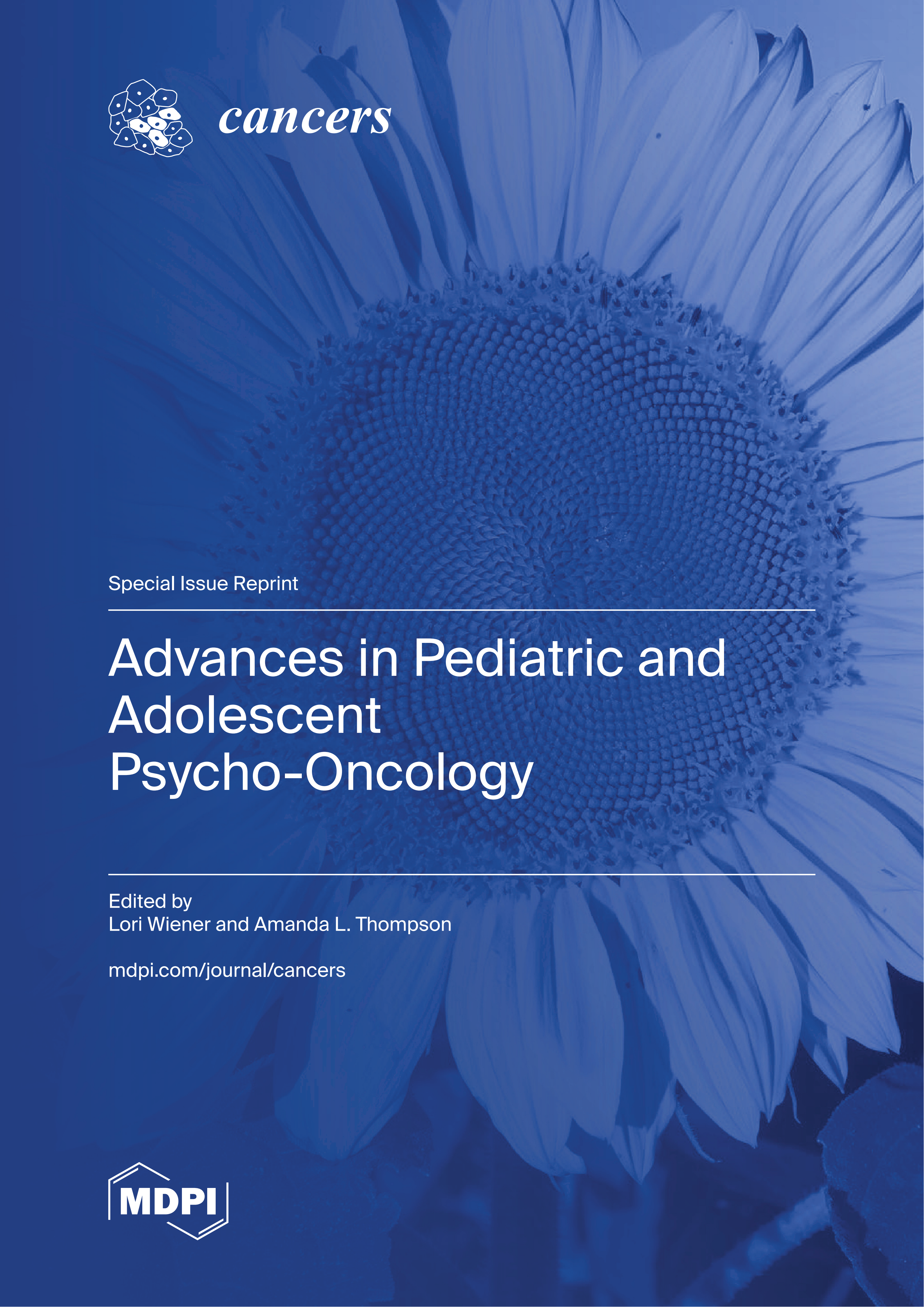

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Special Issue Reprint

Advances in Pediatric and Adolescent Psycho-Oncology

Edited by
Lori Wiener and Amanda L. Thompson

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Guest Editors

Lori Wiener
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Cover image courtesy of Lori Wiener

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About the Editors

Lori Wiener

Lori Wiener is co-director of the Behavioral Science Core and head of the Psychosocial Support and Research Program at the pediatric oncology branch of the National Cancer Institute. As both a clinician and behavioral scientist, Dr. Wiener has developed a robust clinical and research program that has addressed critical clinical issues such as parental coping, lone parenting, transnational parenting, sibling and sibling donor experiences, graft versus host disease, and end-of-life planning. Dr. Wiener has also dedicated a substantial part of her career to applying knowledge from her clinical experience and psychosocial studies to create innovative resources such as books, workbooks, therapeutic games, and an advanced care planning guide for children, adolescents, and young adults. These resources have been distributed worldwide. Dr. Wiener proudly led the team that developed the first evidence-based psychosocial standards of care for children with cancer and their family members. She has published close to 300 publications, spanning peer-reviewed papers and book chapters. Dr. Wiener has also co-edited the textbooks *Pediatric Psycho-Oncology: A Quick Reference on the Psychosocial Dimensions of Cancer Symptom Management* and *Pediatric Psychosocial Oncology: Textbook for Multi-disciplinary Care*, and she has co-authored a storybook, *The Gift of Gerbert's Feathers*, to help children through the psychological and behavioral process of preparing for a natural end of life. Dr. Wiener was recently recognized in the 2025 SIOP Almanac of Women Leaders, with a 2025 Lifetime Achievement Award through the American Pediatric Oncology Social Workers (APOSW), the 2024 Jimmie Holland Lifetime Achievement Award through the American Psychosocial Oncology Society (APOS), and a 2024 Founders Award through the Cancer Support Community. She is a Fellow of both American Psychosocial Oncology Society (APOS) and Div 54 of the Society of Pediatric Psychology, American Psychological Association.

Amanda Thompson

Amanda Thompson first served as the Director of Psychology and Psychosocial Services in the Center for Cancer and Blood Disorders at Children's National Hospital in Washington, DC, and then as Chief of Pediatric Psychology and Director of Pediatric Programs at Life with Cancer, the psychosocial program of the Inova Schar Cancer Institute in Fairfax, VA. For almost two decades, Dr. Thompson has dedicated her career as a pediatric psychologist to improving the lives of children, adolescents, and families impacted by cancer from the time of diagnosis to survivorship or through end-of-life, bereavement, and beyond. She has built world-class psychosocial teams and trained a generation of pediatric psychologists in the evidence-based care of children with life-limiting illnesses. Dr. Thompson has a national presence as an author of the Standards of Psychosocial Care for Children with Cancer and their Families and as a project lead on the Development of Competencies for Psychologists in Pediatric Palliative Care. She currently serves as Co-chair of the American Psychosocial Oncology Society's Pediatrics/AYA Special Interest Group, as the APA's representative to the Pediatrics Division of the National Coalition of Hospice and Palliative Medicine, and as a board member on the Pediatrics Council of the American Academy of Hospice and Palliative Medicine. She has published in numerous peer-reviewed journals, regularly presents at national and international scientific conferences, and frequently presents to medical providers and the wider community on topics related to palliative care and grief and loss. Dr. Thompson was the recipient of the 2024 Outstanding Clinical Care Award from the American Psychosocial Oncology Society and is a Fellow of both American Psychosocial Oncology Society (APOS) and Div 54 of the APA (Society of Pediatric Psychology).

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Editorial

Introduction to the Special Issue on Advances in Pediatric and Adolescent Psychosocial Oncology

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Psychosocial care is critical to supporting the adjustment, coping, and quality of life of children and families from the time of diagnosis throughout treatment and into survivorship or through end of life and bereavement. Now a standard of care in pediatric oncology [1], psychosocial support is considered essential for all children and families. Interprofessional team members collaborate in assessment, interventions, education, and research to advance the science and practice of pediatric psycho-oncology.

The field of pediatric and adolescent psycho-oncology continues to evolve, with new research and innovative interventions. New insights are critical for improving the overall care and long-term outcomes for patients and their families and driving future research in pediatric psycho-oncology. Within this Special Issue, recent innovations in the field are described from a clinical and scientific point of view.

Three papers in this Special Issue present data on caregivers' needs, an emerging and critical area of study that recognizes the impact of diagnosis and treatment on the entire family unit. In "*Somewhat of an Adult": Understanding the "Dance" of Competing Tensions Parents Manage While Caring for an Adolescent or Young Adult (AYA) Diagnosed with Hematologic Malignancy*" (https://www.mdpi.com/2072-6694/17/8/1299), Mullis et al. [2] describe how they conducted in-depth interviews with 20 parents. Tensions were identified regarding the "dance" of being the parent and caregiver of an AYA during the developmental stage when they are seeking more independence. The authors recommend psychosocial education that normalizes these tensions and teaches parents to navigate them in ways that enhance connection with their AYA.

In "*Psychosocial Outcomes in Parents of Children with Acute Lymphoblastic Leukaemia in Australia and New Zealand Through and Beyond Treatment*" (https://www.mdpi.com/2072-6694/17/7/1238), Parker et al. [3] present the results of a prospective longitudinal study conducted across eight sites in Australia and New Zealand. The Emotion Thermometer (ET) tool and Patient-Reported Outcome Measurement Information System (PROMIS) questionnaires were used to quantify the psychological symptoms and need for help of parents whose children had been newly diagnosed with ALL. A total of 117 parents completed 327 surveys over a period spanning 0 to 62 months post-diagnosis. Their distress peaked within the first 6 months, with 40% of parents reporting clinically significant symptoms. Anxiety was the most consistently elevated symptom, with over 50% of responses being above the clinical cut-off and a second peak around the two-year mark (i.e., the time treatment ended). Depression and the parents' need for help also peaked closer to the diagnosis and declined over time. In contrast, anger remained consistently present, with 27% reporting clinically significant scores across all the time points. While many other studies conducted outside Australia and New Zealand have found that a notable minority

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of parents continue to report elevated distress levels over time, this study identified a specific need for intervention throughout the ALL treatment trajectory.

A third study by Bemis et al. [4], "*Problem-Solving Skills Training for Parents of Children Undergoing Hematopoietic Stem Cell Transplantation*" (https://www.mdpi.com/2072-6694/17/6/930) used a mixed-methods approach to examine the feasibility and acceptability of a specific intervention to address distress in the caregivers of children receiving hematopoietic stem cell transplantation (HSCT). In an intensive HSCT setting, the study tested Bright IDEAS®, an evidence-based cognitive–behavioral problem-solving skills training intervention with demonstrated efficacy for caregivers of children newly diagnosed with cancer [5]. Caregivers were assigned Bright IDEAS® with usual care or usual care alone. The intervention involved six-to-eight sessions which empowered caregivers to manage challenges throughout transplantation. The findings suggest that Bright IDEAS® may be a promising way to help caregivers during their child's HSCT.

This Special Issue also includes four papers addressing programmatic approaches to both program development and the provision of psychosocial support within a pediatric oncology setting. Three papers specifically address the implementation of the 15 published, evidence-based Standards for Psychosocial Care for Children with Cancer and their Families. A paper by Foster et al. [6], "*Development of an Evaluation Tool for Monitoring the Delivery of Psychosocial Care in Pediatric Oncology Settings*" (https://www.mdpi.com/2072-6694/17/9/1550), described how a pediatric oncology program utilized the standards as a foundation for psychosocial program development. Ongoing gaps in care were identified, along with the need for greater progress toward achieving these standards. Reviewing and analyzing the Pediatric Psychosocial Standard of Care Institutional Assessment Tool (Matrix) [7] further highlighted the need for program development, as well as the design of institutionally specific objective measures to monitor program improvements over time. The processes used by the authors set an example for other centers interested in evaluating how efficiently their programs currently implement psychosocial standards of care.

In the paper "*An Interprofessional Approach to Developing Family Psychosocial Support Programs in a Pediatric Oncology Healthcare Setting*" (https://www.mdpi.com/2072-6694/17/8/1342), Turner et al. [8] examine the framework provided by the Standards for the Psychosocial Care for Children with Cancer and the Pediatric Psychosocial Preventative Health Model (PPPHM). Employing both the standards and the PPPHM, the authors developed a comprehensive tiered approach, with input from both parent advisors and staff, to support the psychosocial needs of families in a pediatric oncology setting. This then led to the development of over ten new programs to enhance support for families facing pediatric cancer at the Universal, Targeted, and Clinical tiers. The paper illustrates how an interdisciplinary approach that combines the expertise and strengths of diverse disciplines with the perspectives of patients and families is critical to successfully providing support throughout the treatment trajectory.

Bernstein et al. [9] also looked at ways to align psychological care with the established evidence-based Standards of Care in their paper, "*Integrated Psychological Services in Pediatric Oncology: Caregiver Perspectives at Diagnosis*" (https://www.mdpi.com/2072-6694/16/18/3137). They evaluated the New Oncology Program in Psychology (NOPP), designed to provide psychoeducation about and anticipatory guidance for coping with diagnosis and treatment. Caregivers who participated in the NOPP felt more prepared and equipped with strategies to manage difficult emotions over time than those who did not, and those who completed a cognitive assessment also felt more informed and prepared to deal with the potential effects of diagnosis and treatment on the patient's cognitive/academic functioning. The results highlight important domains for universal assessment and interventions at the time of a new cancer diagnosis and suggest that psychological services are associated with positive caregiver perceptions of feeling informed, prepared, and equipped for managing the psychosocial and cognitive impacts of disease and treatment.

Programs that combine conventional treatment with complementary therapies remain limited. In a retrospective study, "*Patient Acceptability of the First Integrative Pediatric Oncology Unit in Spain—The Pediatric Cancer Center Barcelona Experience*" (https://www.mdpi.com/2072-6694/17/2/222), Martínez García et al. [10] describe the feasibility of implementing such a program, the acceptance of the interventions, and early data on the various care activities. Acupuncture, aromatherapy, and reflexology had high acceptance rates and support the feasibility of implementing an Integrative Pediatric Oncology Unit within a patient-centered care model in a comprehensive pediatric cancer center in Spain.

The remaining papers within this Special Issue address different aspects of care. The first addresses germline genetic testing, something increasingly being integrated into pediatric oncology, with little known about its impact on the family unit. A paper by Van Hoyweghen et al., "*Family-Level Impact of Germline Genetic Testing in Childhood Cancer: A Multi Family Member Interview Analysis*" (https://www.mdpi.com/2072-6694/17/3/517) [11], describes the experience of six families who opted for germline genetic testing for a cancer predisposition. Germline genetic testing was generally viewed as a valuable and straightforward step in their child's oncology trajectory, though parents found it difficult to distinguish its impact from that of the overwhelming stressors of their child's cancer diagnosis and treatment. Several themes emerged from the interviews, included familial communication about genetic testing, differences in parental coping, feelings of guilt and forgiveness, and worries about the future health of the family. The authors suggest that proactively addressing these challenges could improve the support provided for and experience of families undergoing germline genetic testing for cancer predisposition.

A second paper addressed a unique aspect of family communication that has received limited attention—the characteristics of therapeutic parent–child communication. Son and Kim [12] examined 10 papers in their scoping review, "*Therapeutic Parent–Child Communication and Health Outcomes in the Childhood Cancer Context*" (https://www.mdpi.com/2072-6694/16/11/2152), and identified characteristics of therapeutic verbal and nonverbal communication. Positive psychological health outcomes included less distress, a lower level of PTSS, less internalization and externalization of symptoms, improved social–emotional competencies, better peer relationships, and more cooperation during the procedure at the individual level, while the family-level outcomes included increased family cohesion and adaptation. The authors noted the need for longitudinal studies to identify specific aspects of communication that predict better psychological outcomes.

It is well-known that adolescents and young adults with cancer have unique psychosocial needs that often go unmet by healthcare systems. Korenblum et al.'s study, "*Factors Affecting Psychosocial Distress in Adolescents and Young Adults with Cancer: BRIGHTLIGHT Cross-Sectional and Longitudinal Cohort Study Results*" [13], contributes to a deeper understanding of the risk of distress and protective factors in AYAs. The study examined a large cohort of patients (over the 3 years following their diagnosis) and provided a longitudinal description of their psychological outcomes, with depression improving over time but anxiety remaining stable. A unique aspect of this study was that a patient and public involvement group (Young Advisory Panel, YAP) helped to contextualize and interpret the study results during a series of online focus group discussions. Like many of the papers in this Special Issue, this study demonstrates the vital role of the participation and engagement of the patients themselves in research, as the importance of considering the patient experience cannot be overemphasized.

Fair et al. considered how to prepare youth with osteosarcoma for amputation, a topic not yet discussed in the literature. Their paper, ""*There are two healing processes in cancer* *care. There's a physical healing and a mental adaptation process": A Pilot Study for Preparing Children and Adolescents with Osteosarcoma for Limb Amputation*" [14], is based on in-depth structured interviews with nine survivors. The participants described the type of support and guidance they received before surgery, including contact with amputation-related organizations and exposure to tangible tools, such as a physical model of a knee joint. Their need for emotional support was found to be unmet, and when it was available, support from fellow amputees and surgeons was the most meaningful. This is the first paper to describe ways to provide holistic, patient-centered care throughout the amputation process.

Finally, two commentaries provide new insights into the field of pediatric and adolescent psycho-oncology. In the first, "*Leveraging the Patient and Family Voice in the Development of Patient Education: Supporting the Pediatric Oncology Experience*" (https://www.mdpi.com/2072-6694/17/7/1201), Jones et al. [15] explore how patients and their families can be actively involved in the creation, assessment, and implementation of patient education materials, fostering a collaborative partnership between families and clinicians. Detailed and concrete examples of this collaboration are provided, including the creation of patient education materials and a podcast. The commentary emphasizes the importance of partnering with patients and families to ensure that their voices remain central to developing and implementing interventions.

In the second commentary, "*Bridging the Gap: Embedding Psychosocial Oncology Research into Comprehensive Cancer Care for Children and Young People*" [16], Sansom-Daly et al. describe a rare opportunity to develop comprehensive psychosocial programming. Located in Australia, Minderoo Children's Comprehensive Cancer Centre will be the first of its kind in the southern hemisphere, providing state-of-the-science medical and psychosocial care for children and their families regardless of their background or location. Recognizing the need for integrated care and the barriers to providing it, similarly to Jones et al., the authors highlight the importance of investing in partnerships, including families in research, and reforming funding so that mental health support becomes a routine part of cancer treatment.

Collectively, the papers in this Special Issue shed light on a broad range of timely and clinically relevant topics within pediatric and adolescent psychosocial oncology. Several themes emerge across this body of work, including the centrality of caregiver support, the importance of aligning psychosocial services with established evidence-based standards, and the value of integrating diverse voices (particularly those of patients and their caregivers and siblings) into the development and delivery of psychosocial resources and interventions. These studies also underscore the unique psychosocial needs of adolescents and young adults, the evolving nature of communication and education in cancer care, and the feasibility of employing integrative care models that include complementary therapies. Clinically, this Special Issue calls for continued efforts to embed psychosocial care into the standard pediatric oncology workflow, ensuring timely access to support across the continuum of care and for all family members.

Future research should prioritize the use of longitudinal and prospective designs to better elucidate the psychosocial trajectories of patients and their families, especially as the current treatments evolve and new ones emerge. Greater inclusion of underrepresented populations and attention to vulnerable patient groups will be critical to improving equity and relevance in the field. Finally, integrating the voices of patients and their caregivers and siblings into research and clinical innovation must remain a guiding principle, ensuring that psychosocial care is responsive and rooted in lived experience. The articles in this Special Issue lay a strong foundation for this next phase of discovery, programmatic innovation, and patient- and family-centered care in pediatric and adolescent psycho-oncology.

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Conflicts of Interest: The authors declare no conflicts of interest.

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Article

"Somewhat of an Adult": Understanding the "Dance" of Competing Tensions Parents Manage While Caring for an Adolescent or Young Adult (AYA) Diagnosed with Hematologic Malignancy

M. Devyn Mullis [1], Carma L. Bylund [1], Diliara Bagautdinova [2], Emma G. Bryan [1], Maria Sae-Hau [3], Elisa S. Weiss [3], Joanne P. Lagmay [4] and Carla L. Fisher [1,*]

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Simple Summary: Parents are often the primary caregiver and source of support for their adolescent or young adult living with blood cancer. In these roles they navigate tensions or competing needs in online, clinical, and family care contexts. This experience can feel like being pulled in two directions. Parents described four tensions that illustrate being pulled in two directions simultaneously, tensions they must constantly navigate as they care for and cope with their child: (1) being the driver versus passenger in their child's care; (2) coping with cancer as a family versus individually; (3) deciding whether to reveal versus conceal information; and (4) expecting typical developmental or disease experiences for their child versus managing unexpected experiences. These findings can be used as the bedrock of resources for parents to help normalize the conflicting realities with which parents cope in their cancer caregiving role and foster health-promoting approaches to navigating these tensions.

Abstract: Background: Parents supporting AYAs with blood cancer juggle dual, competing roles as cancer caregiver and parent, which may heighten distress as they feel pulled simultaneously in two opposing directions. Likewise, AYAs encounter paradoxical needs as they revert to being more dependent on their parents to prioritize their survival while their developmental trajectory toward independence is disrupted. Parents need help understanding the underlying tensions they face in caregiving to reduce their distress and promote their connectedness with their AYA. Using a dialectical lens, we identified tensions parents encountered while caregiving in three contexts (clinical, family, and online communication) to inform a targeted psychosocial intervention. Methods: In partnership with The Leukemia & Lymphoma Society, we recruited 20 parents for in-depth interviews. Parents cared for adolescents aged 15–18 ($n = 10$) or emerging adults aged 19–29 ($n = 10$) diagnosed >3 months prior and in active treatment or within 2 years since treatment ended. Transcripts were thematically analyzed. Results: Parents described four ongoing tensions they needed to negotiate as they cared for their AYA: (1) *being the driver versus passenger in their child's care*; (2) *coping with cancer together as a family versus separately*; (3) *deciding to reveal versus conceal information*; and (4) *expecting normative developmental and disease trajectories versus disrupted trajectories*. These tensions characterize the complex caregiving "dance" parents navigate in all three care contexts. Conclusions: Psychosocial education can normalize these tensions for parents to promote healthier coping and reduce distress while enhancing connectedness with their AYA. As caregiver–patient outcomes are interrelated, it may improve AYAs' well-being.

Keywords: adolescents and young adults; parent caregivers; caregiving; blood cancer; dialectical tensions; qualitative research; family communication; healthcare communication

1. Introduction

Parents are oftentimes the primary caregiver and source of support for their adolescent or young adult (AYA) living with hematologic malignancy (blood cancer)—the most common cancer type diagnosed from childhood through young adulthood [1]. Caregiving parents of AYAs report significant levels of distress, including symptoms of post-traumatic stress disorder, and their diagnosed AYAs report similar outcomes indicating their distress is interrelated [2–4]. Parents' distress is not surprising given the threat to their child's life in addition to blood cancer caregiving being especially turbulent. In comparison to solid tumor cancers, blood cancer care demands can be characterized by immediate and long-term hospitalizations, years of or indefinite costly treatment(s), multiple treatment transitions, travel or relocation for treatment, employment and/or financial hardship, stress on the family's functioning, as well as challenging physical, psychological, and behavioral treatment side effects [5–7]. These care stressors are compounded by the trauma of an AYA facing a life-threatening disease so early in the lifespan, when they are just beginning to develop a sense of self, independence, and separation from their parent [8,9]. Like their caregiving parents, AYAs report significant psychological distress and are at a higher risk of distress, depression, and anxiety in comparison to older adults living with cancer [10].

Diagnosed AYAs often look to their parents as their primary source of support in navigating this traumatic life experience within a complex healthcare system. While supportive care resources for family caregivers in acute or chronic illness are scarce [11], caregiving support for parents is especially limited in AYA oncology care [12]. A recent review showed that family caregivers need supportive resources to help them manage distress by enhancing their caregiving ability and that caregivers especially want education on what to expect in their role and how to navigate it relationally with their diagnosed loved one [11,12].

Parents of diagnosed AYAs would benefit from a "psychosocial map" of psychosocial support and education that teaches them about the "oppositional tensions"—feeling pulled in two directions simultaneously—that they must learn to navigate while parenting and caring for an AYA with blood cancer [13,14]. A dialectical perspective in both psychotherapy practice and behavioral research highlights the importance of normalizing for individuals and families that dual or competing forces define their illness experiences and that it is important to make sense of the "parts" to navigate the "whole" [13]. This cognitive processing or sense-making should be embedded within the relational communication and care contexts in which the tensions emerge, such as online, clinical, or family communication contexts of care [13]. Studies demonstrate that when individuals can identify, acknowledge, and reconcile these tensions, they can embrace a more adaptive mindset and flexible coping approach that is tied to better emotion regulation and less distress [15,16].

Both caregiving parents and their AYAs characterize their cancer experience as fraught with paradoxes, though this research has typically centered on tensions encountered in individuals' coping experiences and not within caregiving contexts. For instance, parents describe themselves simultaneously living in fear of their child's survival while trying to be positive and hopeful, especially in their child's presence [17–19]. Likewise, AYAs describe themselves longing for their pre-cancer self while grappling with their identity as patient or survivor taking center stage [20]. Research informed by a relational dialectics theoretical approach (RDT) has helped unveil and normalize the conflicting nature of illness caregiving and coping at the relational level as caregivers and patients co-navigate multiple and, at times, competing roles (e.g., cancer caregiver versus parent of an AYA; patient versus son/daughter) as they take on the illness together. These studies have highlighted the contradictory nature of living with various types of cancers by identifying common discursive struggles—referred to as dialectical tensions—that parents and family members often encounter, like dualities of autonomy–control, openness–closedness, connection–separation, and the individual–collective [21–26].

In using a dialectical approach, we can help normalize the distinct paradoxical experience of AYA caregiving for both parents and AYAs—evidence that can become the bedrock of future interventions. Psychosocial interventions developed or adapted for specific types of caregivers are needed to reduce their psychological distress by providing them with a roadmap of targeted guidance for their unique circumstances, like the disease type (e.g., blood cancers) and relationship type (e.g., parent–child), as well as the phase of the lifespan in which cancer is diagnosed (e.g., AYA) [27,28]. A recent cancer caregiver intervention (*Healthy Communication Practice*) addresses and normalizes common tensions that emerge in the parent–child bond (e.g., independence–dependence) to help adult child caregivers develop communication skills central to their caregiving role. This is done in three communication contexts in which they engage in caregiving: online, clinical, and family care contexts [29–31]. The online intervention was both feasible and acceptable for adult child caregivers of diagnosed aging parents living with a blood cancer [29], while also effectively reducing caregivers' distress and improving their communication skills [31]. To adapt and target this intervention for parents of AYAs, we aimed to capture the tensions parents encounter in their role as a caregiver to their AYA living with a blood cancer in each care context to help parents identify, acknowledge, and reconcile these tensions that can complicate their role in the online, clinical, and family communication contexts of caregiving.

2. Materials and Methods

2.1. Sample

In partnership with The Leukemia & Lymphoma Society (LLS), parents caring for an AYA were recruited via email through LLS's constituent database to participate in an individual, in-depth interview. Inclusion criteria were (1) a parent caring for adolescent aged 15–18 or emerging adult aged 19–29 diagnosed >3 months prior with a blood cancer in active treatment or within 2 years since treatment ended, and (2) English-speaking. Interested parents could contact a research coordinator via a link in the email to screen for eligibility and schedule an interview.

2.2. Procedures

In-depth, semi-structured interviews with parent caregivers were conducted and audio-recorded (July–September 2024). The interview guide was previously created and tested in formative research to develop the *Healthy Communication Practice* intervention and then adapted and expanded upon to target the parent caregiver of a diagnosed AYA for intervention adaptation to provide targeted guidance for communication skill development in three communication contexts of care (online, clinical, and family communication) (see [32]). C.L.F. and M.D.M. led the development of the guide, which involved experts in family communication (C.L.F.; M.D.M.; D.B.; C.L.B.), clinical communication (C.L.F.; C.L.B.; D.B.), online communication (C.L.B.), cancer caregiving (C.L.B.; C.L.F.), and blood cancer (E.W.; M.S.; J.P.L.). Participants were asked about various caregiving experiences (e.g., diagnosis, clinical communication, family support). In each communication context, parents were asked about their role, challenges faced, as well as coping responses (e.g., *How do you see your role at your child's clinical appointments? What challenges have you encountered in your caregiving role at your child's clinical appointments? How have you managed this?)*. No direct questions were asked about dialectical tensions (e.g., control–autonomy), which emerged naturally in parents' characterizations of their experiences. On average, interviews lasted 92 min (ranging 25–200 min). Transcriptions resulted in 472 single-spaced pages of data. Participants were compensated with a $50 gift card.

2.3. Data Analysis

A thematic analysis was conducted using a constant comparative method (CCM) approach [33] with RDT as a sensitizing framework to capture the dialectical tensions parents encounter in three caregiving contexts (online, clinical, and family communication). To identify tensions, the analysis was segmented or kept separate for each communication care context to compare emergent tensions in each context. ATLAS.ti 24.1.1 was used to manage the data. A multi-coder approach and triangulation of stakeholder perspectives (i.e., parents caring for EA versus adolescent) were used to promote rigor in analysis. The lead author and qualitative analyst with expertise in RDT (MDM) led the analysis by inductively analyzing all transcripts. The senior author and lead qualitative expert on the team, also with expertise in RDT (CLF), served as a second coder to validate analysis and co-develop a codebook over the course of multiple consensus meetings. The CCM steps were followed in analysis: (1) open coding to find patterns using contextual labels; (2) collapsing emergent patterns to develop themes; and (3) axial coding to further define themes and identify thematic properties using RDT typologies to further define tensions (e.g., control–autonomy). The extent of thematic saturation was identified using Owen's criteria of repetition, recurrence, and forcefulness [34]. Moreover, the sample size (n = 20) exceeds sample sizes used in studies of the same design where saturation is reached, demonstrating extended rigor in this project [35]. To capture similarities and differences in parents' experiences, transcripts for parents of adolescents were kept separate from emerging adults, and typologies were later collapsed given significant overlap in parents' experiences.

3. Results

Twenty parents participated and were mostly mothers (95%) caring for either a son (75%) or daughter (25%), with half in emerging adulthood (19–29) and half in adolescence (15–18). The majority of their AYAs were diagnosed with a leukemia (75%). Most parents were non-Hispanic white (80%), with 50% working full-time. See Table 1 for full demographics.

Parents described four ongoing dialectical tensions—feeling pulled in two directions simultaneously—as they cared for their AYA: (1) *being the driver versus passenger in their child's care*; (2) *coping with cancer together as a family versus separately*; (3) *weighing whether to reveal versus conceal information;* and (4) *expecting normative developmental and disease trajectories versus disrupted trajectories*. These tensions illustrate the complex caregiving "dance" parents must learn to manage in all three care contexts, particularly clinical and family communication contexts of care. Each tension is labeled within the AYA oncology care context and then defined using common RDT constructs that identify the opposing yet unified forces (e.g., control–autonomy). These are also underlined to further illustrate the tension parents face in their AYA oncology care role. Each tension is then depicted in each caregiving context (e.g., online, clinical or family communication) in which parents described the tension using their lived narratives to more comprehensively capture the dialectical pulls parents negotiate across different contexts of care. A translational table (Table 2) also presents the typology of tensions using action statements, a technique that facilitates the translation of findings into practice and interventions to provide supportive care to parents [36,37].

Table 1. Parent caregiver and AYA patient demographics.

Characteristic	*n* (Percent)	Mean (SD)	Min-Max
Age			
Caregiver		52 (6.76)	38-69
Patient Age at Diagnosis		19.9 (3.99)	15-28
Patient Developmental Group			
Adolescent	10 (50%)		
Emerging Adulthood	10 (50%)		
Caregiver Gender			
Female	19 (95%)		
Male	1 (5%)		
Patient Gender			
Female	5 (25%)		
Male	15 (75%)		
Caregiver Race/Ethnicity			
White, non-Hispanic	16 (80%)		
White, Hispanic/Latino	2 (10%)		
African American, non-Hispanic	1 (5%)		
African American, Hispanic/Latino	1 (5%)		
Caregiver Employment Status			
Employed, Full-Time	10 (50%)		
Employed Part-Time	5 (25%)		
Not Employed/Retired	5 (25%)		
Patient Diagnosis [1]			
Leukemia	15 (75%)		
Lymphoma	5 (25%)		
Patient Diagnosis Subtype			
Acute Myeloid Leukemia	4 (20%)		
Acute Lymphoblastic Leukemia	9 (45%)		
T-Cell Leukemia	1 (5%)		
Hodgkin's Lymphoma	4 (20%)		
Non-Hodgkin's Lymphoma	1 (5%)		
Diffuse Large B-Cell Lymphoma	1 (5%)		

[1] One AYA was diagnosed with both leukemia and lymphoma.

Table 2. Tensions parents encounter while caring for their AYA with blood cancer.

Caregiving Parents of AYAs Need Help Managing These Four Tensions in Which They Feel Simultaneously Pulled in Opposing Directions:	Parents Experience the Tension in Multiple Care Communication Contexts	As Each Narrative Illustrates:
being the driver vs. a passenger in their child's care (a tension of control–autonomy)	online	I tried to not let him get online very often, which is very hard for a 16 to 17-year-old who's got a phone. . . . We told him, "You're an adult almost. You need to know the answers."
	clinical	We have to be like, "Yes. I would love to be involved in everything." However, she is a young adult, and I have to respect that choice.
	family	[You] just accept . . . the fact that she's in charge and give her all the support that you can possibly give, even if you don't agree with it. Now, obviously if that's detrimental to her well-being, you got to say something or step in! If [doctors] say, "Don't eat sushi," and [son] goes, "I want sushi, damn it. And go get it for me." No, you got to draw that line.
coping with cancer together as a family vs. separately/individually (tensions of presence–absence and individual–collective)	online	Occasionally approaching it: "Here, this may be something you want to know about." It's hard to find that balance because it depends on how everybody is physically and mentally feeling, so finding the appropriate moment to do so is challenging.
	clinical	He's ready to be done with this whole, with the hospital setting stuff. So, he might rush it more [with clinicians], whereas I might say, "But can you talk to him more about whatever?"
	family	There's times I know she wants us there. I know she doesn't like when we've come home, but she'll even say, "You guys go home. You got to go take care of this, . . . cut the grass and you have the dog and you have the cat and you have your meetings." . . . But you know, she's not happy when we're not there.
weighing whether to reveal vs. conceal information (a tension of openness–closedness)	online	It's kind of a two-way street trying not to share things that are going to be unnecessary or overly negative and not helpful, but in the same time when there is something having an open conversation of, "We found this. You're welcome to go here to look at it for yourself."
	clinical	He ripped his port bandage [golfing]. . . . Instead of saying, . . . "Maybe it was from sweat." No, [we should tell the doctor that] he was golfing. . . . He's like, "Mom! It looks like I'm being irresponsible." Well, they just need to know. I don't know. So, being too honest [with the clinicians] has created tension.
	family	For example, my daughter is very much of a - I just need to know what you know [personality]. [But] I think maybe it's hard for her to hear some of the rougher details of, okay [her brother] had diarrhea today or he threw up or he's [having] fevers.
expecting/wanting normative developmental and disease trajectories vs. facing disrupted trajectories (tensions of typical–atypical and stability–instability)	clinical	Young adults are not supposed to be going through these things, . . . [and] it's not something you should go into blind. And for somebody like him, his level of education and his mentality, he would not have been able to . . . figure it all out on his own.
	family	We hate it for them because they're newlyweds, right? And you want your newlyweds to go thrive and go on and live your life. . . . [But] he just needs to take care of himself right now he's not focused on anything else but that so that's hard to watch.

3.1. Being the Driver Versus Passenger in Their Child's Cancer Care

Parents described a paradox of being the leader (i.e., driver) of their AYA's care as their caregiver, while also functioning as a passenger, albeit sometimes reluctantly. At times, parents were (or wanted to be) in control of their child's care. Yet, concurrently parents wanted to promote (or needed to respect) their AYA's autonomy in their decision making. Being a driver versus a passenger in their child's care was especially complex given their child's developmental phase. As one parent described it, "It's a dance. . . . He's somewhat of an adult" (1).

3.1.1. Online

A parent explained how this tension characterized her experience with online information management and sharing. While she wanted to control her AYA's access to online information, at the same time, she acknowledged being a passenger in that experience given her son had a smartphone. Even though she tried to limit her AYA's time online, she also promised to respect his autonomy by letting him set the tone for his information needs:

> I tried to Inot let him get online very often, which is very hard for a 16 to 17-year-old who's got a phone. . . . We were very honest with him about everything. We told him, "You're an adult almost. You need to know the answers. We're never going to lie to you. If you have questions, we're going to try and find the answer together." (8)

3.1.2. Clinical

While parents often wanted to be the driver of clinical interactions to advocate for their AYA as their caregiver, they also described the importance of their AYA's need for autonomy from their parent in their healthcare management. This tension, at times, created a tug and pull dynamic during medical encounters:

> There's some things that I want to push to have addressed [in the appointment] and [my daughter's] like, "No! I don't want you to do that." That does occur and again—it's walking that fine line of advocating but not [being] overbearing. (20)

Parents described how their AYA's developmental phase or maturity played a role in this paradox:

> He's a typical teenager or young adult. But it's fine. He is very responsible and has had to become extremely responsible in this whole process over this past year. But yet, he is still a young adult. So, I still need to ask him for some reminders or make sure he goes in and checks to let us know what's coming up next. (1)

This tension could sometimes be further complicated when parents wanted to be included in their AYA's care discussions and have some control, which clinicians did not always support, positioning the parent as a passenger:

> I got weird vibes from the doctors, and I felt like I was super mindful of I got to remember [my daughter] is an adult. She asks the questions. . . . I was so mindful not to over-talk or I would literally sometimes kind of raise my hand and be like, "Is now a good time to ask a question?" But it kind of rubbed me the wrong way during some of our conversations. There were certain doctors that wouldn't even look at me! They would look directly at [my daughter]. As a family, we had so many questions. . . . Some of [the doctors] were looking at us like, "This is a package deal. . . . This is Mom, and she's a worried mom. I'll have a conversation with her." But, when you have a young adult, especially when

> you're in a children's hospital, they're not used to dealing with young adults as much. I felt like I was ousted as a parent, and I didn't have the support. (13)

3.1.3. Family

Parents shared similar tensions of autonomy–control with health-related decisions made within their family environment. Parents wanted to make decisions that prioritized their AYA's physical health or survival but also wanted to respect or promote their AYA's autonomy as "the captain" of decision making. This meant parents sometimes reluctantly took on the passenger role in decision making:

> [My daughter] says, "I want to go see the Barbie movie." Her doctor said, "Well, if you're going to go try to go like maybe during the day when it's not as many people in the theater and make sure you wear the mask and do all that." I would have said, "The movie's going to be here for a while. It'll probably eventually be on Netflix or something. Is it worth waiting? Just at least wait till after those hundred days!" . . . We suggested that [and she] maybe thought about it for a couple of minutes. [Daughter said] "Nope." Okay, we'll go Wednesday for the matinee. Maybe it's not as crowded. . . . They'll all have their masks on and I'll be careful. They know if any of them don't feel well that day or the day before, they're not coming. . . . Realizing, accepting, and welcoming the fact that she's the captain. (6)

3.2. Coping with Cancer Together as a Family Versus Separately

Parents were torn between wanting to cope together as a family and the realities of coping apart from one another as individuals. This paradox manifested in two ways. Parents wanted to be present and communally cope with cancer together but circumstances forced family members to be absent and cope apart. Parents also shared instances in which individual coping needs conflicted with the collective family's ability to cope.

3.2.1. Online

One parent described this tension emerging with online cancer information. They wanted to manage information as a family. However, coping together with the overwhelming and often negative nature of online information as a collective family unit could be counterproductive to an individual family member's coping needs in that moment:

> Occasionally [we are] approaching it with the appropriate frame of mind, both for us and her, of being able to take that information in without it being preachy or judgey, but an open [approach like], "Here, this may be something you want to know about." It's hard to find that balance because it depends on how everybody is physically and mentally feeling, so finding the appropriate moment to do so is challenging. (20)

3.2.2. Clinical

As a cancer caregiver, parents wanted to be present in clinical settings to support their child but for some discussions, as a parent, they wished they were absent. These included encounters about issues that parents and AYAs would not typically have discussed, like their AYA's fertility and sexual behavior. They wanted to be present to provide caregiving support, yet also absent to protect their child's privacy or reduce discomfort:

> I didn't feel awkward about [talking about sperm banking], but I felt for him because he even said, "That was really embarrassing." When he's going in that room and doing that, [it] is super embarrassing. I felt for him because it is! What

> kid does that? And they wanted me to ask him some private questions and I'm like, "I think he probably has it under control." (10)

Parents also shared a coping paradox when individual approaches to clinical information collided and took precedence, thereby inhibiting communal coping. Parents and AYAs at times differed in the amount of information desired during clinical encounters. This became more complicated during the COVID-19 pandemic:

> I'm definitely an information seeker. ... This was also during COVID ... so [during] that first phase of him being in the hospital, we weren't allowed to visit him. It was very hard. That added a layer of complexity of trying to get information to talk to people because it was only when the doctors would come in and he would FaceTime us and so it was about five minutes a day that we had time to actually talk to people there. Otherwise, I was reaching out to the nurses to check on him to find out what was going on to get more in-depth information [because my son] was good in communication, but he didn't want to know or didn't care about the details—the level of details that I wanted to know. (4)

3.2.3. Family

In the family setting, parents described multiple ways the tension between coping together and apart emerged, both as an individual–collective tension and absence–presence tension. For instance, at times, parents' desire for communal coping clashed with individual coping responses. In such instances, they prioritized their AYA's individual needs over others':

> It was more so me breaking down than [my son] did. His attitude towards this really made this situation. ... I wanted to do the whole breakdown thing, the pity party, ... the "Why is this happening?" ... So, for me, it was more so trying not to break down in front of him and trying not to feel because he had the right attitude. I just needed to get on board. (14)

Additionally, parents shared that because they needed to be present and cope together with their AYA as their parent and primary caregiver, this competed with other roles, like being absent from parenting their other children:

> I remember just feeling resentful that I had turned my life upside down and moved, and I did not have time with my [other] daughter, and she was struggling. So, there were times when I was listening for is this a time that I can go home and be a mother to my daughter? Is it safe for me to do that? Which of course I didn't ask directly because [my diagnosed son] needed me to be there. He did not need to be responsible for my experience. (17)

Finally, parents wanted to cope as a family by being present together, yet, physically could not due to lost shared time: "We were running like separate, almost separate lives. Like if I was at work, [spouse] was at the hospital. If he was at work, I was at the hospital" (2). When a parent and AYA had to relocate for treatment, this tension was further challenging and fraught with emotional and physical distance as they struggled to connect as a family system:

> When we're together, we don't really have a family life right now. ... The last time we did something together, all four of us, was right before [older son] left. We spent three days together, and it was horribly awkward because the boys don't know how to act around each other, because they haven't been together, and they're 15 months apart! And my husband and I are like best friends who happen to be married. We don't talk about finances. We don't talk about normal

> marriage things. He just knows he has his role. I have my role, and let's not burden each other. . . . It's affected everything. I mean, we're not a four-part family anymore until maintenance [therapy] maybe hits because we're so distant. There's so much distance, physical distance in between us. Yes, we make the phone calls every day, but it's just different. (9)

3.3. Weighing Whether to Reveal Versus Conceal Information

Parents wanted to reveal or share disease-related information with their child and, thus, be open, to protect their AYA's physical health and promote their long-term survival. Concurrently, parents wanted to withhold distressing information, or be closed, to protect their child's current psychological well-being. Parents reflected on a paradox between prioritizing their AYA's physical versus psychological health needs.

3.3.1. Online

Parents struggled with wanting to openly share online information they perceived as potentially powerful knowledge in their fight against cancer. Still, information was also perceived as potentially psychologically harmful by inducing fear or sadness. Thus, parents also wanted to conceal information to buffer their child from distress:

> It's kind of a two-way street—trying not to share things that are going to be unnecessary or overly negative and not helpful but, in the same time, when there is something, having an open conversation of, "We found this. You're welcome to go here to look at it for yourself." It's just one of the things that we're keeping that communication open. It's her life. It's her health. We just want to be here to support and help guide. (20)

3.3.2. Clinical

In clinical settings, parents encountered a similar tension of wanting to share information with the clinician to better ensure their AYA received quality care. In contrast, AYAs wanted their parent to withhold information they viewed as private or vulnerable or that disclosure was not necessary or pertinent. This information was related to physical symptoms, mental health issues, logistical challenges, and personal matters:

> I think the one time I did irritate [my son] was when he went on his honeymoon, and I mentioned that he was going out of the country. He wasn't going to bring that up! [I said to him] "Are you kidding? Are you kidding me? That's kind of an important thing that you're leaving the country! I'm nervous for you!" (4)

3.3.3. Family

Within the family environment, parents worked to keep communication open with their child to provide emotional support as their primary caregiver. However, parents' openness competed with AYAs' avoidance, which parents attributed to their developmental phase in life:

> I think [son] and I have been through something together that's really important. I would not say that we're closer. . . . He's just kind of got his head down right now getting through the mental health aspect of remission, but he is holding a lot of things closed right now and not sharing a lot. He is seeing a social worker, so some of that is cancer, but . . . [he's a] 19-year-old too because who wants your mother in every aspect of your life when you're in college? (17)

Parents also shared how the reveal–conceal tension emerged in their communication with other family members. They wanted to be open with family about their AYA's cancer experience to obtain support themselves, but their openness was sometimes met with closed responses from family resistant to hearing the disclosures:

> My one sister pretty much let me down, but she apologized later. She went dark on me for a while. I know one time we were in the hospital and there was a baby, like a one-year-old. He was by himself. He was crying nonstop across the hall, and I asked the nurses, like, can I just go hold him? Like, he's alone. He's here. He's scared and I couldn't because of COVID. And I tell that story and my oldest sister then lashes out at me, like, "Quit telling us these horrible stories." It made me feel so alone. That's when I learned I could tell my mom things, but not them. (10)

3.4. Expecting Normative Developmental and Disease Trajectories Versus Disrupted Trajectories

Parents described two related paradoxes that evolved from disrupted expectations. Parents and their AYA struggled with trying to have a typical developmental trajectory, or what would be expected as an adolescent/young adult, versus the reality of being an AYA with cancer—an unexpected life event and atypical experience early in the lifespan. Parents also struggled with disrupted expectations of the cancer trajectory: they encountered a tension of wanting/expecting stability as their AYA transitioned to survivorship while unexpectedly facing ongoing instability. Parents described these tensions characterizing their clinical and family communication experiences.

3.4.1. Clinical

In clinical settings, parents described the conundrum of an AYA not having the typical maturity needed to navigate oncology care while needing their child to be more mature because of their diagnosis:

> He's a young adult. You know, young adults are not supposed to be going through these things! [But] I mean, you could get blown away by the things that could be provided and done to you without proper education and without asking proper questions. It's not something you should go into blind. (5)

Parents wanted their AYA to be more mature to promote their survival during this atypical experience, yet, recognized their ability was limited given their age: "His using his voice is so important but, at the same time, on the exact same note, a 17-year-old boy can only get to do so much" (9).

Additionally, parents shared how clinically they expected stability in their AYA's health status once they ended active treatment or entered survivorship. However, their cancer trajectory expectations were disrupted and their expectation for stability was met with ongoing instability and an uncertain future:

> Everyone thinks it's just a switch. I thought the same thing. Treatment will be over. We get to resume normal life like it never happened. . . . That's not the way it is. Secondary cancers can come from all [the treatment]. And [there are] other things that we have to constantly be on the lookout for and all the organ damage. (10)

3.4.2. Family

Parents also wanted their AYA to have typical socioemotional experiences as a developing adolescent or emerging adult, but developmental milestones were disrupted due to the cancer diagnosis (e.g., "He's missed most of his high school experience."). While parents wanted their AYA to have typical social experiences like time with peers, that desire sometimes conflicted with prioritizing their AYA's survival. A parent explained this paradox regarding her newly married son who was now living with them: "You want your newlyweds to go thrive and go on and live your life! . . . That's the hardest part to watch. . . . He just needs to take care of himself right now. He's not focused on anything else" (4).

Parents also shared that sometimes they needed to encourage their AYA to engage in typical social activities to promote their ability to have some normative experiences in the midst of their atypical reality of living with a blood cancer and loss of other typical developmental milestones:

> Four of his friends on spring break went on a guy's trip. I kind of forced him to go. He didn't want to go. He just kept talking himself out of it. But I forced him. I was paranoid, too, at first! I can't let him go six hours away without me! . . . [He went and] I was on the phone with him when he needed it. . . . It was such a great thing for him to do! He managed all the symptoms. . . . [Over the phone] I didn't talk about cancer once or how he was feeling—nothing. I just wanted to hear what they were doing. I took the focus away from that. In the meantime, . . . there was a lot of drama with his school. He just wanted to walk at graduation with his friends. He didn't have enough credits because he was behind. . . . They wanted him to finish all these assignments the last two weeks before graduation. There was no way he could do it. . . . He was managing all this mental and emotional pain that he was going through, and then they tried to give him all Fs in his classes. They caused so much other stress and pain in our life. . . . I finally got them to remove the Fs, and I withdrew him, so it didn't count against him. But they didn't let him go walk at graduation. . . . He had a pretty good summer. . . . They did have one more guy's trip in August. . . . That, again, was awesome. A really great experience for him. . . . It was nice to see him doing normal things. But when he got home, he did tell me, he said one day I had to sleep the whole day. I was so exhausted. He tried to keep up with his friends, but he can't. (10)

As parents helped their AYA navigate the ongoing disruption to their developmental trajectory, parents described how their AYA faced ongoing instability even during transitions in which they expected stability to resurface. As their AYA transitioned to survivorship after active treatment, they shared how their expectations were disrupted yet again, thereby prolonging instability:

> I think a challenge is that you expect, well, now [son's] home, and he's in remission and he's all better. And it's a challenge to know that he is not better. He's not 100%. He still is weak. . . . I think that's a challenge—just knowing that he looks good on the outside. He's going to tell you he's good. You have to know that his body is still really recovering. . . . We're not able to do everything that you used to do. (2)

4. Discussion

Parent caregivers of AYAs diagnosed with a blood cancer constantly feel pulled in two, opposing directions as they navigate their cancer caregiving and parenting roles in online, clinical, and family care contexts. These tensions illustrate the difficult dialectical "dance" parents must learn to navigate after their child's cancer diagnosis as they weigh dual, conflicting perspectives that co-exist within a given caregiving situation. Collectively, these tensions demonstrate that parents' priorities as a cancer caregiver (i.e., helping their child survive cancer and receive quality care) can compete with their priorities as a parent (i.e., nurturing and parenting their child to have typical human development including shielding them from distress).

Dialectical tensions experienced by caregivers can be informed by the type of caregiving relationship or relational dynamics (e.g., parent of a child/intergenerational relationship), type of disease (e.g., blood cancer), and developmental context (e.g., AYA) [10,19,38–42]. In particular, parents of diagnosed AYAs struggle with dilemmas of *control and autonomy* in their child's care, wanting to lead clinical conversations while also wanting to respect and foster their son/daughter's growing independence as their parent. They also are challenged by tensions of *presence and absence* in their *individual versus communal* coping with cancer that is, at times, determined by caregiving demands, which can separate family relationships emotionally and physically. Parents also constantly balance *openness with closedness*, as they frequently make decisions about sharing or withholding information and the benefits to their child's cancer care versus the risk to their emotional state. At the same time, parents are in a fight for the *typical versus atypical* or *stability versus instability* as they long for typical age-related experiences for their child with peers and education, while wanting to prioritize their child's survival in both care and parenting decision making.

Results demonstrate that parents encounter and navigate the tug and pull of these challenging forces across online, clinical, and family care contexts, indicating the consistent nature of tension in all facets of caring for an AYA with blood cancer. It is noteworthy that these tensions can emerge in each context and, yet, parents' management of each tension may manifest differently depending on the communication context and parenting or care goals. For instance, while parents want to be open in clinical communication encounters to promote quality care and survivorship (a caregiving goal), they may be less open in sharing online information about cancer to buffer their child from distress (possibly a parenting goal). At home, parents may want more openness than their adolescent or young adult child is willing to give (for both parenting and caregiving motivations), as their AYA engages in more avoidant behavior with their parent relationally at this phase of the lifespan.

Collectively, these findings can be used in developing psychoeducation and supportive care resources in AYA oncology. For instance, psychotherapists in private practice and oncology settings (e.g., psychologists, mental health counselors, licensed clinical social workers) may utilize a dialectical behavioral therapeutic approach that brings awareness to the tensions identified in this study and normalizes the experience of feeling simultaneously pulled in two directions when caring for their AYA. Programs for individuals and families can be developed that incorporate DBT skills for emotional coping and communication (e.g., see [43]) and that normalize parents' opposing feelings—that it is possible to think, feel, and act in more than just one way—as their child's parent and caregiver [44]. Supportive groups or individual therapy could also incorporate reflective written activities (like a caregiver workbook [44]) to help parents process their feelings related to each side of the tension, which can be used to talk through constructive ways to adaptively navigate them. Furthermore, online self-paced caregiver tools, like the *Healthy Communication Practice*, can be adapted to incorporate these findings and used by caregivers on their own (or in conjunction with supportive cancer care in AYA programs) to help parents develop communication skills for navigating the tensions in online, clinical, and family care contexts—skills that will also enhance their decision making [29]. These resources are imperative as evidenced in psychotherapy-focused research and psychosocial coping research, both of which have both demonstrated better coping and health outcomes when individuals facing traumatic experiences like cancer better understand the dual forces that define their illness experience [14–16]. In understanding these tensions and normalizing them, studies show that caregivers, family members, and patients can adopt more flexibility in how they cope with illness and care-related stressors, which promotes better health outcomes like emotion regulation, reduced distress, and better relational functioning [16].

Findings can also be used in medical education for clinicians to educate them on the challenges that parents are grappling with so that they can better understand these dualling dynamics during clinical encounters. A dialectical framework has been argued for in nursing practice to help clinicians make sense of clinically complex experiences when more than one contradictory view exists within a situation [38–40]. In knowing the tensions parents are managing, clinicians can better equip parents in their caregiving role and, at the same time, better approach challenging situations that may arise within clinical encounters and medical decision making in triadic clinical communication (between AYA–parent–clinician).

Limitations

Parents were mostly White mothers; further research with racially and ethnically diverse parent populations is needed to fully address differences related to culture that may inform parent–child dynamics and relational expectations. Furthermore, while mothers often take on primary caregiving responsibilities for a child with cancer, fathers' voices are imperative to capture, as are dyadic perspectives. A recent study indicated that recruitment materials that include father-targeted materials versus only parent-targeted may enhance recruitment of caregiving fathers [45]. Additionally, given the financial hardship families face and disruption to their lives separate from cancer (e.g., having to relocate, leave jobs, pay for lengthy treatments), families representing diverse socioeconomic backgrounds should also be heard from to provided targeted guidance. Partnerships with targeted recruitment with oncology social workers may enhance the ability to identify families facing such hardships. Future research should also include the diagnosed AYA's perspectives to triangulate parent–child findings and broaden our understanding of dialectical tensions that occur within this relationship.

5. Conclusions

Parent caregivers of AYAs diagnosed with a blood cancer must navigate tensions or competing needs in online, clinical, and family care contexts. Psychosocial education and interventions that teach parents about and normalize these tensions may help them manage them in ways that promote supportive parent–child connections. Considering caregiver–patient outcomes are interrelated [2–4], such educational resources may contribute to the reduction of distress for both the parent and their diagnosed AYA.

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Article

Psychosocial Outcomes in Parents of Children with Acute Lymphoblastic Leukaemia in Australia and New Zealand Through and Beyond Treatment

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Simple Summary: Over recent decades, outcomes for children with acute lymphoblastic leukaemia (ALL) have significantly improved. However, parents often experience distress and psychological challenges throughout their child's treatment. Our study explores the levels of distress, anxiety, depression, anger and the need for help among parents in Australia and New Zealand who have a child diagnosed with ALL. We enrolled parents prospectively and had them complete surveys at pre-defined time points to track their psychological experiences across the patient journey.

Abstract: Background/Objectives: Parents of children with acute lymphoblastic leukaemia (ALL) experience emotional distress throughout their child's treatment course. This study describes the psychological experience of Australian and New Zealand parents of children diagnosed with ALL. **Methods:** This prospective, longitudinal study assessed distress, anxiety, depression, anger and the need for help in parents of children with newly diagnosed ALL across eight sites between October 2018 and November 2022. Psychological symptoms were quantified using the Emotion Thermometer (ET) tool and Patient-Reported Outcome Measurement Information System (PROMIS) questionnaires. Participants were recruited within ninety days of chemotherapy commencement, with surveys distributed bimonthly thereafter. **Results:** One hundred and seventeen participants completed 327 survey responses spanning 0 to 62 months post-diagnosis. Parental distress peaked within the first

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6 months and 40% of parents reported clinically significant symptoms across four or more domains as measured by our questionnaires. Anxiety was the most consistently elevated symptom, with over 50% of responses above the clinical cut-off. Depression and the need for help also peaked closer to diagnosis and declined over time. In contrast, anger remained stable, with 27% reporting clinically significant scores across all time points. Increased time since diagnosis was significantly associated with reductions in distress, anxiety and depression scores. **Conclusions:** Australian and New Zealand parents experience high levels of psychological distress within the first six months following their child's diagnosis of ALL. A notable minority continue to report elevated distress levels over time, identifying a need for improved psychological support for family wellbeing throughout the ALL treatment trajectory.

Keywords: childhood acute lymphoblastic leukaemia; parental distress; health-related quality of life; psychosocial

1. Introduction

Significant advances in treating paediatric patients with acute lymphoblastic leukaemia (ALL) have led to a five-year overall survival rate of $\geq$ 90% in high and middle-income countries [1,2]. Therefore, there is growing emphasis on reducing morbidity associated with this condition, which includes a need to explore wellbeing for the entire family unit. Among these efforts, addressing the psychological and emotional outcomes of parents and caregivers of children with ALL is a key area of focus.

Parents and caregivers of children with cancer, including ALL, experience heightened levels of distress compared to the general population [3,4]. The treatment process often involves prolonged hospitalizations and invasive procedures and there is a high burden on parents who often deliver daily medications and monitor side effects. Parental function plays a pivotal role in the child's experience of cancer treatment, and the bidirectional relationship between parent and child wellbeing has been well described [5–7]. The distress and emotional burden on parents has been shown to negatively impact the quality of life for both parents themselves as well as children [8–10]. In contrast, effective parental coping strategies and emotional resilience have been shown to positively influence the child's adaptation to their illness and overall health-related quality of life (HRQoL) [11].

