of the acute BMT period), six months, 9 months, and one year after transplant (designed to capture the late complications and recovery period). The data from the 9-month time point is not included in this analysis because it was not collected in both studies. Both source studies were approved by the Tufts Medical Center Institutional Review Board (IRB) and the IRB at each research center. IRB approval was also obtained to create the dataset for this analysis.

Survey data in this study were collected using the Child Health Ratings Inventories (CHRI)-General, a generic health-related quality of life (HRQL) measure with child, adolescent, and parent versions. The CHRI contains scales to measure HRQL and related constructs: for this analysis global QOL - measuring physical, emotional, and social health; general health; and Parent Impact (PARImpact) (Heinze et al., 2015) were used. The CHRI scales have been extensively validated within the pediatric BMT population (Parsons et al., 2005).

Study Sample

Accompanying parents of pediatric patients aged two months to 18 years who were scheduled to undergo BMT at study sites were recruited for the JTR and HSCT-CHESS™ studies. Dyadic participation of the parent and child was required. Inclusion criteria for parents were: ability to speak/understand English, at least 18 years old, parent or legal guardian of the BMT patient, and ability to provide consent to participate both for him or herself as well as for the BMT patient. Only parent data were included in this analysis; however eligible child participants also provided assent/consent for their participation. Overall, 62% of eligible participants consented to participate. Common reasons for non-participation included parent refusal (44%), child refusal (27%), and medical reasons (3%). Participants did not differ from non-participants based on child age, gender, or race/ethnicity.

A total of 423 parents were enrolled in the JTR and HSCT-CHESS™ studies; 363 are included in this analysis, and the remaining 60 parents were excluded because in the JTR study parents of children less than 5 years old did not complete the CHRIs, completing instead a multidimensional QOL measure, specifically for parents of infants and toddlers.

Measures

Global Quality of Life

The parent version of the Global Quality of Life (QOL) scale of the CHRIs consists of nine questions designed to capture the multidimensionality of QOL as physical, mental, and social well-being. Responses are measured with a Likert-type scale with five response options ranging from “poor” to “excellent.” The scale is

scored from 0-100, with higher scores indicating higher QOL (Rodday, Terrin, & Parsons, 2013).

General Health

The General Health item in the CHRIs is a single summary item of parents' general health appraisal. Parents were asked, "Overall, how would you rate your health"; response options include a five-point Likert-type scale, which is scored on a 0-100 scale with higher scores indicating better health. This item has been used extensively in clinical practice and research (Bailis, Segall, & Chipperfield, 2003), and has been found to be associated with multi-item scales of general health and other markers of disease and clinical outcomes (Bowling 2005).

Parent Impact Scale

The PARImpact was developed as a separate scale within the CHRIs-General Parent Report to measure the response of the parent to the child's physical and emotional health. The scale consists of four items, two that measure the parent's response to the child's physical health and two that measure the parent's response to the child's emotional health. PARImpact items have five response options on a Likert-type scale ranging from "none of the time" to "all of the time." The scale scores range from 0-100 with higher scores indicating greater (more negative) parent impact.

Clinical Variables

Clinical data were collected at all assessment time points by trained study staff, using standardized data collection tools. All clinical data were reviewed by the study PI (SP) for completeness and consistency. Baseline information included time

since initial diagnosis (months), disease category (malignant or non-malignant), and transplant type (related allogeneic, unrelated allogeneic, or autologous).

In follow up, specific clinical outcomes variables included: the presence and severity of acute graft versus host disease (GVHD) through day 100 (Glucksberg et al., 1974); end organ toxicity as measured by the Bearman Toxicity Scale, which is ranked as good, intermediate, or poor, through day 100 (Bearman et al., 1988); local and systemic infection based on the National Cancer Institute Common Toxicity Criteria for Adverse Events v. 3.0 (all time points) (Zhang, Chen, & Wang, 2016); and the extent of chronic GVHD starting at day 100 (Shulman et al., 1974)

Demographic Variables

Parents' demographic data, including sex, age, race/ethnicity, education, marital status, and household income were collected from parent participants at baseline. Parents also reported their child's race/ethnicity, sex, and insurance status.

Statistical Analysis

Demographic variables and clinical characteristics were described for the combined study sample using medians (interquartile range [IQR: 25th-75th percentile]), means (standard deviations [SD]), frequencies, and percentages at baseline. Outcome variables (QOL and general health) were assessed for normality and described using means, medians, IQR, and SD at each time point. Spaghetti plots were created to visualize the relationships between dependent and independent variables.

Cross Sectional Analysis

A General Linear Model was created for each time point (pre BMT baseline, day 45, three months, six months, and one year) and for parent QOL and parent general health to analyze the significant covariates at key transition points.

Independent variables were included in the model based on the Individual and Family Self Management Theory and then backwards elimination with a cut-off of $p \leq 0.2$ was used to identify significant independent variables.

Repeated Measures Analysis

We built two models for regression analysis - one for parent QOL and one for parent general health. In both models, univariate longitudinal regressions were completed with demographic, clinical, and PARImpact variables using Residual maximum likelihood (REML). Timing of repeated assessments was calculated as the number of days since BMT. Clinical variables were also tested with time interactions. All variables that had estimated coefficients with $p \leq 0.2$ were included in a final model and then backwards elimination was used to eliminate variables until all variables in the model had an estimated coefficient with $p \leq 0.1$.

Unstructured and compound symmetry correlation structures were compared using Akaike information criterion (AIC). AIC is a way of selecting a model which is the best fit from a set of models. (Hedeker & Gibbons, 1997) To explore the possibility that QOL and general health responses may have been missing not at random (MNAR), we stratified the final model by the presence and causes of missing data, defining strata as follows: (1) those with missing data due to a medical reason (e.g. child too sick) and (2) those with complete data or those with

missing data not due to a medical reason (e.g., logistical reasons, such as transportation or work-related issues). The stratified models (called pattern mixture models, PMM) (Hedeker) assume the data are missing at random (MAR) within strata. We compared the stratified to the unstratified model using the likelihood ratio test to assess for the presence of MNAR. SAS version 9.3 was used for all statistical analyses; alpha was set at 0.05.

RESULTS

Pre-BMT baseline demographic and clinical characteristics are shown in Table 1. The majority of the sample was female and Caucasian with at least some college education. Most parents reported being married or living with a partner (80%), and reported having at least one child in addition to the BMT recipient (84%). Parent general health was not normally distributed so the scale was reversed and square rooted (Polit & Lake, 2010).

Parent QOL and general health means and standard deviations are listed in tables 2 and 3.

Cross Sectional Results

These results examine the associations between the context and outcomes of parent QOL and parent general health.

Pre-BMT baseline: At pre-BMT baseline, parent QOL was significantly lower among parents who were not married or partnered compared to those who were ($b=5.5$; $p=0.04$); as well as those who had public insurance compared to those with private insurance ($b=-4.7$; $p=0.04$); QOL was significantly higher among non-Whites compared to Whites ($b=5.3$; $p=0.03$). Parent general health (results reported using

the reversed and square rooted scale) was significantly worse among parents who had a high school education or less compared to parents who had a college education or more ($b=0.71$; $p=0.03$); and among parents of children undergoing BMT for malignant diseases compared to parents of children undergoing BMT for non-malignant diseases ($b=-0.58$; $p=0.04$).

Day 45: By day 45 many BMT patients had been discharged from the hospital. At this time point, for each year child age increased, parent QOL also increased by 0.53 ($p=0.03$). Additionally, compared to parents of children who were diagnosed with an infection, parents of children without an infection had higher QOL ($b=5.3$; $p=0.04$). Length of illness negatively affected parent QOL, with each addition month lowering QOL by 2.5 points ($p=0.02$).

Day 90: Day 90 is around the time that most families are able to move back to their family home after living in temporary housing near the hospital. As at day 45, parent QOL was negatively affected by length of illness ($b=2.4$; $p=0.02$), and parents of children without infection had higher QOL than parents of children with infection ($b=9.7$; $p<0.01$). Unlike day 45, at day 90, fathers had higher QOL than mothers ($b=7.7$; $p=0.02$). Parent general health was lower among parents who were not married or partnered compared to those who were ($b=0.81$; $p=0.02$), and higher among parents whose children were undergoing BMT for a non-malignant diagnosis ($b=-0.64$; $p=0.02$).

Day 180: Parent QOL was higher among non-Whites compared to Whites ($b=8.5$; $p=0.01$), and among parents whose children did not have an infection ($b=9.1$; $p=0.02$). Parent QOL was lower among parents who did not report whether they

were local or referred compared to those who were local ($b=-17.2$; $p=0.01$). Parent general health was lower among parents who were not Hispanic compared to those who were ($b=0.92$; $p=0.03$).

Day 365: At one year post BMT, parent QOL was significantly lower among parents whose family income was <\$20K-\$39K compared both to families who made \$40k-79K ($b=11.8$; $p<0.01$) and families who made >\$80K ($b=12.1$; $p<0.01$). In addition, parent general health was higher among families with one child (i.e. the BMT patient; $b=-1.04$; $p=0.02$) compared to families with three or more children, and parent general health was also higher among families with higher income compared to families whose income was <\$20K-\$39K (\$40-\$79K $b=-0.88$; $p=0.02$; >\$80K $b=-1.01$; $p<0.01$).

