2023

Profiles of Family Functioning in Pediatric Hematology-Oncology Patients: Longitudinal Associations with Child Well-Being

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April, 2023

Acknowledgements

I am extremely grateful to everyone who has supported me through this project. Thank you to my committee co-chairs, Dr. Winter and Dr. Rohan, for your generous mentorship and patient direction at every step of this paper. Thank you also to Dr. Corona for providing me with valuable insight to improve the quality of our research. Thank you to Dr. Rohan's research team for their many hours spent collecting this study's data and thank you to all the families who gave their time to participate in this study. Thank you to my professors at Emory and VCU for showing me how exciting psychological research is. Thank you to my family for being our most constant and loving supporters. Thank you to my friends for working long hours along with me and for bringing so much joy along the way.

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Abstract

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By Sydney Sumrall, B.A.

A thesis submitted in partial fulfillment of the requirements for the degree of Master of Science at Virginia Commonwealth University.

Virginia Commonwealth University, 2023

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This study aims to assess 1) patterns of family functioning and 2) longitudinal associations between family functioning patterns and well-being in a vulnerable cohort of children with chronic illness. Caregivers of hematology (16.4%) and oncology (83.6%) patients ages 7-20 (N=55; M_{age} = 13.3 [SD = 2.7]; 52.7% female; 45.5% non-Latinx White, 38.2% Black or African American, 12.7% Latinx, 1.8% Asian, and 1.8% multi-racial,) reported on family functioning via the Family Assessment Device. Cluster analyses identified three mutually exclusive clusters: one high adaptive group, one moderate adaptive group, and one maladaptive group. Group membership was not significantly associated with child psychological distress or quality of life. Results provide insight into family processes for children with a blood disorder or cancer and offer direction for future work that aims to identify predictors of resilience in this population.

Introduction

Approximately 20% of children in the United States have a chronic illness that interferes with their daily functioning (NIH, 2016). Although many children with chronic illness are able to cope well with their diagnosis, a subset of children consistently report physical and mental health concerns (Blackwell et al., 2019; Noll & Kupst, 2007). It is important to understand barriers to their overall well-being to develop effective interventions. Pediatric oncology and hematology patients undergo a variety of stressful medical treatments and procedures and endure physical symptoms that may persist across the developmental lifespan, including experiencing late effects for cancer treatment long into survivorship. These conditions, procedures, and symptoms negatively affect children's overall well-being including quality of life and mental health (Stokoe et al., 2022; Kunin-Batson et al., 2016; Zheng et al., 2018). When families work well together and have positive relationships with one another, they can offer an important source of resilience to the psychological impact of children's illness (Van Schoors et al., 2015). Identifying aspects of family life that enable children to thrive may help researchers and clinicians guide families in effectively providing support to their child during periods of intense medical stress. The primary goal of this study is to identify profiles of family functioning in a sample of children, adolescents, and young adults with oncological and hematological disorders. A secondary aim is to determine whether baseline patterns of family functioning predict subsequent youth well-being (i.e. quality of life, psychological distress).

Literature Review

Prevalence, Survival Rates, and Treatment for Cancer and Blood Disorders

Cancer is a disease which causes cells to multiply at a rapid rate and spread to other areas of the body (NIH, 2021). Approximately 10,470 children in the United States will be diagnosed with cancer in 2022. The five-year survival rate for children with cancer has increased to around

85%, up from 58% in the 1970's; cancer remains the second leading cause of death for children ages one to fourteen (American Cancer Society, 2022). The most common types of cancer include acute lymphocytic leukemia (ALL) and acute oureloid leukemia (AML). Brain and spinal cord tumors are the second most prevalent types of pediatric cancer. Other types of pediatric cancer include neuroblastoma, Wilms tumor, lymphoma, rhabdoourosarcoma, retinoblastoma, and bone cancers such as osteosarcoma and Ewing sarcoma. Pediatric cancer causes a variety of adverse physical symptoms. For example, ALL and AML may cause bone and joint pain, fatigue, weakness, bleeding, fever, and weight loss. Brain and spinal cord tumors can cause headaches, dizziness, seizures, and trouble walking (American Cancer Society, 2022).

Treatment for most pediatric cancers could involve surgery, radiation therapy, chemotherapy, or a combination of these treatments. Surgery may be used to remove tumors or to diagnose cancer by removing a piece of tissue and testing it, known as a biopsy (American Cancer Society, 2022). Radiation therapy works by using X-ray beams to damage the DNA found in cancer cells to kill them or prevent their growth (NIH, 2019). Chemotherapy refers to the administration of drugs to kill cancer cells or alleviate symptoms associated with cancer (American Cancer Society, 2022). In some cases, children with cancer receive stem cell treatment following chemotherapy or radiation therapy. Stem cells are cells in the early stages of becoming blood cells, i.e., hematopoietic cells. Stem cells are found in the bone marrow (center of bones) where they divide to make new blood cells. Once stem cells become mature blood cells, they then leave the bone marrow and enter the bloodstream. Stem cell transplant works by increasing the number of stem cells in the patient's bone marrow. Stem cells then become blood cells which can destroy existing cancer cells or cells damaged due to radiation or chemotherapy (American Cancer Society, 2022). Following stem cell transplantation, patients may develop

infections or experience stomach, heart, lung, or kidney problems. Patients must return to the hospital daily for several weeks as healthcare providers monitor their recovery.

Following cancer treatment, children need regular follow-up exams, often referred to as surveillance monitoring. Survivors are at an increased risk for cancer recurrence as well as developing other kinds of secondary cancers (American Cancer Society, 2022). Recurrence only occurs in about 6% of survivors, and over time the likelihood of cancer recurrence decreases, and doctor's visits become less frequent. Late effects from cancer diagnosis and treatment are possible across their lifespan (Tutelman et al., 2022). These late effects include psychological distress, heart and lung problems, delayed growth, changes in sexual development, cognitive changes (slower processing speed, executive functioning difficulties), and learning problems.

Hematological disorders, or blood disorders, are defined by problems of blood cells and blood-forming organs such as bone marrow, lymph nodes, and spleen. These include sickle cell disease, anemia, hemophilia A and B, and other rare diseases. These diseases are genetically inherited and diagnosed at a young age. Prevalence varies by disorder. For example, there are about 30,000 cases of Hemophilia A and 100,000 cases of sickle cell anemia, also known as sickle cell disorder (SCD), in the United States. Hematological disorders may also occur as a result of other illnesses such as leukemia. The exact number of people in the United States with any blood disorder is not known (CDC, 2020). Symptoms of anemia include tiredness, weakness, painful swelling of the hands and feet, jaundice, and poor appetite. Complications for patients with anemia may result in pain, slow or delayed growth, cancer, and congenital defects. Hemophilia results in symptoms such as bleeding within joints, bruising, and nose bleeds. While treatment for hematological disorders varies across diagnoses, all blood disorders will require life-long disease monitoring and daily disease self-management, including lifestyle changes or taking prescribed treatments (CDC, 2020). Although many children born with hematological

disorders survive into adulthood, life expectancy may be shortened. For children born with SCD, life expectancy is estimated to be 20 to 30 years shorter than for typically developing adults (Sedrak & Kondamudi, 2022).

As with some cancers, treatment for hematological disorders may include a stem cell transplant. For example, patients with Blackfan Anemia, a blood disorder that causes bone marrow to produce too few red blood cells, may receive stem cell therapy to increase the number of red blood cells in the patient's bone marrow (Miano et al., 2021). Similarly, stem cell transplant for SCD, a disease defined by abnormally shaped red blood cells, replaces these abnormal cells with new, healthy ones (CDC, 2020). While prognosis for blood disorders varies by disease and severity, most patients do not ever achieve disease remission following treatment, and hospital readmissions are common. These disorders tend to be life-long diseases that are accompanied with disability and require long-term treatment adherence (Badawy et al., 2017). Children with these conditions and their families must therefore acquire knowledge about their condition and adhere to treatment into adulthood. These children also may also experience chronic pain (Martin et al., 2018) and have increased risk for intellectual disability, cognitive impairment, and hearing impairment (Boulet et al., 2010).

Although there are key differences in age of onset, symptoms, and prognosis for children with oncological and hematological disorders, prior research shows that both types of disorders impact children's psychological functioning in similar ways. Children with these disorders have lower school attendance, greater internalizing difficulties, and poorer academic performance than children without a chronic illness (Trzepacz et al., 2004; Patterson et al., 2012). Additionally, both disorders often necessitate inpatient hospital admissions and invasive procedures such as stem cell therapy. Hospitalizations disrupt these children's everyday activities and increase rates of psychological distress, behavior difficulties, and medical trauma (Wilson et al., 2010; Stark &

Tye, 1994; American Cancer Society, 2022). Challenges faced during medical treatment for chronic illness may further impact family dynamics, thus the importance of investigating not only individual factors but also family-level factors like family functioning.

Family Functioning in Children with Chronic Illness

As treatment for pediatric illness evolves and the number of children living with a chronic illness increases (Wright et al., 2022), research addressing quality of life for this population becomes essential in helping children and their families cope with the challenges associated with the illness. The family is one target identified in the literature to improve children's resilience in the face of a chronic illness. Family systems theory provides a framework for viewing the family as a whole. Individual family members are part of multiple subsystems within the family such as parent-child and sibling relationships. The interactions between these subsystems result in family-level processes (Fiese et al., 2019). Family systems theory posits that interconnections between family members' influence individual behavior, including health and well-being. Interactions between family members can alter or reinforce existing family dynamics and behaviors, which may be adaptive or maladaptive (Kerr & Bowen, 1988).

Family functioning is one measure of family systems that is commonly used in the pediatric illness literature. The concept has been used to measure the ways that family members relate to one another through rituals and routines as well as family-level patterns such as how members work together to resolve stressors, communicate with one another, manage conflict, and divide household labor (David, 1978; Fiese et al., 2002). However, there is currently no standard operational definition of family functioning, and the aspects of the family system used to measure family functioning vary by study. In this study, family functioning is defined by the ways in which families are structured and organized and the interactions between family members, as measured by the Family Assessment Device (FAD; Herzer et al., 2010). The FAD

assesses six dimensions of functioning including problem solving (resolving problems effectively), communication (exchanging clear and direct verbal communication), roles (sharing responsibility of family tasks), affective responsiveness (giving the proper emotional response), affective involvement (being involved and interested in one another), and behavior control (expressing and maintaining standards of behavior). Good family functioning may therefore mean that family members are able to effectively make decisions together, communicate about their feelings, divide household labor fairly, and display interest and care in each other's lives. In cases where family functioning is poor, stress can exacerbate emotional reactivity and hostile interactions, and families may be unable to work together to solve problems (Alderfer et al., 2008). A family systems perspective emphasizes the reciprocal relationship between family functioning and individual behavior. Having a child with a chronic illness could positively or negatively influence family dynamics, including aspects of family functioning.

A variety of changes occur following a child's diagnosis that may impact family functioning. Illness may lead to changes in bedtime, mealtime, and school routines as well as new rules and routines regarding treatment and medication for children with cancer and blood diseases. Parents may also experience lower energy and time constraints due to additional strain of caregiving demands (Bates et al., 2021). Factors such as greater illness severity and lower maternal education are associated with worse family functioning (Al Ghriwati et al., 2020; Huang et al., 2018). Despite the challenges families of children with a chronic illness face, some families report improvements in family functioning following a child's diagnosis (Beek et al., 2015; Herzer et al., 2010). These results indicate that the family could be an important point of resilience for children with these conditions. A family systems framework can therefore guide the identification of predictors and mediators of psychological well-being that allow researchers

to better understand what aspects of the child's environment might be protective in the context of having a chronic illness.