The previous literature has documented the emotional response of parents and carers to having a child diagnosed with leukaemia and explored the HRQoL of parents in a variety of geographical and socio-economic settings [12–20]. However, most existing leukaemia-specific studies are either qualitative or cross-sectional, retrospective and involve small population sizes, resulting in a lack of longitudinal data [8]. Furthermore, there is a lack of standardization regarding the time points at which distress is measured and the methods used to report the prevalence, severity and duration of symptoms [21]. Notably, the contemporary psychological experiences of Australian and New Zealand parents and caregivers remain underexplored; prior studies are now over 20 years old and included fewer than 40 participants [22,23].

Parents' and caregivers' experiences are influenced by a range of positive and negative factors. Factors such as higher income, better baseline mental health, lower levels of care burden with less daily care time and having more co-caregivers such as extended family and social supports have been shown to positively impact parents' emotional wellbeing [18,24]. For example, qualitative data from parents of children diagnosed with cancer suggest that the experience can strengthen family cohesion and draw family members closer together [11]. Conversely, social isolation, increased sleep disruptions, negative attachment style and female gender have been previously associated with negative parental HRQoL [25–27]. For example, data from a cross-sectional survey of 44 caregivers demonstrated significantly higher anxiety scores (measured by the Beck Anxiety Inventory) in women compared to men [28].

Psychosocial assessment is a standard of care in paediatric oncology practice, and existing evidence supports the use of tools such as the Psychosocial Assessment Tool (PAT) and Distress Thermometer (DT) for early identification of risks and resiliencies [29]. Additionally, a large body of evidence supports the use of the Patient-Reported Outcome Measurement Information System (PROMIS) in paediatric oncology trials, with strong evidence for validity and responsiveness [30].

Our study uses a prospective, longitudinal approach to assess parental distress, anxiety, depression, anger and the need for help at multiple time points during diagnosis and treatment. We have previously shown that prospectively monitoring the incidence of treatment-related toxicities and the longitudinal impacts on HRQoL throughout ALL therapy is feasible in the Australian/New Zealand setting [31]. This work found that parents' emotional wellbeing was poorest in the first 6 months post-diagnosis, with anxiety being the most highly rated concern. In this study, we expand on our previous work with a longer duration of follow-up (5 years) and a larger parent cohort (117 participants). The primary aim was to describe the psychological experiences of Australian and New Zealand parents of children diagnosed with ALL, focusing on the prevalence and time course of psychological comorbidities including distress, anxiety, depression, anger and the need for help. Secondary aims included (1) identifying potential risk factors for worsened parental psychosocial outcomes, such as time since diagnosis, child demographics (age and sex), socio-economic status and disease-related variables (risk stratification and presence of toxicity), and (2) evaluating the correlation between the ET tool responses and PROMIS responses.

2. Materials and Methods

2.1. Participants

The *Acute Lymphoblastic Leukaemia Subtypes and Side Effects from Treatment* (ASSET) study is a prospective registry study conducted across nine sites in Australia and New Zealand, designed to identify ALL patients at increased risk of treatment-related toxicity and capture whole-of-life impacts from ALL and its treatment. Patients and their parents were identified by their treating clinicians and invited to participate by a research coordinator. Patients were recruited within 90 days of commencing chemotherapy. The ASSET study opened to recruitment in January 2016, and the HRQoL opened to recruitment in October 2018 at eight of the nine sites. Here, we report on data obtained from the HRQoL sub-study between October 2018 and November 2022. Eligibility criteria for the HRQoL sub-study included parents/guardians of children aged $\leq$ 18 years old with newly diagnosed ALL or mixed phenotype acute leukaemia (MPAL). The study was approved by the HNE HREC (2019/ETH00693) and conducted according to the Australian National Statement on Ethical Conduct in Human Research [32]. Informed voluntary consent was obtained from participants or their parents and/or legal guardians.

2.2. Measures

Following consent, eligible parents were sent an invitation email to complete their study questionnaire via Research Electronic Data Capture (REDCap). Families nominated one parent or carer to complete all assessments, which were completed online using any internet-enabled device. Surveys were distributed at set time points using two tools to measure parental psychosocial wellbeing. Parents were invited to complete surveys every two months during the first two years of their child's treatment, followed by surveys every six months for the next three years. The central ASSET study team managed survey distribution. We extracted demographics and clinical factors including risk stratification and treatment-related toxicity from the larger ASSET database.

The Emotion Thermometer (ET) tool is an adaptation of the distress thermometer, which was developed and validated for the evaluation of distress in cancer [33–35]. The ET assesses distress, anxiety, depression, anger and the need for help, and is scored on a 0–10 scale [36]. The ET tool has been validated in an Australian oncology population [37]. The ET tool is a single-item questionnaire used for screening.

Participants also completed Patient-Reported Outcome Measurement Information System (PROMIS) questionnaires for anxiety, depression and anger [38,39]. The PROMIS surveys consisted of 29 anxiety statements, 28 depression statements and 22 anger statements. Participants rated their response to each statement on a scale from always, often, sometimes, rarely to never. PROMIS questionnaires have been validated in the Australian adult population [40]. As they consist of multiple questions, PROMIS questionnaires are a more thorough assessment compared to the ET screening tool.

In summary, we used five ET measures (distress, anxiety, depression, anger and the need for help) and three PROMIS measures (anxiety, depression and anger). To minimise the burden of participation for parents, PROMIS anger questionnaires were removed from the study in December 2021, and anger was subsequently only measured using the ET tool.

2.3. Statistical Analysis

We conducted all data analysis using IBM SPSS Statistics Version 29.0.2.0. To measure trends over time, we grouped survey responses based on time since diagnosis (0–6 months, 6–12 months, 12–18 months, 18–24 months, 2–3 years and $\geq$3 years). We conducted single and multiple variable mixed model analyses to analyse for associations with increased parental distress responses.

We classified ET tool scores greater than or equal to 4 as clinically significant and warranting further follow-up according to previously published clinical cut-offs [41–43].

PROMIS T-scores were assessed based on the developers' instructions. T-scores 0.5–1 standard deviation above the mean T-score were classified as mildly elevated. Scores between 1 and 2 standard deviations were classified as moderately elevated. Scores 2 or more standard deviations above the mean were classified as severely elevated [38,44,45].

We used Pearson correlations to evaluate the relationship between mean ET tool responses and mean PROMIS questionnaire responses for anxiety, depression and anger. We calculated Kappa statistics to determine the level of agreement between clinically elevated responses measured by the ET tool and PROMIS questionnaires. We interpreted the Kappa statistic of < 0.0 to demonstrate no agreement, 0–0.20 as slight agreement, 0.21–0.40 as fair agreement, 0.41–0.60 as moderate agreement, 0.61–0.81 as substantial agreement and 0.81–1.0 as almost perfect agreement.

Regionality and socioeconomic data were determined based on patient postcode. For Australian participants, the Accessibility/Remoteness Index (ARIA) was used to classify parents as residing in metropolitan, inner regional, outer regional or remote areas [46]. Postcode was also used to determine the parent Socio-economic Index for Areas (SEIFA) score, which was used for socioeconomic classification [47]. A lower SEIFA score indicates a higher level of socio-economic disadvantage.

2.4. Psychological Safeguards

For parents indicating a clinically elevated need for help, they received a follow-up phone call from a research psychologist and were offered information resources and information about local supportive services via e-mail. Parents were also given the opportunity to have their concerns raised with their child's local treating team, or the opportunity to discuss their need for help with a study psychologist via telephone conversation.

3. Results

Of the 297 patients enrolled in the ASSET registry between October 2018 and November 2022, 119 parents (40%) participated in the opt-in HRQoL sub-study. Two patients were excluded due to being over 18 years old. From 117 eligible parents, 327 survey responses were collected (Table A1), ranging from 1 to 8 responses per participant, with a median of 2 surveys per parent. One-third (34.2%) of parents completed only one survey. The survey responses spanned from 0 to 62 months since diagnosis (Figure A1). There were 67 responses recorded from 0–6 months post-diagnosis, 46 from 6–12 months, 48 from 12–18 months, 48 from 18–24 months, 63 for 2–3 years and 55 for >3 years. There were a few missing responses (<2% for each measure).

3.1. Demographics

The demographics of study participants are shown in Table 1. The median age of children at ALL diagnosis was 7 years, ranging from 6 months to 18 years, with the majority (n = 85, 73%) being under 10 years old. Most children with ALL (n = 71, 61%) were male. Approximately two-thirds of children with ALL (n = 75, 64%) were treated on Children's Oncology Group (COG) treatment protocols, while 42 (36%) received treatment on Berlin–Frankfurt–Münster (BFM) protocols. Children's diagnoses were similarly distributed across risk categories, with 41 (35%) low/standard risk, 35 (30%) medium risk and 41 (35%) high risk. Most families (n = 90, 77%) resided in metropolitan areas, which is broadly representative of urban–rural distributions in Australia (where 73% of the total population reside in metropolitan areas [48]) and New Zealand (where over 80% of the population reside in urban areas [49]). The median SEIFA centile was 60.5 (range 5–100), with about half of the participants (n = 59, 50.5%) falling in the 60th centile or above. During the data collection period, four children (3%) experienced disease relapse and two children (2%) died.

Table 1. Demographics of the patient cohort in the ASSET HRQoL sub-study (n = 117 children with ALL).

Patient Information	n	%
Median age in years at diagnosis (range)	7 (0.5–18)	
Age < 10	85	72.6
Age ≥ 10	32	27.4
Male	71	60.7
Female	46	39.3
Metropolitan *	90	76.9
Inner regional *	22	18.8
Outer regional or remote *	5	4.3

Table 1. *Cont.*

Patient Information	n	%
SEIFA [#] rank <20th centile	7	6.0
SEIFA rank 20–40th centile	19	16.2
SEIFA rank 40–60th centile	21	17.9
SEIFA rank 60–80th centile	27	23.1
SEIFA rank >80th centile	32	27.4
Unknown (New Zealand)	11	9.4
Treated on COG protocols	75	64.1
Treated on BFM protocols	42	35.9
Low/standard risk ALL	41	35.0
Medium risk ALL	35	29.9
High risk ALL	41	35.0
Presence or development of treatment-related toxicity	35	29.9
Child relapsed during study	4	3.4
Child died during study	2	1.7

[*] Accessibility/remoteness index (ARIA) [46]. [#] Socio-economic indexes for areas (SEIFA) score in Australia. The SEIFA score indicates relative socio-economic advantage and disadvantage using Census data, with a lower score indicating a higher level of socio-economic disadvantage [47].

3.2. Trends over Time

The proportion of parents demonstrating clinically elevated emotional scores reduced over time, as shown in Table 2. Across the eight measures (five ET and three PROMIS measures), fewer participants reported clinically elevated scores as time since diagnosis increased. Within the first months following diagnosis, 40.3% of parents reported four or more clinically elevated measures, which decreased to 23.6% beyond three years. Only one-third (33.6%) of parents reported no clinically elevated measures at any time point.

Table 2. Prevalence of clinically significant distress, anxiety, depression, anger and the need for help as measured by the Emotion Thermometer (ET) and PROMIS questionnaires in the ASSET HRQoL sub-study (eight measures in total) compared to time since diagnosis.

Time Since Child's Diagnosis	Total Number of Survey Responses	Number of Clinically Elevated Measures in Parent Responses = n (%)				
		0	1	2	3	≥4
0–6 months	67	9 (13.4)	11 (16.4)	10 (14.9)	10 (14.9)	27 (40.3)
6–12 months	46	15 (32.6)	6 (13.0)	2 (4.3)	6 (13.0)	17 (37.0)
12–18 months	48	21 (43.8)	6 (12.5)	3 (6.3)	2 (4.2)	16 (33.3)
18–24 months	48	17 (35.4)	9 (18.8)	4 (8.3)	3 (6.3)	15 (31.3)
2–3 years	63	27 (42.9)	8 (12.7)	1 (1.6)	8 (12.7)	19 (30.2)
>3 years	55	21 (38.2)	9 (16.4)	6 (10.9)	6 (10.9)	13 (23.6)
All time points	327	110 (33.6)	49 (15.0)	26 (8.0)	35 (10.7)	107 (32.7)

ET tool responses for parent QOL measures peaked within the first 6 months and gradually reduced over time (Figure 1A). The mean score for distress and anxiety was above the clinical cut-off within the first 6 months, highlighting the significant emotional response during this time point.

As measured by PROMIS questionnaires, anxiety and depression levels were highest within the first six months, followed by a gradual reduction over time (Figure 1B). In contrast, anger remained relatively stable throughout the study period, showing no significant temporal variation.

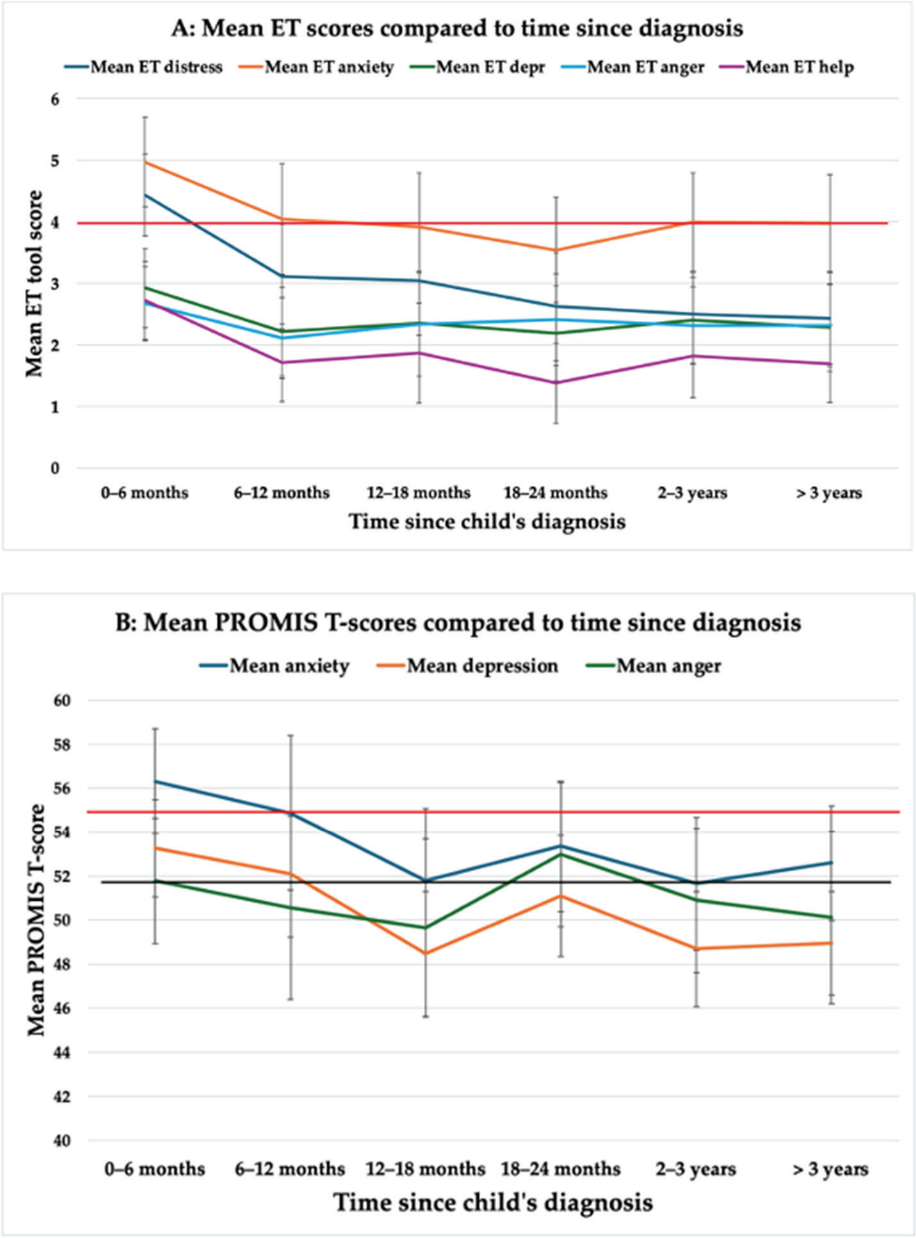

Figure 1. (**A**) Mean Emotion Thermometer (ET) scores compared to time since diagnosis. Red horizontal line indicates the clinical cut-off of ≥ 4. Error bars indicated 95% confidence interval. (**B**) Mean PROMIS T-scores compared to time since diagnosis. Red horizontal line indicates the clinical cut-off of 0.5 SD above the mean. Error bars indicate 95% confidence interval.

3.3. Emotion Thermometer Responses

As measured by the ET tool, anxiety was the most frequently elevated measure. Anxiety scores were highest within the first 6 months post-diagnosis, with a mean ET score of 5.0 and 64% of parents scoring ≥ 4 (Figure 2A). By 18–24 months, the mean ET anxiety score decreased to 3.5, although nearly half (48%) of responses remained above the clinical cut-off. From 2 years post-diagnosis and beyond, the mean anxiety score rose slightly to 4.0, with a similar proportion of responses scoring above the clinical cut-off (51% between 2 and 3 years, 47% beyond 3 years).

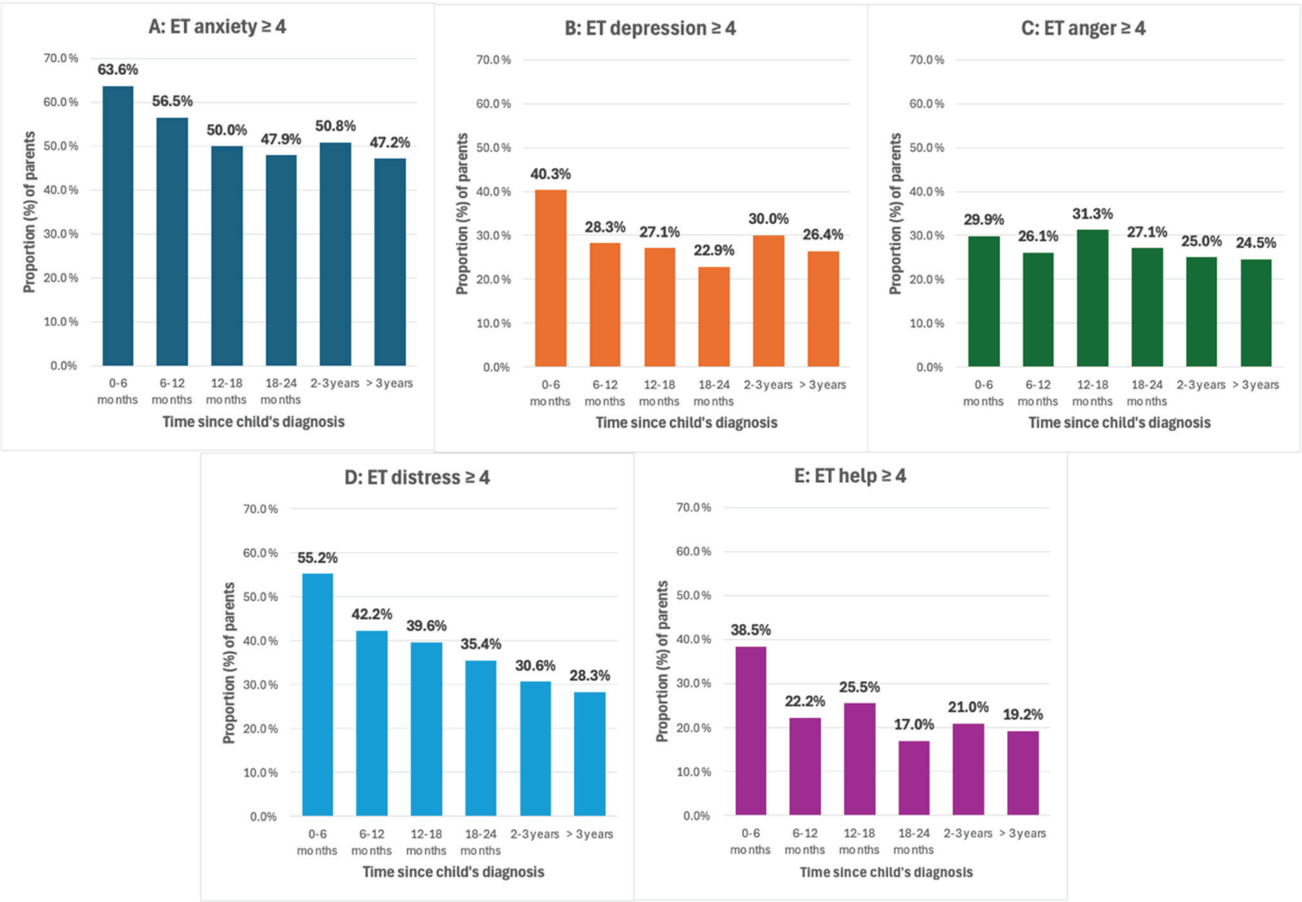

Figure 2. Proportions of parents reporting Emotion Thermometer (ET) scores above the clinical cut-off ($\geq$ 4) across time since their child's diagnosis. (**A**) ET anxiety, (**B**) ET depression, (**C**) ET anger, (**D**) ET distress and (**E**) ET need for help.

Mean ET scores for depression remained relatively stable over time (range 2.19–2.93 across periods), though the proportion of parents reporting clinically significant depression decreased from 40% within the first 6 months post-diagnosis to 26% beyond 3 years (Figure 2B).

Mean ET anger scores remained consistent over time, ranging from 2.11 to 2.67 across different time periods. The proportion of parents reporting clinically elevated anger (score $\geq$ 4) varied between 25% and 31% without a significant peak at any specific time period (Figure 2C).

Distress scores were highest within the first 6 months of diagnosis, with 55% of parents/carers reporting scores above the clinical cut-off (Figure 2D). A gradual reduction in distress scores was observed over time, with 28% of parents reporting clinically elevated distress more than 3 years post-diagnosis.

The need for help scores peaked within the first 6 months post-diagnosis, with 39% of parents/carers reporting scores above the clinical cut-off (Figure 2E). This proportion decreased over time, reaching 19% for those who were more than 3 years post-diagnosis, reflected by a gradual decline in mean scores to 1.69 after 3 years.

3.4. PROMIS Questionnaire Responses

Anxiety was also the most commonly elevated measure on PROMIS questionnaire responses. In the first six months from diagnosis, the mean PROMIS T-score was 56.3, with 34% in the moderate to severe range (Figure 3A). In difference to ET tool responses, there was a larger proportion of responses indicating clinically elevated anxiety between 6 and

12 months from diagnosis. There was a secondary peak 18–24 months from diagnosis, reflecting a time approaching the end of treatment in most ALL protocols. The mean PROMIS anxiety T-score decreased to 50.8 between 2 and 3 years.

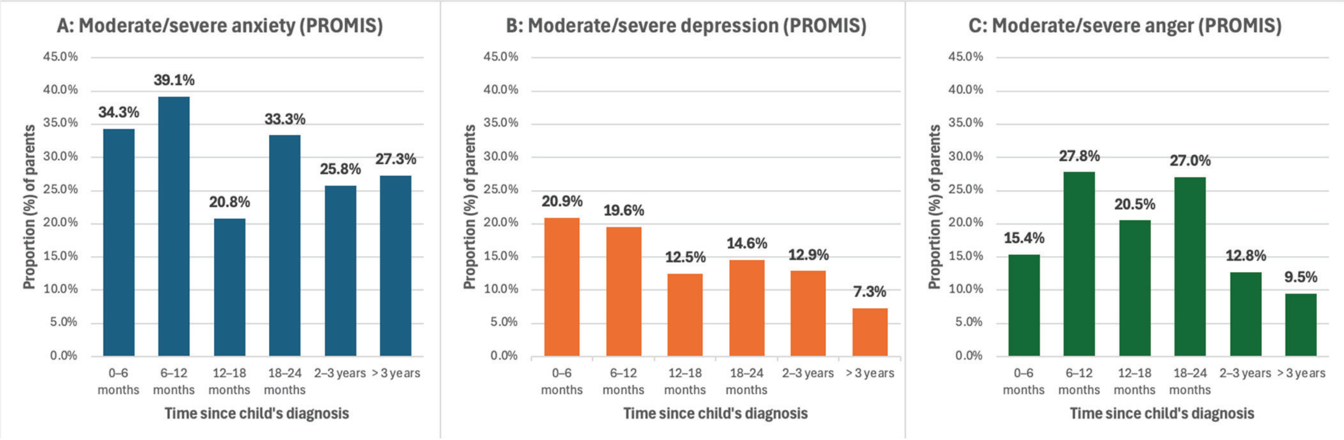

Figure 3. Proportion of parents reporting PROMIS questionnaire responses in the moderate to severe range. (**A**) PROMIS anxiety, (**B**) PROMIS depression and (**C**) PROMIS anger.

PROMIS depression scores were highest within the first 6 months of diagnosis, with a mean of 53.3 and 21% in the moderate to severe range (Figure 3B). By 3 years post-diagnosis, the mean score dropped to 49.0, with only 7% of parents scoring in the moderate to severe range.

Mean PROMIS anger T-scores ranged from 49.2 to 51.6 across time blocks. The highest proportion of anger scores occurred between 6 and 12 and 18 and 24 months post-diagnosis, with over 27% indicating moderate to severe anger (Figure 3C). This contrasts with 15% within the first 6 months post-diagnosis and less than 13% from 2 years onwards.

3.5. Risk Factor Analysis

As time from diagnosis increased, and adjusting for other variables in the model, scores significantly decreased for PROMIS anxiety (estimate −0.18, 95% CI −0.28 to −0.086, *p*-value <0.001), PROMIS depression (estimate −0.13, 95% CI −0.22 to −0.0036, *p*-value = 0.006) and ET distress (estimate −0.073, 95% CI −0.13 to −0.015, *p*-value = 0.013) (Table 3). Using a single variable analysis, increasing time was associated with reduced need for help scores (estimate −0.018, 95% CI −0.036 to 0.00, *p*-value = 0.045), though this association was not statistically significant on multiple variable mixed model analysis.

In this dataset, no statistically significant associations were found between the measured emotional responses and other independent variables including the child's age at diagnosis, the child's sex, ALL risk stratification, or the presence of toxicity (Table 3). We found a statistically significant association between the ET anger score and the SEIFA index, whereby a higher SEIFA index score was associated with a lower ET anger score (estimate −0.02, 95% CI −0.03 to −0.001, *p*-value = 0.038). In our single variable mixed model analysis, we noted a statistically significant association with the study group protocol (COG or BFM) the child was treated on and ET anger score (mean difference estimate −0.76 for children treated on BFM study group protocols, 95% CI −1.49 to −0.04, *p*-value = 0.040); however, this did not remain significant when adjusting for other variables in the multiple variable mixed model analysis.

Table 3. Single variable and multiple variable mixed model analysis of PROMIS questionnaire compared to variables including time since diagnosis, child's age at diagnosis, child's sex, SEIFA index, ALL study group (COG or BFM), ALL risk stratification and presence of toxicity.

		Single Variable Analysis		**Multiple Variable Analysis**	
		Estimate (95% CI)	***p*-Value**	**Estimate (95% CI)**	***p*-Value**
ET Distress	Time since diagnosis	−0.04 (−0.06 to −0.01)	**0.001** *	−0.07 (−0.13 to −0.02)	**0.013** *
	Child's age at diagnosis	0.02 (−0.08 to 0.13)	0.67	0.10 (−0.02 to 0.36)	0.49
	Child's sex	0.40 (−0.43 to 1.23)	0.34	0.06 (−1.47 to 1.59)	0.94
	SEIFA index	−0.01 (−0.03 to 0.004)	0.13	−0.01 (−0.04 to 0.02)	0.38
	ALL study group (COG/BFM)	−0.35 (−1.19 to 0.49)	0.41	−0.02 (−1.75 to 1.35)	0.80
	Risk stratification	−0.33 (−1.35 to 0.70)	0.53	0.73 (−1.29 to 2.74)	0.48
	Presence of toxicity	0.29 (−0.65 to 1.23)	0.54	0.22 (−1.26 to 1.70)	0.77
ET Anxiety	Time since diagnosis	−0.02 (−0.05 to 0.001)	0.06	−0.06 (−0.12 to 0.01)	0.11
	Child's age at diagnosis	−0.03 (−0.14 to 0.09)	0.63	0.04 (−0.29 to 0.38)	0.80
	Child's sex	0.44 (−0.45 to 1.33)	0.33	0.68 (−1.21 to 2.56)	0.48
	SEIFA index	−0.01 (−0.03 to 0.004)	0.15	−0.02 (−0.05 to 0.02)	0.38
	ALL study group (COG/BFM)	0.09 (−0.91 to 0.99)	0.85	0.003 (−1.90 to 1.90)	1.00
	Risk stratification	−0.38 (−1.46 to 0.70)	0.49	−1.05 (−3.18 to 1.08)	0.33
	Presence of toxicity	0.32 (−0.68 to 1.33)	0.53	0.28 (−1.51 to 2.06)	0.76
ET Depression	Time since diagnosis	−0.01 (−0.03 to 0.01)	0.33	−0.02 (−0.06 to 0.01)	0.21
	Child's age at diagnosis	0.02 (−0.08 to 0.12)	*0.74*	0.03 (−0.12 to 0.18)	0.69
	Child's sex	0.40 (−0.41 to 1.21)	0.33	0.22 (−0.89 to 1.32)	0.70
	SEIFA index	−0.01 (−0.02 to 0.01)	0.42	−0.001 (−0.02 to 0.02)	0.95
	ALL study group (COG/BFM)	−0.36 (−1.18 to 0.46)	0.39	−0.04 (−1.21 to 1.12)	0.94
	Risk stratification	−0.36 (−1.34 to 0.62)	0.47	−0.39 (−1.79 to 1.01)	0.59
	Presence of toxicity	0.13 (−0.79 to 1.05)	0.78	0.50 (−0.76 to 1.76)	0.44
ET Anger	Time since diagnosis	−0.01 (−0.03 to 0.01)	0.53	−0.006 (−0.03 to 0.02)	0.62
	Child's age at diagnosis	−0.01 (−0.11 to 0.09)	0.84	−0.05 (−0.16 to 0.07)	0.42
	Child's sex	0.38 (−0.35 to 1.11)	0.31	0.30 (−0.47 to 1.07)	0.45
	SEIFA index	−0.02 (−0.03 to −0.004)	**0.013** *	−0.02 (−0.03 to −0.001)	**0.038** *
	ALL study group (COG/BFM)	−0.76 (−1.49 to −0.04)	**0.040** *	−0.48 (−1.27 to 0.32)	0.24
	Risk stratification	0.21 (0.68 to 1.10)	0.64	0.15 (−0.79 to 1.09)	0.76
	Presence of toxicity	0.39 (−0.42 to 1.20)	0.34	0.14 (−0.73 to 1.02)	0.75
ET Need for Help	Time since diagnosis	−0.02 (−0.04 to 0.00)	**0.045** *	−0.04 (−0.08 to 0.01)	0.11
	Child's age at diagnosis	−0.06 (−0.16 to 0.03)	0.19	0.07 (−0.23 to 0.38)	0.63
	Child's sex	0.090 (−0.68 to 0.86)	0.82	0.23 (−1.47 to 1.92)	0.79
	SEIFA index	−0.01 (−0.03 to 0.003)	0.11	−0.01 (−0.04 to 0.02)	0.40
	ALL study group (COG/BFM)	0.12 (−0.65 to 0.89)	0.75	0.15 (−1.54 to 1.84)	0.86
	Risk stratification	0.11 (−0.81 to 1.04)	0.81	−0.68 (−2.56 to 1.20)	0.48
	Presence of toxicity	0.10 (−0.75 to 0.95)	0.81	0.15 (−1.34 to 1.64)	0.84
PROMIS Anxiety	Time since diagnosis	−0.19 (−0.28 to −0.10)	**<0.001** *	−0.18 (−0.28 to −0.09)	**<0.001** *
	Child's age at diagnosis	−0.29 (−0.75 to 0.16)	0.21	0.14 (−0.38 to 0.66)	0.60
	Child's sex	2.83 (−0.70 to 6.36)	0.11	2.17 (−1.59 to 5.92)	0.26
	SEIFA index	0.001 (−0.07 to 0.07)	0.97	−0.002 (−0.08 to 0.07)	0.95
	ALL study group (COG/BFM)	2.20 (−1.38 to 5.78)	0.23	3.30 (−0.62 to 7.22)	0.10
	Risk stratification	0.69 (−3.62 to 4.99)	0.75	0.54 (−4.17 to 5.26)	0.82
	Presence of toxicity	−2.60 (−6.42 to 1.21)	0.18	−2.57 (−6.79 to 1.65)	0.23

Table 3. *Cont.*

		Single Variable Analysis		**Multiple Variable Analysis**	
		Estimate (95% CI)	***p*-Value**	**Estimate (95% CI)**	***p*-Value**
PROMIS Depression	Time since diagnosis	−0.12 (−0.21 to −0.04)	**0.004** *	−0.13 (−0.22 to −0.04)	**0.006** *
	Child's age at diagnosis	−0.15 (−0.58 to 0.27)	0.48	0.28 (−0.21 to 0.77)	0.26
	Child's sex	1.48 (−1.71 to 4.67)	0.36	0.41 (−3.04 to 3.85)	0.82
	SEIFA index	−0.007 (−0.07 to 0.06)	0.82	0.003 (−0.06 to 0.07)	0.92
	ALL study group (COG/BFM)	−0.23 (−3.47 to 3.01)	0.89	0.30 (−3.29 to 3.89)	0.87
	Risk stratification	−0.85 (−4.73 to 3.04)	0.67	−1.60 (−5.85 to 2.66)	0.46
	Presence of toxicity	−2.28 (−5.56 to 1.00)	0.17	−2.53 (−6.18 to 1.13)	0.18
PROMIS Anger	Time since diagnosis	−0.024 (−0.14 to 0.09)	0.68	−0.03 (−0.16 to 0.10)	0.63
	Child's age at diagnosis	−0.17 (−0.67 to 0.32)	0.50	−0.02 (−0.60 to 0.56)	0.94
	Child's sex	1.64 (−2.44 to 5.72)	0.43	2.05 (−2.39 to 6.49)	0.36
	SEIFA index	0.004 (−0.07 to 0.08)	0.92	−0.02 (−0.10 to 0.07)	0.67
	ALL study group (COG/BFM)	1.38 (−2.69 to 5.45)	0.50	1.37 (−3.16 to 5.90)	0.55
	Risk stratification	0.11 (−4.69 to 4.91)	0.96	−0.89 (−6.28 to 4.51)	0.75
	Presence of toxicity	−3.28 (−7.33 to 0.78)	0.11	−4.24 (−8.81 to 0.34)	0.07

CI = confidence interval; SEIFA = Socio-Economic Indexes for Areas; ALL = acute lymphoblastic leukaemia; COG = Children's Oncology Group; BFM = Berlin–Frankfurt–Münster; ET = Emotion Thermometer; PROMIS = Patient-Reported Outcome Measurement Information System. *: bolded results indicate statistically significant associations with *p*-value < 0.05.

3.6. Correlation Between the Emotion Thermometer Tool and PROMIS Questionnaires

There was a moderately strong correlation between anxiety and depression scores when measured by the ET tool compared to PROMIS measures (Pearson correlation 0.44 and 0.34, respectively), whereas the correlation between anger scores was less strong (Table 4). However, the correlation between measures was statistically significant (*p*-value <0.05) for all measures.

Table 4. Correlation between anxiety, depression and anger scores when measured by the ET tool or the PROMIS questionnaires.

	Pearson Correlation (95% CI)	***p*-Value**
Anxiety	0.44 (0.35–0.52)	**<0.001** *
Depression	0.34 (0.24–0.43)	**<0.001** *
Anger	0.17 (0.05–0.30)	**0.008** *

* bolded results indicate statistically significant correlations with *p*-value < 0.05.

There was statistically significant agreement between anxiety and depression scores above clinical cut-off when measured by both the ET tool and PROMIS questionnaires (Table 5). Anger demonstrated lower level of agreement that was not statistically significant.

Table 5. Measure of agreement (Kappa statistic) between clinically elevated anxiety, depression and anger as measured by the ET tool or the PROMIS questionnaires.

	Kappa Statistic	***p*-Value**
Anxiety	0.14	**<0.001** *
Depression	0.16	**<0.001** *
Anger	0.09	0.060

* bolded results indicate statistically significant agreement with *p*-value < 0.05.

4. Discussion

Our results describe the psychological morbidity experienced by parents of children diagnosed with ALL in the Australian and New Zealand context. Anxiety emerged as the predominant psychological burden, with significant levels observed in over 50% of ET responses and moderate to severe symptoms observed in 30% of PROMIS questionnaires. Substantial rates of other psychological symptoms were also demonstrated: depression (29% ET, 15% PROMIS moderate–severe), anger (27% ET, 19% PROMIS moderate–severe), distress (39% ET) and the need for help (24% ET). Temporal patterns of emotional distress were observed, with anxiety and depression peaking within the first six months post-diagnosis, followed by a gradual decline over time. Conversely, parental anger, which has been less well explored in the existing literature, remained relatively stable throughout and did not exhibit an early peak. Despite the overall reduction in average distress, anxiety, depression and the need for help scores over time, a significant minority of parents continued to experience psychological challenges beyond two years post-diagnosis. Additionally, a second peak in anxiety was identified around the two-year mark, which is when ALL treatment often approaches a conclusion. Temporal proximity to diagnosis emerged as the only risk factor for increased psychological morbidity in this cohort. For the psychological symptoms measured by both the ET and PROMIS questionnaires, strong correlations were observed between anxiety and depression responses on the different tools, whereas anger exhibited a more moderate but statistically significant correlation. Furthermore, there was significant agreement between clinically elevated measures across both assessment instruments.

Our study is the largest prospective investigation of parental psychological wellbeing in paediatric ALL within Australia and New Zealand, providing robust evidence of the significant emotional comorbidities in this population. The results of the ASSET parental HRQoL sub-study build on what has been shown in the literature in other populations, with high levels of parental anxiety, depression and distress. Population-level data from Australia [50] and New Zealand [51] suggest prevalence of mental health disorders is around 20% among adults. In contrast, our results revealed a higher prevalence of clinically significant anxiety and depression, underscoring the heightened psychological burden in this group compared to the general Australian and New Zealand populations. Prior studies, noting that many group together cancer types, have similarly demonstrated a high prevalence of negative psychological wellbeing among parents; for example, in a cross-sectional study, 41% of 518 surveyed caregivers reported always/often experiencing depressive or anxiety symptoms [52].

Our study is unique due to its prospective, longitudinal design, which included up to five years of follow-up for parents. This design enabled the identification of chronological trends in emotional responses and allowed for the analysis of potential demographic or disease-related correlations with psychosocial outcomes. The temporal patterns of emotional distress observed in this study build upon the existing literature, which has often grouped all cancer types together, providing new insights specific to this population. For example, in a previous longitudinal investigation of the trajectory of caregiver psychological symptoms of anxiety, depression and distress, caregivers endorsed more elevated symptoms at diagnosis compared to 6 months later [53,54]. The second peak of anxiety observed towards the end of treatment likely reflects the new emotional challenges arising during this transition. Previous studies have identified the unmet needs faced by families as they prepare for post-treatment challenges, which has been hypothesised to contribute to increased distress at this time point [55,56].

The association between increased time since diagnosis and reduced anxiety, depression and distress scores suggests a degree of emotional adjustment over time. The intensity of ALL treatment in the first 6 months is also a likely contributing factor to high levels of psychological distress during that time. This interpretation is consistent with findings of previous prospective studies in which anxiety symptoms declined over the first year of treatment but remained elevated compared to parents who did not have a child diagnosed with cancer [57,58]. The persistent psychological challenges faced by a subset of parents highlight the importance of early and broad psychological screening at the time of diagnosis to identify parents experiencing significant distress [4,59]. Such screening could enable the timely identification of parents at higher risk of distress, facilitating targeted interventions and appropriate resource allocation to support the entire family's psychological wellbeing.

We also assessed parental anger in our population, a novel area of investigation that has been largely unexplored in previous studies on parental emotional wellbeing. Anger scores measured using the ET screening tool showed a significant correlation with those measured by the PROMIS questionnaire (Pearson correlation coefficient: 0.17, 95% CI: 0.05–0.30, $p = 0.008$). However, there was limited agreement between clinically significant anger as identified by the ET tool and the PROMIS questionnaire (Kappa statistic: 0.09, $p = 0.060$). The ET tool, as a screening measure, appears to provide a quick assessment of anger but may lack the nuance of the PROMIS questionnaire. This could explain the limited agreement in identifying clinically significant anger. The PROMIS tool is designed to evaluate emotions across multiple dimensions, allowing for greater precision in identifying thresholds of clinical significance. Anger may differ from depression and anxiety in how it manifests and is reported, potentially influencing the utility of a screening tool like ET. Anger might be more situational or reactive, whereas depression and anxiety often reflect more pervasive emotional states. This variability could reduce the sensitivity and specificity of simpler tools in detecting anger compared to other emotions.

Our study identified an association between lower socio-economic status, as measured by the SEIFA index, and higher ET anger scores. This finding may highlight the systemic barriers faced by different patient cohorts when navigating the healthcare system. In a single variable analysis, we observed an association between the treatment protocol the child received and ET anger scores. However, the small sample size of patients treated with BFM protocols, coupled with the lack of a consistent association in the multivariable analysis, suggests that this finding may be attributable to limitations in sample size.

4.1. Strengths and Limitations

This is the first bi-national study investigating the longitudinal emotional wellbeing of Australian and New Zealand parents and carers of children with ALL. We included both screening measures (the ET tool) and a more comprehensive evaluation (PROMIS questionnaire) of emotional wellbeing and followed parents for more than 3 years post-diagnosis. However, there are some limitations worth noting. Longitudinal studies are affected by retention and subsequent representation of the population. Of the eligible ASSET population, whilst 40% participated in the HRQoL sub-study, there was a high rate of attrition with one-third of the participants not completing more than one survey. Parents with children in more critical health conditions may have lacked the time or ability to participate fully, as their energy may have been directed towards care for their unwell child, suggesting that our results may not reflect the experiences of all parents. Also, our study was conducted in English, so our results do not offer insight into non-English-speaking parents.

Additionally, the data collected were limited with regard to parent demographics and psychological history. Information regarding parent gender and pre-existing mental health diagnosis would be useful for further understanding the emotional experience of the parent cohort. Previous research identifies gender as an important predictor of psychological outcomes [25]. Although we lacked information on premorbid anxiety and depression, the significantly higher prevalence of these conditions observed in our study population compared to the general population supports the robustness of our findings. This emphasises the considerable psychological burden experienced by parents and highlights the critical gap in resources and support needed to address these challenges over time.

4.2. Future Research Directions

One area for further investigation is exploring the sub-populations of parents who exhibit different trajectories of psychological distress. Our study contributes to the existing literature in that the majority of parents adjust well over time, but a significant number have ongoing poor emotional wellbeing [58,60,61]. Further understanding the characteristics of parents who remain distressed compared to those who demonstrate improved emotional experience could provide valuable insights into risk and resilience factors, enabling tailored interventions to better support vulnerable groups. Future research into institutional factors such as available psychological services would also lead to improved understanding and support of the psychosocial experience of parents.

Expanding on the patterns observed in this study, future research should also focus on developing interventions to address specific aspects of parental HRQoL, particularly regarding the second peak of anxiety towards the end of treatment or the sustained levels of anger observed in this population. Interventions could be assessed using the ET tool and PROMIS questionnaires which have been shown to be a valid and reliable option for monitoring parental psychological experience in this population [31]. For example, with ongoing parent recruitment through the ASSET HRQoL sub-study, we could assess how parents who initially have clinically elevated need for help scores subsequently respond following an intervention.

4.3. Clinical Implications

The high prevalence of anxiety, depression, anger, distress and the need for help among parents of children with ALL support the incorporation of emotional screening at diagnosis and at critical treatment milestones such as the end of treatment and transition to survivorship care [59].

There is robust evidence supporting the use of tools such as the PAT for screening in paediatric oncology to identify family psychosocial risks [29,43]. Our findings, including the correlation between the ET tool responses and more extensive PROMIS questionnaires, support the use of the ET tool for screening for impaired emotional wellbeing and documenting trends over time [62]. The ET tool could be used to evaluate the effectiveness of psychosocial interventions within an institution and serve as an adjunct to tools like the PAT to identify parents and carers experiencing heightened distress.

Recognizing the high psychological burden carried by parents reinforces the importance of a holistic, family-centred approach to paediatric oncology care. Providing resources not only for the child but also for their caregivers ensures that the emotional and social needs of the entire family unit are met, ultimately improving overall health outcomes and quality of life [21,31,63].

5. Conclusions

Our results highlight the significant and often enduring psychological burden experienced by parents of children with ALL. These results emphasise elevated levels of anxiety, depression, distress, anger, and the need for help compared to the general Australian and New Zealand populations. Despite a general trend toward reduced distress, anxiety and depression over time, a substantial proportion of parents continued to experience clinically significant challenges beyond two years post-diagnosis, underscoring the long-term psychological impact of caregiving.

Further research is needed to explore risk and resilience factors, develop tailored interventions addressing specific emotional needs and evaluate the impact of these interventions using validated tools like the ET tool and PROMIS questionnaires. Addressing identified gaps in parental emotional support would improve the overall wellbeing of families navigating childhood ALL treatment, thereby reducing the morbidity associated with paediatric ALL.

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Appendix A

Table A1. Summary of quality-of-life scores across all survey time points (n = 327 survey responses [1]).

Emotion Thermometer (ET) Scores [2]	**n**	**%**
Median (IQR) ET distress	2 (0–5)	
Above clinical cut-off	126	38.5
Missing response	4	1.2
Median (IQR) ET anxiety	4 (1–7)	
Above clinical cut-off	171	52.3
Missing response	5	1.5
Median (IQR) ET depression	1.5 (0–4)	
Above clinical cut-off	96	29.4
Missing response	5	1.5
Median (IQR) ET anger	2 (0–4)	
Above clinical cut-off	88	26.9
Missing response	5	1.5
Median (IQR) ET need for help	1 (0–3)	
Above clinical cut-off	78	23.9
Missing response	9	2.8
PROMIS scores [3]	**n**	**%**
Median (IQR) PROMIS anxiety T-score	54.7 (46.2–61.5)	
Anxiety within normal limits	164	50.2
Mild anxiety	64	19.6
Moderate anxiety	86	26.3
Severe anxiety	12	3.7
Missing response	1	0.3
Median (IQR) PROMIS depression T-score	51.3 (44.4–56.9)	
Depression within normal limits	219	67.0
Mild depression	59	18.0
Moderate depression	46	14.1
Severe depression	2	0.6
Missing response	1	0.3
Median (IQR) PROMIS anger T-score	54.3 (44.7–58.8)	
Anger within normal limits	132	57.1
Mild anger	55	23.8
Moderate anger	44	19.05
Severe anger	0	0
Missing response	1	0.4

[1] PROMIS anger surveys removed from December 2021 onwards. Total PROMIS anger responses n = 231; [2] Emotion Thermometer (ET) tool score 0–10 for each measure, clinical cut-off $\geq$ 4. [3] Patient-Reported Outcome Measurement Information System (PROMIS) measures are scored on the T-score metric, in which 50 is the mean of a relevant reference population and 10 is the standard deviation (SD) of that population. A higher T-score denotes more of the concept being measured. Clinical cut-off $\geq$ 0.5 SD (mild = 0.5–1 SD above mean (T-score 55–60), moderate = 1–2 SD above mean (T-score 60–70), severe = $\geq$ 2 SD above mean (T-score $\geq$ 70).

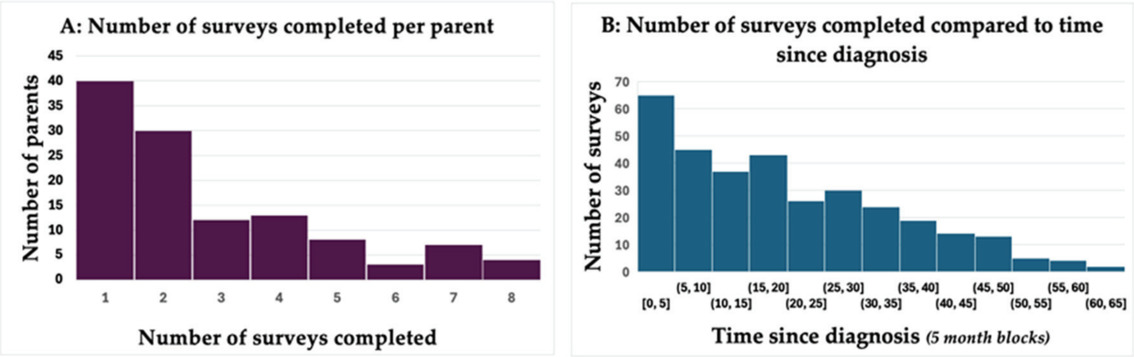

Figure A1. Distribution of surveys completed by parents. (**A**) Number of surveys completed per parent, ranging from 1 to 8 with a median of 2 surveys per parent and (**B**) number of surveys completed according to time since diagnosis, categorized as 5-month intervals.

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Problem-Solving Skills Training for Parents of Children Undergoing Hematopoietic Stem Cell Transplantation: A Mixed Methods Feasibility Study

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Simple Summary: Caring for a child receiving hematopoietic stem cell transplantation (HSCT) can be stressful. This study tested Bright IDEAS®, a problem-solving skills program, to see if it is feasible and acceptable for caregivers in the HSCT setting. Caregivers were assigned by chance to receive Bright IDEAS® with usual care or usual care alone. Bright IDEAS® involved six-to-eight sessions to empower caregivers to manage challenges. IDEAS stands for I—identify problems, D—define options, E—evaluate pros and cons, A—act with a plan, and S—see if it worked. Most caregivers assigned to Bright IDEAS® completed the program and found it helpful. Interviews showed that caregivers appreciated the program's flexibility and support. Overall, the study results suggest that Bright IDEAS® is a promising way to help caregivers during their child's HSCT, and it may decrease symptoms of distress, but needs to be tested in a larger study.

Abstract: Background: Caregivers of children undergoing hematopoietic stem cell transplantation (HSCT) are at risk for psychological distress. Bright IDEAS® (BI) is an effective and acceptable modification of problem-solving therapy for caregivers of children with new cancer diagnoses, but it has not been tested on caregivers of children undergoing HSCT. This randomized controlled pilot examined the feasibility, acceptability, and preliminary efficacy of BI among caregivers of children undergoing HSCT. Methods: English- and Spanish-speaking caregivers of children aged 2–21 years were recruited within 10 days of their child's autologous or allogeneic HSCT. Participants were randomized 1:1 to receive six-to-eight individualized BI sessions or usual care. Participants completed self-report measures of psychological distress at enrollment (pre-intervention) and on days 60, 90, and 180 post HSCT. Qualitative interviews were conducted among a subset ($N = 7$) of intervention completers to assess caregiver perceptions of BI. Results: A total of 107 caregivers were screened for participation; 64 were eligible. Thirty-eight were enrolled (59.4%) and randomized. In the BI group, 14/20 participants (70%) completed six or more sessions. The completion rate for distress measures was at least 78% for both study groups across timepoints. Despite higher baseline scores, caregivers in the BI group reported lower anxiety and depression scores at follow-up timepoints compared to the control group, based on observed trends. Qualitative interviews reflected acceptability of BI. Conclusions: The

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results support the feasibility and acceptability of BI for caregivers of children undergoing HSCT. A larger efficacy trial of BI in the pediatric HSCT setting is warranted.

Keywords: pediatric stem cell transplantation; parent psychological distress; problem-solving skill training

1. Introduction

Approximately 6200 allogeneic and 560 autologous hematopoietic stem cell transplantation (HSCT) procedures were performed for children and adolescents aged <18 years between 2018 and 2022 in the United States [1]. HSCT is a curative therapy for children and adolescents with serious, potentially life-limiting illness, including high-risk, refractory, or recurrent malignancies, immunodeficiencies, and hematologic disorders. However, HSCT procedures are associated with compromised physical, psychological, and social wellbeing [2]. Common medical complications of HSCT procedures include infection, pain, graft failure, and graft-versus-host disease, among other distressing symptoms. Although symptom management strategies are implemented, children undergoing HSCT continue to report a high symptom burden at the time of cell infusion and through the 90 days post HSCT [3].

Alongside their child's intensive treatment, caregivers of children undergoing HSCT face significant sources of stress, including prolonged and repeated child hospitalizations, engaging in complex medical care for their child, disruptions to family routines, and the threat of the possible death of their child. Caregivers in this population are at increased risk of psychological distress [4], with one study reflecting elevated rates of suicidal ideation, ranging from 27% to 39% of caregivers across the time period from stem cell infusion to 90 days later [5]. Caregiver distress has also shown to be significantly associated with poor child health-related quality of life and an increased symptom burden across this time period [6].

Evidence-based interventions are needed to reduce caregiver distress and improve child outcomes for children undergoing HSCT, yet few have been adapted for this population, partly due to the complexities of implementation in this intensive setting. The present study is a pilot randomized controlled trial to test the feasibility and acceptability of delivering an evidence-based cognitive–behavioral intervention in this underserved, high-need setting. Psychoeducational interventions (PEIs) for parents of sick children aim to reduce adverse psychological outcomes, decrease maladaptive parenting behaviors, improve family functioning, and promote the child's health and wellbeing [7]. A recent meta-analysis of randomized controlled trials (RCTs) concluded that PEIs can lead to significant reductions in symptoms of post-traumatic stress, improved mood, and acquisition of problem-solving skills in caregivers of children with cancer [8]. Problem-solving skills training (PSST), specifically, yielded superior effects on caregiver distress and child outcomes for caregivers of children with serious illnesses when compared to other PEIs [9].

PSST is based on the social problem-solving model and emphasizes the importance of fostering enhanced problem-solving skills to support constructive problem-solving and subsequent reduction in emotional distress [9]. Studies of PSST for caregivers have demonstrated improvements in quality of life (QoL) in children with asthma [10], have reduced negative affectivity and distress amongst mothers of children newly diagnosed with cancer [11,12], and have decreased distress in caregivers of adults undergoing HSCT [13,14].

To date, PSST interventions have not been studied in caregivers of children undergoing HSCT. Other caregiver-directed PEIs studied in the pediatric HSCT setting such as massage with relaxation training and guided imagery [15] or stress-reduction education sessions [16] have yielded minimal beneficial effects and documented high drop-out rates with significant missing data. A social–cognitive processing intervention for caregivers of children undergoing HSCT reported an attrition rate of approximately 30% over the course of the intervention, with beneficial effects on caregiver distress that did not endure beyond the short term [17].

Bright IDEAS® (BI) is a cognitive–behavioral PSST intervention with demonstrated efficacy for caregivers of children newly diagnosed with cancer [12], but has not been trialed in the unique, intensive HSCT setting. BI applies a specific method of problem-solving [11,12], where Bright is optimism and IDEAS mark the steps of problem-solving (Identify the problem, Define your options, Evaluate the pros and cons of each option, Act with a specific action plan, and See if it worked).

The primary objective of this research study is to determine the feasibility and acceptability of BI for caregivers of children undergoing HSCT, using a mixed methods approach within a socioeconomic and ethnically diverse population. The secondary objective is to evaluate the preliminary effectiveness of BI in improving problem-solving skills and reducing psychological distress compared to a control group of caregivers receiving the usual psychosocial care. By integrating a mixed methods approach to evaluate feasibility and acceptability, caregivers' voices are incorporated. Caregiver qualitative feedback may be utilized in future iterations of the intervention to refine content, features, and procedures.

2. Methods

2.1. Design

This study employed a randomized, controlled pilot design to test the feasibility, acceptability, and preliminary effect of Bright IDEAS® among primary caregivers of children and adolescents undergoing autologous or allogeneic HSCT with myeloablative or reduced intensity conditioning. The six-to-eight-session intervention and data collection started upon admission to the HSCT inpatient unit, after randomization to intervention or control condition. Repeated psychological outcome measures were collected from caregivers at baseline (prior to their child's stem cell infusion), then 60, 90, and 180 days after cell infusion (four time points in total; Figure 1).

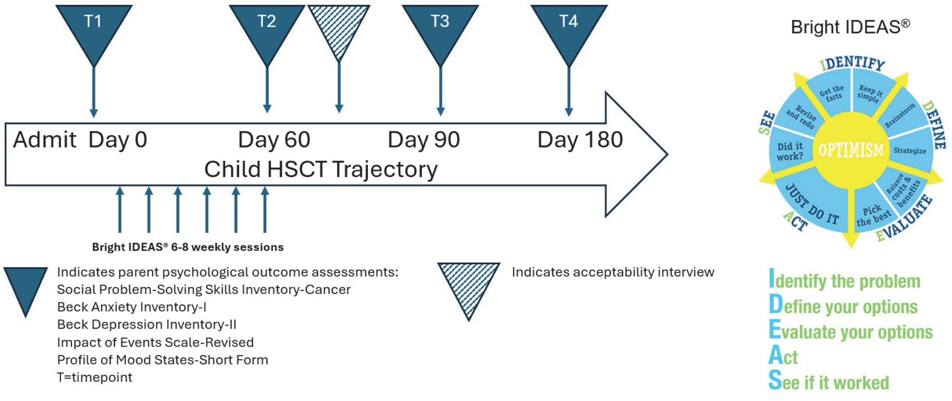

Figure 1. Study schema.

2.2. Setting and Sample

Using a convenience sampling method, participants were recruited from the Transplantation and Cellular Therapy Program at a large urban children's hospital where approximately 100 allogeneic and autologous HSCTs are performed annually. The target sample size was 50 participants, randomized 1:1 to the Bright IDEAS® or control (usual care) group. A formal power analysis was not conducted; rather, the target sample size was estimated to provide sufficient feasibility and preliminary effect data. A 12-month recruitment period was planned. Caregivers of male or female children and adolescents of any race or ethnicity, aged 2 to 21 years, and scheduled to receive allogenic or autologous HSCT using myeloablative (high-intensity chemotherapy, with or without total body irradiation) or reduced-intensity chemotherapy conditioning were eligible to participate. One English- or Spanish-speaking primary caregiver per child was eligible to participate. Caregivers were excluded if they (or their child) were concurrently enrolled on a research study testing psychoeducational interventions for caregivers and/or patients.

2.3. Procedures

Institutional review board approval was obtained. Eligible caregiver–child dyads were identified from the transplantation referral list. Caregivers and children were invited to participate in the study after the child was hospitalized for the HSCT admission, prior to the initiation of reduced intensity or myeloablative conditioning and stem cell infusion. English and Spanish consent and assent forms were available. Caregiver informed consent and permission for child participation was obtained for all study participants. Assent was obtained as per institutional guidelines. Caregiver and children baseline screening data were collected at the time of study enrollment.

Stratified randomization, using the REDCap® (Nashville, TN, USA) randomization module [18], was performed to ensure that an equivalent number of autologous and allogeneic HSCT dyads were assigned to the intervention and control groups. The intervention was initiated after baseline measures were completed. Caregivers completed psychological assessment measures electronically via links sent to their computer, smart phone, or tablet, using REDCap®, regardless of their physical location (inpatient unit, clinic, home). Trained study personnel were available to answer questions or provide support as caregivers completed measures at each timepoint. If electronic measure completion was not possible or preferred by the caregiver, paper measures were provided.

