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Standards for psychosocial care for children with cancer and their families

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Standards for the Psychosocial Care of Children With Cancer and Their Families: An Introduction to the Special Issue

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Pediatric oncology psychosocial professionals collaborated with an interdisciplinary group of experts and stakeholders and developed evidence-based standards for pediatric psychosocial care. Given the breadth of research evidence and traditions of clinical care, 15 standards were derived. Each standard is based on a systematic review of relevant literature and used the AGREE II process to evaluate the quality of the evidence. This article describes the methods used to develop the standards and introduces the 15 articles included in this special issue. Established standards help ensure that all children with cancer and their families receive essential psychosocial care. Pediatr Blood Cancer 2015;62:S419–S424. © 2015 Wiley Periodicals, Inc.

Key words: cancer; child; family; pediatric; psychosocial; standards

INTRODUCTION

A large body of research documents the psychosocial risks for children and their families during and after cancer treatment and approaches to reduce distress and support patients and families.[1–3] Yet, there is a significant variability in psychosocial services offered to patients in different pediatric oncology settings. Furthermore, there are no published, comprehensive, evidence-based standards for pediatric psycho-oncology care.[4] To address this critical gap, 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. This special issue of *Pediatric Blood and Cancer* is a comprehensive set of short articles that describe the standards that have been identified as essential for psychosocial care and summarizes the relevant supporting evidence. This introductory article provides the background for the initiative and describes the methodology used to develop the standards.

METHODS

The formation of the PSCPCC and development of psychosocial standards of care for pediatric cancer have been dependent upon the collaboration and support from The Mattie Miracle Cancer Foundation (www.mattiemiracle.com). Mattie Miracle was started by Vicki and Peter Brown in memory of their son Mattie who died of multi-focal osteosarcoma at the age of 7 years. The foundation is dedicated to "addressing the psychosocial needs of children and families living with childhood cancer as well as educating healthcare providers on the impact of such a diagnosis on children and their families." On March 20, 2012, Mattie Miracle sponsored a congressional symposium and briefing on Capitol Hill stressing the importance of universal services to address the psychosocial needs of children with cancer and their families. The Browns identified five leaders in psychosocial aspects of pediatric cancer; each presented research data at the briefing related to standards for psychosocial care (Anne E. Kazak, PhD, ABPP [Chair]; Robert B. Noll, PhD, Andrea Farkas Patenaude, PhD, Kenneth Tercyak, PhD, Lori Wiener, PhD). A panel of parents and survivors further emphasized the need for psychosocial care for children with cancer and their families. It became clear in conversations with members of Congress and their staffs that any legal or government support for such universal psychosocial care would require clear, widely accepted, well-supported standards for the psychosocial support of children with cancer and their families. Development of these standards based on existing research and existing consensus became a priority of Mattie Miracle and the group leaders.

The existing literature on guideline development informed our development of standards for the psychosocial care of children with cancer.[5,6] (Fig. 1). The group leaders completed a systematic review of current pediatric psychosocial published guidelines, recommendations, standards, and consensus reports.[3] The review not only highlighted the notable past efforts to define and characterize standards of psychosocial care for children with cancer and their family members, but also showed the lack of a widely accepted, up-to-date, evidence and consensus-based, comprehensive standard.

To ensure coverage of all critical areas of psychosocial care, we next constructed and administered an online survey to 20 additional

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Abbreviations: AGREE, The Appraisal Guidelines for Research and Evaluation; APOS, American Psychosocial Oncology Society; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses; PSCPCC, Psychosocial Standards of Care Project for Childhood Cancer

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Conflict of interest: Mary Jo Kupst has served as a consultant to the Mattie Miracle Cancer Foundation for the Standards Project. Mattie Miracle has also provided some travel and conference support for the think tanks.

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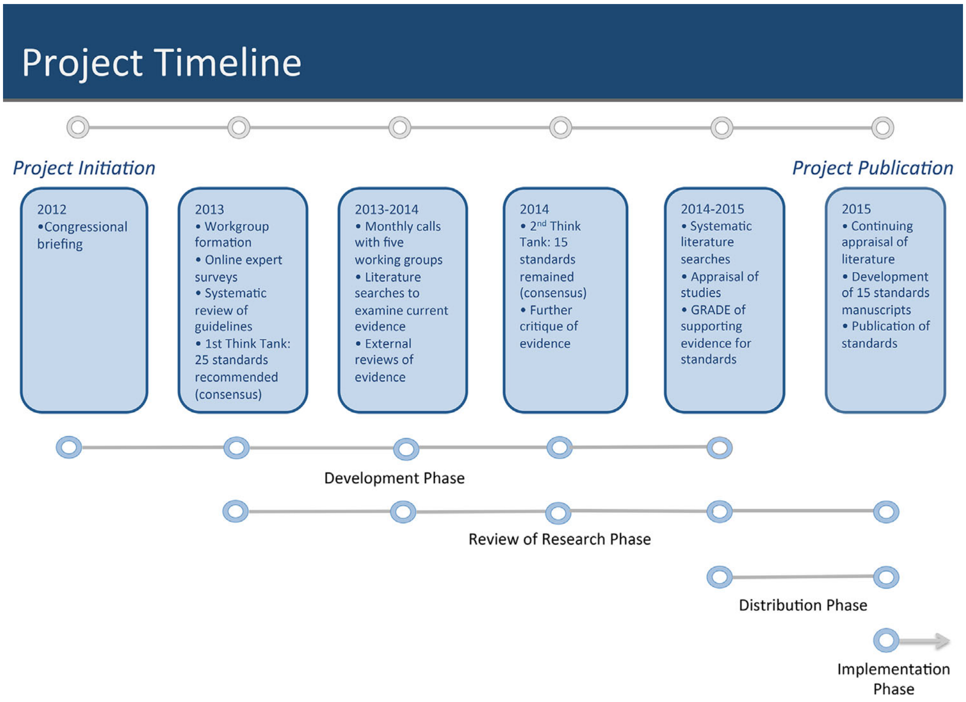

Fig. 1. Phases in the development of standards for the psychosocial care of children with cancer and associated tasks.

psycho-oncology experts across a range of clinical and geographic settings, asking the following questions: (1) What are the five most important issues that we should know about families in order to provide optimal psychosocial care?; (2) What are the most essential services/interventions that should be provided to families throughout the cancer treatment trajectory?; (3) In your setting, what do administrators need to know about psychosocial services that should (and could) be provided to all families and are not currently available or need improvement?; (4) Please list up to five challenges to developing and implementing psychosocial standards/guidelines; and (5) What are some of the most innovative and/or effective ways you or others have discussed or utilized to implement psychosocial care? Three independent psychosocial clinicians reviewed the survey data. Consensus was obtained to define five distinct critical areas wherein standards are needed for satisfactory provision of psychosocial care for children with cancer. These are as follows: (1) Assessment of Child and Family Well-Being and Emotional Functioning; (2) Neurocognitive Status; (3) Psychotherapeutic Interventions; (4) School Functioning; and (5) Communication, Documentation, and Training of Psychosocial Services.

The PSCPCC held two in-person meetings ("think tanks"), each at an annual meeting of the American Psychosocial Oncology Society (APOS) with the sponsorship of Mattie Miracle (travel and logistics) and APOS (meeting space). Between these meetings, PSCPCC held monthly teleconferences focused on organizing reviews of literature in these five areas.

The first think tank meeting occurred on February 14, 2013. In attendance were 20 experts in the field of adult and pediatric psycho-oncology from the majority of relevant professional groups (oncology, psychiatry, psychology, social work, and nursing) and four parent stakeholders. The purpose of the meeting was to determine the scope of the standards and to reach agreement about elements of essential, high-quality psychosocial care that can be implemented in all pediatric oncology settings. Using Livestrong's criteria for an Essential Element of care,[7] it was decided that each proposed standard would be evaluated for its "positive impact on quality of life for all cancer patients and their family members," and potential for utilization in a wide variety of settings. Further, each element required documented support from an existing behavioral science evidence base. Recognizing that a strong evidence base did not exist for some elements of psychosocial care, alternative sources of data that clearly described services widely utilized and are valued by a consensus of the provider community and which could be evaluated in future research were also viewed as providing an acceptable basis for inclusion of a standard of care.

During the think tank, each of the five groups reviewed qualitative data from our online survey and contributed their clinical knowledge and understanding of the supporting literature to make recommendations for elements considered "essential" for psychosocial care in their domain. This was followed by a consensus session wherein all meeting participants reviewed recommendations from the individual working groups. At the conclusion of the meeting, 25 Essential Elements for the psychosocial care of children with cancer and their families were identified.

In the year between the two think tank meetings, working groups were charged with investigating and critiquing the related professional literature to determine whether there was sufficient and compelling evidence or consensus to support each of the essential recommendations generated during the think tank. Leaders from the working groups invited additional interdisciplinary experts and stakeholders to join their groups, as needed. During the first 6 months, the working groups held monthly conference calls wherein they reviewed inclusion and exclusion criteria for their individual literature reviews; conducted systematic literature searches; and identified and defined additional clinical issues not previously noted. The working groups also documented and critiqued available evidence. Each group decided whether they had agreement about whether an explicit link existed between each recommendation and the related evidence, including the potential barriers to implementation of the standard. During the next 6 months, tables of evidence were created and the quality of the literature was rated. To avoid the risk of bias, experts in the field reviewed each other's content and informed a second review and/or revision of the standard. This process continued until no new revisions were recommended.

The Appraisal Guidelines for Research and Evaluation (AGREE II)[5] were used by each group to rate the evidence for their standard. We specifically addressed the following areas: Identification of Target Population; Essential Element, Rationale, Key Evidence, Literature Search Strategy, Organizational Barriers, Response to Barriers, and Literature Cited. Using a rating form, each working group sent their findings to non-member experts who had agreed to review the interim guidelines to determine whether the evidence supported the recommended standard (Table I). Data from the five working groups were combined into a single document that formed the basis for discussion at the second think tank meeting.

Throughout the year, there was a conscious effort to include representation from multiple relevant disciplines within the working groups. Consequently, the working groups consisted of 22 psychologists, three psychiatrists, five social workers, one advanced practice nurse, and two oncologists from the United States, Canada, and the Netherlands and five parent advocates. The working groups also represented members from numerous professional groups: American Psychosocial Oncology Society (APOS), International Psychosocial Oncology Society (IPOS), International Society of Paediatric Oncology (SIOP), Children's Oncology Group (COG), Association of Pediatric Oncology Social Workers (APOSW), Society of Pediatric Psychology (SPP, Division 54 of the American Psychological Association [APA]), Association of Pediatric Hematology/Oncology Nurses (APHON), American Academy of Child and Adolescent Psychiatry (AACAP), Association for Psychological Science (APS), and the Association of Pediatric Hematology Oncology Education Specialists (APHOES).

The second PSCPCC think tank was held on February 13, 2014, in Tampa, Florida. In attendance were 15 of the participants from the initial meeting and four additional experts with specific clinical and research expertise in areas not previously represented. Each of the 25 recommendations was further evaluated in connection to the related evidence. During this meeting, each standard was reviewed and rated by a different working group than the one that had created the standard. Working groups each included a pediatric oncologist, psychiatrist, psychologist, social worker, and a parent or survivor stakeholder. Working group members reviewed each individual standard, the corresponding evidence table, external reviews, and barriers to implementation. Standards without sufficient evidence were eliminated and those with apparent overlap were combined. A shortened list of 15 standards was developed via a consensus process with the full group during the meeting. The wording of each standard was further refined via conference calls.

For each of these final 15 standards, individual members were charged with re-reviewing the literature to assure all relevant and/or new evidence was included. PRISMA guidelines were used to conduct the systematic reviews.[8] For consistency, all authors were instructed to include studies published from March 1995 to March 2015. Search terms and inclusion criteria were specified in advance. Group members used the Critical Appraisal Skills Programme[9] checklists to assess individual study rigor, through examination of study design, analysis, and results. In standards for which there was limited evidence, expert opinion or consensus reports were included and described.

As guidelines can be inconsistent in how they rate the quality of evidence and grade the strength of their recommendations,[10] several journals now require authors submitting clinical guidelines to use a formal system known as Grading of Recommendations Assessment, Development, and Evaluation (GRADE). In summarizing the evidence for each standard, the authors were required to independently appraise their body of evidence as a whole using the GRADE system.[10] Specifically, the GRADE system classifies the quality of evidence in one of four levels—high, moderate, low, and very low. Evidence based on randomized controlled trials begins as high-quality evidence, but confidence in the evidence may be decreased for reasons, including inconsistency of results and reporting bias. Ratings reflect specific methodological considerations. For example, a case–control study may be rated as having a higher level of evidence if the treatment effect is large. The GRADE system also classifies recommendations as strong or weak. The strength of the recommendation reflects confidence that the desirable effects of an intervention outweigh the undesirable effects. For example, desirable effects of an intervention include improvement in the quality of life, reduction in the burden of treatment, reduced resource expenditures, whereas undesirable consequences include adverse effects that have a deleterious impact on quality of life, morbidity, mortality, or increase use of resources.[11] The individual papers in this special issue summarize the evidence base for the full set of consensus standards.