For families of children with cancer and blood diseases, there are unique challenges that may interfere with adaptive family functioning. These families must communicate about the diagnosis and treatment, adjust roles to meet the needs of the child with cancer or blood disease, and make lifestyle changes to support treatment objectives (Long & Marsland, 2011). Parents experience a number of changes that may influence family functioning. Compared to a control group of parents with healthy children, parents of children with cancer experienced lower persistence, determination, and tolerance for failure (Dabrowska & Malicka, 2022). Parents report increases in emotional distress following a child's cancer diagnosis, with a significant proportion of parents experiencing anxiety or post-traumatic stress following the diagnosis (Erickson et al., 2022; Alderfer et al., 2009). However, this distress tends to subside over their child's disease course, and some parents report post-traumatic growth, an increase in meaning and optimism, over time (Czyzowska et al., 2021).

Quantitative research with families of children diagnosed with cancer has suggested adaptive, relatively high levels of family functioning. On the other hand, in qualitative studies family members have reported negative feelings and that cancer disrupted family life and brought distressing changes to family relationships (Van Schoors et al., 2015). Evidence for changes in family functioning in families of a child with cancer varies by dimension of family functioning, and these variations may offer insight into how family functioning is impacted by pediatric cancer. Van Schoors et al. (2015) conducted a systematic review of family functioning following a child's cancer diagnosis. Most studies suggested that higher resilience was related to higher levels of emotional closeness between family members. Furthermore, results suggested that parent-child bonds may become even closer following diagnosis of a chronic illness.

Previous studies also demonstrated that family discord and opposition may increase during treatment but often return to previous levels following treatment completion (Van Schoors et al., 2015). Additionally, marital problems following diagnosis may spillover into the parent-child relationship, creating discord at the family level (Katz et al., 2018).

A comprehensive model of the family environment for child cancer survivors demonstrates that even after treatment is completed, families of cancer survivors often experience heightened levels of stress, especially as it relates to family management and communication of late effects (Peterson & Drotar, 2006). Individuals' roles within the family unit may need to shift in response to the challenges of cancer, and previous research suggests that families are largely able to make these changes (Van Schoors et al., 2015). Open and effective communication between family members is another critical component of family functioning. Prior literature illustrates deficits in effective communication for families of a child with cancer, even though these families may not have worse communication than families of healthy children (Van Schoors et al., 2015). Though communication patterns may not decrease following a child's cancer diagnosis, these results suggest families may need to to express themselves differently and share more information in order to function well. Family functioning is also better when family members are able to respond to each other's affect appropriately and provide emotional support and encouragement when needed. In fact, some children with cancer reported feeling cared for and better understood by their parents following a stem cell transplant (Barrera et al., 2007), and overall family levels of support tended to be higher for these families (Van Schoors et al., 2015).

There is a smaller body of literature on family functioning patterns in pediatric hematology disorders. Given the similarities in challenges and rates of parent's emotional distress across pediatric chronic illness, our understanding of family systems for children with

blood disorders is largely informed by pediatric cancer and other chronic illnesses. However, existing literature on hematological disorders suggests unique risk factors. These disorders are life-long incurable chronic illnesses that present at an early age, so parents' rates of emotional distress are often high (Furmedge et al., 2014; Bioku, 2021), especially given the heritability of these disorders, which may cause parents increased feelings of guilt (Wiedebusch et al., 2013). Sickle cell disease may be particularly difficult for families as it disproportionately affects African Americans who also tend to face a multitude of environmental stressors along with their child's illness (e.g., financial hardship and racial discrimination; Bills et al., 2020). Socioeconomic disadvantage for families of children with SCD is associated with worse parenting behaviors and higher rates of maternal depressive symptoms, which may negatively impact family functioning (Robinson et al., 2015). Additionally, families of children with SCD often report lower adherence rates and worse disease self-management, with more than 50% not adhering to prescribed medication. This finding may be attributed to aspects of family functioning including conflict management, working together to accomplish tasks, communication, and roles (Psihogios et al., 2018).

Some research in the hematology literature evaluated family functioning directly. A study conducted in a sample of children with hemophilia found these children experienced significantly worse family functioning compared to their healthy peers; although, sample size was limited (Evans & Shiach, 2000). On the other hand, other researchers found that families were able to work together to adapt to their child's hemophilia diagnosis and report optimal levels of emotional closeness (Torres-Ortuno et al., 2014). More research is needed to understand how hematological disorders impact family functioning and how these patterns may differ from patients and families managing oncology disorders.

Family Functioning and Child Well-Being

Family functioning impacts quality of life for children with chronic illness. Quality of life (QoL) encompasses several dimensions of health including physical, psychological, and social well-being. Research consistently identifies lower rates of QoL in children with chronic illness, including children with oncological and hematological disorders (Fardell et al., 2017; Hala et al., 2013; Stokoe et al., 2022). A large study (N = 594) of children with cancer assessed different subscales of quality of life in children. Lower family functioning scores were associated with some but not all aspects of quality of life for children 26 months after diagnosis (Zheng et al., 2018). Among children recently diagnosed with leukemia, lymphoma, or a solid tumor, family conflict was associated with poorer child QOL six months later (Desjardins et al., 2022). Another study found that family functioning mediated the association between children's neurocognitive difficulties and QoL six months following completion of cancer treatment (Al Ghriwati et al., 2020). These results demonstrate that the severity of symptoms the child experiences can worsen family functioning which in turn, puts children at greater risk for lower quality of life. Quast et al. (2018) found that child-reported family functioning following treatment significantly predicted parent and child-reported QoL from baseline (five months after completing therapy) to nine months later in a sample of pediatric brain tumor survivors.

Less is known about how family functioning impacts QoL for children with blood disorders. One relevant study by Psihogios et al. (2018) found that among children with SCD, greater family efficacy predicted better disease self-management which in turn led to better QoL. Some studies did not examine family-wide processes, but they examined components of the family system (e.g., parent functioning or parenting) that contribute to family functioning (Delvecchio et al., 2016). For example, parental stress has been associated with poorer QoL (Moody, 2021). Further, Barakat et al. (2005) found that parental locus of control (i.e., perceived

control in the parent-child relationship) was negatively correlated with child QoL. Other work that did not assess QoL directly speaks to outcomes that are known to relate to QoL. For example, Barakat and colleagues (2007) assessed how family functioning was associated with objective measures of health including disease severity, healthcare utilization, hemoglobin levels, and SCD complications, which may be associated with QoL (Megari, 2013). Their results showed that children with SCD who had poorer family functioning reported greater disease severity and greater healthcare utilization. Together, the current literature on pediatric chronic illness supports the link between family functioning and pediatric QoL.

Psychological distress is another important aspect of well-being for children with chronic illness defined by mental suffering that may be indicative of a variety of diagnoses (Thelin et al., 2017). There is a large body of literature indicating children with chronic illness are at risk for psychological distress (Katz et al., 2018), and healthy family functioning may act as a protective factor (Winter et al., 2019). Among children with cancer, poorer family functioning was associated with depressive symptoms but not anxiety symptoms following completion of treatment (Kunin-Batson et al., 2016). A meta-analysis on the association between child and parent psychological distress in pediatric cancer found that child distress was associated with overall parent distress as well as parent distress subtypes including depression, anxiety, and post-traumatic symptoms (Bakula et al., 2019). These results indicate the importance of examining family factors in children's psychological distress. Other research has evaluated psychological distress in the context of stressful aspects of treatment. In a sample of children with cancer, family cohesion and family expressiveness were significantly associated with increased child distress before undergoing stem cell transplantation (Jobe-Shields et al., 2009). One study following 159 children after a cancer diagnosis found that family functioning predicted greater risk for anxious and depressive symptoms immediately following diagnosis (Ourers et al., 2014).

Family functioning is also important after children complete treatment. A systematic review on family support following treatment completion for adolescents and young adults found that better family functioning was associated with lower patient emotional and psychological distress. Other research suggests family functioning may affect long-term stress reactivity. Erickson et al. (2022) found that poorer family functioning was correlated to cortisol concentration, a biomarker of long-term stress, in pediatric cancer survivors but not healthy controls.

There is also evidence that worse family functioning puts pediatric hematology patients at risk for greater psychological distress. Most of this evidence comes from the literature on children with SCD. Children with SCD who had poorer family functioning reported more anxious and depressive symptoms (Burlew et al., 2000). Family functioning as defined by conflict predicted lower scores on the Child Behavior Checklist, a measure of both internalizing and externalizing behavior problems, including anxious, depressive, somatic, and conduct problems (Thompson et al., 1999). Another study found that family functioning mediated the association between the number of emergency room visits reported by children with SCD and their psychosocial adjustment. Higher levels of family support and lower levels of family conflict were linked to better psychological outcomes (Gold et al., 2011). Overall, the existing literature suggests that children with chronic illness who experience challenges to family functioning may be at greater risk for psychological distress.

Patterns of family functioning

Identifying patterns of family functioning can provide a more comprehensive understanding of how family members relate to one another. Whereas a variable approach to measuring family functioning assumes that different dimensions of the construct relate to each other in the same ways and can be evaluated linearly for every family, a typological approach describes these patterns of functioning across dimensions of family functioning (McQuitty,

1987). This approach allows researchers to describe types of families and compare the various typologies that emerge (Mandara & Murray, 2002). The FAD is a well-validated and reliable measure that assesses six domains of family functioning. By evaluating what combinations of these dimensions families are similarly high or low on, we can define patterns in family relationships that simultaneously demonstrate the strengths and weaknesses of families of children with chronic health conditions.

A small body of research has investigated patterns of family functioning in pediatric hematology-oncology patient cohorts and how these patterns relate to overall child well-being. Ozono et al. (2010) identified clusters based on measures of family expressiveness, cohesion, and conflict in child cancer survivors and their parents. They found that "conflict-type" families, as in those with greater conflict but also greater closeness and openness in communication, had higher levels of post-traumatic stress symptoms, anxiety, and depression. Likewise, a study of parents and their children with asthma assessed patterns of family functioning and identified four profiles. The majority (60%) of families that fell in the "cohesive" profile demonstrated better family functioning across domains of the Family Environment Scale including closeness, openness in communication, conflict, organization defined by clear roles and structure in family responsibility, and control defined by rigidity of family rules and procedures. On the other hand, "permissive", "controlling/disengaged", and "controlling/enmeshed" families demonstrated mixed scores of family functioning across these domains. Permissive families were defined by high levels of closeness and open communication but low levels of control (i.e. rigidity and rules in the family) and less clear delineation of family member roles. Controlling/ disengaged families had low levels of closeness and open communication and high levels of control and conflict. Finally, controlling/enmeshed families had moderate levels of closeness and open communication and high levels of conflict and control. Further analyses revealed that children in

families defined as cohesive were less likely to present with externalizing and internalizing symptoms (Al Ghriwati et al., 2017). Another profile analysis from Al Ghriwati and colleagues (2021) identified four profiles of family relationships. To measure family relationships, children with cancer reported on discord and closeness with each sibling and caregiver. The majority of families fell in the "high closeness/ low discord" (47.6%) or "moderate closeness/ moderate discord" (33.4%) profile. A smaller portion of families experienced more problematic functioning defined by "low closeness/ high sibling-only discord" (12.4%) or "low closeness/ high discord" (6.6%). Children in the "low closeness/ high discord" profile experienced greater externalizing symptoms but not internalizing symptoms or quality of life. No study to date has assessed profiles of family functioning using the FAD. Given the ubiquity of the FAD in the literature, understanding how multiple dimensions of the measure relate to one another in a sample of children with chronic illness can help researchers better identify profiles of families that may be at more or less risk for poor child adjustment.

The Present Study

This study seeks to extend the existing literature by identifying typologies of family functioning, using the FAD, among children with chronic health conditions, including oncology and hematology patients. Few studies to date have assessed profiles of family functioning in pediatric chronic illness populations. This study represents a unique opportunity to assess patterns of family functioning in a vulnerable cohort of pediatric patients, especially as it relates to identifying at-risk patients who are early on in treatment, so that family-centered interventions can be implemented. Additionally, there is mixed evidence on the link between family functioning and child well-being for children with chronic illnesses, and there are limited studies conducting this research in hematology populations. We will build upon the existing literature by

examining how longitudinal child adjustment (i.e., child psychological distress and quality of life) differs based on family functioning typology.

