

2022

A mixed methods analysis of the psychosocial functioning of siblings of youth with cancer: involvement in cancer treatment and cancer-related post-traumatic stress

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BOSTON UNIVERSITY
GRADUATE SCHOOL OF ARTS AND SCIENCES

Dissertation

**A MIXED METHODS ANALYSIS OF THE PSYCHOSOCIAL FUNCTIONING
OF SIBLINGS OF YOUTH WITH CANCER: INVOLVEMENT IN CANCER
TREATMENT AND CANCER-RELATED POST-TRAUMATIC STRESS**

by

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Submitted in partial fulfillment of the
requirements for the degree of
Doctor of Philosophy

2022

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DEDICATION

I would like to dedicate this work to the siblings and parents of youth with cancer who shared their personal experiences with me.

ACKNOWLEDGMENTS

I am incredibly grateful to everyone who supported me in this process. To my primary mentor, Dr. Kristin Long, I am honored to have been your first graduate student and to have had the opportunity to build the Child and Family Health Lab with you. You lead by such strong example, with work ethic, respect, and humility, and approach all that you do with a tremendous energy that motivates those around you. You created an environment within our lab where I felt simultaneously challenged and supported, and helped me to grow as a researcher, clinician, and human in so many ways. To my committee members, thank you for your thoughtful guidance and support throughout this process. I am grateful to have learned from you and benefited from your expertise. To the Clara Mayo Memorial Fellowship, thank you for providing the financial support for this research. To Phoebe, thank you for dedicating hours to coding qualitative interviews with me. I could not have done this research without you. To my amazing fellow lab members, Monica, Jenna, Katie, Ariel and Nicole, thank you for helping get this project over the finish line, and for being my family at BU. I am in awe of all of you and feel so lucky to have worked with you. To Lydia, thank you for paving the way for me. I am so grateful BU brought us together. To my BU and BCH cohorts, I could not have asked for brighter, stronger women to walk this path with me. I cannot wait to see the innovative ways you all shape the field of psychology. To my family, thank you for always supporting my dreams, and for the innumerable ways that you have helped me to reach this point. And finally, but certainly not least, thank you to my girlfriend Meesh and cat Stygg, your love made this all possible. I cannot wait to start our next adventure in Philadelphia.

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ABSTRACT

Siblings demonstrate variable psychosocial adjustment to childhood cancer, ranging from cancer-related post-traumatic stress symptoms (PTSS) to resilience. Theoretical frameworks suggest that PTSS are influenced by appraisals (e.g., fear of mortality) and coping. Siblings' presence within clinical settings and participation in cancer treatment may allow them to appraise cancer-related experiences as less threatening and utilize engagement coping strategies, but the nature and effect of siblings' involvement in cancer treatment remains unknown, especially in the context of COVID-19. This mixed-methods research (a) characterized siblings' involvement in cancer treatment, (b) investigated how siblings' involvement in cancer treatment, demographics, cancer characteristics, appraisals and coping relate to PTSS, and (c) characterized COVID-19's impact on siblings. Thirty-eight participants (22 siblings, 16 parents) from 16 families of youth with cancer completed qualitative interviews and quantitative measures. Study 1 qualitatively illustrated treatment involvement. Within the hospital and clinic, siblings emotionally supported patients, observed treatment, and learned about cancer. At home, siblings continued to support patients, assumed helper

roles with treatment, and adjusted to new cancer-induced norms (e.g., family diet, cleaning practices). Study 2 used a cross-sectional, convergent, mixed methods design to identify factors related to siblings' PTSS. Quantitative analyses revealed that siblings' PTSS were significantly related to siblings' age, appraisals and coping strategies. Mixed methods analyses showed that siblings who qualitatively described more consistent involvement in treatment, including access to information and opportunities to express emotions, had lower quantitative PTSS scores. Study 3 qualitatively described siblings' treatment involvement and experience of cancer in the context of COVID-19. Siblings worried about contracting and transmitting COVID-19 to patients, but also described that cancer-related experiences prepared them for pandemic-related precautions. Due to increased time at home, siblings encountered more cancer reminders and had fewer opportunities to "take a break" from cancer. Siblings' exclusion from clinical settings led to reduced understanding of cancer and increased cancer-related stress. Together, findings suggest that treatment involvement offers opportunities for siblings to process cancer-related cues and use engagement coping techniques, which may facilitate positive adjustment to cancer. Findings have important clinical implications related to (re)establishing family-centered care practices that explicitly include siblings.

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LIST OF ABBREVIATIONS

CPSS	Child PTSD Symptom Scale
CSI	Coping Strategies
FCC	Family-Centered Care
PICC	Peripherally Inserted Central Catheter
PTSS	Post-Traumatic Stress Symptoms

Chapter 1. General Introduction

Siblings' Psychosocial Adjustment to Cancer

Approximately 16,000 youth in the United States are diagnosed with cancer each year (American Cancer Society, 2014). Average 5-year survival rates have increased to over 80% with treatment (American Cancer Society, 2014); however, even in the context of cancer survival, the experience of cancer treatment has a considerable emotional impact on the entire family, including siblings (Long & Marsland, 2011). Cancer treatment is often associated with shifts in family roles, such as changes in child caregiving responsibilities, decreased parental time or attention for siblings, and fewer family activities (Long et al., 2015; Prchal & Landolt, 2012; Williams et al., 2009). Siblings of youth with cancer often experience emotional distress following the patient's cancer diagnosis, and endorse a range of negative emotions, such as fear, jealousy, loneliness, anger, and worry (Long et al., 2018). For some siblings, the psychosocial effects of cancer appear to influence functioning in other domains (e.g., poorer school performance, Alderfer et al., 2015) and may extend long after treatment has ended (Alderfer et al., 2003). However, some siblings demonstrate resilience in response to cancer and exhibit positive outcomes, such as improvements in maturity and responsibility (Long et al., 2015). A recent review of the literature (Long et al., 2018) suggests that risk factors for poorer sibling adjustment include lower social support, poorer family functioning, lower family income, racial or ethnic minority status, and shorter time since diagnosis; however, findings are inconsistent and further research is necessary to understand the processes that influence siblings' adjustment to cancer.

Sibling psychosocial risk processes may be related to the extent to which siblings experience cancer-related events as traumatic. Cancer treatment often involves potentially traumatic experiences for the patient (e.g., life threat, painful or frightening procedures) and their family members, who may worry that the patient will die (Alderfer et al., 2003; Goldenberg Libov et al., 2002). In response to potentially traumatic cancer-related events, patients and their family members may develop cancer-related post-traumatic stress symptoms (PTSS; National Child Traumatic Stress Network, 2003).” Cancer-related PTSS refers to a cluster of posttraumatic stress symptoms, specifically arousal (heightened startle reactions, irritability, trouble sleeping), re-experiencing (intrusive thoughts, nightmares) and avoidance (avoiding people, places, and things) related to medical events (Kazak et al., 2017). PTSS is normative in the short-term (e.g., first month after diagnosis), and may even serve adaptive purposes; for example, frequent thoughts about an event can facilitate processing, and periodic avoidance can be helpful in managing distress (Kazak et al., 2006). However, when these symptoms do not resolve and go untreated over weeks or months, they can create substantial distress, impair daily functioning, and develop into serious comorbid psychological disorders (Goenjian et al., 2005).

Although the vast majority of the research on PTSS in response to pediatric cancer has been conducted with patients and their parents, a majority of siblings experience PTSS (Kaplan et al., 2013), highlighting the importance of additional research to better understand PTSS in siblings. In the first few years following diagnosis, up to two-thirds of siblings endorse moderate to severe levels of PTSS (Kaplan et al., 2013).

Although not directly compared, these estimates are approximately 2.5 to 5 times higher than those of patients (across studies, 12-25% of patients endorse moderate to severe symptoms; Barakat et al., 1997; Butler et al., 1996; Kazak et al., 2004; Stuber et al., 1996) and 1.5 to 4 times higher than those of parents (across studies, 15-44% of parents endorse moderate to severe symptoms; Barakat et al., 1997; Brown et al., 2003; Kazak et al., 2004; Landolt et al., 2003). Siblings' PTSS include increased arousal and irritability, intrusive thoughts about cancer, and attempts to avoid thoughts and feelings related to cancer (Alderfer et al., 2003; Kaplan et al., 2013), all of which appear to interfere with siblings' overall functioning (Kaplan et al., 2013). Furthermore, approximately one-third of siblings demonstrate persistent PTSS, reporting moderate to severe symptoms 5 years (on average) after cancer treatment has ended (Alderfer et al., 2003).

The seemingly higher prevalence of PTSS in siblings (as compared to patients and parents) may be related to siblings' unique experience of cancer. One notable difference between siblings' experience and those of other family members is siblings' often limited or less comprehensive involvement in cancer treatment (Franklin et al., 2018, Long et al., 2017), warranting further research on the potential relationship between siblings' involvement in treatment and psychosocial adjustment. In the remainder of this introduction, I will first summarize what is known about siblings' involvement in cancer treatment and identify aspects of sibling involvement that warrant further research, informed by conceptualizations of other family members' involvement in treatment (e.g., fathers of children with chronic illness) and research on similar constructs (e.g., children providing care for family members). Next, drawing from current frameworks related to

traumatic stress, coping, and family systems theory, I will describe the various mechanisms through which siblings' involvement in treatment may influence their psychosocial functioning, highlighting gaps in the literature that will be addressed by the proposed exploratory study.

Siblings' Involvement in Cancer Treatment

While it can generally be assumed that patients and at least one parent are present and involved in cancer-related medical events and treatments, the same is not necessarily true for siblings. Although siblings' involvement in cancer treatment has not yet been systematically assessed, healthcare providers and parents report that siblings are routinely absent from clinical settings (Franklin et al., 2018, Long et al., 2017). Parents report that siblings often remain at home with extended family members or other caregivers during patients' hospital stays while parent(s) are at the hospital with the patient, and that siblings seldom attend clinic visits, which often occur during the school day (Long et al., 2017). Although these findings suggest that siblings as a whole demonstrate limited treatment involvement in hospital and clinic settings (in comparison to patients and parents), individual siblings likely vary in the extent to which they are present and participate in cancer treatment, and siblings may be involved in treatment in other ways, such as caring for patients at home.

Furthermore, while research has yet to characterize how siblings of youth with cancer have been impacted by the COVID-19 pandemic, it is likely that siblings have experienced changes within multiple domains of life, including restricted access within the patient's cancer treatment setting. When the COVID-19 pandemic started, healthcare

leadership were tasked with the challenge of how to balance pediatric cancer patients' critical need for treatment with the healthcare systems' need for policies that minimized the spread of COVID-19 (Sullivan et al., 2020). Many children's hospitals instituted stricter visitation policies, such as only allowing one adult caregiver at the patient's bedside, either at a time or for the duration of the patient's hospital stay, and excluding siblings and other visitors. Across the healthcare system, these pandemic-related changes to prioritize infection control have been associated with tradeoffs (Hugelius et al., 2021; Virani et al., 2020) that de-prioritized family-centered care (FCC), and how these tradeoffs impact siblings of youth with cancer and their involvement in treatment is in not yet understood.

Minimal research has focused on siblings' involvement in pediatric medical treatment, including childhood cancer treatment. Sibling treatment involvement is likely a complex, multidimensional construct; however, research has yet to propose a conceptual framework of the construct or to establish valid and reliable measures thereof. Research on other family members' involvement in treatment can be used to inform factors that may be relevant to consider related to siblings' involvement. Among pediatric chronic illness populations, a larger body of research exists on patient and maternal involvement in treatment (i.e. as primary caregivers), and recently paternal involvement has received increased attention (Taylor et al., 2020). Broadly, findings (e.g., Crespo et al., 2016; Phillips-Salimi et al., 2011; Taylor et al., 2020) suggest that when conceptualizing treatment involvement, it is important to consider not only the practical or logistical ways that one helps or participates in care, but also the reciprocal influence of one's treatment

involvement on relational factors (e.g., family functioning, family relationships) and their own psychosocial functioning (e.g., cancer-related stress, use of coping skills).

The small number of studies that have been conducted on siblings' preferences related to treatment involvement in pediatric cancer and related populations (i.e., bone marrow transplant) suggest that siblings' involvement in treatment likely also encompasses interrelated care management, relational, and sibling psychosocial factors. Frequent separations from family members due to cancer treatment (e.g., patient and parent(s) are at the hospital while sibling is at home with an extended family member) can result in siblings feeling distanced from the family and isolated (O'Shea et al., 2012). Additionally, limited involvement in medical appointments and interaction with healthcare providers may hinder siblings' understanding of cancer. Siblings frequently indicate uncertainty related to patient's cancer diagnosis or treatment (Jenholt Nolbris & Ahlström, 2014), and retrospectively, some siblings express that increased involvement in cancer treatment and opportunities to learn about cancer from the treatment team would have been helpful for their psychosocial adjustment (Lövgren et al., 2016). One study documented that siblings report a desire to do whatever they can to help the patient feel better, and describe strategies such as providing companionship, comforting them, and being more careful around them (Wilkins & Woodgate, 2007). However, research also documents that some siblings express a need for supports that help them "take a break" from cancer (Patterson et al., 2011), possibly suggesting that sustained treatment involvement can be experienced by some siblings as overwhelming.

Research on youth as family caregivers (i.e., providing care to family members

such as siblings) within the general population suggests that when caregiving responsibilities are excessive (20 hours or more per week) and prolonged (extending over several years), caregiving is associated with negative psychosocial outcomes for the child providing care, including stress, depression, and academic difficulties (East, 2010).

Alternatively, when responsibilities are more moderate, caregiving has been linked with positive outcomes such as improvements in maturity, self-confidence, and social skills (East, 2010). In addition to quantity and duration of caregiving, other factors appear to influence the relationship between caregiving and child adjustment, such as the extent to which the caregiving is voluntary and matched to the child's developmental abilities and needs (East, 2010). Research documents that the likelihood and extent of child caregiving is influenced by child demographic factors, such as age and gender. For example, girls routinely engage in more caregiving than boys (East, 2010), particularly when the cultural values of the family prescribe that girls should take on more household responsibilities (e.g., Latino families; McHale et al., 2005). Given that caregiving is a key component of sibling involvement in cancer treatment (i.e., assisting with patient's care), the relationship between treatment involvement and sibling psychosocial functioning may be influenced by similar factors (e.g., quantity of involvement, voluntariness of involvement). Additionally, the extent and nature of siblings' treatment involvement may be influenced by sibling demographic factors.

In sum, research has begun to identify siblings' preferences (e.g., desire to provide support; Wilkins & Woodgate, 2007) and unmet needs (e.g., knowledge about cancer; Jenholt Nolbris & Ahlström, 2014) related to treatment involvement. To date, no

research has documented the ways in which siblings actually report helping or participating in cancer treatment across settings (e.g., hospital, clinic, home) such as assisting with care or providing support. Additionally, research has yet to explore how siblings' treatment involvement relates to their family relationships and psychosocial functioning (e.g., adjustment to cancer, PTSS), or how siblings' treatment involvement and broader lives have been impacted by the COVID-19 pandemic. These gaps in the literature are important to address, as siblings' involvement in treatment may play a key role in influencing the extent to which siblings demonstrate resilience and/or emotional difficulties in response to cancer. In the following sections, I will describe the various mechanisms through which siblings' treatment involvement may influence their psychosocial functioning, informed by frameworks related to traumatic stress and coping.

Traumatic Stress Frameworks

The integrative trajectory model of PTSS (Kazak et al., 2006), originally developed with the patient in mind and extended to describe the experiences of the family as a whole, is the predominant model used to conceptualize PTSS in families of children with cancer. The model suggests that an individual's PTSS trajectory is influenced by the cancer-related events that occur throughout treatment and the individual's subjective appraisals of those events (Kazak et al., 2006; Price et al., 2016). A study done with patients (treated in oncology, cardiology, endocrinology, orthopedics, or pulmonology) and their caregivers and siblings reported that multiple aspects of treatment are potentially traumatic, including: receiving the diagnosis, being at the hospital, going through procedures, enduring medical emergencies, experiencing symptoms and physical

changes (first- or secondhand), and communicating with providers (Christofferson et al., 2020). However, findings were reported for the sample as a whole, and it remains unknown how potentially-traumatic events for siblings (17% of the sample) may differ from those of other family members, particularly given that siblings' direct involvement in the events identified may be more limited than that of patients or parents. Another study documented differences in patients' and parents' reports of the most prominent cancer-related stressors, as well as which stressors predict PTSS (Rodriguez et al, 2012), raising the possibility that for siblings the most prominent stressors may also be unique. No research has sought to identify the events that siblings specifically experience as traumatic.

The integrative trajectory model highlights that an individual's subjective appraisals of cancer-related events, or how they cognitively process events, also influence their PTSS trajectory (Price et al., 2016). Cognitive-behavioral models of traumatic stress (Ehlers & Clark, 2000) suggest that post-traumatic stress symptoms (PTSS) develop when individuals cognitively process an event in a way that results in a sense of serious, current threat. Once established, PTSS are maintained through maladaptive behaviors (e.g., avoidance of reminders) which reduce distress in the short-term but maintain symptoms over time. Alternatively, repeated exposure to reminders can facilitate more adaptive cognitive processing of the event and reduce PTSS over time (Ehlers & Clark, 2000). Applying this theory to siblings of children with cancer, siblings likely experience PTSS in response to specific cancer-related experiences (e.g., seeing patient look sick or lose their hair) that they process as a serious, current threat (e.g., patient will die), which

may or may not align with the patient's prognosis. Siblings' PTSS may then be maintained if siblings are not exposed to cancer-related reminders over time and given the opportunity to process their experiences.

The nature and extent of siblings' involvement in treatment likely influences their overall exposure to cancer-related reminders and opportunities to process cancer-related experiences. In line with this hypothesis, one study found that children who were prepared before visiting an ill family member and had opportunities to process their experiences demonstrated improvements in emotional functioning (Nicholson et al., 1993). In a similar way, siblings who are more frequently involved in treatment and who have more opportunities to process treatment-related experiences may demonstrate reduced PTSS over time.

Coping Frameworks

Siblings' involvement in treatment likely influences not only their exposure to cancer-related reminders but also their cancer-related coping. Coping refers to the cognitive and behavioral strategies that an individual uses to manage or tolerate stress, either by changing the stressor or their emotional reaction to the stressor (Lazarus & Folkman, 1984). Research with children identifies eight primary coping strategies, which are classified as either engagement (problem-solving, cognitive restructuring, emotional expression, social support) or disengagement (problem avoidance, wishful thinking, self-criticism, and social withdrawal; Tobin et al., 1991). Substantial evidence suggests that engagement coping strategies are associated with more positive psychosocial outcomes, whereas disengagement strategies are associated with more negative psychosocial

outcomes (Zimmer-Gembeck & Skinner, 2016).

For siblings of children with cancer, the nature and extent of their involvement in treatment may hinder or support their use of certain coping strategies. Specifically, siblings with limited involvement in treatment may primarily rely on disengagement coping strategies, whereas siblings with increased quantity and depth of involvement may have more opportunities to use engagement coping strategies. While minimal research has specifically examined siblings' cancer-related coping strategies, siblings report emotions (e.g., isolation, guilt; Long et al., 2018) that may reflect disengagement coping (i.e. social withdrawal, self-criticism), endorse trying to avoid thinking about cancer (Kaplan et al., 2013), have more difficulty communicating emotions than peers (Mishra et al., 2012), and express feeling like they need to cope with cancer without parental support (Pariseau et al., 2020). Increased quantity and depth of treatment involvement may be associated with increased opportunities for siblings to access social support from family members and medical providers, express cancer-related emotions, enact problem-solving strategies (e.g., provide emotional support to patient), and as previously discussed, cognitively process cancer-related experiences so that experiences can be interpreted as less threatening.