RESULTS

The 15 standards for psychosocial care of children with cancer and their families (Table II) represent the results of what is, to our

TABLE I. Items From the AGREE II Rating Forms Used to Rate Evidence for Each Standard

1. The strengths and limitations of the body of evidence are clearly described.
2. There is an explicit link between the recommendations and the supporting evidence.
3. The potential organizational and logistic barriers that could prevent successful implementation of this element at every pediatric cancer center have been addressed.
4. The recommendation provides advice and/or tools on how it can be put into practice.
5. The potential resource implications of applying the recommendations have been considered.
6. The literature search strategy is adequate.
7. There is enough evidence to support this Recommendation as a Standard of Care at every center where a child with cancer is treated.
8. Rate the overall quality of this recommendation.

TABLE II. Pediatric Psychosocial Standards With Quality of Evidence and Strength of Recommendations

Standard		Studies Reviewed	GRADE* Quality of Evidence	GRADE* Recommendation
1.	Youth with cancer and their family members should routinely receive systematic assessments of their psychosocial health care needs.	149	High	Strong
2.	Patients with brain tumors and others at high risk for neuropsychological deficits as a result of cancer treatment should be monitored for neuropsychological deficits during and after treatment.	129	High	Strong
3.	Long-term survivors of child and adolescent cancers should receive yearly psychosocial screening for: a) adverse educational and/or vocational progress, social and relationship difficulties; b) distress, anxiety, and depression and c) risky health behaviors.	93	Moderate to High	Strong
	Adolescent and young adult survivors and their parents should receive anticipatory guidance on the need for life-long follow-up care by the time treatment ends and repeated at each follow-up visit.**		Low to moderate	Strong
4.	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.	173	High	Strong
5.	Pediatric oncology families are at high risk for financial burden during cancer treatment with associated negative implications for quality of life and parental emotional health. Assessment of risk for financial hardship should be incorporated at time of diagnosis for all pediatric oncology families. Domains of assessment should include risk factors for financial hardship during therapy including: pre-existing low-income or financial hardship, single parent status, distance from treating center, anticipated long/intense treatment protocol, and parental employment status. Targeted referral for financial counseling and supportive resources (including both governmental and charitable supports) should be offered based on results of family assessment. Longitudinal reassessment and intervention should occur throughout the cancer treatment trajectory and into survivorship or bereavement.	24	Moderate	Strong
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.	138	Moderate	Strong
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.	23	Moderate	Strong
8.	Youth with cancer should receive developmentally appropriate preparatory information about invasive medical procedures. All youth should receive psychological intervention for invasive medical procedures.	65	Low (education) High (interventions)	Strong Strong
9.	Children and adolescents with cancer should be provided opportunities for social interaction during cancer therapy and into survivorship following careful consideration of the patients' unique characteristics, including developmental level, preferences for social interaction, and health status. The patient, parent(s) and a psychosocial team member (e.g., designee from child life, psychology, social work, or nursing) should participate in this evaluation at time of diagnosis, throughout treatment and when the patient enters survivorship; it may be helpful to include school personnel or additional providers.	64	Moderate	Strong
10.	Siblings of children with cancer are a psychosocially at-risk group and 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.	125	Moderate	Strong

TABLE II. (*Continued*)

Standard		Studies Reviewed	GRADE* Quality of Evidence	GRADE* Recommendation
11.	In collaboration with parents, school-age youth diagnosed with cancer should receive school reentry support that focuses on providing information to school personnel about the patient's diagnosis, treatment, and implications for the school environment and provides recommendations to support the child's school experience. Pediatric oncology programs should identify a team member with the requisite knowledge and skills who will coordinate communication between the patient/family, school, and the health care team.	17	Low	Strong
12.	Adherence should be assessed routinely and monitored throughout treatment.	14	Moderate	Strong
13.	Youth with cancer and their families should be introduced to palliative care concepts to reduce suffering throughout the disease process regardless of disease status. When necessary youth and families should receive developmentally appropriate end of life care [which includes bereavement care after the child's death].	73	Moderate	Strong
14.	A member of the health care team should contact the family after a child's death to assess family needs, to identify those for negative psychosocial sequelae, to continue care, and to provide resources for bereavement support.	95	Moderate	Strong
15.	Open, respectful communication and collaboration among medical and psychosocial providers, patients and families is essential to effective patient- and family-centered care. Psychosocial professionals should be integrated into pediatric oncology care settings as integral team members and be participants in-patient care rounds/meetings.	35	Moderate	Strong
	Pediatric psychosocial providers should have access to medical records and relevant reports should be shared among care team professionals, with psychological report interpretation provided by psychosocial providers to staff and patients/families for patient care planning. Psychosocial providers should follow documentation policies of the health system where they practice in accordance with ethical requirements of their profession and state/federal laws.		Low	Strong
	Pediatric psychosocial providers must have specialized training and education and be credentialed in their discipline to provide developmentally-appropriate assessment and treatment for children with cancer and their families. Experience working with children with serious, chronic illness is crucial as well as ongoing relevant supervision/peer support.		Low	Low

*Quality of Evidence: High, Moderate, Low or Very Low. Strength of Recommendation: Strong or Weak (based on GRADE quality criteria).[10,11] **Papers reviewed from January 2011–April 2015 to supplement the COG LTFU guidelines.

knowledge, the largest, comprehensive review of this large psychosocial literature. The systematic reviews conducted across the standards involved 66 authors and a total of 1,217 studies. The evidence included quantitative, qualitative, and mixed method studies. Meta-analyses, systematic reviews, and consensus reports and recommendations from relevant professional organizations provided additional evidence.

Table II also summarizes the systematic assessment of the quality of the evidence and strength of each of the recommendations. The strongest evidence (e.g., high quality) was found for four standards: Psychosocial assessment during cancer treatment[12] and in survivorship;[13] neurocognitive monitoring for children at risk;[14] psychosocial support;[15] and interventions for painful procedures.[16] Although based on a less rigorous literature, moderate evidence was found for assessment of financial issues;[17] addressing behavioral health issues of parents;[18] psychoeducation;[19] social interaction;[20] supportive services for siblings;[21] assessment and monitoring of adherence;[22] early integration of palliative care;[23] and bereavement.[24] Mixed moderate and high quality of evidence was found for survivorship.[13] and moderate-to-low quality of evidence was found for communication, documentation, and training.[25] Low-quality evidence was found for school re-entry[26] and information about invasive medical procedures.[16]

As noted earlier, the GRADE system also classifies recommendations as strong or weak, with the strength of the recommendation reflecting confidence that the desirable effects of an intervention outweigh the undesirable effects.[10,11] Even in the absence of strong research evidence, recommendations can be strong if there are multiple expert groups coming to highly consensual conclusions. Although there is variability in the quality of evidence across standards, based on the risk-benefit ratios, practice-based evidence, and consensus, strong recommendations were made for the implementation of each of the 15 standards.

DISCUSSION

The goal of this process was to provide evidence- and consensus-based standards for services considered essential for all children diagnosed with cancer and their families regardless of treatment setting. Through this rigorous process, standards of care for children with cancer and their families were developed. The standards provide a starting point for cancer centers to identify essential elements of comprehensive psychosocial care.

Although it is recommended that these standards be followed at all sites where children with cancer are treated, full implementation will occur at variable rates in different centers, with some already easily concurring and others needing changes to come up to this minimally acceptable level. Pediatric cancer programs can utilize these standards to identify their program strengths and areas where improvements and/or resources are most needed. Having the evidence underpinning each standard available in the Supplementary Evidence Tables will provide support and compelling evidence for implementation of the standards. The articles that follow discuss potential challenges with implementation and provide suggestions for reducing organizational barriers. Each article also clearly addresses areas where additional evidence-based data are needed to strengthen recommendations for a specific psychosocial intervention(s) for children with cancer and their family members.

There are limitations worth noting. First, addressing the needs of young adults with cancer was beyond the scope of this project and special issue. We recommend that similar methods be used to develop psychosocial standards of care for young adults living with cancer.

Second, implementation is likely to occur first in developed countries with established pediatric oncology programs. In low-resourced nations, psychosocial services may differ and develop in concert with the development of high-level medical care for children with cancer in these countries.

Third, the standards do not elucidate optimal care for children with cancer, only essential psychosocial care. There are valued, evidence-based treatments or interventions of known value, which go beyond a minimum standard of universal care. In some centers, it is reasonable to expect provision of services that exceed the essential standards. Fourth, the think tanks did not include child life specialists, educators, or hospital administrators, although we did engage these professionals in the working groups and as reviewers.

Next steps in this project involve the development of recommendations to improve guideline implementation and utilization. With support from Mattie Miracle and APOS, the PSCPCC group leaders have devised a strategic plan to meet yearly at the APOS annual meetings to evaluate implementation of these standards and encourage broader dissemination. New research will also be reviewed annually and the guidelines updated as needed.

CONCLUSION

A lack of standardized psychosocial standards in childhood cancer results in inconsistent access to behavioral healthcare for pediatric cancer patients and their families. The evidence-based standards presented in this special issue include strong recommendations for basic elements of psychosocial care for all children with cancer. These include both well-researched interventions proven effective in clinical trials and other consensus-based widely used interventions with less research support. These broadly implementable standards are sufficiently general to be tailored to the resources of individual sites that treat childhood cancer and to the needs of individual children and families. With evidence that such care contributes to positive quality of life outcomes of children with cancer and their family members, it is hoped that universal access to psychosocial support and intervention for patients and family members can be guaranteed for all 21st century families who face childhood cancer and its sequelae.

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SPECIAL REPORT
Interdisciplinary Collaboration in Standards of Psychosocial Care

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Many disciplines contribute significantly to the emotional well being of children with cancer and their families. Individuals who provide supportive care include child life specialists; nurses; pediatric oncologists; spiritual leaders; physical and occupational therapists; nutritionists; psychologists, social workers, and psychiatrists. Mutual respect for professional roles and communication to families of each profession's role and responsibilities enhances patient and family centered care. Team members collaborate to enhance communication to patients and parents, observe changes in behavior, support decision making, provide empathetic listening, maximize adherence, minimize distress for children and their caregivers related to illness and treatment, and optimize quality of life. They may need to initiate referrals to community psychosocial care providers to initiate assessment or treatment of patients and their families. They may also participate in follow-up or school re-entry activities.

In this special issue, psychosocial care providers refer to behavioral health professionals who assess children and adolescents with cancer and their family members and offer appropriate psychological and/or psychopharmacological treatment. Their responsibilities may include conducting diagnostic evaluation and assessments of children and family members; development and implementation of treatment plans including psychotherapeutic individual and family treatment and evidence- or guideline-based interventions; administration of neuropsychological and/or psychological testing; and participation in team communication to the patient and family. Psychosocial care providers offer consultation on psychological management of patients to members of the pediatric oncology team. Psychiatrists may also evaluate patients and sometimes, parents, provide recommendations for psychopharmacological intervention and monitor effectiveness of the medication in managing pain or distress. Essential training requirements and credentialing for the psychosocial care providers are described in the following article, in this special issue, by Patenaude, Pelletier, and Bingen.[1] Although some roles overlap, respect for the essential training of all team members and the special skills and strengths of each professional group is most beneficial to children with cancer and their family members.

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Psychosocial Assessment as a Standard of Care in Pediatric Cancer

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This paper presents the evidence for a standard of care for psychosocial assessment in pediatric cancer. An interdisciplinary group of investigators utilized EBSCO, PubMed, PsycINFO, Ovid, and Google Scholar search databases, focusing on five areas: youth/family psychosocial adjustment, family resources, family/social support, previous history/premorbid functioning, and family structure/function. Descriptive quantitative studies, systematic reviews, and meta-analyses ($n = 149$) were reviewed and evaluated using grading of recommendations, assessment development, and evaluation (GRADE) criteria. There is high quality evidence to support a strong recommendation for multifaceted, systematic assessments of psychosocial health care needs of youth with cancer and their families as a standard of care in pediatric oncology. Pediatr Blood Cancer 2015;62:S426–S459.