Longitudinal Results

Selecting the Final Models

After the initial models were selected using backwards elimination, pattern mixture models were created to test whether data were missing not at random (MNAR). Likelihood ratio tests showed that the unstratified models fit as well as the stratified models (Parent QOL: $\chi^2 = 14.4$, $df = 24$, $p=0.94$; Parent general health: $\chi^2 = 13.7$, $df=18$, $p=0.75$) therefore the unstratified models were selected. Based on AIC, an unstructured covariance matrix was selected, and time was treated as a main effect.

Parent Global QOL

Compared to Hispanic parents, non-Hispanic parents children reported lower QOL ($b=-3.95$; $p=0.04$); and parents who were not married or partnered reported lower QOL than parents who were ($b=-4.1$; $p=0.04$). Additionally, parents of only children

reported significantly higher QOL than parents of three or more children ($b=4.8$; $p=0.03$).

For each one-point increase in parent impact (indicating a more negative impact), parent QOL decreased 0.34 points ($p<0.01$). And at day 45, parents of children that had a “good” Bearman score had significantly higher QOL than parents of children with a “poor” Bearman score ($b=8.2$; $p=0.02$).

Parent General Health

Parent general health was worse among parents whose educational level was high school or lower than parents whose education was college or more ($b=0.43$; $p=0.05$). And parents of children who were undergoing BMT to treat malignancies had lower general health than parents whose children did not have a cancer diagnosis ($b=-0.43$; $p=0.03$). As parent impact increased (worsened), parent general health decreased (per half standard deviation $b=0.20$ $p<0.01$). Surprisingly, at day 90, parents’ general health was lower among parents whose children had “good” ($b=1.03$; $p=0.05$) or “intermediate” ($b=1.33$; $p=0.04$) Bearman toxicity scores than those with “poor” Bearman toxicity scores.

DISCUSSION

This study examined parent QOL and general health among parents of children age 0-18 years undergoing blood and marrow transplant, and was guided by the Individual and Family Self-Management Theory (Ryan & Sawin, 2009). To our knowledge this is the first study to specifically investigate parents’ general health during their child’s BMT. We found that parents’ QOL and general health tended to improve over time, but there were several factors that may affect this trajectory.

QOL was significantly lower among parents who were not married or partnered. This is consistent with other findings that a spouse or partner can provide support (Rini et al., 2004.), and highlights the vulnerability of parents who do not have this support. QOL was also lower among parents with three or more children (e.g. the BMT patient and at least two siblings) compared to parents whose only child was the BMT patient. This finding is consistent with parent reports that caring for a pediatric BMT patient while caring for other family members can be challenging (Mayer et al., 2010).

The finding that being married or partnered was associated with a higher parent QOL is consistent with the concept of family context in the Individual and Family Self-Management Theory guiding this study (Ryan & Sawin, 2009). Spouses can support each other in caring for their child undergoing BMT; therefore, spousal support can be strength within the context of a family (Rini et al., n.d.). The support of a spouse may facilitate individual and family self-management. More research is needed to explore whether this caregiver strain could be mitigated with more family support during the BMT.

We also found parents of Hispanic patients had higher QOL than parents of non-Hispanic patients. While Hispanic patients can be from very diverse cultures, another study of Hispanic adolescents with cancer revealed that many of the adolescents and their families trusted that God would care for them and took great comfort in this faith. Also, support from the extended family was reported (Jones et al., 2010). It is possible that Hispanic family background is characterized by family resilience to distressing experiences such as having a child undergoing BMT. Family

cohesion has also been correlated with parent distress in another study (Phipps, Dunavant, Lensing, & Rai, 2005). However, in a recent study (Virtue et al., 2014) Hispanic parents of pediatric BMT patients were found to have higher depression scores than non-Hispanic parents. More research is needed to better understand the interplay of ethnicity, culture, and other factors during pediatric BMT.

End organ toxicity was also related to parent QOL, which is understandable considering that end organ toxicity is related to the need for escalating care and critical illness (Bearman et al., 1988). This provides further evidence that the child's BMT course affects the parent's emotional health (Terrin et al., 2013).

Parent general health was significantly lower among parents with lower educational attainment, which is not surprising given that education can be a proxy for socioeconomic status, and is a commonly identified social determinant of health (Lynch 2003). There also may be specific BMT factors that may contribute to this finding such as the stress of caring for a child with complex medical needs with fewer resources (Mayer et al., 2010). Parent general health was also lower among parents whose children needed BMT to treat a malignancy versus those who needed BMT for a non-malignancy. It's possible this finding is related to the length of illness – a child with cancer may be receiving BMT after multiple rounds of previous treatment, and the family may have less control over the timing of the transplant than a family of a patient with a non-oncologic diagnosis (e.g. an immune disorder). Taken together, these findings indicate the vulnerability of parents before, during, and after their child's BMT. Finding better ways to support families during pediatric

BMT may improve parents' emotional and physical health outcomes, which may in turn improved the child's outcomes as well.

Strengths and Limitations

This study is strengthened by the large sample size and longitudinal design, which represents eight years of research. Obtaining study participants from pediatric BMT centers across the country provided a more heterogeneous sample than is often obtainable in pediatric BMT studies, and resulted in the final sample being 27% non-White and/or Hispanic. Even with these inclusive recruiting techniques, the sample was above average for educational level and income. The study team collected clinical variables through chart review even if surveys could not be administered, which added to the completeness of the data. Study drop out was mainly related to a child's relapse or death, and PMMs were tested to ensure rigorous analysis despite missing data.

Another key limitation is that this study relies on self-reported surveys from parents, which can result in shared variance and inflate associations.

Conclusion

In this study we found family contextual factors such as parents' ethnicity, education, number of children, and marital status are related to parent QOL and general health. The child's clinical course (e.g. organ toxicity) is also related to parent QOL and general health. Understanding the family context of pediatric BMT may help clinicians to provide more comprehensive care to patients and their family members. Family focused interventions may improve parent QOL and general health following a child's BMT.

Table 1: Participant characteristics

	n	%
Parent Race		
White	282	78
Other	81	22
Child Race		
White	296	82
Other	67	18
Income		
<\$39K	120	33
\$39K-\$79K	114	31
>\$80K	129	36
Parent Education		
HS or less	112	31
Some College	108	30
College +	143	39
Marital Status		
Married/partnered	292	80
Other	71	20
Insurance		
Private	235	66
Public	121	34

Table 2: Mean Parent QOL

Variable	N	Mean	Std Dev	Minimum	Maximum
QOL Day 0	363	50.3	19.7	2.8	100.0
QOL Day 45	316	48.4	20.6	2.8	100.0
QOL Day 90	298	50.4	20.8	2.8	100.0
QOL Day 180	264	54.3	21.8	5.6	100.0
QOL Day 365	232	61.1	22.7	0	100.0

Table 3: Mean Parent General Health

Variable	N	Mean	Std Dev	Minimum	Maximum
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Variable	N	Mean	Std Dev	Minimum	Maximum
Parent General Health Day 0	363	63.4	22.6	0	100.0
Parent General Health Day 45	316	58.6	23.0	0	100.0
Parent General Health Day 90	297	57.7	21.6	0	100.0
Parent General Health Day180	266	60.1	22.4	0	100.0
Parent General Health Day 365	235	63.4	22.1	0	100.0

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Chapter 4: Manuscript Three: “Nothing Could Really Prepare You for This”: The Experience of Family Self-Management During a Child’s BMT

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INTRODUCTION

Background

Pediatric blood and marrow transplantation (BMT) gives families the hope of cure for their seriously ill children with oncologic, immunologic, and hematologic illnesses, but it is also an intense treatment with life-threatening side effects. The BMT process is also complicated because it requires patients to spend about a month in the hospital, followed by two additional months in close proximity to the transplant center, either at home or in temporary housing near the hospital so the patient has around the clock access to specialty care. For pediatric patients, this means that a parent or guardian must accompany them at all times.

The challenges of BMT plus the transitions between hospital, temporary housing, and home can be taxing on a family's physical and emotional resources (Mayer et al., 2010; Packman, Weber, Wallace, & Bugescu, 2010). Pediatric BMT patients and their family members have reported decreased QOL, anxiety, depression, post-traumatic stress symptoms, and decreased self-esteem (2010). Furthermore, these effects can linger beyond the acute transplant period; family members have reported long-term physical, emotional, psychosocial, and mental health sequelae (Barrera, Atenafu, Doyle, Berlin-Romalis, & Hancock, 2012a; Clarke 2011; Drew, Goodenough, Maurice, Foreman, & Willis, 2005; Manne et al., 2003).

Despite the intensity of BMT that is experienced by all family members, the family experience of BMT has not been robustly explored in the literature. Variables that have been linked to family outcomes include patient and parent age; income level and

employment status; and number of siblings (Barrera, Atenafu, Doyle, Berlin-Romalis, & Hancock, 2012b; Hovén, von Essen, & Norberg, 2013; Jobe-Shields et al., 2009; Manne et al., 2003; Phipps, Dunavant, Lensing, & Rai, 2004; Zois 2011). A critical review of the literature concluded that more research is needed to examine family variables that could be targeted for family level interventions during BMT (Packman et al., 2010). This is consistent with other calls for inclusion of family level research in the oncology literature (Hinds 2012). Therefore the purpose of this study is to explore the family experience of self-management in the first year after their child's BMT.

Theoretical Framework

The Individual and Family Self-Management Theory (IFSMT) (Ryan & Sawin, 2009) was chosen to guide this study. The IFSMT is a mid-range descriptive systems theory with three main components: context, process, and outcomes. In the IFSMT, self-management is described as a process by which individuals and families achieve health-related outcomes. Examining self-management among families of pediatric BMT patients will provide vital information regarding the family experience of pediatric BMT, and may lead to the development of family level interventions.