Deidentified child demographic, underlying diagnosis (indication for HSCT), and HSCT data were extracted from the electronic medical record and entered into REDCap® case report forms by study ID number. Caregivers were eligible to receive up to USD 100 in gift cards for measure completion (USD 20 at baseline, USD 30 after 3 months, and USD 50 after 6 months). A subset of caregivers who completed the intervention were invited to participate in semi-structured interviews. Those who completed the interview received an additional USD 50.

2.4. Intervention and Control Condition

The study intervention, Bright IDEAS®, is a problem-solving skills training program (Figure 1) developed by physicians and psychologists working in pediatric oncology [19]. Bright IDEAS® materials are available in English and Spanish in the public domain at https://ebccp.cancercontrol.cancer.gov/programDetails.do?programId=546012 (accessed on 3 March 2025). Consistent with the established Bright IDEAS® manual [12], intervention participants attended up to eight individual, face-to-face sessions with a trained BI interventionist over eight weeks. Sessions were scheduled weekly, with flexibility to participant needs. Sessions took place at bedside or in a private room in the transplant unit, per availability and participant preference. Telehealth sessions were offered as needed based on caregivers' preference during hospitalization, as well as after discharge. Caregivers were considered to have completed the intervention if they participated in six sessions [12], with the option to complete up to eight determined by participant interest and progress.

Intervention sessions and all BI materials were available in English and Spanish [11,12]. Sessions entailed the use of worksheets teaching a stepwise process to assist and empower caregivers to contemplate and document challenges and goals in their life (worksheet #1), focus on and identify elements of a single problem to solve (worksheet #2), define and evaluate potential solutions (worksheet #3), and trial an action plan with the opportunity to review the impact and need for revision of the plan (worksheet #4; Table 1). Caregivers were prompted to identify one caregiver-level problem, such as relational, logistical, or financial, and one child-level problem that related to a distressing or persisting symptom the child was experiencing. Bright IDEAS® was individualized to address specific problems identified by the participating caregivers. Spanish-speaking interventionists or Spanish interpreters (in-person, via telephone, or via iPad when bilingual interventionists were not available) were used to conduct intervention sessions for Spanish-speaking caregivers.

Table 1. Bright IDEAS® session structure.

Session	Content	Materials
1	Establishing rapport; learning about the family's medical journey • Explaining the Bright IDAS model • Identifying current challenges/problems	Provider manual User manual and brochure Worksheet #1
2–5	Teaching and practice of the BI steps • Identifying the core elements of the chosen problem or challenge • Defining and evaluating the pros and cons of potential solutions • Creating an action plan to implement the chosen solution(s) • See/assess if the plan worked, revise as needed	Provider and user manuals Worksheet #2 Worksheet #3 Worksheet #4 Worksheet #4
6–8	Review of the BI steps and continued practice for mastery • Applying steps to additional chosen challenge as needed	Provider and user manuals Worksheets #2–4

Usual care was used as the control condition, offered to all study participants, including those randomized to Bright IDEAS®, and entailed pre-HSCT assessments done by a dedicated HSCT-trained social worker followed by ongoing social work support, as needed, for the duration of the child's transplantation journey. In addition, psychologists and psychiatrists were available on a consultative basis for children and adolescents in need, but did not provide direct services for caregivers. All study participants were informed about available psychosocial resources for patients and families. Comparison to usual care was used to evaluate the effects of Bright IDEAS® as an adjunctive intervention.

Interventionists on the study team received Bright IDEAS® training prior to engaging in sessions with caregivers. Training materials and procedures were developed and standardized by members of the Bright IDEAS® leadership team who have trained over 300 professionals. Training for the study team consisted of didactics, case-based scenarios, and mentored role-play experiences.

2.5. Baseline and Psychological Outcome Measures

Caregiver outcomes were assessed at baseline (T1, prior to stem cell infusion and BI intervention), then 60 days (T2), 90 days (T3), and 180 days (T4) post stem cell infusion (four time points in total). The Caregiver Demographic and Health Questionnaire (previously described) was used to measure parent sociodemographic and health information [20].

Caregiver problem-solving skills were evaluated using the Social Problem-Solving Skills Inventory Revised Short Form, a 25-item tool that measures five dimensions of problem-solving using a five-point Likert scale [21]. Higher scores indicate better problem-solving skills. Strong reliability and validity have been documented in similar samples of caregivers caring for children with serious illnesses [22].

The Beck Anxiety Inventory (BAI)-I was used to measure caregiver anxiety. The BAI-I is a 21-item, multiple choice, self-report instrument of adult anxiety symptoms (somatic, subjective, and panic-related) [23]. A four-point Likert-type scale structure is used. Item responses range in intensity from 0 (not at all) to 3 (severe). Previous use of the BAI-I in a study involving outpatient adults with psychiatric conditions demonstrated an internal reliability coefficient of 0.92 [23]. Construct validity of the BAI-I has been supported by its convergence with other validated measures of anxiety in studies of adults with anxiety disorders [24].

The Beck Depression Inventory (BDI)-II, a 21-item, multiple choice, self-report measure [25], was used to measure depressive symptoms. A four-point Likert-type response structure is used with items ranging in intensity from 0 (not at all) to 3 (severe). An internal consistency of approximately 0.93 was demonstrated in a prior study of adults with depression, treated in inpatient and outpatient settings [26].

Item #9 of the BDI-II evaluates suicidal ideation. The response options are as follows: (1) I don’t have any thoughts of killing myself, (2) I have thoughts of killing myself, but I would not carry them out, (3) I would like to kill myself, (4) I would kill myself if I had the chance. A standard operating procedure (SOP) for the BDI suicide item was in place for the duration of this study, as previously described [5].

The Impact of Events Scale Revised consists of 22 items that uses Likert-type responses to measure perceived post-traumatic stress and has acceptable psychometrics [27]. The instrument is organized into three subscales (intrusion, avoidance, and hyperarousal). Higher scores indicate increased severity of distress and more symptoms of post-traumatic stress.

The Profile of Mood States (POMS) Short Form, a 30-item self-report instrument with excellent psychometric properties [28], was used to measure negative affectivity. This tool includes five-point, Likert-type items and seven subscales (tension/anxiety, depression/dejection, anger/hostility, fatigue, confusion, vigor, and friendliness). Higher scores represent a greater mood disturbance.

2.6. Qualitative Acceptability

Semi-structured acceptability interviews were performed to detect themes that represent participant perception of the intervention. To meet thematic saturation within time and budgeting considerations, fifty percent of the intervention completers ($N = 7$) were offered to participate in the semi-structured interviews. Interviewees were representative of the full BI group with respect to language, race, ethnicity, gender, and child age. The duration of each interview was approximately 30–45 min. An interview guide was used and audio recordings were transcribed. All interviews were conducted by a member of the study team unfamiliar to the participant (i.e., not their interventionist).

2.7. Analyses

Quantitative analyses included descriptive statistics to summarize the feasibility of the intervention as assessed by enrollment rates, withdrawal rates, intervention completion, and survey completion. Rates of missing data and attrition were analyzed. Caregiver psychological outcome measures were scored as per the published scoring instructions. Central tendency analyses were performed for sociodemographic data, child HSCT data, and caregiver psychological outcomes. Anxiety and depression scores were evaluated based on clinical cutoffs for each severity level. All data analyses were conducted using the R software version 4.4.2 [29].

To execute the qualitative analyses, all interviews were digitally recorded and transcribed verbatim by a professional transcription service, including professional translation for interviews conducted in Spanish. Three members of the research team performed a thematic analysis [30] of the interview transcripts. Data were coded deductively using the interview guide for overall satisfaction, perceived change, and planned skill use, as well as inductively using concepts introduced by participants regarding their experience. Coders independently reviewed transcripts to identify initial codes, then met to resolve discrepancies and reach a consensus on operationally defined codes that were entered into a formal codebook. Dual coders then applied the coding framework in a second cycle to all interview transcripts, and the team subsequently met to reach a consensus on final themes.

3. Results

3.1. Recruitment, Enrollment and Retention

One-hundred seven dyads were screened and 64 were eligible and approached by the study team (Figure 2). Thirty-eight dyads were enrolled and randomized (59.4%); a total of 20 were randomized to the BI intervention arm and 18 were randomized to the control arm (Figure 2). Primary reasons for caregiver exclusion were prior patient enrollment in a conflicting research study (*N* = 12, 27.9%), language criteria for survey completion (*N* = 5, 11.6%), and study staff inability to contact the caregiver within the approximately 10-day pre-transplant enrollment window (time from admission to the HSCT unit and duration of conditioning to cell infusion; *N* = 15, 34.8%). Of the 26 who declined participation, 10 (38.5%) indicated not having time or being too overwhelmed, while 16 (61.5%) were uninterested in the study and/or research in general.

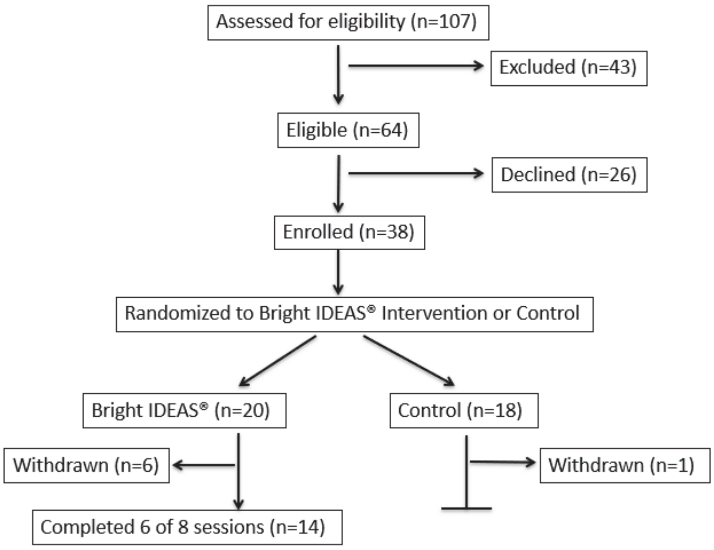

Figure 2. Study enrollment and randomization consort diagram.

Of the 38 dyads enrolled, seven participants later withdrew from the study (18.4%), comprising six withdrawals from the BI group and one from the control group (Figure 2). Two caregivers were withdrawn for failing to complete the baseline surveys, including the caregiver randomized to the control group, one caregiver was withdrawn due to lack of sufficient reading comprehension in English or Spanish, two caregivers withdrew due to feeling overwhelmed and not having enough time to participate, and two caregivers were lost to the follow-up.

Of the 20 caregivers randomized to Bright IDEAS®, two were withdrawn before any sessions could be completed (due to difficulty with reading comprehension or failure to complete baseline surveys, as above). Of the remaining 18 participants, 14 (77.8%) completed the intervention (≥six sessions). Participants attended an average of 5.7 sessions, with 16 caregivers attending at least two. The primary reason for not completing the intervention was caregiver time constraint. The majority (76.6%) of sessions took place in person.

In the BI group, questionnaire completion rates across the four follow-up time points were as follows: 90% (baseline), 89.5% (day 60), 84.2% (day 90), 84.2% (day 180). In the control group, questionnaire completion rates were: 94.4% (baseline), 100% (day 60), 94.4% (day 90), 77.8% (day 180).

3.2. Sample Characteristics

The majority of children enrolled had an underlying diagnosis of cancer (85% in the BI group, 55.5% in the control group) and received an allogeneic HSCT (55% in the BI group, 66.7% in the control group) (Table 2). All caregivers in the BI group were female; two caregivers (11.1%) in the control group were males. The majority of the caregivers were white (40% in the BI group, 33.3% in the control group) and spoke English (65% in the BI group, 77.8% in the control group). The participant sub-sample for qualitative interviews ($N = 7$) was similar to the overall BI group with respect to demographic characteristics; however, interviewees' children all received malignancy diagnoses and allogenic transplants (vs. 85% and 55%, respectively, in the full BI group).

Table 2. Sample characteristics ($N = 3.8$).

		Mean ± SD/*N* (%)	
		Bright Ideas (*N* = 20)	**Control (*N* = 18)**
Child Demographic, Diagnosis, and Therapy			
Age	Child	6.0 ± 5.3	6.8 ± 5.0
Gender	Male	8 (40.0)	10 (55.6)
	Female	12 (60.0)	8 (44.4)
Diagnosis	Malignancy	17 (85.0)	10 (55.5)
	Red cell disorder	0 (0.0)	3 (16.7)
	Immune deficiency	3 (15.0)	2 (11.1)
	Metabolic disorder	0 (0.0)	1 (5.6)
	Other	0 (0.0)	2 (11.1)
Treatment Type	Allogeneic	11 (55.0)	12 (66.7)
	Autologous	9 (45.0)	6 (33.3)
Parent Self-Report Demographic Information			
Age	Parent	37.6 ± 7.3	36.5 ± 6.7
Gender	Male	0 (0.0)	2 (11.1)
	Female	20 (100.0)	16 (88.9)
Race *	White	8 (40.0)	6 (33.3)
	African-American	0 (0.0)	1 (5.6)
	Asian	0 (0.0)	4 (22.2)
	Native Hawaiian or other Pacific Islander	0 (0.0)	0 (0.0)
	American-Indian or Alaska Native	0 (0.0)	0 (0.0)
	Other	3 (15.0)	5 (27.8)
	Do not wish to answer	5 (25.0)	1 (5.6)
	Missing	4 (20.0)	1 (5.6)

Table 2. *Cont.*

		Mean ± SD/*N* (%)	
		Bright Ideas (*N* = 20)	**Control (*N* = 18)**
Ethnicity	Not Hispanic or Latino	3 (15.0)	8 (44.4)
	Hispanic or Latino	10 (50.0)	6 (33.3)
	Other	1 (5.0)	1 (5.6)
	Do not wish to answer	4 (20.0)	0 (0.0)
	Missing	2 (10.0)	3 (16.7)
Primary Language	English	13 (65.0)	14 (77.8)
	Spanish	5 (25.0)	3 (16.7)
	Missing	2 (10.0)	1 (5.6)
Highest Education Achieved *	Grade school	1 (5.0)	1 (5.6)
	Some high school	2 (10.0)	1 (5.6)
	High school diploma or GED	1 (5.0)	1 (5.6)
	Vocational degree	3 (15.0)	4 (22.2)
	Some college	4 (20.0)	7 (38.9)
	Associate degree	0 (0.0)	1 (5.6)
	Bachelor′s degree	3 (15.0)	2 (11.1)
	Graduate degree	4 (20.0)	3 (16.7)
	Do not wish to answer	0 (0.0)	0 (0.0)
	Missing	2 (10.0)	1 (5.6)
Relationship Status	Single, never married	2 (10.0)	1 (5.6)
	Married	9 (45.0)	14 77.8)
	Living with someone as if married	4 (20.0)	1 (5.6)
	Widowed	0 (0.0)	1 (5.6)
	Divorced	1 (5.0)	0 (0.0)
	Separated	2 (10.0)	0 (0.0)
	Do not wish to answer	0 (0.0)	0 (0.0)
	Missing	2 (10.0)	1 (5.6)
Household Income	Less than USD 10,000	1 (5.0)	0 (0.0)
	USD 10,000 to USD 29,999	4 (20.0)	6 (33.3)
	USD 30,000 to USD 49,999	1 (5.0)	1 (5.6)
	USD 50,000 to USD 69,999	1 (5.0)	3 (16.7)
	USD 70,000 to USD 89,999	0 (0.0)	0 (0.0)
	USD 90,000 to USD 149,999	5 (25.0)	3 (16.7)
	USD 150,000 or more	2 (10.0)	4 (22.2)
	Do not wish to answer	3 (15.0)	0 (0.0)
	Missing	3 (15.0)	1 (5.6)
Unable to pay rent/mortgage ^	Yes	7 (35.0)	3 (16.7)
	No	10 (50.0)	15 (83.3)
	Do not wish to answer	1 (5.0)	0 (0.0)
	Missing	2 (10.0)	0 (0.0)
Unable to pay utilities ^	Yes	7 (35.0)	1 (5.6)
	No	9 (45.0)	16 (88.9)
	Do not wish to answer	2 (10.0)	0 (0.0)
	Missing	2 (10.0)	1 (5.6)
Unable to seek medical care #,^	Yes	2 (10.0)	1 (5.6)
	No	15 (75.0)	16 (88.9)
	Do not wish to answer	1 (5.0)	0 (0.0)
	Missing	2 (10.0)	1 (5.6)

Table 2. *Cont.*

		Mean ± SD/*N* (%)	
		Bright Ideas (*N* = 20)	**Control (*N* = 18)**
Unable to seek dental care [#,^]	Yes	41 (29.3)	3 (16.7)
	No	90 (64.3)	15 (83.3)
	Do not wish to answer	1 (0.7)	0 (0.0)
	Missing	8 (5.7)	0 (0.0)

Notes: * some parents selected more than one response, ^ in the past 12 months, # individual(s) in the household unable to seek care due to the inability to pay or lack of insurance.

3.3. Quantitative, Psychological Outcome Measures

Preliminary efficacy analyses probed for changes to self-reported psychological outcome measures across time among participants in the intervention group versus the control, following established guidelines on efficacy reporting in behavioral feasibility studies that are not powered to detect statistically significant changes [31]. Table 3 presents the mean (standard deviation) of caregiver psychological outcome measures over time. Anxiety and depression scores at baseline were higher in the BI group compared to caregivers in the control group. A downtrend in anxiety and depression scores was observed at the follow-up timepoints in the BI group. Impact of Events and Profile of Mood States scores were similar between groups at baseline. These scores remained stable in the BI group, with a downtrend noted in the control group at the follow-up timepoints. Problem-solving skill scores were similar between groups at baseline and remained stable over time.

Table 3. Caregiver psychological outcome scores, mean (SD).

Psychological Outcome Measure	**Arm**	**Pre-Child HSCT**	**Day +60**	**Day +90**	**Day +180**
BAI	BI	11.3 ± 9.6	5.3 ± 4.8	6.5 ± 3.8	6.7 ± 5.5
	Control	7.4 ± 8.4	9.5 ± 8.5	8.0 ± 8.1	8.2 ± 10.3
BDI	BI	14.3 ± 10.4	10.2 ± 8.5	8.5 ± 7.3	10.6 ± 8.6
	Control	9.6 ± 5.8	10.1 ± 10.3	9.7 ± 7.5	9.6 ± 9.6
SPSI	BI	13.2 ± 1.8	13.9 ± 2.0	15.0 ± 2.4	14.4 ± 2.0
	Control	13.6 ± 2.4	14.4 ± 2.6	14.0 ± 2.6	14.0 ± 2.9
IES	BI	29.0 ± 20.0	25.6 ± 13.5	29.5 ± 19.9	29.3 ± 12.4
	Control	29.0 ± 19.8	27.8 ± 18.4	24.6 ± 16.5	21.3 ± 19.5
POMS	BI	22.9 ± 25.5	15.1 ± 13.5	16.5 ± 15.3	24.2 ± 9.6
	Control	21.3 ± 19.7	23.6 ± 27.8	21.6 ± 24.7	15.1 ± 26.5

Notes: SD = standard deviation, BI = Bright IDEAS®, BAI = Beck Anxiety Inventory-I, BDI = Beck Depression Inventory-II, SPSI = Social Problem-Solving Skills Inventory Revised Short Form, IES = Impact of Events Scale Revised, POMS = Profile of Mood States Short Form.

Table 4 displays caregiver anxiety and depression scores based on severity categorization determined by clinical cutoffs. Moderate and severe anxiety (Table 4a) was observed in the BI and control groups at baseline; no participants in the BI group reported moderate or severe anxiety on day 90 or 180. Fewer caregivers in the BI group reported moderate depression on day 90 and 180 compared to baseline (Table 4b). No caregivers in the BI group reported severe depression on day 180.

Suicidal ideation was also assessed as a proxy for caregiver distress, using responses to BDI item #9. Responses of 1 or greater were considered to be indicative of suicidal ideation. In the BI group, suicidal ideation was as follows: pre-infusion: *N* = 1 (5.6%); day 60: *N* = 1 (8.3%); day 90: *N* = 0 (0.0%); day 180: *N* = 0 (0.0%). The frequency of suicidal ideation in the control group was as follows: pre-infusion: $N = 0$ (0.0%); day 60: $N = 2$ (18.2%); day 90: $N = 0$ (0.0%); day 180: $N = 0$ (0.0%). Frequencies were calculated based on the total number of participants who completed BDI #9 at each time point. No caregivers endorsed responses of 2 or 3 at any timepoint.

Table 4. Caregiver anxiety and depression scores by clinical cutoff.

(**a**) Caregiver Anxiety Scores by Clinical Cutoff, N (%).

Clinical Cutoff	BAI-I Range	Arm	Pre-Child HSCT	Day +60	Day +90	Day +180
Minimal	0–7	BI	6 (35.3)	9 (75.0)	6 (54.5)	6 (54.5)
		Control	11 (64.7)	6 (50.0	9 (60.0)	8 (66.7)
Mild	8–15	BI	5 (24.7)	2 (16.7)	5 (45.5)	5 (45.5)
		Control	3 (17.6)	4 (33.3)	4 (26.7)	2 (16.7)
Moderate	16–25	BI	5 (29.4)	1 (8.3)	0 (0.0)	0 (0.0)
		Control	2 (11.8)	1 (8.3)	2 (13.3)	1 (8.3)
Severe	26–63	BI	1 (5.9)	0 (0.0)	0 (0.0)	0 (0.0)
		Control	1 (5.9)	1 (8.3)	0 (0.0)	1 (8.3)

(**b**) Caregiver Depression Scores by Clinical Cutoff, N (%).

Clinical Cutoff	BDI-II Range	Arm	Pre-Child HSCT	Day +60	Day +90	Day +180
Minimal	0–13	BI	6 (35.3)	6 (50.0)	7 (63.6)	5 (45.5)
		Control	12 (70.6)	6 (54.5)	9 (60.0)	8 (66.7)
Mild	14–19	BI	2 (11.8)	2 (16.7)	2 (18.2)	4 (36.4)
		Control	1 (5.9)	3 (27.3)	2 (13.3)	0 (0.0)
Moderate	20–28	BI	5 (29.4)	3 (25.0)	0 (0.0)	2 (18.2)
		Control	2 (11.8)	0 (0.0)	2 (13.3)	1 (8.3)
Severe	29–63	BI	4 (23.5)	1 (8.3)	2 (18.2)	0 (0.0)
		Control	2 (11.8)	2 (18.2)	2 (13.3)	3 (25.0)

Notes: BI = Bright IDEAS®, BAI = Beck Anxiety Inventory, BDI = Beck Depression Inventory.

3.4. Qualitative, Semi-Structured Interviews

Participant feedback from semi-structured interviews supported acceptability to caregivers, with 100% ($N = 7/7$) of those interviewed endorsing overall satisfaction with their experience, perceived positive change in wellbeing following the intervention, and plans to continue using BI skills.

A thematic analysis of qualitative interviews further evaluated caregivers' appraisals of feasibility, implementation, and acceptability (Table 5). An unexpected theme emerged, indicating that BI exceeded participant expectations, as over half ($N = 4/7$) of the interviewees expressed unprompted appreciation or gladness to have participated in BI despite initial reticence to enroll in the study. Similarly to enrollment barriers, participants indicated that the primary challenges to session participation included time and competing medical needs (e.g., interruptions for routine medical cares, meetings or appointments with other providers), while responsiveness to scheduling needs facilitated completion. Relatedly, participants expressed mixed preferences regarding timing and length of the intervention and appreciated that BI provided flexibility and individual choice around scheduling. Most interviewees ($N = 5/7$) expressed a preference for in-person participation versus telehealth, while two were uncertain or preferred a mix of both formats. Participants indicated that they found the one-to-one interventionist role helpful in providing supportive listening and guidance or accountability in using the BI model.

Table 5. Qualitative themes from semi-structured feedback interviews (*N* = 7).

Acceptability		
Theme	**Definition/Subthemes**	**Example**
Satisfaction	Positive experience; appreciation; would recommend to others	"I found it very resourceful to problem solve and navigate certain issues and emotions. So in general, I really liked it and felt very appreciative about it." "I feel like this should be offered, because I'm so glad I did it, because, you know, it really helped me. . .I just feel as though it's a really, really good program."
Exceeded Expectations	Gladness to have participated in BI despite an initial reticence to enroll	"So it seemed like I had a lot in my plate, and I was doubtful about participating in this program. But it helped." "I almost said no because I was like. . .I have so much going on. But I'm glad I said 'yes'."
Feasibility		
Theme	**Definition/Subthemes**	**Example**
Barriers	Time; interruptions; childcare	"There were several times that we started and it just didn't work because the baby was very, very fussy or there was a lot going on."
Facilitators	Flexible scheduling; participant choice	"[The interventionist] was understanding and would wait for me." "I was given the option to, at any point, request more visits or space them out, and I just felt for me once a week was a good fit."
Implementation		
Theme	**Definition/Subthemes**	**Example**
Format	Feedback regarding delivery format (in person vs. telehealth)	"Applying pro and con, it is sometimes better in person because it is easier to express ideas, express feelings face to face." "I didn't mind the Telehealth because it was wonderful to have that flexibility. But me personally, I like it better in person."
Timing of intervention	Feedback regarding timing or number of BI sessions	"Starting it before that [transplant] process probably would have helped make the process, like, easier. Instead of saying okay, we're at this critical point and now we're going to start this...don't let it get to the critical point." "[If] you eventually went to offer it to me before, that would have also been very overwhelming." "I was comfortable with [the BI model] by the time I came home. . .I don't know that I needed the follow up. . .I'm ready to, like, fly and do this on my own."
Use of Worksheets	Feedback regarding BI worksheets	"And even writing down other solutions was very interesting because you could see the amount of solutions this problem has and just get the best one in the moment."
Role of BI Interventionist	Active listening; support	"I felt like all over the place sometimes, and then [the interventionist] would kind of like, on the head, like this is what I hear." "And it felt like I had someone to rely on when I was alone or feeling down, or you know, overstressed."
	Guiding BI skill use	"Having someone guide me through the thought process and have a plan to deal with my issues, that helped me." "I tend to overthink, so. . .it was helpful to have someone kind of simplify things."

4. Discussion

4.1. Summary and Synthesis of Findings

The goal of the current study was to examine the feasibility and acceptability of delivering BI to caregivers of children undergoing HSCT. This randomized, controlled pilot represents a novel application of an evidence-based, structured cognitive–behavioral intervention to improve problem-solving skills and alleviate psychological distress within this uniquely intensive setting in which caregiver support is highly needed yet potentially difficult to access [32].

Results of this BI pilot suggest that the intervention was feasible and acceptable for caregivers of children undergoing HSCT. A priori targets were not set given the dearth of existing PEI studies in pediatric HSCT; however, rates of enrollment (59.4%), retention (81.2%), and survey completion (89.3% across timepoints) were slightly favorable compared to other psychosocial RCTs involving patient–caregiver dyads in cancer populations [33]. Among the caregivers randomized to BI, the majority (70%) completed the intervention. Participant withdrawal from the study was primarily due to time constraints. No participants cited dissatisfaction with BI as a reason for withdrawal.

The current study focuses on feasibility and acceptability of BI in the HSCT setting and did not incorporate formal hypothesis testing of intervention effectiveness [31]. However, trends across timepoints cautiously suggest reductions in symptoms of depression and anxiety in the BI group versus the usual care control with stable follow-up effects. Initial changes in post-traumatic stress symptoms and negative mood were greatest at the first post-intervention follow-up. Notably, the intervention group demonstrated higher levels of psychological distress at baseline, suggesting that BI may be most impactful among caregivers with elevated symptoms of distress. This would be consistent with recent literature suggesting that a resilience skill-building intervention for adolescents and young adults receiving HSCT would be most beneficial for participants with preexisting anxiety and depression [34]. Other recent studies have indicated a risk of caregiver distress not only at the time of HSCT transplant but longitudinally [5,35], while caregivers who utilize approach-oriented coping strategies demonstrate lower distress in adult HSCT populations [36].

All the caregivers interviewed reported high satisfaction with BI, including a perceived positive experience, change, and durability of skill engagement. Though participants appreciated the flexibility afforded by the option to conduct sessions via telehealth, and despite prior research demonstrating the effectiveness of web-based delivery of BI [37], most interviewees in the current study reflected a greater sense of subjective benefit from in-person sessions. Interviewees reflected upon an initial reticence to enroll due to competing priorities at the time of recruitment, alongside gladness or gratitude for having completed BI.

Qualitative and quantitative findings suggest that caregivers benefited from participating in BI. This finding is similar to recent intervention studies involving caregivers of adult patients facing HSCT or advanced cancer [38,39]. Utilizing a “traffic light” model to evaluate feasibility [40], the present results point to an “amber” status, highlighting potential need for revision to the recruitment/engagement approach to support upfront intervention accessibility within the intensive setting of pediatric HSCT. One such strategy could entail expanding the initial engagement window to allow for greater flexibility and responsiveness to individual needs regarding the timing of the intervention.

The results of this pilot trial are consistent with prior studies that have demonstrated the feasibility and acceptability of BI among caregivers of children recently diagnosed with cancer [12]. Recent caregiver interventions with demonstrated feasibility in the HSCT setting have focused on caregiver wellbeing via a positive psychology intervention among caregivers of HSCT survivors [41] and via music play experiences in a small sample of caregivers of young children [42].

4.2. Strengths and Limitations

The present study has several strengths that positively impact its generalizability and support the case for a larger, statistically powered efficacy trial. First, this study included a diverse sample of both English- and Spanish-speaking caregivers. Second, the current pilot is the first to use a mixed methods design incorporating validated quantitative outcome measures of caregiver anxiety, depression, post-traumatic stress symptoms, and negative affectivity in addition to qualitative semi-structured interviews to evaluate a PSST intervention in the HSCT setting. Third, the employment of a narrow pre-transplant recruitment window, a pre-transplant baseline assessment, and multiple follow-up time points allowed for a controlled, longitudinal examination of the feasibility of a BI trial during the most intensive window of HSCT treatment.

This study also included several limitations that are important to consider when interpreting the results and that provide additional direction for future research. Though qualitative satisfaction with BI was high, it is possible that this could reflect selection bias, as interviews involved a subset of caregivers who completed at least six BI sessions. Intervention completers may have been more likely to be satisfied with the intervention than non-completers. While interviewees matched the broader BI cohort with respect to demographics, they comprised all parents whose child had a malignancy diagnosis and allogenic transplant, and it cannot be ruled out that their experience differed due to factors related to their child's diagnosis or treatment. Although no non-completers cited dissatisfaction as a reason for withdrawal, a larger trial could strengthen this evaluation by attempting to include standardized satisfaction assessments for non-completers. Conversely, though feasibility metrics were acceptable for this trial, it is possible that the rates reported underestimate the enrollment and retention potential in this population due to barriers impacting eligibility rates, staffing, and patient access and staff availability during the COVID-19 pandemic. Further, male caregivers were underrepresented in this study and may be particularly at risk of emotional challenges following their child's HSCT [35].

This pilot trial was not powered to assess efficacy of the intervention nor potential mediators or moderators of effect using statistical significance testing and did not meet the target enrollment of 50 participants. Despite reductions in psychological distress being observed descriptively in the BI group versus the control group, minimal differences were noted in participants' self-reported problem-solving skills. Therefore, it cannot be determined whether changes observed in the BI group were due to the intervention or extraneous variance related to other factors. Previous work has demonstrated that improved problem-solving skills mediated less distress in caregivers of children with cancer [12]. Further, more children of caregivers in the BI group had a diagnosis of malignancy compared to children of caregivers in the control group. This observation may contribute to the higher baseline anxiety and depression reported by caregivers in the BI group.

4.3. Future Research

An in-depth analysis of the rich qualitative data obtained in the study is needed to better understand elements of the BI intervention that influence acquisition and maintenance of problem-solving skills in this caregiver population. Additionally, further research employing a randomized controlled design with a larger sample is needed to rigorously examine the short- and long-term effectiveness of BI and the mechanism of the potential effect on caregiver and child outcomes in the HSCT setting. Future research should consider incorporating rigorous examinations of potential moderators to best predict BI's effectiveness; for example, utilizing an a priori cutoff score on baseline measures of distress to guide participant eligibility and randomization procedures might assist in further illuminating which caregivers would benefit most from this resource. Further, prospective and objective measures of caregiver stress and psychosocial support should be added for a comprehensive assessment of caregiver psychological status.

The inclusion of child diagnosis, transplant course, and health care utilization cofactors will be important in future trials with respect to both quantitative and qualitative outcomes. Transplant type and complications such as infection, graft-versus-host disease, graft failure, veno-occlusive disease, post-HSCT disease recurrence, and survival are such examples. Hospital length of stay, intensive care unit transfer, emergency department visits, and readmission may play a role in post-HSCT caregiver distress and may impact BI utilization and effectiveness over time. Measurements of child symptom burden and health-related quality of life should be included in future studies, as these constructs may influence caregiver psychological distress and effectiveness of BI.

5. Conclusions

This study is the first to examine the feasibility and acceptability of Bright IDEAS®, an evidence-based problem-solving skills intervention, to address psychological distress in caregivers of children undergoing HSCT. A rigorous, mixed-method approach was utilized, including a randomized and longitudinal design, semi-structured interviews, and validated psychological measures to pilot BI with a diverse sample of English- and Spanish-speaking caregivers. The results indicate that Bright IDEAS® is feasible and acceptable to caregivers in this setting; therefore, further studies among larger, more balanced samples within multiple HSCT settings are warranted to ensure the intervention's generalizability across diverse demographic groups. In particular, including greater representation of fathers will help provide a more comprehensive understanding of how BI impacts various caregiver subgroups and identify potential variations in effectiveness. Future randomized controlled trials should also consider including greater flexibility in the recruitment window. These adjustments will allow for increased access, understanding of BI's effectiveness, and factors that might impact its effect in order to best tailor this intervention to patient needs in this intensive, underserved setting.

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Data Availability Statement: The data presented in this study are available on request from the corresponding author due to the inclusion of limited identifying information within the dataset.

Conflicts of Interest: Jessica Ward is currently employed at Amgen. Her employment at Amgen began after completion of the study, and Amgen had no input in the study design, analysis, manuscript preparation, or decision to submit. No other authors have conflicts of interest to disclose.

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cancers

Article

Development of an Evaluation Tool for Monitoring the Delivery of Psychosocial Care in Pediatric Oncology Settings

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Simple Summary: Research indicates that the psychosocial and neurocognitive consequences of cancer and its treatment can have adverse lifelong effects, particularly when diagnosed and treated during childhood. After diagnosis, early psychosocial care is important for the timely identification of psychological and neurocognitive concerns and the initiation of appropriate interventions. A proactive approach involving assessment, psychosocial education, and intervention can reduce the intensity and severity of long-term psychosocial effects. The aim of our study was to use the 15 evidence-based standards of psychosocial care established by the Psychosocial Standards of Care Project for Childhood Cancer (PSCPCC) to develop a system for evaluating institutional progress in implementing these standards at the University of Iowa Stead Family Children's Hospital (UI SFCH). This project resulted in the development of a REDCap® version 10.4.1 electronic database, which allowed the pediatric oncology program to not only evaluate its current progress in implementing the PSCPCC standards at the time of this project but also continue to serve as a data repository for the longitudinal evaluation of the program. The initial data imported to the database included two cohorts of patients diagnosed with cancer in 2017–2018 (n = 68) and 2022–2023 (n = 82) for the comparison of psychosocial care delivery over time. The database is designed to track progress in each of the PSCPCC's 15 standards of care. For the purpose of this article, data on Standard 1.A. will be shared to demonstrate the metrics produced by this tool and how they can be used to monitor the provision of services by the institution. With the ongoing use of this tool, the healthcare team at UI SFCH will be able to monitor program progress, identify gaps in psychosocial care, and evaluate specific interventions implemented to address these gaps.

Abstract: In January of 2019, the University of Iowa Stead Family Children's Hospital (UI SFCH) formalized their Pediatric Psychosocial Oncology Program by utilizing 15 evidence-based Standards for Psychosocial Care for Children with Cancer and Families as a foundation for program development. The psychosocial oncology clinical team members identified ongoing gaps in care and a need to improve progress toward achieving these standards. Reviewing and analyzing the Pediatric Psychosocial Standard of Care Institutional Assessment Tool further highlighted the need for program development but also demonstrated the need to design institutionally specific objective measures to monitor program improvements over time. The current project focused on the creation of a program evaluation system with objective measures specific to the UI SFCH practice setting. Barriers such as staffing and cost were identified and addressed. Additionally, a REDCap® database using a structured chart review as its foundation was initiated, which permitted the comprehensive evaluation of the standards of care at UI SFCH. The Matrix and Guidelines included in the Pediatric Psychosocial Standard of Care Institutional Assessment Tool comprised the framework to develop institution specific objective measurements for each standard of care. The objective measures of interest were social work assessments and provider biopsychosocial assessments. Data were exported and uploaded to a statistical program for data analysis. The statistical significance of percentage changes was evaluated with a one-tailed *t*-test; p values < 0.05 were considered significant. The development of this REDCap® database project allowed for the evaluation of the program's current efficiency in implementing the PSCPCC standards of care. Using the database in the future will allow psychosocial oncology team members to easily identify other areas for improvement and to ensure that all 15 standards of psychosocial care are being comprehensively addressed in the care of pediatric oncology patients and interactions with their families.

Keywords: pediatric oncology; psychosocial oncology; program development; program evaluation; database; standards of care

1. Introduction

Despite significant increases in treatment effectiveness and marked improvement in long term survival rates, the diagnosis and treatment of cancer remains one of the most emotionally distressing events in medical care [1–6]. According to the National Comprehensive Cancer Network (NCCN), distress is defined as "a multifactorial unpleasant experience of a psychological (i.e., cognitive, behavioral, emotional), social, spiritual, and/or physical nature that may interfere with the ability to cope effectively with cancer, its physical symptoms and its treatment. Distress extends along a continuum, ranging from common normal feelings of vulnerability, sadness and fears to problems that can become disabling, such as depression, anxiety, panic, social isolation, and existential and spiritual crisis". [4–7] fact, a large body of research documents the psychosocial risks for children and their families during and after cancer treatment, as well as approaches to reducing distress and supporting patients and families [2,8–11]. The Psychosocial Standards of Care Project for Childhood Cancer (PSCPCC; a group of pediatric oncology psychosocial professionals) collaborated with a larger interdisciplinary group of experts and stakeholders to develop evidence- and consensus-based standards for pediatric psychosocial care [10,12,13]. In 2015, this group published 15 evidence-based Standards for the Psychosocial Care of Children with Cancer and their Families (SPCCCF) [12].

The University of Iowa Stead Family Children's Hospital (UI SFCH) pediatric oncology clinical care team has been dedicated to improving psychosocial care delivery in a manner that is consistent with these evidence-based standards. The clinical care team evaluated what resources were currently available and prioritized areas for improvement. Specifically, the team found that UI SFCH did not have a comprehensive team dedicated to the *routine and systematic assessment* of psychosocial healthcare needs and interventions, a situation reflective of national challenges in institutional psychosocial staffing and programmatic implementation [14–17]. The team responsible for caring for cancer patients included only one psychologist and one social worker, which was not enough staff to assess the number of patients in need or to follow the guidelines written within the standards. The team responsible for caring for cancer patients included only one psychologist and one social worker, which was insufficient to adequately assess the number of patients in need or to meet the standards outlined in guidelines. Although the team psychologist attempted to conduct an initial provider biopsychosocial assessment (PBA; focused on the holistic evaluation of biological, psychological, and social factors) for each patient in the early weeks following diagnosis, in reality, the demand for neuropsychological testing in other patients often prohibited the initial PBA of all new diagnoses. Even when an initial assessment was completed, unless a need for psychotherapy was identified at that time, follow-up evaluation did not typically occur due to limited provider resources [14,17,18]. In sum, the lack of a comprehensive psychosocial healthcare team resulted in reactive care as opposed to a proactive approach (taking initiative, preparing, and preventing future distress from occurring). A similar issue was found with respect to the social work assessments. While all newly diagnosed patients received at least one social work assessment (SWA; focused on social and environmental factors) shortly after diagnosis, there was no system in place for structured repeat assessments. Follow-up only occurred if needs were identified in the initial assessment or if the medical team later noted needs arising, typically in the case of an incipient crisis. As a result of its review, the clinical care team determined that an increase in psychosocial services was necessary and that the first step was to shift from a reactive to proactive model of care to align with SPCCCF Standard 1: “Youth with cancer and their family members should routinely receive systematic assessments of their psychosocial health care needs”. [12,18,19].

In January 2019, one full-time nurse practitioner was added to the existing clinical psychologist and medical social worker roles on the oncology team. The nurse practitioner was responsible for the PBA of all patients. A proactive model for these assessments was developed, with the goal of seeing all newly diagnosed patients within 4 weeks and at multiple time points throughout the cancer trajectory, including survivorship years [11,18–21]. The specific time points will be focused on in the Methods section. From these assessments, referrals to additional supportive services or interventions such as individual psychotherapy, neuropsychological testing, Child Life, or music therapy were made. The team’s clinical psychologist was able to focus her effort on neuropsychological testing and psychotherapy. Social work now had an added layer of support to turn to when patients and families were in need. Additionally, with the addition of the nurse practitioner, the oncology team now had a point person for prescribing and managing psychiatric medications [22,23].

Throughout the first two years of having a full-time nurse practitioner, there was a notable increase in the identification of psychosocial care needs in patients, resulting in further demand for intervention and resources. However, the team did not have a specific measurement and evaluation system in place to gather data that would objectively demonstrate the need for increased resources. The lack of such a system also prohibited measuring progress and identifying other areas for improvement in implementing the standards [18,24]. Ultimately, this prevented patients from receiving the levels of support set out in the SPCCCF standards.

Further guidance on implementation and evaluation was disseminated in 2020 by the psychosocial experts who had published the original SPCCCF standards of care [18,19,25]. In consultation with patient advocates, additional pediatric oncology experts and other stakeholders, they developed a matrix and guidelines to help healthcare providers identify and overcome barriers to the implementation of the SPCCCF standards of care. This was achieved through a rigorous, iterative review process including inputs by multidisciplinary psychosocial experts and focus groups and several rounds of revisions following additional expert reviews. This process resulted in the publication of a matrix and guidelines to help healthcare providers identify and overcome barriers to the implementation of the SPCCCF standards of care [18,19,25]. The matrix was designed to help clinicians assess the current levels of psychosocial care at their treatment sites, and the guidelines provide a variety of recommendations to help teams identify a pathway to achieving optimal psychosocial care. The matrix was designed with a scoring rubric with levels 1–5 aligned with each individual standard, with several broken down into sub- components of each standard. A score of 1 indicated a lack of implementation and a 5 indicated the complete implementation of the standards. Given the high level of variability in psychosocial resources across institutions, the matrix was designed with broad measurement criteria, leaving the flexibility for individual institutions to further define specific data points that fit within the broader definitions as they strive to evaluate their program successes and gaps.

To address the goal of developing a robust, comprehensive Pediatric Psychosocial Oncology Program in which a level of excellence in all 15 standards was achieved, the UI SFCH team utilized the rubric matrix and guidelines as tools to develop an institution-specific system for the evaluation of program progress in the implementation of psychosocial care delivery at UI SFCH. In this paper, we describe how objectives from the rubric matrix were framed and documented within a REDCap® database (Research Electronic Data Capture, Vanderbilt University) [26–28], how data were extracted from medical records into this database, the current progress of our program, and the exploration of changes in effectiveness from prior cohorts. This system will continue to serve as a tool for the longitudinal evaluation of the program [24,29].

2. Materials and Methods

2.1. Human Subject Protection

Our prospective study was approved by the University of Iowa Institutional Review Board [IRB Form #202309467] with a partial waiver of HIPPA Authorization for all patients diagnosed prior to 7 November 2023. Children and adolescents who were diagnosed with pediatric cancer during the years 2017–2018 (n = 68) and 2022–2023 (n = 82) were recruited for our study, for a total of 150 patients. Figure 1 shows the process of recruitment for our study.

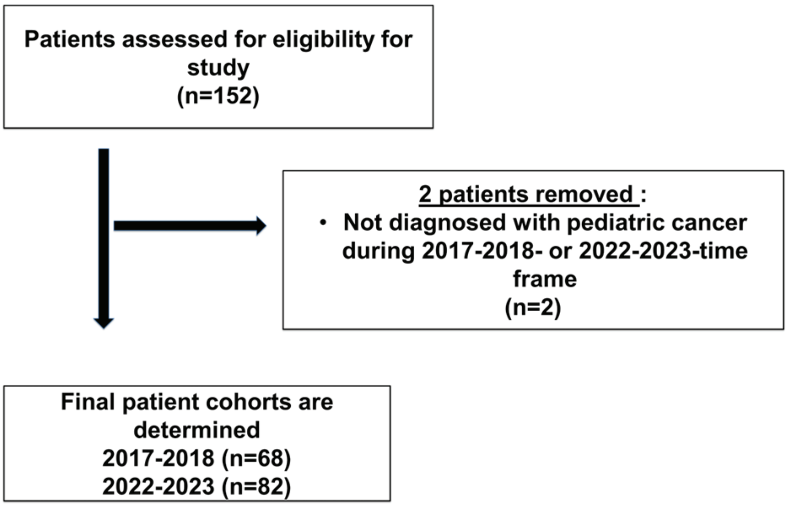


Figure 1. Flow chart of patient population/participant enrollment.

2.2. Procedures/Methods

This project began in 2017 (see Figure 2), when the first steps were taken to establish a psychosocial oncology program at UI SFCH. Using the 15 Standards for Psychosocial Care of Children with Cancer and their Families as the framework for overall program design, key gaps in care were identified. The current article focuses on Standard #1 (Systematic Assessment of Psychosocial Needs). This standard encompasses routine systematic assessments of both youth with cancer and their caregivers [6,7,30]. However, the metrics and data of focus in this article are specific to Standard 1.A., focusing on youth assessments. Gaps for this standard included (a) the timing and breadth of the initial assessment and (b) the frequency of follow-up assessments.

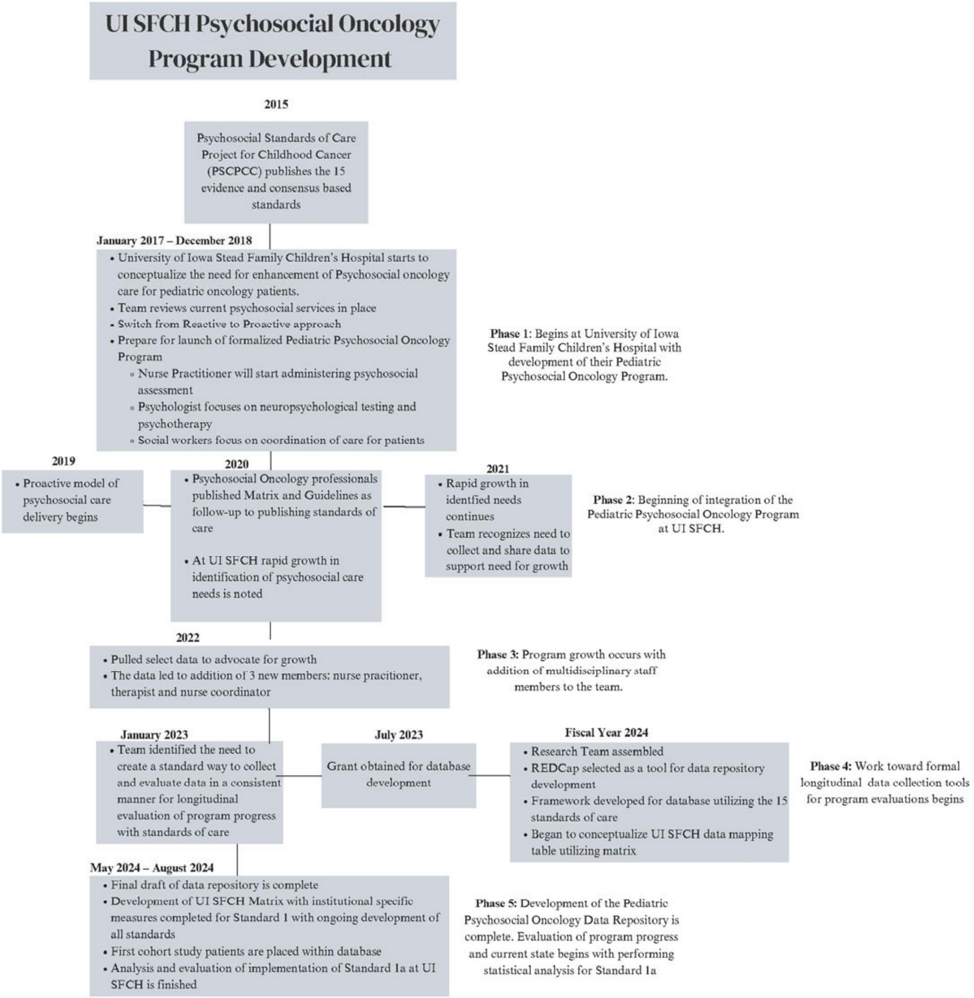


Figure 2. Development of Psychosocial Oncology Program at UI SFCH and development of program evaluation tool and data depository. The timeline includes the steps of development of the program, first cohort study, and first analysis using the evaluation tool that was designed by the team.

2.2.1. Development of an Institution-Specific Matrix

The UI SFCH team modified the published matrix and guidelines [19] based on their existing capabilities to implement the standards (Table 1) and used the same Likert scale of 1–5 as the original matrix. The team began by scoring patients on Standard 1.A., which measures the team status on routine systematic assessments of youth with cancer throughout their cancer trajectory, specifically focusing on the provision of SWAs and PBAs.

2.2.2. Chart Review Development

Using the Iowa-specific matrix as a guide, the team determined what datapoints available in the medical records would best provide information on how well the requirements within Standard 1.A. were addressed. These elements (Table 1) were extracted from the chart review and used as a foundation to structure a REDCap® database.

2.2.3. REDCap® Database Collection

REDCap® (Research Electronic Data Capture, Vanderbilt University) [25] is a secure and widely used online database system. Utilizing REDCap® for data collection allowed the team to test draft designs and pick a design that fit the study and represented the UI data best [26]. Branching logic was used for yes or no questions and made data collection more efficient [27].

Table 1. University of Iowa Stead Family Children's Hospital Matrix and Guidelines.

Standard 1. ***Youth with Cancer and Their Families Routinely Receive Systematic Assessments of Their Psychosocial Healthcare Needs.***		
1: Consider that each of these have dimensions of the following: (a) periodicity (specified as at diagnosis, relapse/disease progression, and at end of treatment), (b) standardized process (systematic assessment), (c) content (see specified domains)		
1.A.: Assessment domains: Youth • Youth pre-morbid and current adjustment • Cognitive and academic functioning/concerns • Developmental level and issues • Family relationships • Quality of social interactions • Disease and treatment related concerns		
Level	**Original Matrix Scoring**	
1	No organized process in place for systematic assessments	
2	* *To be defined at an institution specific level*	
3	There is a system in place to assure that all youth receive assessment of psychosocial functioning early in the treatment trajectory and again only if clinically indicated	
4	* *To be defined at an institution specific level*	
5	All youth receives a comprehensive assessment at regularly scheduled points in their care	
Level	**UIHC Modified Scoring**	**Chart Review Elements**
1	No assessments completed on a child: Social Work Assessment (SWA) OR Provider Biopsychosocial Assessment (PBA)	Was a social work assessment completed? Y/N a. Date of social work assessment completion b. Dates of any repeated full SW assessments Frequency of SW check in during therapy Frequency of SW check in post therapy Psychosocial work assessment completed? Y/N a. Date of the initial assessment b. Dates of any follow-up assessment Time between visits (e.g., time in between first assessment and second assessment) Completed Oncology Treatment? Y/N (Date of completion) Psychosocial oncology assessment at the end of treatment? Y/N (Date of assessment)
2	Assessments completed as follows: Initial SWA completed at any time following diagnosis AND No PBA completed	
3	Assessments completed as follows: Initial SWA completed at diagnosis (within 1 month) AND One PBA completed any time after diagnosis	
4	Assessments completed as follows: SWA completed at least once at diagnosis (within 1 month) AND at least one SWA completed within the first year following the end of treatment (12 ± 3 months) AND PBA initial assessment completed AND at least one additional follow-up PBA completed	
5	Assessments completed as follows: SWA completed at least once at diagnosis (within 1 month) AND Annually (12 ± 3 months) following the end of treatment and continuing for a lifetime AND PBA at the following time points: Initial (within 4 weeks of diagnosis), minimum of every 3 months throughout treatment, end of treatment (±2 months of treatment completion), and at least twice within the year following treatment completion (±3 months), and Annually (12 ± 3 months), continuing for a lifetime	

The REDCap®-based data repository has a total of 17 sections. The initial section is for basic demographic information related to the patients. The second section includes information regarding psychiatric diagnoses and identifies whether each psychiatric diagnosis occurred before or after the cancer diagnosis. Following the first two sections, the database includes individual sections dedicated to each of the 15 standards of psychosocial care. Each dedicated section is designed to collect important data to evaluate program performance with respect to the standard covered. The titles of each section within the database reflected the main points listed within each respective standard. Specific items from the chart review that were relevant to data collection for a given standard (e.g., the frequency of SWAs, the frequency of PBAs) were built into each dedicated section.

2.2.4. Data Collection and Creation of a Training Manual

Data collectors used the EPIC® healthcare records to extract patients' information to answer questions within the section of the REDCap® database pertaining to SPCCCF Standard 1 (see Table 1). To ensure that all collectors were retrieving information from the same place in EPIC®, a data collection manual was developed (available upon request from the corresponding author). This manual was used during collection and will be used to train future research assistants in data collection. Using the data import feature of REDCap® allowed the team to download an Excel file from EPIC® that could be reformatted for upload into the database. After the relevant data were transferred, the collectors would look at patients' answers to questions and score them on the UI SFCH Matrix and Guidelines (Table 1).

2.3. Statistical Analysis

Scores from the UI SFCH Matrix and Guidelines were then exported from REDCap® and imported to SPSS (Version 29.0) statistical software for all statistical analyses. To better understand the team's functioning in relation to Standard 1.A., three one-tailed *t*-tests were used to determine whether there was a difference in the frequency of administration of the SWA and/or PBA between the two cohorts. The first looked only at the frequency of SWAs. The second looked only at the frequency of PBAs. The third looked at the combined score for the receipt of SWAs and PBAs. For each analysis, ninety-five percent confidence intervals were calculated, and significance was set at $p < 0.05$.

3. Results

3.1. Demographics

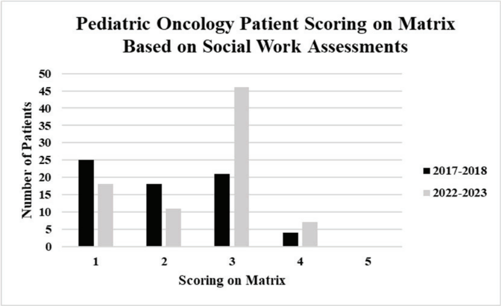
Two cohorts were included in this study, for a total of 150 patients. Table 2 summarizes the characteristics of each cohort. The participants' mean age, averaged over both cohorts, was 8.93 (SD = 6.02), ranging from <1 year old to 25 years old. Most participants were male (56.67%, n = 85) and were white (82.67%, n = 124). In terms of disease state, most patients were part of the solid tumor group (38.67%, n = 58).

Table 2. Demographics of patients.

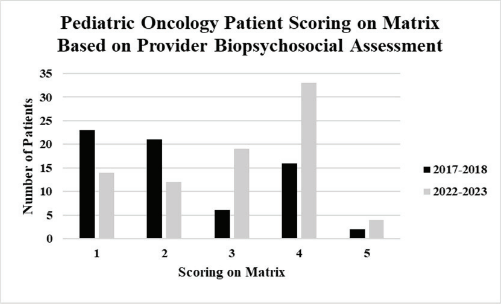
	2017–2018 Cohort		**2022–2023 Cohort**	
	N	**%**	**N**	**%**
Age at Diagnosis				
<1 year old	3	4	9	11
1–2 years old	9	13	11	13
3–4 years old	11	16	8	10
5–7 years old	13	20	7	9
8–10 years old	7	10	12	15
11–14 years old	12	18	16	20
15–18 years old	9	13	19	22
19–25 years old	4	6	0	0
Diagnosis Group				
Non-oncology transplant	1	1	1	1
Leukemia/Lymphoma	22	32	20	24
Neuro-oncology	14	21	34	41
Solid tumor	31	46	27	34
Racial Identity				
White	54	79	70	85
Asian American	0	0	0	0
African American	5	7	4	5
Hispanic/Latino	5	7	6	7
American Indian/Alaskan Native	0	0	0	0
Native Hawaiian/Pacific Islander	0	0	0	0
Multiracial	4	6	2	3
Sex				
Female	24	35	41	50
Male	44	65	41	50
Intersex	0	0	0	0

3.2. Standard 1.A. Matrix Scoring for Social Work Assessments

For each cohort, the social work assessments received by the patients were scored in accordance with the UI SFCH Matrix and Guidelines (Likert scale 1–5, where 5 is complete adherence) (Table 1). The mean social work score for the entire sample, that is, both cohorts, was 2.19 (SD = 0.880; 2.05–2.33, 95% Cl). This corresponds to patients receiving one social work assessment any time throughout their cancer trajectory. There was a significant difference between the two cohorts; the (2017–2018) cohort had a lower score on the matrix (mean = 1.0 [SD = 0.99]) compared to the (2022–2023) cohort (mean = 3.0 [SD = 0.93]; t(df) = 148.00, $p < 0.01$). Closer evaluation indicated that, while many patients received a social work assessment at some point during their treatment, it was rarely within one month of diagnosis. We did see improvement in this standard across two cohorts, where more patients were seen within one month of diagnosis in the 2022–2023 cohort. However, the mean score of this cohort was still at 3.0, indicating that timely follow-up assessment continues to represent a gap in care that needs to be addressed. Figure 3A shows the cohort scoring (Likert 1–5) for the UI SFCH Matrix and Guidelines.



(**A**)



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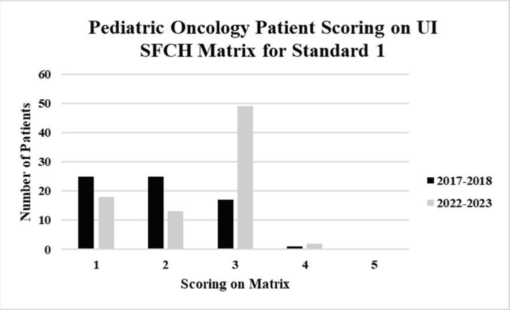

(**C**)

Figure 3. (**A**) The scoring of the patients within the cohorts based on receiving social work assessments according to the UI SFCH Matrix and Guidelines. (**B**) The scoring of the patients within the cohorts based on receiving provider biopsychosocial assessment according to the UI SFCH Matrix and Guidelines. (**C**) The scoring of patients within the cohorts based on receiving social work and provider biopsychosocial assessments according to the UI SFCH Matrix and Guidelines.