In sum, no research to date has documented the variable ways in which siblings are involved in cancer treatment across settings, or how siblings' involvement has been influenced by the COVID-19 pandemic. A thorough conceptualization of sibling treatment involvement necessitates the consideration of interrelated care management, family relationship, and sibling psychosocial factors. Sibling treatment involvement

represents an important gap in the literature, as it may play a key role in determining sibling adjustment to cancer. Specifically, elevated PTSS in siblings may be related to siblings' uniquely limited involvement in cancer treatment (as compared to patients and parents). Siblings with greater quantity and depth of involvement in treatment may have more opportunities to a) process cancer-related experiences and b) utilize engagement coping strategies, both mechanisms expected to support siblings' psychosocial adjustment.

Current Research

In sum, siblings of children with cancer often experience emotional distress following the patient's cancer diagnosis (Long et al., 2018), and a majority experience PTSS. Research is needed on the specific aspects of siblings' cancer experiences that influence their adjustment to cancer and development and/or maintenance of PTSS. The seemingly higher prevalence of PTSS in siblings (as compared to patients and parents) may be related to siblings' unique experience of cancer. Siblings' initial stress responses to cancer-related experiences may be maintained if siblings are not exposed to cancer-related reminders over time and given the opportunity to process their experiences. In this way, the extent to which siblings are involved in cancer treatment (i.e., ongoing exposure to cancer-reminders) and how they are involved in treatment (i.e., opportunities to process their experiences and use engagement coping strategies) may influence their adjustment to cancer and PTSS trajectories. As a first step, it is important to document the nature of siblings' involvement in treatment and the potential relationship between treatment involvement and psychosocial functioning, in order to refine theories of sibling

risk and resilience, and adapt clinical recommendations to promote sibling resiliency. The current exploratory research project aims to address these gaps in the field by a) characterizing the nature of siblings' involvement in treatment, b) investigating how sibling PTSS relates to treatment involvement, sibling demographics, and cancer characteristics and c) characterizing the social-ecological impact of the COVID-19 pandemic on siblings of youth with cancer.

Primary Aims

Aim 1: Characterize sibling treatment involvement. Through qualitative interviews with siblings and parents of children with cancer, I will characterize the variable ways in which siblings help or participate in cancer treatment across the hospital, outpatient clinic and home settings (e.g., assist with medical treatment, provide support, communicate with health professionals), as well as the influence of siblings' treatment involvement on their psychosocial functioning and family relationships.

Aim 2: Explore the relationship between siblings' treatment involvement and cancer-related post-traumatic stress symptoms. First, I will qualitatively characterize the cancer-related experiences that are potentially traumatic for siblings. Next, I will quantitatively examine how sibling demographics and cancer characteristics relate to siblings' cancer-related appraisals and coping strategies. I will also quantitatively examine how sibling demographics, cancer characteristics, cancer-related appraisals and coping strategies relate to siblings' PTSS. Finally, using mixed methods analyses, I will explore the relationships between siblings' and parents' qualitative descriptions of siblings' involvement in cancer treatment and siblings' quantitative reports of PTSS.

Aim 3: Explore the impact of the COVID-19 pandemic on siblings of youth with cancer. I will use a social-ecological approach to characterize the impact of the COVID-19 pandemic on siblings of youth with cancer across family, school, community and healthcare settings.

Chapter 2: Siblings' Involvement in Cancer Treatment and Psychosocial

Adjustment to Cancer

In the United States, approximately 1 in every 285 children are diagnosed with cancer before the age of 20 (Ward et al., 2014). With treatment advances, average 5-year survival rates have increased to over 80% (American Cancer Society, 2014), some patients require shorter inpatient hospitalizations, and there is a growing emphasis on treatment management within outpatient clinics and home settings (Branowicki et al., 2016). Even in the context of improving survival rates, cancer treatment is a life-changing experience for the entire family, including siblings (Long & Marsland, 2011). Siblings of pediatric cancer patients often experience emotional distress (Long et al., 2018), and for some siblings, the psychosocial effects of cancer extend long after treatment has ended (Alderfer et al., 2003). However, some siblings also demonstrate resilience in response to cancer and exhibit positive outcomes, such as improvements in maturity and responsibility (Long et al., 2015). Further research is necessary to understand the processes that influence siblings' adjustment to cancer.

The psychosocial standards of care in pediatric oncology recommend that siblings be involved in care (Gerhardt et al., 2015). Specifically, psychosocial professionals are advised to help siblings feel more engaged, to introduce siblings to the medical team when possible, and to support families in aligning siblings' involvement with their level of interest in involvement (Wiener et al., 2020). Although the psychosocial standards of care endorse the inclusion of siblings in cancer care, it remains unclear how siblings are actually involved in cancer treatment in practice and whether that involvement promotes

more positive adjustment to cancer. Although siblings' involvement in cancer treatment has not yet been systematically assessed, healthcare providers and parents report that siblings are routinely absent from clinical settings (Franklin et al., 2018, Long et al., 2017). Parents report that siblings often remain at home with extended family members or other caregivers during patients' hospital stays while parent(s) are at the hospital with the patient, and that siblings seldom attend clinic visits, which often occur during the school day (Long et al., 2017). Although these findings suggest that siblings as a whole demonstrate more limited treatment involvement in hospital and clinic settings, individual siblings likely vary in the extent to which they are present and participate in cancer treatment, and siblings may be involved in treatment in other ways (e.g., caring for the patient at home).

Research has yet to propose a conceptual framework of the construct or to establish valid and reliable measures thereof. Research on other family members' involvement in treatment can be used to inform factors that may be relevant to siblings' involvement. Among pediatric chronic illness populations, a larger body of research exists on patient and primary caregivers' (generally mothers') involvement in treatment, and recently paternal involvement has received increased attention (Taylor et al., 2020). Broadly, findings (e.g., Crespo et al., 2016; Phillips-Salimi et al., 2011; Taylor et al., 2020) suggest that when conceptualizing treatment involvement, it is important to consider not only the practical or logistical ways that one helps or participates in care (e.g., delivering medical treatment, providing support), but also the reciprocal influence of one's involvement in treatment on relational factors (e.g., family relationships) and

one's own psychosocial functioning (e.g., cancer-related stress).

Within pediatric cancer and related populations, the limited research on siblings' treatment involvement preferences and caring responsibilities suggest that siblings' involvement in treatment likely also encompasses interrelated care management, relational, and sibling psychosocial factors (Kelada, 2021; Lövgren et al., 2016; Patterson et al., 2011; Wilkins & Woodgate, 2007). Siblings of youth with chronic illnesses commonly report engaging in caring responsibilities, such as looking after patients or reminding patients to take their medications, and siblings who are engaged in caring responsibilities report lower anxiety and more positive relationships with patients than siblings not engaged in caring responsibilities (Kelada, 2021). These findings support the rationale that siblings of youth with cancer are involved in treatment through caring responsibilities and that their involvement likely influences their psychosocial functioning and family relationships; however, siblings' specific caring responsibilities across hospital, clinic and home settings remain unknown. Additionally, further research is needed to understand the likely nuanced relationship between siblings' involvement in cancer treatment and psychosocial functioning. Siblings of youth with cancer report a desire to support patients (Wilkins & Woodgate, 2007) and help with treatment (Patterson et al., 2011); however, they also express needing to "take a break" from cancer (Patterson et al., 2011), suggesting that sustained involvement may be experienced by some siblings as overwhelming. In addition to caring responsibilities, siblings' involvement in treatment likely includes other components, such as learning about cancer and communicating with medical providers. Siblings frequently indicate uncertainty

related to the patient's cancer diagnosis or treatment (Jenholt Nolbris & Ahlström, 2014), and retrospectively, siblings express that increased involvement in cancer treatment and opportunities to learn about cancer from medical providers would have been helpful for their psychosocial adjustment (Lövgren et al., 2016).

In sum, psychosocial standards of care state that it is important to involve siblings in cancer treatment (Gerhardt et al., 2015), yet the limited research on siblings' involvement suggests that siblings are routinely absent from clinical settings. To date, no research has documented the specific ways that siblings are involved or participate in cancer treatment across hospital, clinic, and home settings or how siblings' involvement in cancer treatment may influence their family relationships and psychosocial functioning. Through qualitative interviews with siblings and parents of pediatric cancer patients, the present study sought to address these gaps in the literature by characterizing: (1) the specific ways that siblings are involved or participate in cancer treatment within hospital, clinic, and home settings, (2) the influence of siblings' involvement in treatment on their individual and interpersonal psychosocial functioning.

Method

Participants

Individual interviews were conducted with 38 participants, including 22 siblings and 16 parents from 16 families of youth with cancer. Eligible families had a youth who was diagnosed with cancer between three months and five years prior to study enrollment, who was under the age of 18 at the time of diagnosis, and who received cancer treatment in the United States (subsequently referred to as the "patient"), and at

least one other youth (i.e., a sibling) who was eligible to participate. Siblings were eligible to participate if they were living with the patient during some or all of cancer treatment, were under the age of 18 at the time of cancer diagnosis, and were between the ages of 8 and 18 at the time of study participation. No more than two siblings per family participated. All participants spoke English fluently. Demographic and cancer-specific characteristics of the sample are summarized in Table 1.

Recruitment

Information about the research study was distributed to potentially eligible families using a number of recruitment methods. First, participants were recruited by re-contacting families who participated in a previous study within the PI's research lab, provided written consent to be contacted for future studies, and met eligibility criteria for the current study. Second, information about the study was distributed through two pediatric cancer hospitals. At one hospital, the study recruitment flyer was hung in the clinic waiting room. At the other hospital, psychosocial providers shared study information with potentially eligible families by email. Finally, information about the study was distributed via social media (i.e., Twitter and Facebook). The recruitment materials directed interested families to contact the PI or visit the study webpage for more information. The study webpage contained information about the research study, the PI's contact information, and a contact field, where families could enter their contact information to request more information.

Regarding the families who had participated in past research, the PI contacted eight families. Three were enrolled, one expressed interest in participating but was lost to

follow up, and four did not respond. Regarding the families recruited through pediatric cancer hospitals and social media, 27 families expressed interest in participating by contacting the PI by email/phone or entering their information into the webpage contact field. The PI made outreach to interested families by phone to explain study procedures and ask screening questions to determine eligibility. Seven families were unable to be reached, five families who heard about the research study on social media were determined ineligible during screening (two due to the patient's age, two due to time since cancer diagnosis, and one due to cancer treatment not occurring within the United States), two expressed interest in participating but were lost to follow up, and 13 were enrolled. The final sample (22 siblings and 16 parents from 16 families) included three families recruited from past research studies, four families recruited from pediatric cancer hospitals, and nine families recruited via social media and/or word of mouth. Enrollment continued until saturation was reached on all primary research questions, meaning that participants continued to be interviewed until the themes that emerged were fully explored (Morse, 1995).

Procedure

Informed consent and assent were obtained for all families. First, parents completed an online background form about family demographics and cancer characteristics using REDCap. Next, parents and siblings completed remote qualitative interviews. Parents and siblings were interviewed separately using Zoom. Interviews lasted approximately 60 minutes and were conducted by the PI, a psychology doctoral student with intensive training in qualitative interviewing methods and pediatric cancer.

Parents and siblings each received a \$25 gift card as a token of appreciation for participation. All study procedures were approved by the Boston University Institutional Review Board.

A detailed semi-structured interview guide, with parallel sibling and parent versions, was created for this study. Interviews began with introductory questions about the family and cancer experience, and then transitioned to questions about siblings' involvement in cancer treatment within the hospital, outpatient clinic, and home settings. Siblings and parents were asked questions about the quantity of siblings' involvement within each setting (e.g., "How many times did [sibling] visit [patient] in the hospital?"), how siblings were involved (e.g., "Tell me about what you would do at the clinic" "How well did [sibling] know [patient]'s doctors?"), siblings' family relationships (e.g., "How did [sibling] and [patient] get along in the hospital?") and siblings' reactions to involvement (e.g., "How did you feel about helping with [patient]'s treatment at home?").

Data Analysis

Interviews were recorded, transcribed verbatim, and cleaned. Interview transcripts were entered into NVivo 12 (QSR, 2018). An initial qualitative coding structure was developed based on *a priori* research questions and interview probes. The coding structure was then iteratively updated to include themes that emerged from the interviews.

The coding structure included each code's name and instructions regarding when it should be applied. The coding team included the PI (EP) and another psychology doctoral student with research experience in applied thematic analysis and pediatric

cancer (PB), with close oversight from a psychologist with expertise in qualitative methods and siblings of youth with cancer (KL). All members of the coding team (EP, PB, and KL) are trained as clinical psychologists, and have experience providing evidence-based treatment, such as Cognitive Behavioral Therapy and Parent-Child Interaction Therapy, to youth and families. EP and KL have clinical experience working with families of youth with cancer, including patients, parents, and siblings. No members of the coding team have personal experience with pediatric cancer.

The first two interviews were coded together, as the coding structure was refined and finalized, and the final coding structure was then used to code all transcripts. Weekly meetings were held to discuss application of the coding structure and to resolve any discrepancies in applying the coding structure. Twenty percent of transcripts were double coded to ensure reliability across coders. Inter-coder concordance, calculated by dividing the number of concordant codes by the total number of codes assigned (Morse et al., 2002) was 94%. Consensus was reached on all codes prior to data analysis.

Transcripts were analyzed using applied thematic analysis methodology (Guest et al., 2012). For Aim 1, qualitative codes that were used to capture siblings' and parents' descriptions of siblings' involvement in cancer treatment were queried and stratified by setting (i.e. hospital, outpatient clinic, home). Data were reviewed, and a matrix method was used to organize data into themes based on commonalities in the experiences described. For example, descriptions of siblings receiving information about cancer and asking questions about cancer were grouped together, as they both relate to the theme "learning information about cancer." For Aim 2, qualitative codes that were used to

capture siblings' and parents' descriptions of siblings' psychosocial functioning and family relationships were queried. Data were reviewed, and organized into themes using a matrix method. For all three Aims, themes were reviewed and revised to ensure that all themes were adequately supported by the data and distinct.

Results

Themes emerged related to siblings' involvement in cancer treatment and the influence of treatment involvement on siblings' individual and interpersonal psychosocial adjustment to cancer (Figure 1). Regarding siblings' treatment involvement, the higher level theme is "Siblings Were Actively Involved in Learning About Cancer and Helping with Emotional and/or Cancer Management Aspects of Treatment Across the Cancer Trajectory," which is characterized by four sub-themes: (1) witnessing cancer symptoms and treatment, (2) learning information about cancer, (3) providing patient with emotional support, and (4) contributing to cancer management. Example quotations are presented in Tables 2–4.

Regarding the influence of siblings' treatment involvement on their individual and interpersonal psychosocial adjustment to cancer, the first higher level theme is "Siblings' Initial Cancer-Related Distress is Reduced Over Time with Ongoing Treatment Involvement," which is characterized by two sub-themes: (1) treatment involvement initially triggered cancer-related distress, and (2) treatment involvement over time reduced cancer-related distress and increased familiarity. The second higher level theme is "Treatment Involvement Enhanced Feelings of Inclusion and Family Relationships," which is characterized by two sub-themes: (1) treatment involvement increased feelings

of inclusion, and (2) treatment involvement enhanced closeness of family relationships.

Example quotations are presented in Table 5.

Siblings Were Actively Involved in Learning About Cancer and Helping with Emotional and/or Cancer Management Aspects of Treatment Across the Cancer Trajectory

Witnessing Cancer Symptoms and Treatment

As patients progressed through cancer treatment, siblings were exposed (either directly or indirectly) to patients' treatment settings, cancer-related symptoms, medical care, and providers. Many families described how *witnessing* these aspects of treatment was an important component of siblings' involvement.

Hospital. Witnessing the inside of a pediatric hospital was often a novel experience for siblings. Siblings described the sensory-intensity of initial hospital visits, such as hearing machines beeping, seeing teams of providers quickly moving from room to room, and witnessing other hospitalized children. Many siblings saw the patient for the first time since they had been diagnosed with cancer. Families described that it was an adjustment for siblings to witness patients in hospital beds, often connected to machines and exhibiting physical indicators of cancer. Siblings also often experienced their first exposure to patients' cancer-related medical care. Some siblings were routinely present for aspects of patients' medical care and would sit in the hospital room while patients received medicine, blood transfusions, shots, or had vitals taken. Other siblings did not as frequently witness the patient's cancer-related care, related to factors such as the timing of siblings' visits to the hospital, parents intentionally "shielding" siblings from

witnessing procedures, siblings personally deciding to step outside of the room during aspects of care, or medical providers instructing siblings to leave the room. Relatedly, siblings generally interacted with members of the patient's medical team. Some siblings described pleasant interactions with medical providers, such as doctors saying "hello" to them, or nurses talking to them about shared interests. Alternatively, other families identified aspects of the hospital environment that could hinder siblings' ability to become familiar with providers, such as providers rounding early in the morning when siblings were not present, appearing rushed in their interactions with families, and frequently rotating.

Clinic. As compared to the acute hospital setting, families described outpatient clinics and care as more "routine." Some families highlighted that while the primary purpose of siblings' hospital visits had been to see the *patient*, the primary purpose of siblings going to clinic was for siblings to see and gain familiarity with the patient's routine care. Patients' health tended to be better, and siblings commented that seeing patients' progress helped them to feel more hopeful. Relatedly, some siblings felt reassured in observing the other patients and families, and seeing that other patients are surviving cancer. Some siblings were present to observe aspects of patients' medical care, such as patients receiving chemotherapy or having ports accessed, whereas other siblings tended to stay in the waiting room during these procedures. Families reported that logistical aspects of the outpatient care setting could interfere with siblings' ability to go to clinic and witness aspects of care. For example, parents reported that without private space (offered by patients' hospital rooms), it could be challenging to supervise siblings

while simultaneously caring for the patient. Additionally, patients' clinic appointments would at times be multiple hours long, which hindered families' ability and interest in having siblings join.

Home. When families transitioned to home treatment, siblings often experienced multiple changes to their home environments. Areas within families' homes were adjusted to address treatment-related needs, such as converting guest rooms into nursing rooms to store treatment-related supplies, and siblings often commented on the sudden increase in cancer-related reminders within their day-to-day life (e.g., toxic signs on medications, increased exposure to conversations about cancer). Families either had a visiting nurse come to the home or one of the parents took on the primary role of managing the patient's treatment. Many families also described that patients tended to experience their most apparent cancer-related symptoms at home, and that siblings had increased exposure to these symptoms by nature of living in the same home with patients and sharing certain living spaces, such as bathrooms.

Learning Information about Cancer

Hospital. Within the hospital, many families described that siblings met with child life specialists who provided siblings with opportunities to learn about cancer in developmentally appropriate ways. Child life specialists helped siblings understand the patient's cancer diagnosis and what treatment would entail through child-friendly teaching materials such as puppets, stuffed animals, and other medical play toys. Siblings also described learning about cancer within the hospital through asking parents questions. Siblings less routinely described learning about cancer from medical providers in the

hospital, sometimes noting that they did not ask doctors or nurses questions in this setting because they did not want to bother providers who appeared busy seeing many patients.