METHODS

Overall Approach

Family-as-group interviews were selected to allow the inclusion of the maximum number of perspectives in each interview (Eggenberger, 2007). Family interviews have been conducted in other settings, such as intensive care and adult oncology, and provide insight into the family experience and family dynamic. Interviews were conducted using a phenomenological method and were guided by Max Van Manen's human science

approach (Van Manen, 1990). This approach is widely accepted as an appropriate method when little information is known about a topic, and the researchers wish to understand the experience or perspective of others (Rose et al. 1995).

Participant Recruitment

Participants were recruited from the pediatric BMT department at an urban academic medical center in the Mid-Atlantic region of the United States that completes approximately 50 BMTs per year among pediatric and young adult patients. The researcher identified possible participants by searching electronic medical records, searching lists of recent pediatric BMT recipients, and attending a weekly case conference. Inclusion criteria included: families with a child age 0-17 who had undergone BMT in the previous year; able to speak and understand English; at least six years old to participate in the interview; and able to provide consent (participants 18 years or older) or assent (participants younger than 18 years old).

Once an eligible family was identified, the interviewer asked the child's clinician (typically an RN, PA, or PNP) to ask the child's parent if the family might be interested in talking to a research nurse about a study involving families with a child who has undergone BMT. Once verbal consent was obtained to approach the family, the interviewer gave a brief description of the study requirements, possible risks, and study aims. Any questions were answered and if any family members indicated they were interested in joining the study a time and place were identified to complete the interview.

Conducting the Interview

All interviews were conducted in a private setting of the participants' choice, which may include the inpatient, outpatient, or home setting. When children were present, prior to

the interviews, the interviewer asked parents if there were any topics that she should not bring up during the interview. Parents consented both for themselves, and for any minor children who chose to participate. Assent forms were given to any participants younger than 18 years old. Crayons and paper were supplied for anyone who was interested in drawing during the interview and particularly for younger children. All interviews were recorded digitally. All participants were informed that if they did not wish to answer a question they could ask to move to the next question and parents were given a signal to give the interviewer if they wanted to move to a different topic. A semi-structured interview guide, which was developed based on the IFSMT and quantitative analyses, was used to conduct the interviews. Pediatric BMT social workers were on call in case a situation arose in which a participant became upset and needed additional support.

Analysis

Interviews were transcribed verbatim and each interview was read multiple times by the interviewer to obtain a sense of important themes. Each interview was then coded line by line and sections that seemed particularly salient to the study question were highlighted as prescribed by Van Manen's approach (1990). A priori coding was used with Van Manen's suggested categories of lived space, lived body, lived relationship, and lived time (1990). These a priori themes were a propos to the family BMT experience and provided a starting framework for coding. If a passage did not fit in one of these themes, a new theme was created rather than trying to create a way to make it fit into the a priori codes. After initial line-by-line coding was complete, themes were compared across the four main categories and similar themes were combined into larger categories.

After the first three interviews were coded, the interviewer met with a qualitative expert (JW) to discuss the analysis and findings. The interview guide was reordered and shortened to focus on the most salient topics. All interviews were coded using *f4analyse* software. Modified member checking was used by asking families about emerging themes at the conclusion of each interview. In order to maintain anonymity during the analysis period, each family was given a pseudonym family name in alphabetical order – so the first family was given a pseudonym that started with an “a”, the second a pseudonym that started with a “b” and so on. However, the study team decided not to report these pseudonyms because they felt too personal, and may have been identifying to the clinical staff at the study center.

The institutional review board approved this study.

RESULTS

Participants

Sixteen families were identified as eligible to join the study, and all sixteen agreed to meet the interviewer. Although all sixteen families expressed interest in joining the study, scheduling logistics were insurmountable for three families. The remaining 13 families joined the study and completed the interview (See Table 1 for participant information).

The thirteen participating families represented 11 mothers, 5 fathers, 4 BMT patients, and 4 siblings. Among the family members who could not participate, 2 mothers were unavailable at the time of the interview; 7 fathers did not participate (1 declined and 6 were unavailable, mostly due to work schedule); 9 patients did not participate (2 refused to participate because they did not feel well enough, and 7 were not old enough); and 17

siblings did not participate (9 were living out of state, 4 were unavailable, and 4 were not old enough).

Eight interviews were completed in the outpatient setting; two interviews were completed in the inpatient setting; two were completed on the phone, and one was completed in the family's home. Interviews ranged from 39 minutes to 125 minutes.

Themes

As the interviews were analyzed, it became apparent that even though these families' experiences overlapped due to the shared context of BMT, the processes for managing the demands and challenges of BMT were vastly different.

Making a Plan

Once families were told their child would need a BMT, they began to rearrange their lives to accommodate the demands of BMT. This included every facet of life, from making sure the family dog was taken care of, to making decisions about employment through BMT.

BMT Timing: Some families were able to schedule the BMT for a time that was best for them, "...we wanted to wait out the flu season, just because we do have the luxury of time, we're fortunate in that respect, so we thought May would be a good time to go ahead and start the process" (Mother, 43 years old; paragraph 16).

Other families did not have the opportunity to plan ahead such as one family whose son hadn't left the hospital since his diagnosis:

Father (46 years old): "It was about four days of like 103 fevers, so we knew it wasn't - the Tylenol wasn't getting rid of it at all, so it seemed like there might be something more going on. It was like let's not wait another day to take him to the

pediatrician. We should go tonight. I got home from work and I took him [to urgent care].”

Mother (42 years old): “Never again did we see my son and my husband at home ever since then” (paragraphs 18 & 19).

Families who had been undergoing chemotherapy prior to BMT described using the chemotherapy experience as a guide for preparing for BMT.

Mother (52 years old): “I think because we did that first salvage where he was in the hospital five days at a pop, five or six days, and then he was also that other time in the hospital and, you know, we kind of had the drill down.... if we went straight from [outpatient chemotherapy] to BMT that would have been a big shock I think, but we had already had practice sessions “ (paragraphs 858 & 869).

Even though families agreed that previous hospital experience was helpful during BMT, they also described feeling unprepared for the intensity of the BMT:

Mother (43 years old): “Yeah, I think part of it was helpful knowing, having expected, I pretty much stayed with [patient] with all the hospital stays. So the two of us were like twins at that point as far as, you know, she's very comfortable with me in the hospital setting and she was very comfortable being there, helping her and talking with the doctors. I understood like what she needed comfort-wise and to get her through certain procedure and stuff. So that was really good. I think as far as preparing for it, that helped a little, but I don't, even when I look at it now, like nothing could really prepare you for this” (paragraph 104).

Preparing for the Hospital Stay: Many families did their best to think of ways to make the hospital stay more enjoyable for their child, and older patients also participated in planning for the hospital stay. Some families made extensive preparations:

Mother (40 years old): “You know most the things I prepared was for [patient]. I was trying not to let him feel that he is at the hospital environment. So what I did, I did a lot of shopping for him, so I took him, if you noticed he is in a different theme every day. So I tried to match the theme, the bed sheets with his shirts, his hats, because I knew he was going to lose his hair, his hats, his socks, his shirt, and his PJs, and I chose the ten different themes for him... I was just, I didn't want him to stay in the white sheets and feel he is sick and he is at the hospital environment.” (paragraphs 120 & 122).

While others planned more simple ways to pass the time in the hospital, such as the one family who made a game out of placing as many sets of little plastic toy eyes around the hospital as possible, making it seem like inanimate objects had come alive:

Interviewer: “So when you were in the hospital what do you feel like helped you?”

Patient (17 years old): “Googly eyes.”

Mother (48 years old): “We sort of figured it out before we even came in. “

Patient: “Yeah, I was like, it'd be fun to place random eyes around here”

(Paragraphs 736 & 800).

Making Plans for other Children: In addition to preparing for the hospital stay, many families had to make a plan for other children to be cared for during the BMT. Some families who lived near the transplant center were able to find friends or family members

to care for the children on a part-time basis. For example, one sibling was able to be with her stepfather after school every day. In another family, there were grandparents available to care for their well son when neither parent was available.

However, not every family has this option. One family lived in a different state from the transplant center, and in addition to the patient, the family had eight other children:

Mother (43 years old): “It was the summer so [some] kids went to camp and some of my kids went to like siblings in [a different state], but my baby still - like I was very concerned about him... I got to know somebody here very well ...and she offered to take my baby...so then I could see him much more often and take care of him and hug him and kiss him, what a baby needs...from a mother”

(Paragraphs 50, 52, & 54).

There was one family that chose to send their well son to stay with out of state relatives for the first 100 days of their son’s BMT:

Mother (45 years old): “I was just grateful that our son was away all summer having a massive adventure with various family members doing all kinds of fun stuff. You know, he went to football camp, fishing, boating, driving the tractor, on the river, just, I mean just he was having a great time. And that was really the first and foremost, that was most important, was to make sure he was completely taken care of while we go through this so that we could focus on [the patient] and not worry about [his sibling] being neglected and not getting all the love and attention that he rightfully deserves” (Paragraph 44).

Other families described how important it was for them to stay together through the transplant. One family was homeschooling their four children before their son became ill, and they chose to continue homeschooling from the hospital:

Mother (42 years old): “We're [in the hospital] with him every day and we really try to manage every day just like we do at home. By that, I mean, we do homeschooling here, homework...they do piano, too, so I teach them piano so they practice here every day...We just try to instill childhood as much as we can, normalcy.”