The specific aims of the present study are to: (1) Identify typologies of family functioning in a sample of pediatric oncology/hematology patients. Cluster analysis will be conducted to describe the dimensions of family functioning that typically co-occur and how these typologies reflect unique patterns in the ways family members relate to one another. (2) Determine whether baseline patterns of family functioning predict subsequent QoL at 6 -month follow-up in pediatric oncology/hematology patients. (3) Determine whether baseline patterns of family functioning predict subsequent psychological distress at 6 -month follow-up in pediatric oncology/hematology patients.

Although no studies to date have examined family typologies using the Family Assessment Device, previous research on family systems gives insight into what patterns may emerge. Based on previous research, we predict that analyses will show at least three different profiles. Studies that have taken a family systems theory approach consistently identify a profile defined by relatively high scores across domains of functioning, and research shows that most families fit this pattern (Al Ghriwati et al., 2017). Thus, we predict a majority of families in this sample will fit a typology of high family functioning across subscales. In addition, past research has typically found a category comprising a small number of families with relatively poor functioning across domains. Thus, we predict that a profile defined by low affective responsiveness, affective involvement and poor problem solving, behavior control, roles, and communication will also emerge. Finally, past studies typically have identified one or more mixed profiles in which families show some strengths and some weaknesses; we tentatively predict that at least one mixed profile will emerge in this dataset. Furthermore, we predict that

children from families in clusters with relatively healthy family functioning will have higher QoL and better psychological functioning six months later.

Methods

Participants

The proposed study was a secondary analysis of data from the *Promoting Avatars in Clinical Experience (PACE) Study*. The PACE Study is an observational investigation of the patient's experience with having a chronic illness including the psychological impact of chronic illness on the patient and family. In PACE, participants were prompted to share narrative stories and create virtual avatars that described their chronic illness journey. The PACE study recruited pediatric hematology-oncology children, adolescents, and young adults either during a clinic visit or during an inpatient admission at a large hospital in the southeast United States. Inclusion criteria included: 1) diagnosed with cancer or a blood disease requiring medical visits at least every 3-months, 2) between the ages of 7 and 20 years old, 3) speak English, 4) have at least one caregiver involved, and 5) followed by the pediatric hematology/oncology/stem cell transplant service in a pediatric academic medical center in the southeast United States.

The PACE Study is an observational, single arm study that was designed to inform development of eHealth interventions targeting improved psychosocial and health outcomes in children, adolescents, and young adults diagnosed with a chronic illness. The study aimed to recruit a total of 75 participants to take part in this study. Sixty-eight patients were identified as eligible and completed the recruitment process, including signing informed consent and parental permission. The final sample for the secondary data analysis described here consists of participants who remained eligible at time of the baseline visit and had parent baseline data available (N=55). At 6-month follow-up, 12 participants did not have 6-month data available; thus, the sample size for the 6-month parent cohort analyzed in Aim 2 was 43 parents. A post-

hoc power analysis indicated that we have at least 90% power to detect an effect of longitudinal change over time.

The majority of patients (83,6%; *n*=46) had a history of cancer (active treatment, survivorship); whereas 16.4% (*n*=9) were diagnosed with a blood disorder. Pediatric patients were 52.7% female and 47.3% male with an average age of 13.3 years (*SD* = 2.7). Children were 45.5% non-Latinx White, 38.2% Black/African American, 12.7% Latinx, 1.8% Asian, and 1.8% multi-racial. On average, children were 54 months (4.5 years) out from their diagnosis date at study baseline, but this time point varied by hematology versus oncology diagnosis given most hematology diagnoses are diagnosed at birth or shortly thereafter. In fact, hematology patients had a disease duration of 165 months (13.8 years), while oncology patients' disease duration was 35.6 months (3.0 years). Caregivers were primarily adoptive/biological mothers (86.0%) or fathers (7.0%). Other caregivers were step-mothers (1.8%) or grandmothers (5.3%). Caregivers' average age was 44.7 years (*SD* = 8.0). Caregivers were 92.7% female and 7.3% male. It is notable that while some of the patients enrolled and completed data collection prior to March 2020, there was a cohort of patients from whom data was collected after March 2020 during the height of the COVID-19 pandemic.

Procedures

Participants and their parents provided informed consent, assent, and parental permission prior to data collection. Participants were followed over 6 months with data collection occurring at baseline, 3- and 6-months after baseline. At baseline, patients and parents provided demographic information. At each timepoint, patients and parents completed survey data about their well-being and family life and received compensation ($15 cash) at completion of each study visit. The parent study was approved by an Institutional Review Board at VCU as well as the Massey Cancer Center Protocol Review and Monitoring Committee. This study uses parent-

reported demographic and family functioning data from baseline and parent-reported child/adolescent psychological distress and quality of life data from the six-month follow up.

Measures

Psychological Distress

The DSM-5 Level 1 Cross-Cutting Symptom Measure was used to measure patient psychological distress using parent-proxy reports (American Psychiatric Association, 2013). The measure assesses mental health across multiple domains. Subscales include depression, anger, mania, anxiety, somatic symptoms, psychosis, sleep problems, memory, repetitive thoughts and behaviors, dissociation, and personality characteristics. There are 19 items in the parent-proxy report version (ages 6-18+). The items ask how often the person has been bothered by a specific symptom during the past two weeks on a scale of 0 (*none*) to 4 (*severe*). A total score was calculated by summing all items. Higher scores indicate greater overall psychological distress ($\alpha = .90$).

Quality of life

The Pediatric Quality of Life Inventory: General Core was used to assess quality of life in children and adolescents via parent-proxy report (Varni et al., 2007). In the current study, young child (ages 5 to 7), child (ages 8 to 12), adolescent (ages 13 to 18), and young adult (ages 19 to 25) versions were used. Each version contains 23 items reporting on patient functioning in the past month. Parents rated items on a 5-point Likert scale from 0 (*never*) to 4 (*almost always*). Items were reverse-scored and linearly transformed to a 0–100 scale across all age groups. A total score was calculated by totaling scores across all items ($\alpha = .93$). Higher scores indicate better QoL.

Family Functioning

Family functioning was assessed using the Family Assessment Device: Parent report (FAD; Herzer et al., 2010). The FAD measures structural and organizational properties of families and the patterns of transactions between family members. The FAD is made up of 60 items comprising six domain subscales: problem solving ("we confront problems involving feelings"), communication ("we are frank with each other"), roles (Family tasks don't get spread around enough), affective responsiveness ("we cry openly"), affective involvement ("we are too self-centered"), and behavior control ("there are rules about dangerous situations"). The FAD uses a 4-point Likert scale from 0 (*strongly disagree*) to 4 (*strongly agree*) with higher scores indicating more problematic family functioning. It has been shown to be a reliable and valid measure (Hamilton & Carr, 2015) including high test-retest reliability and internal consistency across different family types (Epstein et al, 1983). Dimension scores were calculated by averaging all items within each dimension. Internal consistency for each dimension was tested using Cronbach's alpha: problem solving ($\alpha = .63$), communication ($\alpha = .62$), roles ($\alpha = .73$), affective responsiveness ($\alpha = .68$), affective involvement ($\alpha = .58$), and behavior control ($\alpha = .59$).

Demographic and Medical Data

Parents completed a demographics form at baseline, which included information related to patient- and family-level factors (e.g., patient age, ethnicity, race, gender; primary and secondary caregivers; family income; etc.). Medical charts were reviewed at baseline, 1 month, 3 months, and 6 months using standardized data collection forms. Data was collected on disease type, treatment protocol (if applicable), diagnosis date, prescribed treatments (medication name, dosing), height, weight, BMI, and health care utilization (e.g., clinic visits, urgent care and emergency room visits, and hospitalizations).

Data Analysis

Quality Control of Data Used in the Proposed Study

All study data was cleaned by research coordinators under the supervision of A. Jewell and J. Rohan. All questionnaire data was reviewed for quality and data integrity and issues were addressed as needed. Data was stored in a secured database and double-checked for accuracy.

Data Analytic Plan

Patient and parent baseline descriptive statistics (i.e., means, standard deviations, and ranges) were calculated. These descriptive statistics included family functioning, QoL, psychological distress, patient diagnosis, patient age, patient gender, patient race/ethnicity, parent relationship to child, parent age, and parent gender. All variables of interest were examined for normality and homogeneity of variance. Bivariate correlations were conducted to assess multicollinearity. Data was transformed as needed for assumption violations. Baseline demographic and medical characteristics - e.g., patient gender, age, ethnicity/race - were examined for collinearity with outcome measures to determine which variables should be included as covariates. General linear models were used for continuous variables, which are appropriate for normal and non-normal distributions.

Family Functioning Profiles at Baseline: Two-Step Hierarchical Cluster Analysis: Aim 1

Cluster analysis is a "person-oriented" analysis, meaning it finds similar patterns in participant data to group individuals together who score similarly on different subscales of a measure. Standardized z scores were used as the unit of analysis because cluster analysis requires commensurability (i.e., equal scale units) (Aldenderfer & Blashfield, 1984). Cluster analysis generates a series of solutions, and each solution has one more class (i.e., cluster) than the previous. Researchers then use statistical and theoretical criteria to decide which solution is the

best fit for the data (Weller et al., 2020). Cluster analysis can be used in cohorts with heterogeneous medical diagnoses to identify homogeneous subgroups on a specific variable of interest (e.g., family functioning).

Hierarchical two-step cluster analysis was conducted to identify family functioning profiles in hematology-oncology youth based on the seven subscales of the parent-reported FAD collected at baseline (N=55). In the first step of the analysis, "pre-clusters" were identified by determining if each observation should be merged with previously formed clusters or if a new cluster should be created. Variable means were used to determine whether an observation is similar enough to other observations to be in the same cluster (Weller et al., 2020). The second step of the cluster analysis then uses these "pre-clusters" as single cases that are used to create the desired number of clusters. The ideal number of clusters was based on recommendations from Aldenderfer and Blashfield (1984). Predictor importance and a dendrogram also were examined to determine the strongest predictor of cluster membership and the sample sizes of clusters that emerged.

Prediction of Baseline Patterns of Family Functioning to Subsequent Clinical Outcomes at 6-months: Aim 2.

Longitudinal mixed effects models were used to determine whether baseline patterns of family functioning predicted subsequent clinical outcomes at 6-month follow-up in a cohort of pediatric oncology/hematology patients (N=43). *Aim 2a* examined whether baseline patterns of family functioning predicted parent-proxy reported patient quality of life (QoL) 6-months later. *Aim 2b* determined whether baseline patterns of family functioning predicted subsequent patient psychological distress (parent-proxy report) at 6-month follow-up. General linear mixed models were used for normal/continuous outcomes. Working correlation structures were examined for all models and the "best" model was chosen using the appropriate model fit statistics (R^2) , which

are dependent on the model type used for the analysis (Cui, 2007; Vaida & Blanchard, 2005). This kind of model works well for analyses with small sample sizes (Bell et al., 2010). The model uses a maximum likelihood approach which can accommodate missing data. All analyses were conducted with SPSS, R, and SAS 9.4.

Results

Descriptive Statistics

Descriptive statistics for participant characteristics are presented in *Table 1*, including means and standard deviations for continuous variables and *n* and percentage for categorical variables. Participants included pediatric patients with cancer or blood diseases ages 7 to 19 years and their parents (N = 55). Missing data was handled via listwise deletion and therefore sample size varied for different analyses. The majority of patients (83.6%) were diagnosed with cancer or a cancer predisposition disorder (neurofibromatosis), while 16.4% were diagnosed with a benign blood disorder (e.g., sickle cell disease, hemophilia).