Clinic. Within the outpatient clinic, siblings reported that because care was less fast-paced than the hospital, they felt more willing and able to talk to medical providers and ask them questions. Some siblings also met with psychosocial providers, such as child life specialists or social workers within the clinic, to whom they were able to ask questions; however, these interactions appeared to be more family- or sibling-initiated and less routinely accessible (as compared to within the hospital).

Home. During home treatment, siblings described learning information about the patient's treatment through family members, mainly parents. Some siblings received regular updates from parents about the patient's treatment schedule, such as when the patient would be taking steroid medication that impacted their mood.

Providing Patient with Emotional Support

Hospital. Siblings primarily helped with cancer treatment by providing patients with emotional support. Siblings spent the majority of their time during hospital visits with the patient, either in the patient's room or another location (e.g., resource room). Siblings' hospital visits allowed siblings and patients to spend time together, sometimes following a period of separation. Parents reported that siblings' visits positively impacted patients, who felt comforted and distracted by siblings' presence.

Clinic. Siblings' involvement was less motivated by having the chance to see patients, because siblings and patients were living together at home. However, when present in the clinic, siblings often continued to play a crucial role in providing patients

with emotional support and companionship. For example, siblings entertained patients while waiting to see doctors and provided patients with distraction and comfort during distressing medical procedures.

Home. Siblings continued to provide patients with emotional support at home. For example, siblings engaged in active listening and provided emotional support related to patients' negative cancer-related emotions (e.g., worries about prognosis, low self-esteem related physical indicators of cancer).

Contributing to Cancer Management

Hospital. There were minimal opportunities for siblings to contribute to patients' cancer management in the hospital. Some parents thought of ways for siblings to help with tasks peripherally related to treatment to help them feel included, such as asking siblings to run an errand for them in the hospital.

Clinic. Similarly, the majority of siblings were not involved tasks related to cancer management within the clinic. Some siblings who were old enough to drive took on the primary role of bringing patients to clinic appointments when their parents were not able to do so.

Home. Families emphasized that siblings helped with cancer management during the patient's home treatment. Some families reported that siblings took on active roles in helping with the patient's medication, such as helping to flush the patient's PICC line or to administer medication. Alternatively, other parents reported not feeling comfortable having siblings involved with the patient's medications, due to factors such as siblings' age or that medications were controlled substances. Families allowed younger siblings

who were interested in helping with treatment to assist in more minor roles, such as getting the space ready for the visiting nurse.

Siblings were also involved in caring for patients in other ways at home, such as supporting patients to complete activities of daily living. Siblings cooked healthy meals for patients and adjusted their own eating habits to fit with patients' new cancer-related diets. Siblings also took on leading roles in ensuring patients engaged in recommended physical activities, such as physical therapy exercises, walks, and yoga. Additionally, siblings thoughtfully adjusted their behavior to keep patients safe, such as regularly disinfecting things when patients were immunocompromised, or interacting with patients in a gentle way so as not to hurt them or disrupt aspects of treatment (e.g., PICC line, port).

Siblings' Initial Cancer-Related Distress is Reduced Over Time with Ongoing Involvement in Treatment

Treatment Involvement Initially Triggered Cancer-Related Distress

Many siblings experienced distress in response to witnessing patients' cancer-related symptoms and medical treatment. This was particularly the case during siblings' initial hospital visits, when siblings first observed patients in hospital beds and exhibiting physical indicators of cancer (e.g., weight loss, fatigue). Although distressing, some families described these experiences were simultaneously reassuring for siblings because seeing patients in-person helped affirmed for siblings that patients were alive in a way that may have not been possible otherwise.

Treatment Involvement Over Time Reduced Cancer-Related Distress and Increased Familiarity

Although the majority of siblings experienced some level of distress in response to patients' cancer-related symptoms and medical care, siblings' distress tended to decrease over time with ongoing exposure. Siblings felt less distressed by cancer-related experiences as these experiences became increasingly familiar. Learning about cancer, and gaining a more comprehensive understanding of the things that they were witnessing also helped to reduce siblings' cancer-related distress.

Treatment Involvement Enhanced Feelings of Inclusion and Family Relationships

Treatment Involvement Increased Feelings of Inclusion

Siblings' visits to the hospital, attendance at clinic appointments, and opportunities to support patients' care at home helped siblings to feel more included in their families' cancer experience. Through these treatment-involvement experiences, siblings had opportunities to be together with their family, as well as participate in patients' treatment, which both impacted the extent to which siblings felt included in their families' adjustment to cancer.

Siblings' visits to the hospital were important as they allowed time for all family members to be together and reduced siblings' feelings of isolation. Families identified specific times that were especially important to be together, such as in the beginning of treatment when family members were initially adjusting to the cancer diagnosis, and during holidays. Family-centered activities and resources within the hospital and clinic helped siblings to feel "welcome," such as family pizza nights within the hospital that

siblings were encouraged to attend, and crafts for siblings and patients to do together within the clinic

Within the hospital and clinic, siblings' feelings of inclusion were also impacted by their interactions with medical providers. For some siblings, interacting with the medical team helped them to feel included and enhanced their trust in the patient's care. Siblings' feelings of inclusion and trust were facilitated by providers acknowledging siblings' presence, introducing themselves to siblings, allowing siblings to remain present for aspects of medical, and engaging siblings in conversation during "down time" (e.g., about siblings' school, hobbies). Occasionally, families reported unique circumstances in which siblings could be involved in patients' actual cancer treatment. For example, one family reported that when the patient received a stem cell transplant from one sibling, they decided to have the other sibling contribute by pressing the button to start the transplant. When families were able to involve siblings in these sorts of ways, families reported that the experiences were quite powerful and that siblings seemed to appreciate being included.

During home treatment, siblings spent more time with their family members and took on active roles in caring for patients, which facilitated feelings of inclusion.

Treatment Involvement Enhanced Closeness of Family Relationships

Hospital visits offered siblings the opportunity to see patients and provide them with emotional support. These visits were described as enhancing the closeness of sibling-patient relationships. Similarly, siblings reported that providing patients with emotional support and helping with cancer management at home helped them to feel

closer to patients.

Hospital visits also helped siblings feel closer with parents, particularly if there were brief opportunities for siblings to connect with parents one-on-one during these visits (e.g., going on a walk to get food).

Discussion

Aligned with family-centered care principles, the psychosocial standards of care in pediatric oncology recommend that siblings be involved in cancer care (Gerhardt et al., 2015). However, what it means in practice for siblings to be “involved” in cancer treatment has remained ambiguous, as has the influence of siblings’ treatment involvement on their psychosocial adjustment to cancer. The current study begins to address these research gaps by qualitatively characterizing siblings’ involvement in cancer treatment within the hospital, clinic, and home settings and describing how involvement may lead to improved psychosocial adjustment.

Findings show that siblings were actively involved in cancer treatment through learning about cancer and helping with emotional and/or cancer management aspects of treatment. Across hospital, clinic, and home settings, witnessing aspects of cancer treatment, such as the patient’s cancer treatment setting, symptoms and medical care, were important components of siblings’ involvement in treatment. Families described that seeing these aspects of treatment in-person helped facilitate siblings’ understanding of cancer. Although these experiences initially triggered negative cancer-related distress for many siblings, continued engagement in treatment (e.g., repeated visits to the hospital) led to reduced cancer-related distress and increased comfort over time.

Learning information about cancer emerged as another key element of siblings' involvement in cancer treatment. Past research shows that siblings frequently indicate uncertainty related to the patient's cancer diagnosis or treatment (Jenholt Nolbris & Ahlström, 2014), and retrospectively, siblings express that increased involvement in cancer treatment and opportunities to learn about cancer from medical providers would have been helpful for their psychosocial adjustment (Lövgren et al., 2016). Building on these findings, the present study highlights that within the hospital and outpatient clinic, siblings had opportunities to learn about cancer through receiving developmentally appropriate information from child life specialists and asking medical providers questions; these experiences enhanced siblings' understanding of cancer and reduced siblings' cancer-related distress. These findings provide additional evidence that opportunities to learn about cancer from providers has benefits for siblings' psychosocial adjustment.

Past research suggests that siblings of youth with cancer want to support patients (Wilkins & Woodgate, 2007) and help with treatment (Patterson et al., 2011). Aligned with these findings, the present study found that an integral way that siblings were involved in treatment was through providing patients with emotional support (across settings) and through taking on more active roles in cancer management at home. Quantitative research suggests that siblings of youth with chronic illness who are engaged in caregiving responsibilities reported lower anxiety and more positive relationships with patients (Kelada, 2021). The present study builds on these findings by qualitatively describing the processes through which siblings' involvement (including caregiving) may

influence their individual and interpersonal psychosocial adjustment. Specifically, ongoing involvement may facilitate lower anxiety and distress through repeated exposure to illness-related reminders (e.g., witnessing patient's symptoms) and opportunities to reappraise these experiences as less threatening. Furthermore, siblings' involvement in treatment, particularly their opportunities to provide patients with emotional support and contribute to cancer management, may enhance siblings' relationships with patients by allowing siblings to spend time with patients and while feeling helpful.

In addition to strengthening siblings' family relationships, involvement in treatment helps siblings to feel more included in their families' adjustment to cancer. Findings highlighted the importance of sibling-inclusive activities and resources within the hospital and clinic that allow siblings to feel "welcome." Additionally, siblings' feelings of inclusion were influenced by their interactions with medical providers. Siblings' feelings of inclusion were facilitated by providers saying "hello" to siblings, introducing themselves, allowing siblings to remain present for aspects of medical care, and engaging siblings in conversation during "down time."

Findings from the present study have important clinical implications. First, it is important for providers and families to be aware that siblings' cancer-related distress appears to reduce over time with ongoing involvement. Psychosocial providers can play an important role in providing families with psychoeducation related to exposure and supporting families to make informed decisions about how to involve siblings in treatment. Specifically, families should be aware that siblings' initial distress in-response to cancer-related experiences is normative, and that through ongoing involvement

siblings' distress will likely subside and they will become increasingly comfortable. Families should also be educated that limiting siblings' ongoing opportunities for involvement may likely perpetuate their distress.

Additionally, findings can be used to inform and support clinical recommendations related to engaging siblings' in cancer care. The psychosocial standards of care in pediatric oncology recommend that professionals working with siblings of youth with cancer introduce siblings to the medical team to help siblings feel engaged in treatment (Gerhardt et al., 2015; Wiener et al., 2020). Findings from the present study provide empirical support for this recommendation. Even in the fast-paced setting of the hospital, medical providers are encouraged to acknowledge siblings and briefly introduce themselves to siblings, as these quick strategies facilitate siblings' feelings of inclusion as well as their trust in the patient's medical care.

Strengths of the current study include the consideration of both sibling and parent perspectives, the use of rigorous qualitative methods, and the enrollment of a diverse sample relative to race, ethnicity, sibling age and gender, geographic region, and time since diagnosis. However, findings must be interpreted in the context of the study limitations. While we enrolled families of patients diagnosed with multiple cancer types, the majority of participants were families of youth with leukemia/lymphoma. Future, larger scale research should be conducted to explore how siblings' involvement in treatment may vary based on the patient's specific cancer diagnosis and treatment protocol. Research should also explore the current findings with larger, more diverse samples to understand the influence of cultural and family diversity on siblings'

involvement in treatment.

Given current retrospective, qualitative findings that cancer-related distress appears to reduce over time with ongoing involvement, future research should confirm current findings with longitudinal quantitative work and explore the potential relationship between siblings' involvement in cancer treatment and cancer-related post-traumatic stress symptoms (PTSS). If ongoing involvement in treatment reduces cancer-related distress over time, it is possible that limited involvement may perpetuate siblings' distress. This may be a particularly important potential risk factor to understand within the context of siblings' recent restriction from healthcare settings related to the COVID-19 pandemic (Sullivan et al., 2020).

Additionally, future research should explore how specific characteristics of siblings' treatment involvement influence psychosocial adjustment, such as quantity of involvement (e.g., number of hospital visits, length of hospital visits), duration of involvement, and the extent to which involvement is voluntary and matched to the siblings' developmental abilities and needs. Previous research on siblings' caregiving more broadly suggests that these factors can influence siblings' subjective experience of caregiving as well as the impact of psychosocial adjustment health (East, 2010), and the same may be true for siblings' treatment involvement.

In sum, siblings were actively involved in cancer treatment across the cancer trajectory through learning about cancer and helping with emotional and/or cancer management aspects of treatment. Siblings' going involvement in treatment appears to reduce cancer-related distress, increase siblings' feelings of inclusion, and enhance

family relationships. Implementation of clinical recommendations that support siblings' ongoing involvement in cancer treatment is expected to foster siblings' more positive adjustment to cancer.

Chapter 3: Involvement in Cancer Treatment and Cancer-Related Post-Traumatic Stress Symptoms in Siblings of Youth with Cancer

Pediatric cancer treatment has a strong emotional impact on the patient and their family members, including patients' siblings (Long & Marsland, 2011). Cancer treatment is often characterized by potentially traumatic experiences for the patient (e.g., threat to life, frightening procedures) and their family members, who worry the patient may die (Alderfer et al., 2003; Goldenberg Libov et al., 2002). In response, some patients and family members develop cancer-related post-traumatic stress symptoms (PTSS), including heightened arousal, re-experiencing, and avoidance of cancer-related experiences and reminders (Kazak et al., 2017). These symptoms are normative in the short-term (e.g., first month after diagnosis), and may even serve adaptive purposes; for example, periodic avoidance can be helpful in managing distress (Kazak et al., 2006). However, extended unresolved PTSS can create substantial distress, impair daily functioning, and develop into serious comorbid psychological disorders (Goenjian et al., 2005).

Although the vast majority of the research on cancer-related PTSS has focused on patients and their parents, the more limited sibling-focused research suggests that a majority of siblings experience PTSS (Kaplan et al., 2013). In the years following diagnosis, up to two-thirds of siblings endorse moderate to severe PTSS (Kaplan et al., 2013). Although not directly compared, these estimates are approximately 2.5 to 5 times higher than those of patients (across studies, 12-25% of patients endorse moderate to severe symptoms; Barakat et al., 1997; Butler et al., 1996; Kazak et al., 2004; Stuber et

al., 1996) and 1.5 to 4 times higher than those of parents (across studies, 15-44% of parents endorse moderate to severe symptoms; Barakat et al., 1997; Brown et al., 2003; Kazak et al., 2004; Landolt et al., 2003). Siblings' PTSS include increased arousal and irritability, intrusive thoughts about cancer, and attempts to avoid thoughts and feelings related to cancer (Alderfer et al., 2003; Kaplan et al., 2013), all of which appear to interfere with siblings' overall functioning (Kaplan et al., 2013). Furthermore, approximately one-third of siblings demonstrate persistent PTSS, reporting moderate to severe symptoms five years (on average) after cancer treatment has ended (Alderfer et al., 2003).

Despite emerging findings on the high prevalence of PTSS in siblings, research has yet to identify the cancer-related experiences that siblings specifically find traumatic. A study done with patients, parents, and siblings reported that potentially traumatic experiences included receiving the cancer diagnosis, being at the hospital, going through procedures, enduring medical emergencies, experiencing symptoms and physical changes (first- or secondhand), and communicating with providers (Christofferson et al., 2020). However, findings were reported for the sample as a whole, and it remains unknown how potentially-traumatic events for siblings (17% of the sample) may differ from those of other family members. To our knowledge, no research has sought to identify the events that siblings specifically experience as traumatic.

Additionally, little is known about the factors that influence siblings' risk for PTSS onset or duration. Preliminary longitudinal research (Alderfer et al., 2020) suggests that PTSS decrease over time for the majority of siblings. However, approximately one-

fourth of siblings display less favorable trajectories, and it remains unknown which factors distinguish these siblings from those with more resilient trajectories (Alderfer et al., 2020). The few existing studies report inconsistent findings on the relationship between PTSS and sibling demographic variables, such as age and gender (Alderfer et al., 2003; Kaplan et al., 2013; Long et al., 2013). Additionally, siblings' PTSS do not appear to be significantly related to cancer treatment characteristics, such as treatment intensity (Long et al., 2013).

The unclear relationship between siblings' PTSS and cancer treatment characteristics may be related to siblings' more variable involvement in treatment, as compared to patients and parents. Patients and at least one parent are generally present and involved in cancer-related medical events and treatments. However, the same is not true for siblings, possibly making siblings' involvement in treatment a better predictor of psychosocial adjustment than the specific aspects of cancer treatment itself. As findings from Study 1 highlight (Pariseau et al., in prep), siblings who are routinely present in cancer treatment settings (i.e. hospital, clinic) witness patients' symptoms and medical care, interact with medical providers, and learn about cancer in child-friendly ways. Other siblings are largely absent from cancer treatment settings, potentially limiting their exposure to cancer-related events, as well as their access to adaptive coping resources.

Application of the transactional model of stress and coping (Lazarus & Folkman; 1984) to siblings' experiences during cancer treatment may elucidate the processes that influence siblings' PTSS, as well as how these processes may be shaped by siblings' involvement in treatment. This model posits that individuals' stress responses are

influenced by their appraisals of stressors (i.e. whether there is a serious, current threat) as well as their determination of how to cope based on available options, and that both processes are influenced by personal and environmental factors (Ehlers & Clark, 2000; Lazarus & Folkman; 1984; Price et al., 2016). When applying this theory to siblings of youth with cancer, siblings' involvement in cancer treatment may be an important environmental factor to consider, given the potential impact on siblings' appraisals. Siblings likely develop PTSS in response to cancer-related experiences (e.g., seeing patient lose their hair) that they appraise as a serious, current threat (i.e., thinking patient's life is in danger), which may or may not align with the patient's prognosis. Study 1 offered preliminary evidence that siblings' involvement in cancer treatment may influence their cancer-related appraisals (Pariseau et al., in prep). For some siblings, seeing patients in the hospital and gaining familiarity with cancer treatment helped them feel reassured, possibly suggesting that through ongoing involvement in cancer treatment these siblings appraised cancer experiences as less threatening. However, involvement in cancer treatment also heightened feelings of stress for some siblings. For example, some siblings felt more concerned about the patient's health after seeing them appear sick in the hospital. These examples highlight that siblings' involvement in cancer treatment may influence siblings' cancer-related appraisals in multiple ways, and further research is needed to understand these processes.

Additionally, the nature and extent of siblings' involvement in treatment may hinder or support their use of certain coping strategies. Coping refers to the cognitive and behavioral strategies that individuals use to manage or tolerate stress, either by changing

the stressor or their emotional reaction to the stressor (Lazarus & Folkman, 1984). Disengagement coping strategies, such as problem avoidance, wishful thinking, self-criticism, and social withdrawal, are generally associated with more negative psychosocial outcomes than engagement strategies, such as problem solving, cognitive restructuring, emotional expression, social support (Tobin et al., 1991; Zimmer-Gembeck & Skinner, 2016). PTSS are maintained through disengagement strategies, which reduce distress in the short-term but maintain symptoms over time. Alternatively, engagement coping strategies and repeated exposure to reminders can facilitate re-appraisal of stressors as less threatening and reduce PTSS over time (Ehlers & Clark, 2000). Past research suggests that siblings of youth with cancer use disengagement coping strategies, such as trying to avoid thinking about cancer (Kaplan et al., 2013). The cancer experience may also pose unique barriers to siblings' use of engagement coping. For example, siblings have more difficulty communicating emotions than peers (Mishra et al., 2012), and express feeling like they need to cope with cancer without parental support (Pariseau et al., 2020). However, research has yet to explore how siblings' coping strategies may be influenced by their involvement in cancer treatment. Siblings who are more involved in treatment may have more opportunities to use engagement coping strategies. For example, when siblings are present at the hospital or clinic they may have increased opportunities to access social support from family members and medical providers.