Father (46 years old): “You're not always feeling like she's going to rush to get here in the morning or figuring out what to do about after school and that because we had that built-in flexibility already in our day, so it makes the managing of that daily juggling of the kids a lot easier.”

Sibling (9 years old): “It feels a little bit different than our home. Like except for that, it's basically the same, doing the same things. We do it with our family”

(Paragraphs 69, 84 & 91).

Taking Care of Basic Needs

Even though the acute period of BMT treatment lasts about three months, it is not possible for families to put their lives on hold for this time. There are still bills to be paid, work to attend to, and meals to be made. Families often needed the support of their friends, family, and communities to manage the intense demands of this time.

Working out Work: Two-parent families reported it is nearly impossible for both parents to remain employed during a child's cancer treatment and BMT without accommodation from the employer. This can be stressful for families because their financial security can

depend on the willingness of a supervisor or an organization to be flexible with work arrangements, but this is never guaranteed. Most families reported prioritizing the employment of the family member that held the health insurance because that was foundational for the child's treatment.

Among the families in this study, there were three parents who owned a business, which afforded some flexibility, but it was also stressful for parents to be caring for an ill child in addition to running the business.

Father (51 years old): "We are probably one of the few families that could get through it with both of us still working. It was only because of the situation where I had the ability to say... "I need off." I'm one of the owners of the business. I said 'I'm not coming in. I know I can't be away all the time, but I'm not going to be here.' Her grandmother and her uncle are the other partners in the business. It's not like they're going to say, 'Hey, you can't go do this.' We were a unique situation, but even that said, it takes a lot of people to help through when you're talking about 90 days, 90-100 days. That's if everything goes well" (Paragraph 238).

Three parents got permission from their supervisor to telecommute from the hospital, but these agreements were usually temporary and somewhat tenuous.

Father (46 years old): "Initially, I used up basically almost all my vacation time at the beginning because we didn't know what was going on. I talked to my supervisor [and] I was able to try to set up with him, some long-term projects we had, so things that I could do remotely that I didn't feel like I had to physically be there in the office to do some of this project kind of work. To me, it was working

good. I had worked out a sort of time and schedule here that worked... finally that situation ended and [I have been] forced to start that FMLA clock... basically there seemed to be this one individual who decided that, 'I don't want him to work remotely, or to telework the whole time.'" (Paragraphs 111 & 117).

Father (51 years old): "We really didn't have anything in place other than I said to my wife, 'I've got flexibility, but I can't be down here nonstop for three months. You're going to have to help.' She worked it out with her boss where she could work Thursdays and Fridays remotely. I don't think that she explained it to her boss that it was going to be as long-term as it was. That caused some problems after awhile" (paragraph 111).

Eight parents reported they had to either scale back or quit work all together to accommodate the caregiving needs of their sick child.

Mother (43 years old): "I worked from home as a consultant and then I basically stopped it after probably two weeks or so after [daughter] was diagnosed and we started treatment" (paragraph 36).

Most often the person to reduce work hours was the mother, but in one family the mother carried the health insurance so it was the father that had to cut back on his work hours. However, the mother still wanted to be involved in her son's care so she split her time between work and the hospital:

Mother (39 years old): "I stayed with [patient] at night, and the morning we switch because I have to go to [work]."

Father (41 years old): "So I woke them up, I woke them up at five, got various food for [sibling], and then for my wife to get whatever she needs, got my son off

at six [to the grandparents' house], and go drive all the way to [the hospital]. Then we switch. I stay with [the patient] from then on until she gets back from work” (Paragraphs 278 & 291).

Some parents' employers were very accommodating to the parents' needs during their child's BMT:

Interviewer: “Did he take any formal time off or was it just kind of the job flexibility?

Mother (45 years old): “I don't know how they worked that out. He's been with the company such a long time, he's been with them for like 20 years. So he's up there on the totem pole, so he was, he's definitely able to, you know, to just work it out. Although I think, it's such a nice company, I can't imagine, no matter where you are, them not understanding that. They've been beyond gracious about [patient's] situation. Like they could have moved us so many times and they didn't because they want us close to [the hospital].

Most participants in this study reported that they were able to make ends meet even though they were not earning their usual income. However, not being able to work was devastating for one family:

Interviewer: “So when she got sick did you kind of have to put work on hold?”

Mother (21 years old): “Yeah. First I was trying to do overnight [shifts] because [patient] was living at the hospital and then it wasn't working because she didn't want me to start leaving. So I had to stay, so I had to stop everything... I didn't have nobody. I was a single mom” (Paragraphs 42 & 52).

Two families that were interviewed were receiving public assistance because their child's illness made it impossible for the parents to work.

Mother (19 years old): "like [the patient] basically has checks for herself, actually, it's like social security. She has checks for herself so that's like, if anything, once a month if she needs anything I get it with all her money. So she really is set, it's just us eating and everything, but that's fine" (paragraph 287).

Relying on Others: Every family that participated in this study spoke of relying on friends and family to help with childcare, meals, pet sitting, yard work and more. Some families had vast networks of helpers, and some had very few, but no one was able to live through their child's BMT without help.

Mother (43 years old): "Yeah. No, [the community] was amazing. I mean, they are. They still are, the people. I mean, I have like - right, just even just my neighbors, they're amazing. Like my four-year-old, he was three going on four, so he would finish school at 2:00 so he's - every day at one of my neighbors until 5:00 until the rest of the kids came home and he says, 'Yeah, she told me that I'm like a triplet to her kids.' She has like two boys his age or like around his age. 'I'm like her triplet.' I'm like okay. And he was so comfortable there." (paragraph 494).

Mother (34 years old) "[Our friends] did like a meal train thing and like we had food for a long, long time, more food really than we ever needed. And they would - just come visit [my son] and visit me and bring him gifts, decorate - they made him - when we first admitted - for like induction they made his whole room into a

bat cave. There were bats everything and so they were very helpful” (paragraph 246).

Mother (21 years old): “[We had] a lot of support on the outside...I didn't have no money for Christmas.were just coming home. It was a charity. It helped us out. Bought my daughter toys, clothes, shoes, everything - I was so thankful” (paragraph 140).

The pediatric BMT patients also appreciated the support and camaraderie they had from friends during their BMT even though at times it was difficult to stay connected when they weren't feeling well.

Patient (17 years old): “[I'm] also really happy that I'm at an age where my friends can drive themselves and have their own cars and stuff, because like if I had to rely on my friends to have someone to drive them up here and wait while they visited and stuff, I think I'd have a lot less visitors. But with my friends driving themselves it's really been nice, because I get a lot of visitors paragraph 385).

Interviewer: “So have you kept in touch with your friends on social media or how do you manage that? “

Patient (17 years old): “Mostly when I was in the hospital was through texting and some social media. And my best friends visited me.”

Mother (48 years old): “And Instagram. One of his friends kept everybody up to date” (paragraphs 574 & 587).

Patient (14 years old): “I kept up with my friends.”

Father (51 years old): “She did that some. She didn't have her own phone at the time, but we did a lot of Facetiming. My sister has five kids, so she has five cousins that are pretty close to her and they would Facetime her, maybe not every day, but a good, at least five times a week or so.”

Patient: “And then, when I got my head shaved, [my uncle] and [my cousin] both got their heads shaved. Now, theirs grew back in like three or four days. Mine is still growing” (paragraphs 172-174).

Even though families described feeling grateful for the help they received, some families decided to limit the amount of information they shared, and limit the people involved to trusted friends only.

Mother (42 years old): “The reason why we kept it private is because we felt that we needed positive energy and those people just saying, ‘Oh, my gosh, I can't believe it. What a nightmare! Oh, what you're going through.’ We don't need those kind of comments. We needed somebody who's very spiritual, who has good positive energy and was very helpful without these negative extra comments, with them noticing they're doing that. I know they mean well and also, because we felt that we're dealing with so much here, why should we spend time on the phone talking to people about how he's doing every minute and not only that, there were times where every hour, his situation would change. So imagine if we had to tell people, ‘Oh, now he has a fever. No, he doesn't have a fever. Now he's throwing up. Now he cannot walk. Now he can do this.’ No way” (paragraph 133).

The experience of the BMT Community: Many families described finding support among the other families whose children were undergoing BMT at the same time. They often met each other while in the hospital kitchen, laundry room, at the temporary BMT housing near the hospital, or in the outpatient clinic. For many participants, having a friend who was in a similar situation was very comforting, and having someone to talk to who understood all the medical jargon was refreshing.

Mother (45 years old): “So it was just really, you know, your heart grows bigger for these other families, and you learn from other families too about how they deal with stuff, and what they're doing. And like this one family, we became good friends with, you know, I just thought it was so great, they had visitors almost every night...so I really learned from that too, it taught me that, you know, just because your bone marrow transplant doesn't indicate fun on any level, but you can find joy in it. And so they did teach me that while I was there” (paragraph 286).

However, for some participants, it was difficult to bear the emotional burden of their own child's illness, plus the emotional burdens of other families.

Mother (52 years old): “Yeah, there's another family there - Oh, actually first when we were in the [temporary BMT] house here, right we got to meet a lot of families, that was really, it was nice to just, I don't know, you don't feel like you're alone. I don't know-

Father (54 years old): “I think it was nicer for you than it was for me...I didn't really need to listen to other people's misery, to be honest. I mean it doesn't really change my life when someone else has a cancer too... “

Patient (17 years old): “I’ve always been of that mindset too, like I, the social workers really tried to get me involved in lots of things and I have absolutely no interest in doing any of that” (paragraphs 553-559)

And for some participants, being a patient on the BMT unit was a lonely experience.