3.3. Standard 1.A. Matrix Scoring for Provider Biopsychosocial Assessments

Patients were scored (Likert 1–5) with respect to their receiving PBAs according to the UI SFCH Matrix and Guidelines. The mean PBA score for the entire sample was 2.60 (SD = 2.27; 2.49–2.89, 95% Cl). There was a significant difference between the patients of the two cohorts, with those seen earlier (2017–2018) having a lower score (mean = 2.0 [SD1.24]) than those seen later (mean = 4.0 [SD = 1.20]; t(df) = 149.00, $p < 0.01$). Patients in the second cohort typically received an initial assessment, as well as at least one follow-up assessment. Figure 3B shows the scoring (Likert 1–5) for the UI SFCH Matrix and Guidelines.

3.4. Standard 1.A. Combined Matrix Scoring

Patients were scored on the combined score (Likert 1–5) with respect to their receipt of both assessments (SWA + PBA), as is written within the UI SFCH Matrix and Guidelines. The mean combined score for the entire sample was 2.10 (SD = 0.88; 2.05–2.33 95% Cl). There was a significant difference between the two cohorts, whereby those seen earlier had a lower score (mean = 2.0 [SD = 0.82]) than those who were seen later (mean = 3.0 [SD = 0.86]; t(df) = 148.00, $p < 0.01$). The second cohort of patients had a slightly higher mean score because they received both an initial SWA and one PBA. However, the timing of the SWA was delayed longer than was optimal (i.e., the 2022–2023 cohort patients were seen on average for the SWA over 1 month past their initial cancer diagnosis). Figure 3C shows the combined matrix scoring for both cohorts with the Likert scale.

4. Discussion

The present study aimed to evaluate the two patient care components of SWAs and PBAs in relation to the UI SFCH Matrix Guidelines. While the receipt of assessments did significantly improve between the two cohorts studied, data from all time points collectively support the hypothesis that Standard 1.A. is still not being fully implemented within our clinic. This highlights a specific gap in supportive care that needs further attention.

To the best of our knowledge, this is the first analysis exploring a way to create a data repository to perform evaluations of the implementation of the 15 evidence-based Standards of Care for Pediatric Oncology patients. As proof of principle, this approach successfully allowed the UI SFCH team to evaluate how well they were currently implementing Standard 1.A., that is, administering SWAs and PBAs as they are written within UI SFCH Matrix and Guidelines. As a result, the team was able to identify specific gaps in supportive care. A key aspect of the approach is the use of a database design (REDCap®) that facilitates longitudinal comparisons of cohorts. In summary, our analysis provides a practical demonstration of how, through the development of an electronic database, a clinical care team can adapt the Matrix and Guidelines included in the Pediatric Psychosocial Standard of Care Institutional Assessment Tool to perform an institute-specific evaluation of the implementation of the SPCCCF standards of care.

4.1. Receipt of Social Work Assessments

The qualitative results of our study identified key areas of importance to be used for evaluation and areas of improvement. The analysis of the receipt of SWAs showed that parents were only receiving one assessment any time throughout the cancer trajectory. Improvements were made for the later cohort in terms of a higher likelihood of a visit occurring within a month of diagnosis. However, the overall score for this standard remains low (mean = 3.0 for the most recent cohort), and ensuring that all parents of patients receive a visit within a month of diagnosis remains an unmet goal. To further improve implementation, the team should administer the first assessment at the time of diagnosis, have a follow-up assessment within 12 ± 3 months of the completion of treatment, and have annual assessments for the remainder of the individual's lifetime. From a clinical perspective, if patients received more SWAs, this would allow the team to ensure that all the needs of the patients and caregivers were being accounted for, from the time of diagnosis, through the course and completion of the cancer treatment, and annually, over lifetime follow-up. For our clinic, a realistic next step will be ensuring that patients receive an assessment within one month of diagnosis and at least one additional follow-up.

4.2. Receipt of Provider Biopsychosocial Assessments

Given the increasing number of pediatric patients diagnosed with cancer who need supportive care *throughout* their cancer trajectory, the provision of PBAs is essential. This ensures that the patient's need for support is identified early and interventions can be provided in a timely manner.

The analysis of the receipt of provider biopsychosocial assessment showed that patients in the earlier cohort were only receiving one assessment after the time of diagnosis. Patients seen later did have more evaluations, but their overall scores remained low due to the timing of evaluations—none were completed within one month of diagnosis. To improve the implementation of this standard, our team will need to reduce the time between diagnosis and the date of first PBA. From our clinical perspective, administering earlier initial assessment with frequent follow-up will help address needs proactively as they arise and may prevent a patient from experiencing a crisis.

4.3. Combined Matrix Scoring

The results of our current study, based on the combined score (SWA + PBA), suggest that, while at least one social work evaluation is being performed for all patients, the timing needs to improve. When evaluating Standard 1.A., achieving a score higher than 2 is not possible if the SWA is not completed within one month of diagnosis. With the PBA, one cannot obtain a higher score unless the provider(s) has completed an initial assessment at the time of diagnosis, during treatment, at the end of treatment, within the first year of completion, and annually over a lifetime. To improve standard-of-care implementation for future cohorts, the team's initial goal would be to ensure that (a) patients were receiving an SWA within one month of diagnosis and (b) the PBAs are being administered at the time of diagnosis with at least one follow-up.

Limitations

Limitations in the study of these cohorts included a small sample size, a lack of patient demographic diversity, and the inability of the scoring system to capture all relevant services that had been received. The small sample size (n = 150) increases the risk of type II error (accepting a null hypothesis when a difference truly exists) and sampling bias. Our healthcare center serves a high proportion of patients from predominately White rural communities, this limited both the number and diversity of available patients. Healthcare centers that serve a more diverse population would have more patient representation in many different racial and ethnic groups. This may make the identification of other barriers (e.g., language barriers that may inhibit social work contact and the administration of assessments) more apparent. Another limitation was related to scoring constraints within the UI SFCH Matrix and Guidelines: many patients were reported as receiving multiple PBAs, but they were not administered within the time points recommended in the matrix, resulting in a lower overall PBA score. With SWAs, not receiving a follow-up SWA further prevented any PBAs from being represented in the combined scoring.

Future Directions

The findings of our evaluation study will be used to monitor the continuous improvement of the psychosocial oncology program here at UI SFCH. Specific attention will be focused on the identified areas for improvement, including the administration of SWA follow-up assessments and ensuring that the initial PBA is completed within a month of diagnosis-with regular follow-up. These data will be used as a baseline for future evaluative cohort comparison studies on Standard 1.A. While the team works on continuous improvement in relation to Standard 1.A., future research is needed to gain a deeper understanding of the impacts of social work and provider biopsychosocial assessments in relation to the caregivers of patients (Standard 1.B.).

5. Conclusions

The current research project demonstrated the lack of implementation in relation to Standard 1.A. for two cohorts of pediatric oncology patients and identified areas for improvement in terms of providing SWAs and PBAs for future patients. The monitoring of services was made possible using the electronic database REDCap®. The establishment of this database provided an organized way to compare cohorts and identify the gaps in care. Overall, this study showed the utility of implementation of a standardized system within a REDCap® database project for evaluation of standard of care. This system will continue to allow the team to evaluate the success of new interventions and to identify areas of weakness that can be targeted for improvement to achieve the full implementation of the SPCCCF care standards.

Author Contributions: Conceptualization K.F., methodology K.F. and B.S., protocol writing B.S., database design K.F., B.S., A.L.C. and A.G. The writing for original draft and editing K.F., B.S., A.L.C. and A.G. Figures and tables designed by B.S. and A.L.C. All authors have read and agreed to the published version of the manuscript.

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Institutional Review Board Statement: The study was approved by the Iowa Institutional Board Review (IRB #20230947, 7 November 2023).

Informed Consent Statement: A partial waiver of consent was approved by Iowa IRB for all participants included in the current study.

Data Availability Statement: De-identified data available upon request and with approval from respective institutional human subject's research review boards.

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Conflicts of Interest: The authors declare no conflicts of interest.

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Communication

An Interprofessional Approach to Developing Family Psychosocial Support Programs in a Pediatric Oncology Healthcare Setting

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Simple Summary: When a child is diagnosed with cancer, the entire family is impacted. Depending on the diagnosis and course of treatment, a variety of psychosocial stressors may be present, including but not limited to learning to care for a medically complex child, separating from other family members, loss of income or employment, or internal family conflict. Effective treatment includes psychosocial support for the entire family. Using both the Standards for the Psychosocial Care of Children with Cancer and their Families and the Pediatric Psychosocial Preventative Health Model (PPPHM), we created a unified Psychosocial Services department and Scope of Practice Grid to ensure interventions were developed by each discipline to comprehensively meet the needs of our families. Interventions were established across the framework of a three-tiered model of support based on the family's level of need. This was guided by the development of a multidisciplinary task force that includes parent advisors and staff.

Abstract: Background: The Standards for the Psychosocial Care of Children with Cancer and their Families provide a framework for the delivery of psychosocial care to families experiencing pediatric cancer. Similarly, the Pediatric Psychosocial Preventative Health Model (PPPHM) guides intervention approaches by identifying three tiers of psychosocial support based on a family's level of risk. Employing both the Standards and the PPPHM, we developed a comprehensive three-tiered approach to support the psychosocial needs of families in a pediatric oncology setting. Methods: After publication of the Standards, our institution merged existing psychosocial disciplines into one unified Psychosocial Services department. The new department worked to clearly define the role and scope of each discipline's practice to ensure the psychosocial needs of patients and families were being comprehensively met. Interprofessional workgroups were established to evaluate and enhance the psychosocial services offered to patients, siblings, and caregivers using a three-tiered model of support. Membership included representation from patients and parent/caregiver advisors to ensure their perspectives were included in program development. Results: Over ten new programs have been developed to enhance the support of families facing pediatric cancer. At the Universal tier, new programs available to all families include

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caregiver and sibling support groups, a caregiver podcast, and a relationship health toolkit and workshop. At the Targeted tier, psychosocial interventions and parent mentor supports were implemented. At the Clinical/Intervention tier, a partnership was developed with an external tele-mental health company to provide mental health services to caregivers with significant needs or preexisting mental health disorders. Conclusions: Given the complex needs of families facing pediatric cancer, use of an interdisciplinary approach is paramount to successful support throughout the treatment trajectory. By leveraging the expertise and strengths of diverse disciplines with the perspectives of patients and families, new psychosocial programs can comprehensively address the unique challenges of patients and families impacted by illness.

Keywords: psychosocial standards; pediatric oncology; preventative health model; psychosocial support programs; scope of practice; psychosocial oncology

1. Introduction

The psychosocial needs of children with cancer and their families are extensive [1–4]. Addressing these needs requires a multidisciplinary and systematic approach. Our institution has used the Pediatric Psychosocial Preventative Health Model (PPPHM) [5] and the Standards for Psychosocial Care of Children with Cancer and their Families [6] to guide us in the development and implementation of psychosocial support initiatives that will best support our patients and their families.

The PPPHM [5] employs a public health framework to consider both the challenges and resilience experienced by children and families in pediatric healthcare settings. Based on the unique presentation of each pediatric patient and their family, evidence-based psychosocial care can be implemented along different tiers to match individual risks and strengths. The Universal tier represents the largest group of patients and families, those with understandable distress but also many strengths and abilities to successfully cope. At this level, interventions typically include psychoeducation, family-centered support, and ongoing screening for indicators of higher levels of risk. The Targeted tier includes a smaller group of patients and families with acute distress and identifiable risks and needs. Interventions are specific to these risks and needs, and distress and coping continue to be monitored. Patients and families presenting with persistent and/or escalating distress and high risk are served by interventions within the Clinical/Treatment tier, which often includes involvement of a behavioral health specialist (i.e., psychologist and/or psychiatrist) and more intense psychosocial services. Utilizing this type of framework allows for comprehensive and preventative care for all families, with allotment of specialized services to those most in need. Moreover, systematic screening and implementation of risk-based intervention allows for more equitable and bias-free provision of care [7].

In 2012, the Psychosocial Standards of Care Project for Childhood Cancer (PSCPCC) was formed to identify and address existing gaps in psychosocial care for children with cancer. Following extensive research and collaborative meetings, the PSCPCC published the Standards for Psychosocial Care of Children with Cancer and their Families in 2015 [6,8]. These 15 evidence-based standards outline the specific psychosocial needs of pediatric oncology patients and their families, and all pediatric oncology institutions and care centers are encouraged to follow them. The Standards can also serve as a guide for institutions seeking to expand their existing psychosocial programs or create new initiatives.

A key aspect of the Standards for Psychosocial Care is their emphasis on the importance of interdisciplinary collaboration [9]. While influenced by each cancer center’s size, patient volume, funding, and staffing, various clinical disciplines can contribute to implementing the Standards. In a survey of 144 pediatric oncology programs, the majority (over 93%) reported employing at least social workers and child life specialists to meet the psychosocial needs of patients and families [10]. Other psychosocial disciplines often represented in care teams include psychology, psychiatry, neuropsychology, spiritual care, creative arts, and school personnel [10,11]. With multiple disciplines contributing to the care of pediatric patients and their families, there are rich opportunities for interprofessional collaboration.

In addition to engaging multiple disciplines in the delivery of health care and development of new programs, inclusion of the patient-family voice is critical to the success of a health care system. A partnership between health care staff and families is mutually beneficial and underscores the importance that families play in the quality and safety of a health care system [12,13]. Moreover, a health care system's commitment to patient-family-centered care (PFCC) reflects efforts to be continually responsive to the needs of each patient and family, recognizing that each family's needs are different. Importantly, collaboration with patients and families is one of the core tenets of PFCC, reflecting the vital role of the family voice in a health care system, including its development, implementation, and evaluation of programs [14,15].

Using both The Standards and the PPPHM, the Psychosocial Services department at our institution collaborated with parent/caregiver advisors to develop and implement various programs to support caregivers and siblings of our patients. Psychosocial support programs were established across three tiers of support, based on the family member's need. This paper outlines the development and implementation of these programs to provide a guiding reference for other institutions interested in establishing similar services.

2. Materials and Methods

Our children's hospital serves children and young adults with cancer, blood disorders, and other catastrophic diseases. Patients are referred to our institution from all 50 states and internationally. In 2024, we served almost 9870 patients from over 64 countries: 62% of patients identified as White, 32% as Black or African American, and 6% identified as Other. Of the total patients served, 6241 (63%) were treated for a cancer diagnosis. Most pediatric patients at our institution are accompanied by at least one adult caregiver for all outpatient appointments and inpatient hospitalizations. Often, patients are accompanied by multiple caregivers (e.g., parents, grandparents, etc.) and child or adolescent siblings. These immediate and extended family members often benefit from psychosocial support and programming.

Two years following the publication of the Standards [6], our children's hospital merged existing psychosocial disciplines into a unified department. These include child life, music therapy, psychology, school, social work, and spiritual care. Programs for teens and emerging adults, oncology transition, and staff support were later added. While each department and program has its own discipline leaders, they share a reporting structure directed by a psychosocial vice president, who is a pediatric psychologist. The department maintains close relationships with other services, such as psychiatry and palliative care. This restructure led to opportunities for aligned strategic planning, program development, clinical care delivery, and research and quality improvement efforts. To create caregiver and family supports, the newly formed department focused on clarifying scopes of practice of the different disciplines as well as the creation of a task force to specifically focus on identifying and meeting caregiver needs.

2.1. Development of a Scope of Practice Document to Enhance Collaboration

To enhance interprofessional collaboration and ensure observance of the Standards for Psychosocial Care, our Psychosocial Services department worked to clearly articulate the scope of practice for each psychosocial discipline at our institution [16]. In 2018, the directors of each psychosocial department convened multiple times alongside the department chief to discuss scopes of practice. Through this process, the group gained a better understanding of each discipline's roles while identifying areas of overlap and potential programmatic gaps in care. Over a period of six months, staff meetings and feedback sessions provided opportunities for open discussion and consensus. Clinical disciplines also ranked their expertise and effort in each service area to reduce effort duplication. The resulting Scope of Practice Grid (SPG) is a "living" document that is reviewed and modified when needed to reflect new programs or services, evolving research, or changes in clinical practices.

Ultimately, the SPG enhances interprofessional collaboration by highlighting the strengths and areas of practice expertise for each psychosocial discipline at our institution. This shared understanding and clear role delineation also helps reduce any potential for professional "turf wars". As we consider ways to enhance existing services or address gaps in psychosocial care, the identified areas of shared overlap help us determine which disciplines might be poised to implement new interventions. For example, our child life and social work departments recently partnered to launch a co-facilitated support group for siblings. Each clinician brings their discipline's unique strengths and expertise into the group sessions, ultimately enriching the psychosocial support provided.

Areas of overlap are also addressed in a weekly psychosocial rounds meeting. Psychosocial rounds provide an opportunity to review the needs of patients and families and plan for co-treatment opportunities. Psychosocial Services clinicians can meet separately with patients and family members to provide clinical support, or they may collaborate, using their combined skill sets to enhance a family's wellbeing. Psychosocial rounds are also a structured opportunity for staff to discuss any conflict or issues that arise due to role overlap and plan for how to best support the patient and family moving forward.

2.2. Development of Caregiver Support Task Force

A Caregiver Support Task Force was established to enhance the supports provided to caregivers to ensure alignment with Standard 6 ("Parents and caregivers of children with cancer should have early and ongoing assessment of their mental health needs. Access to appropriate interventions for parents and caregivers should be facilitated to optimize parent, child, and family well-being" [17]). Initially, the task force included one representative from each of the psychosocial services and three representatives from our PFCC program, including a caregiver advisor. As new programs were implemented and additional needs were identified, representation of the task force expanded to include additional staff from the Psychosocial Services department and the PFCC program, as well as members of Information Services, Patient Experience, and the palliative care service. Program development and implementation continued at a steady rate, and additional workgroups were established to capture the varying needs of different family members. While initial efforts focused exclusively on caregivers, programs were ultimately developed to address the needs of families more broadly, which dovetails nicely with Standard 4 ("All youth with cancer and their family members should have access to psychosocial support and interventions throughout the cancer trajectory and access to psychiatry as needed" [11]), Standard 7 ("Youth with cancer and their family members should be provided with psychoeducation, information, and anticipatory guidance related to disease, treatment, acute and long-term effects, hospitalization, procedures, and psychosocial adaptation. Guidance should be tailored to the specific needs and preferences of individual patients and families and be provided throughout the trajectory of cancer care" [18]), and Standard 10 ("Siblings of youth with cancer should be provided with appropriate supportive services. Parents and professionals should be advised about ways to anticipate and meet siblings' needs, especially when siblings are unable to visit the hospital regularly" [19]).

3. Results

To effectively address the psychosocial standards of care, we followed the PPPHM three-tiered model of support [1] to develop a three-tiered model of caregiver support (see Figure 1) tailored to meet each family's unique needs [20]. Recognizing that individuals and families have different preferences for receiving psychosocial support, we have been intentional to ensure programming opportunities are diverse. Some of the programs developed include psychoeducational materials and workshops, podcasts, support groups, animal-assisted therapeutic offerings, and individual mentoring or counseling. Clinicians across psychosocial services were instrumental in the development, implementation, and support of all programming, in collaboration with additional family-serving departments, such as Family Guest and Volunteer Services (see Table 1).

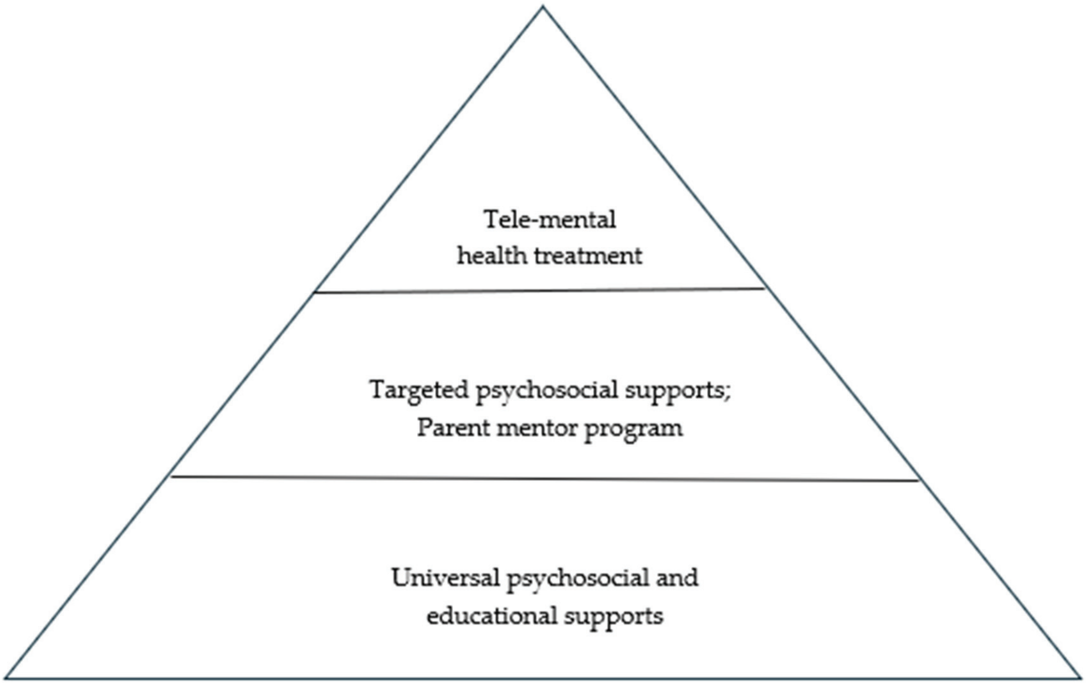

Figure 1. Three-tiered model of caregiver supports.

3.1. Universal Tier Programs

Caregiver Connections is a weekly support group for parents or other primary caregivers to find connection and meaning through shared experiences. The group aims to help decrease caregiver isolation by providing a space for caregivers to share about their child's illness and treatment. Caregivers also receive psychoeducation and anticipatory guidance, including coping skills to manage common emotions like fear, stress, and/or fatigue. Licensed social workers facilitate the groups to help ensure a safe and supportive environment. When possible, a parent advisor attends, reinforcing our model of interprofessional program development and support. Since the inception in spring 2023, 109 sessions of Caregiver Connections have been scheduled. While some sessions have had limited or no attendance due to various challenges (e.g., patient/caregiver schedule conflicts, hesitation to attend a group support offering, patient treatment demands, etc.), there have been 107 attendees in total. A recent caregiver attendee encouraged other parents to participate in Caregiver Connections by posting a written testimonial to a social media page whose membership comprises caregivers of patients at our institution.

Table 1. Involvement of psychosocial services clinicians in the conceptualization, implementation, and support of new programming.

	Child Life	Music Therapy	Psychology	Resilience Center	School Program	Social Work	Spiritual Care
Universal Tier							
Caregiver Connections	S	S	S	S	S	C/I	S
Sibling Connections	C/I	S	S	S	S	C/I	S
The Caregiver Project	C/I	C/I	S	S	S	S	S
Psychoeducational Workshops	C/I	C/I	C/I	C/I	C/I	C/I	C/I
Podcast	C/I	C/I	C/I	S	S	C/I	C/I
Paws at Play	C/I	C/I	C/I	C/I	C/I	C/I	C/I
Targeted Tier							
Individual Interventions	C/I	C/I	C/I	C/I	C/I	C/I	C/I
The Mentor Program	S	S	S	S	S	S	S
Clinical Tier							
Tele-Mental Health Program	S	S	C/I	S	S	C/I	S

Note: Conceptualization (C), Implementation (I), and Support (S).

While Caregiver Connections focuses on adults, Sibling Connections is designed to provide the siblings of our patients an opportunity to make meaningful connections with others and receive psychosocial resources and education to support their unique needs. This programming is a collaboration between two disciplines, with groups co-facilitated by a child life specialist and a licensed social worker. Six sessions of Sibling Connections have been scheduled since its launch in summer 2024, with 12 children and adolescents participating thus far.

To protect the privacy of our caregivers and siblings who are not identified patients of our institution, we do not document sessions of either group in an electronic health record or collect specific identifying data. As a result, we do not have a measure of how often an individual caregiver or sibling returns to group as a measure of success. We have considered exploring methods of measuring progress in the future, but at this time, our primary focus has been on implementing the programs and making them available to our families.

The Caregiver Project is another collaborative initiative aimed at meeting the individualized needs of caregivers. Facilitated by child life and music therapy, caregivers record personal stories that honor and capture their experiences. This serves as a form of memory-making and legacy building for the caregivers of our patients. Beginning in May 2024, the Caregiver Project has been offered monthly for an hour and encourages caregivers to participate as their schedule allows.

The Paws at Play program was created to provide support to patients and families through one-on-one and group visits with one of three trained facility dogs. The dogs are each utilized for specific interventions and work alongside clinicians in the following departments: child life, psychology, social work, and the school program. While the Paws at Play program was created as a Universal tier support, the facility dogs can be utilized within Targeted and Clinical/Treatment tiers as well. When providing feedback about this program, one of our facility dog handlers noted that "[the dog] provided a calming presence for a patient whose anxiety impacted their treatment...". Another handler recently shared the following about their patient: "A teenager recently received bad news about his illness. In the past, he never spoke to the doctor himself, and always asked his caregiver to do it for him. However, when the facility dog was there, he asked all his questions and was able to provide his input regarding his treatment preferences. The patient and his caregivers stated they felt it was because the facility dog was in the room providing him the comfort he needed to speak up". A third handler shared, "the facility dog served as the conduit for me to provide anticipatory grief counseling and support to a family whose patient was at the end of life".

To provide anticipatory guidance, we recently developed a psychoeducational toolkit and workshop to promote relationship health, resilience, and healthy communication. Content for the toolkit and workshop was developed by psychosocial providers utilizing interviews from couples with lived experience to create practical suggestions for supporting a partnership during a child's treatment. We plan to develop additional psychoeducational toolkits and accompanying workshops on various psychosocial topics in the future. Additionally, members of the Psychosocial Services department have worked collaboratively to contribute articles to a freely available website, *Together by St. Jude*™ [21], that provides anticipatory guidance and psychoeducation to patients and families within and outside of our institution. The site's content is aimed at supporting patients and families impacted by childhood cancer, blood disorders, and other catastrophic illnesses and is translated into 12 languages.

Finally, the Caregivers SHARE podcast [22] was created to provide a place for family caregivers to hear advice, reflection, encouragement, and real-life stories from families and hospital care providers. As of January 2025, two seasons of the podcast have been created. Over the past year, episodes have been downloaded 1746 times in 21 countries around the world. One caregiver listener shared the following feedback and consented to have it included in this article: "We are inpatient this week, and I found myself needing something for encouragement at a time when we are struggling. So much of [the podcast] resonates with me, and I was impressed by the honest and hard, but hopeful way, in which the information was shared".

3.2. Targeted Tier Programs

While all families can access programming from the Universal tier, families needing additional support can also access individual interventions with our psychosocial service clinicians as part of the Targeted tier. Because families often present with complex needs, interventions at the Targeted tier may also consist of co-treatment sessions between two or more psychosocial clinicians. Moreover, it is possible that individual sessions may build on information provided through programming at the Universal tier, in more depth.

A parent/caregiver mentor program was designed to meet the individualized needs of parents or adult family members of patients. Through the program, caregivers are matched with another caregiver whose child may have had a similar illness or treatment experience. Mentors provide insight into the treatment experience by drawing on skills or techniques they have learned to support coping. Both the mentor program and Caregiver Connections are scalable to serve a broad population. Patients and families at our institution have reported that one of their strongest supports is the friendship and connection with other families made through such programming or while at the hospital during treatment. A bereaved caregiver provided the following feedback about the mentor program and consented for it to be included in this article: "My mentor was a great asset to have during this difficult journey. . . We shared the stories of our children's cancer journeys and shed a few tears. She gave me insight on what grief could look like in the future".

3.3. Clinical/Treatment Tier Programs

The Clinical/Treatment tier addresses the most significant level of individual need. Caregivers with preexisting mental health problems may require continued treatment so they can best care for their child [23]. Alternately, they may become aware of the need for mental health services due to their change in circumstances [24]. It is often difficult for caregivers to access services in an unfamiliar community, and childcare resources may be limited [25,26]. For these reasons, we have partnered with an outside agency to provide tele-mental health services, including psychotherapy and medication management, to parents or primary caregivers with significant needs. Caregivers are referred to the tele-mental health provider by our social work department and are eligible for ten sessions per calendar year while their child is on active treatment. They are also eligible for ten additional sessions if their child is enrolled in hospice and an additional ten sessions if they become a bereaved caregiver. Privacy concerns are limited, as minimal information is shared between the telehealth company and our institution, and all documentation related to services is performed within the tele-mental health company's medical record.

This program began in late 2019. To date, a total of 488 referrals have been made to the tele-mental health program: 386 referrals were for therapy, 17 were for hospice caregivers, 81 for bereavement services, and 4 additional referrals were made without a specific designation. Not all caregivers who are referred to the program ultimately proceed with services; however, as of this date, the program has met the needs of 203 total families. This includes 155 caregivers whose children were receiving active treatment, 8 for hospice caregivers, 38 caregivers receiving bereavement services, and 1 referred without designation. Primary reasons caregivers did not receive services included caregiver refusal and inability of the tele-mental health company to reach them. Continuity of services has also been challenged by telehealth licensing laws, which require that clinicians be licensed in the state where a client is physically located [27]. Because our population is unique in that patients and families may reside anywhere in the United States or internationally, recipients of these services may have to shift providers when they return home from our institution. Fortunately, the tele-mental health agency we currently utilize has clinicians licensed in every US state, as well as those who are bilingual in Spanish and English. Because the agency is large, provider availability has not been a concern. While access to technology has not been an issue for most of our caregivers, we can provide a loaner device owned by the institution, if needed. In addition, flexible appointment times (e.g., evenings, weekends) and availability of hospital volunteers to watch the caregiver's child allow for increased practicality of receiving services.

4. Discussion

After merging existing psychosocial services into a unified department, we have worked to steadily refine and improve the care we provide to pediatric oncology patients and their families. We utilized the Standards for Psychosocial Care of Children with Cancer and their Families [6,8] to identify outcomes, and our system was modeled after the three-tiered structure discussed in the PPPHM [5]. Within this, psychosocial supports for caregivers were created to provide a wide array of resources in various routes or formats (e.g., written material, podcasts, in-person interactions, and virtual visits; individual versus group formats), delivered by various individuals (e.g., peer versus psychosocial clinician), and with different timing (e.g., drop-in groups, scheduled groups/activities, individually scheduled visits, and on-demand content like podcasts and written material). This allows caregivers some degree of freedom to self-identify interventions that could work best for them, consistent with prior research demonstrating the importance of caregiver choice and recognition that individual family needs will vary [28,29]. It also allows for a more equitable approach to caregiver support and the ability to increase the level of services for those with higher needs, while providing targeted interventions to those with moderate needs and universal support to all caregivers. Many of the supports offered at the universal and targeted tiers are of low or no cost to our institution. They are implemented by our psychosocial clinicians using materials that are already available (i.e., space for group sessions) or present within their respective departments.

While we believe there are many strengths to the psychosocial support programs we have created for our caregivers, there are several limitations to our approach that we must acknowledge. First, our hospital serves primarily pediatric oncology patients. Given that we do not specifically recognize caregivers as patients, and to avoid encountering concerns related to dual roles, provision of the Clinical/Targeted tier of individualized mental health care for caregivers has been delivered through referral to tele-mental health providers. While this eliminates the potential for dual roles and may make it easier for those referred to feel comfortable that their own care will not influence their child's care, it also may require a caregiver to take time away from their child to receive this needed treatment. While the use of telehealth limits this, not all caregivers are amenable to tele-mental health. We are aware that some other institutions will provide direct counseling and medication management to caregivers and identify them as patients of their facility as well. This approach has the benefit of ease of access and possibly greater familiarity of caregivers with the person and place delivering this care.

Another significant limitation to our work is that many hospitals and psychosocial service providers working within them will not have access to the resources to implement all of these supports, making exact replication of our model challenging. However, there are likely some supports that could be added to almost any other facility without a significant strain on limited resources. Our Caregivers SHARE podcast [22] and *Together by St. Jude*™ website [21] are both freely available to the general public and are one way to provide or guide improvements to caregiver supports. In addition, printed education materials are an easy-to-develop and cost-effective intervention approach [30]. Lastly, development of a Scope of Practice document could allow psychosocial providers at other institutions to evaluate their current practices and identify areas of improved psychosocial supports to prioritize.

5. Conclusions and Future Directions

Overall, our work to develop and implement programs to better support families of youth with cancer provides the groundwork for future clinical and research endeavors. While we recognize not all of our interventions may be feasible or replicable at other institutions, we hope that our work can be a model for other institutions and psychosocial clinicians to develop and refine their own approaches to the psychosocial care of patients and their families alike. In the future, it will be important to perform prospective studies and to include objective measurement of program efficacy. As an example, our institution is currently piloting a caregiver distress screening tool that could provide insight into changes in caregiver distress associated with program usage.

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Article

Integrated Psychological Services in Pediatric Oncology: Caregiver Perspectives at Diagnosis

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Simple Summary: Increased distress is a common initial response for youth and their families following the diagnosis of childhood cancer. The New Oncology Program in Psychology (NOPP) was established to provide education and information on what to expect at diagnosis, as well as ways to support coping with treatment. This project examined how NOPP participation relates to caregivers' perceptions surrounding the navigation of difficult emotions, and of the awareness of potential cognitive/academic challenges their child may experience. Caregivers completing NOPP visits felt more informed about difficult emotions and how these may change over time. They felt more prepared for and equipped with strategies and support to manage difficult emotions. Lastly, caregivers felt more aware of and prepared for the potential effects of the diagnosis and treatment on patient cognitive/academic functioning. The results highlight important domains for universal assessment and intervention with pediatric patients and families at the time of a new cancer diagnosis.

Abstract: Background/Objectives: Pediatric oncology patients and families are at risk for increased distress at diagnosis. The New Oncology Program in Psychology (NOPP) aligns psychological care with the established standards of care at diagnosis. This project aimed to evaluate NOPP and understand the differences between caregivers' perceptions of feeling informed and prepared to navigate psychosocial concerns for those who did and did not receive psychological services at diagnosis. Methods: A survey was administered via a virtual platform. Frequency analyses summarize caregiver experiences and concerns. Mann–Whitney U tests assess the differences in caregiver knowledge and preparedness between caregivers who did and did not receive psychological services. Results: Caregivers reported difficult emotions at diagnosis and expressed concern for the impact of diagnosis and treatment across broad domains of patient functioning. Caregivers of patients who received psychology consultation felt more informed about difficult emotions and how these may change over time. They felt more prepared and equipped with strategies to manage difficult emotions. Caregivers of patients who completed a cognitive assessment also felt more informed and prepared regarding the potential effects of the diagnosis and treatment on patient cognitive/academic functioning. Conclusions: Psychological services were associated with caregivers' positive perceptions surrounding the management of difficult emotions and with their knowledge regarding the cognitive/academic impact. The results inform the ongoing modification of NOPP.

Keywords: psychosocial care of children with cancer and their families; psychosocial concerns; universal assessment and intervention; psychoeducation; anticipatory guidance

1. Introduction

In 2024, it is estimated that 9620 children (birth to age 14) and 5290 adolescents (age 15–19) will be diagnosed with cancer in the United States [1]. The psychosocial impact of a cancer diagnosis on pediatric patients and their families is well-documented. The impact following a pediatric oncology diagnosis is variable and can include mood, behavior, and cognitive concerns for the patient [2,3]. Similarly, a pediatric cancer diagnosis can affect caregivers. Specifically, caregivers of pediatric oncology patients experience more symptoms of depression and anxiety following diagnosis compared to the general population [4]. While most pediatric patients and their families go on to experience healthy adjustment and coping [5], it is normative for families facing life-threatening illness to exhibit distress. There are multiple disease-related milestones throughout the cancer trajectory that are characterized by increased distress and the potential for emotional, behavioral, and social difficulties [2,4,6,7]. A targeted psychosocial assessment at these critical timepoints fosters the provision of evidence-based intervention [8], which, in turn, promotes improved adjustment and engagement in adaptive coping [9]. Diagnosis is a particularly salient timepoint for patients and families as they come to terms with a serious illness and adapt to new routines and the medical environment. As such, the timely provision of education and support is warranted to optimize coping and facilitate positive adjustment [8].

The Pediatric Psychosocial Preventative Health Model is a tiered approach to care that stipulates all families should receive universal intervention and support, and additional intervention should be provided based on need [8,10]. This model aligns with the Standards for the Psychosocial Care of Children with Cancer and Their Families (The Standards) which were published in 2015 [11] and provide a framework for the provision of psychosocial services throughout the cancer trajectory. Standards of particular importance to the current project include Psychosocial Assessment [8], Monitoring and Assessment of Neuropsychological Outcomes [12], Anticipatory Guidance and Psychoeducation [13], and Psychosocial Interventions and Therapeutic Support [9]. The Psychosocial Assessment Standard urges the assessment of all families early in the oncology experience to identify the risk and resiliency factors that may impact coping and adjustment, as well as opportunities to provide intervention to families with warranting needs [8]. Similarly, The Psychosocial Interventions and Therapeutic Support Standard stipulates that intervention and support should be available to pediatric oncology patients and their families across the cancer trajectory, including at the time of diagnosis [9]. The Monitoring and Assessment of Neuropsychological Outcomes Standard calls for cognitive surveillance for patients at risk of experiencing disease- or treatment-related changes in functioning [12]. Lastly, The Anticipatory Guidance and Psychoeducation Standard states that patients and families should be given information on all aspects of their diagnosis and treatment, including information on what they may expect throughout the pediatric cancer trajectory. This education can reduce distress and negative symptomology and improve relationships and communication between the family and medical team [13].

At St. Jude Children's Research Hospital (SJCRH), the New Oncology Program in Psychology (NOPP) was developed to align psychology services with the aforementioned standards, offer standardized assessment and intervention services, and improve the psychological care of patients and their families at diagnosis. Through NOPP, all new oncology patients are offered the opportunity to be systematically and prospectively assessed and provided with appropriate intervention in response to presenting concerns near the time of diagnosis or initiation of care at SJCRH. Importantly, although the offering of these services is universal, the delivery of services is tailored and individualized to ensure that a patient's specific disease and treatment characteristics are considered, that services are culturally responsive and demonstrate cultural humility, and that services are consistent with trauma-informed care principles. Within that context, NOPP focuses on the identification of individual and familial factors that may impact the child's adjustment to and coping with diagnosis and treatment. Importantly, NOPP extends beyond distress screening and includes a psychology consultation for all oncology and transplant/cellular therapy patients and a brief cognitive assessment for those patients who do not receive serial cognitive monitoring as a part of their therapeutic protocol. The initial consultation through NOPP (NOPP Consult) includes a clinical interview which assesses patient/family history, current functioning, sociocultural factors, and patient health-related behaviors.

Additionally, patient functioning is assessed via validated self- and parent-report measures. Universal intervention which aligns with family values and preferences, including developmentally tailored anticipatory guidance and psychoeducation, is also provided at this visit. Neurocognitive and academic functioning are assessed through NOPP via a brief, targeted evaluation (NOPP Brief Cognitive Assessment). Following the NOPP Consult and Brief Cognitive Assessment, a psychosocial support plan is developed, including plans for ongoing psychological intervention and referrals to other psychosocial disciplines, as indicated. NOPP aims to transform psychology services at SJCRH from a consult-based model to an integrated model of psychological care consistent with The Standards [14], ensuring all oncology and transplant patients receive equitable psychological support.

NOPP was developed and evaluated following the CDC's Framework for Program Evaluation in Public Health [15]. Stakeholder input was a cornerstone of NOPP's early development. Prior to the launch of NOPP, the SJCRH Patient Family Advisory Council (PFAC), a group of advisers who partner with staff to share the patient and family perspective on various initiatives, provided support for NOPP and offered feedback regarding the scope of the program and timing of services. Additionally, input and feedback from patients, families, hospital administration, medical teams, psychosocial clinicians, and subject matter experts was obtained. The current project further engaged stakeholders more broadly by seeking feedback regarding NOPP from the larger patient and family community. In NOPP's development, the senior author, psychology leadership, and hospital administration collaborated to create and support a program to systematically and prospectively provide universal assessment and intervention to all new patients within 1–2 months of their diagnosis or initiation of care. The goals of NOPP include providing an assessment and intervention to reduce distress and increase families' perception of feeling informed, prepared, and equipped to manage potential psychosocial and cognitive sequela associated with a new pediatric oncology diagnosis. Following the implementation of NOPP, the authors deemed it critical to evaluate the impact of the program and solicit feedback from stakeholders in the context of ongoing program evaluation. Therefore, the current survey was developed with input from subject matter experts and feedback from interdisciplinary collaborators. One goal of the survey was to assess the concerns of families and identify the differences between those who did and did not participate in NOPP. The survey was distributed to stakeholders (i.e., caregivers and young adult patients) to assess their perceptions of feeling informed, prepared, and equipped to manage potential psychosocial concerns. The survey was hosted on St. Jude Voice, a virtual advisory platform where patients and families are surveyed for their input regarding clinical initiatives and other patient-care-related issues. To justify the conclusions, the survey results were statistically analyzed and interpreted. From these findings, recommendations will be made to improve the impact of the program. Additionally, this article serves to disseminate the findings and share the recommendations to the larger academic community. Lastly, the project results and the value of families' participation will be shared with stakeholders in a newsletter to the larger St. Jude Voice community [15].

The current project represents the first effort since the launch of NOPP to understand family perceptions of psychology services near the time of diagnosis or initiation of care at SJCRH. Specifically, the current project aims to understand the psychosocial concerns and difficulties experienced by patients and their families near the time of diagnosis. The second aim of the current project is to examine the differences in feeling informed of and prepared to navigate potential psychosocial challenges between those who met with a psychologist for a consult near the time of diagnosis or initiation of care at SJCRH and those who did not. Finally, this project seeks to investigate the differences between those who completed a neurocognitive assessment near the time of diagnosis or initiation of care at SJCRH and those who did not as it relates to caregiver perceptions of feeling informed of and prepared to navigate potential cognitive/academic challenges.

2. Materials and Methods

2.1. Human Subjects Protection

Although program evaluation projects do not require approval by the Institutional Review Board at the authors' institution, the Institutional Review Board was consulted, and the project was determined to be non-human subject research.

2.2. Materials

The survey consisted of 38 questions. A five-point Likert scale was used for most questions and collapsed during analysis into a three-point scale (strongly agree/agree, neither agree or disagree, and strongly disagree/disagree). Strongly agree and agree were indicative of endorsement of the experience.

2.3. Procedures

The survey was developed with input from subject matter experts and feedback from interdisciplinary collaborators to be consistent with surveys used in program evaluation projects examining clinical programs within pediatric oncology [16]. The survey was administered via an online platform (St. Jude Voice) in English. St. Jude Voice has 581 caregivers, and 41 young adult patients enrolled. The majority of enrolled caregivers identify as female (90.19%), identify as White (78.31%), reside in the United States (93.29%), and have completed treatment (53.97%) The survey link was emailed to St. Jude Voice members and remained open for three weeks. One week prior to the survey closing, a reminder was emailed to eligible caregivers and patients who had not completed the survey. No incentives were provided for participation. NOPP was gradually implemented across diagnostic oncology clinics and reached full implementation (i.e., services offered to all new oncology and transplant/cellular therapy patients) in 2023. As such, caregivers and patients on St. Jude Voice are varied regarding whether or not they were offered NOPP services. This allows for a comparison between participants who completed a Consult and/or Brief Cognitive Assessment near the time of diagnosis or initiation of care at SJCRH (i.e., within 1–2 months of arriving to SJCRH) and those who did not. Furthermore, this allowed us to understand whether those who did not participate in NOPP believe a consult and/or an assessment would have been beneficial near diagnosis.

2.4. Analyses

To better understand if participation in the NOPP Consult and Brief Cognitive Assessment differed based on demographic variables (i.e., age, gender, and race), ANOVAs were utilized. For aim 1, frequency analyses were used to calculate the percentage of participants who expressed worry in the assessed domains and the percentage of individuals who expressed experiencing difficult emotions at the time of diagnosis. For aims 2 and 3, two Mann–Whitney U tests were used to compare the differences in feeling informed of and prepared to manage potential challenges for those who did and did not complete a Consult and Brief Cognitive Assessment near time of diagnosis or initiation of care at SJCRH. Those who could not recall their participation in psychology services near the time of diagnosis or initiation of care at SJCRH were not included in the analyses for aims 2 and 3. Mann–Whitney U tests were selected as the data were not normally distributed [17]. Five caregiver participants were removed prior to any analysis as they did not answer any of the questions related to the three aims. Additionally, all six young adult patients were removed, given the small sample size.

3. Results

3.1. Demographics

In total, 115 individuals completed the survey, with 109 identified as primary caregivers for the patient and six identified as young adult patients. Given the small number of patient respondents, this report will focus only on the response of caregivers. The majority of the caregiver sample identified as White (n = 98) and female (n = 95). See Table 1 for additional demographic information. The response rates for caregivers who began the survey and completed the survey were 19.62% and 18.76%, respectively. There were no significant differences in Consult and Brief Cognitive Assessment participation based on race or gender; however, there was a significant difference in participation in the Consult and Brief Cognitive Assessment based on age (F(7,79) = 2.51, $p = 0.022$; and F(7,82) = 3.51, $p = 0.002$, respectively). Younger children were more likely to participate in the Consult and Brief Cognitive Assessment.

Table 1. Demographic Information.

	N	%
Age at Diagnosis		
Less than 1 year old	13	12.04
1–2 years old	18	16.67
3–4 years old	8	7.41
5–7 years old	18	16.67
8–10 years old	13	12.04
11–14 years old	21	19.44
15–17 years old	13	12.04
18–23 years old	4	3.70
Missing	1	0.01
Primary Clinic		
Leukemia/Lymphoma	30	27.52
Transplant	11	10.09
Solid Tumor	33	30.28
Neuro-Oncology	32	29.36
Genetic Predisposition	3	2.75
Radiation Oncology	10	9.17
Survivorship	11	10.09
Ethnicity		
White	98	89.91
Black	3	2.75
Hispanic	4	3.67
Asian/Pacific Islander	3	2.75
Native American	0	0.00
Prefer not to answer	1	0.01
Gender		0.92
Female	95	87.16
Male	13	11.93
Other	1	0.92

Note. Participants were permitted to belong to multiple primary clinics. Therefore, the percentage of individuals in primary clinic sums to greater than 100%.

3.2. Psychosocial Concerns and Difficulties at Diagnosis

Almost all caregivers (99.00%) reported experiencing difficult emotions at the time of their child's diagnosis. Many caregivers expressed worry regarding the impact the diagnosis and treatment might have on the patient's friendships and relationships (73.27%), patient's emotional wellbeing (94.06%), patient's behavior (80.20%), patient's health-related behaviors (i.e., sleep, appetite, or pain; 93.07%), and patient's cognitive skills (82.00%). The frequency of caregiver worry is depicted in Table 2.

Overall, caregivers reported the following domains to be a very or somewhat important concern they held for the patient at diagnosis: meeting developmental milestones (81.37%); experiencing difficulties related to emotional (95.10%), behavioral (63.72%), or social (73.53%) functioning; understanding medical diagnosis and experience (86.27%); coping with medical experiences (91.18%); sleep (75.49%); fatigue (86.28%); appetite (84.32%); pain (90.20%); parent coping (94.12%); and sibling coping (83.33%). See Figure 1 for additional details regarding caregivers' identified concerns at diagnosis.

Table 2. Caregiver Worry for Impact of Diagnosis and Treatment on Patient Functioning.

	n	%
Friendships and relationships	74	73.27
Emotional wellbeing	95	94.06
Behavior	81	80.20
Health-related behaviors (sleep, appetite, or pain)	94	93.07
Cognitive skills	82	82.00

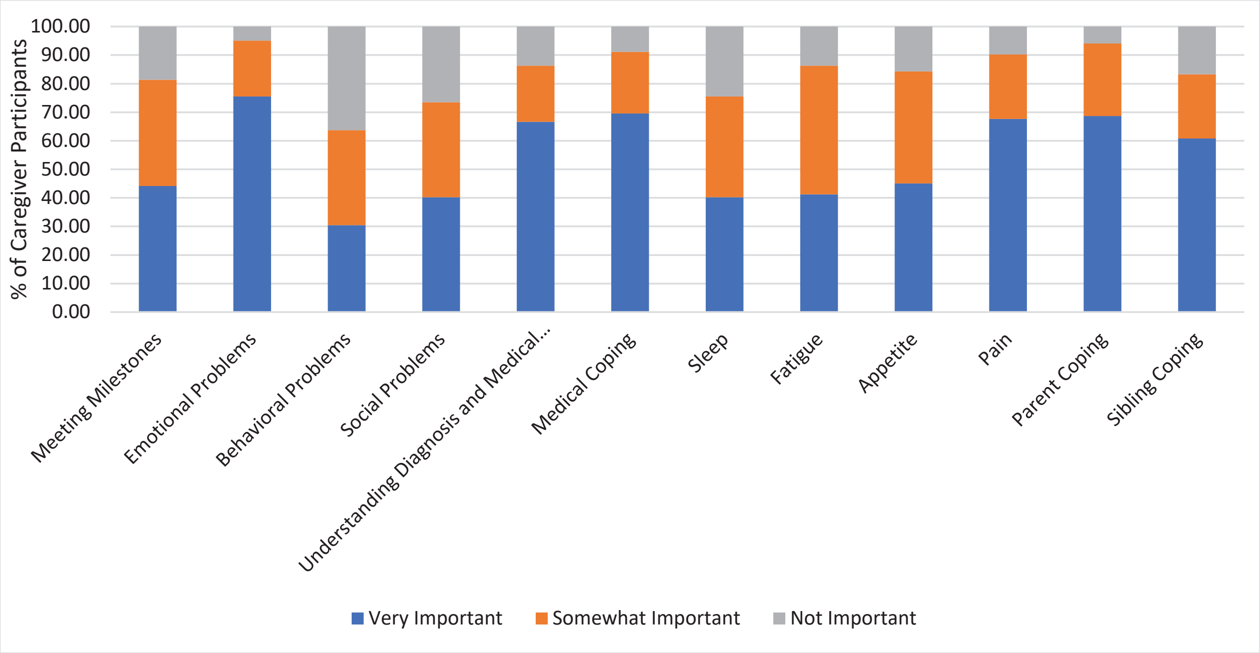

Figure 1. Caregiver concerns at diagnosis.

3.3. Psychology Consult

Fifty-one caregivers (46.79%) reported the patient met with a psychologist for a consultation within two months of initiating care; of these caregivers, 86.00% reported the timing of this visit was appropriate. Of the caregivers who reported the patient did not meet with a psychologist or were unable to recall their participation, 80.70% ($n = 46$) thought meeting with a psychologist would have been helpful.

Caregivers of patients who met with a psychologist, in comparison to those who did not, were more likely to report feeling informed about difficult emotions often experienced by patients and families near the time of diagnosis and how these emotional experiences may change over time (U = 490, $p = 0.03$). Relatedly, they more often reported feeling like they knew what to expect regarding their emotional experience (U = 503, $p = 0.04$) and were equipped with strategies and access to available supports to help manage difficult emotions (U = 507, $p = 0.05$). Additionally, they were more likely to report feeling informed about the potential impact of diagnosis and treatment on the patient's emotional wellbeing (U = 520, $p = 0.03$), and equipped with strategies and access to available supports to improve the patient's emotional wellbeing at diagnosis and throughout treatment (U = 537, $p = 0.05$). No differences were found regarding caregivers feeling informed, prepared, or equipped with strategies to manage patient concerns related to social functioning, behavior changes, and health-related behaviors (Table 3).

Table 3. Caregiver Perceptions of Being Informed, Prepared, and Equipped at Diagnosis. * $p \leq 0.05$.

	Consult		**No Consult**		**Brief Cognitive Assessment**		**No Brief Cognitive Assessment**				
	M	**SD**	**M**	**SD**	**M**	**SD**	**M**	**SD**	***U***	***p***	**η^2**
Informed											
Difficult emotions and the emotional experience might change	1.67	0.77	2.11	0.83					490.00	0.026 *	0.06
Impact on the friendships and relationships	2.00	0.84	2.36	0.83					526.00	0.070	0.04
Emotional wellbeing	1.58	0.77	2.03	0.89					520.00	0.025 *	0.06
Impact on the patient's behavior	1.94	0.80	1.97	0.87					699.50	0.904	0.00
Impact on the patient's sleep, appetite, and/or experience of pain	1.47	0.73	1.63	0.81					601.50	0.356	0.01
Impact on the patient's thinking skills and school performance					1.54	0.79	1.90	0.86	611.50	0.047 *	0.05
Prepared											
Regarding the emotional experience adjusting to diagnosis and treatment	1.94	0.85	2.6	0.83					503.00	0.038 *	0.06
Regarding friendships and relationships with others	1.98	0.78	2.25	0.80					556.00	0.143	0.03
Regarding the emotional wellbeing	1.85	0.82	2.17	0.83					573.00	0.109	0.03
Regarding the patient's behavior	2.02	0.83	2.27	0.78					615.00	0.197	0.02
Regarding the patient's sleep, appetite, and/or experience of pain	1.62	0.83	1.93	0.91					548.50	0.131	0.03
Regarding the patient's thinking skills and school performance					1.56	0.79	2.07	0.91	556.00	0.011 *	0.08
Equipped with Strategies and Supports											
Manage difficult emotions	1.94	0.80	2.32	0.77					507.50	0.045 *	0.05
Improve friendships and relationships with others	2.04	0.76	2.29	0.85					563.00	0.165	0.03
Improve emotional wellbeing	1.77	.78	2.17	0.87					537.00	0.046	0.05
Increase positive behavior	1.83	0.78	2.17	0.87					563.50	0.088	0.04
Improve sleep, appetite, and/or pain	1.56	0.81	1.83	0.83					545.00	0.117	0.03
To help with thinking skills and school performance					1.64	0.81	1.93	0.88	657.00	0.137	0.03

3.4. Brief Cognitive Assessment

Almost half of caregivers (45.56%; *n* = 41) reported the patient completed a cognitive assessment within two months of diagnosis or initiating care; of these, thirty (73.17%) reported the timing of this assessment was appropriate. Of those caregivers who reported the patient did not complete a cognitive assessment or were unable to recall their participation, 28 (57.14%) thought this assessment would have been helpful.

Caregivers who reported the patient completed a cognitive assessment, in comparison to those who did not, were more likely to report feeling informed about the potential impact of diagnosis and treatment on the patient's cognitive skills and school performance (U = 611.50, $p = 0.05$). They were also more likely to report feeling prepared in relation to expectations regarding the patient's cognitive skills and school performance at diagnosis and throughout treatment (U = 556, $p = 0.01$). No differences were found regarding caregivers feeling equipped with strategies and access to available supports in this domain (Table 3).

4. Discussion

The time of diagnosis is known to be one of increased distress for pediatric patients diagnosed with cancer and their families [2,18–20]. Psychologists are poised to offer a psychosocial and cognitive assessment, as well as intervention services, to mitigate distress when integrated into pediatric cancer care. NOPP offers a culturally sensitive assessment and intervention at the universal level [8,10] for all patients and their families, with goals of helping them understand what to expect throughout the cancer trajectory, learn strategies to promote patient coping and overall wellbeing, and achieve a greater understanding of access to available supports. Furthermore, NOPP facilitates the provision of psychological services at the targeted and clinical level [8,10] when indicated.

The results of the current survey suggest that caregivers of patients who receive a psychological consultation near the time of diagnosis or initiation of care were more likely to feel informed of and prepared for challenging emotions often experienced by patients and their families. Importantly, they also noted feeling equipped with strategies and access to available supports to promote coping with these challenging emotional experiences. Similarly, caregivers of patients who received a psychology consultation were more likely to report feeling informed of and prepared for the potential impact of diagnosis and treatment on the patient's emotional wellbeing and equipped with strategies and access to available supports to promote the patient's emotional functioning throughout the cancer trajectory. These findings are consistent with the aims of NOPP regarding the provision of anticipatory guidance and psychoeducation related to patient and family emotional experiences and patient emotional functioning. There was no significant difference between caregivers who reported the patient received a psychology consultation and those who did not on feeling informed, prepared, and equipped across the domains of social and behavioral functioning and health-related behaviors (e.g., sleep, appetite, experience of pain, etc.), suggesting a need for improved anticipatory guidance and psychoeducation across these areas.

Similarly, caregivers who reported the patient received a cognitive assessment near the time of diagnosis or initiation of care were more likely to express feeling informed of the potential impact of diagnosis and treatment on the patient's cognitive functioning, as well as prepared for what to expect over time in this domain. However, there was no significant difference between those who did and did not complete a cognitive assessment on caregivers feeling equipped with strategies and access to available supports to promote the patient's cognitive functioning and academic achievement, suggesting an important area for improved psychoeducation.

Importantly, the target domains for NOPP assessment and intervention align with those identified as important by caregivers, supporting the use of these domains as a model for universal psychological assessment and intervention at diagnosis.

4.1. Limitations

Due to limited participation within the young adult patient population, the patient perspective is not included. This is consistent with the existing literature highlighting the challenges of obtaining AYA participation in oncology research [21]. Limited AYA participation in the current project highlights an area for growth within the institutional virtual advisory platform and subsequently generated interest in intentional recruitment efforts to expand the patient voice, which are currently ongoing. While the platform remains limited to patients aged 18 and older, it is imperative that we seek and maximize direct patient feedback when developing and evaluating clinical programs. Thus, additional avenues to obtain the patient perspective for those under age 18 are needed. Furthermore, the sample was predominantly White and female. The limited racial and gender diversity limits the generalizability of the findings. Future work is needed in order to confirm the current findings in diverse samples.

While the survey assessed perceptions of psychology services near the time of diagnosis or initiation of care, defined as within 1–2 months of arriving to SJCRH, and allowed for a comparison between those who received a psychology consult and/or cognitive assessment and those who did not, it is not known whether patients who received psychological services did so as a part of NOPP or if they were coincidentally referred for psychology services within the NOPP timeframe. Regardless of the mechanism by which patients received these services, the information gleaned from the current survey will serve to inform continued improvement efforts for NOPP and psychology services offered near the time of diagnosis more broadly.

Additionally, the current project did not assess whether families who did not complete NOPP declined to participate or were not offered NOPP based on the timeline of implementation. To ensure the services align with families' needs and values, future modifications to NOPP should track the rate of declined participation and the reason for the decline. Further, it would be of interest to understand if those who decline differ demographically or in their perceived level of knowledge, preparedness, or feeling equipped. This information could be utilized in future quality improvement efforts—efforts that should, undoubtedly, include diverse family representation on the team.