In sum, although a majority of siblings endorse cancer-related PTSS (Kaplan et al., 2013), research has yet to identify the cancer-related experiences that siblings specifically find traumatic, and further research is needed to understand the factors that

influence siblings' risk and resilience relative to PTSS. Involvement in cancer treatment may influence siblings' PTSS trajectories, as increased involvement may support siblings' ability to (a) appraise cancer-related experiences as less threatening and (b) utilize engagement coping strategies. Using the transactional model of stress and coping (Lazarus & Folkman; 1984) as a guiding theoretical framework, the present study begins to address the current gaps in the literature by characterizing siblings' cancer-related traumatic experiences and investigating how siblings' involvement in cancer treatment may influence PTSS. Specifically, the present study aimed to (Aim 1) qualitatively characterize cancer-related experiences that are potentially traumatic for siblings and (Aim 2a) quantitatively explore how siblings' demographic and cancer characteristics relate to siblings' cancer-related appraisals and coping strategies. Given the exploratory nature of these aims, no hypotheses are offered. Additionally, we aimed to (Aim 2b) quantitatively explore how siblings' demographics, cancer characteristics, cancer-related appraisals and coping strategies relate to siblings' PTSS. We hypothesized that PTSS would be higher in siblings who endorsed life-threat appraisals (i.e. thought patient's life was in danger) and who used more disengagement coping. Finally, (Aim 3) we used mixed methods analyses to explore the relationships between siblings' and parents' qualitative descriptions of siblings' involvement in cancer treatment and siblings' quantitative reports of PTSS. We hypothesized that families of siblings with "low" (minimal or mild) quantitative PTSS scores would qualitatively describe more involvement in treatment (as evidenced by visits to the hospital/clinic) and more opportunities to enact engagement coping strategies, as compared to families of siblings

with “high” (moderate or severe) PTSS. Our focus on elucidating the processes that influence siblings’ PTSS is well suited to a mixed-methods approach, as this approach enabled us to measure well established constructs (i.e. coping strategies, PTSS) using quantitative measures, while qualitatively capturing siblings’ involvement in cancer treatment.

Method

Procedure

All research procedures were approved by the Boston University Institutional Review Board. Families of youth with cancer were eligible to participate if the cancer patient was under 18 years old at diagnosis, the diagnosis occurred 3 months to 5 years prior, cancer treatment occurred in the United States, and at least one sibling within the family was eligible to participate. Eligible siblings were 8-18 years old at the time of study participation and had lived with the patient some or all of the time during cancer treatment. All participants spoke English fluently.

A multi-method approach to recruitment was used. First, families who participated in a previous study within the PI’s research lab and provided written consent to be contacted for future studies were re-contacted if they met eligibility criteria for the current study. The PI made outreach to eight families who were identified in this manner; three were enrolled, one expressed interest in participating but was lost to follow up, and four did not respond.

Information about the study was also distributed through two pediatric cancer hospitals. At one site, a recruitment flyer was displayed in the clinic waiting room, and at

the other site, psychosocial providers distributed study information with potentially eligible families by email. Lastly, study information was circulated on Twitter and Facebook. Recruitment materials distributed through the hospitals and social media instructed interested families to either contact the PI directly or visit the study webpage, which contained study information and a contact field for families to enter their contact information if they preferred to be contacted by the PI. Twenty-seven families expressed interest in participating by contacting the PI or entering their information into the webpage contact field. Of those, seven were unable to be reached, five were determined ineligible during screening (two due to the patient's age, two due to time since cancer diagnosis, and one due to cancer treatment not occurring within the United States), two expressed interest in participating but were lost to follow up and 13 were enrolled.

The final sample (22 siblings and 16 parents from 16 families) included three families recruited from past research studies, four families recruited from pediatric cancer hospitals, and nine families recruited via social media and/or word of mouth. Enrollment continued until saturation was reached on all primary research questions, meaning that participants continued to be interviewed until the themes that emerged were fully explored (Morse, 1995).

Informed consent and assent were obtained for all families. Parents and siblings each first completed measures online using REDCap and then completed remote qualitative interviews. Parents and siblings were interviewed separately using Zoom. Interviews lasted approximately 60 minutes and were conducted by the PI, a psychology

doctoral student with intensive training in qualitative interviewing methods and pediatric cancer. Parents and siblings each received a \$25 gift card for participation.

Qualitative Interviews

Detailed semi-structured interview guides were created for this study. Interviews began with introductory questions about the family and cancer experience, and then transitioned to open-ended questions about siblings' involvement in cancer treatment within the hospital, outpatient clinic, and home setting (See Study 1 for more detailed description of complete interview guide). Particularly relevant to the current analyses, siblings and parents were asked about cancer-related experiences that were potentially traumatic for siblings; siblings' emotional and behavioral reactions to cancer-related experiences; siblings' cancer-related appraisals; siblings' cancer-related coping strategies; siblings' preparation for cancer-related experiences; how treatment involvement influenced siblings' cancer-related appraisals and coping strategies; siblings' treatment involvement preferences; family decisions related to treatment involvement; and changes in siblings' involvement across the treatment trajectory. Example interview questions and probes are presented in Table 6.

Quantitative Measures

Demographics and Cancer Characteristics. Parents and siblings completed a background form on which they provided information about their age, gender, race, and ethnicity. Parents also provided information about the patient's cancer diagnosis (i.e. leukemia/lymphoma, central nervous system (CNS) tumor, or solid tumor), time since

diagnosis, relapse status (i.e. relapse, no relapse), and treatment status (i.e. active treatment, maintenance treatment, off treatment).

Cancer-Related Primary Appraisals. Siblings' primary appraisals were assessed via two questions related to life threat (i.e. "When your brother or sister was diagnosed with cancer, did you think his or life was in danger?" and "When your brother or sister was receiving cancer treatment, did you think his or her life was in danger?"). Siblings answered yes or no. These items have been used in past research with siblings of youth with cancer (Kaplan et al., 2013).

Cancer-Related Coping Strategies. Siblings completed the Coping Strategies Inventory (CSI; Tobin, 1991), a 32-item self-report measure (ages 7+) that assesses coping thoughts and behaviors in response to a specific stressor. Instructions were worded so that siblings reported on cancer-related coping strategies. First, siblings described in writing the circumstances of a stressful cancer-related event. Next, they completed the CSI using a 5-point Likert scale for each question. The CSI measures eight primary subscales (problem-solving, cognitive restructuring, emotional expression, social support, problem avoidance, wishful thinking, self-criticism, and social withdrawal). The CSI is classified as a well-established measure with adequate test-retest stability, internal consistency, and predictive validity (Blout et al., 2008). In our sample, internal consistency was acceptable on the emotional expression (.75), problem avoidance (.70) and wishful thinking (.78) subscales; internal consistency was good on the problem-solving (.88), cognitive restructuring (.87), social support (.85) and social withdrawal (.84) subscales.

Cancer-Related PTSS. Siblings completed the Child PTSD Symptom Scale (CPSS-5-SR), a 27-item self-report measure (ages 8–18). As has been done in past research on siblings of youth with cancer (Alderfer et al., 2020), instructions were worded so that siblings reported on symptoms of traumatic stress related to cancer. Siblings indicated how often they experienced each symptom in the past month on a 5-point Likert-scale from 0 (not at all) to 4 (6 or more times a week/ almost always). Total scores correspond with symptom severity categories: minimal (0-10), mild (11-20), moderate (21-40), severe (41-60), and very severe (61-80). The CPSS-5-SR has good to excellent internal consistency, test–retest reliability, and discriminant validity (Foa et al., 2018). Internal consistency in our sample was .95.

Data Analysis

Qualitative (Aim 1)

Qualitative interviews were recorded, transcribed verbatim, and cleaned. Interview transcripts were entered into NVivo 12 (QSR, 2018). An initial qualitative coding structure was developed based on the transactional model of stress and coping and the study’s primary research focus (i.e. siblings’ involvement in cancer treatment and cancer-related PTSS). The coding structure was then iteratively updated to include themes that emerged from the interviews. The coding structure included each code’s name and instructions regarding when it should be applied. The coding team included the PI (EP) and another psychology doctoral student with research experience in applied thematic analysis and pediatric cancer (PB), with close oversight from a psychologist with expertise in qualitative methods and siblings of youth with cancer (KL). All

members of the coding team (EP, PB, and KL) are trained as clinical psychologists, and have experience providing evidence-based treatment, such as Cognitive Behavioral Therapy and Parent-Child Interaction Therapy, to youth and families. EP and KL have clinical experience working with families of youth with cancer, including patients, parents, and siblings. No members of the coding team have personal experience with pediatric cancer.

During coding, all members of the coding team were blind to siblings' scores on quantitative measures. The first two interviews were coded together, as the coding structure was refined and finalized, and the final coding structure was then used to code all transcripts. Weekly meetings were held to discuss application of the coding structure and to resolve any discrepancies in applying the coding structure. Twenty percent of transcripts were double coded to ensure reliability across coders. Inter-coder concordance, calculated by dividing the number of concordant codes by the total number of codes assigned (Morse et al., 2002), was 94%. Consensus was reached on all codes prior to data analysis.

Transcripts were analyzed using applied thematic analysis methodology (Guest et al., 2012). For Aim 1, we queried qualitative codes that were used to capture siblings' and parents' descriptions of the cancer-related experiences that were potentially traumatic for siblings. Data were reviewed and organized into themes based on common characteristics of the experiences described using a matrix method. For example, experiences related to patients' emesis and hair loss were grouped together, along with other experiences that shared the common characteristic of cancer-related symptoms,

resulting in the final theme “observing physical indicators of cancer.” Themes were reviewed and revised to ensure that all were adequately supported by the data and conceptually distinct from each other.

Quantitative (Aim 2)

Descriptive analyses were used to characterize the sample as a whole. The percentages of siblings who retrospectively endorsed life-threat (i.e., thinking the patient’s life was in danger) at diagnosis and during treatment were calculated. Means and standard deviations were calculated for the eight CSI sub-scales and the CPSS-5-SR. The percentages of siblings who endorsed minimal, mild, moderate, severe, and very severe cancer-related PTSS on the CPSS-5-SR were calculated.

For Aim 2a, exploratory quantitative analyses were conducted to identify factors associated with siblings’ cancer-related appraisals and coping strategies. Chi-square tests and independent samples t-tests were used to examine whether siblings’ cancer-related appraisals differed based on siblings’ demographics (i.e., gender, age at diagnosis) and cancer characteristics (i.e., patient cancer type, cancer relapse, treatment status, and time since diagnosis). Similarly, one-way ANOVAs, independent samples t-tests, and Pearson correlations were used to examine the associations between siblings’ demographics, cancer characteristics, and coping strategies (as measured by CSI sub-scale scores).

For Aim 2b, exploratory quantitative analyses were conducted to identify factors associated with siblings’ cancer-related PTSS. Specifically, Pearson correlations, independent samples t-tests, and one-way ANOVAs were used to examine how siblings’

demographics, cancer characteristics, cancer-related appraisals, and coping strategies relate to cancer-related PTSS (CPSS-5-SR total scores).

Importantly, the sample size of the present study (22 siblings) was too small to reliably detect small to medium effects for Aim 2a and 2b. For these analyses, statistical power ranged from 15% to 29% to detect moderately sized effects. Therefore, analyses are considered exploratory; null findings should be interpreted with caution and preliminary significant findings should be used to inform future research.

Mixed Methods (Aim 3)

Mixed methods analyses were used to explore the relationship between siblings' quantitative report of cancer-related PTSS (CPSS-5-SR) and siblings' and parents' qualitative descriptions of siblings' involvement in cancer treatment. Siblings who reported minimal or mild PTSS on the CPSS-5-SR were grouped together (i.e. "low PTSS") and siblings who reported moderate or severe PTSS were grouped together (i.e. "high PTSS"). No siblings within the present sample reported very severe PTSS. Qualitative data were analyzed using applied thematic analysis (Guest et al., 2012). Qualitative codes that were used to capture siblings' and parents' descriptions of siblings' involvement in cancer treatment were queried, and data was stratified by PTSS level (i.e. low PTSS vs. high PTSS). A matrix method was used to organize the data and to systematically compare similarities and differences between the PTSS groups. Qualitative themes were generated to characterize the differences between groups. Themes were reviewed and revised to ensure that all were adequately supported and distinct.

Results

Twenty-two siblings and 16 parents from 16 families completed the study. For sample as a whole, siblings were between the ages of 8 and 18 years ($M = 12.7$, $SD = 3.1$) at the time of study participation, and between the ages of 5 and 15 years at the time of cancer diagnosis ($M = 10.0$, $SD = 3.2$). Sixty-four percent were older siblings (relative to patient) and 36% were younger siblings. Fifty-five percent of siblings identified as female and 46% identified as male. Seventy-four percent of siblings identified as White and 30% identified as Black/African American (percentages reflect that one sibling identified as multi-racial). Fourteen percent of siblings identified as Hispanic/Latinx. Regarding cancer characteristics, 61% were siblings of patients with leukemia/lymphoma, 22% were siblings of patients with central nervous system tumors, and 17% were siblings of patients with solid tumors. Regarding treatment status, 23% were siblings of patients in active treatment, 41% were siblings of patients in maintenance treatment, and 36% were siblings of patients off treatment. Eighteen percent were siblings of patients that had relapsed. Time since cancer diagnosis ranged from 3 to 60 months ($M = 35.6$, $SD = 18.6$). Demographic and cancer-related characteristics of siblings with low PTSS and siblings with high PTSS are presented in Table 7.

Parent participants were predominantly female (88%) and married /partnered (88%). Nineteen percent of parents reported completing high school or less, 69% reported completing some or all of college, and 13% reported completing some or all of a master's or doctoral degree program.

Aim 1: Potentially Traumatic Cancer-Related Experiences

Siblings experienced negative emotions consistent with PTSS, such as fear and helplessness, in response to multiple cancer-related experiences. Potentially traumatic experiences fell into six categories: (1) learning diagnosis, (2) seeing patient in the hospital for the first time, (3) observing physical indicators of cancer (e.g., hair loss, weight loss, vomiting), (4) witnessing or hearing about aspects of patient's medical care (e.g., port accessed, surgeries), (5) witnessing or hearing about medical emergencies, and (6) seeing other pediatric cancer patients. Example quotations are provided in Table 8.

Aim 2: Factors Associated with Siblings' Cancer-Related Appraisals, Coping Strategies, and PTSS

Descriptive statistics regarding siblings' cancer-related appraisals, coping strategies, and PTSS for the sample as a whole are presented in Table 9.

Cancer-Related Appraisals

Retrospectively, 77% of siblings reported thinking the patient's life was in danger when they were diagnosed with cancer, and 41% of siblings reported thinking the patient's life was in danger when they were receiving cancer treatment. Appraisals of life threat did not differ based on siblings' age at diagnosis, gender, time since diagnosis, cancer type, treatment status, or cancer relapse status (Table 10).

Cancer-Related Coping Strategies

Associations between siblings' demographics, cancer characteristics, and coping strategies are presented in Table 11. Siblings' age at diagnosis was positively correlated with their use of problem avoidance and social withdrawal, but not related to other

strategies. Coping strategies did not differ based on siblings' gender. Patients' cancer type was related to siblings' use of problem avoidance. Specifically, siblings of patients with leukemia/lymphoma had higher problem avoidance scores ($M = 21.8, SD = 3.3$) than siblings of patients with solid tumors ($M = 12.7, SD = 2.5, p = .034$), but not significantly different scores than siblings of patients with CNS tumors ($M = 17.0, SD = 9.2$). Cancer type was not related to siblings' use of other coping strategies. Neither cancer treatment status nor time since cancer diagnosis were related with siblings' coping strategies.

Post-Traumatic Stress Symptoms

On the CPSS-5-SR ($M = 25.0, SD = 18.0$), approximately equivalent percentages of siblings reported minimal, mild, moderate, and severe levels of PTSS; no siblings in the sample reported very severe PTSS (Table 9). Associations between siblings' demographics, cancer characteristics, cancer-related appraisals, coping strategies and PTSS scores are presented in Table 12. PTSS was positively correlated with siblings' age at diagnosis. Mean PTSS scores did not vary by sibling gender, time since cancer diagnosis, cancer type, treatment status, or cancer relapse status. Siblings who thought the patient's life was in danger when they were diagnosed with cancer ($n = 17$) had significantly higher mean PTSS scores ($M = 29.1, SD = 18.3$) than siblings who did not think the patient's life was in danger when they were diagnosed with cancer ($n = 5, M = 11.0, SD = 6.0$). However, siblings who thought the patient's life was in danger when they were receiving cancer treatment ($n = 9$) did not have significantly different mean PTSS scores ($M = 30.1, SD = 16.3$) than siblings who did not think the patient's life was in danger when they were receiving cancer treatment ($n = 13, M = 21.4, SD = 18.8$).

PTSS was positively correlated with siblings' use of emotional expression, problem avoidance, wishful thinking, self-criticism, and social withdrawal, but not correlated with siblings' use of problem-solving, cognitive restructuring, or social support.

Aim 3: Differences in Siblings' Involvement in Treatment between Low PTSS and High PTSS Groups

Differences in siblings' involvement in cancer treatment emerged between the low PTSS and high PTSS groups related to four qualitative themes: consistency of siblings' treatment involvement, alignment between siblings' preferences and actual treatment involvement, access to developmentally appropriate cancer-related information, and opportunities to communicate cancer-related emotions. Example quotations are presented in Table 13.

Consistency of siblings' treatment involvement. Within both groups, siblings reported that their cancer-related distress decreased with ongoing involvement in treatment; however, siblings with high PTSS were less likely to be consistently involved in treatment over time than siblings with low PTSS. Siblings with high PTSS more often reported initially visiting the patient in the hospital followed by a period of restricted access to clinical settings. Some siblings were no longer able to go to the hospital or clinic due to system-wide visitation restrictions within these settings (i.e. related to flu season, COVID-19). In other families, parents limited siblings' ongoing involvement because they worried about the potential impact of these experiences on siblings' emotional functioning.

Alignment between siblings' preferred and actual treatment involvement.

Siblings with high PTSS were more likely than siblings with low PTSS to describe that their involvement in cancer treatment was in some way misaligned with their preferences related to involvement. Almost all siblings with high PTSS at some point in treatment were not allowed to go to the hospital or clinic as often as they would have liked to. Additionally, siblings with high PTSS were more likely to describe a lack of control over cancer-related information and reminders, such as ongoing updates via the family's online health journal even when they were at school, or parents not keeping them informed when they were unable to be present at the hospital or clinic. Within families of siblings with low PTSS, parents were more likely to ask siblings if they wanted to go to the hospital or clinic, and to adjust to siblings' preferences when possible.