Both of the younger mothers who participated in this study described feeling lonely on the BMT unit.

Interviewer: “Was there anybody there to support you when you were having a hard time?”

Mother (19 years old): “I’m going to be - I’m going to be honest. I really didn’t have nobody” (paragraphs 205 & 206).

Interviewer: “When you were going through all the hard stuff, do you feel like you were supported by people here?”

Mother: “Yeah, I was supported by people here. But it’s like, I don’t know, I feel like I always, if I talk to somebody, I’m talking to somebody, because I care, I really care. But vice versa, I don’t get that in return, I don’t ever get that, so I don’t know what that feel like. Even I don’t, I don’t need it, but it would be nice to have it, like but, I ain’t never, like I did not ever like care. Like I didn’t, like it’s all right for people to check on me, like my mother and then, they checked and make sure I was all right, but like how I talk to people and made sure they was all right, like their scary situation, nobody never did that to me, or for me, so (paragraph 219 & 220).

Managing Emotional Burdens During BMT

Just as families managed the day-to-day aspects of BMT in unique ways, they also approached coping with the emotional demands of BMT in ways that were congruent with their individual and family values.

Living with Family Separation

Among the 13 families that participated in this interview, four lived close enough to the transplant center to go directly back to their family home after BMT. The remaining nine families needed to live in temporary housing before returning to their family home.

Dealing with this separation, plus the caretaking needs of the patient, was a challenge to many families.

Mother (43 years old): “Like at home I'm very active like with the school and things and picking them up and being home with them. So I naturally am there and hear about what happens in school, you know, what's kind of going on. But now, when we see each other on the weekends, it's great, it's kind of back to normal, but even when we speak on the phone during the week, you don't get that same level of knowing what's going on or sharing is a little different because it's been quite a while...the time we share together are the two to three days here and I actually sleep in the room with [patient] and [father] sleeps in the other second bedroom with [son]. So it's still physically, we're still physically separated and the time we spend is with the kids. But we figure there'll be a point when we can spend time together again and that'll be when we're back in the house and [patient] is, we can do some babysitting option things again. We figure that time will come down the road, yeah” (paragraph 198).

Mother (43 years old): “I guess being like a long-distance mom [is hard]. Picking up the kids in the morning, making sure everybody got out to school. I was very hands-on in that way. I used to make sure like say goodbye to each kid in the morning and to make sure that they had what they needed, even though I wasn't right there. That can sometimes be very stressful because when things weren't working the way they were supposed to be working ... I'm not there. What am I supposed to do? There's no milk in the frig, so what am I supposed to do? I can't make you milk.”

Interviewer: “Do they still call you and say, ‘Mom, there's no milk in the frig?’”

Mother: “Yeah. No, they totally did. Of course, I took care of it. I called a friend or a neighbor or - and people were helpful, but it just - those kinds of things got me really stressed or one of my kids had a bad day and I wasn't there to like give him a good hug and kiss and be there for them. I think that was probably a really hard part” (paragraphs 396-414).

Managing Relationships during BMT

Relationship challenges were related to family separation, but even families who were able to stay in their home experienced relationship strain. Husbands and wives, parents and children, and siblings all experienced changes in their relationships during BMT. Before their child became ill, families generally worked together to make sure that everyone's needs were met, and it seemed realistic to meet everyone's needs. But many families described that during BMT, taking care of the patient's needs and working together for the patient's survival took priority.

Mother (40 years old): “[My husband’s and my] life before the transplant was very, very close, very close. [We were] friends and a couple, were very close to each other, he was everything, really, for me and the same for him. After the transplant, our life is very different. Maybe, I think I am depressed, I do cry, sleep less, eat less, not taking care of myself at all, not even how I look...[Father] is already a quiet personality so he kept it all inside. He turned to be even quieter than what he was. We get further and further from each other. It did affect our relationship a lot, a lot, to the degree that I did not expect...It's a lot of stress that I don't think we did handle it well. We handled it well to the degree that [patient] will survive” (paragraphs 266 & 268).

Mother (34 years old): “Oh, I think [BMT] is the worst thing for your marriage ever in life...And there is no time and...I won't make time because it doesn't – [the marriage] will be there. You know what I mean? Like I need to do what I need to do for [patient] and like my mom is like ‘Yeah, but you need to work on your marriage.’ I'm like ‘In my spare time?’” (paragraph 383 & 389).

Mother (39 years old): “Having a sick son. So it may be very abnormal to others but it's normal now for us and it's a part of our life now. “

Interviewer: “So if you were going to tell other couples, or give them advice on how to keep your relationship strong through an experience like this, what would you say?”

Father (41 years old): “I would say understand each other. I mean try to be patient with each other I guess, try to, I mean if it's not going, just don't feel bad if it ruins your relationship at first. I mean if it's a heated battle, I mean granted everyone

messes up every now and then, it happens, it happens to everybody. Just try to rebound and try to remember the focus is for the child as much as possible” (paragraphs 762-764).

Families described doing their best to make life as normal as possible for siblings through BMT, but even so siblings’ lives changed drastically:

Sibling (10 years old): We'd done everything fun before he came along...for me it went all downhill when [patient] came (paragraphs 3 & 5).

Interviewer: What's a hospital birthday like?

Sibling 1 (9 years old): “It's not the same.”

Sibling 2 (11 years old): “Not exactly the same, but it's still okay. “

Interviewer: “Did you have a family party here?”

Sibling 1: Yeah. We invited friends and some family and stuff and the hospital makes it fun and all the nurses come together and sing us Happy Birthday (paragraphs 158-162).

Mother (40 years old): I'm busy, I'm very, very busy with [patient] and [sibling] is not happy with that” (paragraph 250).

Mother (34 years old): “yeah, [2 year old sibling] is kind of a mess. He still is kind of a mess...he would sleep with [father] every night. So now, even now at home he sleeps with [father] every night or me or both...he won't go to sleep unless he's in our bed... But yeah, he started stuttering for a while when all this happened and - kind of a tick blinking where he blinks his eyes strangely (paragraphs 281-289).

Mother (45 years old): “I felt bad because, you know, I had to tell [sibling], he was crying, we just couldn't even console him, and I said, you know, it's going to be okay, I said, but we're not quite ready for you yet, just want to make sure we're home, we're ready. And he's like, ‘why aren't you ready?’ I'm like, because we're still not home yet. And so I had to explain that to him like six times” (paragraph 276).

Even though the majority of families described relationship difficulties as the result of BMT, some participants reported positive changes as well.

Mother (48 years old): “My marriage is stronger. When [patient] came down with the [cancer] I knew that that type of illness can push you apart, too. So I made a concerted effort or making sure that we talked and luckily, we're talking people...I have - will say is having time away made coming back together sweeter... We never really flirted by text and we started flirting by text on the phone...And then when he came up in - is - a lot of times when he came up is I just sat next to him and held him. I mean - and my husband has high blood pressure and it made a marked difference of just spending time connected to each other – the feeling of just holding each other. [The kids] would play games and we would sit and read with our Kindles and stuff, but I would lean against him and it made a huge difference” (paragraphs 1073-1088).

Finding strength in spirituality

Many families reported that having a faith belief was a source of strength through the BMT experience. Whether it was more abstract, like a general belief that God would help

them, or if it was more concrete, like prayers being part of their daily hospital ritual, spirituality was important to many families' coping.

One family viewed their child's BMT as a spiritual challenge:

Mother (40 years old): "And we do consider it as a test. That's how we took it. It's a big test, it's a big exam, and a big challenge, that we did not prepare well for it, but we were trying to do all the homeworks [sic] during the exam, trying to answer the questions, trying to be closer, and read more, and know more, and hoping for the best. Really, really hope that we will get out of this experience with a very positive thing. And I even told [patient], recently he was like, why did this happen mommy, how could this be a test? I said, you don't see it now but in the future you might realize it. You will be fortunate in something, you will be a unique in something. Because even now you are more mature than other kids in your age, so you will see it later. There is something that you will see later, and we will see that we don't know yet" (paragraph 365).

Other families expressed that, with the challenges that being seriously ill presented, faith became even more central to their lives.

Mother (48 years old): it's a deeper connection with my faith ... if you ask for God to save your son and he dies and you lose God and your son, you're screwed. I mean, if you - you know, you lost both. So we both came to - and we talked about it, is if you had to give up one or the other and I actually made him go first and is - he actually said is he would sacrifice [himself] versus losing God. And I said, I had to call - that's the same I came to is that you have to have that faith. And with the [cancer] I had to come to a point where God's will be done. Because

I can't lose both because if there's a heaven you're not going to see them again. It's gone, so you have to have faith to have heaven (paragraphs 1110-1122).

Mother (42 years old): Yeah. I wouldn't know what to do without faith. I would not know what to do. Yes, I would, like I had moments where I just said, "God, are you really there? Come on, I'm asking you," because he had some really rough moments that was really scary, but then, I realize every time, [patient] was a champ and how he came out of it. Even doctors scratch their heads about things he didn't get that they thought he would get, for sure ... I so see and I so believe in God even more. I always did, but faith is, in my opinion, is the soul of it all.