The perception of psychological services differs based on cultural factors [22,23]. It is acknowledged that psychology services may not be perceived favorably by all families. In an attempt to reduce stigma, NOPP provides psychological services universally, to all new patients. The universal nature of NOPP is shared with patients and families when introducing the service. Nonetheless, clinicians remain mindful that sociocultural differences likely impact families within the medical and psychological setting [24,25]. Therefore, clinicians continue to engage in trainings in cultural humility and seek consultation from other mental health professionals and cultural experts within the institution.

Universal screening and intervention aim to provide equitable and personalized resources to all families [26]. Nonetheless, providing widespread screening and intervention has not yet been implemented by all pediatric oncology centers [27]. At the majority of pediatric oncology centers, most psychological services are provided exclusively on a referral basis. Barriers to the systematic implementation of psychosocial standards of care stem from resource limitations, namely, funding, staffing, and time constraints [28]. Given the barriers, policy changes are necessary in order to ensure resources are available to provide equitable and culturally responsive care to all patients and families [28]. It is recognized that some institutions are more resource-rich than other institutions. In an effort to increase equity in access, psychoeducation and anticipatory guidance, one aspect of the NOPP universal intervention package, is widely available for all individuals regardless of treatment location [29–31].

4.2. Future Directions

The findings of the current project will be used to inform the continuous improvement of NOPP, with specific attention paid to identified areas of needed improvement, including anticipatory guidance and psychoeducation related to aspects of patient behavioral, and social, cognitive, and academic functioning, as well as health-related behaviors, including sleep, appetite, and pain. Some of these data will be used as baseline data in the Plan phase of the Model for Improvement's Plan–Do–Study–Act framework [32].

While the continuous quality improvement of NOPP is underway, with results from the current project informing the Plan phase of the Model for Improvement's first intervention cycle [32], future research is also needed in order to understand the impact of NOPP on patient outcomes, both psychological and medical, extending throughout the cancer trajectory and into survivorship.

5. Conclusions

The current project demonstrates that caregivers experience difficult emotions at the time of diagnosis and express concern for the impact of diagnosis and treatment across broad domains of patient functioning. These domains should be considered in the provision of universal assessment and intervention that align with the Standards for the Psychosocial Care of Children with Cancer and Their Families. The differences (and lack thereof) in feeling informed, prepared, and equipped between caregivers of patients who did and did not participate in psychology services near diagnosis will guide the continuous quality improvement of NOPP.

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Article

Patient Acceptability of the First Integrative Pediatric Oncology Unit in Spain—The Pediatric Cancer Center Barcelona Experience: A Retrospective Study

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Simple Summary: This study explored the introduction of a new approach for treating children with cancer at the Pediatric Cancer Center in Barcelona, Spain. Traditionally, cancer treatment focuses on conventional approaches including surgery, chemotherapy, and radiation, but a significant percentage of families seek additional therapies, such as acupuncture and aromatherapy. This study aims to assess how well these complementary therapies are accepted by patients and their families. Over two years, the center offered these therapies to 433 patients, with the majority of families welcoming additional treatments. Very few rejected them, demonstrating that integrating these therapies into pediatric cancer care is feasible and well-received. The findings suggest that such therapies could play a valuable role in supporting young cancer patients, potentially encouraging more healthcare centers to adopt similar practices.

Abstract: Background/Objectives: Pediatric cancer patients and their families are increasingly combining conventional treatment with complementary therapies. These therapies are not covered by most public healthcare systems, and Spain is not an exception. To address this need, the Pediatric Cancer Center Barcelona, at the Hospital Sant Joan de Déu (Spain), established the first integrative pediatric oncology unit in 2019. The objective of this study is to describe the feasibility of implementing a pediatric integrative oncology unit, in terms of the acceptance of the interventions, as well as to present initial data on the care activities; Methods: This is a retrospective single-center study, conducted in the Pediatric Cancer Center Barcelona, at the Hospital Sant Joan de Déu. Data from patients during a two-year period were collected by reviewing medical records in a pseudonymous manner; Results: From 1 September 2019 to 30 September 2021, the unit was visited by 433 patients. The median age of patients was 9 years [range 0–34 years], with 266 boys (61.4%) and 167 girls (38.6%). Of these patients, 90.1% were in active treatment, 7.6% were survivors, and 2.3% were at the end of life. Acupuncture was recommended to 227 patients, with a 94.7% acceptance rate. Aromatherapy was recommended to 114 patients, with a 100% acceptance. The reflexology team visited 129 patients, delivering a total of 414 sessions, with a 96.1% acceptance.; Conclusions: The findings of our study support the feasibility of implementing an Integrative Pediatric Oncology Unit within a patient-centered care model in a comprehensive pediatric cancer center. The high acceptance rates of various

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complementary therapies highlight their potential role in enhancing supportive care for pediatric oncology patients.

Keywords: acupuncture; children; adolescent; oncology; integrative oncology; pediatrics; cancer; pediatric oncology

1. Introduction

Childhood developmental cancer refers to a group of diseases that arise during the formation of organs and tissues as part of the human developmental process. It can affect infants, children, adolescents, and young adults [1,2]. In most cases, the causes of these cancers remain unknown without obvious predisposition factors, although they may be related mostly to epigenetic factors during cellular development [3,4]. These potential lethal conditions during the first decades of life can have a strong physical and emotional impact on the patient and their family [5]. To cope with these challenges, many patients and families turn to complementary therapies, particularly when conventional allopathic treatments fail to provide sufficient relief. Complementary therapies are defined as medical practices and products not typically part of conventional medical care but used alongside standard treatments to improve patient well-being and quality of life [6]. Studies indicate that between 31 and 84% of families of oncologic patients combine conventional allopathic medicine with complementary therapies such as acupuncture, massage, meditation, herbs, homeopathy, and dietary supplements [7–9].

The World Health Organization (WHO), in its Traditional Medicine Strategy 2014–2023, highlights that the increased interest in traditional and complementary therapies demands its closer integration into healthcare systems [10]. The strategy aims to support Member States in harnessing the potential contribution of complementary therapies to health, well-being, and people-centered healthcare, and to promote the safe and effective use of complementary therapies through the regulation of products, practices, and professionals. This emphasis aligns with a growing acknowledgment of the importance of holistic approaches in addressing not only the physical aspects of diseases but also their emotional, social, and psychological dimensions, particularly in pediatric oncology, where patient vulnerability is compounded by developmental and familial factors [11–14]. The overall use across countries of these complementary treatments has increased in the last 30 years, revealing a real social demand [15–17]. In a recent study conducted in Germany, 40% of interviewed parents reported using complementary treatments and wished for their incorporation into the public health system, even if that would imply the individual coverage of its cost [18].

Integrative medicine is the combination of complementary therapies with conventional medical treatments in a coordinated manner, focusing on a holistic, patient-centered approach to care [6]. In the context of cancer care, integrative oncology extends this concept by incorporating mind and body practices, natural products, and lifestyle modifications with conventional cancer treatments [19,20]. The goal is to optimize health and quality of life across the cancer care continuum while empowering patients to become active protagonists in their care [21]. In 2017, the American Academy of Pediatrics (AAP) published a guide on pediatric integrative medicine, acknowledging the increased use of complementary therapies in this age group [22]. The guide reviewed evidence supporting therapies such as acupuncture, aromatherapy, and reflexology. Acupuncture was highlighted for its efficacy in reducing pain, nausea, and anxiety, particularly in oncology settings. Aromatherapy was recognized for its potential to enhance psychological well-being and alleviate symptoms like stress and fatigue through the use of essential oils. Reflexology, though supported by more limited evidence, was noted for its benefits in promoting relaxation and improving overall comfort. The AAP emphasized the importance of using these therapies alongside conventional treatments, recommending their application in clinical practice when delivered by certified practitioners and supported by emerging evidence [23].

Integrative oncology units are increasingly spreading worldwide, responding to the demand for complementary treatments mostly in adult, but also in pediatric oncology centers [24,25]. This trend is evident not only in high-income countries but also in those classified as low- and middle-income, highlighting a universal desire to explore and integrate a variety of therapeutic approaches applicable in cancer care [19,26]. Prominent U.S. oncology centers offer a range of complementary treatments including nutritional evaluation and advice, acupuncture, mind-body therapies, meditation, mindfulness, massage, reflexology, yoga, tai chi, music therapy, art therapy, aromatherapy, and physical training programs [27,28].

Currently, the Spanish public health system does not cover complementary integrative therapies, leaving patients to bear the cost of these interventions. To address this gap in our region, the Pediatric Integrative Oncology Unit (UOPI, for its abbreviation in Spanish: Unidad de Oncología Pediátrica Integrativa) was established in September 2019 within the oncology area of the Hospital Sant Joan de Déu, currently the SJD Pediatric Cancer Center Barcelona. This unit, the first of its kind in Spain, was made possible through philanthropic support, enabling the incorporation of complementary treatments into all oncologic patients' therapeutic plans at no additional cost. The UOPI treats pediatric patients as well as adolescents and young adults, who may present with developmental cancers. Examples of these cancers include neuroblastoma, medulloblastoma, osteosarcoma, and germ cell tumors.

By offering complementary therapies integrated into clinical care, the UOPI seeks to enhance the overall treatment experience and address the multidimensional needs of pediatric cancer patients. This innovative approach reflects broader global trends and has the potential to serve as a model for expanding access to integrative oncology within national healthcare systems.

The aim of this study is to describe the acceptance rate and clinical activity of complementary treatments offered to patients, including acupuncture, aromatherapy, and reflexology, corresponding to the first two years of implementation of the UOPI. Nutritional counseling is part of the UOPI, and although it was provided to all patients, it was not evaluated in this study, as it is considered standard of the care rather than a complementary intervention.

2. Materials and Methods

This study was approved by the Medical Research Ethics Committee of Hospital Sant Joan de Déu (approval code: PIC-13-22). The study report was prepared in accordance with the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines [29] Table S1.

The study was conducted at Sant Joan de Déu Children's Hospital in Barcelona, in full compliance with local legislation and institutional regulations. Patient health data were retrospectively collected from clinical records. Since many patients no longer had active follow-ups at the hospital, obtaining specific informed consent beyond the general consent provided by oncology patients during their treatment was considered unfeasible. Consequently, a request for consent exemption was submitted, along with a data management commitment document signed by the principal investigator. This was accomplished in accordance with Organic Law 3/2018 of 5 December, on the Protection of Personal Data and the Guarantee of Digital Rights, ensuring that patient privacy and data security were maintained throughout the study.

2.1. Study Design and Setting

This retrospective, descriptive, observational study was conducted at the UOPI within the Hospital Sant Joan de Déu in Barcelona/Pediatric Cancer Center Barcelona, Spain, over a two-year period. The study took place in a single-center setting, focusing on the integrative oncology services provided by the UOPI.

The UOPI provides individualized care to all patients, beginning with personalized nutritional counseling as part of the broader therapeutic plan. Based on their clinical condition, treatment-associated toxicities, or specific therapeutic needs, patients are referred to the UOPI by their oncologist for evaluation and treatment. This may involve a variety of complementary therapies, including acupuncture (body acupuncture, auriculotherapy, moxibustion), Transcutaneous Electrical Nerve Stimulation (TENS), Transcutaneous Auricular Vagus Nerve Stimulation (taVNS), electroacupuncture, stiperpuncture, cross tape application, aromatherapy, and reflexology. These interventions are delivered during hospitalization to help alleviate symptoms and improve the overall well-being of the patients, ensuring an integrative, supportive approach throughout their cancer treatment journey.

Consultations regarding complementary therapies can be initiated at any stage of the patient's illness, whether by healthcare providers, patients themselves, or their families. The UOPI offers a safe and relaxed environment for these discussions, with close and individualized follow-up to monitor and adjust care based on the patient's evolving needs. The multidisciplinary team at the UOPI includes experienced healthcare professionals with over a decade of expertise in complementary treatments for both pediatric and adult patients. This team consists of a pediatrician with additional training in acupuncture, nutrition, and aromatherapy, one nutritionist, and four reflexologists, ensuring a broad spectrum of complementary care tailored to the patients' needs. The team provides services across multiple hospital settings, including inpatient wards, outpatient clinics, daycare hospitals, the hematopoietic progenitor transplant unit, and intensive care units. In addition, cancer survivors have access to these treatments to manage late-stage toxicities arising from their previous cancer treatments.

2.2. Participants

The study included all oncologic patients evaluated by the Pediatric Integrative Oncology Unit (UOPI) between its establishment on 1 September 2019 and 30 September 2021; no additional exclusion or inclusion criteria were settled. The UOPI serves both pediatric patients and young adults who are transitioning from pediatric oncology care. As a result, the study sample comprised both pediatric and adult patients.

2.3. Data Collection

Data were collected retrospectively by three researchers who reviewed the patients' electronic medical records. Patient confidentiality was strictly maintained throughout the data collection and analysis process in accordance with established ethical standards and institutional guidelines.

First, the researchers accessed patients' electronic medical records in a pseudonymous manner, meaning that all identifiable information, such as names, medical record numbers, and other personal identifiers, was replaced with unique study codes. This ensured that the data could not be linked back to individual patients. Furthermore, the data collection form used for recording information was designed to exclude any identifying details, focusing solely on anonymized sociodemographic, clinical, and treatment-related variables. All data were stored in secure, password-protected files, accessible only to the research team.

A standardized data collection form was created using Microsoft Excel to ensure consistency. The extracted data included sociodemographic information (age at the time of appointment, gender, race, ethnicity, country of origin, and cancer type), hospital admission area (inpatients, outpatients, daycare hospital, transplant unit, special techniques areas, and intensive care unit), reasons for consultation, and details of the complementary treatment (type, number of sessions, and instances of rejection). A fourth researcher (S.P.J.) checked the accuracy of the data and reviewed the records for any mistakes, ensuring the reliability of the collected information.

No strategies for handling missing data were necessary, as there were no instances of missing data in the dataset. The completeness of the records ensured that all relevant information was captured and analyzed without the need for imputation or other data handling techniques.

2.4. Statistical Analysis

The demographic and clinical characteristics of the study population were summarized using SPSS software (version 29). Qualitative variables were described using absolute frequencies and percentages, while quantitative variables were summarized using the median, minimum, and maximum values.

3. Results

During the two-year study period, 433 pediatric oncology patients were visited in the UOPI.

3.1. Sociodemographic Data of Patients

The sociodemographic data of patients is summarized in Table 1. The median age at the first visit to the UOPI was 9 years (range: 0 to 34 years). Of the patients who attended, 266 were boys (61.4%) and 167 were girls (38.6%). Most patients (90.1%) were in active cancer treatment at the time of their UOPI visits, with patients receiving care at various stages of their disease across different care areas. Additionally, 7.6% of the patients were out of active therapy, while 2.3% were at the end-of-life care.

Table 1. Demographic characteristics.

Characteristics	Number of Patients (n = 433)	%
Age in first visit (years)		
0–4	128	29.6
5–9	112	265.9
10–14	104	24.0
15–19	81	18.7
20–24	7	1.6
25–29	0	0
30–34	1	0.2
Median (Range)	9 (0 to 33)	
Sex		
Male	266	61.4
Female	167	38.6
Race		
Caucasian	414	95.6
Asian	13	3.0
Black	6	1.4
Origin		
Spain	375	86.6
Europe	25	5.8

Table 1. *Cont.*

Characteristics	Number of Patients (n = 433)	%
Asia	23	5.3
Latin America	7	1.6
Africa	3	0.7
Active treatment		
Yes	390	90.0
No	43	10.0
Out of active therapy	33	7.6
End of life situation		
Yes	10	2.3
No	423	97.7

3.2. Hospital Areas and Reasons for Consultation

Participants were primarily visited in the ward (362), followed by the daycare hospital (107), a specialized unit within the hospital where patients receive care for procedures or treatments that do not require overnight hospitalization, outpatient clinic (87), stem cell transplant unit (7), procedures area (6), and intensive care unit (5) (Table 2). Some patients were visited in multiple areas of the center depending on their clinical status and needs.

Table 2. Area of treatment.

Hospital Area	Number of Patients *
Ward	362
Outpatient clinic	87
Day care hospital	107
Stem Cell Transplant unit	7
Procedures room	6
Intensive care unit	5

* Several patients were seen in multiple hospital areas.

Detailed information regarding the reasons for consultation was only available for acupuncture and aromatherapy. For all other techniques, the consultation reason was not systematically recorded. With respect to acupuncture and aromatherapy, the most common reasons for consultation were chemotherapy-induced nausea and vomiting, gastrointestinal motility disorders, pain, and stress management. Some patients presented with more than one reason for consultation. The remaining diagnoses, along with the complementary treatments received, are detailed in Table 3.

Table 3. Consultation reasons.

Condition *	Acupuncture	Aromatherapy
Gastrointestinal motility		
Chemotherapy-induced nausea/vomiting	113	
Constipation	59	5
Diarrhea	36	
Pain		
Abdominal pain	59	
Pain Immunotherapy related	4	
Headache	29	
Headache lumbar punction related	6	
Neuropathic pain	28	9

Table 3. *Cont.*

Condition *	Acupuncture	Aromatherapy
Arthralgia	18	
Phantom limb	4	24
Scar pain	12	
Musculoskeletal	49	
Stress/Anxiety	61	43
Respiratory symptoms		
Mucus	5	7
Cough	1	
Night terrors	13	
Anal fissure	6	
Allergic reaction	14	
Insomnia	29	
Mucositis		4
Mutism	4	
Asthenia	18	
Loss of appetite	19	
Dermatological issues		26
Neurogenic bladder	1	
Hot flushes	3	
Tinnitus	4	
Bruxism	3	

* Consultation reasons were not systematically reported for reflexology.

3.3. Provided Treatments

A detailed summary of the interventions provided by the UOPI during the study period is shown in Table 4.

Table 4. Provided treatments and acceptance.

Treatment	Patients Offered n (%)	Patients that Accepted n (%)	Rejection of the Treatment n (%)	Number of Treatments Provided n
Acupuncture	227 (52.42)	215 (94.7)	12 (5.3)	1352
Aromatherapy	196 (45.2)	196 (100)	0	196 *
Reflexology	129 (29.7)	124 (96.1)	5 (3.9)	414

* Aromatherapy was a single session intervention.

3.3.1. Acupuncture

Acupuncture was recommended to 227 cancer patients, with 215 (94.7%) accepting the treatment. The rejection rate was 5.3%, with nine parents (4.0%) and three patients (1.3%) declining the treatment. A total of 1,352 acupuncture sessions were performed over the two-year study period, with a median of 4.5 sessions per patient (range: 1 to 43 sessions). In addition to filiform needle acupuncture, non-insertional techniques were also used. These included cross tape, an adhesive tape applied to acupuncture points or areas of muscle tension; stiperpuncture, which involves placing small silicon-based tablets on acupuncture points; and moxibustion. Additionally, Transcutaneous Electrical Nerve Stimulation of acupuncture points and Transcutaneous Auricular Vagus Nerve Stimulation (taVNS) at ear acupuncture points, which were implemented in the last three months of the study period (Table 5).

3.3.2. Aromatherapy

Aromatherapy treatments were recommended to 114 patients, all of whom (100%) accepted and participated in a single information session. This session provided specific instructions on using aromatherapy to alleviate cancer or cancer treatment-related toxicities such as nausea, pain, and anxiety.

Table 5. Description of the acupuncture treatment components.

Acupuncture Technique	Number of Participants that Received Acupuncture, n (%)
Filiform needle	215 (100)
Non-insertional techniques	62 (28.8)
Cross tape	10 (4.6)
Stipper	10 (4.6)
Moxibustion	21 (9.7)
Mixed	21 (9.7)
Electroacupuncture	28 (29.7)
TaVNS	19 (8.8)
TENS	3 (1.4)

TaVNS: Transcutaneous Auricular Vagus Nerve Stimulation; TENS: Transcutaneous Electrical Nerve Stimulation.

3.3.3. Reflexology

Reflexology was recommended to 134 participants, with 129 (96.1%) accepting the treatment. Despite restrictions imposed by SARS-CoV-2, the reflexology team conducted a total of 414 sessions over the two-year period.

4. Discussion

Integrative medicine combines conventional treatments with complementary therapies to holistically address patients' physical and emotional needs. In pediatric oncology, this approach supports symptom management and enhances quality of life, which many families increasingly seek [30]. The Integrative Pediatric Oncology Unit at the Pediatric Cancer Center Barcelona was designed with this goal, offering therapies like acupuncture, aromatherapy, and reflexology as complements to standard care.

The findings of our study demonstrate a high level of acceptance and feasibility of implementing an Integrative Pediatric Oncology Unit within a comprehensive care model in a pediatric cancer center, with a global rejection rate of only 4.77%. Regarding reasons for consultation, acupuncture and aromatherapy were predominantly used for managing chemotherapy-induced nausea and vomiting, gastrointestinal motility disorders, pain (including neuropathic and immunotherapy-related pain), and stress or anxiety. The use of these therapies demonstrated adaptability to a broad spectrum of symptoms, reflecting their versatility in addressing both physical and psychological dimensions of care. Acupuncture was the most widely offered therapy (52.42% of patients) and had a high acceptance rate (94.7%), followed by aromatherapy, which had a perfect acceptance rate of 100%. This suggests that these interventions were particularly well-received for symptom relief and stress management.

To our knowledge, this is one of the first studies assessing the acceptability of complementary therapies within a pediatric oncology setting. Such low rejection rates highlight a growing willingness among families to explore integrative approaches, particularly when these therapies are seamlessly embedded into the care structure by trained professionals, reducing concerns about safety, accessibility, or additional costs.

A previous study conducted by Lim et al. in Singapore in 2006 assessed the prevalence of complementary and alternative medicine (CAM) use among pediatric cancer patients, reporting that 67.1% of patients had used at least one type of therapy [31]. However, a notable distinction of our study is that the complementary therapies were offered and administered within the same hospital by experienced healthcare professionals at no additional cost to the patients. This integrated approach is likely a key factor contributing to our study's higher acceptance rate. As emphasized in previous studies, healthcare service integration and professional competence are crucial determinants of patient satisfaction, underscoring the value of embedding these services within hospital care models to enhance trust and accessibility [32].

Similarly, a 2017 study conducted at Columbia University Medical Center in the United States reported a 54% acceptance rate of acupuncture among 90 acupuncture-naïve children receiving cancer treatment [33]. In contrast, our study observed a much higher acceptance rate of 94.7%. This discrepancy could be attributed to the use of various acupuncture strategies in our setting, such as painless techniques with minimal puncture using semi-permanent needles, as well as non-invasive methods like cross tape, stiperpuncture, moxibustion, and transcutaneous electrical nerve stimulation (TENS). Additionally, the acupuncture sessions in our study were provided by a senior pediatrician acupuncturist, which may have contributed to a higher level of trust and confidence among patients and their families, leading to greater acceptance of the treatment. Evidence suggests that provider expertise and interpersonal communication significantly enhance patient satisfaction, particularly in sensitive populations such as pediatric patients [32]. These findings highlight the importance of tailoring complementary therapies to the pediatric population, ensuring both physical comfort and emotional reassurance through approaches that minimize invasiveness and build trust.

Lim et al.'s study also found that 55.1% of parents had not discussed their CAM usage with their child's physician, which poses potential risks due to a lack of coordinated care [31]. The UOPI model directly addresses this issue by providing CAM services through highly trained healthcare providers who work closely with oncologists and are part of the multidisciplinary team. This collaboration enhances patient safety and satisfaction, as it ensures that complementary therapies are integrated into the patient's overall treatment plan. The implementation of similar models of integrative care in the United States has demonstrated positive outcomes, which served as an inspiration for our own unit [7,34]. By fostering open communication and collaboration among healthcare providers, integrative units like the UOPI can mitigate risks associated with unregulated complementary therapy use, such as potential interactions with conventional treatments, delayed essential medical interventions, and exposure to unsafe practices [35], while promoting a more cohesive approach to care.

Despite the positive results, this study has several limitations. First, the COVID-19 pandemic imposed significant restrictions on our ability to conduct in-person visits. Reflexology sessions were suspended, and acupuncture interventions were limited to those deemed strictly necessary to minimize the risk of patient exposure to the virus. These changes likely impacted the patient care data we collected and may not accurately reflect service utilization in a post-pandemic era. Second, the retrospective nature of our study restricted the scope of factors that could be analyzed. Variables such as patients' educational background, occupational status, type of employment, and symptom severity could not be assessed. Moreover, patient-reported outcomes such as satisfaction or perceived benefits of the therapies were not captured. Capturing these outcomes in future prospective studies will be essential to understand the full scope of benefits and identify areas for further improvement in service delivery.

The inclusion of young adults transitioning from pediatric oncology care also deserves attention. These patients, up to 34 years of age, may present with developmental cancers typically treated in pediatric settings. The inclusion of young adults may have influenced the acceptance rates, as the consent process differs for minors (who require parental consent) compared to adults (who provide independent consent). However, given the distribution of ages, the majority of patients were pediatric, with most participants falling under 18 years of age. Therefore, we do not consider this a significant concern for the interpretation of the results.

Moreover, while the acceptance rates of complementary therapies were high, the short and long-term benefits and possible adverse effects of these treatments remain areas that require further investigation in the pediatric oncologic population. Future research should prioritize longitudinal studies to evaluate the sustained impact of complementary therapies on clinical outcomes, quality of life, and psychosocial health, as well as potential interactions with conventional cancer treatments. Additionally, incorporating feedback from patients and families can provide invaluable insights into optimizing integrative oncology models for broader implementation.

5. Conclusions

The high acceptance rates of the UOPI's complementary therapies, with over 94% for acupuncture and reflexology and 100% for aromatherapy, suggest that integrative oncology can be successfully incorporated into pediatric cancer care. These therapies were primarily offered in the ward, daycare hospital, and outpatient clinic, reflecting their integration into diverse care settings and clinical stages.

In addition to the high acceptance rates, this study highlights important findings regarding the reasons for consultation and types of interventions offered. Acupuncture and aromatherapy were most commonly used to address chemotherapy-induced nausea and vomiting, gastrointestinal motility disorders, pain management (including neuropathic, immunotherapy-related, and musculoskeletal pain), and stress/anxiety. Aromatherapy was also applied for conditions such as insomnia, dermatological issues, and respiratory symptoms. These findings demonstrate the broad applicability of these therapies for symptom management in pediatric oncology patients.

Future research should focus on the efficacy, safety, and long-term outcomes of these therapies, as well as identifying best practices for their implementation in various healthcare settings. Such studies will be crucial for establishing evidence-based guidelines and ensuring that complementary therapies are used effectively and safely in pediatric oncology. Ultimately, this approach may contribute to a more holistic, patient-centered model of care that better meets the needs of pediatric cancer patients and their families.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/cancers17020222/s1, Table S1: STROBE Statement.

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Institutional Review Board Statement: This study was approved by the medical research ethics committee of Hospital Sant Joan de Déu (PIC-13-22) on 19 March 2022.

Informed Consent Statement: In this observational study, which posed minimal or no risk to the subjects, the ethics committee of Hospital Sant Joan de Déu approved the waiver of informed consent.

This decision was based on several factors: (1) the retrospective nature of the study, which spans a long period of time, making it practically impossible to collect informed consent from all subjects; (2) the requirement for individual consent would have made the study impractical; (3) the study could not be conducted with anonymized data; and (4) the confidentiality of personal data was fully ensured throughout the research process.

Data Availability Statement: The original data presented in the study are openly available at https://figshare.com/articles/dataset/Dataset/27118425?file=49445457 (access on 17 december 2024).

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cancers

Article

Family-Level Impact of Germline Genetic Testing in Childhood Cancer: A Multi Family Member Interview Analysis

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Simple Summary: Genetic testing is increasingly used in children with cancer to detect inherited genetic changes that may increase their risk for cancer. Many families are interested in this testing, but little is known about how it affects the entire family. This study explores how parents experience the impact of genetic testing for cancer risk on their family as a whole. We interviewed both parents in six families with a child diagnosed with cancer to understand their perspectives. The testing was performed by analyzing genes linked to cancer risk. Parents found genetic testing valuable and relatively straightforward but noted that it was hard to separate its effects from the emotional stress of their child's cancer diagnosis and treatment. Despite this, they described how the testing influenced family life in significant ways. Key themes included how families talk about genetic testing, the importance of spending time together, differences in coping between parents, feelings of guilt and forgiveness, and worries about the future health of the family. This research highlights that genetic testing impacts families deeply and that healthcare providers should address these family-level challenges to better support families during an already difficult time.

Abstract: Objectives: Germline genetic testing is increasingly being integrated into pediatric oncology and a large number of families are interested. Current research on the psychological impact of germline genetic testing is limited by a main focus on individual outcomes in parents or children and little is known about its impact at the family level. Our study addresses that limitation by exploring parents' lived experiences of how their family—as a whole—is affected by germline genetic testing for cancer predisposition. **Methods:** In six families who opted for germline genetic testing in the context of cancer predisposition, both parents of six ill children (five boys) with an average age of 9.67 years (SD = 3.77 years) were interviewed individually (N = 12). Germline genetic testing was performed by exome sequencing followed by analysis of a panel of childhood cancer predisposition genes in pediatric cancer patients and their parents. Their experiences were elicited through semi-structured interviews and the data were analyzed using Multi Family Member Interview Analysis. This qualitative study was conducted at Ghent University Hospital in Belgium. **Results:** The findings demonstrated that while germline genetic testing was generally viewed as a valuable and straightforward step in the child's oncology trajectory, parents found it difficult to distinguish its impact from the overwhelming stressors of their

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child's cancer diagnosis and treatment. However, parents recognized that the testing also significantly affected various family-level processes. Five main themes were identified: talking about germline genetic testing, being together matters (more), differences in coping with germline genetic testing between partners, feelings of guilt and mutual forgiveness, and concerns about the future health of the family. **Conclusions:** Given the expanded use of germline genetic testing in pediatric oncology, it is critical for clinicians to address the family-level impacts of germline genetic testing. Although families are affected by these issues, they often do not raise them due to the overwhelming challenges posed by the cancer diagnosis and treatment. Proactively addressing these themes could improve the support provided to families undergoing germline genetic testing for cancer predisposition.

Keywords: cancer; cancer predisposition; children; germline genetic testing; oncology; parents; pediatric oncology; psycho-oncology; family impact; multi-family member interview

1. Introduction

Recent studies in Western countries indicate that a growing proportion of children with cancer harbor a cancer predisposition syndrome (CPS), estimated at 10–15% or higher depending on the sample studied [1–5]. The possibility of diagnosing a CPS in pediatric cancer is highly relevant for treatment adaptation, future risk prediction, surveillance, consideration of predictive testing for at-risk relatives, and family planning [6]. Consequently, germline genetic testing is progressively being integrated into standard pediatric oncology care in different parts of the world (e.g., Belgium, The Netherlands, Canada, Australia, USA, etc.) [7,8]. Despite the medical benefits, offering germline genetic testing also presents families with notable challenges. In addition to ethical considerations (such as balancing a child's best interest with their right not to know) [9–13], the psychological impact on the patient and their family can be significant.

First of all, facing childhood cancer is already a life-threatening stressor and family stress models assume that a stressor affecting one family member also affects the other family members and the entire family system [14]. Empirical evidence supports this theoretical perspective. Next to pain, fatigue, and reduced immunity, the ill child often endures anxiety and uncertainty [15]. Siblings frequently report a diminished quality of life and adverse emotional reactions [16] and, in comparison to parents of healthy children, parents of children with cancer often report significantly higher levels of distress, posttraumatic stress symptoms, parental conflicts, emotional difficulties, and physical complaints [17]. Additionally, families—as a whole—often experience altered roles and responsibilities and increased conflict, but also increased cohesion, communication, and family support [18].

Second, the introduction of germline genetic testing into pediatric oncology care may represent an additional stressor affecting these families, namely the possibility of identifying an underlying predisposition to cancer. On the individual level, recent reviews have shown that ill children and their parents experience a wide range of emotions as a consequence of germline genetic testing, including depression, anxiety, distress, uncertainty, and loneliness [19,20]. Although these negative emotions can resolve in the long-term and can co-exist with feelings of empowerment and positive emotions like relief and hopefulness, several individual risk factors (e.g., female gender, having pre-existing mental health issues, and a lower education level) and contextual risk factors (e.g., families receiving different genetic test results across children or children witnessing distress or illness in a parent) have been identified that can make the impact of germline genetic testing more emotionally taxing [19,21–25]. This psychological impact should be carefully considered in oncology care and genetic counseling [26]. In addition, germline genetic testing in childhood cancer likely also generates a broader family-level impact. Especially since a germline predisposition may affect families by influencing their health outcomes (including susceptibility to cancer). Indeed, the results of germline genetic testing for a CPS can have direct and significant implications not only for the affected child but also for parents, siblings, and extended relatives, who may be found to share the same genetic predisposition [7]. The family-level impact of germline genetic testing in childhood cancer, however, remains understudied, so a detailed and systematic examination of shared experiences in families navigating the complexities of childhood cancer and associated germline genetic testing is needed to organize comprehensive support for them. Therefore, the current study aimed to achieve an in-depth understanding of how germline genetic testing in childhood cancer impacts families.

2. Materials and Methods

Focusing on the family as a whole, the parents as a couple, and the parent–child relationship as units of analysis, Multi Family Member Interview Analysis (MFMIA) [27] was used as a guiding methodological framework in this qualitative study. One-to-one in-depth semi-structured interviews were conducted with both parents separately. This allowed each parent to provide their own perspective [28] without having to factor in their partner's feelings [29]. Children were not included in the interviews due to their health status and/or age.

2.1. Participants

Interviews were conducted with both parents in six families who opted for and received the results of extensive germline sequencing for childhood cancer predisposition in a diagnostic setting. The children received three possible outcomes: diagnosis of a cancer predisposition syndrome (CPS) linked to the malignancy they developed (N = 2), diagnosis of a secondary finding (a secondary finding is a (likely) pathogenic variant that is not causative of the patient's presenting condition but that might have implications for the patient's future or for the wider family. In this case the reported secondary findings are all cancer-related actionable, late-onset (likely) pathogenic variants.), i.e., a genetic variant that could not be linked to the malignancy they developed (N = 2), or no genetic diagnosis, i.e., no (likely) pathogenic genetic variant was identified (N = 2). All families were Caucasian, consisting of four Belgian and two Dutch families. The parents were aged between 30 and 50 years old and had attained a middle or higher level of education (educational level was identified as lower (i.e., did not complete secondary education), middle (i.e., completed secondary education but did not pursue higher education), and higher (i.e., completed education beyond secondary level)). More detailed characteristics of the families can be found in Table 1. Approval from the medical ethical committee of Ghent University (Hospital) had been obtained (BC-08213-AM04), and informed consent forms were signed by all participating parents.

2.2. Data Collection

This study is part of the larger ongoing Belgian DHECIPR project (Diagnosing HEreditary predisposition syndromes for Childhood cancer: Implementation in clinical Practice) where extensive germline genetic testing was offered to all patients (0–18 years) diagnosed at the Department of Pediatric Haemato-Oncology of Ghent University Hospital (Belgium) between 2021 and 2025. Depending on their maturity, children were involved in the genetic counseling and, if 12 years or older, they also signed a simplified informed consent form to initiate germline genetic testing. In the informed consent process, parents (and children if 12 years or older) agreed to receive information about all (likely) pathogenic variants related to the disease and actionable secondary findings. This was thoroughly discussed during genetic counseling prior to the testing. Psychological support was available for families interested in germline genetic testing both during consultations and between sessions, should families need it. The germline genetic testing was performed by exome sequencing followed by data analysis focused on a panel of 197 genes associated with a childhood cancer predisposition. The families who opted for germline genetic testing were invited for discussion of the results in a multidisciplinary consultation (with a clinical geneticist with large expertise in oncology, a clinical psychologist, and a pediatric hemato-oncology nurse specialist) at the center for medical genetics. The psychosocial component of the DHECIPR project, consisting of a questionnaire study for both parents and the child (≥12 years) and an interview study (only parents), was offered in parallel with the medical component of the study (i.e., collecting clinical and genetic data to prospectively validate the MIPOGG referral tool [30] in real-world clinical practice). Interviews took place after consent and disclosure of the genetic results. Exclusion criteria were (a) insufficient proficiency in Dutch and (b) prior confirmation of a genetic cancer predisposition in the patient.

Table 1. Characteristics of participating families.

Families	Both Biological Parents Participated	Gender of Ill Child	Age of Ill Child	Time Since First Cancer Diagnosis	Number of (Half-) Siblings (+Carrier Status)	Sequencing Result of Ill Child
Family 1	Yes	Male	12 years	8 months	X (not known)	CPS found
Family 2	Yes	Male	11 years	3 years (This patient developed several metachronous primary tumors, with the most recent 4 months prior to the interview)	1 (carrier)	CPS found
Family 3	Yes	Male	5 years	6 months	1 (carrier)	Secondary finding
Family 4	Mother and stepfather (In this particular family there was no longer any contact with the biological father, and the stepfather had been fulfilling the father role for many years.)	Female	14 years	5 months	2 (no carriers)	Secondary finding
Family 5	Yes	Male	12 years	9 months	2 (N/A)	No CPS found
Family 6	Yes	Male	4 years	1 year	1 (N/A)	No CPS found

Of all nine families who received a diagnosis of a CPS or secondary finding (N = 6 + 3), the parents were subsequently invited by the psychologist to complete an interview about the impact of the germline genetic testing on their family. In five families, one or both parent(s) preferred not to participate in the interview study, citing lack of time and/or interest as primary reasons. The remaining four families (N = 2 + 2), where both parents agreed to participate in the interview study, were included. Of the families where no underlying genetic variant was discovered, two families were randomly selected and contacted by the psychologist; both families agreed to participate. Interviews were conducted with each of the participating parents individually and were held at the patient's home, at the University Hospital, or via video conference. To avoid influencing each other, both parents were interviewed consecutively without having contact, except for one couple. All interviews were conducted by the same interviewer (S.V.H.: a female clinical psychologist with experience in oncology, family therapist in training, and PhD student with experience in qualitative research), were audio recorded, and lasted between 19 and 68 min. Verbatim transcripts of these interviews served as the raw data for this study. The interviews were based on an interview guideline (see Supplementary Materials; developed by S.V.H.; S.H., a female clinical psychologist in the Center for Medical Genetics, family therapist, and researcher with experience in qualitative research; M.V.S, a female clinical psychologist and researcher with experience in qualitative research and pediatric oncology; and L.V., a female clinical psychologist, family therapist, researcher, and professor in Couple and Family Psychology, and based on literature review, other interview guidelines within pediatric oncology [31], and clinical experience) and consisted of open-ended questions about (a) the experience of the germline genetic testing, (b) the impact on the parent as an individual, (c) the impact on the family relationships, (d) the impact on the parents' romantic relationship, and (e) the impact on their environment. Participants' experiential accounts were facilitated by prompts, in order to encourage the participants to give personal accounts [32]. Pseudonyms were used to protect participants' anonymity.

2.3. Data Analysis

Inspired by Interpretive Phenomenological Analysis (IPA) [32] and Dyadic Interview Analysis [28], MFMIA [27] aims to understand broader family dynamics by obtaining and combining the perspectives of multiple family members [33] and enables a detailed and systematic examination of shared family experiences [27,34]. Data were analyzed from a post-positivist position. The central focus is on the meanings that these experiences have for the families and not to produce objective data. To ensure a robust analysis, all phases were completed by two authors (S.V.H. and S.H.), with consensual agreement [35], i.e., working collaboratively to co-construct the best representation of the data by integrating multiple perspectives and resolving coding differences through discussion until consensus is reached.

In the first phase, individual interviews were analyzed separately following IPA principles. S.V.H. and S.H. both analyzed 100% of the interviews and independently read each transcript multiple times to familiarize themselves with the participant's account, then annotated the transcript with initial observations. These observations (e.g., "this mother wants to spare her child's future children from cancer") were then translated into broader themes (e.g., "preventing cancer in future generations"). Parallels between emerging themes were explored, leading to a clustering of themes for each case. This process was repeated for each participant. In the second phase, themes relevant for each parental dyad were identified by combining the themes of both parents after analyzing their individual transcripts. In the third phase, parallel themes between parental dyads from different families were identified and discussed until consensus was reached among two additional authors (L.V. and M.V.S.). This process of investigator triangulation allowed enhancing the validity of the study [36]. The final list of subordinate (e.g., "preventing the inheritance") and superordinate (e.g., "concerns regarding the other children") themes reflected patterns of convergence across different dyads, based on unique aspects of each parent's and couple's experiences. Finally, all themes were translated into a written account, elaborating on the analysis and illustrating it with direct quotes from the participants.

3. Results

3.1. Childhood Cancer Versus Germline Genetic Testing

When parents were asked about their experiences with the diagnostic germline genetic testing, they predominantly characterized it as a beneficial component of their child's oncological treatment process, expressing appreciation for its implementation.

> "For us, it is actually a kind of given. Look, a tumor isn't there for no reason. There might be a cause somewhere, and then it is good to start early to trace the cause or a possible cause. So, in that sense, we were glad that it was addressed quickly." (Father, Family 1)

However, when explicitly asked to describe the impact of the germline genetic testing on their families, parents encountered difficulties in articulating its specific impact. The challenges associated with the cancer diagnosis and subsequent treatment often overshadowed those related to germline genetic testing or the impact of the germline genetic testing was indistinguishable from that of the cancer diagnosis. This resulted in the emergence of an overarching theme: childhood cancer diagnosis versus germline genetic testing. This was consistently evident across all other identified themes.

> "Yeah, you know, from the moment of the diagnosis, you are so overwhelmed that you are not even thinking about it [the germline genetic testing]. You are so consumed every day with going to radiation treatments, you are very busy with everything (. . .), it really fades into the background and I know it's important, but it's more important to focus on the here and now for a while." (Mother, Family 1)

> "It's very difficult to say that it [the experienced changes in the family] is really linked to that genetic result. Because, as I said, it came just a week after the chemo. So, for us, that genetic result is kind of in the same bag. That's just how it is, but our family has changed definitely." (Mother, Family 4)

Despite the challenge of pinpointing the unique impact of germline genetic testing, parents' narratives clearly revealed impact on their families associated with the germline genetic testing. This impact was grouped into five main themes: (a) talking about germline genetic testing, (b) being together matters (more), (c) differences in coping with germline genetic testing between partners, (d) feelings of guilt and mutual forgiveness, and (e) concerns about other the future health of the family (see Figure 1).

3.2. Theme 1: Talking About Germline Genetic Testing

Subtheme 1a: Transparency with children. When offered germline genetic testing, parents found it important to maintain transparency with their children about the germline genetic testing process. They believed that their ill child, as well as the siblings, should be informed about the possibility of testing and that they should be included in discussions if the children were of an appropriate age. In addition—as some children may exhibit more interest than others—parents adjusted their communication to meet each child's needs, providing more information to those who sought it while not overwhelming others.

> "We also told Jules (Pseudonyms were used to protect participants' anonymity.) and the sisters about it. The oldest, for example, is studying pharmacy, so she is very interested in the whole process and how it affects Jules. (. . .) We explained it

> to them. The middle one wants less to do with it. She is sixteen years old and is also less interested in it." (Mother, Family 5)

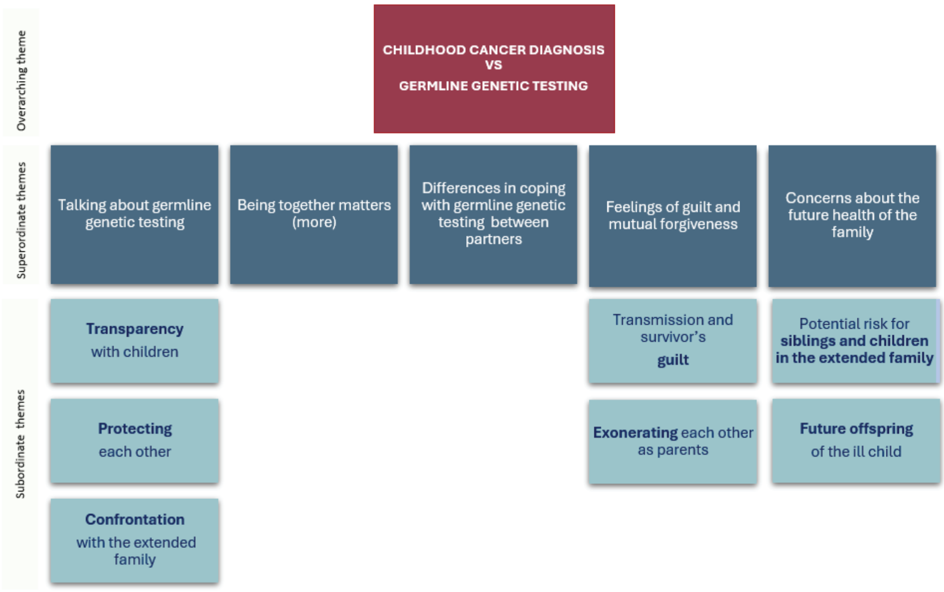

Figure 1. Impact of germline genetic testing: superordinate and subordinate themes.

Families valued open conversations with their children without imposing pressure to participate, especially when discussing germline genetic testing with the affected child. There, parents strived for a balance between informing and supporting their children to reach a shared decision.

> "We talked a lot. It's really difficult because she is still a child and can't really make drastic decisions, but this is about her and her life. And she has to go through all of it. So, I definitely believe that if anyone deserves to have a say or a choice, it is her. And it's not about 'oh, what do you want?' and then 'ok, we'll do it that way'. . . No, we talk about it." (Mother, Family 4)

Even though parents considered shared decision making important and supported their children's autonomy, parents did experience relief if their children's preferences align with their preferred course of action.

> "We also talked about it with the rest of our children because the doctor wanted to have them tested as well. At first, I didn't want it, but I left the decision up to them. Fortunately, they felt the same way and didn't want to know about it right now." (Mother, Family 1)

Subtheme 1b: Protecting Each Other. Discussions regarding germline genetic testing within families were conducted with sensitivity, as both parents and children strived to protect one another. Parents carefully selected both the content and manner of information shared with their children.

> "We discussed more about how we are going to explain it to the children. What are we going to say, what can we not say? What is the best way to phrase it, to talk about the genetics?" (Mother, Family 3)

Parents felt ambivalence and aimed to balance honesty with the desire to protect their children's emotional well-being. They prioritized maintaining a carefree childhood for their children. Indeed, given the significant challenges their child had already faced due to the cancer diagnosis, some parents aimed to shield them from additional stress by selectively withholding information or postponing discussions about the testing.

> "We always decided to let Oscar know as little as possible about everything. Because he had to go through so much with the chemo, we tried to take away as much of the emotional burden as we could for him. So, he knows that he is sick, he knows what he has, but that's it." (Father, Family 6)

Another way of protecting was by maintaining a strong "front". Parents frequently described suppressing their own worries to prevent placing an additional emotional burden on their children.

> "The emotions are intense (. . .) [starts crying], but you still want to be strong for your family, for your child." (Mother, Family 1)

Likewise, parents reported that their children also tried to take on a supportive role by being positive in order to protect their parents from becoming even more distressed.

> "When Emma would sit with me, she would say. . . 'Mom, are you okay?' And she would look at me and say. . . 'Mom, it will be alright. Everything will be alright. Now we know, and it will be all right'." (Mother, Family 4)

Subtheme 1c: Confrontation with the extended family. Parents reported that their extended family members were sometimes concerned about the (results of the) germline genetic testing, regarding the consequences for themselves and their own children. While parents understood the concerns of the extended family, they often were reluctant to be too open and found themselves unsure about how and what to communicate regarding the testing. Therefore, some parents chose to withhold information about the germline genetic testing from their extended family until the results were known to prevent unnecessary worry.

> "I didn't go around telling everyone: 'we're going to get tested because it could be genetic'. That would only make more people worry. We were going to tell them when the time came and we knew the results. (. . .) For example, my brother is someone who immediately thinks it will happen to him too. And if I said it could be genetic, he would start worrying that his daughter and son might have it as well. He wouldn't take it well. So, I prefer to protect him rather than make him worry as, for now, there's nothing to worry about." (Mother, Family 6)

Besides wanting to avoid potential unnecessary worry in the extended family, another reason for not sharing information was to protect their nuclear family. Some parents preferred to focus on their own child and family without the additional emotional strain of dealing with the concerns and reactions of relatives. By withholding information, they shielded themselves and their nuclear family from potential emotional stress.

> "That also opens a whole can of worms regarding the family, instead we can now focus only on this and not worry about the entire family potentially being involved." (Mother, Family 1)

3.3. Theme 2: Being Together Matters (More)

When confronted with a cancer diagnosis and a genetic predisposition, parents reported that their family experienced a shift in values. They placed greater importance on family time and invested more effort in spending moments together. Parents perceived that the shared experience of navigating cancer and germline genetic testing made their families more appreciative of the time they have together. These moments of togetherness had become a source of strength and comfort, reinforcing their bonds and highlighting the importance of prioritizing family over other obligations.

> "I'm not saying we leave everything; we keep going, but now we're more likely to say, 'Does it have to be this week? There's always next week.' We say, 'We can also do something fun with the kids.' (. . .) There was a movie night at school, and this is the first year we've attended. Now we say, 'Yes, we're going to do that, and we're going to go as a family.'" (Mother, Family 3)

3.4. Theme 3: Differences in Coping with Germline Genetic Testing Between Partners

As a couple, some parents dealt with the germline genetic testing in a similar manner, finding alignment in their response to the ongoing germline genetic testing and the subsequent result. However, other couples managed the experience differently. Typically, one partner assumed the role of the "practical level-headed one", approaching the germline genetic testing rationally, searching for a solution, and feeling reassured by receiving adequate information. Conversely, the other partner took on the role of the "emotional thinker," approaching the testing based on feelings, ruminating about potential negative outcomes, and not finding reassurance in information alone. Partners described being very aware of their differences and trying to respect each other's approaches.

> "It did bring a kind of burden with it. Lisa (partner) was a bit more afraid of the result, but in the end, I am just a bit different from Lisa. Lisa would be more likely to say, 'What if,' whereas I would be more likely to say, 'If we know, we can respond to it." (Father, Family 3)

> "For me, it faded into the background and I thought, we'll worry about it when we get the results. You can worry about it now, but that doesn't make any sense. Well, I'm the type of person who moves on quickly and we'll see when it comes. I know that Dave thinks about it and worries about it much more. . . But he's not a talker, so. . ." (Mother, Family 6)

Parents also tended to make assumptions and fill in for each other based on what they observed in their partner's external behaviors. In this way, they assigned roles to each other, where differences in coping seemed to be amplified.

> "I am the one who is more emotional. (. . .) And Bert (partner) is a bit like my dad; he only looks at the positive things. So, if the doctor says, we can do this, we can do that, that is their bit of reassurance. I don't mean to say that he isn't afraid, but he doesn't really think about: oh, what could happen? Or at least, he doesn't talk about it." (Mother, Family 2)

Partners may also be considerate of each other's emotional states and intentionally withhold certain thoughts to avoid causing distress or arguments due to their differences in dealing with the germline genetic testing.

> "That is not something I want to talk about with someone who is emotional. I haven't actually talked about it with anyone except you and the psychologist. I think that I am too dry in my thinking about it. And I don't want to discuss it with anyone, especially not with Valerie (partner). (. . .) I also don't want it to cause any arguments because of what I think." (Stepfather, Family 4)

> "You say. . . I am very rational and very strict in this. On the other hand, it also visibly affects you when speaking about it, doesn't it?" (interviewer)

> "It, it, it definitely affects me. Uh. . . [gets emotional] But in my mind, there is a solution." (Stepfather, Family 4)

3.5. Theme 4: Feelings of Guilt and Mutual Forgiveness

Subtheme 4a: Transmission and survivor’s guilt. Regardless of the test results, the theme of transmission guilt consistently and spontaneously surfaced in all conversations with parents. Parents expressed concerns about the possibility of having transmitted a (likely) pathogenic variant to their offspring, highlighting the emotional burden and feelings of responsibility for their child’s genetic condition.

> “Sometimes you think, I just hope it has nothing to do with the known syndrome [that already runs in the family] (In this family some family members (and not the patient) were known carriers of a predisposition. At the start of the study the carrier status of the child was not known, but the genetic analysis revealed this.) because I would really find that terrible. I mean, otherwise, it comes from me.” (Mother, Family 1)

Moreover, according to some parents, grandparents also shared this burden, feeling responsible for the cancer predisposition syndrome manifesting in their grandchild. This (inter)generational aspect highlights the broader familial impact of the possibility of a genetic predisposition and the accompanying emotional burden.

> “My mother takes this very much to heart; she’s quite affected by it because she, of course, had surgery herself at one point. Fortunately, it always remained just that one tumor, but she is really upset that the youngest grandchild is indirectly affected by what she had. While she knows, rationally, that she can’t feel guilty about it, she still feels it somewhere, yes.” (Mother, Family 2)

> “That was ultimately a big relief for me. I also started to cry because I could reassure my mom that it didn’t come from her side.” (Mother, Family 6)

When germline genetic testing failed to identify a cause for the tumor, many parents initially expressed relief from their feelings of guilt. However, a persistent sense of guilt often remained.

> “It is now clear that something went wrong due to nature and that none of us could do anything about it. We just have to get through it. It was not his fault, or our fault, or my fault that he got it. That is life, but it doesn’t mean I feel any less guilty that he got it.” (Mother, Family 6)

Additionally, some parents who did not carry the (likely) pathogenic variant themselves also experienced a form of guilt, often referred to as survivor’s guilt. This was characterized by a deep sense of injustice and, in some cases, a desire to share the burden with their affected child.

> “I was so angry that Emma had to bear this alone. It’s strange to say, but I felt disappointed that it was her again and not me. They’ve tested me too. I would have preferred to have it myself, along with my two children. Not that I want them to have it, but it was just Emma again. . .” (Mother, Family 4)

Subtheme 4b: Exonerating each other as parents. At the individual level, feelings of transmission and survivor’s guilt were prominently felt. However, at the interpersonal level, mothers and fathers consistently demonstrated mutual forgiveness. This understanding was frequently expressed even before receiving germline genetic testing results.

> “We had already said, ’don’t be angry if it comes from me, okay.’ But then one says, ’well why? You didn’t know there was going to be anything either.’ My parents wouldn’t have known that they passed it on, and it’s been in the genes for generations.” (Mother, Family 3)

> "Well yes, Lisa also had a bit of the mindset of 'if there's anything, then it will come from my side of the family.' But yes, in the end, it's just the bad luck that it came from her side; just as easily, it could have come from my side." (Father, Family 3)

Overall, in nuclear families, parents' primary concern was the health of their child, rather than determining which partner was to blame. They emphasized unity and actively avoided placing blame on one another.

> "You are dealing with your son's illness. I mean, Jules needs to get through it, and where it comes from—whether it's from me or my wife—we're not going to start saying it's your fault or my fault." (Father, Family 5)

In contrast, the dynamics in divorced families may differ significantly.

> "I was so angry. I thought, that man has brought nothing good into her life. Since she was born, he has only brought us misery. I mean, and that's actually not fair, because he probably can't help it. This might be the only thing he can't help. And afterwards, when it all calms down, you know that, but at that moment..." (Mother, Family 4)

3.6. Theme 5: Concerns About the Future Health of the Family

Subtheme 5a: Potential risk for siblings and children in the extended family. Gradually, or sometimes simultaneously with their child's cancer diagnosis, parents began to express concerns about the potential cancer risk for their other children, the children of their family members, and themselves.

> "We actually had that [question] at the time of diagnosis, yes: 'Is it something hereditary? Are we passing on that tumor ourselves? Should we already be looking to check for ourselves or for his brother?'" (Mother, Family 3)

> "Our other son and also within the family. We have more children, more people. So, if there is something genetic, it doesn't stay just within our immediate family. Then, of course, it also affects the extended family, and that can be quite extensive." (Father, Family 6)

Subtheme 5b: Future offspring of the ill child. Parents were not only concerned about the current health of family members but also contemplated the future health of their potential grandchildren. They reflected on whether prior knowledge of genetic risks might have influenced their decisions to have children. Conversely, some parents recognized the challenges but expressed gratitude for modern medical interventions that could help future generations manage these risks.

> "I find it very difficult that he is also a carrier. Then you know that the syndrome doesn't just stop, it continues in their lives, and now he already knows it. Yes, I sometimes say that if I had known this beforehand—especially if this tumor had been related to this syndrome—I would never have had children, but I didn't know. He can now make the choice, you know, to not have children, and I find that quite intense at his age." (Mother, Family 1)

> "If our children want to have children later, then it will have to be with all sorts of modern medical interventions. I find that a shame for them, but on the other hand, it's also something very beautiful that it is possible." (Mother, Family 2)

In addition, parents also seemed considerate of the impact it would have on their child's future partner.

> "It will of course be a bit strange for the children in the long run, because if they have a partner they want to be with, then at some point they will also have to say: well, if we want children, it will never happen in the normal way." (Mother, Family 2)

4. Discussion

This study is, to our knowledge, one of the first to explore how parents experience the family-related impact of germline genetic testing offered as the standard of care in the context of pediatric cancer. Genetic information can affect multiple family members, and adopting a family-wide approach helps us to identify the psychological impact of germline genetic testing on the entire family system.

While germline genetic testing was generally viewed as a valuable and straightforward step in the oncology trajectory, it seems that parents found it difficult to distinguish its impact from the overwhelming experience of their child's cancer diagnosis and the current treatment, which is their primary concern. This is in line with the findings of McGill et al. [37], where the parents of children with cancer described genetic consultations as a secondary concern to the immediate stressors of their child's treatment. This highlights the complex emotional landscape in which families navigate both the immediate challenges of the cancer and the potential long-term implications of germline genetic testing, such as the increased future cancer risk in the child, the increased cancer risk in family members, and the lifelong, intensive surveillance in carriers. An interesting finding from our study is that, despite the overarching stressors related to cancer treatment, parents recognized the role of germline genetic testing in influencing family dynamics such as discussing testing, the importance of togetherness, variations in how partners manage testing, feelings of guilt, and concerns about their other children and family members. These findings provide a nuanced understanding of how families deal with germline genetic testing and its broader implications and differ from previous more descriptive findings by focusing on processes rather than on outcomes [8,9,25,38].

Family communication plays a vital role throughout the germline genetic testing process in cases of childhood cancer. This "*talking about testing*" encompasses not only the disclosure of genetic results but also the pre-test discussions that shape the decision-making process. Our data show that, within nuclear families, parents often emphasize maintaining an open dialogue with their children, encouraging *"transparency with their children"* and promoting shared decision making. However, the inherent emotional complexities of such conversations frequently introduces challenges. The delicate balance between fostering transparency and the instinct to protect one another from psychological distress—referred to as "*protecting each other*"—becomes a central tension. The desire for emotional protection within families aligns with established psychological constructs such as protective buffering [39] and double protection [40]. These concepts describe the tendency of individuals to withhold personal thoughts, feelings, or concerns in an effort to protect a loved one from emotional distress. In the case of double protection, both parents and children engage in this mutual shielding, attempting to spare each other from additional emotional burden. This tension between transparency and protection is particularly pronounced in the context of genetic testing, where the stakes are both emotionally and medically high. Family studies highlight the importance of open communication in coping with illness, noting that transparency can foster collective resilience and meaning-making within the family unit [41]. However, as Afifi et al. [42] emphasize, individuals often face competing desires for information and avoidance when the emotional cost of full disclosure feels too high. This sheds light on the ambivalence parents and children experience in genetic testing discussions, where the imperative for transparency is counterbalanced by the urge to protect.