Access to developmentally appropriate cancer-related information. Siblings with low PTSS tended to have access to more information than siblings with high PTSS, including preparatory information before cancer-related experiences, and opportunities to learn about cancer from parents and providers. Parents of siblings with low PTSS prepared siblings for cancer-related experiences by telling them what to expect (e.g., what the hospital was like, how the patient would look) and preemptively explaining things that might be frightening for siblings. Alternatively, parents of siblings with high PTSS shared less information with siblings, including less information before initial visits and less regular updates throughout treatment. Relatedly, families of siblings with high PTSS were more likely to describe that siblings' access to cancer-related information was at times hindered by parents' cancer-related knowledge. For example, some parents

described that when siblings expressed cancer-related worries, such as concerns that the patient's treatment was actually making them worse, it felt challenging to know how to respond to siblings' worries because parents did not thoroughly understand the details of the patient's treatment themselves. Siblings from both groups expressed that preparation before hospital visits was (or would have been) helpful for their psychosocial adjustment. Siblings with low PTSS felt more prepared for what they saw at the hospital. Alternatively, siblings with high PTSS tended to describe feelings more shocked and confused when they first visited the hospital.

Siblings with low PTSS also described having more opportunities to learn about cancer from medical and psychosocial providers within the hospital and clinic, as compared to siblings with high PTSS. Families of siblings with high PTSS reported less interaction between siblings and providers mainly related to siblings' less consistent presence within clinical settings. Some families also described barriers to siblings connecting with providers when siblings were present, such as parents discouraging siblings from asking providers question, either due to worries about "bothering" providers or about upsetting the patient who may overhear. Although siblings with high PTSS reported fewer opportunities to learn about cancer from providers, siblings from both groups appeared to benefit from these opportunities when they did occur. Siblings across groups reported that having the opportunity to learn about cancer from providers helped shift their appraisals so that they interpreted cancer-related experiences as less threatening. For example, siblings described that learning factual information about the cancer diagnosis and treatment (e.g., prognosis, survival rates) helped them to reassure

themselves that the patient was going to be alright.

Opportunities to express cancer-related emotions within a supportive context. Siblings with low PTSS had more supportive opportunities to express cancer-related emotions to family members than siblings with high PTSS. All siblings with low PTSS had at least one close relationship with a family member (e.g., patient, parent, another sibling) with whom they talked about cancer-related emotions. Siblings described connecting with family members and talking about cancer-related emotions within healthcare settings (e.g., hospital) as well as at home. Alternatively, siblings with high PTSS tended to feel more disconnected and less able to access social support from family members, either in the context of siblings remaining at home while other family members were at the hospital, and/or siblings feeling unable to share their negative cancer-related emotions with family members. Siblings within the high PTSS group were more likely to describe that their parents explicitly encouraged them to stay positive, and limited their ability to go to the hospital or clinic if they could not remain positive in those settings. Alternatively, parents of siblings with low PTSS were more likely to describe modeling emotional expression for siblings, actively encouraging siblings to share their negative emotions, and normalizing siblings' feelings.

Discussion

A majority of siblings of youth with cancer endorse cancer-related PTSS (Kaplan et al., 2013). The present study contributes to our understanding of siblings' PTSS by identifying the cancer-related experiences that siblings specifically find traumatic, as well as exploring factors that may influence siblings' risk and resilience relative to PTSS.

Building on past research documenting cancer-related experiences that are potentially traumatic for patients and their family members (Christofferson et al., 2020), the present study identified cancer-related experiences that are potentially traumatic specifically for siblings. These experiences included learning the diagnosis, seeing the patient in the hospital for the first time, observing physical indicators of cancer, witnessing or hearing about aspects of the patient's medical care, medical emergencies, and seeing other pediatric cancer patients. Findings suggest that many of the cancer-related experiences identified for the patient and broader family extend to siblings; however, there were also differences. New experiences identified as potentially traumatic for siblings included visiting the patient in the hospital for the first time, and seeing other pediatric cancer patients within clinical settings. Findings have clinical implications related to sibling psychosocial assessment and support. The Standards for Psychosocial Care of Children with Cancer and Their Families recommend that professionals and parents take steps to anticipate siblings' needs (Gerhardt et al., 2015). For this reason, it is essential that professionals and parents be aware of the specific cancer-related experiences that may be traumatic for siblings, as siblings may benefit from increased support surrounding these experiences (e.g., preparation beforehand, opportunities to express cancer-related emotions afterwards). Additionally, while past research suggests that communicating with providers can be potentially traumatic for patients and family members (Christofferson et al., 2020), siblings in the present sample did not report this, and in fact expressed that conversations with providers helped reduce cancer-related worries.

Using the transactional model of stress and coping (Lazarus & Folkman, 1984) as a guiding framework, the present study also builds upon previous research by quantitatively exploring how siblings' demographic characteristics relate to their cancer-related appraisals, coping strategies, and PTSS. While siblings' gender was not significantly related to appraisals, coping or PTSS, siblings' age at the time of cancer diagnosis was positively correlated with siblings' cancer-related avoidance, social withdrawal, and PTSS. Past studies report inconsistent findings on the relationship between PTSS and sibling age (Alderfer et al., 2003; Kaplan et al., 2013; Long et al., 2013), and present findings provide additional evidence that older siblings may be at increased risk for PTSS. Additionally, given that siblings' age was associated with not only PTSS but also siblings' disengagement coping strategies, findings may suggest that older siblings' PTSS relate to their use of cancer-related avoidance and social withdrawal. Older siblings may have more opportunities to use disengagement coping strategies within the context of their increased capacity for independence. For example, parents may feel more comfortable leaving older siblings at home when they go to the hospital, enabling older siblings to spend more time alone than their younger counterparts.

Present findings also contribute to the literature by exploring how cancer characteristics relate to siblings' cancer-related appraisals, coping strategies, and PTSS. No significant relationships were identified between cancer characteristics (i.e. cancer type, treatment status, relapse status, or time since cancer diagnosis) and siblings' cancer-related appraisals or PTSS; however, null findings may reflect insufficient power.

Analyses did identify a connection between cancer type and sibling coping. Specifically, siblings of patients with leukemia/lymphoma engaged in more cancer-related avoidance than siblings of patients with solid tumors. Given that observing physical indicators of cancer can be distressing for siblings, and that patients with leukemia/lymphoma tend to display more apparent cancer symptoms than solid tumor patients, one possible explanation of these findings is that siblings of leukemia/lymphoma patients are more motivated to avoid patients' distressing symptoms. Given that avoidance coping can maintain individuals' (initially normative) stress response and contribute to ongoing PTSS, future research with larger samples should explore whether siblings of patients with leukemia/lymphoma are at increased risk of unfavorable PTSS trajectories.

As hypothesized, siblings' life threat appraisals (i.e. thinking the patient's life was in danger when they were diagnosed) and disengagement coping strategies (i.e. avoidance, wishful thinking, self-criticism, and social withdrawal) were related to higher PTSS. Interestingly, higher PTSS was also related to higher emotional expression. Importantly, this CSI subscale captures forms of emotional release (e.g., "I was angry and really blew up"; "My feelings were overwhelming and they just exploded") rather than emotional communication, and many of the items may have captured the heightened arousal component of traumatic stress.

Also as hypothesized, siblings with low PTSS and high PTSS differed relative to how they were involved in cancer treatment. Although siblings from both groups reported that their cancer-related distress decreased with ongoing involvement in treatment, siblings with high PTSS has less consistent involvement in treatment. Specifically,

siblings with high PTSS more often reported initially visiting the patient in the hospital followed by a period of restricted access to clinical settings. These findings make sense conceptually when considered in the context of an exposure framework. Siblings with high PTSS were exposed to a potentially traumatic stressor (e.g., seeing patient in the hospital for the first time) and then, due to restricted access to clinical settings, had less ongoing exposure and opportunities to re-appraise cancer-related stressors as less threatening. These findings have important clinical implications. In some families, parents described restricting siblings' access to clinical settings related to well-intentioned efforts to shield siblings from ongoing emotional distress. Aligned with past research on parental awareness (Pariseau et al., 2020), these parents were often unaware that siblings wanted to be more involved and that siblings' cancer-related distress tended to decrease over time. Professionals that work with families of youth with cancer can address an important unmet need by providing parents with psychoeducation on the influence of disengagement vs. engagement coping strategies following a stressor (e.g., avoidance maintains stress response). Additionally, given that siblings with high PTSS were more likely to describe that their involvement in cancer treatment was in some way misaligned with their preferences, professionals can support parents of youth with cancer to communicate with siblings about their preferences. While families will likely not be able to entirely align siblings' involvement with their preferences, it is important to identify even small opportunities for siblings to make decisions. The experience of pediatric cancer can trigger feelings of helplessness (Long et al., 2018), and allowing siblings opportunities to regain control by deciding how to be involved in treatment may

have an influential impact on their psychosocial adjustment to cancer.

Differences also emerged between low PTSS and high PTSS groups relative to siblings' access to developmentally appropriate cancer-related information. Siblings with low PTSS tended to be better prepared for cancer-related experiences, as well as have more opportunities to learn about cancer from parents and providers. Importantly, families of siblings with high PTSS were more likely to describe that siblings' access to cancer-related information was at times hindered by parents' cancer-related knowledge. Professionals that work with families of youth with cancer should be aware that siblings benefit from being preemptively receiving information about what to expect during cancer experiences, particularly initial hospital visits. Additionally, siblings who have parents with less health literacy may benefit from additional opportunities to learn about cancer from providers.

Finally, the present study revealed that siblings with low PTSS had more supportive opportunities to express cancer-related emotions than siblings with high PTSS. These findings build upon past research identifying that siblings often receive messages from family members about the importance of remaining positive (Pariseau et al., 2017). Importantly, the present findings highlight that these well-intentioned encouragements to stay positive can hinder siblings' communication of negative cancer-related emotions and create a scenario in which siblings' inability to suppress negative emotions triggers self-criticism. Professionals can play an important role with families of youth with cancer by normalizing siblings' negative cancer-related emotions and encouraging all family members to engage in healthy communication of their emotions.

Additionally, professionals can support families to identify opportunities for siblings to share their emotions and to problem-solve around how parents can manage competing demands within clinical settings. For example, if parents are concerned that siblings' sharing emotions or asking questions will upset the patient, families can be supported to identify separate one-on-one times for siblings to ask questions and emotionally connect with parents or other trusted adults.

The findings of the current study need to be considered within the context of its limitations. First, the sample size of the present study was too small to reliably detect small to medium effects for Aim 2a and 2b. Therefore, null findings should be interpreted with caution and significant findings should be used to inform future larger scale research projects. An additional limitation of the present study is that while 22 siblings participated, multiple siblings (up to two) per family were enrolled. Future research should be conducted with larger samples with adequate power to account for nesting within families. Furthermore, given the cross-sectional nature of this study, assumptions cannot be made about the directionality of relationships identified among variables. Future longitudinal studies should investigate whether siblings' involvement in cancer treatment influences their development and maintenance of cancer-related PTSS, or rather that siblings with higher PTSS tend to be more reluctant to engage with treatment in the same way as siblings with lower PTSS. Additionally, while the sample included families of patients diagnosed with CNS and solid tumors, the majority were families of patients diagnosed leukemia/lymphoma. Future research should continue to explore how siblings' involvement in cancer treatment and PTSS may differ based on cancer diagnosis

and associated differences in treatment protocols. Larger research samples would also allow the important investigation of how siblings' involvement in cancer treatment may differ based on intersecting aspects of siblings' and families' cultural identities, including (but not limited to) race, ethnicity, spirituality/religiosity, acculturation (e.g., English language proficiency) and socioeconomic status.

Strengths of the current study include its mixed methods approach to identifying factors that relate to sibling PTSS. This approach enabled measurement of well characterized constructs (e.g., PTSS, coping) with quantitative measures, while also building upon quantitative findings with siblings' and parents' detailed qualitative descriptions of siblings' involvement in cancer treatment. Additionally, the current study incorporated both sibling and parent perspectives, an important methodological decision given parents' often limited awareness of siblings' psychosocial adjustment to cancer (Pariseau et al., 2020).

Overall, findings support the researchers' application of the transactional stress model to siblings' experiences during cancer treatment, and begin to characterize the processes that influence siblings' PTSS. Findings inform best practices in family-centered care for siblings of youth with cancer, by identifying specific cancer-related experiences that are potentially traumatic for siblings, and providing recommendations of how providers and families can involve siblings in cancer treatment in ways that promote resiliency.

Chapter 4: The Social-Ecological Impact of COVID-19 on Siblings of Youth with Cancer

The COVID-19 pandemic has profoundly impacted the mental health of youth and families in the United States and around the world (Feinberg et al., 2022; Samji et al., 2022). In addition to social isolation, major disruptions to daily life, and the loss of loved ones due to COVID-19, the pandemic has posed unique challenges for families of youth with cancer. These families have been faced with ensuring the safety of youth who are immunocompromised due to cancer treatment, while navigating an abruptly changing healthcare system (Sullivan et al., 2020). While parents of youth with cancer did not present with elevated distress during the first few months of the pandemic (van Gorp et al., 2021), longitudinal research documents increasing distress as the pandemic has continued (van Gorp et al., 2022), and additional research is needed to understand the likely complex ways in which these families have been impacted as the pandemic has endured. The limited research that has been done suggests that parents have worried about family members getting the virus and transmitting it to the youth with cancer, and that families have vigilantly followed health guidelines (Chardon et al., 2021; Darlington et al., 2021; Wimberly et al., 2021). Given that cancer treatment is often associated with strict health precautions and isolation, families of youth with cancer felt prepared for pandemic-related health precautions and isolation (Chardon et al., 2021; Darlington et al., 2021); however, those who had recently completed treatment also felt frustration related to the delays in resuming activities of daily life (Darlington et al., 2021).

Despite research beginning to explore how families of youth with cancer have

been impacted by the pandemic, this work has focused almost exclusively on the experiences of patients and parents, and the impact of the pandemic on siblings of youth with cancer remains poorly understood. Of the limited studies completed, most have not included siblings in their findings. To our knowledge, only two studies have reported findings related to siblings of youth with cancer (Davies et al., 2022; Chardon et al., 2021) and neither of these studies focused on siblings as a primary research aim. In one study (Davies et al., 2022), parents of youth with cancer reported that hospitals' pandemic-related visitation restrictions made it more challenging to see siblings during patients' hospitalizations. In the other, parents with a child undergoing pediatric hematopoietic stem cell transplant described that siblings had a more difficult time adjusting to COVID-19 than patients because COVID-19 had a greater impact on siblings' everyday lives (Chardon et al., 2021). While these findings suggest that siblings of youth with cancer have likely been impacted by the pandemic in far-reaching and unique ways, sibling-focused research is needed to characterize siblings' specific challenges and to urgently inform clinical recommendations related to FCC for families of youth with cancer.

While research has yet to characterize how siblings of youth with cancer have been impacted by the COVID-19 pandemic, it is likely that siblings have experienced changes within multiple domains of life, including restricted access within the patient's cancer treatment setting. When the COVID-19 pandemic started, healthcare leadership were tasked with the challenge of how to balance pediatric cancer patients' critical need for treatment with the healthcare systems' need for policies that minimized the spread of

COVID-19 (Sullivan et al., 2020). Many children's hospitals instituted stricter visitation policies, such as only allowing one adult caregiver at the patient's bedside, either at a time or for the duration of the patient's hospital stay, and excluding siblings and other visitors. Across the healthcare system, these pandemic-related changes to prioritize infection control have been associated with tradeoffs (Hugelius et al., 2021; Virani et al., 2020) that de-prioritized family-centered care (FCC), and how these tradeoffs impact families of youth with cancer is not yet understood.

The psychosocial standards of care in pediatric oncology highlight that siblings' psychosocial needs must be considered to provide FCC for families of youth with cancer (Gerhardt et al., 2015). Specifically, it is recommended that siblings are involved in cancer treatment in a way that is consistent with their preferences, provided with developmentally appropriate information about cancer, routinely screened for psychosocial risk factors, and provided with supportive services (Gerhardt et al., 2015; Wiener et al., 2020). Despite the establishment of these recommendations related to FCC for siblings of youth with cancer, research suggests that even before the start of the pandemic these recommendations were rarely actualized in practice, due to barriers including healthcare providers limited interaction with siblings (Brosnan et al., 2021). It is likely that the pandemic has increased these barriers to FCC for siblings of youth with cancer.

It is particularly important to understand how COVID-19 has impacted siblings of youth with cancer given that siblings were recognized as an at-risk population for poor psychosocial outcomes before the pandemic started (Long et al., 2018). Following the

patient's cancer diagnosis, siblings of youth with cancer endorse a range of negative emotions, such as fear, jealousy, loneliness, anger, and worry (Long et al., 2018), and a majority of siblings experience cancer-related post-traumatic stress symptoms (Kaplan et al., 2013). For some siblings, the psychosocial effects of cancer appear to influence functioning in other domains (e.g., poorer school performance, Alderfer et al., 2015) and can extend years after treatment has ended (Alderfer et al., 2003). Similar to cancer treatment, the COVID-19 pandemic may include potentially traumatic medical events (e.g., having COVID-19, being hospitalized; Kazak et al., 2021) and may result in negative emotional reactions for siblings (e.g., fear, loneliness, post-traumatic stress). Siblings of youth with cancer may currently be especially susceptible to poor outcomes given they are enduring stressors associated with the COVID-19 pandemic in addition to stressors associated with cancer.

Given that siblings of children with cancer are at increased risk for negative psychosocial outcomes (Long et al., 2018), and that COVID-19 has resulted in changes for siblings across healthcare, school, peer and family systems, it is critical to clarify the impact of COVID-19 within this population to inform recommendations for sibling psychosocial assessment and family-centered care moving forward. The current study used a social-ecological approach to elucidate the impact of the COVID-19 pandemic on siblings of youth with cancer. Within this lens, siblings' psychosocial functioning is understood to be influenced by the multiple inter-related systems, which siblings are embedded within. Specifically, we aimed to characterize the impact of the COVID-19 pandemic on siblings of youth with cancer across family, school, community and

healthcare settings. Using a social-ecological approach allows us to examine the intersection of systems and multilevel factors that lead to risk and resilience, which are critical to informing clinical recommendations for siblings and families of youth with cancer.

Method

Participants and Recruitment

Thirty-eight participants completed the present study, including 22 siblings and 16 parents from 16 families of youth with cancer. Parents were eligible to participate if they had a child who was diagnosed with cancer between 3 months and 5 years previously, their child's cancer treatment took place within the United States, and they had at least one other child (i.e., a sibling) who was eligible to participate. Siblings were eligible to participate if they were living with the child with cancer during cancer treatment, were under the age of 18 at the time of diagnosis, and were between the ages of 8-18 years old at the time of study participation. Parents and siblings needed to be fluent in English. No more than two siblings per family were enrolled. Families were enrolled between January and August of 2021. Enrollment continued until saturation was reached on all primary research questions, meaning that participants continued to be interviewed until the themes that emerged were fully explored (Morse, 1995). Parents and siblings each received a \$25 gift card for participation. All study procedures were approved by the Boston University Institutional Review Board.

Information about the research study was distributed to potentially eligible families using a number of recruitment methods. First, participants were recruited by re-

contacting families who participated in a previous study within the PI's research lab, provided written consent to be contacted for future studies, and met eligibility criteria for the current study. Second, information about the study was distributed through two pediatric cancer hospitals. At one hospital, the study recruitment flyer was hung in the clinic waiting room. At the other hospital, psychosocial providers shared study information with potentially eligible families by email. Finally, information about the study was distributed via social media (i.e. Twitter and Facebook). The recruitment materials directed interested families to contact the PI or visit the study webpage for more information. The study webpage contained information about the research study, the PI's contact information, and a contact field, where families could enter their contact information if they preferred to be contacted by the PI.