Key words: pediatric oncology; psychosocial assessment; screening; standards of care

INTRODUCTION

The significant psychosocial impact of a diagnosis of childhood cancer on the child and family over the course of treatment and beyond is well established and widely understood. Despite this, psychosocial care for children with cancer and their families is not provided in a systematic or consistent manner across and even within pediatric cancer programs. In a study of Children's Oncology Group institutions, about half of families were offered psychosocial services within the first 30 days after diagnosis. Only 9% of institutions used empirically supported psychosocial evaluations and less than 11% implemented empirically based treatments.[1]

Assessment of the psychosocial needs of the child and family is the first action necessary to determine subsequent steps for delivering treatments addressing psychosocial needs for youth (the terms "youth" and "children and adolescents" are used interchangeably in this paper) with cancer and their families throughout the treatment trajectory. A large literature, dating back to the 1970s, provides support for the importance of recognizing psychosocial concerns both during and after cancer treatment. The psychosocial impact of pediatric cancer on the child, mothers and fathers, siblings, and extended family has garnered the most attention in the research literature.[2] This literature is broad but consistent themes run through it. Most work has argued for the consideration of children in the context of broader systems (e.g., families, schools, healthcare settings, communities, cultures). Outcomes include both psychosocial risks for patients and family members (e.g., anxiety, depression, adjustment problems, posttraumatic stress symptoms, lack of financial resources, poverty, family problems, social isolation) and resiliencies (e.g., coping, well-being).

Support for the importance of assessment or brief screening in pediatric cancer has been articulated by a number of prominent groups and organizations, including the Institute of Medicine,[3] the American Cancer Society,[4] the National Comprehensive Cancer Network,[5] and the Association of Pediatric Oncology Social Workers.[6] Screening, usually for depression or distress, has become more common in adult oncology. The Commission on Cancer guidelines[7] require distress screening, particularly at times of highest distress (i.e., at diagnosis, family meeting with oncologist to discuss treatment, transitions off treatment) and applies to pediatric as well as adult cancer programs. This paper refers to both screening and assessment, with the latter generally involving a more comprehensive and lengthier approach than screening, which is very brief. Assessment may be thought of as the follow-up step to screening, in order to identify specific areas for treatment.

Psychosocial Standard of Care

Youth with cancer and their family members should routinely receive systematic assessments of their psychosocial health care needs.

Abbreviations: DT, distress thermometer; GRADE, grading of recommendations, assessment development and evaluation; PAT, psychosocial assessment tool; PSCPCC, psychosocial standards of care project for childhood cancer; SES, socioeconomic status

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This review was performed as part of the collaborative Psychosocial Standards of Care Project for Childhood Cancer (PSCPCC). For a full description of the methods used to develop each standard, refer to Wiener, Kazak, Noll, Patenaude, and Kupst in this Special Issue.[8] This paper provides a review of the relevant literature with the goal of identifying and summarizing the evidence for the systematic assessment of children with cancer and their families, reflecting the breadth of factors necessary to assure comprehensive care.

METHODS

This review was conducted by an interdisciplinary team from the PSCPCC. The sub-group of the PSCPCC that conducted the reviews for this standard included pediatric psychologists (n = 7), social workers (n = 1), psychiatrists (n = 1), counselors (n = 3), all with experience in pediatric cancer. One parent of a child with cancer also participated. In addition to broad representation across the United States, we included representation from the Netherlands to provide an international perspective.

Given the breadth of this standard, the study team identified distinct areas embedded in the broader content of the standard during its first meeting.[8] The group subdivided accordingly into five groups, each with 2–3 members: 1) *Youth and family psychosocial adjustment* (e.g., depression, anxiety, posttraumatic stress); 2) *Family resources* (e.g., financial, socioeconomic status [SES], language issues, barriers to care); 3) *Family and social support* (e.g., extended family, friends, community resources); 4) *Previous history/premorbid functioning* (e.g., prior stressors, behavioral, and educational problems); and 5) *Family structure and function* (e.g., family dynamics, beliefs, cultural factors).

Each group conducted the initial literature search independently for their subgroup in the spring of 2013. In March 2015, the search was monitored across groups by team members (SD, JC) under the supervision of the senior authors (AK, MJK), including updating the search to capture literature from a broader range of January 1, 1995 through March 31, 2015. The literature search utilized five databases: EBSCO, PubMed, PsycINFO, Ovid, and Google Scholar. Literature search terms included: "ped* cancer" OR "pediatric cancer" OR "childhood cancer" OR "child* cancer" OR "cancer*" AND "PAT" OR "psychosocial assessment tool" OR "ongoing assessment" OR "assessment" or "child adjustment" OR "parent adjustment" OR "family adjustment" OR "adjustment" OR "child understanding" OR "family understanding" OR "understanding" OR "PedsQL" OR "distress thermometer" OR "quality of life" OR "QOL" OR "language barrier" OR "outcome barriers" OR "barriers" OR "barriers to care" OR "psychosocial" OR "SES barrier" OR "SES" OR "healthcare disparities" OR "family resources" OR "barriers to treatment" OR "treatment barriers" "outcome barriers" OR "cancer support" OR "family support" OR "support" OR "child support" OR "peer*" OR "peer support" OR "friend" OR "commun*" OR "community" OR "church" OR "faith" OR "spirit*" OR "pre-morbid" OR "family history" OR "illness history" OR "ill* history" OR "prior loss" OR "behavioral" OR "behavior*" OR "education*" OR "previous history" OR "function*" OR "before diagnosis" OR "family" OR "family functioning" OR "functioning" OR "family structure" OR "cultural factors" OR "culture" OR "family dynamics" OR "dynamics" OR "family belief*" OR "belief." The reference sections of identified studies were hand-searched for additional studies. All studies were reviewed to assure that they included families of children 0–18 years of age. All studies were evaluated for their quality using grading of recommendations, assessment development, and evaluation (GRADE).[9]

RESULTS

A large number (n = 149) of studies/reviews was identified in the literature search. Because of the size and breadth of the literature, the original five areas of psychosocial assessment were considered separately initially (and reported separately in this section) and then combined for the Summary of Evidence Table (Table I). Supplemental Table I includes all of the studies identified and briefly summarizes each study with regard to study design, sample characteristics, main findings related to this standard, study rigor, and level of evidence.[9]

Youth and Family Psychosocial Adjustment

There is strong and highly consistent research evidence that children and parents experience increased distress, poorer quality of life, and difficulties in psychosocial functioning immediately and in the months after the diagnosis of cancer.[10] These papers (n = 29) come from mostly quantitative descriptive studies, conducted at single institutions, with some mixed methods research. These findings are strongly supported by systematic reviews (n = 3), randomized clinical trials (n = 1), longitudinal studies (n = 6), and consensus statements (n = 3).

Family Resources

Cancer care is impacted by family resources, including socioeconomic status (SES), parental education, and income. Families at socioeconomic risk experience more difficulties with respect to access to care and barriers to treatment throughout the course of care.[11–13] There are health care disparities and barriers as well with respect to language.[14] The research in this area provides rigorous and critically important justification for the importance of identifying families that are at risk for poorer outcomes related to pre-existing socioeconomic considerations (n = 38). Most studies are quantitative (n = 25) although there is also evidence from qualitative (n = 5) and mixed methods studies (n = 4). Additional support comes from longitudinal (n = 3) studies, retrospective chart reviews with large samples (n = 6), and systematic and narrative reviews (n = 5).

Family and Social Support

Perceived support from family members and others is related to psychosocial functioning and reduced distress across the course of treatment. Similar to the other areas of psychosocial assessment, the literature (n = 17 studies and reviews) consists primarily of cross sectional studies, including qualitative (n = 6) and quantitative (n = 5, plus two longitudinal) studies. Systematic (n = 4) and narrative (n = 3) reviews provide additional support. This literature documents the association of child and parent social support with other psychosocial outcomes and provides justification for helping families remain connected with important support systems during and after treatment.[15–17] The overall evidence is robust; the literature is highly consistent albeit smaller in size than the other areas.

TABLE I. Summary of Evidence—Assessing Psychosocial Needs in Children and Adolescents With Cancer and Their Families

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
Youth with cancer and their family members should routinely receive systematic assessments of their psychosocial health care needs	The diagnosis and treatment of childhood cancer has a significant impact on the emotional and behavioral functioning of children and their families, including increased distress and lower quality of life. Many well-designed studies have determined risk and protective factors that affect psychosocial outcomes. Pre-cancer stresses and psychosocial functioning are related to psychosocial functioning in children with cancer and their parents. There is some evidence that psychosocial functioning early in treatment is predictive of later functioning during and after treatment. Family factors play an important role in adaptation to childhood cancer. Family resources (SES, education, income) as well as disparities and barriers based on race/ethnicity and language are related to access to care, burden of care, adherence, relapse, and survival. Family cohesion, resources, perceived family, and social support can be protective factors in psychosocial outcomes. There is some evidence that cultural factors are related to access, utilization, communication, and adherence to treatment. These findings strongly indicate the need for early and continued screening and monitoring of factors related to psychosocial functioning of children with cancer and their families. Reliable and valid screening instruments have been developed for this population, as well as use of standard psychological measures	149 studies and reviews Systematic reviews (16), two meta-analyses, 9 narrative reviews, and 3 consensus studies have indicated consistent results. Most studies were quantitative, with qualitative and mixed method cross-sectional descriptive studies. Longitudinal (22) and comparison group (17) studies, two RCTS (one pilot) reviewed. In addition, six retrospective record surveys were conducted with large scale cohorts	High quality evidence based on the results of meta-analysis, systematic reviews, and a large number of well-designed lower level studies	Strong recommendation based on the quality of evidence as well as the balance between desirable (early screening and continued monitoring of risk and protective factors) and undesirable (lowered psychosocial functioning going undetected and untreated) effects

[1]Based on summary of evidence table for that standard; [2]Types of studies: e.g., RCT, cross-sectional, longitudinal; consensus; systematic review articles. Quality of evidence; [3]High = A, moderate = B, low, and very low = C (based on GRADE criteria) strength of recommendation; [4]Strong = 1, weak = 2 (based on GRADE quality criteria).

Previous History/Premorbid Functioning

Stressors and experiences that predate the diagnosis of cancer in a child are associated with functioning after diagnosis. This finding is well supported by the literature (n = 20 studies and reviews), with quantitative (n = 18), one systematic review, and one narrative review. Most studies are descriptive with five cohort/comparison group studies, some are longitudinal (n = 5), and there is one pilot randomized clinical trial. The strongest evidence comes from the longitudinal studies of Kupst[18] and from studies of families entering stem cell transplantation because a true "pre" evaluation can be conducted.[19,20]

Family Structure and Function

There is a substantive and cogent body of evidence to support the roles of both family structure and functioning in relation to psychosocial outcomes in pediatric cancer (n = 45 studies and reviews). Most were quantitative studies, with four qualitative and two mixed methods studies (six case comparison studies and six longitudinal studies); one meta-analysis and seven systematic reviews provided additional support. With respect to family structure, being a lone parent is associated with increased risk.[21,22] Family cohesion, resources, and perceived social support from family and community can be protective factors in psychosocial outcomes across family members, findings that are supported in narrative reviews.[23,24] There is some evidence that cultural factors are related to access, utilization, communication, and adherence to treatment.[25]

DISCUSSION

The evidence for understandable levels of psychosocial distress of patients and families is strong and highly consistent across multiple areas related to child and family functioning during childhood cancer treatment. The evidence all points in the expected directions, demonstrating that cancer is an extreme stressor for children and families and one that is often associated with distress and decreases in overall quality of life. Family structure and beliefs of the family, as well as the family's natural ability to cope and function, are similarly associated with adaptive adjustment to cancer and its treatment. The social support system for the child and family is critically important. Similar to other literatures, social isolation and poverty are risk factors for ongoing and escalating distress. In addition, a prior history of child or family dysfunction, the presence of pre-existing problems, or the presence of other stressors are also associated with less optimal psychosocial outcomes. Finally and notably, the socioeconomic status of the family, the extent to which the family experiences cancer as a financial hardship[26] and other factors associated with health disparities in pediatric cancer are all well documented and critical factors related to overall adjustment and wellbeing.

Although the data presented in this paper and in the accompanying supplemental table show the consistency of findings across study designs and methodologies, this succinct report summarizing a large body of literature precludes a detailed discussion of many of the nuances of work in this field. One of the significant strengths of this literature is its inclusion of the family voice in psychosocial assessment and care delivery. Many of the studies reviewed conceptualize the care of children with cancer from a family systems or socioecological perspective and accordingly include data from multiple members of the family (e.g., mothers, fathers, siblings). We also know, for example, that distress is elevated at diagnosis but that it tends to return to baseline levels after 3–6 months for most, but across multiple studies and reviews 10–30% of families have long-term sequelae.[27–29] Indeed, although many families cope remarkably well with cancer and its treatments, identifying those families who will continue to experience difficulties is a paramount concern, and entirely consistent with recent efforts to mandate screening in cancer programs. The research summarized in this report provides additional strong support for this effort and for the implementation of a standard of care associated with psychosocial assessment: *Youth with cancer and their family members should routinely receive systematic assessments of their psychosocial health care needs*.