Table 1

Descriptives for Patient and Caregiver Baseline Demographic and Medical Characteristics (N=55)

N (%) or M (SD)		*N* (%) or M (SD)	
Patient Diagnosis		**Caregiver Marital Status**	
Blood disorder	9 (16.4%)	Never married	4 (7.3%)
Cancer	46 (83.6%)	Married	38 (69.1%)
Child Gender		Separated	2 (3.6%)
Male	26 (47.3%)	Divorced	10 (18.2%)
Female	29 (52.7%)	Unknown or not reported	1 (1.8%)
Child Age (years)	13.3 (2.7)	**Household Income**	
Child Race		Less than $19,000	8 (14.5%)
Asian	1 (1.8%)	$19,000 to $34,999	4 (7.3%)
Black or African American	22 (40.0%)	$35,000 to $49,999	7 (12.7%)
White or Caucasian	30 (54.5%)	$50,000 to $72,999	9 (16.4%)
More than one race	2 (3.6%)	$73,000 to $126,500	15 (27.3%)
Child Ethnicity (Latinx)		More than $126,500	10 (18.2%)
Latinx	7 (12.7%)	Unknown or not reported	2 (3.6%)
Non-Latinx	46 (83.6%)	**Caregiver relationship to child**	
Unknown/Not Reported	2 (3.6%)	Biological parent	48 (87.5%)
		Adoptive parent	1 (1.8%)
Caregiver Gender		Step-parent	2 (3.6%)
Male	4 (7.3%)	Biological maternal grandparent	2 (3.6%)
Female	51 (92.7%)	Other non-biological relation	1 (1.8%)
Caregiver Age (years)	44.7 (8.07)	Unknown or not reported	1 (1.8%)

Aim 1: Description of Parent-Reported Baseline Family Functioning Patterns.

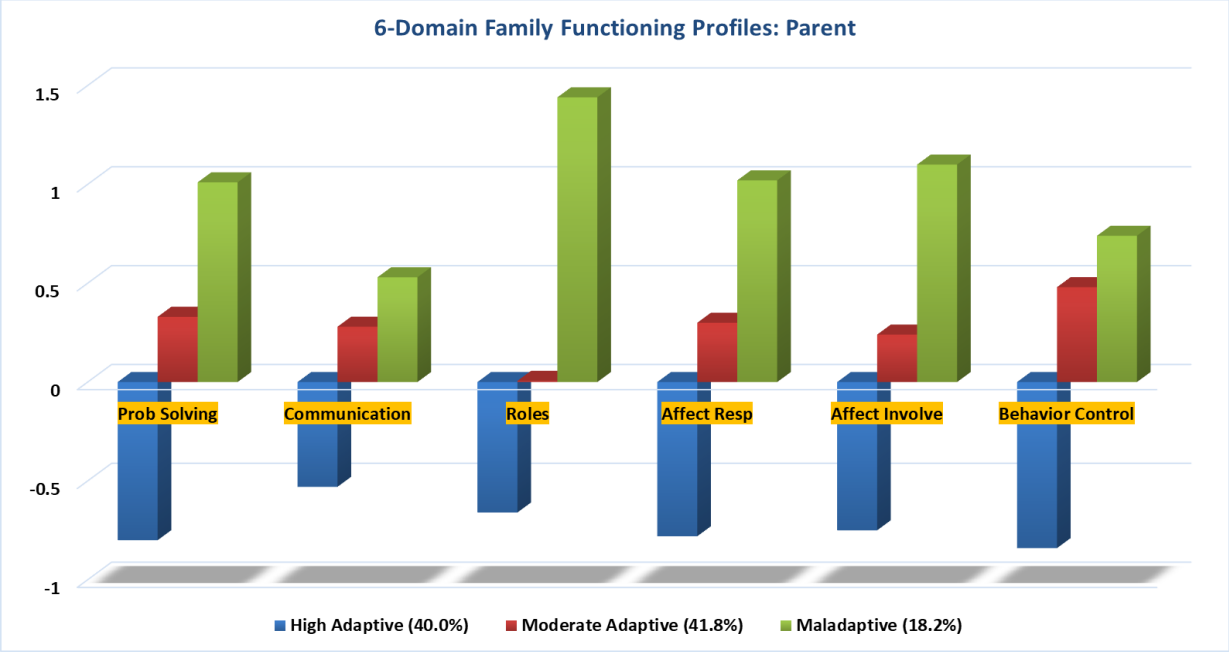
As previously discussed, prior work on family functioning suggested a three-cluster solution was most appropriate for capturing family functioning patterns among families of a child with a chronic illness: (1) an adaptive family functioning group, (2) a maladaptive family functioning group, and (3) a group with variable patterns of family functioning (Al Ghriwati et

al., 2017; Al Ghriwati et al., 2021; Ozono et al., 2010). The present study found a three-cluster solution of family functioning with (1) a *high adaptive family functioning group* demonstrating better functioning across all six domains, including role definition, affective involvement, behavioral control, affective responsiveness, communication, and problem solving (40.0%; *n*=22); (2) a *moderate adaptive family functioning group* demonstrating poorer functioning across all six domains (41.8%; *n*=23); and, (3) a *maladaptive family functioning group* with the least optimal family functioning scores observed across all six domains (18.2%; *n*=10). See *Figure 1*.

Descriptive statistics for the participants in the three groups identified in the cluster analysis are displayed in *Table 1b*. Non-standardized FAD subscale scores for each profile are described in *Table 2* and *Figure 2*. Previously developed clinical cut-off scores for impairment in family functioning were included in *Table 2* for descriptive purposes (Miller et al., 1985). As expected, for the high adaptive family functioning group, no dimension of family functioning was above the clinical cut-off indicating no impairments in any domain of family functioning. On the other hand, the moderate adaptive family functioning group had a clinically elevated score in the domain of roles, suggesting unhealthy family functioning for that domain. As expected, the maladaptive family functioning group reported clinically significant deficits in mean scores of affective involvement, affective responsiveness, roles, communication, and problem solving domains.

Figure 1

Three-Cluster Solution of Family Functioning



Note. Lower z scores indicate better functioning; higher z scores indicate worse functioning

Table 1b

Baseline Demographic and Medical Characteristics of Patients by Baseline Family Functioning Profile (N=55)

	High Adaptive (*N* = 22) *N* (%) or *M (SD)*	**Moderate Adaptive (*N* = 23)** *N* (%) or *M (SD)*	**Maladaptive (*N* = 10)** *N* (%) or *M* (*SD*)
Patient Diagnosis			
Blood Disorder	4 (18.2)	4 (17.4%)	1 (10.0%)
Cancer	18 (81.8%)	19 (82.6%)	9 (90.0%)
Child Gender			
Male	9 (40.9%)	12 (52.2%)	5 (50.0%)
Female	13 (59.1%)	11 (47.8%)	5 (50.0%)
Child Age	12.4 (2.68)	14.2 (2.55)	13.5 (2.62)
Child Race/ Ethnicity			
Person of Color	11 (50.0%)	12 (52.2%)	7 (70.0%)
Non-Latinx White	11 (50.0%)	11 (47.8%)	3 (30.0%)
Household Income			
Less than $50,000	5 (22.7%)	9 (39.1%)	5 (50.0%)
More than $50,000	16 (72.7%)	14 (60.9%)	4 (40.0%)

Table 2

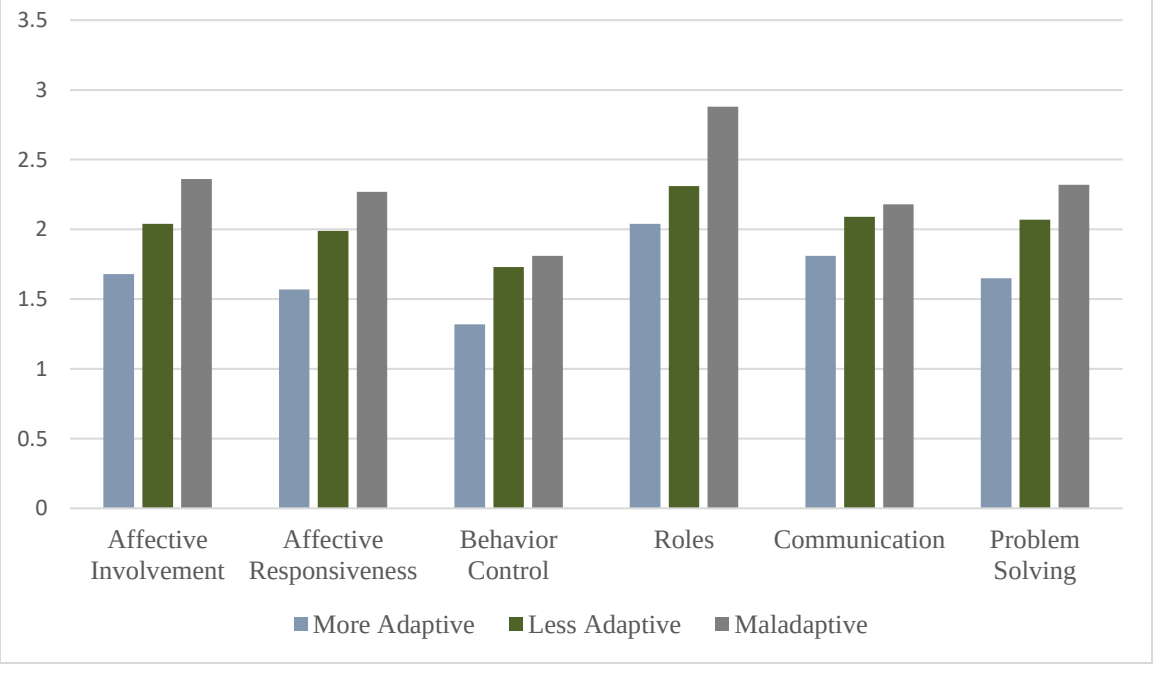
Descriptive Statistics of Baseline Family Functioning Domains by Cluster Assignment (N=55)

	High Adaptive (*N*=22)	**Moderate Adaptive** (*N*=23)	**Maladaptive** (*N*=10)	**1985 Clinical Cut-off**
Affective Involvement	1.7 (0.3)	2.0 (0.2)	2.4* (0.4)	2.1
Affective Responsiveness	1.6 (0.3)	2.0 (0.3)	2.3* (0.2)	2.2
Behavior Control	1.3 (0.2)	1.7 (0.2)	1.8 (0.3)	1.9
Roles	2.0 (0.3)	2.3* (0.2)	2.9* (0.3)	2.3
Communication	1.8 (0.3)	2.1 (0.2)	2.2* (0.5)	2.2
Problem Solving	1.7 (0.3)	2.1 (0.1)	2.3* (0.4)	2.2

Note. Higher scores for each domain indicate poorer functioning; * = above clinical cut-off

Figure 2

Non-Standardized FAD Mean Subscale Scores by Family Functioning Profile



Aim 2: Predictive Models of Baseline Family Functioning Profiles and 6-month Psychological Outcomes

Participants who completed baseline and 6-month data collection were included in models predicting 6-month psychological outcomes from baseline family functioning profiles (*N*

= 43). Due to the smaller sample size of the maladaptive family functioning group (n = 10), the moderate adaptive and maladaptive family functioning clusters were combined into a single group for Aim 2 analyses. These two less adaptive family functioning groups were similar profiles with all families demonstrating poorer family functioning across all domains (see *Figure 1)*. Combining these two less adaptive family functioning groups into a single less adaptive group allowed for comparisons between more adaptive and less adaptive family functioning profiles.

For Aim 2, two family functioning groups, a more adaptive family functioning group (46.5%; n=20) and a less adaptive family functioning group (53.5%; n=23), were examined to determine whether baseline family functioning group membership predicted patient's six-month psychological outcomes, per parent-proxy report. Descriptive statistics for these two family functioning groups are depicted in *Table 3*. Pearson correlation coefficients were calculated for all continuous variables prior to analyses (see *Table 4*). Independent t-tests and chi-square tests were used to determine what baseline demographic characteristics, if any, should be included as covariates in Aim 2. There were no significant differences between family functioning profiles on patient diagnosis, child gender, child race, and household income ($p > .05$). There was a significant difference between family functioning profiles and child age, $t(53) = -2.26$, $p = .01$. Patients in the less adaptive family functioning group were older (M=13.8 years). Patients in the more adaptive functioning group were younger (M = 12.1 years).