Relative to the families who had participated in past research, the PI contacted eight families, three were enrolled, one expressed interest in participating but was lost to follow up, and four did not respond. Relative to the families recruited through pediatric cancer hospitals and social media, 27 families expressed interest in participating by contacting the PI by email/phone or entering their information into the webpage contact field. The PI made outreach to interested families by phone to explain study procedures and ask screening questions to determine eligibility. Seven families were unable to be reached, five families who heard about the research study on social media were determined ineligible during screening (two due to the patient's age, two due to time since cancer diagnosis, and one due to cancer treatment not occurring within the United States), two expressed interest in participating but were lost to follow up and 13 were

enrolled. The final sample (22 siblings and 16 parents from 16 families) included three families recruited from past research studies, four families recruited from pediatric cancer hospitals, and nine families recruited via social media and/or word of mouth.

Procedures

Informed consent and assent processes were completed. Parents provided information about family demographics and cancer characteristics via a brief online questionnaire. Siblings and parents were then scheduled for separate interviews that were held virtually on zoom. Interviews were conducted in English by the PI, a doctoral student with intensive training in pediatric cancer and qualitative interviewing methods.

Detailed semi-structured interview guides were created for the present study. Interviews began with introductory questions about the family and cancer experience, and then transitioned to open-ended questions about how siblings were involved or participated in cancer treatment within the hospital, outpatient clinic and home settings. Given that pandemic-related changes within healthcare settings (e.g., hospital visitation restrictions) would likely influence siblings' involvement in cancer treatment for families of patients currently receiving treatment, probes related to the impact of the COVID-19 pandemic were included throughout the interview guide. Additionally, a separate section of the interview guide included questions specifically about the impact of the COVID-19 pandemic on siblings across settings (i.e., family, school, community, healthcare). Parents and siblings were asked open-ended questions about how COVID-19 impacted their family (e.g., "How has the COVID-19 pandemic changed things for your family?", "In what ways has the pandemic influenced how members of your family get along?"),

the sibling (e.g., “How has COVID-19 impacted life for you?”), the patient’s cancer treatment (e.g., “How has the pandemic influenced [patient]’s care within the hospital/clinic?) and the sibling’s involvement in cancer treatment (e.g., “How was [sibling]’s involvement or participation in [patient]’s treatment impacted by COVID-19?”).

Data Analysis

Interviews were audio recorded, transcribed verbatim, and cleaned. Interview transcripts were entered into NVivo 12 (QSR, 2018) and analyzed using applied thematic analysis methodology (Guest, MacQueen, & Namey, 2012). An initial qualitative coding structure was developed based on *a priori* research questions. The coding structure was then iteratively updated to include findings that emerged from the interviews. The coding structure included each code’s name and instructions regarding when it should be applied. A specific code was included related to the COVID-19 pandemic and related changes. The first two interviews were coded together by the coding team, during which the coding structure was refined and finalized. The final coding structure was then used to code all sibling and parent transcripts. The coding team included the PI and another psychology doctoral student with experience in applied thematic analysis and pediatric cancer, with close oversight from a psychologist with expertise in qualitative methods and siblings of youth with cancer. The coding team included the PI (EP) and another psychology doctoral student with research experience in applied thematic analysis and pediatric cancer (PB), with close oversight from a psychologist with expertise in qualitative methods and siblings of youth with cancer (KL). All members of the coding

team are trained as clinical psychologists, and specialize in youth and family populations. EP and KL have clinical experience working with families of youth with cancer, including patients, parents, and siblings. No members of the coding team have personal experience with pediatric cancer. EP has experience working within a pediatric hospital during a time of heightened visitation restrictions in the context of COVID-19. Weekly meetings were held throughout the coding process to discuss application of the coding structure and to resolve discrepancies that arose in assigning codes. Twenty percent of transcripts were double coded and then compared to ensure reliability across coders. Inter-coder concordance, calculated by dividing the number of concordant codes by the total number of codes assigned (Morse et al., 2002) was 94%. Consensus was reached on all codes prior to data analysis. Transcripts were analyzed using applied thematic analysis methodology (Guest et al., 2012). Data were queried to explore siblings' and parents' descriptions of the impact of the COVID-19 pandemic on siblings across systems (i.e. family, school, community, healthcare). Data were stratified based on families' cancer treatment timing relative to the start of the COVID-19 pandemic in March 2020 (i.e. families of patients that finished cancer treatment before March 2020, families of patients that started treatment before March 2020 and were still receiving treatment after March 2020, and families of patients that started cancer treatment after March 2020). Additionally, data were also stratified based patients' treatment status (i.e. active treatment, maintenance treatment, off treatment).

Results

Thirty-eight interviews in total were completed, including interviews with 22 siblings and 16 parents from 16 families. Siblings and families varied on demographic and cancer-related characteristics (Table 1). Families also varied on patients' cancer treatment timing relative to the start of the COVID-19 pandemic in March 2020. Participants were 2 families (including 3 siblings and 2 parents) of patients that began cancer treatment after the start of the COVID-19 pandemic, 10 families (including 12 siblings and 10 parents) of patients that began cancer treatment before the start of the COVID-19 pandemic, and 4 families (including 7 siblings and 4 parents) of patients that finished cancer treatment before the start of the COVID-19 pandemic.

Social-Ecological Impact of COVID-19 Pandemic

The COVID-19 pandemic resulted in changes for siblings across multiple systems, including family, school, community and healthcare. Given that COVID-19 impacted these systems simultaneously, changes for siblings were often inter-related. For example, stay at home orders resulted in increased time at home, virtual school, reduced community engagement and stricter healthcare policies.

Across families' descriptions of how siblings were impacted by the COVID-19 pandemic, two overarching themes emerged that reflect the bidirectional relationship between factors related to being a sibling of a youth with cancer and factors related to being a youth living during the COVID-19 pandemic. Specifically, the first overarching theme was "Being a Sibling of a Youth with Cancer Exacerbated Encountered Pandemic-Related Challenges." This overarching theme was more fully characterized by three sub-

themes (1) cancer-related worries about contracting COVID-19 increased siblings' health precautions, (2) due to cancer treatment, siblings' social isolation began before pandemic-related social isolation started, and (3) cancer-related demands made it challenging for parents to support siblings' transition to virtual school. The second overarching theme was "The Pandemic Exacerbated Challenges Associated with Being a Sibling of a Youth with Cancer." This overarching theme was also more fully characterized by three sub-themes (1) families had reduced access to resources and supports that siblings previously relied on, (2) stricter visitation policies within healthcare settings excluded siblings from cancer treatment, and (3) increased time at home resulted in fewer opportunities for siblings to "take a break" from cancer.

Being a Sibling of a Youth with Cancer Exacerbated Encountered COVID-19

Pandemic-Related Challenges

Families described how challenges associated with being a sibling of a youth with cancer made changes related to the COVID-19 pandemic even more difficult. Specifically, in the context of cancer treatment, many siblings already experienced health-related worries, social isolation, and reduced parental support. Families reported that these cancer-related factors exacerbated siblings' pandemic-related health concerns, and difficulties related to social isolation and virtual school.

Cancer-Related Worries about Contracting COVID-19 Increased Siblings' Health Precautions. Siblings reported that they were more worried about contracting COVID-19 than their peers because they worried about then transmitting the virus to their brother or sister with cancer. One parent shared, "[Sibling is] kind of like zero

tolerance. 'I don't want to catch it. I don't want to do this. I don't want to do that.' He doesn't want to bring it home to [patient].” With the context of siblings’ heightened concerns about contracting COVID-19 and transmitting it to patients, siblings reported increased worries in response to their communities beginning to relax pandemic-related health precautions, particularly within their school settings. For example, one sibling shared, “I'm definitely worried about [patient] getting sick. I guess you don't have to wear a mask whenever we get back to school. I'm really worried about that.”

Families described pandemic-related changes to their local, state and federal policies, including stay at home orders, restrictions on business and school openings, and mask mandates. Families often reported taking additional precautions above and beyond others within their communities because they had a child diagnosed with cancer. As one parent shared, “We are more locked down than other families... we literally go nowhere.” Parents described their families’ stricter health precautions as necessary due to having a child with a compromised immune system; however, they also highlighted the negative social and emotional consequences of these precautions on siblings. As a result of families’ concerns about patients contracting COVID-19, many families described that siblings had more limited involvement in activities than peers. One sibling reported, “I do not go anywhere. So it's been really difficult because I feel like I don't see any friends, and they don't try and do things that I can do usually, because it's 10 degrees outside and nobody wants to go for a walk... I don't know anyone else in the same situation...it feels very lonely and kind of never ending.”

Due to Cancer Treatment, Siblings' Social Isolation Began Before Pandemic-Related Social Isolation Started. Before the COVID-19 pandemic started, many siblings of youth with cancer already had experience with implementing health precautions, such as frequently washing hands, disinfecting surfaces, and reducing social interaction. Families needed to engage in these practices when patients were receiving cancer treatment that compromised their immune systems, and siblings described that these experiences helped them feel prepared to implement similar practices in the context of COVID-19. Families also reported that siblings' previous cancer-related experiences made the burden of similar pandemic-related practices feel heavier, because siblings had been engaging in these practices for a prolonged period of time. As one parent reported, "It's been years and years, we've been wanting to emerge, like, let us out! Steroids and fear and sickness and disconnection and so much isolation, then to have COVID, it's like, is this ever going to end?" Some parents reported that their family was about to reach the point in the patient's cancer treatment when they would have been able to relax health precautions, and then the COVID-19 pandemic started. Parents and siblings described that it was emotionally challenging for siblings to accept the necessity of more social isolation, when they had been excitedly looking forward to getting back to their "normal" life and socially reconnecting with their friends and broader communities.

Cancer-Related Demands Made It Challenging for Parents to Support Siblings' Transition to Virtual School. The majority of families reported that the transition to virtual school was challenging for siblings; however, families of patients in active treatment described that cancer-related demands made it especially difficult for

parents to support siblings with this transition. In this way, siblings of patients in active treatment described having difficulty with virtual school for non-cancer related reasons, such as reduced structure and less social interaction, and that these challenges were exacerbated by less parental supervision and support due to cancer-related demands. As one sibling of a patient in active treatment described:

“I had to stay on top of my own school work [because] I was virtual and my parents were gone sometimes. Before, they were strict about grades, now it’s like it’s not important anymore. It’s a little annoying, like, I’m working really hard, please give me some credit here.”

The COVID-19 Pandemic Exacerbated Challenges Associated with Being a Sibling of a Youth with Cancer

Changes associated with the COVID-19 pandemic made difficulties related to being a sibling of a youth with cancer even more challenging. Specifically, the COVID-19 pandemic resulted in reduced access to resources and supports, stricter visitation policies within healthcare settings, and increased time at home. These changes impacted siblings of youth with cancer in unique ways given that siblings often utilized external supports (e.g., grandparents), time outside the home, and involvement in cancer treatment to cope with and adjust to cancer treatment.

Families Had Reduced Access to Resources/Supports that Siblings Previously Relied On. The COVID-19 pandemic negatively impacted families’ access to resources and supports in many ways, including reduced family income due to parental job loss, inability to access social supports due to social distancing, and more limited availability of community resources. One sibling described the financial difficulties his family was facing within the context of cancer treatment costs and the pandemic-related financial

insecurity, “The cost of treatment it’s really drained us financially and we had to move to somewhere else and even like forgo some things... we've even had to change our meals.” Families also described reduced access to social supports, such as friends and extended family members. Within some families, grandparents and family friends had served important roles in caring for siblings, but these arrangements were no longer possible due to concerns related to COVID-19 exposure. One parent reported, “My mom, who stayed at the house a lot of those first few months, after COVID, she didn't. She was scared to come over.” Additionally, families commented on reduced access to psychosocial supports within their communities, such as mental health providers and cancer-focused supports. One parent reported, “People seem to be really affected mentally. Before, [sibling]’s therapist was not so busy, but, currently it's so busy and hard to find times.” Another parent shared, “For a couple of years [sibling] participated in [Camp for families of youth with cancer], which has totally changed our lives. [Camp] knows what we need better than we do. Unfortunately, they're closed at this time because of COVID.”

Stricter Visitation Policies within Healthcare Settings Excluded Siblings from Cancer Treatment. Hospitals and outpatient clinics where families went for patients’ cancer treatment introduced stricter family visitation policies that prohibited siblings from entering the hospital or clinic and only allowed one or two adult caregiver(s) to be with the patient at a time. Some hospitals also implemented new rules that made it more challenging for parents to alternate between hospital and home settings. These changes had multiple implications for siblings, relative to their understanding and comfort related to cancer treatment, as well as their family relationships.

When siblings were not allowed within the hospital and clinic, they expressed less understanding of, and comfort with, cancer treatment. Exclusion from these healthcare settings meant that siblings had less opportunities to learn about cancer through observing patients' care and asking providers questions. One sibling whose brother was diagnosed with cancer during the pandemic reported wishing she could go to the hospital:

“I could really see what was happening. I wouldn't have to just go off what my mom was telling me, I could ask my own questions. I wouldn't be as worried, cause I could see him, and know what was happening.”

Some siblings who went to the hospital or clinic pre-pandemic, commented on how they no longer had the same access to healthcare-based supports that had helped them to understand cancer treatment. For example, one sibling reported, “If I had any questions, I would go to [child life specialist]. Then, after COVID, I didn't go to the hospital, so I couldn't ask her.” Similarly, families who finished cancer treatment before the pandemic reported that it would have been challenging if siblings were not allowed to visit the hospital or go to clinic appointments because these experiences helped siblings understand the patient's treatment and feel more comfortable. One sibling shared, “Being at the hospital, it makes it more comfortable...just being around it, so I think it would have been really hard to not be there.”

Visitation restrictions within healthcare settings also impacted siblings' relationships with family members in multiple ways. It was challenging for patients to not be allowed visitors, and siblings experienced negative emotions related to not being able provide patients with emotional support. One sibling who was accustomed to visiting her sister in the hospital before the pandemic reported, “Because of COVID, I couldn't go,

even with a mask, just say ‘hi,’ I still couldn't. So, she'd be in the hospital for like a month and I couldn't even go give her a hug.” Families used video calls and other creative strategies to help patients and siblings connect when siblings were not allowed in the hospital; however, families often described that these approaches (while helpful) failed to provide the same emotional connection and comfort as in-person interactions. One sibling of a patient who was diagnosed during the pandemic reported, “We went to the hospital parking lot and wrote with chalk, like, ‘you got this, you're doing great,’ so [patient] could look from his window, and see us like dancing around. I think it made him a little bit happier.” Families of patients who finished cancer treatment before the pandemic expressed concern for those families currently going through treatment. They reported that it would have been emotionally challenging for siblings, patients, and parents if siblings had not been allowed to visit the hospital. A parent from one of these families shared:

“I think it would have been really hard for [siblings] to cope, not being able to see [patient] or touch him. For them to try to go to sleep at night knowing he's having a tough time and we can't see him or hug mom, I couldn't imagine. I feel so bad for those that are going through it.”

Stricter policies within healthcare settings also limited parents' ability to spend time with siblings. One parent who experienced cancer-related hospitalizations before and during the pandemic reported, “I always relied on another grown up to take my place so I could take a break. During COVID, that was really difficult, I was [at the hospital] nonstop. I wasn't able to leave and go see [sibling].” Parents of patients who finished treatment before COVID-19 reported that the pandemic-related visitation restrictions would have been emotionally challenging for them because they would have needed to choose

whether to spend time with the patient or the sibling. For example, one parent shared:

I would have felt so torn all the time. [Sibling] would have felt so left out. And, if I were to be home with her, I would have felt, like ‘what the hell is going on at the hospital? That would have been absolutely terrible.

Families described that in certain situations staff members had allowed their family to not abide by the pandemic-related restrictions and had allowed siblings to be present within cancer-related healthcare settings. For example, one parent reported that staff within the emergency room allowed the sibling to see her brother who had just received his cancer diagnosis before he went into surgery. This parent shared, “Thankfully the hospital bent the rules and let [Sibling] come into the ER. They were like ‘we’ll say she’s a patient if we have to, but she gets to come in and hang out with her brother for a while.” Another parent reported that the sibling had been allowed to come with her to outpatient clinic appointments.

“[Clinic] made an exception because as a single mom, it’s hard to always find childcare, also, I can keep [sibling] safer from COVID with me, rather than bringing him to different people’s houses, you know, inside not wearing masks. So, I said [to clinic staff] it kind of comes down to either [patient] can have her appointments with [sibling] or I can’t be bringing her all the time.”

Increased Time at Home Resulted in Fewer Opportunities for Siblings to

“Take a Break” from Cancer. Due to increased time at home, siblings had the opportunity to engage in more family activities; however, siblings of patients in active treatment also encountered more cancer reminders and had fewer opportunities to “take a break” from cancer. Families described both benefits and difficulties associated with increased family time, including closer relationships and new family routines (e.g., movies nights, karaoke), as well as decreased frustration tolerance with one another due

to not getting “breaks.” In addition to not getting “breaks” from family members, siblings of patient in active treatment described the transition to spending more time at home as challenging because they were not accustomed to being surrounded by cancer-related reminders all of the time. For example, one sibling reported:

I can't get out of the house and clear my head like I used to. [Cancer has] become my normal now, like hearing about everything, and the nurse comes. It's definitely become more part of my life than it was like a year ago.

Siblings of patients at all treatment stages (i.e. active treatment, maintenance treatment, off treatment) reported that it was challenging to not be able to see friends and engage in extracurricular activities; however, siblings of patients in active treatment described this as especially difficult in conjunction with their increased exposure to cancer-related reminders at home. One sibling of a patient in active treatment described:

I feel like I can never get away from it. You don't get out and do the things that make you happy and feel normal again. It feels overwhelming. I mean, as a parent it's kind of your job to do that. But as a sibling, it's less of that, so [I] feel trapped.

Siblings described how previously they had relied on time with friends and extracurricular activities to help them feel better and that without access to those coping options it at times felt hard to cope with cancer-related thoughts and feelings.

Discussion

From the start of the COVID-19 pandemic in March 2020 to present day, children and families across the United States have been impacted by the pandemic's far-reaching impact (Feinberg et al., 2022). Families have faced challenges associated with social isolation, major disruptions to daily life, and the loss of loved ones due to COVID-19. In addition, families of youth with cancer have had to worry about caring for

immunocompromised patients and navigating an abruptly changing healthcare system (Sullivan et al., 2020). While emerging research has been conducted to understand the impact of the COVID-19 pandemic on families of children with cancer, the present study is the first to our knowledge to characterize the impact of the COVID-19 pandemic on siblings specifically. Siblings were recognized as an at-risk population for poor psychosocial outcomes before the pandemic started (Long et al., 2018), and emerging research suggests that siblings may have been greatly impacted by the pandemic. Current findings suggest that cancer-related and pandemic-related challenges exacerbate each other for siblings of youth with cancer. These findings are consistent with previous work in which parents reported that siblings had a more difficult time adjusting to COVID-19 than patients because COVID-19 had a greater impact on siblings' everyday lives (Chardon et al., 2021).

While emerging research suggests that the psychosocial functioning of youth with cancer may be less affected by the COVID-19 pandemic than the general population (van Gorp et al., 2022), present findings do not suggest that the same is true for siblings of youth with cancer. Some speculate that youth with cancer are less impacted by the pandemic because they are accustomed to practices such as social isolation, health precautions and home schooling, and that in fact, the pandemic has allowed patients to feel less different, as peers have needed to adopt similar practices (van Gorp et al., 2021). However, siblings appear to have had a quite different experience during the pandemic.