Implementation of this standard can facilitate the early identification of risks and resiliencies for families and the delivery of evidence-based treatments to assist all members of the family across the continuum of cancer treatment. How can this be accomplished? Fortunately the literature in the field has shifted more recently in the direction of developing models to guide psychosocial screening and assessment and toward the validation of instruments to assure that screening can be accomplished in an evidence-based practical manner.[30] Kazak et al. conducted a systematic review,[31] which identified the psychosocial assessment tool (PAT)[29,32] and the distress thermometer (DT)[33] as two evidence-based screening approaches used in pediatric cancer.

The conceptual model underlying the PAT is the Pediatric Preventative Psychosocial Health Model[34] a three-tier model, based on a public health approach, identifying families at universal, targeted, and clinical levels of risk. These levels are determined by examining risk across multiple domains (e.g., family resources, family problems, social support, child and sibling problems, etc). Intervention approaches depend both on level of risk and also specifically identified risk areas. The PAT is a parent-report screener that takes 5–10 min to complete and is available in web-based and electronic health record forms. PAT is used currently in 50 sites in the United States and has been adapted/translated for use in a number of other countries,[29] with published reports from Canada [35] and Australia.[36] The PAT is also used in survivorship, with scores associated with survivors requiring psychological consultations.[37]

The DT is a brief screening instrument which provides a rating of distress on a 1–10 scale. The DT was used in a prospective study using multiple respondents (children, parents, and staff), highlighting the importance of each perspective in a thorough understanding of distress.[38,39]

The well-documented evidence for psychosocial risk and the availability of valid instruments for screening can minimize some of the barriers to systematic screening and assessment. This is necessary in order to implement this psychosocial standard and advance pediatric cancer care. Concerns about the time necessary for screening can be addressed by use of the brief PAT or DT. Screening that is family-centered, integrated into the health record, and that quickly flags patients and families at risk is an essential first step in accessing psychosocial care quickly and efficiently by addressing staffing concerns (e.g., who will conduct the screening). It is also important to consider the timing and frequency of screening and assessment. The evidence shows that although distress usually diminishes over the first year after a pediatric cancer diagnosis,[40–42] many disease, treatment, patient, and family factors may contribute to ongoing or even escalating distress over time,[43] including demographic and socioeconomic factors that indicate the importance of delivering early evidence-based interventions.[44]

Although the evidence is strong, it is not without its limitations. There are few randomized clinical trials in this field and a reliance on mostly single institution studies, based in the United States. It is important that screening be viewed as the first step in a process of psychosocial care delivery and that pediatric cancer centers provide adequate levels of appropriate staff to implement interventions pertinent to the identified needs, while at the same time conducting further research to identify optimal evidence-based interventions to promote adaptive functioning and quality of life in youth with cancer and their families.

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Monitoring and Assessment of Neuropsychological Outcomes as a Standard of Care in Pediatric Oncology

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Central nervous system cancers or exposure to CNS-directed therapies increase risk for neuropsychological deficits. There are no accepted guidelines for assessment of neuropsychological functioning in this population. A multifaceted literature search was conducted and relevant literature reviewed to inform the guidelines. Studies of neuropsychological outcomes are widely documented in the pediatric oncology literature. There is strong evidence of need for neuropsychological assessment, but insufficient evidence to guide the timing of assessment, nor to recommend specific interventions. Children with brain tumors and others at high risk for neuropsychological deficits should be monitored and assessed for neuropsychological deficits. Pediatr Blood Cancer 2015;62:S460–S513.

Key words: children; neuropsychology; outcomes

BACKGROUND

Pediatric cancers affecting the central nervous system (CNS) are a broad collection of diseases including brain tumors and others where therapy can directly impact brain function. The American Academy of Pediatrics (AAP) identifies nzeuropsychological follow-up as a critical component to the care of children who have CNS cancers and are cancer survivors.[1] That report, based upon guidelines from the Children's oncology group, identifies that late effect risks are proportional to the intensity of therapy and that, in general, longer treatment with higher cumulative doses of radiation, higher doses of chemotherapy, and multimodal therapies increase the risk of late effects. Neuropsychological late effects of cancer and cancer therapy may not become manifest for a number of years, requiring ongoing health surveillance.[2]

In this article, we propose two broad groups of children where neuropsychological monitoring and assessment have sufficient evidence to warrant guidelines: a) children diagnosed with brain tumors who are currently in treatment or post treatment; and b) children with cancer who receive CNS-directed therapies that are in treatment or post treatment. This latter body of literature is primarily reflective of children with acute lymphoblastic leukemia (ALL). Throughout this article, the terms monitoring and assessment are used as distinct processes. Monitoring refers to a broad range of activities intended to screen for potential neuropsychological changes, and that might be conducted by a variety of disciplines, including physicians, nursing, psychology, and other allied health professionals. Activities might include interview of the patient or parent regarding school performance, use of standardized self- or parent-report measures, or administering of screening tools such as from the NIH toolbox, the CogState battery or abbreviated neuropsychological screenings.[3–6] Assessment is used to denote a more comprehensive procedure involving several performance-based measures that would typically require a licensed psychologist.

Children with brain tumors or other cancers receiving CNS-directed therapies have both immediate and long-term sequelae that impact brain development. The child's age at the time of diagnosis as well as the type and intensity of treatments (neurosurgical procedure, radiotherapy, chemotherapy) are likely to adversely impact both cortical and subcortical pathways of children's brain function.[7–9] Growing evidence indicates that core neuropsychological processes involved in brain function that can be adversely impacted include the following: general intelligence, attention, memory, language, executive functions (e.g., inhibitory control, working memory, cognitive flexibility), neurosensory functions, perceptual processing, and processing speed. These neuropsychological functions appear to directly impact functional outcomes such as academic achievement, adaptive functioning, and psychological adaptation.

Psychosocial Standard of Care

Children with brain tumors and others at high risk for neuropsychological deficits as a result of cancer treatment should be monitored for neuropsychological deficits during and after treatment.

As part of a project to create evidence-based standards for psychosocial care of children with cancer, we systematically examined the body of literature regarding pediatric neuropsychological assessment for children with cancer and CNS-directed therapies (e.g., ALL) to identify evidence in support of a standard of care. Our secondary aim is to link studies of neuropsychological assessment with studies that inform the timing of monitoring and assessment as well as highlight interventions have been developed, utilized, and evaluated with these populations (e.g., cognitive rehabilitation and pharmacotherapy).

METHODS

This review was performed as part of the collaborative *Standards for Psychosocial Care of Children with Cancer and Their Families* effort. For a full description of the methods used to

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Abbreviations: AAP, American Academy of Pediatrics; ALL, acute lymphoblastic leukemia; CNS, central nervous system; TBI, total body irradiation

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develop each standard, refer to Wiener et al.[10] A multifaceted search strategy was implemented that included PubMed and PsychInfo that was further refined through additional author-specific searches. The search strategy included the years 1995 until 2015. Search terms included the following: pediatric brain tumor, cancer, ALL, children, neuropsychology and cancer, neurocognition and childhood cancer, cognitive function and cancer, children and cancer, and (intelligence, attention, memory, language, executive functions, neurosensory, perceptual processing, processing speed, neuropsychological, bone marrow transplantation) cognitive remediation, cognitive intervention, rehabilitation, pharmacotherapy and cognitive and cancer. Search results were inspected for inclusion criteria, including the following: peer reviewed journal, child age birth-18 years (including some follow-up studies where children were diagnosed with cancer and then seen as young adults), and meta-analyses. Resulting studies were reviewed for inclusion criteria as well as bibliographic citations that could further inform the search. Exclusion criteria included non-English language publications, book chapters, and on-line publications that were not indexed in PubMed and PsychInfo.

RESULTS

Studies of neuropsychological outcomes using individual performance-based measures are widely documented in pediatric cancer research. A Summary of Evidence Table is available as Table I. Cross-sectional studies were commonly found in the search process. These studies indicate the presence of deficiencies and deficits in children's neuropsychological function arising from individual and combination treatments for brain tumors and CNS-directed therapies. We have identified 19 studies that have prospectively examined neuropsychological functioning in children with brain tumors and CNS-directed therapies.[11–30] Several general and systematic reviews (n = 24) and meta-analyses (n = 5) provide evidence examining child neuropsychological function following treatment for a brain tumor and other cancers with CNS-directed therapies (i.e., ALL).[31–36]

Neuropsychological Outcomes

Key domains of neuropsychological assessment that were supported by the pediatric cancer literature included the following: intelligence,[7,11,14,17,19,23,32,36–73] attention,[7,24,25,32,47,48,58,62,64,71,74–82] memory,[7,13,18,19,25,36,43,44,58,64,66,67,71,82–90] language,[36,40,66,86,91] executive functions (including inhibitory control, cognitive flexibility, working memory),[9,17,24,25,50,53,67,76,77,80,82,86–89,92–103] neurosensory functioning,[36,41,44,48,61,65,76,83,104] perceptual processing,[7,54,105] and processing speed.[17,24,58,75,76,78,79,84,87,98,106–108]

Studies examining neuropsychological outcomes in survivors of brain tumor have documented that longer times from diagnosis/treatment are associated with greater deficits in global IQ, academic functioning, and specific neuropsychological domains including attention, memory, executive functions, and processing speed.[36,67] There is established evidence for children with brain tumors that child age at diagnosis and treatment impacts observed outcomes, with younger age associated with increased risk for impairment.[8,109,110] Child age and intensity of treatment have also been predictive of neuropsychological outcomes for children with ALL.[111–113] Evidence for the impact of CNS-directed therapy for bone marrow transplantation offers relatively weak evidence for interval neuropsychological change,[15,16,26–30,114–120] although the evidence points to younger children (particularly those <3 years) who receive total body irradiation (TBI) as being the subgroup at highest risk.[30]

Longitudinal studies are more rare, but have also pointed to a decline over time in global IQ and most neuropsychological indices among patients with brain tumors. Linear modeling suggests declines of 1.5–2 points per year in first 5 years post radiotherapy treatment for brain tumors, without consistent evidence of nadir or leveling off.[25,71,110,121–124]

Timing

There is insufficient evidence to guide the specific timing of comprehensive neuropsychological assessment for children with brain tumors and ALL. However, given the available evidence of long-term sequelae, clinicians should be vigilant of possible neuropsychological changes, and engage in frequent monitoring for such changes. Monitoring should occur intermittently during the acute treatment phase, and regularly at follow-up visits following completion of treatment. Monitoring should ideally include a time-efficient yet sensitive screening of neuropsychological function

TABLE I. Summary of Evidence Table—Neuropsychological Assessment

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
Children with brain tumors and others at high risk for neuropsychological deficits as a result of cancer treatment should be monitored for neuropsychological deficits during and after treatment	Empirical research for brain tumors indicates significant impairments associated with tumor and treatment	Cross-sectional; longitudinal studies; significant replication of findings	High	Strong recommendation, given the impact of disease and treatment factors on later neuropsychological functioning
	Evidence gaps: prospective research still needed to assess long-term neuropsychological deficits with other malignancies	Large scale follow-up studies; clinical trials group consensus	Quality of evidence given consistent findings from numerous well-designed studies	

[1]Based on summary of evidence table for that standard; [2]Types of studies: e.g., RCT, cross-sectional, longitudinal; consensus; systematic review articles; [3]Quality of evidence: high = A, moderate = B, low and very low = C (based on GRADE criteria); [4]Strength of recommendation: strong = 1, weak = 2 (based on GRADE quality criteria).

with standardized measures. Questioning of the patient and/or parent regarding any perceived changes, such as declining school performance, memory loss, or difficulty with attention and concentration is critical, especially if screening of neuropsychological function is not feasible. Referral for a comprehensive neuropsychological assessment should be considered when there is evidence of any such changes. The rate of decline documented in the literature for children with brain tumors appears sufficient to indicate that a comprehensive neuropsychological assessment 2–3 years from the time of diagnosis would identify functional deficits, though this timing is not optimal for reintegration planning for school aged children. Thus, consideration should be given to obtaining an assessment upon completion of treatment, in order to facilitate school entry/re-entry, and to obtain a baseline assessment that provides a context for appreciation of any subsequent changes. Completion of treatment will vary by disease and the specific treatment protocol. This will require the attention of the care team to identify when this point occurs for a child so that a referral can be made in a timely manner. Follow-up assessment would then be recommended at 2–3-year intervals, or in the events of suspected cognitive changes, and statistically meaningful changes in a child's neuropsychological profile.