Table 3

Demographic and Medical Characteristics of Patients and Parent-Reported Scores of Child Well-Being at 6-months by Family Functioning Profile (N=43)

	More Adaptive (*N* = 20) *N* (%) or *M (SD)*	**Less Adaptive (*N* = 23)** *N* (%) or *M (SD)*
Patient Diagnosis		
Blood Disorder	3 (15.0%)	3 (13.0%)

	More Adaptive (*N* = 20) *N* (%) or *M (SD)*	**Less Adaptive (*N* = 23)** *N* (%) or *M (SD)*
Cancer	17 (85.0%)	20 (87.0%)
Child Gender		
Male	7 (35.0%)	12 (52.2%)
Female	13 (65.0%)	11 (47.8%)
Child Age	12.1 (2.53)	13.8 (2.38)
Child Race/ Ethnicity		
Person of Color	10 (50.0%)	12 (52.2%)
Non-Latinx White	10 (50.0%)	11 (47.8%)
Household Income		
Less than $50,000	4 (20.0%)	7 (30.4%)
More than $50,000	15 (75.0%)	15 (65.2%)
Psychological Distress	10.9 (10.7)	10.3 (9.98)
Quality of Life	71.8 (17.9)	74.6 (17.2)

Table 4

Means, Standard Deviations, and Correlations with Confidence Intervals of Patient 6-month Quality of Life and Psychological Distress per Parent-Proxy Report (N=43)

Variable	*M*	*SD*	1	2
1. Child Age	12.97	2.57		
2. Quality of Life	73.34	17.36	-.18 [-.45, .13]	
3. Psychological Distress	10.56	10.19	.08 [-.22, .37]	-.66** [-.80, -.44]

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

Aim 2a: Predictive Model of Baseline Family Functioning Profiles and Quality of Life (QoL) 6-months Later

First, t-tests were used to examine mean differences in 6-month QoL between adaptive and less adaptive family functioning profiles. There were significant differences observed in QoL by family functioning profile. Unexpectedly, those in the more adaptive family functioning

profile reported lower QoL (M = 71.8, SD = 17.9) compared to those in the less adaptive profile (M = 74.6, SD = 17.2), $t(42) = 27.10$, $p < .001$. Prior research suggested that patient gender, age, and race may ultimately influence quality of life (Isaac et al., 2020), thus we examined mean differences in QoL based on these factors using t-tests. Specifically, girls, $t(42) = -27.47$, $p < .001$, older children, $t(42) = -27.51$, $p < .001$, and children of color, $t(42) = -27.50$, $p < .001$, experienced lower quality of life.

As there were significant differences between each potential covariate and QoL, models with and without covariates were separately examined to determine the model with best fit. General linear regression models were conducted in R using the package 'saslm' to assess whether baseline family functioning cluster membership predicted parent-proxy report of child/adolescent QoL at six months. Our general linear model without covariates revealed baseline family functioning profiles did not significantly predict differences in mean QoL at 6-months, $F(1, 42) = 0.27$, $p = .61$, $\eta^2_p = .01$ (see *Table 5*).

Table 5

Associations between Baseline Family Functioning Group Membership and Six-Month Child Well-Being per Parent-Proxy Report without Covariates Included (N=43)

	Quality of Life			**Psychological Distress**		
Predictors	*Estimates*	*SE*	*p*	*Estimates*	*SE*	*p*
(Intercept)	74.63	3.65	**<0.001**	10.26	2.15	**<0.001**
More adaptive family functioning	-2.78	5.36	.61	0.64	3.15	.84
Less adaptive family functioning	0.00	0.00		0.00	0.00	
R^2 / R^2 adjusted	.01 / -.02			. 01 / -.01		

As shown below (*Table 6a*), the model with age entered as a covariate demonstrated the best model fit (R^2= .11, R^2-adj = .04), thus those results will be described here. When including

age as a covariate, baseline family functioning profiles did not significantly predict mean differences in 6-month quality of life, F (3, 42) = 1.56, p = .21, η^2_p = .07 (see *Table 6a*). Similarly, baseline family functioning, when controlling for age, was not a significant predictor of 6-month QoL [t = -1.70, p = .10]. On the other hand, baseline age was a significant predictor of 6-month QoL when controlling for baseline family functioning profiles [t = -2.09, p = .04]. QoL decreased as patient age increased (see *Figure 3a*). In contrast, the interaction between family functioning profiles and age was not significant [t = 1.54, p = .13]. Although not significant, there is a trend that those in the more adaptive family functioning group had relatively consistent QoL scores across all patient ages. On the other hand, for the less adaptive family functioning group, parent-reported patient QoL was higher for younger patients and decreased by patient age, such that the oldest patients had the lowest QoL (see *Figure 3b*).

Table 6a

Associations Between Baseline Family Functioning Group Membership and 6-Month Quality of Life with Covariates Included (N=43)

	Model 1 (Age)			**Model 2 (Gender)**			**Model 3 (Race)**		
Predictors	*Estimates*	*SE*	*p*	*Estimates*	*SE*	*p*	*Estimates*	*SE*	*p*
(Intercept)	118.55	21.28	**<.001**	76.07	5.16	**<.001**	72.93	5.12	**<.001**
More adaptive family functioning	-48.41	28.50	.10	-1.85	8.50	.83	-4.40	7.60	.57
Less adaptive family functioning	0.00	0.00		0.00	0.00		0.00	0.00	
Child Age	-3.19	-2.09	**.04**	--			--		
More Adaptive Group x Child Age	3.33	1.54	.13	--			--		
Less Adaptive Group x Child Age	0.00	0.00		--			--		
Child Gender	--			-3.00	7.46	.69	--		

More Adaptive Group x Child Gender	--	-0.65	11.22	.95	--		
Less Adaptive Group x Child Gender	--	0.00	0.00		--		
Child Race	--	--			3.56	7.41	.63
More Adaptive Group x Child Race	--	--			3.08	10.86	.78
Less Adaptive Group x Child Race	--	--			0.00	0.00	
R^2 / R^2 adjusted	.11 / .04	.07 / .00			.03 / -.04		

Figure 3a

Main Effect of Age on 6-month Quality of Life

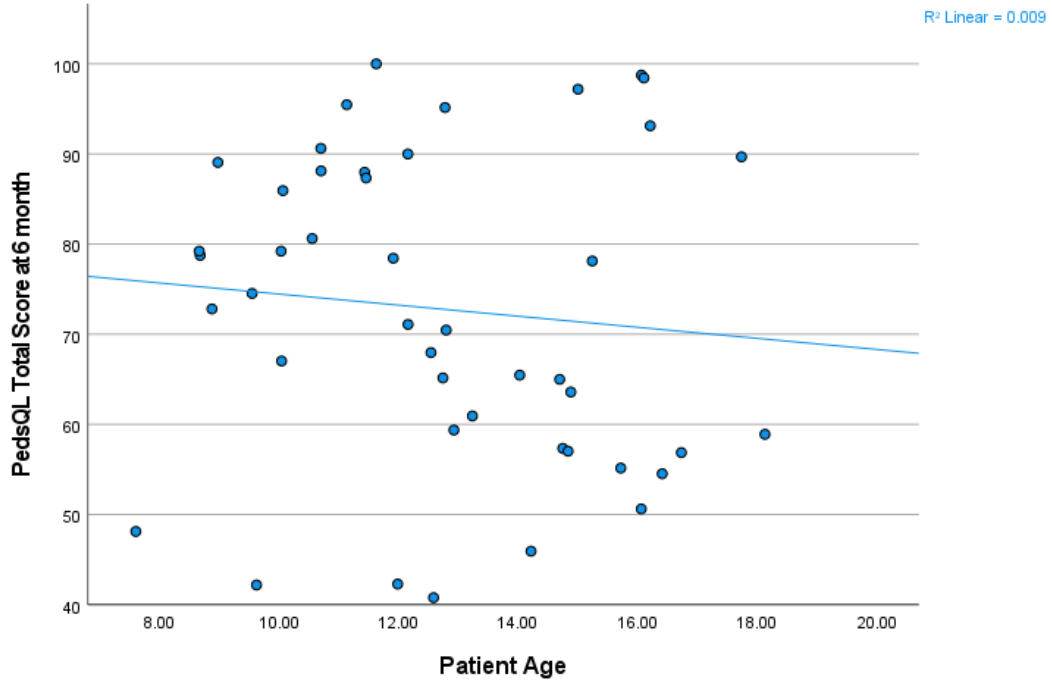
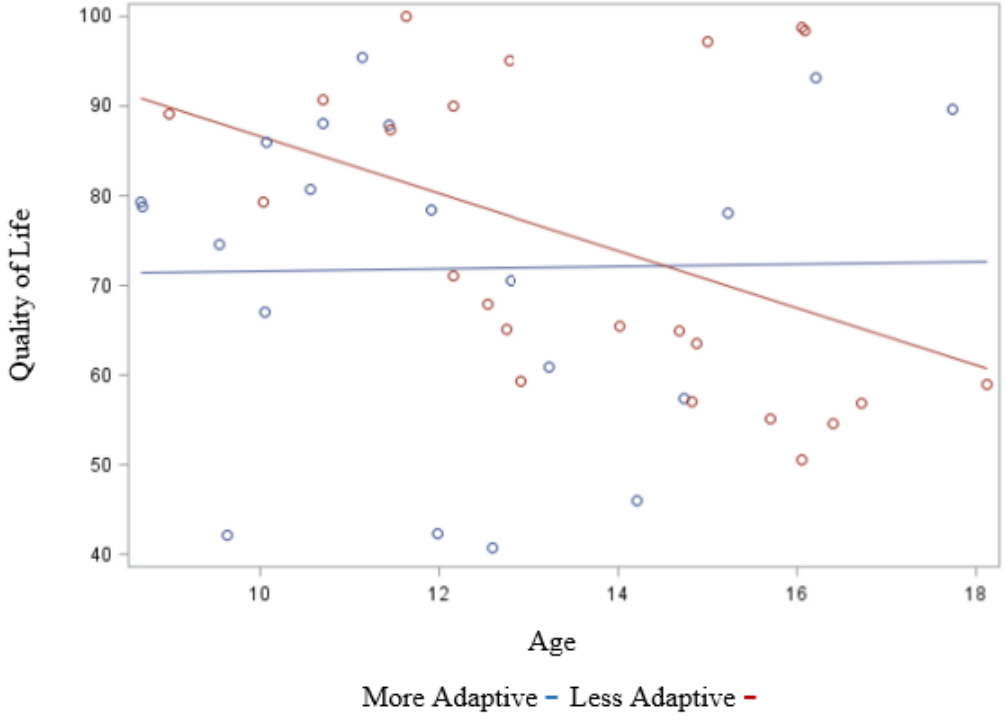


Figure 4b

Interaction between Baseline Family Functioning Groups and Age Predicting 6-month Quality of Life



Note. Interaction was not statistically significant.

Aim 2b. Predictive Model of Baseline Family Functioning Profiles and Psychological Distress 6-months Later

Similar to Aim 2a, a t-test exploring mean differences in psychological distress was conducted first. As expected, there were significant differences observed in psychological distress between the more and less adaptive family functioning profiles. Unexpectedly, those in the more adaptive functioning group had greater psychological distress at 6-months (M = 10.9, SD = 10.7) compared to those in the less adaptive functioning group (M = 10.3, SD = 10.0), $t(42) = 5.80$, $p \leq .00$. It is notable that the mean difference was negligible (0.6 difference). Prior research suggested patient gender, age, and race may ultimately influence psychological distress (Isaac et al., 2020), thus we examined mean differences in psychological distress based on these factors using t-tests. Specifically, girls, $t(42) = 6.43$, $p < .001$, older children, $t(42) = 6.49$, $p < .001$, and children of color, $t(42) = 6.47$, $p < .001$, experienced greater psychological distress. As

differences between covariates and psychological distress were significant, models with and without covariates were separately examined to determine the model with best fit. General linear regression models were conducted in R using the package 'saslm' to assess whether baseline family functioning cluster membership predicted parent-reported child/adolescent psychological distress at six months. Our general linear model without covariates revealed baseline family functioning was not significantly associated with 6-month psychological distress, F (1, 42) = 0.04, p = .84, η^2_p = .00 (see *Table 5*).