Father (46 years old): There's so much that's out of your control. There's so much that, as my wife was saying about the fates, it's knowing that there's some greater power out there, there's something that's beyond you, it helps you stay grounded in that way, if you already know that there's something greater than you and there's so much out of your control, the feeling that sometimes in a situation like this, it helps from getting too overwhelmed. Obviously there's moments where it is, but it helps you stay more grounded and focused, regardless of what you do as a support ... We can't wave our hands and fix things, nor can the doctors in that sense, so knowing that there is - even at some points we were low and getting discouraged and things like that, suddenly something would happen in his body positive, out of the blue. Suddenly, we're at these low points and suddenly that day, he decides to have neutrophils. He doesn't have it for the proceeding days afterwards, but that day, when you needed something good to happen, there's a spark of something. There's things you can't explain.

Sibling (11 years old): Now we even have more faith than - we believe even more from all the things that happened to [patient] and how he's come out of everything and he's been a champ and everything. So, our faith is probably even grown stronger in this situation than worse, I would say.

For other families, faith was a more subtle part of their lives during BMT, but they described feeling supported by prayers, their faith communities, and their faith leaders.

Mother (52 years old): You know I'm very culturally Jewish, I'm not very religious. So it's not like I, it's so funny all the people who say they pray for us from every religion, we've had them all, and I feel like, well let's see, if I didn't really have faith before, but now it's like, well if a thousand people have prayed and we're still here, I don't know, like it really does work. So, but that said, I do love the support and our Rabbi has come to visit a couple times and [patient] appreciates that, but he comes more as like a family friend, not as a spiritual leader, right?

Patient (17 years old): He, in the two visits that he's had he's probably been here a total of three and a half hours and maybe had about eight minutes of Judaism ...He, like I don't think he could come and just completely not mention it (paragraphs 962-966).

DISCUSSION and IMPLICATIONS FOR PRACTICE

This study examined the family experience of self-management during a child's BMT, particularly during transitions in care. Identified themes include Making a Plan, Taking Care of Basic Needs, and Managing Emotional Burdens During BMT. This study was guided by the Individual and Family Self-Management Theory, and themes across

families focused heavily on (1) contextual factors, such as parents' employment and issues surrounding the BMT patient's diagnosis and treatment plan; and (2) the processes families employed to deal with the practical and emotional challenges of pediatric BMT. Even though outcomes were not the main focus of this interview, families were focused on child health outcomes, and how the family had changed as a result of the BMT.

When approaching an impending BMT, families tended to face similar questions: "Where are we going to live?" "Who will take care of siblings?" "How will we handle lower income?" However, even though the context of BMT was fairly stable among families, the processes families chose to manage the BMT experience were vastly different. This is important for the care of BMT families because it means that clinicians need to assess each family's preferences for managing the family during their child's BMT, and then help the family implement plans that are congruent with the family's values.

As would be expected, families with more resources had more options for managing during BMT. Having more financial resources, more job flexibility, and more support from friends and family gives families more options for dealing with the demands of their child's BMT. Understanding that a lack of resources could make a family vulnerable to more stress and difficulty coping is important information for BMT clinicians to consider. This is congruent with a recent study, which found that the financial challenges of BMT disproportionately affects low income families, and that 38% of BMT families experience material hardship (Bona et al., 2015).

There are also policy implications associated with the finding that parents' employment during BMT varies greatly and may depend on the kindness of an individual boss or organization. Even though families may take advantage of the Family Medical Leave Act

(FMLA), this program only provides 12 weeks of leave, and this is not sufficient for a parent who is caring for a BMT patient. There need to be policies in place to provide families the confidence that they will be able to both provide for their family's finances, and care for the needs of their children. Furthermore, if medical insurance for the child was not linked to the parent's employment, it may relieve some of the pressure parents feel to work during their child's BMT.

Another key finding of this study is the difficulties families face while they are separated for the BMT. Families struggled in the short term with practical concerns and lack of emotional closeness, and then after the initial separation was over, the family still had to deal with addressing longer-term relationship challenges. Understanding that these challenges continue past the initial separation may help clinicians identify vulnerable families and provide resources beyond the initial separation.

Even so, the BMT was not a universally negative experience for some families, which may indicate it is possible to experience growth during BMT. Another study identified the concept of post-traumatic growth (Riva et al., 2014) that may be similar to the experiences of these families. More research is needed to more fully understand the factors that are associated with positive outcomes after pediatric BMT with the goal of creating family-focused interventions during pediatric BMT.

Because families manage the challenges of pediatric BMT in differing ways, it will be important for BMT clinicians to assess each family's individual goals and values.

Helping families meet their goals during a child's BMT may also be associated with more positive outcomes. Families in this study also reported that they appreciated any special accommodation that could be made for their family. This is congruent with the finding

that parents of children at the end of life wish for hospital staff to know their family's preferences, and treat them accordingly (Hinds et al., 2009).

Finally, families in this study universally reported that a support system was necessary to handle the demands that BMT placed on their family. Usually this support network was comprised of family members, friends, neighbors, and faith communities. This highlights the importance for clinicians to assess the support networks of BMT patients, and the need to intervene if a family does not have adequate support.

Strengths and Limitations

This study was strengthened by the family-as-group interview method, and development of study questions was informed by the IFSMT. Furthermore, using Max Van Manen's human science approach provided a rich context for the analysis. Obtaining the perspectives of multiple family members provided a more complete understanding of the family experience of pediatric BMT. However, even though the researchers feel saturation was reached, these interviews were cross-sectional, and conducted at one transplant center. It is possible that there are other key concepts that were not discussed in these interviews, and that the concepts discussed in this paper are not applicable to pediatric BMT families in other settings. Finally, even though the phenomenological approach requires researchers to "bracket" their personal emotions and experiences, the interviewer for this study completed the interviews and analysis while she was expecting a baby and was newly postpartum. This life-changing experience most certainly affected the way she thought about the interviews, analysis, and final themes of this study.

Conclusion

We found three main themes from the family-as-group interviews: *Making a Plan*, *Taking Care of Basic Needs*, and *Managing Emotional Burdens During BMT*.

Throughout these themes families described the intense pressures that pediatric BMT places on the family, and they reported diverse methods of family self-management throughout the BMT. It is important for clinicians to both be aware of the challenges of family self-management through the BMT trajectory, and link families to support services. Further research is needed to ascertain whether these themes are consistent over time.

Table 1: Interview Participant Characteristics

Parents' Age (Average)	41.3 years
Patients' Age (Average)	13.5 years
Siblings' Age (Average)	9.0 years
Distance to Transplant Center (Range)	5-460 miles
Time since transplant (Range)	1-12 months
Represented Races	Asian, Black, Caucasian
Represented Faiths	Christianity, Judaism, Islam, Roman Catholicism

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Chapter 5: Discussion

This study used the Individual and Family Self-Management Theory to examine the responses of parents and families to a child undergoing BMT. Below we have summarized the main findings of the study.

Parent Impact

This study introduced the Parent Impact Scale (PARImpact), which was conceptualized in this study as a self-regulation variable, and was designed to measure the direct effects of BMT on the parent. The PARImpact had a single factor solution and displayed convergent and known-group validity. Using pattern mixture models (PMM) we found that PARImpact was associated with BMT complications, such as infection, organ toxicity, and graft vs. host disease. Other studies have shown that BMT complications affect parent outcomes such as emotional functioning (Terrin et al., 2013), and these findings further indicate the vulnerability of parents whose children are experiencing complications.

This study also described parents' reports of missing work or cutting back on other activities during their child's BMT. Two-thirds of parents had to take at least one day off work per week, or cut back on usual activities during the acute phase of the child's BMT, and at one year after BMT, one-third of parents had to take at least one day off per week or cut back on usual activities. Parents of pediatric BMT patients have

reported financial strain during and after BMT, which may be related to employment challenges during a child's BMT.

Parent QOL and General Health

We found that parent QOL and general health were associated with key contextual factors such as marital status, education, and the disease of the patient. At day 45, 3 months, and 6 months post BMT, a child's infection was associated with decreased parent QOL. This finding is congruent with other findings that BMT complications, like infections, are related to the parent impact of transplant and parents' emotional functioning (Heinze et al., 2015; Terrin et al., 2013). Similarly, increasing organ toxicity was also associated with decreased QOL. Longitudinal analyses found QOL is also decreased when the parent is unmarried/not partnered, which is congruent with other findings that a spouse may provide emotional support during a child's BMT (Rini et al., 2004).

Families with one child (the BMT patient) also reported higher QOL than families with three or more children. Our interviews with parents indicated this may be related to the strain of family separation, and feelings of guilt when a parent is unable to provide care for all their children.

Parents' general health was lower among parents with lower educational attainment, which would be expected because education is a known social determinant of health (citation). General health was also lower among parents whose children were undergoing BMT for a malignant disease compared to non-malignant diseases. It is possible that this finding is related the length of the child's illness. Interviews with parents revealed that families of children who were undergoing BMT for immunologic or

hematologic diseases described having more control over the timing of the BMT, and these families did not have to undergo traditional chemotherapy treatment prior to BMT.

Finally, as with QOL, parents' general health was lower among parents whose children experienced organ toxicity. This indicates that when a child has serious BMT complications, not only are they at risk for decreased emotional functioning, but also physical functioning.

Family-as-Group Interviews

The family interviews revealed three themes: making a plan, taking care of basic needs, and managing emotional burdens during BMT. In the interviews families described their processes of addressing the challenges that BMT presented and how they coped with the stress of BMT.