Interventions

A large multisite study of 161 total survivors, along with several studies employing a small number of survivors of brain tumor and/or ALL *randomized* to a cognitive remediation arm showed positive benefits on parent-report measures and selected objective tests compared to a control condition.[125] However, the trial did not demonstrate improvement in the intended outcome of attention functioning. Cognitive intervention programs have thus far included computerized "game-like" activities, various cognitive behavioral and learning strategies, and prescriptive activities focused on a single academic area such as mathematics.[125–136] A pilot parent training directed at promoting effective learning strategies within the home has shown improved child academic performance, the study's primary outcome, and therefore has promise for educational outcomes.[137]

Pharmacotherapy approaches to improve attention dysfunction have included psychostimulant medications (e.g., methylphenidate) and acetylcholinesterase inhibitors (e.g., donepezil).[35,138] Studies involving survivors of brain tumor and ALL demonstrate significant improvement on parent report ratings. Higher baseline IQ, males, and older age at treatment predicted better response to 0.60 mg/kg dose of methylphenidate in the cohort.[20,106,138–143]

Barriers to Care

There are significant clinical care barriers to implementation of the proposed guideline, notably the availability of pediatric neuropsychology specialists, the costs of comprehensive neuropsychological assessment, and inconsistent reimbursement for such services by third party payors. In addition, there is a need to increase capacity within pediatric caner teams through training in screening procedures for neuropsychological functioning that could be employed. Table II provides a brief overview of barriers that have been identified and potential solutions.

DISCUSSION

Assessment of child neuropsychological functioning is supported by empirical findings demonstrating impairments for survivors. Both acute and long-term neuropsychological sequelae are consistently observed among children with brain tumors. In contrast, evidence suggests that neuropsychological impairments for children with ALL are less evident during the period of active treatment, yet long-term sequelae do emerge, albeit at a less severe level compared to children who have brain tumors. Thus to attempt to capture dynamic brain function changes that occur, the currently available evidence supports regular monitoring of neuropsychological functioning integrated with routine clinical care during and particularly after treatment. Monitoring can serve as a valuable tool for early identification and subsequent referral for more comprehensive assessment procedures. In addition to monitoring neuropsychological functioning, we suggest that developmental delays and learning difficulties be included in the monitoring procedures. Where our evidence lags is in determining the specific timing for neuropsychological assessment, and identifying which types of interventions should be implemented. Moreover, a substantial gap exists regarding evidence-based monitoring strategies with known sensitivity/

TABLE II. Barriers and Response to Barriers for Neuropsychological Assessment of Children With Brain Tumors and CNS-Directed Therapies

Barrier	Response to barrier
Multidisciplinary screening with expertise in assessment of neuropsychological functioning and pediatric cancer late effects	1. Training of pediatric cancer team in screening procedures that can identify children with risk factors or acute mental status changes
Pediatric neuropsychologist not available at the center	1. Programs can prioritize this service as an essential part of acute and late effects care 2. Programs can create a systematic way that neuropsychological monitoring and assessment will be provided in the clinical setting 3. Partnerships can be developed with existing pediatric neuropsychological providers from other clinical services (e.g., pediatric neurology)
Reimbursement for neuropsychological monitoring and assessment services vary by state, with some states requiring a mental health diagnosis for reimbursement	1. Children's hospitals need to ensure support for improving billing mechanism and efficiency for reimbursement for clinical services 2. Work with national professional and consumer groups to lobby for mandated coverage for these indicated services

specificity. Yet there is reason to consider that monitoring of neuropsychological functioning could best be completed by a psychologist with expertise in cancer effects and late effects of treatment. The timing for monitoring is debatable, with little evidence to guide this element of care. Nonetheless, we suggest that monitoring begins several months after diagnosis, and using clinical judgment, as necessary during treatment to the time when a comprehensive neuropsychological evaluation is indicated.

A research gap in need of improvement is the development of standardized methods and measures for screening/monitoring neuropsychological functioning in children with cancer. While processes and measures exist for developmental surveillance in pediatrics,[144] no disease-specific childhood cancer tools exist at present. However, measures with validated sensitivity and specificity employed in general pediatrics may be useful for children up to early school age, though are not without some controversy.[145–147] For the larger population of children and adolescents, there are no similar measurement systems for neuropsychological function, though screening and monitoring may be completed with broad based behavioral measures.[96,148–150]

The interventions reviewed typically were provided within the context of a research study, thus ensuring fidelity with the intervention procedures, but with most lacking the practicality of real world interventions. Thus, they are not currently routinely available outside the context of research protocols.

Limitations within this review include no attempt to statistically examine the impact of child age and other influential features (e.g., socioeconomic status) upon groups of children that contributed to the studies presented. Moreover, studies are limited in the information that is provided regarding the content of the interventions that may have occurred for the participants.

In summary, the proposed standard, derived from the existing body of research, serves to provide improvements in care for children with cancer, as well as guide the future of pediatric research in this area. There are barriers to overcome; yet with providing a framework for assessment that is linked to intervention, we can provide an impetus to improve the life of children with cancers affecting the CNS.

CONCLUSION

Children with brain tumors and others at high risk for neuropsychological deficits as a result of CNS-directed cancer treatment should be monitored and assessed for neuropsychological deficits during and after treatment. Domains for neuropsychological monitoring include procedures for mental status changes as a result of treatments received. Domains of neuropsychological assessment via culturally appropriate assessments administered to the youth with cancer need to include the following: intelligence, attention, memory, language, executive function, neurosensory functioning, perceptual processing, and processing speed. Academic achievement should be included as a functional outcome.

The timeline for neuropsychological assessment for children with a pediatric brain tumor and child receiving CNS-directed therapy begins with the multidisciplinary screening at the time of diagnosis and at times of child acute mental status change. A comprehensive assessment should ideally occur after treatment has ended, followed by a re-assessment at 2–3 years after treatment or when monitoring is suggestive of significant neuropsychological or functional changes.

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Psychosocial Follow-Up in Survivorship as a Standard of Care in Pediatric Oncology

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Childhood cancer survivors (CCS) have a high risk of medical late effects following cancer therapy. Psychosocial late effects are less often recognized. Many CCS do not receive long-term follow-up (LTFU) care, and those who do are rarely screened for psychosocial late effects. An interdisciplinary team conducted a systematic review of qualitative and quantitative studies to assess social, educational, vocational, psychological, and behavioral outcomes along with factors related to receipt of LTFU care. We propose that psychosocial screening be considered a standard of care in long-term follow-up care and that education be provided to promote the use LTFU care starting early in the treatment trajectory. Pediatr Blood Cancer 2015;62:S531-S537 © 2015 Wiley Periodicals, Inc.

Key words: long-term follow-up care; survivorship; childhood cancer; psychosocial

INTRODUCTION

Rapid improvement in treatment for child and adolescent cancers has led to greatly increased survival with a growing population of over 300,000 long-term childhood cancer survivors (CCS) in the United States.[1] Sixty percent of CCS report medical morbidities.[2–4] Long-term follow-up (LTFU) care is recommended and involves systematic assessment to detect and treat health problems related to childhood cancer and its treatment.[5] However, late effects of childhood cancer are not limited to physical health problems. Previous studies describe CCS as having psychosocial difficulties impacting academic achievement, employment, social, and family relationships,[6–15] affective distress,[16–18] posttraumatic stress symptoms,[19–21] suicidality,[22] and tobacco and heavy alcohol use.[23,24] Few survivors receive dedicated LTFU care,[25–29] and even fewer receive a comprehensive psychosocial assessment.[30]

The Children's Oncology Group (COG) has outlined LTFU guidelines that provide recommendations for regular surveillance and care for those 2+ years from end of treatment based on specific treatment exposures.[31,32] The COG LTFU guidelines are updated periodically using systematic reviews of published scientific literature. While guidelines primarily pertain to physical late effects, recommendations also include screening for psychological, social, and behavioral difficulties.[31] This manuscript builds on the COG recommendations and provides an additional, updated review of the recent literature focusing on the psychosocial sequelae from childhood cancer and the psychosocial factors influencing uptake of LTFU care. The goal of the review is to identify and summarize the evidence so that screening can target the most relevant psychosocial domains. Screening for distress among cancer survivors has been recommended starting at cancer diagnosis and at appropriate intervals thereafter.[33–35] The provision of screening supports patients' wishes since psychological well-being has been described by CCS as more important than physical quality of life (QoL) dimensions.[36]

Psychosocial Standard of Care

- Long-term survivors of child and adolescent cancers should receive yearly psychosocial screening for: (1a) adverse educational and/or vocational progress, social and relationship difficulties; (1b) distress, anxiety, and depression; and (1c) risky health behaviors.
- (2) Adolescent and young adult survivors and their parents should receive anticipatory guidance on the need for life-long follow-up care by the time treatment ends, and repeated at each follow-up visit.

METHODS

This review was performed as part of the collaborative *Standards for Psychosocial Care of Children with Cancer and Their Families* effort. For a full description of the methods used

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Abbreviations: CCS, childhood cancer survivors; CNS, central nervous system; COG, Children's Oncology Group; HRQoL, health-related quality of life; LTFU, long-term follow-up; PTSD, posttraumatic stress disorder; QoL, quality of life

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to develop each standard, please refer to Wiener et al.[37] Search terms specific to this manuscript included "education," "vocation," "depression," "anxiety," "tobacco," "alcohol," or "long-term follow-up." (The full list of search terms is available in Supplemental Table I.) The search identified 813 articles of which 93 met inclusion criteria and were reviewed. (See Supplemental Figure I, PRISMA.) These articles included six systematic reviews, three narrative reviews, one case-control, 67 cross-sectional/cohort or retrospective chart reviews, 10 qualitative and four mixed methods studies, one opinion piece, and one randomized trial. Studies were published January 2011 to April 2015, supplementing the COG LTFU guidelines, Version 4.[31] Inclusion criteria were: CCS (i) diagnosed between ages 0–18; (ii) completed treatment for initial cancer diagnosis; and (iii) psychosocial issues were key outcome measures. Studies were excluded when CCS data were aggregated with adult cancer survivors, main outcomes were focused on measurement validation or interventions, except where unique data on psychosocial factors was provided. Additional articles were identified through references in the included articles, and by group consensus. Standards were developed following a lengthy procedure as outlined in Wiener et al.[37] adhering to suggested methods for guideline development in the existing literature.[38,39] The goal of the paper was to document the type and extent of psychosocial issues in CCS in order to develop recommendations that can be applied across all cancer treatment centers and begin the process of addressing CCS psychosocial issues.

The study team includes epidemiologists (EAL and ARR), social workers (FP and BJ), a psychologist (LAS), an oncologist (ARR), and a stakeholder (EAL). A nurse and a social worker with CCS specialization externally reviewed drafts of the manuscript.

RESULTS

Table I summarizes findings from the literature review. In general, studies describe most CCS as well-adjusted; however, studies have described poorer psychosocial outcomes compared with the controls.

Social, Academic, and Vocational Difficulties

CCS may be at risk for social and relationship difficulties. For example, CCS who participated in the 2009 U.S. Behavioral Risk Factor Surveillance Study (BRFSS) were significantly more likely to report poor social support compared with their peers.[40] Reports from the Italian, Swiss, and U.S. Childhood Cancer Survivorship Studies (CCSS) described lower marriage rates among CCS compared with the population controls and data from the U.S. cohort suggested CCS have poorer sexual health.[13–15,41]

Educational and vocational disadvantages are also reported in CCS. Although some CCS report greater school satisfaction than controls,[42] CCS generally have lower educational attainment.[15,43–45] Likewise, they are less often in high skilled managerial or professional positions, less likely to work full-time, receive lower incomes than their gender-matched siblings,[46] and are more likely to be unemployed.[15,47] Survivors of Wilms tumors are slightly less likely to go to college or obtain employment.[43] Brain tumor survivors are at risk for poor vocational outcomes;[47] however, special education programs can minimize these disparities.[15] Hence, early detection and referral for services has potential to improve patient outcomes. Risk factors for social, academic, and vocational difficulties include diagnosis or treatment for central nervous system (CNS) tumors, premorbid learning or emotional difficulties, low income or education, hematopoietic cell transplant, and younger age at diagnosis.[31]

Mental Health Concerns

Systematic and narrative reviews describe CCS as experiencing lower psychological well-being, greater anxiety, more problem behaviors, and more PTSS.[48] Brain tumor survivors report greater depression, anxiety, suicidal ideation, and behavioral problems.[49] Studies of acute lymphoblastic leukemia survivors report higher risk for adverse psychological outcomes such as depression and somatic distress.[15]

Large, high-quality studies including the CCSS from the U.S. and Switzerland, the U.S. BRFSS, and the Danish Cohort study provide evidence of greater mental health distress,[50] greater utilization of mental health care,[47,51] greater risk for neurodevelopmental, emotional, and behavioral disorders,[52] more psychoactive medication use,[53] suicidal ideation,[54] and higher rate of posttraumatic stress in CCS compared with sibling controls.[55] In a study comparing CCS to healthy peers, CCS reported more days per month of poor mental health.[40] A longitudinal study noted worsening physical health predicted greater depression, anxiety, and somatization.[56] Some large studies report no differences in mental health outcomes between CCS and siblings,[57,58] and no differences in anxiety and depression scores between CCS and population controls.[59] Negative outcomes are generally associated with a diagnosis or treatment for CNS tumors, premorbid learning or emotional difficulties, perceived poor health, female gender, and low socioeconomic status.[31]