It seems crucial to address this felt contradiction within families in order to enhance shared coping and meaning making of the challenges of germline genetic testing [43].

Additionally, our findings highlight the involvement of *extended family members* in conversations about germline genetic testing in the context of childhood cancer. Extended family members often express both an interest in the germline genetic testing process and concerns about the health implications for the ill child, as well as the potential risks for themselves and their offspring. Similar to communication within nuclear families, our results suggest that discussions with the extended family require a careful negotiation between transparency and emotional protection. On the one hand, initiating conversations with extended family members before receiving germline genetic test results may provide them with an opportunity to process potential outcomes, engage in informed discussions, and offer emotional support to the nuclear family [44]. This approach aligns with findings from studies on family-centered communication, which suggest that early, transparent discussions about genetic risk can reduce uncertainty and enhance coping mechanisms across familial networks [45]. On the other hand, parents are often cautious about the emotional impact such disclosures may have on extended relatives. Parents in our study felt a responsibility to assess the emotional capacity of extended family members before broaching the subject of germline genetic testing. They aimed to assess the emotional readiness of others before sharing sensitive information. Common reasons for delaying discussions about sequencing with extended family members include the relatives' potential anxiety about their own health risks, as well as the emotional toll of navigating a loved one's illness [44]. Some parents therefore prefer to wait until they receive results before disclosing their decision to undergo germline genetic testing, as a way to avoid imposing unnecessary emotional strain on relatives before any concrete information is available.

Furthermore, the theme of "*being together matters (more)*" reflects a shift in (prioritizing) certain family values, often triggered by the experience of a cancer diagnosis [31], but potentially intensified by the experience of a genetic risk diagnosis. Our data show that this shift towards (more) togetherness can be driven by a heightened awareness of the uncertainty surrounding the future. In such situations, families may re-evaluate their priorities, placing greater importance on family-centered activities that reinforce emotional connections. These shared experiences seem to strengthen family bonds, underscoring the importance of time spent together as a source of resilience [46]. However, this shift may not be universal. Other studies have shown that parents of children with cancer predisposition syndromes and the children themselves sometimes report feelings of alienation from other family members. The rarity of such a diagnosis can create a unique emotional and social divide within the family [24,37]. Thus, while a genetic diagnosis can foster family closeness for some, for others, it may lead to a sense of loneliness.

Another prominent theme we identified in the context of germline genetic testing for childhood cancer was the tension between *emotional responses and rational decision making*. In the decision to initiate germline genetic testing, couples typically demonstrated a preference for knowing rather than not-knowing and prioritized the potential medical benefits over fear-driven reactions, arriving at a joint decision grounded in the perceived utility of the results. This principal attitude toward germline genetic testing seems consistent with the previous literature [25,47,48]. In addition, families generally experienced little burden in opting for germline genetic testing [25,49]. However, alignment in decision making does not necessarily translate into a shared coping strategy throughout the germline genetic testing process. Our findings show that one partner may focus more on practical solutions and the other on the emotional ramifications. This aligns with previous studies showing that—even when confronted with the same stressor—coping strategies (e.g., problem-focused versus emotion-focused) can differ significantly between partners [50]. Emotion-focused coping aims to regulate emotional responses during stressful situations, while problem-focused coping targets addressing and altering the source of stress. Problem-focused coping tends to be more appropriate in situations that are changeable (i.e., controllable), whereas emotion-focused coping is more useful when facing inevitable or uncontrollable events [51]. In the context of germline genetic testing, the effectiveness of these strategies seems complex to determine; as stated by Fantini-Hauwel et al. [52], on the one hand the test results may seem uncontrollable due to inherent factors, while on the other hand families can exert some control by engaging in regular screenings to manage their risk. When facing germline genetic testing in childhood cancer, these divergent coping styles become more pronounced as couples have to navigate the psychological complexities of the process together. Here, our data demonstrated that both partners strived to respect one another's approach; sometimes this divergence led to a more balanced way of coping, where partners learned from each other, and other times this divergence also led to unspoken emotional burdens, as some parents withheld their concerns or thoughts to protect their partners or children from additional stress or conflict. These findings emphasize the need for healthcare professionals to offer tailored support to both parents to address emotional and informational needs more comprehensively.

Our results also revealed the pervasive theme of *guilt*, manifesting in concerns over the transmission of (likely) pathogenic variants and feelings of survivor guilt—where parents spared from the (likely) pathogenic variant experience guilt on behalf of their affected child. This finding aligns with other research on hereditary oncologic conditions, which demonstrates that parents often feel a profound sense of responsibility and guilt, despite the genetic risk being beyond their control [19]. Interestingly, parents in our study often preemptively exonerated each other from *blame*, reflecting a mutual understanding of the uncontrollable nature of genetic inheritance. However, the experience of one family, struggling to navigate these emotions, indicates that not all families are able to provide mutual support, suggesting that certain families may require additional psychological support.

Another significant finding was the persistent *concern about the future health of the family*, reflecting the far-reaching emotional and practical implications of germline genetic testing. Parents in our study expressed apprehension not only about the risks faced by their affected child but also regarding the potential impact on their other children, future grandchildren, and extended family members. Indeed, other studies show that gaining knowledge about whether other family members could be at risk and implications for their and their child's future family planning were seen as advantages of testing [48,49]. These findings highlight the intergenerational ripple effect of germline genetic testing and underscore the need for family-centered genetic counseling to address ongoing and future concerns, taking into account the availability of resources and time. Additionally, our results demonstrate that parents communicate little about their own genetic risk. During the germline genetic testing process, they primarily focus on their role as caregivers, rather than viewing themselves as individuals at risk. As a result, they either do not address their personal concerns or choose not to discuss them. This highlights the complexity of parental perspectives, as their focus on caregiving often takes precedence over acknowledging or discussing their own genetic vulnerabilities.

4.1. Clinical Implications

Given the complex emotional dynamics that families experience during germline genetic testing in the context of childhood cancer, there is a clear need for family-centered counseling that goes beyond discussing the individual risk of the ill child. Healthcare providers (e.g., geneticists, but also psychologists and genetic counsellors) could therefore gather the involved family members and facilitate discussions that address both immediate medical concerns and the psychological impact on the entire family system. This includes (a) helping parents navigate discussions with their children, (b) preparing nuclear families for conversations with extended family members, (c) discussing feelings of guilt by normalizing the uncontrollable nature of genetic inheritance and promoting mutual understanding among families, (d) exploring couples' differences and similarities in coping, so partners and clinicians can discover if they have converging or diverging needs and how these can be met, and (e) acknowledging the future-oriented concerns by discussing the preventative health strategies for future generations. This approach could provide a supportive space for families that can foster familial coping and shared understanding.

4.2. Study Limitations and Suggestions for Future Research

A key limitation of this study is the need for caution when interpreting the impact of germline genetic testing on families in the context of childhood cancer, as our findings indicate that this impact is intertwined with the impact of the cancer diagnosis, suggesting that the results should be understood within the broader context of families dealing with the challenges of a cancer diagnosis. Additionally, the sample size was relatively small, which may limit the diversity of family dynamics observed. A larger sample could provide a more comprehensive picture, could potentially uncover additional themes, and could explore differences in the experienced impact of germline genetic testing based on when it was offered to families. Another limitation is that the sample did not include families with children who have a de novo variant. Including such families could help researchers identify whether the emotional and communicative dynamics differ when the risk is not linked to one parent's genetic material. Furthermore, from parents' perspectives, the germline genetic testing process seems to resonate with various family members, which highlights the potential value of future research involving other family members. First, future studies could focus on how to involve children, siblings, and extended family members in research, as their experience of the impact of germline genetic testing on their family may differ from parental views. Second, future studies could explore how to involve extended family members in clinical practice, as it would add to our knowledge and understanding of family dynamics (e.g., the benefits and challenges of family communication about germline genetic testing prior to disclosure of the test results). Finally, it would be of interest to develop guidelines to support clinicians in addressing the described themes with families.

5. Conclusions

Having a child being confronted with a life-threatening cancer diagnosis often makes it difficult for parents to recognize the specific impact of germline genetic testing on their family. However, as healthcare providers engage in more in-depth discussions, a range of important issues tends to come to light. When confronted with germline genetic testing, families encountered complex emotional and communicative challenges. The decision to undergo germline genetic testing sparked various discussions within families, as parents strived to balance transparency, protection, and consideration for their nuclear and extended family. Germline genetic testing also triggered concerns in families about the health of siblings, future offspring, and extended family and made them place greater value on being together. While it elicited experiencing guilt, parents also showed mutual forgiveness and, despite their differences in coping, tried to respect each other's approaches in dealing with the germline genetic testing. The emotional complexities surrounding germline genetic testing for childhood cancer underscore the need for nuanced approaches to family communication by geneticists, genetic counselors, and other involved clinicians (e.g., pediatric oncologists). A deeper understanding of the protective mechanisms families employ, along with interventions designed to encourage shared meaning-making, will be critical in enhancing the psychological well-being of the family as a whole. Future research should continue to explore how families navigate these emotional dynamics, particularly in the context of emerging genetic technologies and evolving ethical considerations in pediatric oncology.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/cancers17030517/s1, Table S1. The interview guide is a prototype for interviewing parents of a child with a blood disease or tumor where germline genetic testing has been initiated. The order of the questions depends on the input from the parent(s). The questions are asked in a very empathetic and sensitive manner, delving deeper into what the parent shares.

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cancers

MDPI

Review

Therapeutic Parent–Child Communication and Health Outcomes in the Childhood Cancer Context: A Scoping Review

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Simple Summary: Family communication in the cancer context has gained attention as a crucial factor related to the quality of life of children and adolescents. The aim of this review was to examine therapeutic parent–child communication and its impact on health outcomes in children and adolescents with cancer. This review identified an emerging body of evidence that therapeutic parent–child communication contributes to better psychological health outcomes at both the individual and family levels in the childhood cancer context. A comprehensive understanding of the impact of family communication may provide knowledge for developing interventions to improve family communication and health outcomes.

Abstract: Family communication has been thought to be an important area to support children's adjustment to a cancer diagnosis. However, the characteristics of therapeutic parent–child communication that contribute to better patient outcomes and the specific patient health outcomes have been less explored. This current review explored the characteristics of therapeutic parent–child communication and its physical and psychological health outcomes. A total of 5034 articles were initially identified, and only 10 articles remained for inclusion in this review after application of the exclusion criteria. Most studies used a cross-sectional design and measured verbal communication characteristics and its psychological outcomes, but no physical outcomes. The characteristics of therapeutic verbal communication (openness, maternal validation, quality of information shared, etc.) and nonverbal communication (eye contact, close physical distance, and acknowledging behaviors) were identified. The psychological health outcomes included less distress, a lower level of PTSS, less internalizing and externalizing of symptoms, increased levels of social emotional competencies, better peer relationships, and more cooperation during the procedure at the individual level. Increased family cohesion and family adaptation were family-level outcomes. Longitudinal studies are needed to identify what qualities of communication predict better psychological outcomes so that interventions can be developed and tested. In addition, physical outcomes should be evaluated.

Keywords: child; adolescent; cancer; communication; health outcomes

1. Introduction

Recently, due to advances in technology, early diagnosis, and treatment, up to 85% of childhood cancer patients look forward to long-term survival [1]. This improvement in survival makes childhood cancer a chronic illness with long-term sequelae from treatment [2] and adds an important component to cancer care of focusing on quality of, life which includes a sense of well-being and coping for cancer patients and their families [3].

Family support is among the diverse factors found to contribute to better quality of life [4–6]. Research has described the importance of the role of family in dealing with cancer-related distress and adjusting well to the cancer diagnosis by overcoming psychological challenges such as internalizing and externalizing behaviors, as well as anxiety in the childhood cancer context [7]. Without proper support, children and adolescents with cancer can be at risk of maladjustment to a cancer diagnosis [7,8], and parents may struggle to provide optimal care due to unresolved stress and fear related to uncertainty [9].

Among the diverse roles that family plays in supporting children and adolescents along the childhood cancer journey, communication has been found to be a potentially important area [10]. Communication is an indication of the quality of dyadic interactions, including the level of intimacy, trust, or conflict. By communicating, parents can share information, offer support, and socialize their children. In the childhood cancer context, family communication is defined as family members' ability to communicate and express their thoughts and feelings in the midst of a stressful situation [11]. Specifically, the ability to share emotional needs has been reported as essential for the development of healthy coping strategies in children and adolescents [12,13].

The relationship between effective family communication and coping or adjustment is well supported in the context of adults' chronic illnesses, including cancer [14,15]. When patients could share their emotions with their family members, they were less likely to experience disease related distress [16]. Positive family communication between the patient and caregiver mediated the relationship between family hardiness, caregiver positivity [17], and reduced caregiver burden [18]. When family members and patients shared all cancer-related information, all were more likely to experience overall adjustment and higher rates of psychological well-being [19,20]. Family members also benefit from the open communication that involves sharing emotions and emotional support [21–23]. Other benefits from family communication include fewer conflicts between family and physician and improved decision making [24,25]. The association between parent–child communication characterized by the openness of emotions and adjustment is also widely accepted in the childhood cancer context [26]. For example, by communicating with their parents, children with cancer were well informed about their condition and showed psychological stability and better adjustment and resilience [27,28].

Despite the evidence demonstrating the importance of communication, there are several gaps that need to be addressed in order to develop and test potentially effective interventions to improve the communication between parents and children/adolescents with cancer. The most important gap is a lack of knowledge around the characteristics of parent–child communication that are associated with better patient outcomes in terms of both psychological and physical outcomes. In addition, the expected benefits other than coping and adjustment have not been fully explored as much as they have been in the adult cancer context. Thus, the purpose of this paper was to explore the characteristics of parent–child communication that result in better physical or psychological patient outcomes in the childhood cancer context.

2. Materials and Methods

This literature review was conducted in accordance with the methodological guidelines outlined in the Principles of the Preferred Reporting Items for Systematic Reviews and Meta-Analysis Extension for Scoping Reviews (PRISMA-ScR) for data extraction and reporting [29]. A scoping review, which falls under the category of systematic literature reviews, delineates the extent of existing research evidence across diverse methods or disciplines. This methodology fit the purpose of this review in that we wanted to understand the breadth of the evidence and highlight the gaps as well as offer recommendations for potential future investigations [29,30].

2.1. Literature Search and Screening

A health sciences librarian with expertise in systematic review methodologies assisted with the selection of databases and the development of search terms. References were obtained by searching in PubMed, CINAHL PsycINFO, and Scopus using the MeSH terms based on keywords, including 'parent-child', 'parent-adolescent', 'communication', and 'malignancy.' The MeSH terms based on each keyword were generated by a health sciences librarian for each database. The search process was conducted from December 2022 to December 2023.

Two reviewers (HS and NK) independently screened the titles and abstracts of the studies obtained through the databases to determine their eligibility for full-text review. The studies included were selected based on the following criteria: (a) written in English and peer-reviewed, (b) published after January 2014, (c) the target population was children and adolescents aged up to 24 years with any type of cancer, and (d) the inclusion of physical or psychological outcomes. Nonresearch articles, such as literature reviews, letters, editorials, dissertations, and opinion pieces, were excluded. Also, studies focusing on disclosing children's cancer diagnoses only were excluded, as the scope of our topic was broader than disclosing the diagnosis. Finally, any studies focused on the effectiveness of the interventions or the validity of psychological measurements were not included. Following the screening process, the two reviewers discussed conflicts in the eligibility, and a consensus was established.

2.2. Data Extraction

The studies selected beyond the initial screening were divided between the two reviewers. Each reviewer read the full-text articles and had discussions with each other to determine whether the articles satisfied the inclusion and exclusion criteria, as well as the strengths and weaknesses of each study to be included. The two reviewers independently completed an investigator-developed table, which integrated the characteristics of parent–child/adolescent communication and the relationships between the communication and health outcomes. The first author subsequently reviewed the table to verify the accuracy of the data extraction process. Discrepancies regarding the decision on which literature should be included were resolved at regular meetings. In this scoping review, a quality assessment score was not determined; rather, we employed critical appraisal to scrutinize the strength and limitations of each study. This approach enabled us to encompass diverse sources of evidence on our broad topic.

3. Results

A total of 5034 articles were identified through database searches. After removing duplicates, 4153 remained. An additional two studies were identified through manual searching reference lists. Abstract and title screening removed 4127 nonrelevant articles, and full-text review removed an additional 18 studies. Out of the 18 articles excluded, 1 article was excluded because of its irrelevance to our research questions and its low quality as evidenced by several errors in text, inconsistency between research questions and findings, and statistical tests. One additional article was excluded because it mainly focused on measuring the parent–child relationship without including the parent–child communication component. As a result, 10 studies were retained. Figure 1 provides our PRISMA flow diagram [31].

3.1. Overview of Included Studies

The included studies spanned quantitative (90%, $n = 9$) and a mixed-method design ($n = 1$). Among the quantitative studies, the majority used a cross-sectional, correlational study design (70%, $n = 7$), with only three using a longitudinal study design. Methodological elements, sample demographics, and other characteristics are summarized in Table 1.

3.2. The Characteristics of Children and Adolescents with Cancer

The age range of patients was broad, from 3 to 24 years old, with a majority of the participants identifying as White [8,26,32–35]. Nine out of ten studies provided demographic information about child/adolescents' biological sex [8,26,32–38]. The majority of studies included a slightly higher number of male participants [32,33,35–37]. However, no influence of sex differences on parent–child communication was reported.

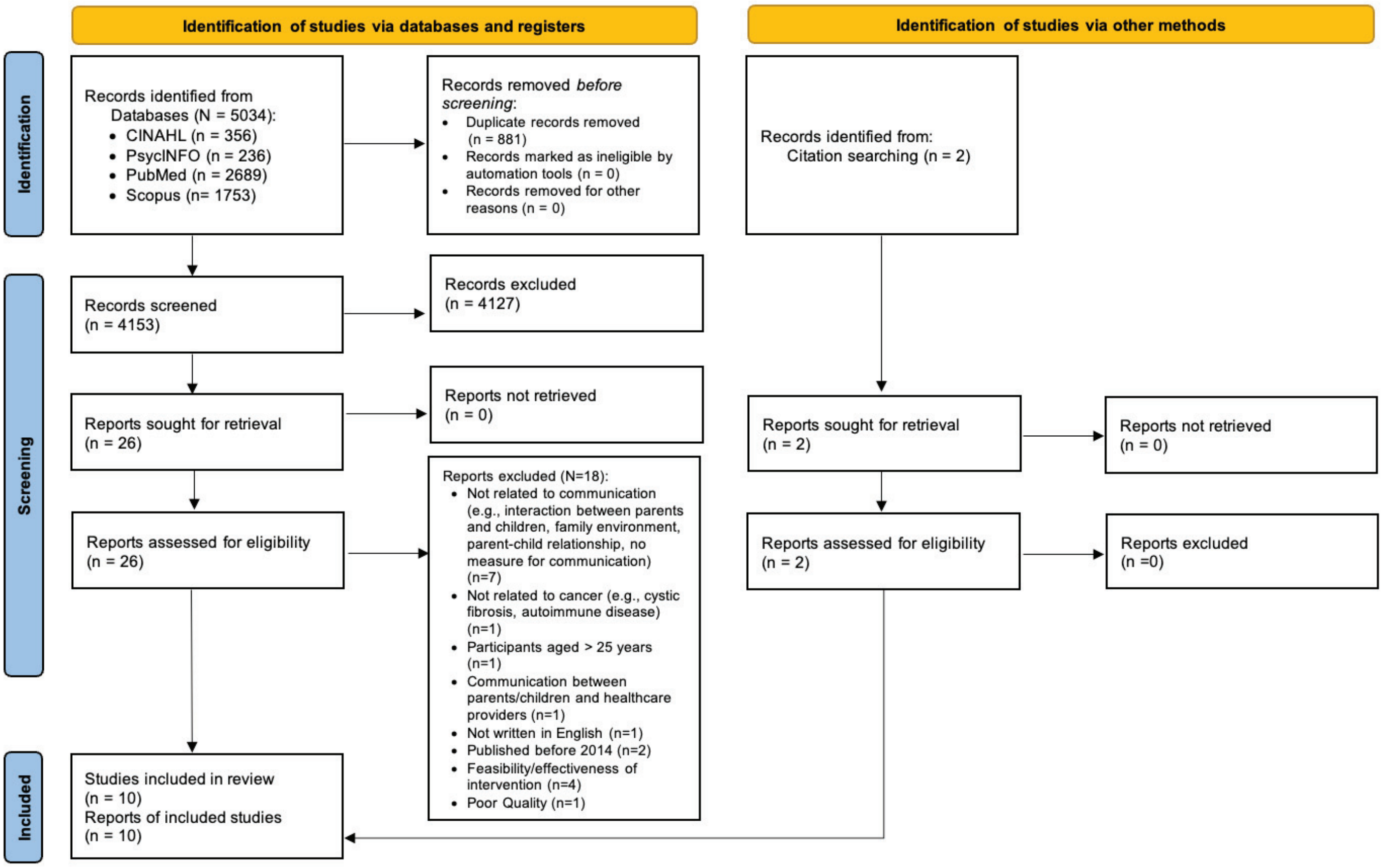

Figure 1. PRISMA flow diagram of study selection [31].

Overall, the participants in the included studies reported diverse types and statuses of cancer diagnoses. Studies mostly included patients who were newly diagnosed with cancer, based on the timeframe ranging from 1 to 18 months from the initial diagnosis [8,33,36,37]. Cancer-related communication was assessed among study participants undergoing cancer treatment in six articles [8,26,33,35–37]. One article provided detailed information regarding the types of cancer and progress (advanced vs. nonadvanced cancer), as well as whether the diagnosis was relapsed or refractory [26]. Four articles identified the types of treatments participants received, such as chemotherapy, surgery, and radiation [8,32,34,36], whereas the remaining six articles did not provide this information [26,33,35,37–39].

It was notable that one article examined illness-related factors, such as years since diagnosis and relapse status, as potential influences on communication. Tillery et al. [34] examined the influence of the time since diagnosis and relapse status on the parent–child relationship, which included parent–child communication practices. An increased time since diagnosis was related to a higher chance of being either struggling (lower than average levels of parent–child relationship) or normative (average levels of parent–child relationship) groups. When children had a relapsed cancer diagnosis, their families were more likely to have a highly involved parent–child relationship, which was characterized by reports of above-average levels of parent–child communication.

Table 1. Summary of studies included in this review.

Study/ Country	Objective	Design	Sample and Age Range (Groups)	Independent Variable (Measurement and Method)	Dependent Variable (Measurement and Method)	Relationship/ Characteristics of PAC
Greeff et al. [38]/ Belgium	Identify resilience factors associated with family adaptation	Cross-sectional, correlational	26 parents and 25 children aged 12–24 (G3 and 4)	Parents' and children's self-report of family communication (affirming/incendiary communication) (FPSC)	Parents' and children's self-report of family adaptation (FACI8)	(1) Positive correlation between affirming communication and family adaptation in the reports from parents (r_s = 0.62) and children (r_s = 0.71) (2) Negative correlation between incendiary communication and family adaptation in the reports from children (r_s = −0.59) (3) Family problem solving communication found to be the predictor of family adaptation in the reports from parents (b = 0.65) and children (b = 0.52)
Phillips-Salimi et al. [33]. USA	Identify the relationships among adolescents' and parents' perceptions on communication, family adaptability, and cohesion	Cross-sectional, correlational	70 dyads: AYA aged 11–19 (G3&4)	Adolescents' and parents' self-reports on perceptions of (a) open family communication, and (b) problems in family communication (PACS)	Identify the relationships among adolescents' and parents' perceptions on communication, family adaptability, and cohesion (FACES-II)	Controlling for age and sex of AYA and parents, (1) Four predictors found for adolescent-perceived communication: (a) adolescent-perceived family adaptability ($^{\circledR}\beta$ = 0.49), (b) adolescent–family cohesion (β = 0.67), (c) parent–family adaptability (β = 0.33), and (d) parent–family cohesion (β = 0.40) (2) Four predictors found for parent-perceived communication: (a) adolescent-perceived family adaptability (β = 0.34), (b) adolescent–family cohesion (β = 0.33), (c) parent–family adaptability (β = 0.37), and (d) parent–family cohesion (β = 0.43)
Murphy et al. [8]/ USA	Examine potential risk factors for adolescent PTSS at T1 (2 months after diagnosis), T2 (3 months after T2), and T3 (12-month follow-up)	Longitudinal, nonexperimental	41 dyads: Adolescents aged 5–17 (G2, 3, and 4)	Observed maternal communication: macro level at T1 (IFIRS): harsh communication and withdrawn communication; observed maternal communication: micro level at T2 (IFIRS): solicit and validation	Adolescents' and maternal self-report of the PTSS (the Impact of Events Scale–Revised) at T1 and T3	(1) No relationships between maternal harsh/withdrawn communication and adolescent PTSS (2) The indirect effect of maternal PTSS on adolescent PTSS through maternal validation (b = 0.01, kappa^2 = 0.10) (3) Controlling for adolescent PTSS, two predictors found for adolescent PTSS: (a) adolescent PTSS (β = 0.37) and (b) maternal validation (β = −0.32)

Table 1. *Cont.*

Study/ Country	Objective	Design	Sample and Age Range (Groups)	Independent Variable (Measurement and Method)	Dependent Variable (Measurement and Method)	Relationship/ Characteristics of PAC
Bai et al. [36]/ USA	Examine the associations between parent interaction behaviors, parent distress, child distress, and child cooperation during cancer-related port access placement across timepoints (T1–T4)	Longitudinal, nonexperimental	43 dyads: Children aged 3–12 (G2 and 3)	Observation of parent caring verbal/nonverbal interactions: caring parent verbal interaction (P-CaReSS) and nonverbal behaviors (duration)	Observations of (a) child distress, (b) parent/child distress, and (c) child cooperation: (1) verbal/nonverbal child distress (the Karmanos Child Coping and Distress scale), (2) Parent/child distress (the Wong–Baker Faces Scale), (3) Child cooperation (CCS)	Children's low verbal/nonverbal distress found following parents' caring behaviors (eye contact, comforting, supporting/allowing, less availability, verbal protecting, avoiding assumption, believing in/esteem), except for verbal forms of care (e.g., criticizing, apologizing) (Yule's Q ranged from −0.85 to −0.99)
Keim et al. [26]/ USA	Examine the relationships between PAC and adjustment at T1 (enrolment) and T2 (one year later)	Longitudinal, nonexperimental	55 children with advanced cancer; 70 with nonadvanced disease; 60 without cancer as the control group and their mothers: adolescents aged 10–17 (G3 and 4)	Children's self-reports on communication with their mother and father, separately (PACS)	Mothers' self-reports on (a) child adjustment, (b) anxious/depressed scores, and (c) withdrawn/depressed scores (the Child Behavior Checklist)	The relationship between parent–child communication at T1 and child adjustment at T2: (1) Higher maternal openness in communication at T1 predicted lower withdrawn/depressed scores for children with advanced cancer at T2 ($b = -0.14$, $t = 2.82$) (2) Fewer problems in communication with mothers at T1 predicted lower withdrawn/depressed scores for children with advanced cancer at T2 ($b = 0.14$, $t = 3.47$) (3) Higher openness in communication with fathers at T1 predicted lower anxious/depressed ($b = -0.12$, $t = 2.51$) and withdrawn/depressed scores ($b = -0.10$, $t = 2.80$) at T2 in advanced cancer

Table 1. *Cont.*

Study/ Country	Objective	Design	Sample and Age Range (Groups)	Independent Variable (Measurement and Method)	Dependent Variable (Measurement and Method)	Relationship/ Characteristics of PAC
Viola et al. [35]/ USA	Examine associations among problem-solving skills, PAC, parent–adolescent dyadic functioning, and distress	Cross-sectional, correlational	39 dyads: Adolescents aged 14–20 (G4)	Parents' and adolescents' self-reports of parent-adolescent cancer-related communication (CRCP)	Adolescents' self-report of the level of adolescent distress (BSI)	No significant relationship between adolescent-reported cancer related communication problems and adolescents' distress
Tillery et al. [34]/ USA	Identify the relationships between PAC and psychosocial outcomes	Cross-sectional, correlational	165 dyads: adolescents aged 10–19 (G3 and 4)	Parents' self-report of the parent–child relationship quality (PRQ): involvement, attachment, communication (quality of information exchange), parenting confidence, relational frustration	Children's self-report of psychosocial outcomes: (1) post-traumatic stress symptoms (22-item UCLA PTSD Reaction Index for DSM-IV), (2) internalizing difficulties (BASC-2), (3) social functioning (self-regulation, empathy, responsibility, and social competence (SEARS)	Adolescents of caregivers who reported struggling relationship patterns (below average levels of parent–child relationship functioning across several domains) were more likely to report (1) increased level of PTSS ($\chi^2 = 35.06$), (2) elevated levels of internalizing symptoms ($\chi^2 = 10.62$), and (3) poorer social functioning ($\chi^2 = 16.38$) compared to youth of caregivers who reported normative or above average levels of relationship function

Table 1. *Cont.*

Study/ Country	Objective	Design	Sample and Age Range (Groups)	Independent Variable (Measurement and Method)	Dependent Variable (Measurement and Method)	Relationship/ Characteristics of PAC
Al Ghriwati et al. [32]/ USA	Identify subtypes of family relationships and the effects of relationships on child adjustment upon treatment completion within 7 months	Secondary analysis, longitudinal data	81 dyads: Children aged 6–14 (G3 and 4)	Children's self-report of (1) closeness (e.g., how frequently you share information about things that you want others to know) and (2) discord (e.g., how frequently you disagree and quarrel with others) (NRI-RQV)	Caregivers' self-report of children's status: (1) internalizing and externalizing symptoms (CBCL), (2) peer relationships (PROMIS), (3) family functioning (FAD-GFS), (4) quality of life (the Pediatric Quality of Life Inventory 4.0)	(1) Children from families characterized by low closeness/high discord reported significantly greater difficulties with peer relationships ($\chi^2 = 4.12$) and higher externalizing symptoms ($\chi^2 = 5.68$) than those with high closeness/low discord in their families (2) Children from families characterized by low closeness/high sibling-only discord reported significantly poor social functioning than those from high closeness/low discord families ($\chi^2 = 3.99$)
Barrios et al. [39]/ Spain	Explore (1) the openness about cancer, (2) the relationships between the types of communication and children's emotion	Cross-sectional, mixed method	52 dyads: children aged 6–14 (G3 and 4)	Self-report of open communication: the degree of information given to the child as categorized by (1) direct honest information, (2) nuanced or distorted information, and (3) no information at all	Self-report of child's emotion (e.g., fear, anger, sadness, happiness, pain, boredom, loneliness, shame) and mother's subjective emotion (e.g., fear, anger, sadness, frustration, anxiety, guilt) during the qualitative interviews	(1) Truly informed children were much less likely to express fear ($p = 0.011$) (2) The willingness to communicate with parents was higher in children whose mothers did not express anxiety ($p = 0.003$)

Table 1. *Cont.*

Study/ Country	Objective	Design	Sample and Age Range (Groups)	Independent Variable (Measurement and Method)	Dependent Variable (Measurement and Method)	Relationship/ Characteristics of PAC
Park et al. [37]/ Republic of Korea	Identify risk and protective factors for family resilience that affect the adaptation of families of children with cancer	Cross-sectional, correlational	111 dyads: children's mean age of 8.3 (N/A)	Parents' self-report of family communication (the Family Problem-Solving Communication Scale)	Parents' self-report of family resilience (adaptation) (APGAR questionnaire)	Family communication skills found to be predictive of family adaptation ($\beta = 0.40$)

PAC, parent–adolescent communication; FPSC, Family Problem Solving Communication; FACI8, the Family Attachment and Changeability Index 8; AYA, adolescents and young adults; PACS, Parent Adolescent Communication Scale; FACES-II, Family Adaptability and Cohesion Evaluation Scales; T1, time 1; T2, time 2; T3, time 3; T4, time 4; IFIRS, he Iowa Family Interaction Scale; PTSS, post-traumatic stress symptoms; P-CaReSS, Parent Caring Response Scoring System; CCS, Child Cooperation Scale; CRCP, Cancer-related Communication Problems Scale; BSI, Global Severity Index of the Brief Symptom Inventory; PRQ, Parenting Relationship Questionnaire; PTSD, post-traumatic stress disorder; BASC-2, Behavior Assessment System for Children, Secondary Edition; SEARS, Social and Emotional Assets and Resilience Scale; NRI-RQV, modified network of Relationship Inventory-Relationship Qualities Version; CBCL, Childhood Behavior Checklist; PROMIS, Patient-Reported Outcome Measure Information System; FAD-GFS, Family Assessment Device-General Functioning Scale; APGAR, Family Adaptability, Partnership, Growth, Affection, and Resolve; G1, infants (~1 year); G2, toddlers/preschoolers (2–5 years); G3, school age (6–12 years), G4, adolescence (13–24 years).

3.3. The Characteristics of Parents of Children or Adolescents with Cancer

Seven out of ten studies included a self-report from either the mothers or fathers of children or adolescents with cancer [32–38]. In the case of proxy reports, the majority of reporters were mothers [8,26,34,35,37–39]. However, none of the included studies provided information regarding the possible influence of parents' sex on communication practice. It is noteworthy that two studies addressed the differences in communication between mothers and fathers [26,33]. In one of the studies, children's reports on mothers' and fathers' communication were analyzed separately, indicating that fathers' patterns of openness and problems in communication were similar to mothers' communication patterns [26]. In another study, only the parent corresponding to the biological sex of the patient was included for analysis [33].

Most articles provided demographic information about parents' sex and age range, but limited demographic information was provided. Regarding parents' marital status, only three articles indicated that a majority of parents were married [32,37,38]. None of the studies included marital status as a factor in assessing its potential influence on parent–child communication.

3.4. Parent–Child Communication in the Childhood Cancer Context

3.4.1. Types of Communication Being Measured

Each of the 10 studies used a different definition of parent–child communication and measured it differently. A majority of studies (*n* = 8) focused on measuring verbal communication only [26,32–35,37–39], while two studies measured both verbal and nonverbal communication [8,36].

The verbal communication measured included the degree of openness of the parent–child communication [26,33,39], the attitude of the persons communicating [32,37,38], or both the attitude and amount of information shared during the communication [34,35]. For example, the degree of openness of communication was assessed based on whether they communicated about their disease or emotions without holding back or had nondistorted communication [26,33,39]. In the articles retained, the attitude during the communication was described as either affirming or incendiary [38] using the frequency of agreement or disagreement [32], and the parent–child relationship was described by the level of involvement or attachment, the amount of information sharing that was present, parenting confidence, and relational confidence [34].

Two studies measured both verbal and nonverbal communication. For example, Bai et al. [36] measured the presence or absence of caring verbal and nonverbal interactions during port insertion, and Murphy et al. [8] observed maternal communication (i.e., whether it was harsh or withdrawn at the macro level) and whether maternal communication involved solicitation, which was defined as questions asked by mothers to elicit children's responses or validation at the microlevel.

3.4.2. Types of Communication Being Measured

The majority of studies relied on self-report measures of children, adolescents, and their parents (proxy) [26,32–35,38,39]. Three studies included both parents' and children's reports of their parent–child communication practices by having participants complete the same measures, which included the Parent-Adolescent Communication Scale (PACS) to assess openness and problems in parent–child communication, the Family Problem Solving Communication (FPSC) to assess affirming and incendiary communication, and the Cancer-related Communication Scale (CRCP) to measure cancer-related communication problems [33,35,38]. In the rest of the studies, the children/adolescents or parents were asked to report parent–child communication characteristics using different measurements, such as closeness and discord in communication, using the subscale of the modified Network of Relationship Inventory-Relationship Qualities Version (NRI-RQV) [32], children's report of their parents' openness and problems in communication practices using PACS [26], and parent reports of closeness and discord in communication using the Parent–Child Relationship Quality Questionnaire (PRQ) subscales on attachment and communication [34]. In one mixed-design study, the types of parent–child communication were assessed through parent interviews and coded as three types of communication including direct, honest information, nuanced or distorted information, and no information [39]. All of the measures were reliable and psychometrically valid. The Cronbach's alpha of reliability ranged from 0.83 to 0.94 in each study, except the CRCP, with a Cronbach's alpha of 0.69 (adolescent) and 0.61 (parents), which was originally developed to measure cancer-related communication problems between partners in the adult cancer context [40].

Among the 10 studies, only 2 studies involved observations performed by trained research assistants [8,36]. In one observation study, Bai et al. [36] used the Parent Caring Response Scoring System (PCaReSS) to measure the presence or absence of caring verbal and nonverbal communication during port insertion, with an 80% agreement between the trained coders. The second observation study by Murphy et al. [8] used the Iowa Family Interaction Scale (IFIRS) to measure harsh or withdrawn communication or communication involving either solicitation or validation, demonstrating an inter-rater reliability of 80%, with average intraclass correlations exceeding 0.60.

3.5. The Influence of Age and Developmental Stage

Among the diverse demographic variables that could possibly impact parent–child communication practices, age and the developmental stage were examined in this literature review due to the inclusion of a wide range of age groups. We found that the age group of study participants in this scoping review was broad, ranging from 3 (toddler) to 24 (late adolescence). One out of ten articles did not provide specific information regarding participants' age range but only the mean. Among the articles that provided the age range, the broadest one included participants aged between 3 and 12 years (from toddler to adolescence) [36]. Despite the broad age range, the influence of age was not the focus of any of the studies included. However, few studies examined or considered the influence of age on communication practices. For example, Murphy et al. [8] found that harsh maternal communication was negatively related to adolescent age, suggesting that mothers of older adolescents demonstrated less irritability and frustration with their children during communication. Phillips-Salimi et al. [33] recognized the influence of age on parent–child communication and controlled for it in a statistical analysis of the relationships among adolescents' and parents' perceptions of communication, family adaptability, and cohesion. Keim et al. [26] also found that age was related to problems in communication with mothers at T2 (one year later), suggesting that older age was related to more problems when communicating with mothers. However, they did not consider age as a covariate in examining the relationship between parent–child communication and child adjustment. Barrios et al. [39] invited children and adolescents aged 6–14 years old to an interview study and asked the frequently reported emotion by themselves according to the age group (younger: 6–9 years, older children: 10–14 years). They found that older children (10–14 years old, 82.1%) were more likely to have received forthright and truthful information about their illness than younger children (6–9 years old, 37%), and children who received truthful information were significantly less likely to mention fear. Finally, two studies conducted by Tillery et al. [34] and Al Ghriwati et al. [32] did not find any differences in communication practices based on participants' age.

3.6. The Relationship between Parent–Child Communication and Physical Health Outcomes

There were no studies that measured the relationship between parent–child communication and physical health outcomes among the articles considered. All studies measured different types of psychological health outcomes at different levels.

3.7. The Relationship between Parent–Child Communication and Psychological Health Outcomes

The majority (90%) of the studies supported a positive association between parent–child communication and psychological health outcomes. The psychological health outcomes found to be beneficial included decreases in externalizing problems [32], internalizing problems, anxiety, [34], child's anxiety level/depression, child's withdrawal/depression scores [26], adolescent's post-traumatic distress [8], child's distress during port insertion [36], and child's report of their perceived fear during treatment or hospitalization [39], and increases in the child's self-report of their peer relationships and their quality of life [32], and enhanced social emotional strengths (self-regulation, empathy, responsibility, social competence). Table 2 provides the summary of the characteristics and behaviors associated with positive psychological outcomes.

Table 2. Characteristics and behaviors associated with positive psychological outcomes.

Characteristics of Verbal Communication	**Associated Psychological–Behavioral Outcomes**
Affirming [37,38]	Better family adaptation
Open [26,33,37]	Less anxiety, less depression, better family adaptation
Satisfying [33]	Better family adjustment and cohesion
Maternal validation [8]	Lower PTSS
Avoiding assumptions [36]	Less behavioral and verbal distress during procedure
Believing what the other says [36]	Less behavioral and verbal distress during procedure Lower level of PTSS
Quality of information shared [34]	Less internalizing symptoms, Increased level of social emotional competencies
Low discord [32]	Better peer relationships, less externalizing problems
Truthful, honest communication [39]	Reduced fear
Characteristics of Nonverbal Communication	**Associated Psychological–Behavioral Outcomes**
Eye contact [36]	Less behavioral and verbal distress
Being physically close enough to touch [36]	More cooperative behavior during procedure
Acknowledging behavior [8]	Lower PTSS

PTSS, post-traumatic stress symptoms.

Bai et al. [36] found that children were significantly less likely to display behavioral and verbal distress during the invasive port insertion procedure following parents' caring verbal (e.g., avoiding assumptions, believing in self-esteem) and nonverbal communication, involving eye contact and being within a distance close enough to touch the child or the child to touch the parent. Barrios et al. [39] reported that children who received truthful information about their diagnosis were significantly less likely to mention fear during treatment or hospitalization in their qualitative design study. The importance of quality of information exchange was also emphasized [34]. They found that youths reported elevated levels of post-traumatic stress symptoms (PTSS), internalizing symptoms, and lower levels of social emotional strengths when they were struggling in maintaining desirable parent–child relationships by exchanging needed amounts of information through open and honest communication. Al Ghriwati et al. [32] examined family relationship profiles on closeness (i.e., talking about things that are supposed to be known) and discordance (i.e., disagreement with each other) and the effects on child adjustment during treatment completion. They found that children from families characterized by low closeness/high discord reported significantly greater difficulties with peer relationships compared to those who were from families with high closeness/low discord in their families.

The positive association between parent–child communication and psychological outcomes was also supported in two longitudinal design studies [8,26]. Murphy et al. [8] examined the effect of maternal PTSS on adolescents through maternal communication practice. Maternal validation mediated the negative effects of maternal PTSS on adolescent PTSS at one year. Maternal validation in communication was described as maternal behavior that acknowledges and validates the child's verbalizations through the use of backchannels and assents. Thus, maternal validation in communication significantly reduced adolescents' PTSS, controlling for adolescents' PTSS at baseline.

Researchers also found some parent–child communication characteristics that were related to better family function, such as family cohesion and adaptability. For example, Philips-Salimi et al. [33] found that adolescents who perceived poor parent–adolescent communication, which is described as poor quality of communication (e.g., satisfaction with the way they communicate with each other), reported lower adolescent-perceived family adaptability, adolescent-perceived family cohesion, parent-perceived family adaptability, and parent-perceived family cohesion, after adjusting for the age and sex of the AYAs and their parents. Thus, the researchers concluded that adolescent–parent communication played a significant role in fostering a positive family environment. Similarly, Park et al. [37] found that affirming family communication was one of protective factors of family adaptation by providing children emotional support and encouragement. Greeff et al. [38] also found that affirming family communication contributed to family adaptability. Affirming family communication is characterized by taking time to hear what each other has to say or feel, working hard to ensure that family members are not emotionally or physically hurt, and respecting each other's feelings.

Only one study did not find a significant association between parent–child communication and psychological health outcomes [35]. Viola et al. [35] evaluated the associations among problem-solving skills, parent–adolescent cancer-related communication, parent–adolescent dyadic functioning, and distress in adolescents with cancer by employing the disability–stress–coping model. They found that there was no association between cancer-related communication and adolescents' distress.

4. Discussion

This review aimed to explore the characteristics of communication that contribute to positive health outcomes in the childhood cancer context and positive physical or psychological health outcomes. Our main finding was that there is preliminary evidence of a relationship between some aspects of parent–child communication and psychological health outcomes in the childhood cancer context, but there is a dearth of evidence related to physical health outcomes.

This review contributes new knowledge by identifying the specific characteristics of therapeutic parent–child communication, which contributes to better patient outcomes in the childhood cancer context. For example, although there are some existing guidelines for parents encouraging honest and open communication with children with cancer and instructions on disclosing a child's cancer diagnoses in a safe manner, there are no specific parent–child communication guidelines that can be applied in daily life to improve patient outcomes. The additional contribution of this review to current knowledge is identifying what positive health outcomes have been studied in relation to parent–child communication. It is surprising that physical health outcomes have not been studied and presents an area of opportunity. The relationship between therapeutic interpersonal communication and positive physical health outcomes in adults with chronic illness has been widely explored. Various research studies have shown the linkage between social interaction with significant others and physiological outcomes in adult cancer survivors, which include neuroendocrine, heart, and immune functioning [41,42]; symptom distress [43,44]; and levels of stress hormones [45]. Thus, expecting a positive relationship between therapeutic parent–child communication and physical health outcomes would be reasonable. Potential physical health outcomes to explore include symptom severity such as nausea or febrile neutropenia or even treatment adherence and disease outcomes.

Most importantly, the results of this review may be meaningful to children and adolescents with cancer and their parents. In previous studies, pediatric patients with cancer and their parents reported a fear of the expected disadvantages of communication, such as hurting each other by sharing sensitive topics, as the main barrier to engagement in parent–child communication [46]. Thus, there is a need to identify the positive outcomes related to communication.

In this review, several weaknesses in parent–child communication studies were identified. Despite the evidence supporting the relationship between parent–child communication and positive psychological health outcomes, there is a lack of understanding about the mechanisms through which this occurs. For example, researchers have pointed out the need to examine the mechanism of the relationship between therapeutic family communication and coping [47,48]. Thus, future studies involving mediator, moderator analysis, or structural equation modeling around potential mechanisms are required to promote our deeper understanding.

Second, most studies focused on different characteristics and different aspects of psychological outcomes; thus, definitive conclusions are not possible. Additionally, we found that most of the studies employed cross-sectional, nonexperimental designs, with only two studies using a longitudinal design. This limitation makes identifying a causal relationship between therapeutic parent–child communication and positive health outcomes impossible.

Wide variations in terms of sample age, time since diagnosis, and diagnosis of the child with cancer were not considered in examining the association between parent–child communication and positive health outcomes. Children and adolescents require different needs in their parent–child relationship and developmental tasks according to developmental stage [49,50]. Age-related cognitive abilities would also impact communication practices with parents. In this review, 6 out of 10 articles considered age/developmental stage as a potential factor that might influence parent–child communication practices; only 2 of them reflected it in their analysis. However, the impact of age on parent–child communication practice was previously supported [8]. Future studies should either consider the influence of age on communication practices, potentially powering studies for subgroup analyses based on age or developmental stage.

In previous studies, children reported that their physical condition clearly affected their preference for being involved in parent–child communication [51]. Future studies might consider providing detailed information about the clinical characteristics of study participants or setting inclusion/exclusion criteria to minimize the influence of clinical status on parent–child communication practice or, at the very least, studies could be designed to control for these differences. Despite including studies from diverse countries, most of the population studied were White and conducted in the United States. Only one study mentioned the potential influence of culture on the relationship between parent–adolescent communication and family adaptation [37]. However, none of the remaining studies reported cultural differences and their influences on parent–child communication. There is a need to include diverse populations considering the impact of cultural background on family communication practices.

Another limitation would be that most of the included studies lack reports of parent–child communication practices from the involved participants, by including responses either from one parent or children and adolescents but failing to provide a full description of parent–child communication practices. In addition, most of the literature relied on self-report measures rather than observation. Among the literature identified, only two articles employed observation with high inter-rater reliability [8,36]. Although all studies used reliable and valid measurements, and the retrospective self-report measure is one of the most widely used methods [52], diverse data collection methods, such as observation, are required to collect objective data. Psychologists have acknowledged the importance of direct observation in assessing family communication patterns, despite some limitations [53]. In a recent topical review, Murphy et al. [54] also emphasized the objective assessment of parent–child communication practices using a reliable and unified framework by observing communication practices.

In the case of proxy reports, the majority of responses came from mothers. This might be reasonable because mothers spend the most time with their children or adolescents with cancer at their bedside or at home. Thus, mothers might be the best person to share their communication experiences. However, considering the role of fathers in parenting, the parent–child communication with fathers also needs to be assessed [55]. In addition, there is evidence showing that parent–child communication practices might be different depending on their parents' sex [56]. In our review, only two studies included assessment from fathers' perspectives [26,33]. Future studies should consider the inclusion of fathers' reports to assess the triangle of communication among the child–mother–father. Additionally, considering the potential influence of parental relationships on parent–child communication [57], future studies should assess the quality of the parental relationship in studying parent–child communication. Although this review focused on parent–child communication in general nuclear families, there is also the issue of defining the parent(s) in different types of families. For example, grandparents play an important role in some extended families [58]. Additionally, it would be possible to have one parent or stepparents in divorced families. The family dynamics and parent–child communication practices might be slightly different in those families. Future research examining parent–child communication practices in the diverse forms of families is required.

Next, when employing self-report measurements, there is a need to employ age- and context-specific measurement to assess parent–child communication practices in the childhood cancer context. Although all of the studies included in this review used reliable and valid measurements to assess parent–child communication practices, we found that no measurement was age- and/or context-specific. For example, 3 out of 10 studies assessed openness/problems in parent–adolescent communication using the Parent–Adolescent Communication Scale (PACS) [26,33,39]. Despite its reliability and validity, the PACS was not specifically developed to assess parent–adolescent communication practices in the childhood cancer context. The Cancer-Related Communication Problem Scale (CRCP) was originally developed to assess whether patients and their partners have difficulty talking about cancer with each other, which had acceptable reliability in the original study [40] but presented poor reliability in the study with adolescents with cancer and their parents.

Despite the contributions of this current study, some limitations should be acknowledged. First, this study only included 10 articles and 9 out of 10 studies supported the relationship between parent–child communication and psychological health outcomes. This small sample size could not fully explain the relationship between parent–child communication and positive health outcomes. Second, this study focused on including only parent–child communication as one core aspect of family support. However, parent–child communication has multifaceted aspects that reflect the parent–child relationship, and a variety of other variables, such as development stage, also affect it. Future studies should consider the impact from other key variables in determining the relationship between parent–child communication and positive health outcomes. This study only included English-written studies. Although the included studies originated from diverse countries, such as the Republic of Korea, Spain, and Belgium, this might have caused bias by over-representing research from English-speaking countries. Finally, this study mainly focused on assessing verbal communication despite 2 out of 10 articles assessing nonverbal communication. Nonverbal communication, however, is also an important aspect in communication practices, which is mainly in charge of expressing emotions [59]. Future studies should consider including further studies assessing both verbal and nonverbal communication.

There is an emerging body of evidence that therapeutic parent–child communication contributes to better psychological health outcomes at both the individual and family levels in the childhood cancer context. The next step is to test these identified characteristics to examine the causal relationship between parent–child communication and positive health outcomes to identify the predictors of positive health outcomes. Once predictors are identified, interventions to teach those characteristics of therapeutic parent–child communication can be developed and tested.

While this review does not provide strong evidence supporting the causal relationship between therapeutic parent–child communication and positive psychological health outcomes, the evidence still supports the importance of parent–child communication in the childhood cancer context. Thus, it is important to identify children/adolescents with cancer and their families who struggle in communicating with each other and to monitor them for poor psychological health outcomes. Expressing and sharing emotions are regarded as some of the most powerful predictors in the adjustment to a cancer diagnosis [60]. Most importantly, parent–child communication does not occur in a vacuum. Thus, assessing overall parent–child relationships with a special focus on parent–child communication is recommended.

5. Conclusions

There is emerging evidence supporting the association between therapeutic parent–child communication and positive psychological health outcomes and the characteristics of that therapeutic communication. Future longitudinal-design studies are warranted to enhance our understanding of the causality and underlying mechanisms of the relationship between the relevant characteristics of therapeutic parent–child communication and its psychological health outcomes. Also, future study is required to explore more physical health outcomes from the engagement in therapeutic parent–child communication. The findings would contribute to the future development of parent–child communication interventions to improve psychological and physical health outcomes in the childhood cancer context. Finally, it is still important to identify children/adolescents with cancer and their families who struggle with engaging in therapeutic parent–child communication and to monitor them for poor psychological health outcomes.

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cancers

Article

Factors Affecting Psychosocial Distress in Adolescents and Young Adults with Cancer: BRIGHTLIGHT Cross-Sectional and Longitudinal Cohort Study Results

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Simple Summary: Adolescents and young adults with cancer (AYAs) have biopsychosocial needs that differ from younger children and older adults. Facing cancer during this complex developmental stage can result in mental health consequences, historically not adequately addressed by pediatric or adult healthcare systems, nor by researchers. This study adds a deeper understanding of risk and protective factors for distress in AYAs, including the paradoxical impacts of social support. It also offers a longitudinal description of symptoms, with depression improving over time while anxiety remained stable. Future implications include informing the design of developmentally tailored screening and intervention tools to mitigate distress in this understudied population.

Abstract: Background/Objectives: AYAs face unique psychosocial challenges compared to younger children and older adults. Little published data exists about symptoms of psychological distress in AYAs, particularly in large cohorts over time. This study aimed to (1) explore relationships between distress and variables known to impact mental health, (2) examine distress over time, and (3) consult with a patient and public involvement group (Young Advisory Panel, YAP) to contextualize the results. Methods: Secondary data analyses were conducted from a multicentre longitudinal cohort study, which recruited 830 participants aged 13–24 years newly diagnosed with different cancers. The mean age was 20.1 (SD 3.3) and 45% were female. Bespoke surveys, including a validated measure of psychosocial distress, were administered five times over 3 years following diagnosis. Participatory workshops were conducted with the YAP to gather their impressions of the study results and inform conclusions. Results: At the first time point, higher social support was associated with higher distress scores ($\beta = 0.202$, $p < 0.001$) and AYAs with higher depressive symptoms were more likely to see a mental health professional ($p < 0.01$). Over time, anxiety symptoms remained stable ($p = 0.090$); however, depressive symptoms improved ($p < 0.001$). The YAP highlighted the mixed effects of social support and the under-recognition and undertreatment of distress. Conclusions: This information identifies key targets, such as social support, that can inform the design of personalized screening and intervention tools. It also highlights specific timepoints at which these efforts can be directed to mitigate distress and improve quality of life. Future research should include

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longer-term follow-up, qualitative explorations of distress along the disease trajectory, and ongoing co-production efforts.

Keywords: cancer; young people; adolescents; young adult; BRIGHTLIGHT; mental health; distress; depression; anxiety

1. Introduction

Each year worldwide, there are approximately 1.2 million new cases of cancer in young people aged 15–39 years, representing 6% of all new cancer diagnoses [1]. After accidents, cancer ranks as the next most common cause of death in young people in the United Kingdom (UK) [2]. Adolescents and young adults with cancer (AYAs) are defined in the UK as those aged 16–24 [3] and new diagnoses number approximately 2400 annually [4]. Cancer in AYAs presents distinct biological and psychosocial considerations that require special attention [5,6].

Cancer biology, genomics, and host pharmacokinetics in AYAs differ from younger children and older adults and are poorly understood [7]. Among the most common cancer types occurring in this age group are lymphomas, thyroid carcinomas, testicular tumours, and melanomas [5,8]. Prolonged and complex pathways to diagnosis are common, due in part to a lack of awareness in the medical community and general public, risks of treatment side effects are higher, and survival rates have not improved the way they have in other age groups [9–12]. Additionally, enrolment in clinical trials is very limited, preventing advances in new drug development, and thus impacting cure rates [8,13,14]. Long-term morbidities from treatment include chronic cardiac, musculoskeletal, neurocognitive, and endocrine conditions, as well as the risk of second primary cancers [15,16]. In those who survive into older age, their life trajectories may be profoundly altered, representing a significant number of disability-adjusted life years impacting individuals, families, and society [17,18].

Although cancer is disruptive at any age, a diagnosis during this phase of life has a particularly acute psychosocial impact, affecting body image, peer and romantic relationships, fertility, family dynamics, education, employment, future life plans, and mental health [19–21]. Adolescence and young adulthood are inherently dynamic periods of development without a cancer diagnosis, and these life stages have become increasingly complex in recent times [22]. Nearly three-quarters of adult mental health problems are thought to start during this pivotal life stage [23]. Cancer fundamentally disturbs this already precarious developmental trajectory, isolating youth precisely when they want to fit in [24]. Furthermore, AYAs have a greater ability compared to younger children to understand the severity of their situation but have fewer emotional, informational, and practical resources as compared with older adults to help cope [25].

Not only do AYAs with cancer have unique biopsychosocial needs, but they fall into the gap between cancer care services designed for younger children, where the average age is 6, and older adults, where the average age is 66 [26,27]. They are “squeezed between, and insufficiently addressed by, the achievements of the pediatric oncology world on the one hand and the weight of cancer burden on adult cancer services on the other” [28]. Described as ‘no man’s land’ or ‘the lost tribe,’ AYAs experience a lack of autonomy, peer connection, support to complete developmental milestones, appropriately trained staff, and tailored healthcare environments [29,30]. The advent of high-quality AYAs specialist services and support organizations in many countries has begun to address AYAs needs, but these services currently only cover a minority of AYAs worldwide and generally require more comprehensive resources to fully support this population. Therefore, it is unsurprising that AYAs with cancer experience high levels of psychological distress [24]. Psychological distress, defined for this paper as an overall measure of anxiety and depression [31], can impact adherence to treatment, overall morbidity and mortality, experience of healthcare services, as well as quality of life [32]. However, evidence about anxiety and depression in AYAs with cancer is limited, including prevalence in large samples and symptom trajectories over time. Understanding the distinct psychological needs of this population may help reveal mechanisms for addressing these disparities and mitigate the negative consequences of cancer care.

The body of evidence examining mental health symptoms in AYAs with cancer is limited but has gradually increased over the last two decades. This growth may be attributed to a rising recognition of the importance of providing supportive care for cancer patients as survival rates improve, in parallel to searching for new cures [33]. Another contributing factor may be the change in Western societal attitudes toward an awareness of mental health issues [34].

However, there are methodological challenges in AYAs cancer research including varying definitions of the AYAs age range, studies that group AYAs in with other age categories, small sample sizes, multiple cancer types, lack of validated measures for this age group, and few longitudinal studies [33,35].

Estimates of the prevalence of distress in AYAs with cancer are based on a small number of systematic reviews and descriptive studies, with age ranges including 12–25, 15–24, and 15–39, among others [20,36,37]. Rates of anxiety range from 8 to 55% and depression from 13 to 47% [31,38–40]. Compared to older people with cancer, siblings, and peers, AYAs face higher risks for these conditions [31,41,42]. Some evidence suggests that AYAs experience improvements in anxiety and depression over time, though the trajectory and timing of these changes vary across studies [43]. The generalisability of much of the research on this issue is questionable, due to small sample sizes, inconsistently applied psychometric measures, variation in diagnoses and complexity of treatment across studies, and primarily cross-sectional designs. However, despite these limitations, there is consensus and concern that psychological needs are high and remain unmet, and that an understanding of the developmental context of this population is crucial and lacking [35,37]. Contextual factors such as patient, social, institutional, and healthcare system level resources are also important considerations [20].