Rather than reducing siblings' cancer-related challenges, present findings suggest that changes associated with the COVID-19 pandemic have exacerbated siblings' unique

vulnerabilities. In the context of cancer treatment, many siblings already experienced worries about family members' health, endured periods of social isolation, and had reduced parental support. Families reported that these cancer-related factors exacerbated siblings' pandemic-related health concerns, made virtual school more challenging (in the context of less parental support), and prolonged siblings' experiences of social isolation.

Siblings were more worried about contracting COVID-19 than their peers because they worried about then transmitting the virus to the patient with cancer. Additionally, siblings reported they were often stricter relative to health precautions than their peers. These findings build upon research documenting that parents of youth with cancer worried about family members getting the virus and transmitting it to the youth with cancer, and that parents vigilantly followed health guidelines (Chardon et al., 2021; Darlington et al., 2021; Wimberly et al., 2021). Within the present study, sibling participants tended to express similar pandemic-related opinions and practices as their corresponding parent participant. These findings raise the possibility that siblings' pandemic-related beliefs and practices may be in part due to parental modeling.

Additionally, findings highlight how cancer-related demands during active treatment reduced parents' capacity to support siblings with pandemic-related changes. These findings align with research suggesting that parents of youth with chronic disease, particularly mothers and single parents, are particularly vulnerable to parental burnout during the COVID-19 pandemic (Wauters et al., 2022). While the present study specifically identified parents' reduced capacity to support siblings' participation in virtual school, findings may reflect a broader trend in parents' reduced capacity to attend

to siblings' needs while simultaneously managing cancer-related demands and pandemic-related stressors. Siblings had unmet needs related to parental support before COVID-19 (Pariseau et al., 2020), and the pandemic may have exacerbated these needs in ways that have worrisome implications for siblings' psychosocial adjustment. Given that present findings specifically identified parents' reduced capacity to support siblings with virtual school, and that even before the pandemic siblings demonstrated poorer academic outcomes (Alderfer et al., 2015), siblings' school functioning may be an important target for assessment and intervention moving forward.

Present study findings also describe how changes associated with the COVID-19 pandemic made difficulties related to being a sibling of a youth with cancer even more difficult. Specifically, the COVID-19 pandemic resulted in reduced access to resources and supports and increased time at home. These changes impacted siblings of youth with cancer in unique ways given that siblings often utilized external supports (e.g., grandparents) and time outside the home to cope with and adjust to cancer treatment. With stay-at-home orders, siblings experienced increased exposure to cancer-related reminders and fewer opportunities to take "breaks" from cancer, which they had previously done through connecting with peers and engaging in activities within their communities. Before the COVID-19 pandemic, siblings expressed a need for supports that helped them to "take a break" from cancer (Patterson et al., 2011), and present findings highlight that the pandemic created an environment in which siblings were less able to access those much-needed "breaks," due to reduced community supports and increased exposure to cancer-reminders at home.

Importantly, findings characterize the impact of healthcare settings' stricter family visitation policies on siblings of youth with cancer. Families reported that in response to the COVID-19 pandemic, siblings were prohibited from entering the hospital or clinic. When siblings were not allowed within the hospital and clinic, they expressed less understanding of, and comfort with, cancer treatment. Exclusion from these healthcare settings meant that siblings had less opportunities to learn about cancer through observing patients' care and asking providers questions. These stricter visitation policies were also described as hindering siblings' relationships with family members who were at the hospital. Before the pandemic started, siblings were documented to have unmet needs relative to both family support and understanding of cancer (Long et al., 2018). It is concerning to consider how siblings' pre-existing unmet needs and pandemic-related barriers to adaptively adjusting to cancer will contribute to siblings' psychosocial functioning moving forward.

Present findings have clinical implications related to sibling psychosocial assessment. The Sibling Standard of Psychosocial Care recommends routine, universal sibling screening for current and anticipated psychosocial difficulties (Gerhardt et al., 2015). As a first step, present study findings underscore the importance of implementing universal psychosocial screening of siblings of youth with cancer to better characterize the psychosocial impact of siblings' intersecting cancer- and pandemic-related vulnerabilities, and to identify siblings in need of psychosocial supports. Healthcare providers reported barriers to implementing screening practices before the pandemic (Brosnan et al., 2021) and siblings' ongoing restriction within (at least) some treatment

settings will likely exacerbate those barriers. However, sibling screening is a crucial component in providing family-centered care for families of youth with cancer, particularly given that siblings may be more negatively impacted by the pandemic than other members of the family. Telehealth and other virtual screening options will likely be important options to explore.

While some pandemic-related changes (e.g., stay at home orders) have been lifted, and communities have slowly tried to resume “normal” activities of daily living, other pandemic-related changes have remained. Specifically, many healthcare settings have continued to enforce stricter visitation policies. For example, (as of July 2022) many pediatric cancer clinics in the United States still prohibit patients from bringing companions under the age of 18, including siblings. Deciding how to balance priorities related to infection control and family-centered care within healthcare settings is certainly not an easy one; however, the present findings highlight negative psychosocial consequences of excluding siblings from cancer treatment settings, and these findings are crucial to consider moving forward.

Strengths of the current study included enrolling families with diverse experiences relative to the intersection of cancer treatment and the COVID-19 pandemic, including families of patients that finished treatment before the COVID-19 pandemic, families of patients whose treatment was ongoing during the COVID-19 pandemic, and families of patients that were diagnosed and started treatment during the COVID-19 pandemic. Together, these families’ perspectives collectively highlight the challenges faced by siblings of youth with cancer during the pandemic. Additionally, the present study

enrolled both siblings and parents, an important methodological decision given that's parents and siblings often have non-overlapping but complementary perspectives (Pariseau et al., 2020).

The present study also had several limitations. Families were enrolled between January and August of 2021; the ever-changing nature of the pandemic and its evolving impact on health care settings may limit the generalizability of findings. Additionally, data collection began when limited resources were available to quantitatively assess the impact of the COVID-19 pandemic on families. Future research should use standardized measures, such as the COVID-19 Exposure and Family Impact Scales (Kazak et al., 2019) to more fully and systematically characterize the ongoing impact of COVID-19 on families of youth with cancer.

The present study highlights the multifaceted ways in which siblings of youth with cancer have been impacted by the COVID-19 pandemic. While some pandemic-related changes have been lifted (e.g., youth have returned to in-person school), siblings of youth with cancer continue to be excluded from many cancer-related treatment settings. Findings suggest that excluding siblings from treatment settings can negatively impact siblings' understanding of cancer treatment, increase siblings' cancer-related worries and interfere with siblings' ability to provide cancer patients with much-needed family support as they cope with cancer treatment. As treatment settings move forward and consider if and how to adjust visitation restrictions, it is crucial that the psychosocial ramifications of excluding siblings of youth with cancer be considered.

Chapter 5: General Discussion

Siblings of youth with cancer demonstrate variable psychosocial adjustment to childhood cancer, ranging from cancer-related post-traumatic stress symptoms (PTSS) to resilience (Long et al., 2018). Theoretical frameworks (Lazarus & Folkman; 1984; Price et al., 2016) suggest that PTSS are influenced by appraisals (e.g., fear of mortality) and coping. Siblings' presence within clinical settings and participation in cancer treatment may allow them to appraise cancer-related experiences as less threatening and utilize engagement coping strategies, but the nature and effect of siblings' involvement in cancer treatment remains unknown, especially in the context of COVID-19.

Broadly, my goals for this three-part mixed-methods research project were to (a) characterize siblings' involvement in cancer treatment, (b) investigate how siblings' involvement in cancer treatment relates to PTSS, and (c) characterize COVID-19's impact on siblings.

In Study 1, I qualitatively characterized siblings' and parents' descriptions of siblings' involvement in cancer treatment. Siblings were actively involved in cancer treatment across the cancer trajectory through learning about cancer and helping with emotional and/or cancer management aspects of treatment. Within the hospital, outpatient clinic and home settings, siblings' involvement in cancer treatment was characterized by witnessing cancer symptoms and treatment, learning information about cancer, providing patients with emotional support, and contributing to cancer management.

Witnessing patients' cancer symptoms and treatment in-person helped facilitate siblings' understanding of cancer. However, these experiences also triggered negative

emotions for many siblings, at least initially. Families largely described that siblings' initial experiences were the most distressing, and that when siblings had the opportunity to continue to engage in treatment (e.g., kept going to the hospital) siblings became more comfortable over time. In fact, continued involvement seemed to lead to better adjustment over time as siblings habituated to initially-distressing cancer cues.

Siblings were also involved in treatment through learning information about cancer. Past research shows that siblings frequently indicate uncertainty related to the patient's cancer diagnosis or treatment (Jenholt Nolbris & Ahlström, 2014), and retrospectively, siblings express that increased involvement in cancer treatment and opportunities to learn about cancer from medical providers would have been helpful for their psychosocial adjustment (Lövgren et al., 2016). Building on these findings, the present study highlights that when present within clinical settings, siblings had opportunities to learn about cancer through receiving developmentally appropriate information from child life specialists and asking medical providers questions. Additionally, present findings provide additional evidence that opportunities to learn about cancer from providers have benefits for siblings' psychosocial adjustment, as enhanced understanding of cancer was described as helping to reduce siblings' cancer-related distress.

Past research suggests that siblings of youth with cancer want to support patients (Wilkins & Woodgate, 2007) and help with treatment (Patterson et al., 2011). Aligned with these findings, Study 1 found that an integral way that siblings were involved in treatment was through providing patients with emotional support (across settings) and

through taking on more active roles in cancer management at home. Quantitative research suggests that siblings of youth with chronic illness who are engaged in caregiving responsibilities reported lower anxiety and more positive relationships with patients (Kelada, 2021). The present study builds on these findings by qualitatively describing the processes through which siblings' involvement in treatment may influence their individual and interpersonal psychosocial adjustment. Specifically, ongoing involvement may facilitate lower anxiety and distress through repeated exposure to illness-related reminders (e.g., witnessing patient's symptoms) and opportunities to reappraise these experiences as less threatening. Furthermore, siblings' involvement in treatment, particularly their opportunities to provide patients with emotional support and contribute to cancer management, may enhance siblings' relationships with patients by allowing siblings to emotionally connect with patients and feel helpful.

Building on Study 1 findings that siblings' cancer-related distress appeared to decrease with ongoing involvement in treatment, I explored the relationship between siblings' involvement in cancer treatment and cancer-related post-traumatic stress symptoms (PTSS) in Study 2. Using a cross-sectional, convergent, mixed methods design, I explored the relationship between siblings' quantitative report of cancer-related PTSS (CPSS-5-SR) and siblings' and parents' qualitative descriptions of siblings' involvement in cancer treatment.

As hypothesized, siblings with low PTSS and high PTSS differed in a number of ways relative to how they were involved in cancer treatment. First, aligned with Study 1 findings, low PTSS and high PTSS groups differed with regard to consistency of

involvement in cancer treatment. Siblings with high PTSS were more likely to describe that they were initially involved in treatment (e.g., visited patient 1-2 times at the hospital) followed by a period of inability to go to the hospital or cancer clinic. These findings make sense conceptually when considered in the context of an exposure framework (Ehlers & Clark, 2000). Siblings with high PTSS were exposed to a potentially traumatic stressor (e.g., seeing patient in the hospital for the first time) and then, due to restricted access to clinical settings, may have had less ongoing exposure to cancer-related reminders and opportunities to re-appraise cancer-related stressors as less threatening.

Additionally, siblings with high PTSS were more likely to describe that their involvement in cancer treatment was in some way misaligned with their preferences. Previous research on siblings' caregiving more broadly suggests that the extent to which caregiving is voluntary influences siblings' subjective experience of caregiving as well as the impact of caregiving on siblings' psychosocial adjustment (East, 2010), and the same may be true for siblings' treatment involvement in the context of pediatric cancer. However, within the sibling caregiving literature, caregiving tended to be misaligned with siblings' preferences because it was too *extensive* (East, 2010), whereas within the present study, treatment involvement tended to misalign with siblings' preferences because it was too *limited*.

High PTSS and low PTSS groups also differed regarding access to cancer-related information and opportunities to express cancer-related emotions. Siblings with low PTSS tended to be better prepared for cancer-related experiences, have more

opportunities to learn about cancer, and receive more encouragement from parents around expressing negative cancer-related emotions. Alternatively, siblings with high PTSS received less information about cancer, at times related to parents' more limited health literacy, and were more often encouraged to stay consistently positive and minimize any negative emotions.

Finally, Study 3 characterized the impact of the COVID-19 pandemic on siblings of youth with cancer across family, school, community and healthcare settings. Given that siblings of youth with cancer were at increased risk for negative psychosocial outcomes before the pandemic (Long et al., 2018), and that the pandemic likely resulted in multi-systemic changes for siblings, including restricted access to cancer treatment settings, it was critical to clarify the impact of COVID-19 on siblings' involvement in cancer treatment and broader lives.

Study 3 findings highlighted that challenges associated with being a sibling of a youth with cancer made changes related to the COVID-19 pandemic even more difficult. Specifically, in the context of cancer treatment, many siblings already experienced health-related worries, social isolation, and reduced parental support. Families reported that these cancer-related factors exacerbated siblings' pandemic-related health concerns, and difficulties related to social isolation and virtual school.

Additionally, changes associated with the COVID-19 pandemic made difficulties related to being a sibling of a youth with cancer even more challenging. The pandemic resulted in reduced access to resources and supports, stricter visitation policies within healthcare settings, and increased time at home. These changes impacted siblings of

youth with cancer in unique ways given that siblings often utilized external supports (e.g., grandparents), time outside the home, and involvement in cancer treatment to cope with and adjust to cancer treatment.

Importantly, in response to the COVID-19 pandemic, siblings were prohibited from entering the hospital or outpatient cancer clinic. Aligned with Study 1 findings that involvement in cancer treatment enhanced siblings' understanding of cancer, Study 3 highlighted that siblings' exclusion from healthcare settings hindered their understanding of, and comfort with, cancer treatment. When considered with findings from Studies 1 and 2, Study 3 findings related to siblings' exclusion from cancer treatment settings during the COVID-19 pandemic are particularly concerning. Specifically, Study 3 highlighted that siblings had reduced opportunities to witness cancer treatment, learn about cancer, or provide emotional support to patients within the hospital. Additionally, siblings' involvement has not been consistent over time, or aligned with their preferences. Together, these findings suggest that pandemic-related restrictions that disallowed siblings' presence within clinical settings may have increased siblings' risk for cancer-related PTSS.

Findings from across all three studies have clinical implications related to sibling psychosocial assessment and support. The Sibling Standard of Psychosocial Care recommends routine, universal sibling screening for current and anticipated psychosocial difficulties (Gerhardt et al., 2015). As a first step, present study findings underscore the importance of implementing universal psychosocial screening of siblings of youth with cancer to better characterize the psychosocial impact of siblings' intersecting cancer- and

pandemic-related vulnerabilities, and to identify siblings in need of psychosocial supports. Sibling screening is a crucial component in providing family-centered care for families of youth with cancer, particularly given that siblings may be more negatively impacted by the pandemic than other members of the family (van Gorp et al., 2022). Telehealth and other virtual screening options will likely be important options to explore.

Additionally, if / when siblings are allowed to return to treatment settings, psychosocial providers can play an important role in providing families with psychoeducation related to exposure and supporting families to make informed decisions about how to involve siblings in treatment. Specifically, families should be aware that siblings' initial distress in-response to cancer-related experiences is normative, and that through ongoing involvement, siblings' distress will likely subside and they will become increasingly comfortable. Families should also be educated that limiting siblings' ongoing opportunities for involvement may likely perpetuate their distress.

Whether or not siblings are physically allowed within treatment settings, professionals can support families to identify and facilitate opportunities for siblings to learn developmentally appropriate information about cancer, provide patients with emotional support and help with cancer management at home. Professionals may consider connecting with siblings via telehealth to provide information, show siblings helpful visual aids, and answer siblings' questions. Additionally, professionals should support parents to communicate with siblings about their preferences related to involvement in treatment, as well as to model healthy emotional expression of cancer-related emotions.

Strengths of the current study include its mixed methods approach, which enabled characterization of siblings' involvement in cancer treatment using rigorous qualitative methods, as well as quantitative measurement of previously well-characterized constructs (e.g., PTSS, coping) with quantitative measures. The present study was also grounded in multiple well-established theoretic frameworks (i.e., Ehlers & Clark, 2000; Kazak et al., 2006; Lazarus & Folkman, 1984). Additional strengths include the consideration of both sibling and parent perspectives and the enrollment of a diverse sample relative to race, ethnicity, sibling age and gender, geographic region, and time since diagnosis. Additionally, participants included families with diverse experiences relative to the intersection of cancer treatment and the COVID-19 pandemic, including families of patients that finished treatment before the COVID-19 pandemic, families of patients whose treatment was ongoing during the COVID-19 pandemic, and families of patients that were diagnosed and started treatment during the COVID-19 pandemic.

Findings must be interpreted in the context of the study limitations. While we enrolled families of patients diagnosed with multiple cancer types, the majority of participants were families of youth with leukemia/lymphoma. Future, larger scale research should be conducted to explore how siblings' involvement in treatment may likely vary based on the patient's specific cancer diagnosis and treatment protocol. Research should also explore the current findings with larger, more diverse samples to understand the influence of cultural and family diversity on siblings' involvement in treatment. An additional limitation is that the sample size of the present study was too small to reliably detect small to medium effects within Study 2. Therefore, null findings

should be interpreted with caution, and significant findings should be used to inform future larger scale research projects. Furthermore, given the cross-sectional nature of this study, assumptions cannot be made about the directionality of relationships identified among variables. Specifically, future longitudinal studies should investigate whether siblings' involvement in cancer treatment influences their development and maintenance of cancer-related PTSS, or rather that siblings with higher PTSS tend to be more reluctant to engage with treatment in the same way as siblings with lower PTSS.

Together, the findings of these studies characterize how siblings have historically been actively involved in cancer treatment across the hospital, clinic and home settings, as well as how siblings have been excluded from healthcare settings in the context of the COVID-19 pandemic. Findings highlight multiple benefits to siblings' involvement in cancer treatment, and make connections between limited involvement and cancer-related PTSS. Within the context of the siblings' ongoing exclusion from healthcare settings two years after the start of the pandemic, these findings are concerning relative to siblings' psychosocial functioning. Universal psychosocial screening of siblings is needed to better characterize siblings' current psychosocial functioning, and clinical efforts are needed to ensure that siblings have opportunities to be involved in cancer treatment moving forward.