In parallel to Aim 2a, the model with age entered as a covariate revealed the best model fit (R^2= .09, R^2-adj = .02), thus those results will be described here. When including age as a covariate, baseline family functioning profiles did not significantly predict mean differences in 6-month psychological distress, F (3, 42) = 1.23, p =.31, η^2_p = .08 (see *Table 6b*). Similarly, baseline family functioning, when controlling for age, was not a significant predictor of 6-month QoL [t = 1.84, p = .07]. When controlling baseline family functioning, baseline age also did not significantly predict subsequent psychological distress [t = 1.72, p = .09]. There also was not a significant interaction between age and family functioning profile [t = -1.80, p = .08]. Although not statistically significant, there is a trend that those in the more adaptive family functioning group had decreasing psychological distress as patient age increased. On the other hand, for the less adaptive family functioning group, parent-reported patient psychological distress was lower for younger patients and increased by patient age, such that the oldest patients had higher levels of psychological distress compared to younger patients (see *Figure 4*).

Table 6b

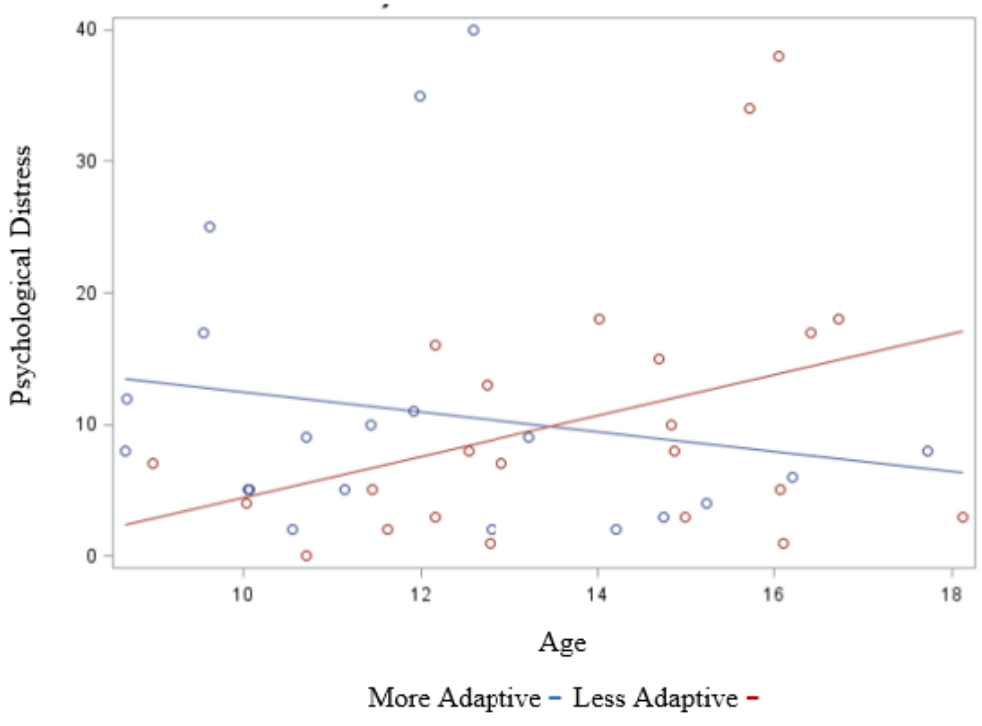
Associations Between Baseline Family Functioning Group Membership and Psychological Distress at 6-months with Covariates Included (N = 43)

	Model 1 (Age)			**Model 2 (Gender)**			**Model 3 (Race)**		
Predictors	*Estimates*	*SE*	*p*	*Estimates*	*SE*	*p*	*Estimates*	*SE*	*p*

(Intercept)	-11.18	12.63	.38	8.33	3.01	**<.001**	12.50	3.00	**.009**
More adaptive family functioning	31.19	16.91	.07	1.38	4.96	.78	-2.30	4.45	.61
Less adaptive family functioning	0.00	0.00		0.00	0.00		0.00	0.00	
Child Age	1.56	0.90	.09	--			--		
More Adaptive Group x Child Age	-2.31	1.29	.07	--			--		
Less Adaptive Group x Child Age	0.00	0.00		--			--		
Child Gender	--			4.03	4.36	.36	--		
More Adaptive Group x Child Gender	--			-2.21	6.55	.74	--		
Less Adaptive Group x Child Gender	--			0.00	0.00		--		
Child Race	--			--			-4.68	4.34	.29
More Adaptive Group x Child Race	--			--			6.01	6.36	.35
Less Adaptive Group x Child Race	--			--			0.00	0.00	
R^2 / R^2 adjusted	.09 / 0.02			.03 / -.05			.03/ .04		

Figure 5

Interaction between Baseline Family Functioning Groups and Age Predicting 6-month Quality of Life



Note. Interaction was not statistically significant.

Discussion

To our knowledge, this is the first study to examine caregiver-reported profiles of family functioning in a cohort of pediatric hematology-oncology patients and to assess whether these patterns were associated with child well-being as defined by parent-proxy report of patient quality of life and psychological functioning. We conducted a cluster analysis to identify mutually exclusive groups of family functioning patterns based on parent-reported ratings of family functioning across domains of affective involvement, affective responsiveness, behavior control, roles, communication, and problem solving. We hypothesized that there would be an adaptive family functioning group with more optimal scores observed across all dimensions, a maladaptive family functioning group with poorer scores observed across all dimensions, and at least one group with variable scores observed across family functioning dimensions (e.g., parent report of adaptive family functioning in some areas and less adaptive family functioning in others). Results from the cluster analyses revealed that the solution with the best model fit

consisted of three family functioning groups: a *high adaptive group* defined by better levels of affective involvement, affective responsiveness, behavior control, roles, communication, and problem solving; a *moderate adaptive group* defined by poorer family functioning across all six domains; and a *maladaptive group* defined by the least optimal levels of family functioning across all six domains. However, we did not identify a mixed family functioning group, as hypothesized.

When comparing family functioning dimensions within each cluster to previously developed clinical cut-offs for the FAD (Miller et al., 1985; see *Table 2*), we found that nearly all dimensions in the maladaptive group were above clinical cutoffs (i.e., role definition, affective responsiveness, affective involvement, communication, and problem solving); whereas only role definition indicated clinical impairment in the moderate adaptive family functioning cluster. As expected, no dimensions were above the clinical cutoff for the high adaptive family functioning group. Thus, our conceptualization of the three clusters as high adaptive, moderate adaptive, and maladaptive appears to be descriptively, and potentially, clinically meaningful.

For purposes of data analysis, we collapsed the moderate and maladaptive family functioning groups into a single group, less adaptive, given both groups demonstrated less adaptive family functioning across all six domains of family functioning. When examining mean differences in 6-month QoL and psychological distress between the more adaptive and less adaptive family functioning profiles using t-tests, the less adaptive family functioning group, which included moderate adaptive and maladaptive family functioning profiles, had higher quality of life and less psychological distress compared to the more adaptive family functioning group. Although these mean differences were negligible for both quality of life, M_{QoL} = 71.8 for high adaptive vs. 74.6 for less adaptive (i.e., a 2.8 difference); and, psychological distress, $M_{DSM5\text{-}total\ score}$ = 10.9 for high adaptive vs. 10.3 for less adaptive (i.e., a 0.6 difference). It is

notable that family functioning profiles and 6-month outcomes were both based on parent-reported family functioning and parent-reported child/adolescent quality of life and psychological distress at 6-months. Future work should examine whether these current findings focused on parent-reported family functioning and parent-report of child psychological outcomes are generalizable to patient-reported family functioning profiles, quality of life, and psychological distress. Similarly, future work should further examine family functioning profiles of patient-parent dyads to determine if parents and patients are reporting consistent or inconsistent family functioning patterns and how these contribute to patient outcomes.

Unexpectedly, our predictive models utilizing general linear modeling indicated that baseline family functioning was not a significant predictor of either psychological distress or QoL at 6-months even when controlling for patient age, gender, and minority status. Prior research has shown an association between family functioning and QoL. Many studies report that children with chronic illness experience better quality of life when families have more supportive and cooperative relationships with one another (Fardell et al., 2017; Stokoe et al., 2022). Although, some research has shown that family functioning may not be associated with all dimensions of QoL (Zheng et al., 2018). Future work should explore how family functioning profiles predict the different aspects of QoL (e.g., social, physical, emotional, school). Further, there is limited research on the link between family functioning and QoL for pediatric hematology patients. Research for this population has addressed parent factors, but not the role of the family system, in enhancing children's QoL. More research is needed to understand how the role of family functioning in promoting patients' QoL may differ across chronic illness populations and developmental phases.

Findings in the current study are surprising given the prior evidence that good family functioning is beneficial for the mental health of children with chronic illness (Ourers et al.,

2014). There are some inconsistencies in past research findings that may provide insight into the present results. For example, although severity of mental health symptoms has been found to increase over the course of cancer treatment, some evidence suggests these increases are not significant or clinically relevant (Katz et al., 2018). Though some studies have found evidence for the role of family functioning in children's anxiety symptoms, others have failed to find an association (Kunin-Batson et al., 2016). It is notable that the DSM-5 measure used in the present study reports on a parent-proxy screening measure of child/adolescent mental health that identifies clinically significant concerns warranting further attention by a mental health clinician. No prior studies have used the DSM-5 cross cutting symptom measure to examine the relationship between family functioning and mental health outcomes. This measure identifies heightened psychological distress by a brief symptom count of multiple psychiatric domains (e.g., depression, somatic concerns, anxiety, anger/irritability, inattention, sleep, etc.), hence providing a more general psychological distress score. Although the DSM-5 screening measure offers insight into overall levels of psychological distress, the measure is not intended to provide a comprehensive evaluation of any one specific mental health domain. As previously described, prior research on family functioning evaluated distress as a narrower construct. Future work should use construct-specific measures of anxiety, mood, disease distress, etc. to capture how family functioning profiles may be predictive of more specific mental health outcomes.

Additionally, baseline child age significantly predicted 6-month QoL but not 6-month psychological distress. Several previous studies have identified age differences in QoL, with most reporting older age being associated with poorer QoL (Stokoe et al., 2022). These results highlight the importance of identifying factors that promote resilience and psychological well-being for adolescents with cancer or a blood disorder. Given the socioemotional turbulence associated with adolescence, both peer support and emotional involvement of parents may be

important for adolescents' ability to adapt to stressful circumstances associated with chronic illness (Juth, 2016).

In the present study, there was not a significant interaction between patient age and family functioning group for either QoL or psychological distress. Although our findings in the predictive models were not significant, there are trends towards significance, thus the non-significant findings will be highlighted. For quality of life at 6-months, those in the more adaptive family functioning group had relatively consistent QoL scores across all patient ages. On the other hand, for the less adaptive family functioning group, QoL was higher for younger patients and decreased by patient age, such that the oldest patients had the lowest QoL. Similarly, although not statistically significant, those in the more adaptive family functioning group had decreasing psychological distress as patient age increased. This suggests that for families with more adaptive family functioning, there could be other protective factors for adolescents that were not assessed in the current study, such as peer support, greater use of adaptive coping strategies, or participation in mental health treatment that younger children may not be utilizing for various reasons. On the other hand, for the less adaptive family functioning group, parent-reported patient psychological distress was lower for younger patients and increased by patient age, such that the oldest patients had higher levels of psychological distress compared to younger patients. If there is increased discord between adolescents and their parents, this is likely to increase psychological distress while also impacting family dynamics and relationships.