Multiple risk factors have been identified in the literature as contributing to increased distress in AYAs with cancer [20,44,45]. Female gender [46–48], older age at diagnosis [47], migration background [46], and having a brain tumour diagnosis [40] have all been implicated. However, factors that feature consistently across multiple studies include various proxies of poor social support (e.g., difficulties with social relationships, not being in a partnership, social isolation, and poor family functioning) as well as disease-related features including illness relapse or a higher number of late effects [20,40,45,49]. Certain psychological characteristics have also been identified, such as maladaptive coping skills, low self-image, and identity issues [40,48]. These commonly arising factors are explored in more detail as follows.

Social support is defined by the National Cancer Institute’s Dictionary of Cancer Terms [50] as “a network of family, friends, neighbours, and community members that is available in times of need to give psychological, physical, and financial help”. Social support has been observed to alleviate distress in AYAs and potentially improve with intervention [51]. A defining developmental task of this life stage is to individuate from family and shift toward peer influences, a challenge even when healthy [52]. A cancer diagnosis and its treatment can disrupt a young person’s evolving social network to varying extents depending on the illness severity, treatment intensity necessitating repeated or prolonged hospital admissions, emotional development of the patient, and capacity of family and friends to cope with the situation [20,51]. Qualitative focus groups with 25 AYAs aged 16–39 at the time of the study identified social isolation as a major psychosocial dilemma, encompassing separation from peers due to treatment, loss of independence due to reliance on caregivers, and feeling like an outsider in the medical setting (either the youngest person in the adult clinic, or the oldest person in the pediatric clinic) [45]. Limited evidence from longitudinal observational studies proposes that social functioning in AYAs at diagnosis is low, and though it improves over time, it remains reduced compared to healthy peers [53]. While the general trend across the literature shows increasing distress with worsening social support, this area remains understudied.

Disease-related features in AYAs associated with higher levels of distress include diagnoses such as brain tumours, head and neck cancers, and testicular cancers [40,54]. In a population-based study of 572,000 AYAs, rates of suicide—a serious potential consequence of extreme distress—were higher among those with metastatic illness [54]. This corroborates evidence that relapsed disease predicts poorer emotional health [55]. Long-term treatment complications detailed above also contribute to worsening mental health [46,56]. Following from the evidence, specific features of an AYA's illness, while not necessarily modifiable, could inform the early stratification of patients to individualized support.

Finally, psychological characteristics have been correlated with increased distress in AYAs [57]. In a systematic review of predictors of psychiatric disorders in AYAs, four studies found that AYAs engaged in maladaptive coping strategies, such as wishful thinking and avoidance, are more often compared to healthy peers [40]. Self-image and identity issues in a cross-sectional, multi-centre study of 196 AYAs were associated with elevated distress [48]. However, few if any studies have directly examined the relationship between pre-existing mental health conditions and distress. These conditions could be screened for at diagnosis, helping allocate support to those who may need it most.

Little is known about the change in symptoms of distress in AYAs over time along the disease trajectory. An older prospective observational study of 61 Swedish adolescents aged 13–19 indicated improved psychosocial functioning over the first 18 months after diagnosis [44]. A follow-up study of the same cohort over a much longer period of 10 years showed a non-linear pattern, with anxiety and depression declining around the 2-year mark, and then increasing gradually over the subsequent 8 years [58]. Another similarly designed study of 514 German AYAs aged 18–39 found rates of anxiety and depression remained stable over two time points (the first within 4 years of diagnosis, and the second 12 months later) [49]. There have been few attempts worldwide to quantify this problem longitudinally. Qualitative exploration is also needed, to examine possible reasons behind observed time course patterns.

The objectives of this study were to illustrate the mental health burden in a large, geographically diverse, well-characterized cohort of AYAs with cancer aged 13–24 at diagnosis and examine risk and protective factors that affect mental health in the cohort. The primary aim was to explore relationships between distress and variables described in the literature known to impact mental health, including social support, disease severity, pre-existing mental health conditions, and contact with mental health professionals. The associated hypotheses were that lower levels of social support, higher disease severity, presence of a pre-existing mental health condition, and contact with mental health professionals would be associated with higher levels of distress. The secondary aim was to examine these associations over time. Distress was hypothesized to decrease with passing time. The final aim was to consult with an established patient group of young people about the results and seek their interpretation based on their own experiences.

A deeper understanding of the distinct psychological needs of this population will help inform the design and evaluation of individualized, effective screening processes and interventions, targeted at key time points, to reduce distress and mitigate the negative biopsychosocial consequences of cancer care during this dynamic life stage.

2. Materials and Methods

2.1. Study Design

To characterize distress in AYAs and examine changes over time from diagnosis, a secondary analysis of raw data from the BRIGHTLIGHT study was conducted. BRIGHTLIGHT was a national programme of research, central to which was a multicentre, prospective, longitudinal cohort study, aiming to determine whether specialist care for AYAs in England was associated with improved outcomes [59–61]. A patient and public involvement group (PPI) Young Advisory Panel (YAP) of AYAs with cancer were involved in BRIGHTLIGHT from the outset as co-researchers, naming the study, developing the design, research questions, and outcome measures, disseminating results, and identifying future areas for research. PPI in this context is understood as conducting research together with the public instead of on them [62].

2.2. Participants and Setting

The BRIGHTLIGHT cohort consisted of 1114 young people aged 13–24 years, recruited within 4 months of a new cancer diagnosis, between 2012 and 2015 from 97 hospitals across England. Recruitment primarily occurred within adult cancer services, but in hospitals offering both child and adult care, individuals aged 13–16 were also enrolled. The cohort represented one-fifth of the total population diagnosed at the time in the UK. Those who did not participate in the cohort were similar in age and ethnicity to cohort participants but had a lower proportion of males and different proportions of cancer diagnoses (lower proportions of leukemia and lymphoma, germ cell tumours and bone tumours and higher proportions of brain tumours, skin cancers, and carcinomas compared to the BRIGHLIGHT cohort).

People with language or sensory impairments impacting communication were included. Young people were excluded if they had a prognosis of less than 6 months from the time of diagnosis, received a custodial sentence, or were unable to complete a survey [60]. The Confidentiality Advisory Group of the Health Research Authority (ECC 8-05[d]/2011) and the London Bloomsbury NHS Research Ethics Committee (reference 11/LO/1718) approved the study, including activity involving the YAP [62].

2.3. Procedures and Measures

Data were collected through a study-specific survey, administered by an independent research organization across five time points: 6, 12, 18, 24, and 36 months following diagnosis [60]. Interviews were conducted in person at the first time point and by telephone or online for the subsequent time points. The co-designed survey (available under licence from https://xip.uclb.com/i/healthcare_tools/brightlight_wave1.html, accessed on 30 January 2025) contains demographic items, validated outcome measures, and questions about cancer care.

This paper reports socio-demographic data and clinical characteristics shown in the literature to impact mental health. Information was collected about participants' age, gender, ethnicity, marital status, geographic location, socioeconomic status, and employment status. Clinical characteristics included cancer type, time to diagnosis, disease severity at diagnosis, prognostic score, and treatment type. The severity of illness was determined using a bespoke grading system incorporating the burden of symptoms, treatment, and late effects, as well as predicted survival. A grade of low, intermediate, or high severity was assigned to each cancer type [60]. An existing scoring system was used to determine patient prognosis at the first time point, using anticipated 5-year survival to separate young people into those with an expected survival of less than 50%, 50–80%, and above 80% [63,64].

This paper also details the psychosocial characteristics of the cohort to approximate their mental health status, including scores on the Hospital Anxiety and Depression Scale (HADS) [65], the Multidimensional Scale of Perceived Social Support (MSPSS) [66], and the self-reported presence or absence of a pre-existing mental health condition.

The HADS was used to measure patient-reported distress, including symptoms of anxiety and depression. Summary scores range from 0 to 21, with 8–10 representing borderline and 11 or higher representing moderate to severe levels of anxiety and depression [67]. The HADS is suitable for individuals with cancer because it does not include potentially confounding questions about physical symptoms [68]. It has been found to be reliable and validated for use in the cancer population [69,70].

Social support was measured using the MSPSS, which identifies an individual's perceived level of social support from family, friends, and significant others [66]. Example items include "I get the emotional help and support I need from my family", "I have friends with whom I can share my joys and sorrows", and "I have a special person who is a real source of comfort to me". The total support score is reported as an average ranging from 1 to 7. Low support is indicated by a total score of 1–2.9, moderate support by a score of 3–5, and high support by a score of 5.1–7. The measure also has domain scores for support from family, friends, and significant others. The MSPSS has been validated and shown to be reliable in people with cancer [71,72].

Data on whether young people were offered and had contact with a mental health professional at the first time point were also collected, to assess referral numbers and service utilization.

Covariates were selected a priori based on established literature linking psychosocial distress in AYA cancer populations to demographic and clinical factors.

2.4. Data Analysis

Descriptive data for demographics were collected, including means and standard deviations, medians and interquartile ranges, frequencies, and percentages. Statistical analyses were performed using IBM SPSS Statistics version 29.0.2.0 software for Mac. A value of $p < 0.05$ was taken as statistically significant with 95% confidence intervals calculated.

Poisson regression was used to examine the association between distress and social support, adjusting for confounders including gender, age, marital status, socioeconomic status, employment status, and ethnicity. This type of analysis was chosen over linear regression as the former summarizes the outcome as an average rate, which is more appropriate for discrete variables, whereas the latter summarizes the outcome as an average.

One-way ANOVAs were performed to elicit the relationship between distress and disease severity, with Bonferroni corrections for post hoc analyses to determine where significant differences lay. Independent samples *t*-tests were used to explore distress and pre-existing mental health conditions, as well as contact with mental health professionals. Mann–Whitney U tests were used for small sample sizes. A repeated measures ANOVA was used to analyze the change in distress across the five time points.

2.5. Patient and Public Involvement

To help inform the interpretation and enhance the validity of the study results, all ten active members of the BRIGHTLIGHT YAP were invited to contextualize the findings through their own cancer experiences. Two participatory workshops were conducted online with the YAP and co-facilitated by the research team. The goals of the workshops were to present the key study findings to the YAP, examine their impressions of the results, and explore insights for future research and clinical directions. All participants consented to participation, recording, and using anonymized feedback. Before each workshop, the YAP were given agendas, consent forms, and a written summary of the study results.

The workshops were 1.5 h each and involved a slide presentation of key study findings, followed by a Zoom (version number 6.3.11 (50104)) interactive whiteboard exercise and a focus group discussion [73]. For the first workshop whiteboard exercise, four headings were posted across the top of the board representing each of the variables to be discussed (social support, gender, disease severity, and pre-existing mental health conditions). Three questions were posted down the left side of the board to stimulate thought and facilitate participation for those with treatment-related cognitive difficulties ('Does this make sense and why?', 'How does this relate to your experience?', and 'Any other thoughts?').

In the second workshop, the five time intervals assessed were posted across the top of the board and 'things I was anxious about' and 'things I was worried about' were posted down the left side. During the whiteboard exercise in both workshops, time was allocated for YAP members to select a 'virtual sticky note' upon which to type their thoughts about the results, and 'stick' on the whiteboard under the relevant heading. The YAP were sent copies of the whiteboards beforehand, in case they wanted to prepare their thoughts in advance.

During the focus group discussion which followed the whiteboard exercise, members of the YAP were asked for any additional interpretations and retrospective views on how the results may or may not have resonated with their own experiences. A topic guide was used to navigate the conversation. During and after the workshops, the BRIGHTLIGHT team monitored for any distress and followed up with a mental health check-in. Participants were remunerated for their involvement, as per the National Institute of Health and Care Research PPI guidelines [74].

The workshops were held on Zoom under a secure subscription account and were digitally recorded. The YAP selected Zoom as their preferred platform due to the user-friendly whiteboard function. Transcripts were produced using Otter AI software (version number 3.52.240617). The content of the whiteboards and workshop transcripts were reviewed independently by three members of the research team, highlighting key themes through the categorization of responses. The themes were then reviewed and summarized by the research team during a group discussion. A summary report on the workshop was prepared and given to the YAP.

3. Results

Demographic, clinical, and psychosocial characteristics will be presented first. This will be followed by the quantitative findings, each of which will then be contextualized with interpretations from the YAP workshops.

3.1. Demographic, Clinical, and Psychosocial Characteristics

Eight hundred and thirty AYAs completed the survey at the first time point. Sociodemographic characteristics are shown in Table 1. The mean age was 20.1 (SD 3.3), 45% were female, the majority were white and 73% identified as single/divorced. Most were working or in school (64%) and socioeconomic status was distributed approximately evenly across five indices of multiple deprivations. Approximately 50% of the cohort-self identified as being on treatment.

The clinical characteristics of young people are outlined in Table 2. The most common cancer types were lymphoma, germ cell tumours, and leukemia, and most had a prognosis >80%.

Table 1. Sociodemographic characteristics of the BRIGHTLIGHT cohort at the first time point (6 months after diagnosis).

Characteristic	Frequency	% ^{a}
Age at diagnosis (years)		
Mean (SD)	20.1 (3.3)	
Gender		
Male	457	55.1
Female	373	44.9
Ethnicity		
White	730	88.0
Mixed	14	1.7
Asian	61	7.3
Black	15	1.8
Chinese	4	0.5
Other	6	0.7
Marital status		
Married/civil partnership	26	3.1
Cohabiting	93	11.2
Single/divorced	606	73.0
Missing data	105	12.7
Geographic location		
North East England	44	5.3
NW	106	12.8
Yorkshire	100	12.0
East Midlands	107	12.9
West Midlands	120	14.5
London	165	19.9
South East England	98	11.8
Sound West England	90	10.8
Socioeconomic status (IMD quintile)		
1—most deprived	184	22.2
2	136	16.4
3	156	18.8
4	182	21.9
5—least deprived	152	18.3
Missing data	20	2.4
Employment status		
Working full/part time	257	31.0
Education	274	33.0
Other work (apprentice, intern, volunteer)	17	2.0
Unemployed	31	3.7
Long term sick leave	126	15.2
Not seeking work	125	15.1

Note. SD, standard deviation; IMD, Index of multiple deprivation. ^{a} Column percentages displayed.

In terms of psychosocial characteristics (Table 3), mean distress scores were below borderline for anxiety and depression, indicating low levels of distress on average at 6 months after diagnosis. Total social support scores were also low, with a median of 1.5 (IQR 1.1–2.2).

Very few young people (n = 7, 0.8%) identified as having a pre-existing mental health condition. Most (n = 628, 74.8%) were offered support from a psychologist, counsellor, psychotherapist, social worker, youth worker, or activity coordinator, and 534 (64.3%) met at least one of these professionals for at least one session.

Table 2. Clinical characteristics of the BRIGHTLIGHT cohort at 6 months after diagnosis.

Characteristic	Frequency	% [a]
Cancer type—Birch classification [75]		
Leukemia	106	12.8
Lymphoma	267	32.2
Central nervous system	33	4.0
Bone	70	8.4
Sarcoma	59	7.1
Germ cell	154	18.6
Skin	31	3.7
Carcinoma (not skin)	100	12.0
Other [b]	10	1.2
Time to diagnosis (days) [c]		
Mean (SD)	125.3 (172.6)	
Median (IQR)	62.0 (29.0, 153.0)	
Disease severity at diagnosis		
Least severe	461	55.5
Intermediate	194	23.4
Most severe	175	21.1
Prognostic score		
<50%	127	15.3
50–80%	239	28.8
>80%	460	55.4
Missing data	4	0.5
Treatment type [d]		
SACT only	271	32.6
RT only	14	1.7
Surgery only	117	14.1
SACT and RT	106	12.8
SACT and Surgery	181	21.8
SACT, RT, and Surgery	77	9.3
RT and Surgery	36	4.3
Transplant	19	2.3
Other [e]	9	1.1

Note. SACT, systemic anticancer therapy; RT, radiation therapy; [a] Column percentages displayed. [b] Other diagnoses included Birch grouping categories 9–12 (see Birch et al., 2008 [75] for more detail). [c] From first symptom to diagnosis. [d] In the first 12 months since diagnosis. [e] Other was recorded if patients were in a watch and wait category at diagnosis and then received SACT, RT, and/or surgery. IQR, interquartile range.

Table 3. Psychosocial characteristics of the BRIGHTLIGHT cohort 6 months after diagnosis.

Characteristic	Value
HADS [a]	
Anxiety score, mean (SD)	6.89 (4.39)
Borderline, n (%)	160 (19)
Moderate/severe, n (%)	172 (21)
Depression score, mean (SD)	4.62 (3.68)
Borderline, n (%)	120 (15)
Moderate/severe, n (%)	55 (7)
MSPSS, median (IQR)	
Support—friends	1.75 (1–2.75)
Support—family	1.25 (1–2)
Support—significant others	1 (1–2)
Total support	1.5 (1.1–2.2)

Note. HADS, Hospital Anxiety and Depression Scale; MSPSS, Multi-dimensional Scale of Perceived Social Support. [a] Borderline = 8–10, moderate/severe > 11.

3.2. Quantitative Findings

There was a significant relationship between social support and anxiety levels ($\beta = 0.202$, $p < 0.001$), with higher total social support scores associated with higher anxiety scores (Table 4). The relationship was significant for depression in the same direction ($\beta = 0.227$, $p < 0.001$), with higher social support scores also associated with higher depression scores (Table 4). Examining the domain scores of social support revealed the same relationships.

Table 4. Associations between distress symptoms (anxiety and depression) and factors known to impact mental health at 6 months after diagnosis [a].

Distress Symptom	Test Statistic	*p*-Value	95% CI
Anxiety			
Age	$\beta = 1.022$	0.002	0.008 to 0.035
Gender	$\beta = 0.696$	<0.001	−0.420 to −0.305
Social support			
From family	$\beta = 1.028$	<0.001	1.696 to 1.799
From friends	$\beta = 1.030$	<0.001	1.631 to 1.732
From significant others	$\beta = 1.009$	0.003	1.818 to 1.915
Total	$\beta = 1.148$	<0.001	0.108 to 0.168
Disease severity			
Between groups	F(2827) = 3.351	0.036	0.000 to 0.023
Low vs. Medium		1.000	−0.774 to 1.024
Low vs. High		0.032	0.0610 to 1.927
Medium vs. High		0.172	−0.226 to 1.964
Pre-existing mental health condition	U = 385.500	0.162	−1.241 to 0.276
Offered contact with a mental health professional	t(827) = −1.617	0.106	−0.287 to 0.028
Had contact with a mental health professional	t(774) = −1.656	0.098	−0.280 to 0.024
Depression			
Age	$\beta = 1.001$	0.186	−0.005 to 0.027
Gender	$\beta = 0.729$	<0.001	−0.388 to −0.246
Social support			
From family	$\beta = 1.033$	<0.001	1.254 to 1.378
From friends	$\beta = 1.041$	<0.001	1.119 to 1.242
From significant others	$\beta = 1.015$	<0.001	1.367 to 1.485
Total	$\beta = 1.192$	<0.001	0.140 to 0.212
Disease severity			
Between groups	F(2827) = 3.999	0.019	0.000 to 0.025
Low vs. Medium		0.020	−1.610 to −0.100
Low vs. High		0.390	−1.270 to 0.290
Medium vs. High		1.000	−0.560 to 1.280
Pre-existing mental health condition	U = 431.000	0.301	−1.167 to 0.349
Offered contact with a mental health professional	t(827) = −3.672	<0.001	−0.452 to −0.136
Had contact with a mental health professional	t(774) = 3.840	<0.001	−0.450 to −0.145

Note. CI, confidence interval. [a] Univariate analyses.

When adjusting for confounders, the female gender was associated with significantly higher anxiety ($\beta = 0.696$, $p < 0.001$) and depression ($\beta = 0.729$, $p < 0.001$), while older age was associated with significantly higher anxiety ($\beta = 1.022$, $p = 0.002$) but not depression ($\beta = 1.001$, $p = 0.186$) (Table 4). Ethnicity, marital status, socioeconomic status, and employment status were not associated with higher distress. Other demographic and clinical covariates are found in Table 4.

There was a significant difference between disease severity groups in terms of anxiety (F(2827) = 3.351, $p = 0.036$) (Table 4). Higher disease severity was associated with lower anxiety, between low and high disease severity groups only ($p = 0.032$). For depression, there was a significant difference between groups (F(2827) = 3.999, $p = 0.019$). Lower disease severity was associated with lower depression, between the low and medium-severity groups only ($p = 0.020$).

Mean anxiety and depression scores were both higher in AYAs with a pre-existing mental health condition, but differences were not statistically significant.

AYAs with higher depressive symptoms were more likely to be offered (t(827) = −3.672, $p < 0.001$) and have contact (t(774)= 3.840, $p < 0.001$) with a mental health professional, as shown in Figure 1. There was no association between anxiety levels and being offered (t(827) = −1.617, $p = 0.106$) or having contact with (t(774) = −1.656, $p = 0.098$) a mental health professional.

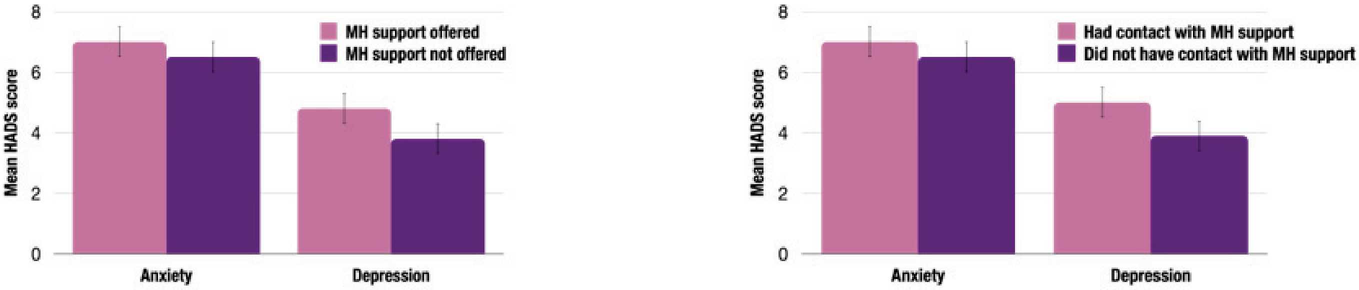


Figure 1. Relationship between distress scores and AYAs who were offered mental health support (**left**) and had contact with mental health support (**right**) at 6 months post-diagnosis.

Across the five time points, anxiety did not change significantly (F(4109) = 2.069, $p = 0.090$) (Figure 2). However, depressive symptoms improved when comparing the first time point (6 months after diagnosis) to the subsequent four time points (12 to 36 months after diagnosis) (F(4109) = 10.460, $p < 0.001$). Of note, sample sizes across the time points were as follows: 6 months, n = 830; 12 months, n = 566; 18 months, n = 460; 24 months, n = 394; 36 months, n = 336.

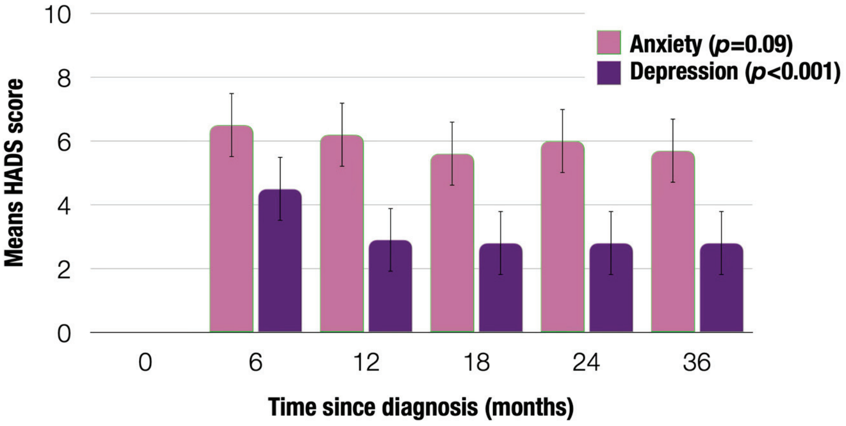
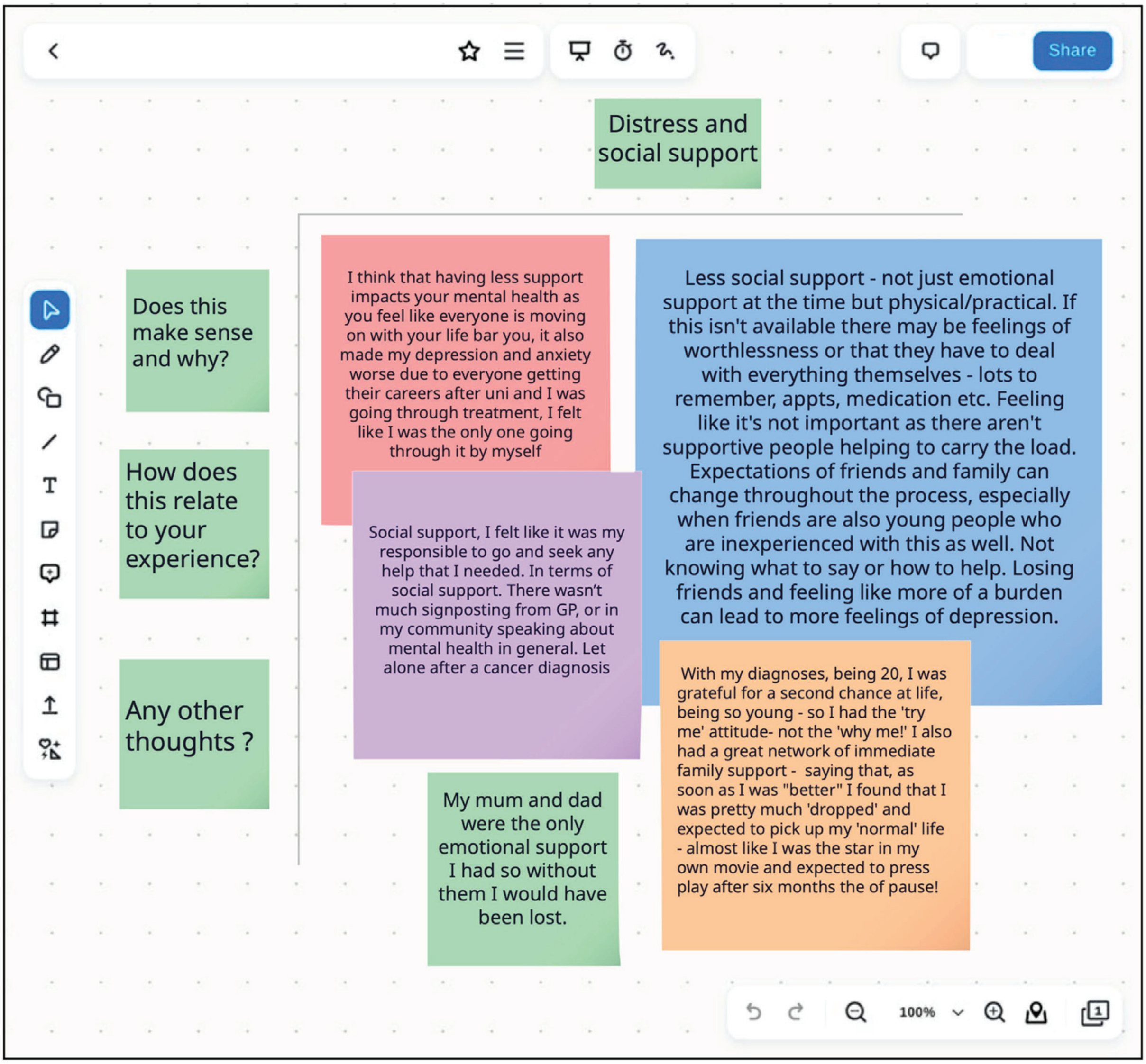


Figure 2. Change in distress across the five time points since diagnosis.

3.3. Findings from the YAP Workshops

To help contextualize and interpret the study results, feedback was garnered through two participatory workshops with the BRIGHTLIGHT YAP in June 2024. Ten members (eight female and two male) were invited to the first online workshop, nine confirmed attendance and seven participated on the day. The second workshop was restricted to those who had attended the first, and all seven were present. Participants were all female, aged 14–20 at diagnosis, and had typical AYA cancers including sarcoma, lymphoma, and brain tumours. The group comprised a mix of patients who had recently finished treatment, those who had been off treatment for some time, and those now being treated for late complications of therapy.

Overall, participants felt the topic of mental health in young people with cancer was important, not well acknowledged by their friends and family, and underexplored by their healthcare team during the first few years following diagnosis. Key study findings were explored in more detail with the YAP, including the above results from the primary and secondary research questions. Their interpretations are detailed below along with exemplar quotes. An example of a whiteboard with the variable 'Social Support' is included below (Figure 3).



Distress and social support

Does this make sense and why?

How does this relate to your experience?

Any other thoughts ?

I think that having less support impacts your mental health as you feel like everyone is moving on with your life bar you, it also made my depression and anxiety worse due to everyone getting their careers after uni and I was going through treatment, I felt like I was the only one going through it by myself

Social support, I felt like it was my responsible to go and seek any help that I needed. In terms of social support. There wasn't much signposting from GP, or in my community speaking about mental health in general. Let alone after a cancer diagnosis

Less social support - not just emotional support at the time but physical/practical. If this isn't available there may be feelings of worthlessness or that they have to deal with everything themselves - lots to remember, appts, medication etc. Feeling like it's not important as there aren't supportive people helping to carry the load. Expectations of friends and family can change throughout the process, especially when friends are also young people who are inexperienced with this as well. Not knowing what to say or how to help. Losing friends and feeling like more of a burden can lead to more feelings of depression.

With my diagnoses, being 20, I was grateful for a second chance at life, being so young - so I had the 'try me' attitude- not the 'why me!' I also had a great network of immediate family support - saying that, as soon as I was "better" I found that I was pretty much 'dropped' and expected to pick up my 'normal' life - almost like I was the star in my own movie and expected to press play after six months the of pause!

My mum and dad were the only emotional support I had so without them I would have been lost.

Figure 3. YAP whiteboard interpretation of the relationship between distress and social support based on their own experiences.

In general, the YAP thought that higher levels of social support would be protective against distress, in contrast with the study findings. However, the discussion on this topic was extensive, and they identified both positive and negative impacts of social support on distress, summarized in Table 5.

Table 5. Impacts of social support from family and friends on mental health.

Source of Support	Positive Impacts	Negative Impacts
Family	● 'Managing the load': Practical support with scheduling and taking to appointments, taking medication	● 'Protecting family', having to manage family emotions and being the strong one ● Having support early on from new family members appearing and then 'dropped' when 'better' ● Lack of available support for family members experiencing distress ● Feeling overwhelmed by 'too many people around', feeling pressure to see them when not feeling well enough ● Loss of identity if forced temporarily to give up a caregiving role
Friends	● Coming to visit when in hospital or at home, when too sick to go out	● Seeing friends move on with their lives but feeling held back due to impact of treatment on activities ● Losing contact with friends ● Feeling like a burden on friends ● Expectations from friends to 'be like before', hard for friends to understand post-treatment symptoms like fatigue, pain ● Friends may not want to discuss difficult feelings or think about 'worst case scenarios which you as the patient may be having to face' ● Having lots of friends around but not wanting to acknowledge what's happening could 'feel more isolating than not having them at all'

An experience highlighted by multiple participants was a strong sense of duty to protect close family and friends from further emotional pain, by keeping their own feelings private. While most participants recognized they had several individuals they could rely on for emotional support, this recognition did not necessarily translate into them actively seeking out and sharing their feelings with those individuals:

> "I'm actually really upset about this, [but] I had to be strong for everybody else. And I think that's something that kind of gets missed a lot because you have a family member there. So my mom sat there crying, but she would cry the whole way home, and I had to make sure she was okay. So without, you know, going into too much negativity, I had to sort of be the strong one. And I think that definitely weighed on my mental health".

Another common observation was that family and friends often anticipated an immediate return to normalcy following the end of treatment, when the young person was only beginning to process the emotions that come with cancer. One participant reflected that,

> "...as soon as I was better I found that I was pretty much dropped and expected to pick up my normal, almost like I was the star in my own movie and expected to press play after 6 months of pause."

The YAP thought the finding that female gender was associated with significantly higher anxiety and depression was not surprising. They reported that women may be more likely to talk about emotions than men, and that it may be more conventional for women to report distress and seek help:

> "...I think it's more socially acceptable to have to need to seek help for mental health and are, like, if I was having a bad day, I'd be a lot more vocal about it

> than my partner would be. And so I think it's probably, personally, I think men try not to talk about things like that. . ."

They also wondered about emotional maturation occurring earlier in females, and about the differential impact of cancer treatment on female reproductive hormones, thereby influencing rates of distress.

The association of disease severity significantly predicting distress resonated with the YAP. Several reported a lack of awareness soon after diagnosis about the severity of their illness, for a variety of reasons. In some cases, information was communicated directly to parents or caregivers from the healthcare team, bypassing the young person. In others, incomplete or surface-level details may have been given to the young people:

> ". . .it wasn't till a few years later that I actually realized the severity [of the diagnosis]."

One participant wondered whether their young age and a retained sense of invincibility, at least initially, may have been protective for their mental health:

> "I was very naïve and unaware of the severity of my cancer. I think this worked in my favour with my mental health."

Others reported that the severity did not matter after a diagnosis was finally confirmed. The relief of having an explanation for their symptoms provided welcome validation. One participant thought that the focus on the physical aspects of treatment and recovery could distract from or protect against the mentally distressing aspects, while another felt conversely that physical limitations such as not being able to dance significantly worsened their mental health. It was reflected that each person's situation and associated emotions were their own.

The YAP had several insights about the finding that there was no difference in distress between people with and without a pre-existing mental health condition. They thought that AYAs with a mental health diagnosis before cancer may already relate to appropriate support, which could position them to have better healthcare system navigation skills, feel more prepared to speak up about physical symptoms and be equipped with tools to cope with distress. Most thought that during the time of recruitment (2012–2014), there may have been under-reporting or under-detecting of mental health conditions, due to higher levels of stigma and barriers to accessing mental healthcare:

> "I think there will be a lot of young people who just haven't received a diagnosis yet. So then down the line, it may appear that if they are struggling mentally, it's been, like, directly because of their cancer and what they've been through. . . But it could be that they may be already suffering with anxiety. But it was just kind of put down to being a teenager and, like, normal worries."

Some participants wondered whether people might have already been imagining the 'worst case scenario', and thus were not taken aback when the diagnosis eventually came:

> "If you're a person who tends to worry a lot, when the thing you're worrying about actually happens, you suddenly become very calm. Because that disaster I've been waiting for is here."

The YAP shared their own experiences of distress over time from diagnosis (responses summarized in Table 6). Most reported higher overall levels of anxiety than depression, in line with the study results. Many reported a decrease in overall distress across the time points, corroborating findings that depression was significantly lower after the 1-year mark.

Table 6. Perspectives of the YAP on patterns of distress over time from diagnosis.

Time	Anxiety (Things I Was Worrying About)	Depression (Things I Was Sad About)
6 months	• Losing friends, becoming disconnected • Treatment (surgery) and its implications • Side effects of chemotherapy: how bad each cycle would feel physically • Fear of recurrence • Returning to school/work	• Feeling left behind socially, academically/workwise • Coping with permanent physical changes • Grieving lost identities, friendships
12 months	• Feeling "normal" again • Adapting to new limitations • Returning to school or work • Needing more treatment, associated uncertainty • Scans	• Coming to terms with new limitations • Difficulties with ongoing treatment, thinking about stopping
18 months	• Needing school/work accommodations • "Normal teenage worries" (e.g., sitting GCSEs) • 'Survivor guilt'	• Struggling with being well, but • 'nowhere near what it was before'
2 years	• Adjusting back at school/work • Fear of recurrence • Starting a 'new life' • Survivor guilt • Late effects starting • Adapting to new limitations	• No depression symptoms
3 years	• Worries waning, 'I can still live a good life' • Late effects progressing • Difficulties securing work due to lack of accrued experience	• No depression symptoms

Note. GCSEs, General Certificate of Secondary Education.

Several participants wished they had been asked about their mental health as they progressed through treatment and in the months and years afterward but they were not given the opportunity. The YAP noted that while there were many common emotional reactions over time, each person's experience and feelings were personal and unique.

4. Discussion

This study contributes novel insights into the mental health of young people with cancer by providing a deeper understanding of risk and protective factors related to psychological distress in a large, longitudinal sample of AYAs. Six months after diagnosis, young people in our cohort reported anxiety and depression rates (borderline plus moderate/severe) similar to other studies of the AYA population, with anxiety ranging from 8 to 55% and depression from 13 to 47% [31,38–40]. Anxiety levels decreased with age, and females reported higher rates of distress, anxiety, and depression, aligning with results from several studies [46–48]. The YAP agreed that females might find it more socially acceptable to express emotions and seek help.

The first aim of this study was to investigate the relationships between distress and social support, disease severity, pre-existing mental health conditions, and contact with mental health professionals during treatment. Contrary to the proposed hypotheses, increased distress was associated with higher levels of social support and less severe illness. Existing research suggests the presence of close family and friends generally mitigates distress [76–78]. The YAP noted both positive and negative impacts of social support, including the benefits of practical help versus the burden of suppressing their feelings to protect loved ones, which are concepts supported by the literature [76,79]. It is also possible that current quantitative measures of social support do not capture the complexity and nuance involved in social interactions young people experience after a cancer diagnosis. A final possibility is that distress itself may have evoked greater social support.

Another unexpected finding was that lower disease severity was associated with higher anxiety scores at the first time point, which persisted after controlling for covariates. There is a dearth of research on this topic in AYAs [80], but the YAP suggested this could be due to an initial lack of awareness about the severity or the invincibility of youth. Another possibility is that young people did not allow the severity to define them, i.e., other illness-independent factors were driving their emotional response [81,82].

Additionally, there was no significant relationship between the presence of a pre-existing mental health condition, a screen-detectable target for early psychological intervention, and distress, likely due to small numbers of AYAs disclosing a pre-existing condition. Response bias should also be noted, as these were self-reported responses to an interviewer-administered questionnaire. Although little evidence exists on this issue, the YAP posited that AYAs with mental health diagnoses before cancer might benefit from existing healthcare connections and coping tools, helping manage distress. They also commented that higher levels of stigma and barriers to accessing mental healthcare during recruitment more than 10 years ago may have led to under-identifying these conditions.

Finally, anxiety levels did not correlate with being offered or having contact with support, but AYAs with higher depression scores were more likely to be referred to and connected with a mental health professional. In this study, distress scores were not reported back to the treating clinicians, indicating the possibility that either clinicians were good at detecting distress in other ways, or patients (and/or caregivers) were good at advocating for their mental health needs, or both. In a large cohort study comparing rates of mental health service utilization between 639 AYAs with cancer and 29,793 without, those with cancer were more likely to undergo psychotherapy [83]. A qualitative study in AYAs with cancer highlighted that offering mental health services to all patients at multiple time points enhanced engagement with and utilization of those services [84].

The second aim of the study was to explore the change in distress along the cancer trajectory, predicting a decrease. This was borne out for depression, which decreased significantly after the first time point, but not for anxiety, which remained stable. Due to a lack of longitudinally designed research on distress in AYAs with cancer, changes in mental health symptoms over time are poorly understood. There are a small number of long-term follow-up studies, each varying in duration, research design, measures of distress, AYAs age range, and cancer type [44,49,58,83]. Evidence from these studies indicates mixed outcomes, with rates increasing, decreasing, or remaining stable over time.

The YAP described highly personalized distress trajectories depending on life stage, specific diagnosis and treatment, and amount of educational or vocational support. Anxiety was the more dominant emotional state over depression, with both declining over time. Nonetheless, the YAP emphasized the need for emotional support beyond the end of the study period, because, despite a general decrease, symptoms did not fully dissipate.

This study encountered common cohort study limitations, such as selection bias, differential loss to follow-up, and missing data [85,86]. Treatment status (on/off treatment) was also not analyzed, and the sample's predominantly white ethnicity from England narrows the generalizability of the findings. Moreover, given that the cohort's data are over a decade old, it represents a historical perspective of medical and mental healthcare, which has undoubtedly evolved [87]. The analyses in this study were confined to the outcome measures captured in the survey, precluding the examination of additional mental health variables and confounders, such as specific diagnoses, or more granular surveys on depression, anxiety, grief, post-traumatic stress, and suicidal ideation. Moreover, the database lacked insights into distress, including narrative accounts of symptomatology at various time points or the effects of mental health interventions. Although few measures have been psychometrically tested within this demographic, often being validated in populations of younger children or older adults, the HADS and MSPSS have been consistently utilized in AYAs cancer research, affirming their effectiveness in assessing the intended constructs [68,88,89].

While this study benefits from a longitudinal design, certain analyses were conducted cross-sectionally (e.g., at 6 months after diagnosis) to provide a detailed understanding of distress and its correlates early in the cancer trajectory. Cross-sectional analyses are particularly valuable in early survivorship, where timely identification of psychosocial risk factors can inform real-time clinical decision-making and targeted interventions. By capturing distress levels at distinct time points, this approach allows for immediate assessment of mental health needs, ensuring that support can be mobilized when most needed.

However, because these analyses were conducted early in the disease course, the positive association between distress and social support may reflect the process of seeking and utilizing support in response to distress, rather than social support acting as a prospective protective factor. Examining these relationships prospectively within the available longitudinal dataset would provide a clearer picture of how social support influences distress over time. Additionally, while cross-sectional analyses allow for efficient subgroup comparisons, they do not capture individual trajectories of distress or establish causality. As distress in AYAs is dynamic, future research could incorporate time-dependent models, such as linear mixed-effects models or growth curve analysis, to better understand long-term psychosocial trends.

Finally, although the YAP's input was vital to the research, their involvement had certain constraints. Notably, due to the absence of male participants and being self-selected and well-versed in cancer research and PPI, the group might not necessarily reflect the broader views of all AYAs [62].

Despite these limitations, the large, longitudinal dataset used in this study allowed for a broad exploration of the mental health characteristics of young people and how distress changed over time. As described in more detail by Taylor and colleagues [90], engaging young people in research from the very beginning and implementing a centralized system for tracking patients seem to be crucial strategies for enhancing recruitment and retention.

Furthermore, the YAP workshops provided crucial interpretations of study findings, filling existing literature gaps with insightful and complex hypotheses. The YAP validated the importance of the topic and the need for continued research in this area. Contrary to prevailing descriptions of this population as vulnerable [91,92], which often leads to their exclusion from research participation [62], the YAP freely shared their feedback, particularly eager that their input would be used to inform future interventions.

Future research on the topic of mental health in AYAs with cancer should include longer follow-up periods and larger sample sizes. Sensitivity analyses examining the differences between distress levels in those who participated across all time points versus those lost to follow-up would enhance the validity of the results. Additional prospective analysis of the longitudinal data could help offer a clearer picture of the impacts of variables such as social support on distress over time. With respect to social support in particular, the surprising finding that higher levels were not protective against distress is worthy of further study. A clearer understanding of that relationship could help minimize harm and harness the benefits of social support for AYAs with cancer.

Prospective qualitative exploration of distress along the disease trajectory should be undertaken, to understand patterns of distress over time and experiences of accessing professional mental healthcare during or after treatment. Synthesizing quantitative and qualitative data would provide a more detailed picture of the psychological burden in this population, helping inform the design and evaluation of individualized screening and intervention tools. While reported rates of pre-existing mental health conditions in this study were low, between suspected underreporting at the time of the study and increasing rates of mental health conditions over time since the study was conducted, current rates are likely to be much higher. Newer studies examining these rates using multiple methods could help strengthen the recommendation that screening should be routinely offered to facilitate early detection and provide proactive interventions.

Clinical care improvements could include facilitating the early detection and treatment of mental health concerns shortly after diagnosis, addressing modifiable risk factors and bolstering protective factors. Findings also underscore the importance of sustained psychosocial support during and after treatment, and enlightening healthcare professionals about developmental explanations for distress in AYAs at different stages of their cancer experience [48]. An open dialogue about mental health in hospitals can not only facilitate better referral pathways but also reveal service shortfalls, thereby informing advocacy initiatives aimed at filling these gaps.

In a recent priority setting exercise, AYAs themselves identified the top AYAs research priority as finding a “psychological support package that improves psychological well-being, social functioning and mental health during and after treatment” [93]. Young people should continue to be included as active co-designers in all elements of research, as they have been in the BRIGHTLIGHT study. Given appropriate scaffolding from researchers, young people can contribute immensely to healthcare research. The advantages to AYAs, the research team, and the broader AYA community far outweigh any obstacles [62].

5. Conclusions

Several key conclusions may be drawn from this study. First, while this large cohort has similar rates of overall distress to other AYA studies, high levels of social support and low disease severity were identified unexpectedly as potential risk factors for distress. Identifying the presence of a support network alone may not capture the distress young people feel about their cancer’s impacts on that network and thus may not necessarily equate to active confiding or soliciting support. Guidance on improving emotional communication can help young people and their support networks navigate the feelings associated with a cancer diagnosis. Furthermore, severe disease does not necessarily register with a young person as emotionally detrimental. These points reinforce the importance of avoiding assumptions about factors affecting a young person’s emotional state. Second, pre-existing mental health conditions are important targets for screening initiatives, particularly due to increasing rates of mental health conditions in young people. Third, while professional mental health support is being offered to AYAs with cancer, rates of referral could be higher. Fourth, young people need emotional support long after their diagnosis, given the persistent nature of symptoms over time. Finally, participation and engagement from young people themselves in research and clinical programming is vital and rewarding for all. Insights from this study will help shape the creation of developmentally tailored screening approaches and intervention strategies, to alleviate distress and improve the overall well-being of this unique and understudied population.

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cancers

Article

"There Are Two Healing Processes in Cancer Care—There Is a Physical Healing and a Mental Adaptation Process": A Pilot Study for Preparing Children and Adolescents with Osteosarcoma for Limb Amputation

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Simple Summary: Pediatric and adolescent patients with cancer who require limb amputation face significant physical and emotional challenges. While medical care has improved survival outcomes, far less is known about how to prepare young patients for the psychosocial impact of amputation and how to support their adjustment afterward. This study draws on the lived experiences of cancer survivors who underwent amputation during childhood or adolescence to identify key elements of informational, emotional, and social support needs. By listening to firsthand accounts, the study aimed to inform the development of more comprehensive, patient-centered care strategies. The findings highlight missed opportunities for improving mental health access, the benefit of tailoring information delivery to individual needs, and integrating peer and family support into clinical practice. These insights can guide clinicians, psychosocial professionals, and healthcare systems in enhancing the quality of care for future patients undergoing similar procedures.

Abstract: Background/Objectives: This study assessed how to best prepare pediatric and adolescent cancer patients for amputation and support them afterward. Methods: This pilot qualitative study explored pre- and post-amputation experiences from the perspectives of nine pediatric and adolescent survivors who underwent amputation. Hour-long audio-recorded semi-structured interviews were transcribed and analyzed using the Sort and Sift, Think and Shift qualitative approach. Results: Participants described the informational supports they received before surgery, including guidance on what to expect, contact with amputation-related organizations, and exposure to tangible tools, such as a physical model of a knee joint. Emotional support from fellow amputees and healthcare providers, particularly surgeons, was also found to be meaningful. Individuals also identified unmet needs and gaps in emotional care. These included clearer guidance on post-surgical adaptations (e.g., basic self-care and navigating physical limitations) and the need for information tailored to their learning styles. Many emphasized the importance of improved pain management resources, expanded access to mental health services for both them and their families, and support in adjusting to changes in body image and social relationships. Participants also shared advice for future patients, recommending strategies such as personalizing hospital rooms, connecting with other amputees through social media, and using art to process their experience and say goodbye to the lost limb. Conclusions: Interviews with nine cancer survivors provide guidance for improving holistic, patient-centered care throughout the amputation process. Informational and emotional support should be tailored to an individual's learning style and specific needs, in addition to their age at the time of surgery.

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1. Introduction

In the U.S., approximately 15,780 children aged 0–19 are diagnosed with cancer annually [1], with about 1 in 285 children receiving a diagnosis before their 20th birthday. Bone sarcomas, primarily osteosarcoma and Ewing sarcoma, represent 3% of pediatric cancers (roughly 400 cases/year in the U.S.) [1]. Although less common than other types, these cancers often require limb amputation in 9–57% of cases [2], along with radiation and chemotherapy, placing patients at a heightened risk for poor psychosocial outcomes [3]. Osteosarcoma is the most common malignancy of bone in children and young adolescents, most often occurring in the lower extremities, and accounts for approximately 60% of all malignant bone tumors diagnosed before young adulthood [4].

Research highlights the importance of pre-operative interventions in promoting coping and adjustment and reducing post-operative distress [5]. However, Weschenfelder et al. [6] identified a critical gap: no psychological interventions are tailored specifically for youth facing amputation, despite the potential traumatic nature of the surgery and aftermath [7]. While existing studies focus on post-operative quality of life, there is limited guidance on pre-operative support [6]. For localized bone sarcomas, the five-year survival rate is 76% [1], underscoring the need for effective psychosocial interventions to support long-term well-being.

This study aimed to identify essential components of pre- and post-operative support for pediatric and adolescent patients undergoing amputation, drawing on insights from individuals with lived experience. Qualitative methods are particularly well-suited for exploring understudied populations [8], as they provide rich descriptions and amplify the voices of individuals often excluded from research [9,10].

2. Materials and Methods

2.1. Participants

Nine English-speaking adolescents and young adults who underwent amputation due to cancer in their childhood or teen years participated in semi-structured, audio-recorded phone interviews. Interviews lasted approximately 60 min. Participants were recruited at the NIH Clinical Center, Dana-Farber Cancer Institute, Sydney Children's Hospital, and Make it Better Agents. The study was open at three sites. Each site identified eligible participants (having undergone an amputation during their childhood or teen years). Due to the high relapse rate, each site had very few participants. There were no participants reported to us who, after being approached, declined to participate. Each site documented the patient's participation and interview within their medical record or required systems.

2.2. Procedure

Clinicians trained in qualitative interviews conducted the interviews. Audio recordings were professionally transcribed. The NIH Office of Human Subjects Research determined the study to be exempt from full IRB review (OHSRP #5736). Participants did not receive any incentives or compensation for participation. Oral consent was obtained before recording each interview. See Supplemental S1 for oral consent procedures.

The interview was developed by psychosocial experts in the field of pediatric oncology. Interview questions (Supplemental Table S1) included how the surgery was explained to them, how they were emotionally and physically supported before and after amputation, and what advice they would give to children facing amputation and their parents. Sample questions included the following:

- "From what you know now, what do you feel would have been helpful for you to have been told before the time of amputation?"
- "Was there anything that the care team did or said that you remember helped you feel more prepared or comfortable? Please explain."
- "Did you feel you were prepared emotionally for the amputation? After the amputation? Please explain."

2.3. Analysis

Transcribed interviews were analyzed using the Sort and Sift, Think and Shift method, an iterative approach that encourages researchers to immerse themselves in the data to uncover its key dimensions and meanings, followed by deliberate reflection to interpret findings in relation to broader scholarly conversations [11]. In the initial phase of analysis, authors independently and then collaboratively read the transcripts in full to gain a deep understanding of the material and to identify emerging patterns related to support received and desired before and after the survey [12]. Summaries of each interview were created and organized into matrix form to facilitate cross-case comparison and identify trends and variations in participants' experiences [13]. A preliminary codebook was developed based on the range and nuance of themes within each analytic domain. All transcripts were uploaded into Dedoose (Version 9.0.107, 2025) [14], and coding was carried out collaboratively through a consensus-driven process.

3. Results

3.1. Subsection

Nine pediatric and adolescent cancer patients with limb amputation completed in-depth interviews. Mean age at amputation was 15.2 years, and at the time of the interview, it was 25.1 years. Mean time between amputation and interview was 10.0 years. One participant identified as Asian, and the rest identified as White. Please see Table 1 for additional demographic information.

Table 1. Participant demographic information.

Participant #	Sex	Age at Surgery	Age at Interview	Type of Cancer	Type of Surgery
1	M	9	25	osteosarcoma	amputation
2	M	10	36	osteosarcoma	amputation
3	F	19	27	osteosarcoma	amputation
4	M	18	24	osteosarcoma	amputation
5	F	12	33	osteosarcoma	amputation
6	F	18	24	osteosarcoma	amputation
7	F	18	20	osteosarcoma	amputation
8	F	19	21	osteosarcoma	amputation
9	M	14	16	osteosarcoma	rotationplasty

3.1.1. Resources Offered: Informational and Emotional Supports in Place

Participants expressed gratitude for the informational and emotional support they received throughout the amputation process. Resources varied, with some receiving "what to expect" information, contacts from amputee organizations, and the opportunity to see a physical knee joint. One participant underscored the importance of timely communication, saying, “I definitely think that once the word is out, definitely someone should talk to the child. There are going to be questions; they are going to be filled with questions.”

Emotional support was offered through peer connections and medical care providers. Meeting other amputees was critical in preparing for treatment. “The idea of being able to connect with other amputees is so tremendously important. The internet affords you the ability to transcend physical separation and connect with other amputees.” One participant remarked that it was important that she spoke with an amputee who was also a woman, as she had specific questions about attire and navigating romantic relationships. Surgeons were seen as particularly helpful, as indicated by the following quote. “She was really good. I remember her sitting and like I was talking to her once. I mean all the doctors were. They really, really drummed into me the message that this would not stop you from doing anything you want to do.” See Table 2 for additional definitions and quotes.

Table 2. Resources offered: informational and emotional supports in place.

Informational Resources		
Theme	**Definition**	**Quotes**
What to expect	Participants appreciated information about how their bodies would look and feel after amputation.	“Yeah, I mean they showed me exactly where the tumor was—the bone growth—the cancer was. And then they showed me exactly where they needed to take—where they needed to amputate—how far up they would need to amputate my leg. And then, um, they also showed me what it would look like and what it would feel like and what my—what the post-surgery would be like if I had gotten it reconstructed.” “. . .with Dr. E [the surgeon] did give me pamphlets of what the stump would look like and how to clean it and all that.” “They told me about the drainage tube during surgery.”
Supportive organizations	Adolescents often mentioned organizations that offered emotional and physical support.	“I think having contact with foundations, like Amputee Coalition and MIB Agents. . .I think having a support network while you’re going through it and being able to have a place where you can ask questions and get opinions from the people who have already been through it is important.”
Physical materials	A few participants appreciated being shown a physical knee joint.	“Dr. M, she showed me, when she was explaining the limb salvage, she showed me the mechanical knee joint, the actual one that she was going to put inside me if I decide on that surgery.”

Table 2. *Cont.*

Emotional Resources		
Theme	**Definition**	**Quotes**
Connecting with others	This theme referred to the importance of connecting with other amputees and having role models with similar experiences for guidance and support.	"Being involved with Special Op is still . . . one of the biggest parts of my joy. It's great to see when you're there to see other people, and then you also have role models. R. was one of my role models. And to see him having life afterwards is a wonderful thing." "There's a guy named L., and he drives a white convertible, and he works for the government in DC and he's very handsome, in his late 20s. And just this picture of this adult guy with one leg who had a job and was very handsome and had a white convertible, just that concept was like amazingly powerful."
Support from medical providers	Participants appreciated the emotional support, particularly from their surgeons, during visits and hospital stays.	"I think just my surgeon was there for me. After my amputation, she gave me a gift and it just meant a lot because she was not just my surgeon, she was human and she saw me as a human, too." "My family and I had talked about amputation. One [children's hospital] was against the amputation, and I had a lot of pushbacks from them. So, I actually left and went to another hospital and had it done. But my decision to [move care] with the healthcare provider was not a good interaction. I walked out very angry and upset [with the first hospital]. But then, when I went to the other one, it was a totally different experience."

3.1.2. Resources Desired: Unmet Needs and Gaps in Emotional Support

Participants identified a range of needs that were not met, including strategies for self-care, adaptation to physical activity, and sensitivity to learning styles. Multiple interviewees mentioned the challenges of maintaining personal wellness after surgery, particularly bathing. "I think something that I didn't know was that walk-in showers and tubs are a lot different with one leg." Identifying strategies to continue participating in physical activities was an area of unmet need, as illustrated by this statement: "Then just learning how to walk again was hard. So, I think that time probably would have been the best to have a mentor. I didn't see her after I had an amputation. Only before."

A clear need for additional emotional resources before and after their amputation was emphasized. They specifically expressed the desire for more assistance in coping with pain and improved access to mental health providers for themselves and their families. Participants felt prepared for the surgical pain but not for phantom pains. For example, "I was really ready for it [the surgery] because I was in so much pain, but I did after have a mental breakdown because I was in so much pain. And the phantom pains were really bad for me." The need for additional emotional support was expressed by multiple individuals. Participants wished for mental health services before and after surgery. One person explained, “But also I thought that that could have been better with like mentors, and psychologists, and other support of like mentally and emotionally because it was a big decision.” The desire for emotional support extended to parents and siblings. “Through my treatment or through my amputation, I think it would have been nice for them to have someone to talk to or like a group about it. I feel like the families are kind of forgotten in the whole process.”

Multiple participants emphasized the importance of adapting information to the learning styles and comfort level of each patient. For example, “I personally did not even want to know. Some people, they look up what an amputation surgery looks like because they want to know. I didn’t because I knew it was going to be not pretty. I liked being explained it better than looking at it”.

Further, participants sought support in building social connections, given their new physical restrictions, and coping with self-image challenges. Maintaining friendships through shared experiences was important to the youth. However, they struggled to find ways to connect with peers due to their new physical limitations, as previous pastimes had included hiking or other physically strenuous activities. Issues of self-image also emerged, as noted in the quotes below:

- “People pay more attention to me now that I have the leg, and I don’t know if I like that.”
- “It was a bit difficult I suppose like when going out in public. Because we were from a small town, so everyone knew what was going on. But, yeah, everyone saw that thing.”

See Table 3 for additional definitions and quotes.

Table 3. Resources desired: unmet needs and gaps in emotional support.

Unmet Needs		
Theme	**Definition**	**Quotes**
Self-care	Basic personal wellness needs, specifically showering, were frequently mentioned as a desired resource.	“I mean we had to modify the bathroom, which we did beforehand. So that was one thing that they had suggested to mom and dad. But I don’t know what they really talked about or said.” “PT started having me learn how to do stairs in adaptive ways. I think there could be more physical therapy towards household ways of doing things. We didn’t talk about showering but maybe how to transfer from a chair into a shower.”
Physical activity	Participants expressed a desire to learn how to modify previously enjoyed physical activities.	“I’m a really active person and having to stay still and lay in bed all day for months at a time was just unbelievable for me.” “The dancing is a big thing to me, ...over sort of college, I started to get more confident, and now I go out dancing with my friends over the weekend.”
Learning style	Participants wished that providers adapted information to different learning styles of patients and families.	“I’m the kind of person who likes people telling me more than reading stuff. But my parents found it very helpful.” “I’m a visual learner. I watched videos after I spoke with the doctors.”