Risky Health Behaviors

Compared with a representative sample of matched peers, smoking is more common among adult CCS and use of smokeless tobacco is more common among a sub-sample non-white men aged 35–49.[40,60] As a whole, CCS are less likely to engage in smokeless and dual tobacco use (smokeless and combustible).[60] Past month and binge drinking is not significantly different between adult survivors and controls.[40] In adolescent CCS, tobacco and alcohol use is comparable to siblings though current beer/wine consumption, binge drinking, and smokeless tobacco use in adolescent CCS.[61] Lower household income or education and older age at diagnosis increase risk for risky health behaviors.[31]

Psychosocial Associates of Engagement in Long-Term Follow-Up Care

Our search methods did not identify evidence-based practices to promote ongoing LTFU care for CCS or youth transition to LTFU care in the adult system. Patient-provider communication about health risks and follow-up care may be important to sustain engagement in care and uptake of recommended screening.[62] Cancer-related anxiety and perceived poor health status relates to the likelihood of

TABLE I. Psychosocial Follow-Up in Survivorship-Summary of Literature

Standard	Evidence summary	Methodology	Quality of evidence	Strength of recommendation
1. Long-term survivors of child and adolescent cancers should receive yearly psychosocial screening for:				
(a) Adverse educational and/or vocational progress, social and relationship difficulties;	Subsets have impaired social relationships, lower educational and vocational attainment, and impaired QoL due to psychological distress, medical late effects, or financial hardship. Multiple studies highlight identifiable and possible modifiable risk factors for poor QoL in CCS.	Cross-sectional survey-based, descriptive, and qualitative studies were most common; several large survivorship cohort studies included. Few systematic reviews of descriptive studies; no experimental or quasi-experimental studies. Broad range of selected variables examined QoL.	Moderate: consistent findings from lower level evidence studies.	Strong: Recommendation applies to most patients in most circumstances, low risk and high benefit associated with guideline with potential health benefit for CCS and family. Further research needed to increase confidence in the estimate of effect and to inform future interventions.
(b) Distress, anxiety, and depression;	Subsets of CCS are at higher risk for poor mental health outcomes, especially survivors of CNS tumors.	Systematic reviews, cross-sectional survey-based, cohort, qualitative, and mixed methods studies. Many large survivorship cohort studies with controls are included of descriptive studies.	High: consistent findings from multiple studies with large cohorts. More longitudinal studies are needed.	Strong: Recommendation applies to all survivors, low risk and high benefit associated with guideline and potential health benefit for CCS. Future longitudinal research could better identify trajectory of distress and critical opportunities for intervention.
(c) Risky health behaviors.	Heavy alcohol use, smoking, smokeless tobacco and dual tobacco use are similar or lower in CCS compared with peers or national norms. Some of the most medically vulnerable groups smoke and drink similarly to peers.	Five of the seven studies were cross sectional, several with large samples and controls. One case-control design and one systematic review (COG) citing additional studies with strong methodology.	High-moderate quality of evidence. Studies identified sub-groups at risk.	Strong: Recommendation applies to all survivors. Low risk associated with guideline, high potential health benefit given possible synergistic health risks for CCS. Brief screeners exist but future research needed to test them in CCS population.
2. Adolescent and young adult survivors and their parents should receive anticipatory guidance on the need for life-long follow-up care by the time treatment ends, and repeated at each follow-up visit.	Psychosocial variables impact uptake of follow-up care and readiness to transition to adult care. These include patient knowledge/perception of late effects, vulnerability, motivation to pursue follow-up care, and disease self-management/self-efficacy.	Cross-sectional cohort, qualitative (focus groups and interviews), opinion	Low to moderate: evidence for critical outcomes, from observational and cross-sectional studies.	Strong: Recommendation applies to most survivors. Future research should assess the impact of transition readiness and receipt of LTFU care on long-term outcomes of CCS.

CCS, childhood cancer survivors; PTSD, post-traumatic stress disorder; CNS, central nervous system; HRQoL, health related; COG, Children's Oncology Group; LTFU, long-term follow-up.

having conversations about risk-based care and screening.[62] Hypothesized psychosocial factors that impact uptake of follow-up care and readiness to transition to adult care include patient knowledge/perception of late-effects, feelings of health vulnerability, motivation to pursue follow-up care, and disease self-management.[63–66] Cancer identity and emotional components such as fear, gratitude, and gaining perspective positively influenced likelihood of transition to adult care.[67]

DISCUSSION

While results from the systematic review show that many CCS are able to adjust and move beyond their cancer experience, psychosocial difficulties are also widely described. Findings from the present systematic review are consistent with previous studies. Our review supported prior reports that CCS have lower educational and vocational attainment compared with the controls.[2] Screening for needed educational support during and after cancer treatment may minimize this disparity.[15] A pattern of greater psychosocial distress,[2,17,68,69] (but not greater psychopathology [70]) in CCS compared with controls was also supported in this review.

Reports that PTSD is more common among CCS may be partially explained by methodological differences.[71–73] For instance, lower rates of PTSD are reported when authors employ stricter criteria for diagnosis, or direct the focus on the cancer as the sole traumatic event.[71,74] Full coverage of the PTSD debate falls outside the scope of the present review. However, because considerable debate remains in the field about whether there is additional risk for PTSD among CCS, the authors have not included such screening in the proposed standards. As more definitive studies clarify the traumatic impact of childhood cancer, screening recommendations should be revisited. Previous studies described tobacco or heavy alcohol use as comparable or slightly lower among adult and adolescent CCS compared to peers or siblings.[23,24,75–77] Unfortunately, tobacco and heavy alcohol use is not lower in the most medically vulnerable CCS.

To combat underrecognition and undertreatment of psychological problems in CCS, a first step involves systematic screening to accurately identify those who most need support.[31,35, 78–80] To be effective, screening must be paired with referral for support, education, and treatment as appropriate. Evidence-based interventions to address psychosocial late effects have been described elsewhere.[81,82] CCS with risky health behaviors rarely receive risk-based medical care.[83]

A recent systematic review described psychosocial screening tools for CCS such as those to assess overall distress,[86] depression, anxiety, and suicidality.[84,87–90] Strong recommendations exist for the use of brief screening tools to identify substance use in primary care settings for healthy adults and adolescents.[93,95] Use of these tools has been shown to be effective in reducing problematic drinking and tobacco use when combined with brief interventions in healthy populations.[76,96,98–100] It is recommended that substance use screening occur in the LTFU setting, in addition to primary care, given synergistic health risks for CCS related to treatment exposures. Providers of LTFU care are likely more attuned to these specific health risks. Delivery of interventions that employ survivor focused counseling have been shown to be more effective in reducing smoking among CCS though substance use treatment remains challenging for survivors, as for other populations.[101,102]

With the development of more effective treatments the population of CCS has grown and so too has the number of survivors suffering from psychosocial sequelae. Psychosocial screening in LTFU settings is effective in identifying distress and CCS report minimal burden and high acceptance.[103, 104] In the absence of data on the most effective screening schedule, practical considerations influence the recommendation that screening should be administered according to existing COG LTFU schedules for care (usually yearly based on exposures) and performed for all survivors, regardless of diagnosis or exposure.[31] CCS who consistently screen negative for substance use could receive reduced screening over time consistent with healthy population data showing declining initiation of substance use with age.[105] Larger studies are needed to describe the implementation and adequacy of specific psychosocial screening instruments in a variety of LTFU care setting [84] and thus future research should identify appropriate, sensitive, and acceptable brief instruments for CCS.

Some authors have questioned the utility of screening and suggested that energies could be put to more productive use in treating distress.[106–110] Screening may improve patient-provider communication,[111,112] but there is not clear consistent evidence that it improves survivor well-being.[113–116] Questions remain on what should be screened, how screening should be implemented (on-line, in-person, nurse vs. doctor), whether to assess felt need or contextual need, and what to do with the results.[117] The addition of information on the context of the distress may reveal need for concrete support rather than psychosocial support. Certainly, screening is unlikely to be effective unless is leads to a response, whether that is further clinical assessment, treatment, or just an acknowledgement of the distress. More research is needed to discern the best methods for screening.[117]

Many survivors are not worried about their health,[118] underestimate their health risks,[63,64,119] know few details of their treatment history,[119] and few engage in detailed discussions about their cancer history with a provider or receive regular cancer specific follow-up care [118] creating a barrier to psychosocial screening.[31,120] Less than 20% of adult CCS reported follow-up care that included advice about risk reduction, or screening tests for physical late effects.[26,121] Patient and parent education that starts early in the treatment trajectory and emphasizes the importance of LTFU is needed. Furthermore, primary care providers and other adult health care providers may need focused education about both the physical and psychosocial sequelae of childhood cancer, and published guidelines must emphasize both realms of whole-patient care.[122]

Certain barriers have been identified and include lack of financial or personnel resources, lack of health insurance reimbursement or psychosocial providers, or low motivation for screening. These barriers may be addressed by use of brief standardized instruments, development of hospital and community referrals, and provider education. Future research must focus on the selection, timing, and efficacy of such screening tools in CCS.[123]

A number of limitations exist in the manuscript. We chose to target specific negative psychosocial late effects most often identified in the literature. We did not include studies assessing global or abstract constructs such as QoL, neurological, or cognitive late effects (addressed within this special issue [124]), or positive outcomes, though promoting such outcomes may contribute to survivor well-being and QoL.[125,126] A full review of assessment instruments and effective interventions for each outcome was beyond the scope of the project and is reviewed elsewhere.[81,82] Finally, given the considerable presence of psychological late effects in CCS, the authors recommend screening while recognizing that future research is needed to determine whether screening lessens this morbidity.

In summary, the burden of chronic health problems among CCS is profound in both prevalence and severity.[127] Receipt of LTFU care that addresses both physical and mental health sequelae of childhood cancer is critical to supporting longevity and well-being.[128] In the face of late medical health effects, some of which are not reversible, health care providers can still attend to the human cost of cancer by asking the patient about his or her experience and listening to the answer.[128]

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SUPPLEMENTARY INFORMATION

SUPPLEMENTAL TABLE I. Psychosocial Follow-up in Survivorship–Methods and Search Terms

The search strategy for this manuscript used electronic databases including PubMed, PsychINFO, Google Scholar, and Academic Search Complete.

Baseline search terms used: Childhood Cancer, Pediatric Cancer, Cancer Survivor, Survivorship, Oncology, Cancer, Intrathecal chemotherapy, Methotrexate, Radiotherapy, Cranial radiation, Antineoplastic, Neoplasms, Leukemia, CNS tumors, and Brain tumors.

Search terms specific to psychosocial issues for long-term survivors include: Depression/Global Distress included baseline terms and: depression, low mood, flat affect, sad, sadness, mood disorder, guilt, uncertainty, isolation, poor self-esteem, poor body image, suicidal ideation, grief, bereavement, distress, global distress, mental health, emotional functioning, psychological outcomes, psychosocial outcomes, psychosocial late effects, somatic complaints, somatization, and pain.

Search terms used for the Anxiety category included baseline terms and: anxiety, stress, post traumatic stress symptoms, post traumatic stress disorder, psychological distress, worry, fear, and phobia.

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Psychosocial Interventions and Therapeutic Support as a Standard of Care in Pediatric Oncology

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Research indicates that a subset of youths with childhood cancer and their parents will experience significant psychological distress throughout the course of their illness. Importantly, the existing literature indicates that psychosocial support is beneficial in decreasing symptoms of distress in these families. The aim of the current review is to determine the extent of the evidence to support a standard of psychosocial care for children and their families throughout the cancer trajectory; thus, we examined the research related to psychosocial outcomes in youth with cancer and their parents. Pediatr Blood Cancer 2015;62:S585–S618.

Key words: cancer; intervention; pediatric; psychosocial; standard of care; support

INTRODUCTION

Position statements from professional organizations, guidelines, and journal articles emphasize that pediatric cancer patients and their families need access to psychosocial support and in tervention throughout the illness trajectory, including at the time of diagnosis and through survivorship. The type of support and interventions provided may vary depending on resource and staff availability, but should be provided by an individual with training and expertise in child development, emotional adjustment to illness, psychological and psychiatric syndromes, family systems, and/or concrete resources the family may need. Refer to Patenaude et al.[1] in this edition for further information about training for professionals.