Examining children's perception of their own well-being may be important to understanding how family functioning impacts psychological distress for children with chronic illness. Child reports would offer insight into how children undergoing treatment perceive how their family works together and provides support. Older children and adolescents, in particular, may possess the self-awareness and insight to effectively judge their family's level of

functioning and their own psychological state. Given their increased need for independence, adolescents are more likely to conceal their emotional states from their parents than younger children. Thus, the importance of peer relationships for adolescence, which are often impacted during the active phases of cancer treatment or during hospitalizations for disease management. Additionally, some research on the FAD has suggested that children rate family functioning as less healthy than parents (Sawyer and Sarris, 1988). Evidence on protective buffering in adults with chronic illness suggests children may conceal their suffering in order to protect their parents from worrying about them (Langer et al., 2009). Even if parents perceive family functioning to be highly adaptive, the child's well-being may be more affected by their own perception of family functioning. Parent reports may therefore fail to accurately represent pediatric patient experiences. Future work should therefore include child reports and consider developmental differences in pediatric hematology and oncology patients' well-being.

These results provide valuable additions to the limited work on patterns in family functioning for children with a hematology or oncology disorder. To our knowledge, this is the first study to evaluate profiles of family functioning using the FAD. Therefore, this work may inform researchers who wish to categorize families using the FAD in chronic illness populations. Since the FAD is a common tool for measuring family functioning in the chronic illness literature, information informing person-based approaches using the scale is highly valuable. Unlike a variable approach, a person-centered approach gives researchers insight into patterns of functioning in the family system. This study builds upon prior research to describe patterns of risk and resilience in family functioning. Given that none of the family functioning profiles identified in the present study indicated clinically relevant impairment in behavior control, it is possible that families of children with a blood disorder or cancer have rules and expectations for how to behave and treat one another even through treatment for the child's illness. Both the

moderate adaptive and maladaptive family functioning profiles demonstrated impairment in roles, while the maladaptive family functioning profile also showed impaired affective involvement, affective responsiveness, communication, and problem solving.

Results that roles are impaired for families of children with cancer or a blood disorder are unsurprising given literature on how family organization changes following diagnosis of a chronic illness. As previously discussed, families of children with chronic illness experience changes in routine and increases in caregiver burden (Bates et al., 2021). When routines are disrupted, it may be more difficult for family members to continue with their previous responsibilities. Roles must also change, as responsibilities shift to accommodate changing needs (e.g., caregiving responsibilities) over time. Similarly, when families experience more stress or exhaustion, domestic tasks may feel more burdensome, and they may feel less satisfied with other family members' contributions. Our results suggest that even families who are demonstrating more adaptive levels of family functioning may still need assistance in role definition as a preventative intervention. Given that the less adaptive and maladaptive family functioning profiles made up over half of the present sample, many families might benefit from assistance with defining clear responsibilities and ensuring that family members feel satisfied with the distribution of domestic labor.

In addition to roles, the other dimensions of family functioning except behavior control (i.e., affective involvement, affective responsiveness, communication, and problem solving) were also impaired for families in the maladaptive family functioning group. Problem solving is an important aspect of family functioning for families of children with chronic illness as they must work together to make decisions about the child's medical care. Additionally, families function better when they express affection and respond to each other with warmth and support (Barrera et al., 2007). Although prior studies have reported increased emotional closeness

following a diagnosis of a chronic illness (Van Schoors et al., 2015), the scores for affective responsiveness and affective involvement in the maladaptive family functioning group suggest that a subset of families with cancer or a blood disorder experience significant impairment in their ability to feel and express care for one another. Similarly, prior research has shown that parents of children with chronic illness tend to report equal or better levels of communication compared to parents of healthy children (Van Schoors et al., 2015). Though many families with hematology or oncology disorders are resilient and maintain positive and cooperative relationships with one another, it is important that psychosocial resources are available to help the subset of families who require support to better work together to solve problems, provide each other with love and care, and communicate effectively.

There were several limitations to the current study that should be considered in future research on family functioning. Internal consistency of family functioning subscales was low in the current sample. Work is needed to evaluate whether all items in the FAD reliably measure their respective construct. Further, some research suggests the FAD may not be a valid measurement of family functioning across racial and ethnic groups (Aarons et al., 2007); thus, results may not accurately represent all families' experiences. Future research should also consider using multi-site research designs to recruit a larger sample with more homogeneity in developmental stage, diagnosis, etc. The majority of the current cohort was diagnosed with cancer. There may be differences in family functioning observed between those with an acute life-threatening illness compared to those with a chronic, life-long illness (de Souza et al., 2019). The percentage of patients with blood disorders in the sample was low, so we were unable to make meaningful inferences about differences between oncology and non-oncology patients. Future work should evaluate the utility of the FAD in diverse samples and assess family

functioning profiles in larger samples that allow for examination of differences between disease groups.

Prior research identified family functioning profiles in which families demonstrate a mix of adaptive and maladaptive areas of functioning (e.g., Al Ghriwati et al., 2017). Work that builds upon the profiles found here will help clarify the diversity of families' strengths and weaknesses. Additionally, qualitative research could also provide additional insight into how families' characteristics and identities shape their perception of optimal family functioning. Given that prior research has suggested significant associations between family functioning and child well-being outcomes, larger samples or qualitative studies are needed to understand how families perceive how well they work together and whether that is relevant to the child's ability to cope with chronic illness.

It is notable that patients and families in the current study had access to a large multidisciplinary psychosocial team of psychology, social work, and chaplaincy. Families who presented with higher levels of adaptive family functioning at baseline may have received less psychological and psychosocial services than families who were functioning less well. Thus, improving psychological well-being and QoL of children in moderate and maladaptive family functioning groups, but not for those in the high adaptive family functioning group, who may have received less services. Additionally, patients included in this study took part in a cognitive behavioral narrative therapy exercise in which they created artistic narratives using avatars of their medical experiences. Processing the emotions associated with their medical challenges and treatment may have naturally improved their well-being over time regardless of their family life. Greater utilization of mental health services and participation in the avatar intervention may explain why better patient quality of life and lower psychological distress was observed six months later. Further, data was collected during the COVID-19 pandemic. Given the variety of

changes impacting families of a child with a chronic illness (Darlington et al., 2019), it is unclear how results may have differed under less disruptive conditions. Work that assesses longitudinal change, including day-to-day fluctuations in family functioning could demonstrate how changes in family functioning might impact later patient well-being outcomes.

Interventions that help families maintain healthy relationships are important given the potential for impairment in family functioning as evidenced by more than half our sample identifying with family functioning profiles of moderate adaptive family functioning (41.8%) or maladaptive functioning (18.2%). Integrative care may help improve family functioning by alleviating strain associated with navigating the healthcare system (Lee et al., 2020). Furthermore, inpatient medical settings and multidisciplinary medical sub-specialty programs that have integrated psychosocial teams might connect families in active treatment to therapists that can help them navigate medical adversity and maintain an adaptive family system (West et al., 2009). However, treatments for children with chronic illness targeting the family system are currently underdeveloped (Law et al., 2019), and very few have been specifically developed to improve family functioning (Winter et al., 2019). Randomized controlled trials evaluating a brief positive parenting intervention (Morawska et al., 2015) and a family empowerment program (Yeh et al., 2016) provide preliminary evidence of effective interventions for improving family functioning in families of children with chronic illness. More work is needed to understand how evidence-based interventions could capitalize on interpersonal strengths to help these families thrive despite medical adversity. The current person-centered approach highlights the importance of developing interventions that assist the subset of families who experience difficulty in multiple areas of family functioning.

The current study provides evidence of three distinct profiles of family functioning among pediatric hematology-oncology patients. However, these profiles did not predict quality

of life or psychological distress as expected, and more research is needed to understand the relevance of family functioning for overall well-being within this population. Although the current study does not provide evidence that membership in a specific family functioning profile predicts subsequent well-being outcomes, previous research supports family functioning as an important predictor for many psychological outcomes in families of children with chronic illness. Research that includes larger and more homogenous samples, multiple reporters, and longitudinal change in family functioning may provide more insight into this association.

It is important to note that family functioning impacts all individuals in the family system, and cascading effects of poor family functioning may not be apparent in the short-term. Furthermore, future studies incorporating multiple predictors of well-being could be beneficial in understanding how to provide care for these families. These include factors such as physical exercise (Dimitri et al., 2020), mindfulness (Stritter et al., 2021), parent mental health (Lewandoska, 2022), and social determinants of health (Bemis et al., 2015). Other forms of social support, such as those from extended family members, may also account for coping processes that family functioning may not encompass (Kelada et al., 2019). A more comprehensive model that incorporates multiple possible determinants of patient resilience could illustrate the relative importance of these factors as possible mechanisms for intervention. Together, findings of this study provide insight into family dynamics among children with a blood disorder or cancer and may inform future studies utilizing the FAD or person-centered approaches for understanding families of children with chronic illness.

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Appendix A

PACE Study: Demographics Form

1. Child/Adolescent's Gender O Male O Female

2. Child/Adolescent's Date of Birth ____ / ____ / _____

3. Child/Adolescent's Ethnicity
- O Hispanic or Latino (Specify in Q. 4)
- O Not Hispanic or Latino (Skip to Q. 5)
- O Unknown or Not Reported (Skip to Q. 5)

4. Specify Child/Adolescent's Hispanic or Latino Ethnicity: Select Only One Category
- O Cuban
- O Mexican
- O Puerto Rican
- O South or Central American
- O Other Spanish Culture or Origin, Specify _____
- O More Than One Hispanic Ethnicity, Specify _____
- O Unknown or Not Reported
- O Not Applicable: Not Hispanic or Latino

5. Child/Adolescent's Race: Select Only One
- O American Indian or Alaska Native
- O Asian
- O Black or African American
- O Native Hawaiian or Pacific Islander
- O White or Caucasian
- O More Than One Race (Specify in Q. 6)
- O Unknown or Not Reported

6. If Applicable: Specify Child/Adolescent's Multiple Races: Select All That Apply:
- O American Indian or Alaska Native
- O Asian
- O Black or African American
- O Native Hawaiian or Pacific Islander
- O White or Caucasian
- O Unknown or Not Reported
- O NA: Not Multi-racial

7. Has your child/adolescent been diagnosed with any learning disabilities? O Yes O No

8. Is your child/adolescent receiving any special services at school?

O Yes: O 504 Plan O IEP O Homebound O Other Services _____
O No

9. Has your child/adolescent received any mental health services in the last year? O Yes O No

10. If your child/adolescent has received any mental health services within the last year, what services has he/she received? Select All That Apply.

O Counseling
O Medication
O In-Patient Program
O Day Treatment Program
O Other, Specify _____
O Not Applicable: Has Not Received Mental Health Services in Last Year

11. What is the highest grade or level of schooling that your child/adolescent has completed?

O 1 - 6th grade (elementary school)
O 7th grade (middle school)
O 8th grade (middle school)
O 9th grade (high school)
O 10th grade (high school)
O 11th grade (high school)
O 12th grade (high school)
O Training after high school, other than college
O Some college
O Other (Specify) _____

III. Demographics: Primary Caregiver:

12. Primary Caregiver's Gender O Male O Female

13. Primary Caregiver's Date of Birth ____ / ____ / _____

14. Primary Caregiver's Relationship to Adolescent

O Biological Parent
O Adoptive Parent
O Step-Parent
O Foster Parent
O Biological Maternal Grandparent
O Biological Paternal Grandparent
O Full Sibling
O Other Biological Relative, Specify _____
O Other Non-Biological Relation, Specify _____

15. Primary Caregiver's Marital Status

- O Never Married
- O Married
- O Separated
- O Divorced
- O Widowed

16. Highest Grade or Level of Education Completed (Primary Caregiver):

- O 6th Grade or Less Than 6th Grade
- O 7th to 8th Grade
- O 9th to 11th Grade
- O High School Diploma or G.E.D.
- O Vocational, Trade School, or Associate's courses after high school
- O Vocational, Trade School, or Associates Degree
- O Courses toward Four-Year College Degree
- O Bachelor's Degree or Four-Year College Degree
- O Master's Degree
- O Professional Degree (M.D., Ph.D., J.D.)