Families' plans for BMT were influenced by the diagnosis of the patient and the timing of the transplant, and the desires of the patients. Ensuring that siblings were taken care of was important to parents, but the ways that families accomplished this varied greatly. Some families chose to stay together in the hospital, while others chose to arrange for the sibling to be cared for by other friends or family members. This is congruent with other studies that have shown siblings experience distress during their ill siblings' BMT (Packman, Weber, Wallace, & Bugescu, 2010).

Some parents were able to make arrangements with their employers to keep working through their child's BMT, but this depended on the individual situation of each parent. There were also a few parents who were business owners and were able to manage their business while participating in the care of their child. However, the majority of families relied on one parent to be the primary caretaker, and the other parent was the

primary breadwinner and insurance carrier. Other studies of pediatric BMT families have shown that these families are vulnerable to financial strain both during BMT and after (Vrijmoet-Wiersma et al., 2010). Single parents were especially vulnerable because it was impossible to split caretaking and breadwinning responsibilities with a partner.

Families universally reported that help from extended family and friends was necessary to meet the families' needs through their child's BMT. While some families had a vast network of helpers, and others had a single support person, it was necessary to have that support. Families also reported that they gained strength from their spiritual beliefs, and some families reported that having hope was important through the BMT.

Limitations & Strengths

The participants for the quantitative and qualitative portions of this study were predominantly Caucasian and had high educational attainment. The quantitative analysis included self-reported variables, which may introduce measurement error. The qualitative interviews used a cross sectional design, which prevented us from exploring concept stability over time. The interviews were also conducted at one pediatric BMT center, which may limit transferability to pediatric BMT families in other locations.

The study had several strengths. First, we used a mixed methods design, and the Individual and Family Self-Management Theory guided variable selection and analysis. The robust sample size for the quantitative analysis provided power for the analyses, and collecting the data at eight different pediatric BMT centers across the United States increases the generalizability of the findings. Even though missing data is unavoidable in longitudinal studies, the study team worked to mitigate loss to follow up. The qualitative study used a family-as-group interview method, which included viewpoints from multiple

family members. Additionally, using Max Van Manen's (1990) human science approach provided a rich context for analysis.

Implications

Taken together these findings illustrate the intense challenges that BMT places on pediatric patients and their family members. As suggested by the Individual and Family Self-Management Theory (Ryan & Sawin, 2009), there were a number of contextual factors that were associated with both self-regulation, and the outcomes of parent general health and QOL. Assessing families undergoing BMT for these factors may help clinicians better care for these families, and allow them to intervene at critical points to improve QOL and general health outcomes.

Some of these contextual factors may be assessed before the transplant begins, such as the parents' marital status, child age, and length of illness. Yet others may occur later on in the transplant trajectory, such as infection and graft vs. host disease. It is important for clinicians to be sensitive to these factors that may indicate vulnerability, and understand that the stress of an adverse event, such as an infection or graft vs. host disease may continue after the treatment has ended.

Furthermore, the finding that even though families experience similar contextual challenges during their child's transplant, such as the need for living for an extended time in the hospital and temporary BMT housing, families chose very different ways to manage these challenges based on their preferences and values. This seems to indicate that there is not one preferred or best method for addressing the contextual challenges of BMT, and clinicians should seek to understand the goals of each family, and support the family as they endeavor to achieve their desired outcomes. Families described how

important it was for hospital staff to understand their family needs and accommodate them as much as possible. Families also agreed that having assistance from friends and family was essential to surviving a child's BMT. Many families also reported that they found strength in their faith beliefs through the BMT trajectory, and ensuring families are supported in these beliefs throughout the transplant trajectory may help to mitigate some of the stresses families face.

The findings in this study that parents need to cut down on work and other usual activities not only in the acute stage of transplant, but also up to a year after transplant highlights the need for policies to ensure ongoing employment for these families (Bona et al., 2015). Many patients reported working with their employer to enable the parent to continue working during their child's BMT. But this option was not universal, and many parents had to quit their jobs to take care of their child.

The science of family self-management during pediatric BMT could be furthered by the development of interventions to assist families through the BMT trajectory. Additionally, longitudinal family interviews would provide more insight into the stability of these concepts over time.

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Appendix A: Sample Questions from Study Instruments and Variable Definition

	Question	Possible Answers
Parent Health	Overall, how would you rate your health?	5 choices ranging from “excellent” to “poor”
Parent Quality of Life	During the past week, how would you rate the general quality of your life in each of the following areas? <ul style="list-style-type: none"> •Physical Health •Mental health •Family life •Friendships •Work life (and/or homemaking) •Responsibilities outside work •Self confidence •Free time or fun •General life enjoyment 	5 choices ranging from “excellent” to “poor”
Parent Impact	During the past week, how many days have you: 1. Missed work or cut down on usual activities because of your health? 2. Missed work or cut down on usual activities because of this child’s health? 3. Arranged for someone to help out because of this child’s health?	Fill-in-the-blank
	During the past week how often did your child’s health: 4. Cause you emotional suffering or worry? 5. Limit the amount of time you have for your own personal needs? During the past week how often did your child’s emotional well-being: 6. Cause you emotional suffering or worry? 7. Limit the amount of time you have for your own personal needs?	5 choices ranging from “a whole lot” to “very little”

Variable Definition and Measurement				
Variable	Theoretical Definition	Operational Definition	Instrument	Measurement Time Points
Parent and Family Characteristics	Characteristics of the parent and family that could influence self-management outcomes	Parent and patient age; parent education; parent marital status; number of siblings	CHRIs demographic form	Pre transplant
Physical and Social Environment	Contextual factors that may influence the patient and family BMT course	Race; ethnicity; income; employment status; site of care;	CHRIs demographic form; medical record	Pre transplant
Complexity of Condition and Treatment	Condition-specific factors that may impact self-management	Type of transplant; duration of illness; diagnosis and stage; BMT complications	Medical record; bearman toxicity scale	Pre transplant, days 45, 90, 180 & 365
Self-Regulation (Parent Impact)	The management of physical and emotional responses to the BMT course	Parent emotional and physical response to child's physical and emotional health; number of days missed from work; number of days asked for help	CHRIs Parent Impact Scale	Pre transplant, days 45, 90, 180 & 365
General Health	Subject assessment of one's general health status	Rating of overall health	CHRIs general health – parent version	Pre transplant, days 45, 90, 180 & 365
Quality of Life	Physical, mental, and social functioning	Physical health; mental health; family life; friendships;	CHRIs global QOL scale – parent version	Pre transplant, days 45, 90, 180 & 365

		work life; responsibilities outside work; self- confidence; free time/fun; general life enjoyment		
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Appendix B Missing Data

	Baseline	45 days	3 months	6 months	12 months
Parent General Health, n (%)	0	47 (13%)	66 (18%)	97 (27%)	128 (35%)
Parent QOL, n (%)	0	47 (13%)	65 (18%)	99 (28%)	131 (36%)

Appendix C Comparison of Quantitative and Qualitative Findings

Quantitative Findings	Qualitative Findings
Changing work patterns and usual activities	Every family had to either change employment or get an accommodation to fit the BMT schedule
Length of Illness	Sometimes having prior experience in the hospital was helpful, but it was challenging to undergo BMT after having previous treatment.
Malignant Diagnoses	Families reported more flexibility among families with children who had non-malignant diseases
BMT Clinical Complications aGVHD Organ toxicity Infection	BMT clinical complications were painful for patients, and emotionally challenging for families
Education/Income	A child's BMT caused two parents to delay school and training programs. Higher income was associated with more options
Insurance	Insurance coverage varied; having a sick child made it more difficult to complete the process to sign up for public insurance
Having a partner	Having a partner provided more resources
Race/Ethnicity/Parent Sex	Families spoke more about the characteristics of their communities and their family interests
Number of Children	Having more children required more planning for the sick child's hospital stay
	Changing relationships

Relying on spirituality to cope

Sample Interview Guide

Protocol Name: Parent and Family Responses to a Child Undergoing BMT during Transitions in Care

Introduction: Thank you for sharing your time with me to talk about your experiences with your child's BMT. We are so grateful that you are willing to share your story, and we hope that we will be able to use this information to help other families experiencing BMT. Please remember that you do not have to answer questions you do not wish to answer and you may stop the interview at any time.

Warm-up:

- Please tell me about your family.
 - What did your family enjoy doing before your child was sick?

Context of Family Self-Management:

- How did your child come to need a bone marrow transplant?
- How did your family prepare for the BMT?
 - Work, school, childcare
- Did your child experience any complications during the BMT (infections, pain, GVHD)?
- What help you to cope during the BMT?
 - If you have a faith belief, did this change how you coped?

Process of Family Self-Management:

- In this interview we are particularly interested in the transition periods from the hospital to the temporary housing, and then the temporary housing to your home.
- What do you remember about your child's time in the hospital?
 - What was challenging to your family?
- Thinking about the transition from the hospital to the temporary housing, what do you remember about that first transition from the hospital to the temporary housing?
 - How did it feel to leave the hospital? What medications/treatments was your child receiving at the temporary house?
- (for families with more than one child) What were some of the ways your family worked out taking care of children in different locations?
- What was helpful to your family during the time at the temporary housing?
 - Was there anyone who helped you during this time?
- (For families who are no longer in temporary housing) What advice would you give to a family leaving the hospital after BMT?
 - What do you wish someone had told you when you were discharged from the hospital?

- What was it like to transition from the temporary housing to your family home?
- What, if any, were the positives of being home again? What if any, were the negatives?
- After being apart for a little while, what was it like to have everyone back together?