Although pediatric cancer survival rates have increased in the past four decades, these treatments place patients and families at increased risk for coping difficulties throughout the treatment course, survivorship, and at end of life. Although many childhood cancer patients and their families cope effectively during the illness course,[2,3] it is quite clear that a significant subset of patients and their parents will indeed experience significant clinical levels of psychological distress at the time of diagnosis, during the course of treatment, and after treatment ends.[2,4] Numerous studies have highlighted the importance of providing access to resources at the time of diagnosis as the majority of patients and parents directly benefit from support from a psychosocial provider.[5–7] As such, access to and provision of psychosocial services is clearly necessary throughout the cancer experience from the time of diagnosis throughout survivorship.

METHODS

This review was performed as part of the collaborative *Standards for Psychosocial Care of Children with Cancer and Their Families* effort. Articles were reviewed using GRADE methodology. For a full description of the methods used to develop each standard, refer to Wiener et al.[8] in this special edition. Computerized literature searches of OVID, PSYCINFO, and PUBMED were performed. Searches were limited to articles published in the English language from March 1, 1995 to March 1, 2015. Search terms included combined neoplasm terms with child and follow-up-related terms and MeSH headings. See Supplemental Table SI for a full list of search terms used. Search results were supplemented with hand searching of the bibliographies of systematic reviews and selected seminal articles, and contributions from personal files.

Psychosocial Standard of Care

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.

Consultation With Other Experts/Groups

The study team comprised three pediatric clinical psychologists and a child and adolescent psychiatrist who jointly reviewed all articles. As part of the development of standards process, expert opinion of this work was sent to various individuals for their review and commentary; these individuals included pediatric oncologists, pediatric psychologists, and child psychiatrists. Minor changes, such as reviewing additional psychosocial literature and making the scope of the standard broader, were made following feedback from reviewers.

Abbreviations: ALL, acute lymphoblastic leukemia; CBT, cognitive behavioral therapy; CNS, central nervous system; GRADE, grading of recommendations assessment, development, and evaluation; PSST, problem solving skills training; PTSD, post-traumatic stress disorder; PTSS, post-traumatic stress symptoms; RCT, randomized controlled trial

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RESULTS

The search strategy identified 173 peer-reviewed papers, including 19 randomized controlled trials, 109 quantitative studies, 8 qualitative studies, 36 systematic reviews of the literature, and one consensus report evaluating various aspects of psychosocial interventions. See Supplemental Table SII for a description of articles reviewed. This review resulted in three key findings: (i) youth diagnosed with cancer are at risk for negative adjustment difficulties; (ii) parents are at risk for negative adjustment outcomes; and (iii) support and intervention provides clear benefit in helping youth and parents adjust. These findings are discussed in more detail below.

Child Adjustment Outcomes

For well over four decades, researchers have examined the psychosocial functioning of children with cancer and their families.[9,10] Numerous studies document that children diagnosed with cancer experience various forms of distress associated with the cancer experience.[11] Cancer and its associated treatment can have a serious social and emotional impact on the child and the parent.[12] Much data indicate that child functioning is closely associated and often dependent on parent and family functioning.[13] The physical symptoms of cancer and associated treatment can have serious social and emotional consequences for the child and parent.[14] Further, children treated for acute lymphoblastic leukemia (ALL) or lymphoma report poorer emotional functioning, cognitive skills, autonomy, and family interaction than do children treated for non-CNS solid tumors.[15] For children whose disease or treatment directly involves the CNS, the risk of developing social isolation and peer difficulties appears much greater than children whose disease or treatment does not, and these children are less likely to be endorsed as friends by their peers.[16]

Specifically, children with cancer are at relative risk of experiencing anxiety, inhibited and withdrawn behavior, behavior problems, intense stress, depression, post-traumatic stress disorder (PTSD), academic difficulties, peer relationship difficulties, and worries about the future in relation to career and relationships.[17–20] Importantly, childhood cancer survivors have been identified as being particularly at risk for anxiety and somatic concerns. For more information about survivorship, refer to Lown et al.[16] in this special edition.[21] It should be noted, however, that some studies have not identified children to be at higher risk of experiencing post-traumatic stress (PTSS).[22]

Parent Adjustment Outcomes

Caregivers of children with cancer are also at risk for a variety of poor adjustment outcomes. Approximately 40% of caregivers of children with cancer meet criteria for acute distress disorder, a possible precursor to PTSD, within the first 2 weeks following their child's diagnosis; these symptoms appear to continue through the course of their child's illness.[11,23] Subclinical levels of symptoms of PTSS have also been documented in both mothers and fathers of childhood cancer survivors, and mothers of children newly diagnosed with cancer report significantly higher levels of PTSS than parents of long-term cancer survivors.[24] Other research has found that parents can experience moderate to severe post-traumatic stress at time of diagnosis in comparison to caregivers of long-term survivors.[25] Thus, parents of children with cancer appear to be at risk for experiencing distress both during the early diagnosis phase and after their child's cancer treatment ends. For more information related to psychosocial support for parents, refer to Kearney et al.[26] in this special edition.

General psychological distress in parents of children with pediatric cancer has also been documented. Rates of global psychological distress (e.g., a combination of depressive and anxious symptoms) have been estimated to be as high as 51%.[27] Numerous studies indicate that parents experience high levels of general distress during the initial weeks of treatment,[28] and that this distress remains stable or moderately decreases through the course of treatment.[29] Other studies would similarly indicate that both fathers and mothers have self-reported levels of psychological distress above normative means[30] with large sample studies demonstrating that mothers in particular evidence moderate levels of distress (Table I).[6,7,31]

In sum, both children and parents are at risk for poor adjustment outcomes throughout the illness course. The many other psychosocial challenges involved in pediatric cancer clearly warrant intervention to facilitate positive psychosocial adjustment.[32,33]

Psychosocial Interventions

Due to challenges associated with a diagnosis of childhood cancer and subsequent treatment, the development and implementation of effective psychosocial support resources is of the utmost importance. Such psychosocial support can be tailored to pediatric patients and their families as they face the challenges of cancer treatment.[34] Although little research has been conducted to assess practice patterns, traditional individual therapy appears to be broadly utilized in pediatric cancer centers. Individual therapy may take many forms, but most often, it appears to involve crisis intervention approaches or supportive therapy, especially in the time period following the initial diagnosis.[35] Traditional marital therapy and medical family therapy may be utilized as well.[36,37]

Efforts have been made to establish the efficacy of specific psychosocial interventions within pediatric cancer populations. Traditional cognitive behavioral therapy (CBT) approaches have witnessed empirical examination in pediatric cancer populations. CBT can include a variety of strategies, including the encouragement of emotional expression, identification of distorted automatic thoughts, use of problem-focused coping skills, discussion of psychosocial impact on the family, and training in assertiveness and communication skills, among others.[38–40] CBT has been utilized in working with both children with cancer as well as their parents. For example, Marsac et al.[41] provide them with strategies to deal with cancer-related stressors, such as medical procedures and hospital visits. Similarly, Wiener et al.[42] developed an innovative board game called "ShopTalk" that can be used in individual or group therapy sessions to assist in starting conversations about difficult emotional issues related to the child or adolescent's medical illness and the impact this illness has in various situations (e.g., school, home, peer relations).

PSST has been shown to be feasible and efficacious in reducing parental distress[6,7,43] compared to standard care as well as to non-directive supportive therapy in two large RCTs. This training involves teaching a very broad, general strategy that is designed to address a given problem situation. The client is taught to effectively utilize all available strategies that will address or solve the problem, evaluate the specific pros and cons of each possible solution to the

TABLE I. Psychosocial Interventions and Therapeutic Support as a Standard of Care: Summary of Evidence Table

Standard	Evidence summary	Methodology	Quality of evidence	Strength of recommendation
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	• Empirical evidence exists for providing access to psychosocial resources, as the majority of pediatric cancer patients and their families experience increased distress and benefit from such support • Research indicates that a subset of children and families will experience clinically significant adjustment issues. Additionally, psychosocial interventions can produce clinically meaningful decreases in distress Evidence gaps: • Prospective research and more randomized clinical trials could add to existing evidence base	• Consensus • Systematic review articles • Randomized controlled trials • Replication of findings	High quality of evidence given with consistent findings from numerous well-designed studies • Cross-sectional studies	Strong recommendation, given the impact of disease and treatment factors on the patient and family

problem, select a solution and take action, evaluate the effectiveness of that action, and determine whether another option or course of action is needed in order to effectively solve the problem.[44]

Mullins et al.[45] developed an interdisciplinary CBT-based intervention that targeted helping mothers learn to cope with the uncertainty of the cancer experience. This intervention was shown to be feasible, acceptable, and tended to improve adjustment in mothers. Cognitive behavioral approaches have also been integrated with other therapeutic approaches, such as family therapy, and have demonstrated feasibility and efficacy. The best example of such an intervention is the Surviving Cancer Competently Intervention Program developed by Kazak et al.[46,47] It should be noted that while the above interventions are highlighted, there is still the need for further research and development of evidenced-based interventions for children diagnosed with cancer and their parents.

Children with cancer may also have pre-existing psychiatric diagnoses, and are vulnerable to developing a range of psychiatric disorders related to psychological challenges of cancer treatment, as well as the physiological effects of the illness and cancer-directed therapies.[17,48,49] Psychiatric assessment and pharmacologic treatment should be available when other approaches are not sufficient, and medications should be used in conjunction with other psychotherapeutic modalities. Effective psychopharmacologic treatments are available for depression, anxiety, and delirium, and can be used safely in children receiving oncology treatment, although randomized controlled studies are lacking in this population. SSRIs are effective and well-tolerated first line treatments for depression in the general pediatric population, especially in combination with CBT.[50–52] Small studies demonstrate tolerability and efficacy in children with cancer.[48,51–55] There is widespread prescribing of anti-depressants by mental health professionals and pediatric oncologists, sometimes without mental health follow-up,[49,56–58] highlighting the need for access to specialized psychiatric consultation. Refer to Weaver et al.[59] in this special edition for information about palliative care standards.

DISCUSSION

Although accessible psychosocial resources for children and their families are a widely held expectation in pediatric oncology, models of assessment and delivery of services vary widely across centers, as does the availability of psychosocial staff.[35] Additionally, barriers may be present that hinder centers from providing comprehensive psychosocial care. For example, pediatric cancer centers vary in size and location, and as such have varying amounts of resources and funding. Some centers may not treat a sufficient number of patients to justify the financial cost of supporting a comprehensive multidisciplinary psychosocial team (e.g., a social worker, pediatric psychologist, and psychiatrist).

Centers can provide training for members of their staff on how to effectively support patients and families. Examples include ongoing trainings on family centered care, empathic listening, communication, child development, problem solving, and health literacy, among others.[60–62] Centers can build strong referral bases within the community through community support agencies, psychologists, psychiatrists, religious support, and can direct patients and families to these trained professionals. Centers can appoint or hire a staff member to oversee psychosocial support for families who would monitor supportive care given to patients and families and facilitate referrals. Centers can provide formal consultation services with local professionals to assist patients and families during clinic or hospital visits.

Additionally, staff at treatment centers may have limited access to training in supporting the specific needs of pediatric cancer patients and families. Those centers should ensure access to appropriate multimedia resources for both parents and children throughout the course of illness, including, but not limited to, disease education, procedure preparation videos, games, books, and handouts. Books and videos that normalize the cancer experience and feelings associated with different stages of illness could also be available. Centers should make a commitment to having at least one staff member with education or training in supportive care (social work, psychology, or counseling) to assist patients and families.

Based on the available high quality of evidence in the literature, it is a strong recommendation that pediatric oncology centers should provide psychosocial support for patients and families throughout the illness course. Child life specialists, social workers, psychologists, psychiatrists, clinical educators, patient/nurse navigators, and spiritual leaders may all serve in this role.[63] Services from social work and psychology, tailored to the specific needs of each family, are especially valuable at initial evaluation and are critical to the family's active participation in the treatment of their child. Identifying and addressing risk factors early is essential.[64]

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Assessment of Financial Burden as a Standard of Care in Pediatric Oncology

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Family financial hardship has emerged as a burden of pediatric cancer treatment with negative implications for family well-being. As part of an extensive project to create evidence-based standards for the psychosocial care of children with cancer, we performed a literature review of pediatric cancer-associated financial hardship utilizing six databases, and identified 24 publications for incorporation into this review. Financial hardship during childhood cancer was found to affect a significant proportion of the population and to negatively impact family well-being. Existing literature supports a strong recommendation for assessment of financial hardship as a component of comprehensive psychosocial care in pediatric oncology. Pediatr Blood Cancer 2015;62:S619–S631. © 2015 Wiley Periodicals, Inc.