17. What is your employment status?

- O Unemployed (Skip to #19)
- O Laid off
- O Self-employed
- O Employed

18. What kind of work do you do (what is your occupation)?

_____

(for example: electrical engineer, machinist, stock clerk, farmer)

19. Is there a spouse, partner, or secondary caregiver in the home? ***(If NO, skip to Q #27)***

☐ Yes (Q. 20) ☐ No

IV: Demographics: Secondary Caregiver:

20. Secondary Caregiver's Gender O Male O Female

21. Secondary Caregiver's Date of Birth ____ / ____ / _____

22. Secondary Caregiver's Relationship to Adolescent

- O Biological Parent
- O Adoptive Parent
- O Step-Parent
- O Foster Parent
- O Biological Maternal Grandparent
- O Biological Paternal Grandparent
- O Full Sibling
- O Other Biological Relative, Specify _____
- O Other Non-Biological Relation, Specify _____

23. Secondary Caregiver's Marital Status

- O Never Married
- O Married
- O Separated
- O Divorced
- O Widowed

24. Highest Grade or Level of Education Completed (Secondary Caregiver)

- O 6th Grade or Less Than 6th Grade
- O 7th to 8th Grade
- O 9th to 11th Grade
- O High School Diploma or G.E.D.
- O Vocational, Trade School, or Associate's courses after high school
- O Vocational, Trade School, or Associate's Degree
- O Courses toward Four-Year College Degree
- O Bachelor's Degree or Four-Year College Degree
- O Master's Degree
- O Professional Degree (M.D., Ph.D., J.D.)

25. What is the secondary caregivers employment status?

- O Unemployed
- O Laid off
- O Self-employed
- O Employed

26. What kind of work does the secondary caregiver do (what is their occupation)?

_____

(for example: electrical engineer, machinist, stock clerk, farmer)

27. Does the child/adolescent live with (select all that apply):

- O Two caregivers
- O One Caregiver
- O Other (Specify) _____

28. Who is/are the primary caregiver(s) / guardian(s) with whom the child/adolescent lives?

- O With both Biological Parents in the same house
- O With both Biological Parents who share custody in separate homes
- O With One Biological Parent and One Step-Parent
- O With Other Non-Relative(s) (Specify) _____
- O With Single Parent
- O With Grandparent(s)
- O With Other Relative(s) (Specify) _____

29. Please list all of the adults and children, now living at home with the child/adolescent (e.g., parents, grandparents, siblings, etc.):

Relationship to Child	Gender	Age
_____	O Male O Female	_____
_____	O Male O Female	_____
_____	O Male O Female	_____
_____	O Male O Female	_____
_____	O Male O Female	_____
_____	O Male O Female	_____
_____	O Male O Female	_____

30. Total annual Household Income Before Taxes

- O Less than $19,000
- O $19,000 to $34,999
- O $35,000 to $49,999
- O $50,000 to $72,999
- O $73,000 to $126,500
- O More than $126,500

31. What is your religious affiliation?

- O Baptist
- O Buddhism
- O Catholic
- O Judaism
- O Lutheran
- O Methodist
- O Islam
- O Presbyterian
- O Other (Specify) _____
- O None
- O Decline to Respond

Appendix B

Family Assessment Device: Parent Report (Herzer et al., 2010)

INSTRUCTIONS

This questionnaire contains a number of statements about families. Read each statement carefully, and decide how well it describes your own family. You should answer according to how you see your family. For each statement are four (4) possible responses:

SA	Strongly Agree	The statement describes your family very accurately.
A	Agree	The statement describes your family for the most part.
D	Disagree	The statement does not describe your family for the most part.
SD	Strongly Disagree	The statement does not describe your family at all.

1.Planning family activities is difficult because we misunderstand each other.	SA ☐ A ☐ D ☐ SD ☐
2.We resolve most everyday problems around the house.	SA ☐ A ☐ D ☐ SD ☐
3.When someone is upset the others know why.	SA ☐ A ☐ D ☐ SD ☐
4.When you ask someone to do something, you have to check that they did it.	SA ☐ A ☐ D ☐ SD ☐
5.If someone is in trouble, the others become too involved.	SA ☐ A ☐ D ☐ SD ☐
6.In times of crisis we can turn to each other for support.	SA ☐ A ☐ D ☐ SD ☐
7.We don't know what to do when an emergency comes up.	SA ☐ A ☐ D ☐ SD ☐
8. We sometimes run out of things that we need.	SA ☐ A ☐ D ☐ SD ☐
9. We are reluctant to show our affection for each other.	SA ☐ A ☐ D ☐ SD ☐
10. We make sure members meet their family responsibilities.	SA ☐ A ☐ D ☐ SD ☐
11. We cannot talk to each other about the sadness we feel.	SA ☐ A ☐ D ☐ SD ☐
12. We usually act on our decisions regarding problems.	SA ☐ A ☐ D ☐ SD ☐
13. You only get the interest of others when something is important to them.	SA ☐ A ☐ D ☐ SD ☐
14. You can’t tell how a person is feeling from what they are saying.	SA ☐ A ☐ D ☐ SD ☐

15. Family tasks don't get spread around enough.	SA ☐ A ☐ D ☐ SD ☐
16. Individuals are accepted for what they are.	SA ☐ A ☐ D ☐ SD ☐
17. You can easily get away with breaking the rules.	SA ☐ A ☐ D ☐ SD ☐
18. People come right out and say things instead of hinting at them.	SA ☐ A ☐ D ☐ SD ☐
19. Some of us just don't respond emotionally.	SA ☐ A ☐ D ☐ SD ☐
20. We know what to do in an emergency.	SA ☐ A ☐ D ☐ SD ☐
21. We avoid discussing our fears and concerns.	SA ☐ A ☐ D ☐ SD ☐
22. It is difficult to talk to each other about tender feelings.	SA ☐ A ☐ D ☐ SD ☐
23. We have trouble meeting our financial obligations.	SA ☐ A ☐ D ☐ SD ☐
24. After our family tries to solve a problem, we usually discuss whether it worked or not.	SA ☐ A ☐ D ☐ SD ☐
25. We are too self-centered.	SA ☐ A ☐ D ☐ SD ☐
26. We can express feelings to each other.	SA ☐ A ☐ D ☐ SD ☐
27. We have no clear expectations about toilet habits.	SA ☐ A ☐ D ☐ SD ☐
28. We do not show our love for each other.	SA ☐ A ☐ D ☐ SD ☐
29. We talk to people directly rather than through go-betweens.	SA ☐ A ☐ D ☐ SD ☐
30. Each of us has particular duties and responsibilities.	SA ☐ A ☐ D ☐ SD ☐
31. There are lots of bad feelings in the family.	SA ☐ A ☐ D ☐ SD ☐
32. We have rules about hitting people.	SA ☐ A ☐ D ☐ SD ☐
33. We get involved with each other only when something interests us.	SA ☐ A ☐ D ☐ SD ☐
34. There is little time to explore personal interests.	SA ☐ A ☐ D ☐ SD ☐
35. We often don't say what we mean.	SA ☐ A ☐ D ☐ SD ☐
36. We feel accepted for what we are.	SA ☐ A ☐ D ☐ SD ☐
37. We show interest in each other when we can get something out of it personally.	SA ☐ A ☐ D ☐ SD ☐
38. We resolve most emotional upsets that come up.	SA ☐ A ☐ D ☐ SD ☐
39. Tenderness takes second place to other things in our family.	SA ☐ A ☐ D ☐ SD ☐
40. We discuss who are responsible for household jobs.	SA ☐ A ☐ D ☐ SD ☐
41. Making decisions is a problem for our family.	SA ☐ A ☐ D ☐ SD ☐
42. Our family shows interest in each other only when they can get something out of it.	SA ☐ A ☐ D ☐ SD ☐
43. We are frank (direct, straightforward) with each other.	SA ☐ A ☐ D ☐ SD ☐
44. We don't hold to any rules or standards.	SA ☐ A ☐ D ☐ SD ☐

45. If people are asked to do something, they need reminding.	SA ☐ A ☐ D ☐ SD ☐
46. We are able to make decisions about how to solve problems.	SA ☐ A ☐ D ☐ SD ☐
47. If the rules are broken, we don't know what to expect.	SA ☐ A ☐ D ☐ SD ☐
48. Anything goes in our family.	SA ☐ A ☐ D ☐ SD ☐
49. We express tenderness.	SA ☐ A ☐ D ☐ SD ☐
50. We confront problems involving feelings.	SA ☐ A ☐ D ☐ SD ☐
51. We don't get along well together.	SA ☐ A ☐ D ☐ SD ☐
52. We don't talk to each other when we are angry.	SA ☐ A ☐ D ☐ SD ☐
53. We are generally dissatisfied with the family duties assigned to us.	SA ☐ A ☐ D ☐ SD ☐
54. Even though we mean well, we intrude too much into each other's lives.	SA ☐ A ☐ D ☐ SD ☐
55. There are rules in our family about dangerous situations.	SA ☐ A ☐ D ☐ SD ☐
56. We confide in each other.	SA ☐ A ☐ D ☐ SD ☐
57. We cry openly.	SA ☐ A ☐ D ☐ SD ☐
58. We don't have reasonable transport.	SA ☐ A ☐ D ☐ SD ☐
59. When we don't like what someone has done, we tell them.	SA ☐ A ☐ D ☐ SD ☐
60. We try to think of different ways to solve problems.	SA ☐ A ☐ D ☐ SD ☐

Appendix C

DSM-5 Parent-Rated Level 1 Cross-Cutting Symptom Measure—Parent Report of Child
(American Psychiatric Association, 2013)

INSTRUCTIONS

The questions below ask about things that might have bothered your child. For each question, circle the number that best describes how much (or how often) your child has been bothered by each problem during the past TWO (2) WEEKS.

		During the past **TWO (2) WEEKS,** how much (or how often) has your child…	**None** Not at all	**Slight** Rare, less than a day or two	**Mild** Several days	**Moderate** More than half the days	**Severe** Nearly every day	**Highest Domain Score** (clinician)
I.	1.	Complained of stomachaches, headaches, or other aches and pains?	0	1	2	3	4	
	2.	Said he/she was worried about his/her health or about getting sick?	0	1	2	3	4	
II.	3.	Had problems sleeping—that is, trouble falling asleep, staying asleep, or waking up too early?	0	1	2	3	4	
III.	4.	Had problems paying attention when he/she was in class or doing his/her homework or reading a book or playing a game?	0	1	2	3	4	
IV.	5.	Had less fun doing things than he/she used to?	0	1	2	3	4	
	6.	Seemed sad or depressed for several hours?	0	1	2	3	4	
V. & VI.	7.	Seemed more irritated or easily annoyed than usual?	0	1	2	3	4	
	8.	Seemed angry or lost his/her temper?	0	1	2	3	4	
VII.	9.	Started lots more projects than usual or did more risky things than usual?	0	1	2	3	4	
	10.	Slept less than usual for him/her, but still had lots of energy?	0	1	2	3	4	
VIII.	11.	Said he/she felt nervous, anxious, or scared?	0	1	2	3	4	
	12.	Not been able to stop worrying?	0	1	2	3	4	
	13.	Said he/she couldn't do things he/she wanted to or should have done, because they made him/her feel nervous?	0	1	2	3	4	
IX.	14.	Said that he/she heard voices—when there was no one there—speaking about him/her or telling him/her what to do or saying bad things to him/her?	0	1	2	3	4	
	15.	Said that he/she had a vision when he/she was completely awake—that is, saw something or someone that no one else could see?	0	1	2	3	4	
X.	16.	Said that he/she had thoughts that kept coming into his/her mind that he/she would do something bad or that something bad would happen to him/her or to someone else?	0	1	2	3	4	
	17.	Said he/she felt the need to check on certain things over and over again, like whether a door was locked or whether the stove was turned off?	0	1	2	3	4	
	18.	Seemed to worry a lot about things he/she touched being dirty or having germs or being poisoned?	0	1	2	3	4	
	19.	Said that he/she had to do things in a certain way, like counting or saying special things out loud, in order to keep something bad from happening?	0	1	2	3	4	

Vita

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