Table 1
Demographic and Cancer-Specific Characteristics

	Participating Siblings (N = 22 Siblings)	Participating Parents (N = 16 Parents)
Age at Study Participation, <i>M (SD)</i>	12.7 (3.1) Years	41.6 (6.2) Years
Age at Cancer Diagnosis, <i>M (SD)</i>	10.0 (3.2) Years	38.8 (6.3) Years
Gender ^b	55% Female	88% Female
Race ^c	74% White 30% Black/African American	75% White 25% Black/African American
Ethnicity	14% Hispanic/Latinx	13% Hispanic/Latinx
Parent Education		19% High School or Less 69% Some College / College
Graduate		13% Master's or Doctoral Degree
Parent Marital Status		88% Married/Partnered
Geographic Region within United States	39% South 35% Northeast 26% West or Midwest	
Healthcare Insurance	44% Medicaid 44% Private Insurance 13% Military Health Care	

Cancer Type 61% Leukemia/Lymphoma
 22% Central Nervous System Tumor
 17% Solid Tumor

Cancer Treatment Status 23% Active
 41% Maintenance
 36% Off Treatment

Time Since Diagnosis, *M (SD)* 35.6 (18.6) Months

Relapse Status 18% Relapse

^a “Participating Siblings” column includes cancer characteristics relative to siblings (e.g., 23% of siblings had a brother/sister in active treatment)

^b All participants identified all either female or male

^c Adds to more than 100% because one sibling identified as multi-racial

Table 2*Witnessing Cancer Symptoms and Treatment*

Setting	Example Quotation
Hospital	<p>Parent: “[Patient]’s health was poor at the hospital, and I think it was scary for [sibling], in a sense. But I think that seeing [patient], being in his proximity, was also really calming for him. To know that [patient]’s still there, and he still looks familiar.”</p> <p>Sibling: “Being at the hospital makes it more comfortable because you get to understand who’s taking care of your sibling.”</p>
Clinic	<p>Sibling: “[Clinic] was better because how [patient] was doing and like the surrounding things...you could just hang out, without worrying about anything because the people around weren’t as sick.”</p> <p>Sibling: “[Patient] had a clinic appointment and my mom just wanted me to see what it was like, see where she was going every week or every couple days. So, I went to clinic with her to get her lumbar puncture...Being at the place where she’s going for this stuff, that was a cool experience for me because I could just, like, understand it more from [patient]’s perspective.”</p>
Home	<p>Parent: “In the hospital and getting infusions, [Patient] always felt pretty good. It was when he was home that things went bad, and in the middle of the night, he would yell that he was throwing up...It was those times that everybody was more scared because you could physically see him getting sick and losing his hair and stuff.”</p> <p>Parent: “[Sibling’s] old bedroom...it’s turned into a nursing room, so, I’ve got all the supplies and sharps containers and everything in there.”</p>

Table 3*Learning Information About Cancer*

Setting	Example Quotation
Hospital	<p>Sibling: “[Child life specialist] did a puppet presentation with a stuffed bone with things in it that were the blood cells to explain Leukemia.”</p> <p>Sibling: “The port was the biggest thing I was scared about. [Child life specialist] explained what it did and how it helped. She helped me not be so freaked out.”</p>
Clinic	Sibling: “If I had a question, I would ask when I went for his clinic appointments because they didn’t seem like they're really busy...[Doctor] definitely helped a lot, for me to understand.”
Home	Sibling: “My parents usually gave me updates, about like [patient]’s treatment schedule, what weeks were steroid weeks and stuff like that. That was good to know, because steroids weeks she was kinda moody.”

Table 4
Providing Emotional Support and Contributing to Cancer Management

Setting		Example Quotation
Hospital	Emotional Support	Parent: “[Sibling] literally was in the bed with [patient] playing games, and it just made things more peaceful for him. [Sibling] tried to swallow you know his fear, trying to keep some normalcy like as if we're at home. We tried to just make it feel as comfortable as possible and [patient] was such a trooper. The only thing he wanted was his siblings.”
Clinic	Emotional Support	Sibling: “You could bring things like games back to the chair [patient was] sitting in. [Patient] liked doing crafts at the time, so I would like pick out crafts to do, like decorating cookies. So I would bring them back to him and we would just like, do them and hang out.”
Home	Emotional Support	Parent: “[Sibling] started climbing into bed with [patient], almost every single night, snuggling together, because [patient] was really scared. It really brought them closer”
	Cancer Management	Parent: “[Sibling] would help flush [PICC line] out and put the shot in.” Sibling: “I’ve learned to make some healthy meals, because [patient] can’t be eating junk foods, and when he sees someone else eating junk food he might feel like he’s missing out. So I tend to just eat whatever he’s eating.”

Table 5*Treatment Involvement Influences on Siblings' Individual and Interpersonal Psychosocial Adjustment to Cancer*

Theme	Example Quotation
Cancer-Related Distress is Reduced Over Time with Ongoing Involvement	Sibling: "At first I had a weird feeling about [being at the hospital]. Like 'what's going on? Is this bad?' I didn't really know what was happening. And the noises and people and medicine, I just didn't like it...[Later in treatment] we were there when [Patient] would get his vitals checked, sometimes he would get medicine. I didn't like seeing that happen, but I knew why [he needed it] and I got used to it. It was just like a normal thing when we were there."
Enhanced Feelings of Inclusion and Family Relationships	<p>Sibling: "I felt included because like I was in the room where [patient] was going through his battle with cancer and, I just really felt like we could help [patient] like emotionally, so he wouldn't feel lonely and sad."</p> <p>Parent: "For the stem cell transplant, [younger sibling] donated, but [older sibling] turned on the switch to start the transplant...to know that he was starting this important process of putting [younger sibling]'s blood into [patient]...that will forever be an important memory."</p> <p>Parent: "[Sibling] wouldn't call it 'visiting' [the hospital], especially at the beginning, that's where her family was. That was going 'home' to be with family...She'd come in for dinner. She'd take a nap [laughs]. We were often there for family dinners they have on the sixth floor, so we would have dinner together."</p>

Table 6*Example Qualitative Interview Questions and Probes*

Topic	Example Sibling* Questions and Probes
Potentially traumatic cancer-related experiences for siblings	What parts of [patient]'s cancer treatment were the scariest or the most upsetting for you?
Sibling reactions to cancer-related experiences	How did you feel when you saw [patient] in the hospital for the first time?
Sibling cancer-related appraisals	When you learned that [patient] had cancer, what worries did you have?
Sibling cancer-related coping strategies	When you felt confused or sad or scared about [patient]'s cancer treatment, how did you cope with those feelings? What helped you feel better?
Sibling preparation for cancer-related experiences	When you went to the hospital for the first time, how prepared did you feel for what you saw and did there?
Influence of treatment involvement on cancer-related appraisals and coping strategies	Did going to the hospital make you feel more worried, less worried or have no impact? Why do you think that was?
Sibling preferences and family decisions related to treatment involvement	Did you go to the clinic more than you wanted to, less than you wanted to, or about the right amount? How did your family decide how often you went?

*Parents were asked parallel questions about each topic

Table 7*Characteristics of Low PTSS and High PTSS groups*

Characteristic	PTSS Group								<i>t</i> test	p-value	Chi-Square	p-value
	Low PTSS (n=11)				High PTSS (n=11)							
Sibling Characteristics	Valid %	N	Mean	SD	Valid %	N	Mean	SD				
Age at Diagnosis (years)			9.1	3			10.9	3.1	-1.355	.19		
Female Gender ^a	45.5	5			63.6	7					.733	.67
Race											.917	.64
White	81.8	9			63.6	7						
Black/African American	18.2	2			36.4	4						
Hispanic/Latinx Ethnicity	0.0	0			27.3	3					3.474	.214
Cancer characteristics												
Time since diagnosis (months)			34.5	21.0			30.7	16.6	.461	.65		
Cancer Type											.819	.664
Leukemia/Lymphoma	54.5	6			72.7	8						
CNS Tumor	27.3	3			18.2	2						
Solid Tumor	18.2	2			9.1	1						
Treatment Status											3.911	.141
Active Treatment	9.1	1			36.4	4						
Maintenance Treatment	36.4	4			45.5	5						
Off Treatment	54.5	6			18.2	2						
Cancer Relapse	9.1	1			27.3	3					1.222	.586

^a All siblings identified as female or male

Table 8

Potentially Traumatic Cancer-Related Experiences for Siblings

Experience	Example Quotation
Learning diagnosis	Sibling “When he was diagnosed, I was quite scared, of what would happen to him, like would he be around for much longer.”
Seeing patient in the hospital for the first time	Sibling: “She's my big sister, like my role model. She had always been super active, she loved to swim, she was really big with volleyball...And then she went from doing that to being in a hospital bed with her hair falling out.”
Observing physical indicators of cancer	Sibling: “Seeing her very weak and having lost a lot of weight and, losing her hair. Those things really made me cry.”
Witnessing or hearing about aspects of patient’s medical care	Sibling: “I was kind of nervous. I felt bad and scared for him because they had to poke that needle and they had to put the button inside of his chest.”
Medical emergencies	Parent: “The kids are at school. Everyone's assuming this is a normal day, we'll see mom after school. But [the day] turned out completely different. So we're racing down the highway in the back of an ambulance and [siblings are] following behind us. [Sibling] really didn't understand. He was afraid [patient] was going to die.”
Seeing other pediatric cancer patients	Sibling: “When you go there and see people in the hallway. It's like not what you expected at all. Grandparents have cancer, but it's weird to experience like younger people that have cancer. You walk in and it's everywhere. They have monitors with IVs that they walk with. It’s just a lot at once, seeing young people sick.”

Table 9*Summary of Cancer-Related Appraisals, Coping Strategies, and PTSS*

	N (%)	Mean	SD
Cancer-Related Appraisals			
Life threat at diagnosis			
Yes	17 (77%)		
No	5 (23%)		
Life threat in treatment			
Yes	9 (41%)		
No	13 (59%)		
Coping Symptoms Inventory (CSI)			
Problem Solving		18.0	7.3
Cognitive Restructuring		22.1	8.0
Emotional Expression		20.8	6.5
Social Support		25.6	8.1
Problem Avoidance		19.3	6.0
Wishful Thinking		28.8	7.3
Self-Criticism		14.3	8.2
Social Withdrawal		20.3	7.6
PTSS (CPSS-5-SR Total Score)		25.0	18.0
“Minimal” PTSS (Scores 0-10)	6 (27%)		
“Mild” PTSS (Scores 11-20)	5 (23%)		
“Moderate” PTSS (Scores 21-40)	6 (27%)		
“Severe” PTSS (Scores 41-60)	5 (23%)		
“Very Severe” PTSS (Scores 61-80)	0 (0%)		

Table 10*Cancer-Related Appraisals: Associations with sibling demographics and cancer characteristics*

	Sibling Demographics						Cancer Characteristics					
	Age at diagnosis		Gender		Time since diagnosis		Cancer Type		Treatment Status		Cancer Relapse	
	t-test	p	Chi-square	p	t-test	p	Chi-square	p	Chi-square	p	Chi-square	p
Life threat at diagnosis	1.061	.301	.552	.457	-.929	.364	.227	.893	.046	.977	.014	.905
Life threat in treatment	.638	.531	.627	.429	-.053	.958	2.459	.292	.489	.783	.167	.683

Table 11*Cancer-Related Coping Strategies: Associations with sibling demographics and cancer characteristics*

	Sibling Demographics						Cancer Characteristics					
	Age at diagnosis		Gender		Time since diagnosis		Cancer Type		Treatment Status		Cancer Relapse	
	r	p	t-test	p	r	p	F-test	p	F-test	p	t-test	p
Problem Solving	.322	.154	.187	.855	-.094	.684	1.312	.294	.111	.896	-2.391*	.027
Cognitive Restructuring	.089	.701	.062	.952	-.085	.715	2.200	.140	.184	.833	-.750	.462
Emotional Expression	.221	.335	-1.080	.294	.127	.584	.732	.495	.702	.509	-2.161*	.044
Social Support	.009	.969	-.562	.580	.086	.709	.092	.912	2.865	.083	-1.088	.290
Problem Avoidance	.517*	.016	-.953	.353	-.345	.125	4.429*	.027	2.345	.124	-2.592*	.018
Wishful Thinking	.135	.560	-.311	.759	-.230	.316	.371	.695	.181	.836	.836	.266
Self-Criticism	.289	.204	-.347	.732	-.255	.266	1.492	.251	.530	.597	-2.284*	.034
Social Withdrawal	.440*	.046	-1.996	.060	-.259	.258	1.734	.205	1.596	.230	-2.750*	.013

Note. * $p < .05$. Statistically significant findings are bolded.

Table 12

Cancer-Related PTSS: Associations with sibling demographics, cancer characteristics, cancer-related appraisals and coping strategies

Characteristic	r	p	t-test	p	F-test	p
Age at diagnosis	.46*	.033				
Gender			-1.706	.103		
Time Since Diagnosis	-.28	.216				
Cancer Type					.503	.613
Treatment Status					1.620	.224
Cancer Relapse			-1.287	.213		
Cancer Appraisals						
Life threat at diagnosis			3.472**	.002		
Life threat in treatment			1.127	.273		
Coping Strategies (CSI)						
Problem Solving	.42	.056				
Cognitive Restructuring	.10	.662				
Emotional Expression	.58**	.006				
Social Support	.14	.549				
Problem Avoidance	.50*	.020				
Wishful Thinking	.59**	.005				
Self-Criticism	.55**	.010				
Social Withdrawal	.81**	<.001				

Note. * $p < .05$, ** $p < .01$. Statistically significant findings are bolded

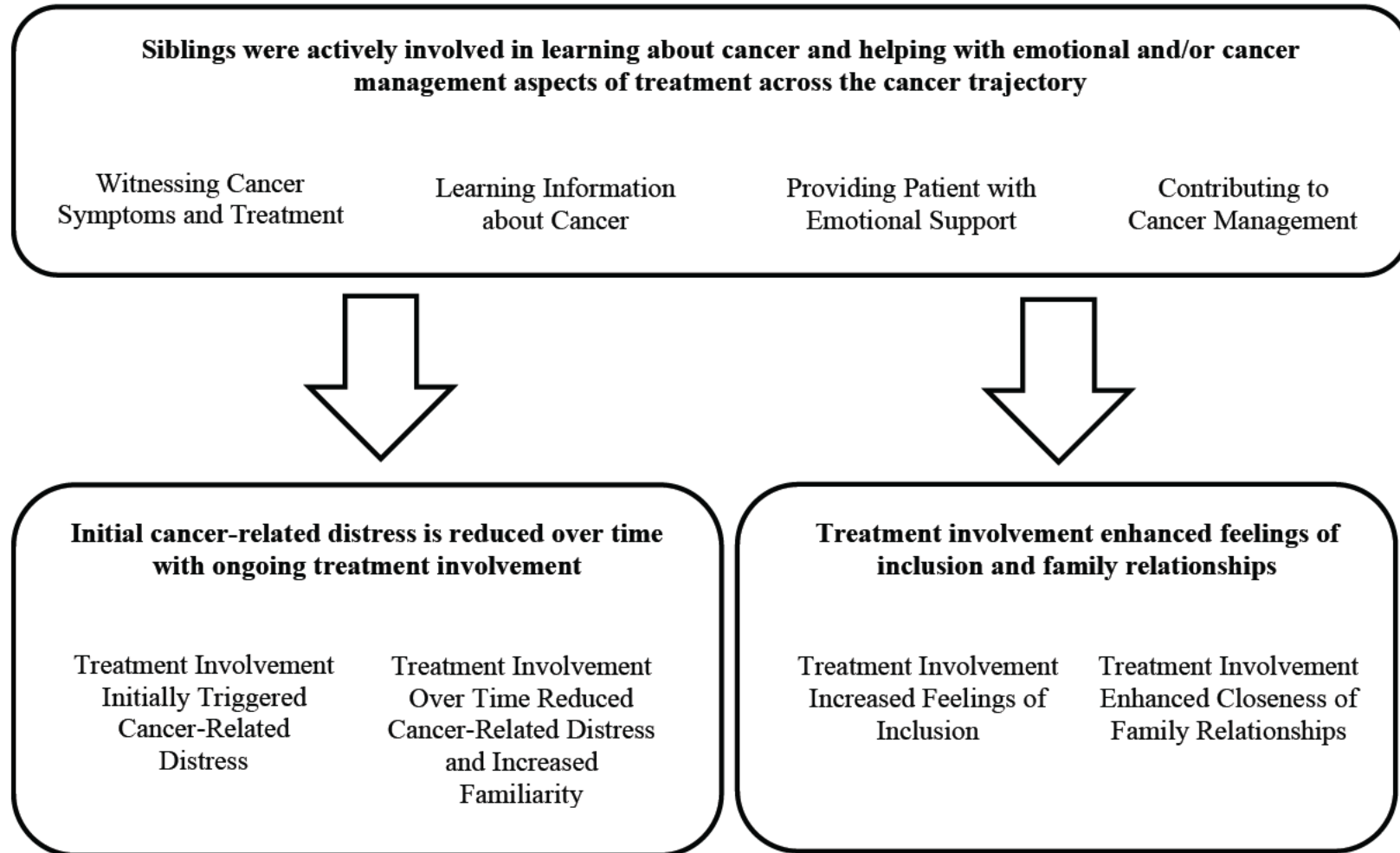
Table 13*Differences in Treatment Involvement between Low PTSS and High PTSS Groups*

Theme	Low PTSS	High PTSS
Consistency of Siblings' Treatment Involvement	Parent: "[Siblings] were uncertain as to what to say, what to do, more in shock. They weren't really able to process it, at least the first time [they went to the hospital]. The second time they were more relaxed, more willing to engage with [patient]."	Sibling: "The first time, I was nervous and scared. I didn't know what to expect since I had never been in a hospital like that. But after the first visit and the second visit, I kind of got the hang of it."
	Parent: "I didn't want to shield them from it, because I thought that would make it more scary, to not see him and know what's going on. I felt like when they got to know the nurses and doctors it made it less scary."	Parent: "I always tried to shield [siblings] off from seeing certain stuff. So I don't believe they ever saw the doctors or nurses access him, because [patient] would always cry, so I just didn't want them to see."
Alignment between Siblings' Preferred and Actual Treatment Involvement	Parent: "I always gave [siblings] a choice. I would say 'who wants to come? If you don't want to, you can stay home' and then they would do whatever. They usually came with me."	Sibling: "I didn't feel like I went [to the hospital] enough. My parents didn't want to do the driving. And I didn't have like much of a choice. That was kind of hard for me because I was like, you don't want to drive, but I also don't want to never see my family."
Access to Developmentally Appropriate Cancer-Related Information	Parent: "My husband's a doctor, so he was able to talk to [siblings]. He's used to hard conversations about health and so I feel like he was able to be open enough with them and factual. Like, 'we'll be okay, we have great care.' So I think [siblings] felt okay."	Parent: "The first time, I didn't share a lot of information with [sibling]. I had not shared [the diagnosis], although, she could see the symptoms."
	Sibling: "[Worries about patient dying] got better as I understood more about it. I was able to know that this kind of cancer is one of the easiest to cure, and like, he has the best chance of survival."	Parent: "[Other parent] had a hard time relaying information as to what was going on. I think a lot of times he chose to maybe not speak to [sibling] about it, just like '[patient] is fine, mom and him are at the hospital.'"

Opportunities to Communicate Cancer-Related Emotions	Parent: "I would tell [sibling], if he need to cry 'you can cry' and so he would cry and he'd be like 'is he gonna die? Why is this happening?' I tried to make sure [siblings] understood it's okay to be scared, you know, this is new for all of us."	Sibling: "[Parents] told me if I could just keep it all together, they would really appreciate that because they didn't want me to show [patient] that I was weak, because that would make him feel weak. So I had promised myself that I wouldn't cry, but, when I saw him, I cried. I felt so bad and just ashamed of myself."
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Figure 1.

Siblings' involvement in cancer treatment and the relative influence on their individual and interpersonal psychosocial adjustment to cancer



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Curriculum Vitae