Outcomes

- In what ways has your child's BMT impacted your family?
 - How has the BMT changed family relationships (marriage, parent/child)
- Is anyone still taking time off work/cutting back work hours to take care of [child who had BMT]?
- Our research has shown that when children experience complications during BMT (like serious infections and GVHD) that parents continue to be affected after the crisis is over. What is your opinion on this?
- What do you consider to be your family's greatest triumphs during the BMT? Probe: What makes you proud when you look back over your family's BMT experience (so far)?
- How do you foresee your child's BMT impacting your family in the future?

Closing

- If you were in charge of making the transition from the hospital to the temporary housing, and then the temporary housing to home as easy as possible for families like yours, what you do to improve these transitions?
- Thank you so much for sharing your story with me today. Is there anything else you feel doctors and nurses should know about helping families after BMT?
- Please feel free to contact me if you have any questions or concerns.

Curriculum Vitae

Curriculum Vitae Format

Part I

PERSONAL DATA

Katherine E. Heinze

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525 N. Wolfe St. Baltimore, MD 21205
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Home address: 12 Knoll Ridge Ct. Apt 1931
Baltimore, MD 21210
719.351.8377

EDUCATION

Year	Degree Earned	Institution, Location
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2012	PhD Candidate	School of Nursing, Johns Hopkins University Baltimore, MD
2009	Business of Nursing Graduate Certificate	School of Nursing, Johns Hopkins University Baltimore, MD
2004	Bachelor of Science in Nursing	School of Nursing University of Colorado Colorado Springs, CO

LICENSE AND CERTIFICATION

Year	Source, Type	License Number
	Expiration Date	

Current

2004	Present Registered Nurse- 10/31/2017 Maryland	R165486
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Not Current

2011 Certified Pediatric Hematology/Oncology Nurse
12/31/2015

2011 APHON Pediatric Chemotherapy and Biotherapy Provider
12/31/2015

PROFESSIONAL EXPERIENCE

Years	Position Institution, Location	
2011-2015	Supplemental RN-Pediatric Hematology/ Oncology Clinic	University of Maryland Baltimore, MD
2006-2011	Nurse Clinician IIE-Pediatric Oncology Clinic	Johns Hopkins Hospital Baltimore, MD
2008-2010	Per Diem Nurse clinician- Pediatric Emergency Department	Johns Hopkins Hospital Baltimore, MD
2004-2006	Nurse Clinician IM-Pediatric Oncology Inpatient Unit	Johns Hopkins Hospital Baltimore, MD

HONORS AND AWARDS

2015	Recipient of the Nurses Educational Funds Evelyn Barclay Scholarship
2013	Recipient of the Professional Development Award, School of Nursing, Johns Hopkins University
2012	Recipient of NIH-funded T32 Predoctoral Training Fellowship, School of Nursing, Johns Hopkins University
2012	Recipient of the Jonas Nurse Leaders Scholar Award
2011	Recipient of the Caylor Predoctoral Award, School of Nursing, Johns Hopkins University
2008, 2010	Recipient of the BRYCE foundation Excellence in Nursing award

RESEARCH

- 6/2014- Predoctoral National Research Service Grant Award, Katherine Heinze, PI, NINR, NIH, F31 NR014751-01A1
- 5/2014- Special Interest Research Group in Pediatric Palliative Care,
Present Member, Children's
National Medical Center, Washington, DC.
- 7/2013 – Doctoral Degree Scholarship in Cancer Nursing DSCN-13-268-01-
SCN,
6/2016 American Cancer Society
- 1/12 – Trial of Ascertaining Individual preferences for Loved Ones' Role in
8/2016 End-of-life Decisions (The TAILORED Study), Research Nurse,
Principal Investigator, Marie T. Nolan, National Institute of Nursing
Research, NIH, 1R01 NR010733, \$2,489,786, total direct costs.
- 1/2012 - Health Disparities Predoctoral Training Grant, Fellow, Jerilyn
1/2013 Allen PI, NINR, NIH, T32 NR07968

PRACTICE

Practice Initiatives

- 2009-2011 Pediatric Oncology Outpatient Electronic Documentation
Initiative, The Johns Hopkins Hospital, member
- 2007-2011 Patient Education Committee Pediatric Oncology Clinic, The
Johns Hopkins Hospital, Chair
- 2006-2008 The Johns Hopkins Hospital Children's Center Service of
Remembrance planning committee, member and performer

SCHOLARSHIP

Publications

- 2012 **Heinze KE** & Nolan MT. Parental Decision Making for Children with Cancer at
the End of Life: A Meta-Ethnography. *Journal of Pediatric Oncology Nursing*,
29(6), 337-345.

- 2015 **Heinze KE**, Rodday AM, Nolan MT, Bingen K, Kupst MJ, et al. The impact of pediatric blood and marrow transplant on parents: Introduction of the Parent Impact Scale. *Health and Quality of Life Outcomes* 13(46), 1-8.
- 2015 Weaver MS, **Heinze KE**, Bell CJ, Wiener L, Garree AM, et al. [epub ahead of print} Establishing Psychosocial Palliative Care Standards for Children and Adolescents with Cancer and their Families. *Palliative Medicine*.
- 2015 Weaver MS, **Heinze KE**, Kelly KP, Wiener L, Casey RL, et al. Pediatric Palliative Care as a Standard of Care in Pediatric Oncology. *Pediatric Blood and Cancer* 62(suppl 5).
- 2015 Xu J, Lee MC, **Heinze KE**, Kub J, Hughes M, et al. (in press). Symptom Frequency, Severity, and Quality of Life among Persons with Three Disease Trajectories: Cancer, ALS, and CHF. *Applied Nursing Research*.

Book Chapters

- 2013 Heinze, KE, Chapter 51, Pediatric Oncology in: Nettina, S. (Ed.) *Lippincott Manual of Nursing Practice*. Lippincott, Williams & Wilkins, Almbler: Pennsylvania.
- 2013 Heinze, KE, Chapter 52, Pediatric Hematology in: Nettina, S. (Ed.) *Lippincott Manual of Nursing Practice*. Lippincott, Williams & Wilkins, Almbler: Pennsylvania.

Abstracts

- 2015 **Heinze KE**, Budhathoki C, Rodday AM, Nolan MT, Parsons SK. Quality of Life and General Health among Parents of Pediatric BMT Recipients (accepted). Sigma Theta Tau International 43rd Biennial Convention. Las Vegas, Nevada.
- 2015 **Heinze KE**, Rodday AM, Nolan MT, Parsons SK. Parents' Self-Reported Stressors and Associated Coping Strategies During their Child's BMT (accepted). International Family Nursing Association 12th International Family Nursing Conference. Odense, Denmark.

Presentations

- 9/2012 Xu J, Nolan MT, **Heinze KE**, Yenokyan G, Hughes MT, Johnson J, Kub J, Tudor C, Sulmasy, DP, Lehmann L, Terry PB, Rockko F, Lee MC. Symptoms severity and quality of life in persons with advanced cancer, ALS, and CHF. Annual Conference of Council of Advances in Nursing Science, Washington, DC. Poster (Refereed).

- 8/2014 **Heinze KE**, Rodday AM, Nolan MT, Parsons SK. Measuring Parental Distress Across the BMT trajectory: Introducing the Parent Impact Scale. Annual Conference of the Japanese Association for Research in Family Nursing, Okayama, Japan. Podium Presentation.
- 6/2015 **Heinze KE**, Rodday AM, Nolan MT, Parsons SK. Family Related Stressors and Associated Coping Strategies before Pediatric BMT. International Family Nursing Association Biannual Conference, Odense, Denmark. Podium Presentation.

Editorial Activities

Ad-Hoc Reviewer

2013 *Research in Nursing and Health*

2012 *Progress in Transplantation*

PROFESSIONAL ACTIVITIES

Society/Association membership and leadership

2013-present	Eastern Nursing Research Society, member
2013-present	International Family Nursing Association, member
2008-present	Association of Pediatric Hematology/Oncology Nurses
(APHON), member	

Curriculum Vitae Format Part II

EDUCATIONAL ACTIVITIES

Spring, 2016 **Ethics Seminar**, Teaching Assistant, Master's level, 30 students

Fall, 2015 **Ethics Seminar**, Teaching Assistant, Master's level, 30 students.

Spring, 2015 **Ethical, Philosophical, and Theoretical Basis for Nursing** (NUR 110.500), Teaching Assistant, 3 credits, Master's level, 45 students.

Fall, 2014 **Applications of Research to Practice** (NUR 110.503), Teaching Assistant, 3 credits, Master's level online course, 40 students.

Summer, 2012 **Acute Care of Children** (NUR 110.410), Guest Lecturer, "Febrile Neutropenia and Acute Sepsis in the Pediatric Oncology Population" 2 Credits, BSN level, 25 students.

Spring 2012, **Acute Care of Children** (NUR 110.410), Teaching Assistant, 2 credits, BSN level, 20 students.

Fall, 2011, **Nursing for Child Health**, (NUR 110.402), Guest Lecturer, "An Introduction to Pediatric Oncology" 3 credits, BSN level, 45 students.

ACADEMIC SERVICE

2014-2015 Doctoral Student Organization, Johns Hopkins University School of Nursing, Curriculum Committee Student Representative

2013-2014 Doctoral Student Organization, Johns Hopkins University School of Nursing, President

2012-2013 Doctoral Student Organization, Johns Hopkins University School of Nursing, Ethics Chair

2003-2004 Student Government Association, University of Colorado, Colorado Springs, Student Dean of Nursing