Table 3. *Cont.*

Desired Emotional Resources		
Theme	**Definition**	**Quotes**
Coping with pain	Participants expressed the need for more information about managing physical and phantom limb pain.	"I guess the nerve pain was really bad. I still deal with it to this day. I guess that could've been explained better just because it's such a weird sensation. I didn't expect to feel so terribly from that." "I think one of the biggest shocks was the phantom pain. I don't think that term gives it enough credit because it's not fake pain. It's very much real and I think I didn't have the coping skills that I wish that I did have for that."
Access to mental health providers for patient and family -Patients -Families	Participants emphasized the need for mental health professionals to provide dedicated emotional support, noting that while medical providers were supportive, they lacked specialized training in addressing the psychological impact of limb amputation. Many also wished their families had received more emotional support during the process.	"I think definitely the biggest thing would be a mentor and social support. I think it would be good if a prosthetic company came before and actually had you come and look. I feel like the surgeon though was just doing the surgery, but there was no psychologist. It is such a big emotional change." "There are two healing processes in cancer care. There's a physical healing and a mental adaptation process and I wished I had more emotional help." "There was no counseling. There was nothing at all." "I feel like if there was a team of people, that would help out a lot to get them prepared and explain to them what's going to happen. Because the unknown is a lot scarier than knowing what's going to happen." "They have seen me having two legs all my life. So, seeing me without one of them would be like a very hard thing on them basically." "It was really, really, hard for him (7 years old brother) I think probably just because I was getting all this attention, and he wasn't and partly because he didn't quite understand what was really going on and he thought I might die, and we were like best friends."
Body change support -Social connection -Self-image	Participants highlighted social connection and self-image as key post-amputation challenges, citing isolation from reduced peer activities and emotional difficulty adapting to bodily changes and others' perceptions.	"My friend, she was going to Aruba. She was like 'I just want you to know that you're completely invited. But I've scoped it out. It's not very wheelchair accessible.'" "My friends right now are planning a hike to the Grand Canyon. That really hurt me because they didn't even invite me obviously. There's just stuff that I can't do which I'm still not really knowing how to deal with yet because every amputee that I talk to deals with the same thing." "Because I have the prosthetic they are looking at me and like trying to figure out what I am doing." "The limb doesn't stop changing for the first year to two years. I was really expecting it to stop in six months. And my limb is still changing for almost two years now. I think, just to know that that's normal and that it is a long process, but it will eventually even out, I think would have been helpful."

3.1.3. Reflections and Advice

Participants enjoyed the opportunity to reflect upon their journey and offer advice to other pediatric and adolescent cancer survivors with limb amputation. They suggested other cancer survivors “keep a journal throughout the process” and personalize their hospital room. Maintaining connections with other amputees through social media was also encouraged. Several noted the importance of finding creative ways to say “goodbye” to their limb. For example, “Then I also had people sign my leg, which I know is very popular. That was really helpful because it was just like saying goodbye to it. Instead of hating it and hating that the tumor was there, being thankful which was very, very helpful for me”.

See Table 4 for additional definitions and quotes.

Table 4. Reflections and advice.

Theme	**Definition**	**Quotes**
Personalization	Participants found personalization helpful as a coping strategy, engaging in creative activities, customizing their environments, and connecting with others through support groups or social media.	“Every single hospital stay I have different decorations in my room for something to look forward to. Because you don’t want the room to look so blah. When you’re going in for amputation, you want a nice and bright comforter and a couple of decorations here and there.” “I know there’s a couple [amputation groups] on Facebook. Some people have osteosarcoma, some people have been in accidents, but they’re all dealing with amputation. Everyone always has all these different facts and all these useful links. It’s just very useful.”
Closure	Several participants discussed the importance of saying goodbye to their limb through creative activities.	“I think the most important thing to start off with is how can you get closure for the process. The way that I did closure was saying goodbye to my leg, and being thankful, and doing all those things. Like parasailing and going up in a helicopter. Just really having fun before things got serious basically. Putting paint all over my foot and doing this really cute picture so I’ll always have my footprint. Making little jokes. Like, hey, you can get 50 percent off of pedicures now. Little jokes like that and finding the right person to talk to.” “I painted my foot with different watercolors and then I put it on a piece of paper. I have that in my room. It’s really powerful to me to just see. It’s not just like a footprint on the wall. It’s like watercolor and it’s beautiful. That was really helpful. It was kind of like a fun activity to do as well.”

Table 5 summarizes the helpful and unhelpful experiences reported by pediatric and adolescent cancer survivors who underwent limb amputation. Helpful experiences included access to clear information, supportive providers, opportunities for connection, and emotional validation, all of which facilitated adaptation and healing. In contrast, participants also described unmet needs, including insufficient self-care guidance, limited pain management strategies, difficulty maintaining social connections, and poor access to mental health resources. Together, these themes highlight both the strengths of current care approaches and the critical areas requiring further attention to optimize patient support.

Table 5. Summary of helpful and unhelpful experiences.

Helpful Experiences	Unhelpful Experiences
Clear and timely informational resources about care (pre-surgery)	Educational materials not adapted to age, learning style, or developmental stage (pre-surgery)
Opportunities for personalizing the care process and achieving closure (e.g., memory-making, rituals) (pre-surgery)	Poor access to mental health services for patients and families (pre- and post-surgery)
Supportive, attentive medical providers (pre- and post-surgery)	Lack of practical information on self-care and recovery (post-surgery).
Connection with peers and others with similar experiences (pre- and post-surgery)	Limited strategies offered to manage acute and chronic pain (post-surgery)
Access to supportive counseling and emotional care (pre- and post-surgery)	Difficulty regaining physical activity and mobility (post-surgery)
Encouragement and validation from family and providers (pre- and post-surgery)	Challenges maintaining social connections and coping with body image/self-esteem concerns (post-surgery)

4. Discussion

Little is known about how to best support pediatric and adolescent cancer patients facing limb amputation. Our findings suggest that informational resources, including access to cancer-related organizations, were readily available. Developmentally sensitive and honest communication is vital. Ongoing assessments of their level of interest and preferred learning style are necessary. Age is not always a reliable indicator of a child's informational needs, as they can change over time. Assessment should take place before and after surgery, not only to evaluate a patient's understanding of the treatment, but also to explore its emotional implications [15].

Participants found emotional support by communicating with other individuals who have undergone similar experiences. Peer modeling and connection helped foster a sense of normalcy and belonging [16]. Participants also received comfort from their medical care providers, who sought to underscore the message that their patients could lead rich and fulfilling lives. However, while positive messaging is essential, the data suggest that providers must avoid overly optimistic assurances that may downplay the challenges of recovery and adjustment. Realistic preparation, including discussions of adaptation and loss, is essential for psychological resilience [17].

Participants highlighted the urgent need for emotional support related to pain management and mental health access. While most anticipated surgical pain, they were unprepared for phantom limb pain, which has been described in 60–80% of amputees [18]. Phantom limb pain often requires a multimodal approach, including pharmacologic agents, such as gabapentin or opioids [19], and non-pharmacological approaches, such as acupuncture or mirror therapy, where patients perform symmetrical movements in front of a mirror to "trick" the brain into perceiving the movement in the missing limb [20]. Finally, psychological approaches to pain management, such as guided imagery or cognitive-behavioral interventions, have been recommended for pediatric cancer patients [21].

Mental health support was repeatedly cited as essential but underprovided. Youth expressed a need for access to psychologists before and after surgery to help process fear, grief, and identity changes. Family members also experienced distress, yet few had access to mental health support. This aligns with broader pediatric oncology literature showing that family-centered psychosocial interventions improve coping outcomes and reduce distress in parents and siblings [22]. Support groups, both online and in-person, can also foster a sense of community and reduce isolation. In recognition of these needs, the Standards for the Psychosocial Care of Children and their Families (the Standards), published in 2015, emphasized the importance of integrating mental healthcare throughout the cancer trajectory [23]. However, many cancer centers lack the staffing needed to translate the Standards into routine clinical practice [24].

Analyses revealed a tension between feeling visible and feeling vulnerable. This highlights the importance of providing anticipatory guidance on body image changes [25]. Pediatric studies show that children with visible differences often experience greater social anxiety and stigma, particularly during adolescence, when peer identity becomes paramount [26]. Interventions should include opportunities for open dialogue about body image, reassurance of worth beyond appearance, and guidance on how to discuss their bodies with peers.

Many participants reflected on the importance of personalization and closure. Small gestures, such as decorating their hospital rooms or journaling, offered agency during a time of medical powerlessness. Encouraging creative rituals, such as having peers sign a cast or taking a photo with the limb pre-surgery, helped promote closure and symbolic transition [27]. These practices can be incorporated into clinical preparation protocols.

Maintaining peer connections, particularly with other amputees, was another consistent theme. Participants noted that social media provided an accessible means to exchange advice, vent frustrations, and gain emotional validation. Structured peer mentorship programs may offer a formal avenue for these benefits [28]. Providers can also create or refer interested families to curated networks of age-matched peers who have undergone similar procedures. See Supplemental Table S2 for a list of nationally available resources.

Limitations and Future Research

Several limitations need to be considered. The small sample size makes definitive statements about guidance challenging. Because interviews were conducted over the phone, nonverbal cues could not be captured. However, this qualitative study offers a springboard for future research with larger and more diverse samples.

Many participants discussed the importance of attaining good prosthetic function, but we did not capture detailed accounts from each individual regarding their prosthetic experiences. Challenges related to body changes and self-image also emerged as central concerns, underscoring the need for longitudinal research to better understand the physical and psychological adaptation to prosthetic use. Retrospective recall is another possible source of bias, though it offered participants the benefit of temporal distance from the immediate trauma of amputation, allowing for a more nuanced reflection on resources that supported their healing.

Selection bias is possible, as participants not only survived cancer but may also have been better adapted than those who did not participate. Another consideration is that participants underwent amputation in different calendar years, during which models of care may have varied. Prior exposure to intensive chemotherapy may have also influenced participants' perspectives on amputation.

Our sample was relatively homogeneous in terms of racial and ethnic composition, compared to the broader epidemiologic distribution of pediatric and adolescent osteosarcoma, which disproportionately affects Black and Hispanic youth [29]. Efforts to include perspectives from a more diverse patient population are warranted, as existing literature demonstrates that marginalized groups often face discrimination in healthcare settings, which compromises access, quality of care, and outcomes [30,31]. Finally, perspectives of family members were not included. Retrospective and prospective research should seek to broaden the understanding of the needs of parents and siblings.

Despite these limitations, this study has important strengths. To our knowledge, it is among the first to elicit in-depth perspectives from pediatric and adolescent survivors of osteosarcoma who experienced limb amputation. By centering the voices of individuals with lived experience, the study provides valuable insights into both pre- and post-amputation needs. These findings offer a rare and meaningful foundation for developing patient-centered interventions that can improve preparation, rehabilitation, and long-term psychosocial support for this population.

5. Conclusions

Research suggests that amputations in childhood and adolescence can be particularly traumatic, yet no prior studies were found that examined what pre-surgery interventions youth find helpful. It was also not known whether individuals who had an amputation during their childhood or adolescence found being involved in the planning and decision-making process useful. This pilot study is the first available literature to describe preparation for amputation in youth with cancer. The findings underscore the need for holistic, developmentally tailored, and emotionally attuned care before and after amputation. Future intervention models should integrate psychoeducation, peer support, and mental health resources across the cancer journey.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/cancers17172755/s1. Supplmentary S1: IRB notification and Oral Consent Procedures; Table S1: Interview Protocol; Table S2: Current Resources for Children and Adolescents with Osteosarcoma.

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Commentary

Leveraging the Patient and Family Voice in the Development of Patient Education: Supporting the Pediatric Oncology Experience

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Simple Summary: The inclusion of patient families in clinical program development and research design has received growing recognition in recent years. While the existing literature highlights broad involvement methods, such as focus groups and patient and family advisory councils, detailed examples of how these collaborations shape intervention development and execution—particularly in the context of patient education materials in pediatric oncology—remain limited. This commentary aims to highlight the importance of leveraging the patient and family voice in the development of patient education materials to further support the pediatric oncology experience. By examining the research to date and offering concrete examples of these collaborations, the authors hope to highlight not only practical approaches but also the critical importance of keeping the patient and family voice at the center of this work.

Abstract: Given the understandable patient and caregiver distress that often fluctuates across treatment, especially at diagnosis and with major disease milestones, it is important to prioritize interventions that are known to best support families during these key times of transition. Individualized and time-sensitive psychoeducation and anticipatory guidance are effective interventions in reducing distress and fostering adaptive coping across the cancer trajectory and are a standard for the psychosocial care of children with cancer and their families. Patient education materials have proven to be a flexible, accessible, cost-effective, and easily adaptable tool for delivering these interventions to this population. Moreover, within the field, there has been a call to action for the inclusion of patients and families in the development of research study design and clinical programming, recognizing them as the experts of their own experiences. This commentary aims to explore how patients and families can be actively involved in the creation, assessment, and implementation of patient education materials, fostering a collaborative partnership between families and clinicians. We provide detailed examples of this collaboration in the creation of patient education materials focused on promoting coping at diagnosis, supporting siblings and assisting with social reintegration at the end of treatment, promoting responsive caregiving for young children, and supporting caregiver coping through the creation of a podcast. Ultimately, this commentary seeks to emphasize the importance of partnering with patients and families to ensure their voices remain central to intervention development and implementation.

Keywords: childhood cancer; psychosocial care; psychoeducation; patient education; patient-family collaboration; anticipatory guidance

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1. Background

Approximately 15,000 children and adolescents in the United States are diagnosed with cancer each year [1]. Understandably, the receipt of a childhood cancer diagnosis is often met with heightened distress by the child and their family [2]. With appropriate psychoeducation and anticipatory guidance, as well as time to process the diagnosis and determine a treatment plan, distress levels decrease [3–6]. This is not to say that heightened distress is only present at diagnosis. It is not uncommon for levels of distress to ebb and flow throughout treatment, with major milestones and transitions [7–10]. In fact, the Standards for the Psychosocial Care of Children with Cancer and their Families [11] tout the importance of access to anticipatory guidance and psychoeducation to promote adaptive coping across the cancer care continuum [12].

The Pediatric Psychosocial Preventative Health Model (PPPHM) [13] provides a framework for understanding and stratifying psychosocial risk using three distinct tiers, which then guides intervention implementation. Those at the lowest risk are captured within the universal level (i.e., exhibiting expected levels of distress); those at moderate or high risk fall within the targeted (i.e., elevated distress in the context of identified risk factors) or clinical level (i.e., significant and persistent distress), respectively. This screening approach allows for tailored interventions based on psychosocial risk, such that targeted (e.g., collaborative identification of adherence barriers and strategies to improve adherence) or clinical (e.g., cognitive-behavioral therapy for anxiety) interventions can be offered alongside universal interventions when indicated [13]. Recent findings suggest that the implementation of this universal screening process followed by personalized care, such as psychoeducation at the universal level, is crucial in promoting equitable access in pediatric health care [14]. Further, systematically delivering psychoeducation serves as a platform for meeting the related standard for psychosocial care of patients and families facing pediatric cancer [12].

1.1. Benefits of Using Patient Education to Facilitate Psychoeducation

One tool for supporting the delivery of psychoeducation is patient education materials. This mechanism for intervention implementation offers many benefits, including portability, affordability, broad reach, and tailorability.

1.1.1. Portability and Flexibility of Format

Patient education materials can be shared in a paper-based format (e.g., booklet, handout) or electronically (e.g., through the EHR, e-mail, website, QR code). The ability to offer these materials in various formats increases their accessibility and portability [15], allowing for a greater number of patient families to obtain and benefit from these interventions.

1.1.2. Affordability and Cost-Effectiveness

Additionally, compared to other intervention modalities, patient education materials are relatively low in cost and resource burden [16]. There is also a plethora of data indicating a positive return on investment of patient education materials, suggesting a cost-effective and impactful avenue for universal intervention [17].

1.1.3. Reach

Efforts to more consistently use a plain language editing process have improved comprehension and uptake of information and recommendations, increasing access to those with limited health literacy [18]. Further, with advances in technology, there is now the ability to translate web-based patient education materials into additional languages [19].

1.1.4. Tailorability

When considering childhood development broadly, there is uniform recognition of unique differences and needs of children across developmental stages as they make their way toward adulthood [20]. It is important for this to also be reflected in patient education materials. Content can, and often should, be adapted to address the unique needs of patients across developmental stages (e.g., infancy, early childhood, school-age, adolescence, etc.), ensuring applicability and appropriateness of anticipatory guidance and psychoeducation provided through these materials [12].

1.2. The Patient and Family Voice

There has been a relatively recent recognition of the importance of the inclusion of patient families in the development of clinical programming, as well as research study design [21]. This has led to a wider representation of patient families within national and international scientific and clinical societies. Further, as patient families are the experts on their experiences, there has been an uptick in the creation of patient and family advisory councils and boards to review and provide comment on institutional and clinical initiatives [22]. The engagement of patient families as key collaborators in the development of patient education materials is paramount in facilitating the acceptability of these interventions and optimizing their benefits. While there is a dearth of literature outlining established frameworks for patient-family engagement in the development of patient education materials specifically, the extant literature provides frameworks that can be adapted for this purpose. The Patient-Centered Outcomes Research Institute (PCORI) details six core expectations for patient-family collaboration in research. These include: (1) broad representation, (2) continuous engagement of patient-family collaborators at each step in the research process, (3) funding for patient-family engagement in research, (4) leveraging unique skillsets and providing training/support for teamwork, (5) the inclusion of patient families in decision making, and (6) eliciting feedback assessing engagement [23]. While developed specifically for patient-family engagement in research, many of the core principles within PCORI can be adapted to patient-family engagement in clinical programming. Considering intervention development more specifically, intervention mapping is a process that was developed to engage patient families and key collaborators in intervention and program development. It is comprised of six unique steps: (1) conducting a needs assessment, (2) identification of objectives, (3) theoretical framework/justification for the chosen intervention, (4) intervention design and planning, (5) intervention implementation, and (6) intervention evaluation [24]. These steps can be applied to the development of various interventions, including the creation of patient education materials to deliver psychoeducation. Although this commentary does not represent original research or quality improvement methodology, it seeks to outline ways in which patient families can be actively engaged in the development, evaluation, and implementation of patient education materials through an intervention mapping approach, representing a partnership between patient families and clinical staff.

2. Collaborative Needs Assessment and Identification of Objectives

Prior to the development of any patient education materials, it is strongly recommended that an assessment with key collaborators be conducted to ensure the materials developed will meet the needs of and be acceptable to the individuals who will be providing, as well as receiving, the intervention [25]. Clinicians are likely to be knowledgeable about various content domains and are responsible for considering theoretical frameworks for intervention development, as well as the theoretical and empirical justification for the chosen intervention. Collaboration between clinicians and the patient's family is critical in identifying which aspects of a content area are likely to be most needed, desired, and impactful to patient families [21]. For example, while *all* aspects of sleep hygiene are important, patient families may identify that there is one aspect that is universally challenging that they would like to prioritize (e.g., balancing a consistent sleep/wake schedule in the context of medications that frequently induce drowsiness). The collaborative needs assessment can take many forms (e.g., patient-family advisory council, focus group, online advisory community), and the modality for the assessment largely depends on which avenues are accessible to the clinician. The needs assessment should be conducted once an avenue is selected for patient-family engagement and prior to intervention development [24]. Importantly, following the needs assessment, ongoing collaboration with patient families at each step of intervention development, implementation, and refinement is essential in creating interventions that are seen as acceptable to patient families [25].

In addition to the collaborative identification of the most salient content areas for patient education materials, adequate theoretical justification for intervention development is paramount to the effectiveness and utility of the intervention [24]. Further, ongoing conversations with collaborators, especially patient-family collaborators, are important in guiding other aspects of intervention development. These include when the intervention should be delivered, how the intervention should be delivered, and the target audience for the intervention. The timing of intervention delivery within the cancer care continuum is critical to the uptake of patient education materials and the optimization of intervention effectiveness [26]. For example, the consideration of a patient and family's cognitive load is imperative in intervention timing [27]. As such, collaboration among psychosocial disciplines to minimize duplication of services and protect against the redundancy effect, as well as to spare cognitive load, is critical. This allows patient education interventions provided by each discipline to be optimally consumed and utilized. Additionally, the timing of the intervention should be informed by when the information is likely to be most beneficial for patients' families. For example, patient education outlining strategies for coping with radiation therapy should ideally be given prior to the start of radiation therapy but not so far in advance that families may not recall the information provided.

Furthermore, content area, target audience, and patient-family collaborator input should be considered when determining what amount of information should be included and how it should be presented [16]. If the content is being created for consumption by children, emphasis on graphics over written text is likely to be most appropriate and beneficial [28]. Additionally, there has been some literature to suggest increased involvement in intervention consumption by children when there is a gamified component to the intervention [29].

3. Collaborator Engagement in Intervention Development, Implementation, and Evaluation

The partnership between clinicians and patient families should extend beyond the initial collaborative needs assessment. The inclusion of patient families on the intervention development team is crucial to creating an intervention that will be acceptable to and meet the identified needs of patient families while being grounded in evidence-based practice [21]. Although the engagement of one patient or family member in informing intervention development may lead to novel insights not thought of by others on the development team, the absence of input from multiple patient families is likely to decrease the generalizability of the intervention to a broad range of patient families [30–32]. For example, the inclusion of a caregiver of a school-aged child on the development team would provide excellent insight based on this patient family's experience; however, it would not be representative of caregivers of all school-aged children and certainly not representative of caregivers of young children or adolescents. As such, the engagement of diverse patient families in intervention development is essential in optimizing the acceptability and appropriateness of the intervention for a broader audience. Diversity in representation should be considered broadly and may include age, sex, gender, primary language, cancer diagnosis, and socioeconomic status, among others. The avenue for engaging patient families in intervention development, implementation, and refinement processes will depend on which modes are available to the clinician. Below, three potential avenues are discussed, including the benefits and drawbacks of each approach (see Table 1 for direct comparisons). As emphasized previously, patient families should be involved from the beginning of the intervention development process, starting with the needs assessment, with continued involvement through intervention evaluation and optimization [24].

Table 1. Avenues for Patient-Family Engagement.

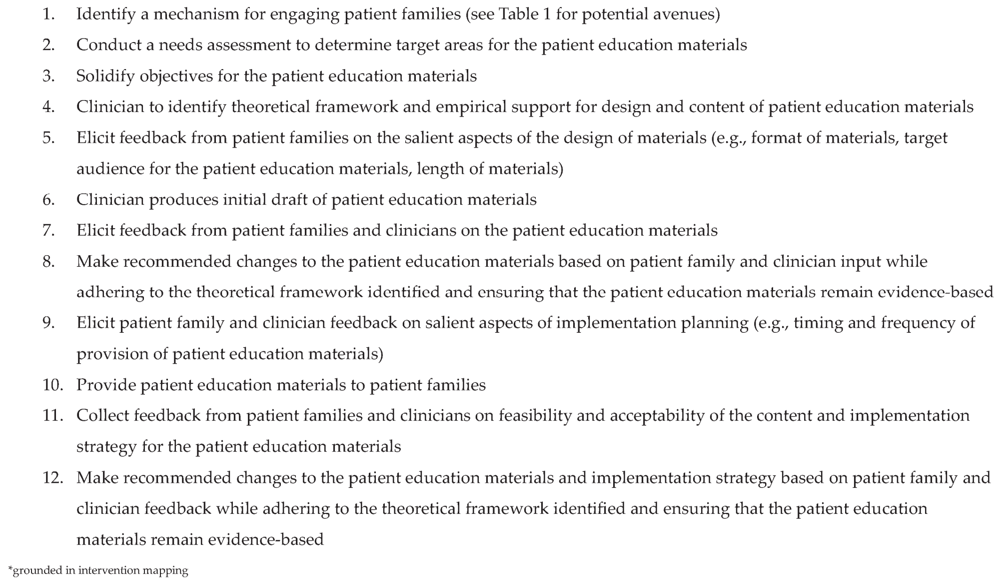
Avenue	Benefits	Drawbacks
Focus Groups	• Discussion among multiple patient families • Diverse family perspectives • Ideas/thoughts may be triggered during the group based on something another participant shares • Discussions may be guided or open-ended	• Resource intensive • Time intensive • Potential participant discomfort sharing opinions in a large group
Patient-Family Advisory Councils (PFACs)	• Institutionally supported • Aim to be representative of the patient population • Diverse family perspectives • Discussion among multiple patient families	• May not always be representative of the patient population • Time intensive • Resource intensive
Online Advisory Communities—Forum	• Specific questions able to be posed • Allows for unsolicited feedback • Patient families can provide feedback at their leisure • Wider reach • Possibly more representative of the patient population • Not time intensive for patient families	• Resource intensive (moderator) • Time intensive (moderator) • Difficult to regulate and for moderator to respond to potentially harmful comments in real time
Online Advisory Communities—Surveys	• Patient families can provide feedback at their leisure • Allows for anonymity • Responses are structured and answer specific questions • Allows for a variety of methods for obtaining feedback (e.g., likert scale, ranking, open-ended) • Wider reach • Possibly more representative of the patient population	• Clinicians cannot follow up on anonymous replies for clarification • No back-and-forth discussion among participants

One mechanism for gathering input from a wide variety of patient families is through focus groups [31]. Focus groups allow for discussion among multiple patient families. These discussions can be guided, with set questions posed by the moderator of the focus group, or open, with little input and direction from the moderator [33]. Focus groups offer the opportunity for diverse patient families to discuss and offer input and direction on intervention development. As multiple patient families are present, focus groups offer the advantage of an idea or thought to be triggered by something another patient family says, therefore, enriching the discussion and taking the conversation in directions it may not have gone otherwise. While there are many positive aspects to focus groups, drawbacks can also be present. Focus groups are frequently resource and time-intensive, both in terms of scheduling and facilitating, as well as in synthesizing the information shared [34]. Further, depending on the dynamic of the group, some patient families may feel uncomfortable sharing their perspective, which would then potentially limit the generalizability of the information gleaned during the group [35].

Another system for receiving ongoing input from patient families is through patient-family advisory councils (PFACs). PFACs typically support the mission of hospitals by providing a family voice in health care decision-making to improve quality, safety, and experience of care [36]. PFACs promote an environment where families and professionals work together to ensure optimal patient care, and they represent an effective mechanism to partner with families to develop and implement new programs and interventions. Frequently, there are efforts to ensure PFACs are representative of the patient population and include patient families from various backgrounds [37]. PFACs can be time intensive and require significant commitment from patient families, which may preclude the involvement of some families.

Lastly, online patient-family advisory communities offer a unique approach to soliciting feedback from a large group of patient families. These can be constructed in a multitude of ways. Online advisory communities can be structured to allow for free-flowing conversation among members of the community, often overseen by a moderator of the forum [38]. The moderator can pose specific questions or elicit feedback on a particular topic. Members of the online advisory community may also be able to offer feedback or input unprompted, should the forum be designed in this way. Other online advisory communities are structured with patient families as respondents to more traditional surveys [39]. While this method allows patient families to respond to calls for feedback and input at their leisure and frequently with the option for anonymity, one significant drawback is the inability to engage in back-and-forth discussion among the group of patient families or between patient families and the clinician or researcher eliciting feedback. Further, should the input be provided anonymously by a member of the online advisory community, this precludes clinicians and researchers from seeking clarification or asking follow-up questions of specific individuals within the online advisory community.

As no one avenue for patient-family engagement is likely to provide a comprehensive picture of the patient-family voice, utilization of multiple methods for patient-family collaborator engagement and input is recommended [25]. In addition to patient-family involvement in crafting the intervention, it is important to elicit feedback and input again from patient families once the intervention development is complete and prior to its implementation. Further, ongoing feedback following intervention implementation using the above-mentioned avenues for engagement to determine acceptability and incorporate additional feedback, in turn, optimizing the intervention, is paramount to its success [23]. See Figure 1 for a list of steps to consider in collaborating with patient families in the development of patient education materials.



1. Identify a mechanism for engaging patient families (see Table 1 for potential avenues)
2. Conduct a needs assessment to determine target areas for the patient education materials
3. Solidify objectives for the patient education materials
4. Clinician to identify theoretical framework and empirical support for design and content of patient education materials
5. Elicit feedback from patient families on the salient aspects of the design of materials (e.g., format of materials, target audience for the patient education materials, length of materials)
6. Clinician produces initial draft of patient education materials
7. Elicit feedback from patient families and clinicians on the patient education materials
8. Make recommended changes to the patient education materials based on patient family and clinician input while adhering to the theoretical framework identified and ensuring that the patient education materials remain evidence-based
9. Elicit patient family and clinician feedback on salient aspects of implementation planning (e.g., timing and frequency of provision of patient education materials)
10. Provide patient education materials to patient families
11. Collect feedback from patient families and clinicians on feasibility and acceptability of the content and implementation strategy for the patient education materials
12. Make recommended changes to the patient education materials and implementation strategy based on patient family and clinician feedback while adhering to the theoretical framework identified and ensuring that the patient education materials remain evidence-based

*grounded in intervention mapping

Figure 1. Steps for Collaborative Development of Patient Education Materials *.

4. Putting It into Practice: Examples of Patient-Family Collaboration in the Creation of Patient Education Materials

At our institution, nine steering councils (or subcommittees) report to the PFAC, with the staff champion and caregiver lead of each council sitting on the PFAC. This ensures an optimal bi-directional flow of communication between the PFAC and the steering councils. The focus of the steering councils spans hospital efforts, with some examples including patient experience, caregiver support, quality and patient safety, and nursing. The Patient Education Steering Council is comprised of eight caregiver advisors and three staff members who are intimately involved in hospital communication and education efforts, such as development of a learning management system for caregivers, creation of educational content for an online resource platform, review of written educational materials, and development of educational videos for caregivers about infection control, medication safety, and other treatment-related topics. The engagement of caregiver advisors through the Patient Education Steering Council and other councils has contributed directly to hospital planning, evaluation, and policy to improve the care of patients and to guide best practices. Our institution is also fortunate to have a diverse and active online advisory community. We have partnered with patient-family collaborators on the PFAC, through online advisory communities, and as formal project team members in the creation of patient education materials to promote coping throughout the illness trajectory (i.e., at diagnosis and completion of cancer-directed therapy) and to support the unique needs of patients and their caregivers.

The examples of patient education materials developed at our institution are discussed below and are representative of a universal intervention for patient families within the PPPHM. While the patient education materials described next are offered to all families when meeting with psychology at specified timepoints in the cancer care continuum (i.e., at diagnosis and end of therapy, at admission to the hospital, as indicated), they are also readily available in our waiting room to any interested patient family and digitally to patient families outside of our institution through together.stjude.org. These materials are available in printed form in English and Spanish and digitally in 12 languages. Although patient-family feedback has yet to be collected in a systematic fashion regarding these materials, initial anecdotal feedback has been uniformly positive. Please see Supplemental Material for hyperlinks to these published patient education materials.

4.1. Patient Education Materials to Promote Coping and Adjustment at Diagnosis

Patient education materials outlining strategies to support psychosocial health during treatment were developed as a component of our clinical program that offers universal psychological assessment and intervention to all oncology and transplant patients near the time of diagnosis. The patient-family voice was central to the development of this program, with aspects of intervention mapping utilized (i.e., interventions grounded in theory and best practice, patient-family engagement in program development and evaluation). The PFAC provided support for the program, as well as guidance regarding the scope of services offered within the program. Building upon advisement from the PFAC, patient education materials were created with input from subject matter experts, including pediatric psychologists who specialize in assessment and treatment across developmental stages (i.e., early childhood, school age, adolescence and young adulthood) and clinical domains (e.g., sleep, pain, etc.) in pediatric oncology. In addition to clinical training and practice, content derived from subject matter experts was born out of years of input received from patients' families who were willing to share the challenging aspects of their oncology journey. These patient education materials were developmentally tailored and created to augment in-person intervention, with the goal of sharing broadly applicable strategies to promote patient coping and adjustment near the time of diagnosis and throughout cancer-directed therapy. Following the launch of this clinical program and the implementation of the patient education materials, we leveraged our online advisory community to examine the patient-family perspective of psychology services near the time of diagnosis. Please see Bernstein et al. [40] for a comprehensive description of methods and results. Caregivers identified multiple psychosocial areas to be very important or somewhat important [40]. While many of these domains are addressed in current versions of our patient education materials, the patient-family perspective will shape the development of future education to support patient families at diagnosis.

4.2. Patient Education Materials to Support Sibling Coping at End of Treatment

Consistent with the recognized need for systematic psychosocial support at various points throughout the cancer care continuum, a new institutional program aimed at providing support to patient families during early survivorship was created. Shortly following the launch of this program, a survey was sent out to the patient-family online advisory community seeking feedback on the quality of support received during the transition off therapy. Forty-three caregivers completed the survey. One tangible need identified through this survey that was still unmet with the creation of the early survivorship program was systematic support for siblings during this transition. This need was echoed by 63 caregiver respondents in another survey sent to the patient-family online advisory community seeking insight on the patient-family experience with psychological and psychosocial support during the transition off therapy. As such, patient education materials were created by a team of psychosocial providers (i.e., psychologists, social workers, child life specialists) to address this identified gap. While general strategies were included in these patient education materials to support siblings of all ages, additional developmentally tailored strategies were also provided for young children and adolescents. This educational intervention is provided to all families of patients with siblings when meeting with psychology prior to the transition off therapy. Consistent with aspects of an intervention mapping approach, ongoing efforts to engage patient families in intervention evaluation and optimization are needed.

4.3. Patient Education Materials to Support Social Reintegration at End of Treatment

A survey was sent to the patient-family online advisory community to seek feedback and inform the optimization of existing services aimed at supporting patient social reintegration following completion of cancer-directed therapy and/or return to the child's home community. This survey sought to better understand the positive and negative aspects of current processes and mechanisms for supporting social reintegration, as well as allow families to provide suggestions on how our institution might improve these services. Results from caregivers (N = 55) provided meaningful guidance on the development of patient education materials outlining strategies to promote positive social development and functioning during this reintegration. This education was developmentally tailored and shared with all patient families when meeting with a psychologist at the time of transition off therapy and/or back into the home community. Consistent with aspects of intervention mapping, plans are in place to evaluate this intervention's acceptability and seek additional patient-family collaborator feedback to optimize the effectiveness of the patient education materials.

4.4. Patient Education Materials to Promote Responsive Caregiving for Young Children

Guided by the literature and an identified need, an institutional quality improvement project was initiated to increase responsive caregiving for infants and toddlers during hospitalizations [41,42]. The patient-family voice was essential at every step of the process, and an intervention mapping approach was utilized. A caregiver from the PFAC served as a member of the core project team, offering insight from her experiences. Prior to the development of any interventions to promote responsive caregiving, the project team presented to the PFAC for general guidance and recommendations. Additional information regarding caregiver experiences during their young child's hospitalizations was gleaned from caregiver responses (N = 18) to a survey distributed through the patient-family online advisory community. Feedback indicated that a substantial portion of caregivers did not receive education on the importance of responsive care or safe provision of responsive care (e.g., how to safely hold, diaper, or feed) for their child, but such education would have been helpful. As such, several educational interventions were collaboratively developed and reviewed by subject matter experts (e.g., nurses, critical care physicians, child life specialists, and psychologists) and caregivers. Caregiver input also guided the implementation of these interventions, which are provided to all families of patients under age 3 years at the time of inpatient hospital admission via electronic healthcare record and via display on the TV screen in the patient's room. The very positive impact of these interventions in increasing responsive caregiving behaviors was measured using a QI framework, and findings are under preparation for hopeful publication. At the suggestion of the caregivers involved in this project, the next educational intervention being developed is aimed at training bedside nurses to provide individualized in vivo education and modeling to families during hospitalization.

4.5. Collaborative Creation of a Podcast to Support Caregiver Coping

As part of ongoing efforts to enhance caregiver support programs and consistent with an intervention mapping approach, the institutional caregiver support committee surveyed our patient-family online advisory community. Questions were posed regarding the acceptability, feasibility, and utility of various caregiver support programs being considered for development. One hundred fifty-nine caregivers completed the survey. While most respondents indicated interest in support groups and social events, they also reported attendance barriers, including timing of the event, illness of their child, and lack of childcare. Moreover, many caregivers indicated that informational and educational support available through a podcast would be desirable and would eliminate most of these barriers. As such, the committee partnered with key collaborators in hospital administration and medical communications to begin the development of a podcast series for caregivers of children with serious illnesses. Patient-family collaborators were directly involved in content development, the selection of staff moderators and caregiver participants, and the naming of the podcast. Caregivers S.H.A.R.E. aims to support, honor, advise, reflect, and encourage. The content presented in the first two seasons included navigating a new diagnosis, handling logistics during treatment, seeking support, maintaining healthy relationships during treatment, supporting siblings, parenting during illness, and transitioning off active medical treatment. Consistent with both the mission of the institution and the universal level of the PPPHM, the podcasts are distributed widely on most major podcast platforms and are available to caregivers around the world. To date, the podcast has been downloaded 1746 times and in 21 countries.

5. Conclusions

The extant literature suggests that patient education materials are an effective means of providing universal interventions, including anticipatory guidance and psychoeducation, for pediatric oncology patient families [12,16,43]. There are multiple benefits to using patient education materials to provide psychoeducation to patient families, including the portability, affordability, reach, and tailorability of these interventions [12,15,16,18]. Additionally, the use of patient education materials to provide psychoeducation is consistent with the PPPHM and the Standards for the Psychosocial Care of Children with Cancer and their Families [11,13]. While patient education materials offer a robust mechanism for intervention delivery, the success of these interventions depends on their acceptability by patients’ families [15,16,18,43]. As such, it is critical to involve patient families in the intervention development process from the beginning, including in choosing content, working through logistical considerations, and intervention development and implementation [24]. Engagement with patient-family collaborators can take many forms, such as through engagement with PFACs, conducting focus groups, seeking input from online advisory committees, and having patient families as formal members of the project team. Intervention mapping is a recommended empirical approach to collaborative intervention development with patient families that should be considered [24].

While the benefits of using patient education materials in the provision of psychoeducation are significant [12,16,43], limitations must also be acknowledged. Patient education materials require routine review and revision to ensure that materials are in line with current evidence and best practices. Further, it can be challenging to create patient education materials that are broadly applicable to a wide audience while also acknowledging unique cultural factors and lived experiences of individuals. This highlights the importance of having diverse representation from patient-family collaborators in the intervention development process. Further, there are often individualized preferences for the delivery of patient education materials, with some individuals expressing a preference for tangible written education, while others desire to receive this information electronically or audiovisually [43]. Many of these limitations can be minimized by clinicians coming alongside patient families when providing psychoeducation through patient education materials to flexibly adapt and individually tailor the psychoeducation to meet the needs of each patient family.

An important limitation to note is that this commentary does not represent original research or quality improvement methodology but seeks to encourage engagement with patient families in the creation of patient education materials, with recommended steps provided based on the extant literature [24] and tangible examples of how this has been accomplished at our institution. Strict adherence to intervention mapping was not utilized in the development of patient education materials at our institution, and we recognize that this is a limitation in drawing specific and robust conclusions from the examples provided. As such, the conclusions presented are based on the extant literature. Ongoing assessment of patient education materials at our institution presented as examples in this commentary is needed to empirically examine the acceptability and effectiveness of these interventions. Further, examples provided are based on the experience of one institution, which may limit generalizability. For those interested in engaging families in the development of patient education materials, Figure 1 provides a recommended process grounded in intervention mapping.

Going forward, continued collaboration with patient families to guide the expansion and refinement of established patient education materials is essential. Further, while there has been increased attention to the incorporation of the patient-family voice in clinical efforts, there are still areas for improvement. Formalized guidelines for the incorporation of patient families in *all* aspects of clinical programming and intervention development are essential to ensure that patient families have a voice in aspects of their care that will directly impact them and are the next step in systematically including patient families in this way.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/cancers17071201/s1, Published patient education materials.

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Commentary

Bridging the Gap: Embedding Psychosocial Oncology Research into Comprehensive Cancer Care for Children and Young People

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Simple Summary: Cancer care has advanced dramatically in recent decades, yet psychosocial care—support for the mental, social, and emotional health of young patients and their families—remains under-resourced and often disconnected from medical treatment. Australia is building the Minderoo Children's Comprehensive Cancer Centre, the first of its kind in the southern hemisphere, to provide world-class cancer care for children and their families, regardless of background or location. The aim of the Centre is to fully integrate psychosocial care with cutting-edge medical research and treatment, ensuring that every child receives not only the best physical care but also support for their emotional and social needs. Research shows that when psychosocial care is included, children and families experience better quality of life and improved outcomes. However, barriers such as limited funding, fragmented research, and a lack of collaboration slow the translation of research into everyday care. This commentary highlights that to truly achieve comprehensive care, it is essential to invest in partnerships, include families in research, and reform funding so that mental health support becomes a routine part of cancer treatment. This approach will help ensure that advances in science lead to real improvements in the lives of children with cancer and their families, benefiting society as a whole.

Abstract: The National Cancer Institute designated models of comprehensive cancer care centres endeavour to enable the delivery of high-quality, holistic cancer care, informed by research evidence across the cancer care trajectory. These comprehensive cancer centers have typically been adult-oncology-focused, leaving an important gap and opportunity to consider what a model of comprehensive cancer care might look like for children and

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young people. With the advent of the opening of the first comprehensive children's cancer center in Australia and the southern hemisphere, this commentary considers the important role that psychosocial oncology needs to play in driving high-quality, person-centered comprehensive cancer care for all.

Keywords: pediatric and adolescent cancer care; psychosocial support; integrated care; family-centered care; quality of life

1. Introduction

This commentary was written, quite literally, while staring out the window at the construction site of the Minderoo Children's Comprehensive Cancer Centre [1] (MCCCC). Due to be completed in late 2025, the building will house Australia's first comprehensive children's cancer center, the first of its kind in the southern hemisphere [1,2]. The energy, hope, and excitement surrounding the new building is palpable. Inside its doors, scientists, clinicians, and researchers will sit side-by-side, buoyed by a vision of collaborative, multidisciplinary, research-driven healthcare that ensures Australia's youngest cancer patients, and their families, receive cutting-edge treatment and care regardless of where they live, their families' financial situations, or cultural background.

The word "comprehensive", in this instance, is doing some heavy lifting. A National Cancer Institute-designated comprehensive cancer center requires that research is comprehensively integrated into care, at all levels, and across the entire cancer care trajectory [2]. Relevant to psychosocial oncology, key hallmarks of a comprehensive cancer center include excellence in multidisciplinary cancer treatment and patient care; the development of empirically driven clinical innovations; translational science spanning preclinical science to clinical implementation; educational programs to train cancer clinicians and scientists; education and support for patients and their caregivers; and finally, a commitment to linking patients and their caregivers with primary, supportive, and palliative care services [3].

Against this backdrop, this commentary aims to examine the evolving role of psychosocial research within pediatric oncology, highlighting both its essential contributions to holistic cancer care and the unique challenges faced in translating research into practice. We discuss the integration of psychosocial research into multidisciplinary treatment models; explore barriers to implementing evidence-based psychosocial interventions; and consider the new opportunities and persistent gaps that arise as comprehensive cancer centers strive to deliver equitable, family-centered care. In doing so, we seek to underscore the importance of embedding psychosocial research and support at every stage of the cancer care continuum, ensuring that advances in medical treatment are matched by progress in addressing the psychological and social needs of young patients and their families

2. Comprehensive Cancer Care—Tailored to Child and Family Needs

Several notable gaps appear when considering how we might build on international definitions to develop a local model of comprehensive cancer care for children. Firstly, to date, definitions of comprehensive cancer have mostly been age-agnostic and primarily adult-oncology-focused [3,4]. Secondly, while psychosocial oncology research can play a critical role in advancing each of these clinical aspirations, most definitions of comprehensive cancer care make little reference to the important role that psychosocial care plays in patient care [3,4]. This is a critical gap that challenges the delivery of comprehensive cancer care. However, it also presents an opportunity for groundbreaking research to advance our understanding of psycho-oncology and develop clinical care.

In addition to providing gold-standard clinical care aligned with Australia's National Cancer Plan [5], the MCCCC will formalize collaborations between three world-leading research centers—the Kids Cancer Centre at Sydney Children's Hospital, which provides a comprehensive service for children and adolescents with cancer and hematologic diseases and conducts the largest bone marrow transplant program in the region; the Children's Cancer Institute, which is the largest children's cancer research facility in the southern hemisphere and an internationally recognized leader in child cancer research; and the Behavioural Sciences Unit (BSU), Sydney, Australia's largest pediatric psychosocial oncology research team. Together, the Children's Cancer Institute and the Kids Cancer Centre lead ZERO Childhood Cancer (ZERO) [6–9], Australia's national child cancer precision program, which is at the international forefront of child cancer precision medicine programs and has a strong psychosocial program of research led by the BSU [6–16]. The BSU was incepted as a complementary entity to the Kids Cancer Centre at the Sydney Children's Hospital in 2001 by pediatric oncologist Professor Richard Cohn AM [Member of the Order of Australia]. Professor Cohn was driven by the principle that "cure is not enough" [17] and led critical pediatric psycho-oncology work, alongside the BSU's inaugural Director, Professor Claire Wakefield, to understand and document the experiences, suffering, and needs of patients and their families and the effect on the wider community [18–28]. The BSU has since developed many theoretically driven, evidence-based interventions to address these identified needs—with meaningful lived-experience input and co-designs conducted with young people and their families [29–34]. The timely investment in constructing the MCCCC is both a celebration of the progress made in children's cancer care and an opportunity to continue to integrate psychosocial research into the care and lives of young people and families living with cancer—acknowledging how much more needs to be done to achieve "zero childhood cancer" [35]—with zero long-term complications.

Watching the MCCCC emerge before our eyes has caused us to reflect on the progress of our field, articulate tensions often felt but not discussed, and develop a roadmap for the challenges and opportunities that lie ahead. In the spirit of progress, we are using this opportunity to ask ourselves some difficult questions as we cross the threshold of our new, comprehensive children's cancer center. Though the building itself has a structural blueprint, and the center has a clear vision, as a field in psychosocial oncology, do we have a similarly clear roadmap or vision? What does a child-friendly, family-centered model of comprehensive cancer care look like? And what can we do to bridge the gap between innovative psychosocial research and translation to sustained, meaningful changes in the healthcare system?

3. Truly Integrating Supportive Care

While progress has been made in *recognizing* psychosocial care as a critical component of cancer care [36–40], there is much-needed work to be done to reflect change in clinical practice, including protecting resources for a sufficient and skilled workforce and support of clinician–researcher roles in order to mainstream psychosocial services, ensure accessibility for all cancer patients, and advance the field [41]. Evidence-based models of mental health and psychosocial care have consistently emerged as top priorities for young people living with cancer and their families, both in Australia [42] and overseas [43,44]. And as a field, we have been able to develop psychosocial standards of care built on an extensive empirical literature base that can now drive our clinical care—and implementation and auditing tools—to help us map the path to bridge the gaps to get there [36,45].

Yet, internationally, even in high-income countries, psychosocial care often remains under-resourced in pediatric oncology [46–48]. Despite efforts to screen for, and prevent, mental health problems, psycho-oncology services often remain more reactive than proactive. Healthcare systems, including in Australia, also frequently do not have the necessary resourcing, managerial support, organizational ideology or culture, or governance (bureaucracy) to enable a clinician–researcher model to flourish in the psychosocial disciplines [49,50].

Nevertheless, encouraging progress is being made. Clinical trials are increasingly including quality-of-life end-points and psychosocial sub-studies in their programs of work. For example, PRISM-IMPACT was a sub-study that aimed to understand families and health professionals' experiences with PRISM, ZERO's first national clinical trial [6–9]. By better understanding families' experiences associated with the delivery of precision medicine, we can then generate resources and supportive interventions that better meet their needs—including during their precision medicine trial participation [8]. Comprehensive cancer programs that complement and enhance "usual practice" are being trialed, such as the "Engage" survivorship program. Engage provides a comprehensive assessment of survivors' medical and psychosocial concerns and facilitates access to care to survivors otherwise "lost to follow-up" through remotely delivered assessment and personalized, risk-stratified recommendations [51]. The program highlights the interrelated nature of physical and mental health problems and the need to address both simultaneously to overcome common barriers to accessing care. Yet, progress in these trials does not always translate to success in the "real world", despite illustrating effectiveness in the research setting.

4. Bridging the Gap Between Psychosocial Oncology Research and Comprehensive Cancer Care

Psychosocial oncology researchers have the potential to play a critical role in translating gold-standard research into standard-of-care practices to improve the experiences of cancer patients, survivors, and their families [52]. This imperative is reflected in research funders increasingly requiring evidence of real-world impact, both anticipated and realized, reflective of the "social return on investment" of this research [53]. Patients and families have also, rightly, come to expect research to translate into real changes in their care. Yet, there remain key systemic barriers to the translation of psychosocial oncology research into comprehensive cancer care, including the academic nature of research, the fragmented research recruitment processes, and reliance on individual clinical champions (that is, clinicians who help drive research within their organization and advocate for its integration into everyday practice). First, research is inherently academic, focused on generating knowledge rather than implementing systemic change. Funding models reflect this, supporting research activities but not operational costs for integrating findings into clinical care. Without resources for training, staffing, and sustaining psychosocial care models, even well-evidenced interventions may never be adopted in practice, and the potential benefit of research for the intended population is not fully realized. Addressing this gap necessitates prioritizing and funding translational research, as well as providing adequate support for services to enact and sustain these evidence-based changes.

Second, study-specific recruitment and ethics approvals restrict generalizability. Studies are often limited to single institutions, creating isolated, institution-specific changes rather than scalable solutions. While smaller-scale proof-of-concept and pilot studies have a critical place in the scientific pipeline, reliance on single-site studies alone can ultimately limit the broader applicability of interventions. To develop interventions that are effective across jurisdictions, it is important to build on the insights gained from these initial studies and progress them toward larger, multi-site research that can address cohort- and site-based variability and support wider implementation. Moreover, inconsistent research settings and participant pools complicate the creation of universal psychosocial care models, limiting their applicability. Patients and healthcare professionals may face uncertainty when navigating varied psychosocial care practices across different institutions, highlighting the need for cohesive, system-wide integration.

Third, while local clinical champions are essential for tailoring and driving implementation, their involvement is inherently uncertain, typically hinging upon some combination of collegial goodwill, in-kind contributions, and inputs that occur after hours due to a lack of protected time to engage in research activities. Competing clinical demands, limited funding after research grants expire, and staffing changes jeopardize the longevity of psychosocial programs that depend on clinical champions.

Each of these research gaps/challenges offers an opportunity for new approaches (see Table 1). For example, to address these challenges, we have trialed several key strategies at the BSU. One solution is fostering partnerships with not-for-profit cancer-support organizations to deliver psychosocial interventions developed through academic research at scale [54,55]. These collaborations leverage community infrastructure for national delivery, though they require researchers to become advocates and network-builders, roles for which they often lack training and resources. Another solution is embedding implementation scientists within research teams to enhance dissemination and scalability. Despite their potential, implementation scientists remain underrepresented in psycho-oncology, highlighting the need for greater focus and investment [52]. Additionally, integrating patients and families as co-leaders in research can assist with policy and advocacy efforts. Patient-inclusive advisory committees have been shown by health-services researchers to improve psychosocial service availability, fostering comprehensive care [56–58]. Health professionals and professional associations can also play a pivotal role in addressing these gaps by advocating for dedicated funding streams and protected time for psychosocial research translation within clinical settings. Additionally, policymakers and healthcare funders could establish incentives and policy frameworks that prioritize the system-wide integration of evidence-based psychosocial care, setting new objectives for equity, sustainability, and measurable patient outcomes.

Table 1. Gaps and opportunities in the integration of psychosocial research into comprehensive models of cancer care for all children/adolescents with cancer.

Translation Gap/Challenge	Opportunity
Evidence-based models of psychosocial care are identified as research priorities for young people/families, yet in practice, psychosocial care is often under-resourced and often reactive in nature.	Psychosocial research methods and principles should be integrated alongside clinical pediatric cancer research, including using lived experience and co-design methods to ensure psychosocial concerns are addressed in how medical care and clinical trials are explained and delivered.
Scientist–practitioner models that integrate research and practice are limited in psychosocial oncology due to a lack of funding for dual clinician–researcher positions and a lack of career pathways in the health system.	Competitive research funding schemes should include dedicated and specific opportunities for clinician–researchers. Likewise, a greater number of clinician–researcher positions—including for non-medical disciplines such as psychosocial/allied health and nursing disciplines—are needed to embed research translation into the health system.
Psychosocial interventions are often tested in isolation, unintegrated with the clinical care systems in which they are hoped to be integrated.	Research should focus on testing complex interventions, including more explicitly testing referral and access pathways driven by patient and family preferences.

Table 1. *Cont.*

Translation Gap/Challenge	Opportunity
Individual clinical champions at individual institutions can become overburdened or may not have the capacity to consistently support psychosocial oncology research.	Partnerships with the broader health system and community, including non-profit organizations, can expand the reach of psychosocial oncology research and increase access to evidence-based interventions. Future research should examine how digital technologies and artificial intelligence can be used to enhance how families can access high-quality support and information to supplement their clinical care.
The vast majority of psychosocial oncology research is conducted in high-income, English-speaking, Westernized countries, limiting the relevance and cultural appropriateness of psychosocial models/interventions. There has been little research examining how social determinants of health intersect to promote or threaten psychosocial outcomes.	Psychosocial oncology researchers should prioritize building partnerships with community groups to increase the reach and impact of their work. Researchers (and the funding schemes that fund their work) should also incorporate strategies to address current disparities in research participation/lived experience involvement, such as approaches that support the financial remuneration of consumers/participants.
The voices of young people and their families have traditionally been left out of the research development and translation pipeline.	Ensuring that research priority-setting is part of the research pipeline is critical to ensuring studies focus on what matters most to people with the lived experience of cancer. Furthermore, upskilling and reimbursing people with lived experience to contribute their expertise is a necessary step. Partnering with people with lived experience as co-experts and co-investigators from the study idea-generating phase through to implementation/dissemination is critical to ensuring their priorities remain central to the entire research translation pipeline.

Bridging the gap between psychosocial oncology research and clinical care necessitates structural funding reforms, cross-sector partnerships, embedded implementation science, and amplified patient voices to ensure equitable, sustainable, and holistic cancer care.

5. Comprehensive Cancer Care for All Young Australians

At the core of our advocacy efforts, we must focus on the integration of children, young people, and their families as co-leaders in the research we conduct. Amplifying their voices in the development, conduct, and dissemination of research studies and output is critical to ensuring that we conduct meaningful, impactful research that can generate the improved psychosocial outcomes and health system change that families want to see [59]. Families' voices are vital to generating the health system and policy change needed to implement psychosocial interventions into practice. Increasing evidence suggests that hospitals that involve patients and families in advisory committees or roles are significantly more likely to conduct psychosocial needs screening, offer more psychosocial support programs, and offer more community support partners to families to address needs [56–58]. The BSU has, therefore, together with the Kids Cancer Centre, been working to develop a youth-inclusive patient and family advisory committee to ensure families' voices will be amplified to advance research and improve care.

Despite the Australian healthcare system delivering some of the best survival outcomes for children and adolescents with cancer, equity of care in childhood cancer remains a challenge [5]. Childhood cancer does not discriminate based on social class, culture, or location; so, neither should access to cancer treatment or psychosocial oncology support.

The MCCCC is a critical step toward improving access to care. Yet, undeniable inequities do exist for our most vulnerable children and families—families living in rural/remote areas of Australia [60,61], those with lower socioeconomic backgrounds [62], people living with disability [63,64], gender- and sexuality-diverse young people [65,66], Aboriginal and Torres Strait Islander families [67,68], and families with culturally and/or linguistically diverse backgrounds (CALD) [43]. Over the past decade, psychosocial researchers across Australia have begun to improve reach to rural/remote families through the development and implementation of digital interventions to address psychological distress both during [69] and beyond active cancer treatment [29,70], physical activity [33], and long-term survivorship follow-up care [71].

For lower education and health literacy levels, our research has focused on improving consent processes and patient information and support resources, with the aim of empowering patients and caregivers at some of the most challenging treatment-related junctures, including in making complex treatment decisions [72] and when approaching end-of-life [73]. Yet, much child cancer research has excluded priority populations and missed explicitly examining important social determinants of health [74]. Common to the limitation sections of many psychosocial manuscripts are statements such as, "*Our sample was limited to English-speaking participants, with few perspectives from culturally and linguistically diverse (CALD) communities.*" With Australia's rich cultural diversity, where 3 in 10 people were born overseas [75], and Australia's Aboriginal and Torres Strait Islander peoples and communities experience documented systemic inequities/barriers to care [76], it is difficult (perhaps impossible) to develop a study that represents all unique communities. Encouragingly, however, our field has increasingly used participatory research methods involving individuals and family members with lived experience from these priority groups. Given the intersectional nature of many social determinants of health, future research will need to better leverage these methods to examine multiple, co-occurring sources of disadvantage and health inequity to truly develop models of comprehensive cancer care for all children and young people with cancer.

6. A New Paradigm? Researchers as Translators

As we look out over the scaffolding and cranes next door, what steps should we take to move toward comprehensive cancer care for children and young people? The building under construction provides us with an important mandate to develop—and action—a new blueprint.

Two decades ago, Sean Phipps wrote of how, as psychosocial researchers, we were "yoked" to the increasingly fast pace of medical progress and running to keep up [77]. This remains true in 2025, where precision medicine and other novel therapeutics have continued to challenge the abilities of our colleagues to keep up across all disciplines. The promise of new technologies and therapies also carries the threat that access to these new technologies may widen existing disparities. In this era of technological advancements and the emerging integration of artificial intelligence into healthcare domains, maintaining the patient- and family-centered focus of psychosocial research and ethical considerations will remain a priority for psychosocial researchers. There is untapped potential for psychosocial researchers to serve a mediating role between the lived experience of patients and families and the health-system changes that need to occur to ensure that each young person and family's experience is better than the last.

7. Conclusions

To realize the potential of comprehensive cancer care, our opportunity—and moral imperative—is to consider how we translate our field's research achievements into real-world practice change to reduce suffering for patients, families, and the health professionals who care for them. Over the past 20 years, as a field, we have been increasingly writing about the process of *research translation*. Yet, what of the potential role of the "researcher as *translator*"? As psychosocial oncology researchers, we occupy a unique and privileged position—hearing directly from children and families about their lived experiences and the "pain points" that they face across the cancer care trajectory [7,20,30,51,62]. Subtly re-envisioning the role of the psychosocial researcher as a research translator calls for us to leverage this unique position to act as conduits to translate this knowledge among key parties—not only elevating the voices of young people and families to inform our clinical practice—but also driving the changes needed at the health system and policy levels. Success in this endeavor will enable comprehensive, humanistic, and person- and family-centered cancer care. Beyond survival, what could be more important than that?

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