Key words: pediatric oncology; psychosocial; quality of life; support care

INTRODUCTION

Family financial hardship as a consequence of childhood cancer treatment has been documented over the past three decades,[1–6] and the economic burden of caring for a child with cancer has been described as a significant source of distress for families.[3,7–9] Data suggest that financial hardship during childhood cancer treatment has implications for family economic status,[5,6,10–12] parental emotional and mental health,[3,8,13] and potentially child well-being. Efforts to identify and address family financial hardship are essential to the provision of comprehensive care for children with cancer. The primary objective of this review was to assess the existing literature on family financial burden in childhood cancer to determine whether the evidence-base supports incorporation of financial hardship assessment into pediatric cancer care, and to evaluate the quality of existing assessment tools.

METHODS

This review was performed as part of the collaborative *Standards for Psychosocial Care of Children with Cancer and Their Families* effort. For a full description of the methods used to develop each standard, refer to Wiener, Kazak, Noll, Patenaude, Kupst.[14] PubMed, Medline, CINAHL, PsychINFO, Google Scholar, OVID were searched using the terms [“economic aspects of illness” OR “cost of illness” OR “health care costs” OR “financial burden” OR “economic burden” OR “out of pocket”] AND [“neoplasms”]. Results were limited to English-language only publications from March 1, 1995 to March 1, 2015 including data on children aged less than 18 years with cancer. Due to the focus on family financial hardship, studies reporting solely on hospital costs or cost-effectiveness without consideration of family financial impact were excluded. A hand-search of the reference lists from relevant review articles and all eligible studies was additionally performed. The title and abstract of all citations obtained through the search strategy were screened, and full texts were obtained for in-depth review of potentially eligible studies.

The study team comprised a pediatric oncology social worker and pediatric oncology physician–researcher who jointly reviewed all articles. Expert opinion was elicited from additional representatives of pediatric oncology and pediatric oncology social work. Consultative reviewers indicated that the strengths and limitations of the body of evidence were clearly described and that there was an explicit link between the recommendations and the supporting evidence. Utilizing the GRADE (Grading Recommendations Assessment, Development and Evaluation) system, evidence tables were rated for the quality and strength of evidence.[15]

Psychosocial Standard of Care

Pediatric oncology families are at high risk for financial burden during cancer treatment with associated negative implications for quality of life and parental emotional health.

- Assessment of risk for financial hardship should be incorporated at time of diagnosis for all pediatric oncology families. Domains of assessment should include risk factors for financial hardship during therapy including: pre-existing low-income or financial hardship, single parent status, distance from treating center, anticipated long/intense treatment protocol, and parental employment status.
- Targeted referral for financial counseling and supportive resources (including both governmental and charitable supports) should be offered based on results of family assessment.
- Longitudinal reassessment and intervention should occur throughout the cancer treatment trajectory and into survivorship or bereavement.

Abbreviations: GRADE, grading recommendations assessment, development and evaluation

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RESULTS

Of the 42 initially identified studies, 14 failed to meet inclusion criteria based on abstract/title alone and 28 were potentially eligible. Of these 28, two were excluded because they fell outside of the acceptable publication dates, one was excluded as it was not published in a peer-reviewed journal, and one was excluded for failure to include a consideration of family financial impact.

Twenty-four studies were ultimately incorporated into this review. Of these, two articles were systematic reviews or meta analyses of controlled studies or evidence-based clinical practice guidelines, one article was a quasi-experimental study, four articles were non-experimental studies (case-control, cohort), and 17 articles were individual descriptive studies.

Supplemental Table I includes the 24 studies identified and briefly summarizes each study with regard to study design, sample characteristics, main findings related to this standard, study rigor, and level of evidence. All publications report findings of family financial burden due to childhood cancer treatment across a range of healthcare and insurance models which support the need for standardized incorporation of financial assessment into pediatric cancer care models.[4,5,12,16] Studies considered the economic and financial impact of childhood cancer on families across two primary domains: direct costs including monetary expenditures due to illness, and indirect costs including the value of lost wages or productivity.

Out-of-pocket expenses due to travel, accommodation, child-care, food, gifts, and treats were identified as significant sources of financial burden.[4,16–18] Work disruptions as a result of a child's treatment were ubiquitously reported,[4–6,12,13,19,20] including at least one parent quitting a job to provide care for their child in 30–50% of families.[5,6,12,19,20] Variations in study design—including duration of follow-up and included costs—make precise reporting of the magnitude of financial costs to families challenging. However, income loss due to work disruptions and out-of-pocket expenses were estimated at over 30% of after-tax family income in two Canadian cohorts,[10,12] and over 50% of lower income families experienced annual income losses of >40% in a U.S. cohort.[6] Two studies exploring the economic consequences of such losses reported that approximately 15% of U.S. families fell from above to below the poverty line due to treatment-related financial burden.[5,6]

Characteristics at the time of diagnosis associated with highest risk for treatment-related financial burden included baseline low-income, single-parent status, longer treatment protocols, and receipt of care far from home.[3–5,16,20,21] One population-based study identified families of younger children and those with poorer prognoses as being at risk.[22] Caregivers who experienced employment disruptions reported higher financial burden suggesting a need for ongoing reassessment of risk.[23]

A handful of studies explored the consequences of family financial burden with uniform findings of negative impact on parental and family well-being. Financial hardship during a child's cancer care was documented to result in increased emotional distress for families,[3,13] increased burden on parental relationships,[3,24] and increased risk of serious psychological distress for parents.[8,9] These findings support a recommendation for standardized efforts to evaluate family financial needs as part of comprehensive psychosocial care in children with cancer.

Numerous studies explored parental patterns of coping with financial stressors. Parents reported use of governmental supports,[4,13] community resources and foundation funds.[5,13,25] Additional coping strategies included borrowing money from family and friends, selling property or mortgaging homes, taking on loans or credit card debt and reduced spending.[5,13,17,25] Despite these efforts, parents of children with cancer report a need for more practical guidance and financial support to mitigate the financial consequences of childhood cancer treatment and facilitate their care-giving capabilities.[1,5,13,17]

Data from a handful of reviewed studies suggest that financial burden persists well beyond the initial diagnostic and treatment time period. Income losses due to work disruptions were identified in numerous studies as significant contributors to family economic hardship,[5,6,10,18] and represent a variable which cannot be assessed at the time of diagnosis. Investigations of families of children with advanced cancer[5,6,8] report that approximately 25% of families endorse experiencing great economic hardship due to their child's illness. These data support the recommendation for longitudinal reassessment of family financial burden (Table I).

There were four notable limitations to the body of literature reviewed. First, no study in our review examined the impact of family financial hardship on child outcomes or well-being. Emerging evidence suggests that that the interrelated domain of family poverty may impact child cancer outcomes. Recent publications have identified family low-income as an independent predictor of both poor adherence to oral chemotherapy[26] and decreased overall survival for children treated for cancer.[27–29] Although it is plausible that poverty or low-income resulting from financial hardship during treatment may similarly mediate child cancer outcomes, this area requires further investigation. Second, no studies in our review reported on the success of efforts by care providers to prevent or ameliorate family financial hardship in pediatric cancer. It is notable that available governmental "safety-net" programs, including examples such as Medicaid and Supplemental Nutrition Assistance Program in the United States, and child support and employment insurance benefits in Canada[13,30] vary by country and healthcare system. Evidence from general pediatrics suggests that successful enrollment in such programs is beneficial to children and families.[31–34] Third, review of the literature demonstrates significant variability in domains of hardship assessed. Fourth, the appropriate interval and duration of assessment for family financial burden has not been identified in the literature, though multiple publications identify persistent hardship during survivorship and bereavement. Alignment of financial hardship assessments with standard time-points for supportive care or late-effects evaluation is thus reasonable until further research clarifies the trajectory of financial burden.

DISCUSSION

Review of data from 24 peer-reviewed studies published over the past 20 years demonstrates that pediatric oncology families are at high risk for financial hardship during cancer treatment with associated negative implications for quality of life and parental emotional health. Studies primarily included small, cross-sectional quantitative investigations.

Lack of awareness of the scope of family financial burden in pediatric cancer by health care providers, treating institutions, governmental agencies, pediatric cancer foundations, and pediatric

TABLE I. Summary of Assessment of Financial Burden as a Standard of Care in Pediatric Oncology

Standard	Evidence summary[a]	Methodology[b]	Quality of evidence[c]	Strength of recommendation[d]
Pediatric oncology families are at high risk for financial burden during cancer treatment with associated negative implications for quality of life and parental emotional health. Assessment of risk, referral to supports, and reassessment should occur across the treatment trajectory and into bereavement or survivorship.	Research on families of children with cancer indicates that financial hardship associated with treatment increases emotional distress for families, increases the burden on parental relationships, and increases the risk of serious psychological distress for parents.	Quantitative cross-sectional, and longitudinal studies; qualitative studies; mixed-methods; systematic literature review. Majority of studies were cross-sectional retrospective survey, and in-depth interviews. Consistent replication of findings evident across all reviewed papers.	Moderate quality of evidence given methodological limitations of most studies. Consistent findings across numerous investigations and methodological approaches (qualitative, cross-sectional, systematic review) bolster the overall strength of evidence.	Strong recommendation, given the serious consequences for parents and families of financial hardship associated with pediatric cancer treatment.

[a]Based on summary of evidence; [b]Types of studies: e.g., RCT, cross-sectional, longitudinal; consensus; systematic review articles; [c]Quality of evidence: High, moderate, low, and very low; [d]Strength of recommendation: Strong or weak (based on GRADE quality criteria).[15]

cancer families represents the most significant barrier to incorporation of routine assessment during the provision of psychosocial care to pediatric cancer families. Efforts to disseminate existing evidence on the high prevalence of financial hardship during treatment could feasibly be achieved through provider educational seminars and inclusion in family educational materials. Although families already overwhelmed by a child's diagnosis and care may not be in a position to identify and advocate for their financial needs, education of patient advocacy groups and foundations aimed at supporting pediatric cancer families have the potential to facilitate family advocacy. The lack of a standardized and comprehensive instrument with which to assess financial hardship is a major barrier to incorporation into standard care.

Finally, availability of psychosocial support resources varies significantly by institution. Recommendations within this standard are achievable within a variety of clinical contexts, and can be adapted to the availability of institutional resources. Addressing financial burden represents an essential component of comprehensive psychosocial care for pediatric cancer families, thus in settings with limited social work supports routine assessment could be performed by other clinical providers, including nurses, physicians or psychologists, with subsequent referral to social work as needed. Ongoing research to develop standardized screening tools and identify evidence-based interventions is needed. In the interim, attention to this domain of family burden within pediatric cancer has the potential to significantly improve the care of patients and families.

CONCLUSION

Although evidence for this recommendation is of moderate quality secondary to the methodological limitations of reviewed studies, the robustly congruent findings of significant financial burden across numerous investigations and diverse methodological approaches strengthen the data as a whole. Taken together, existing literature supports a strong recommendation for inclusion of a formal assessment of family financial hardship as detailed in the recommended standard in the provision of comprehensive psychosocial care to pediatric oncology families.

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SUPPLEMENTARY INFORMATION

SUPPLEMENTAL TABLE I. **Assessment of Financial Burden as a Standard of Care in Pediatric Oncology**

Study	Design	Sample	Findings	Study Rigor	Level of Evidence
Aung L, Saw SM, Chan MY, Khaing T, Quah TC, Verkooijen HM (2012) [1]	Cross-sectional Primary aim: To assess the financial, psychological, social and emotional impact of childhood cancer in Singapore. Self-administered survey study	Parents of children (age <21 years) with cancer receiving care at a regional referral center in Singapore N = 79 Single center	Financial Burden represented the second highest weighted score (3.41) of 4 domains on the Impact on Family Scale after Familial/Social Burden When compared to caregivers who remained employed, those who were asked to quit their job, or who took voluntary leave experienced a higher financial burden ($P = 0.03$) Recipients of financial aid experienced lower Financial Burden impact	Sample size sufficient Data collection appropriate to study method Descriptive statistics appropriate to sample; statistical methods for associations poorly described Good response rate (82%) Missing data not discussed	6
Barr R, Furlong W, Horsman J Feeny D, Torrance G, Weitzman S (1996) [2]	Two-part design: Retrospective, cross-sectional survey; Prospective, longitudinal diary collection Primary aim: To describe the monetary costs borne by pediatric cancer families and determine whether these costs represent an important component of illness burden	Families of children treated for high risk leukemia, Wilm's tumor (stages 2-5), and Neuroblastoma (stages 3 and 4) at one of two major referral centers in Canada N = 40 off-treatment families N = 64 on-treatment families Multi-center	Despite universal first dollar coverage for medical care in Canada, family-borne costs during the course of these three illnesses are at least one-third of the average family's after-tax income The mean total expenses in 1986 Canadian dollars incurred by families of childhood cancer patients over the entire course of therapy were \$26,000 for leukemia, \$20,074 for Wilm's tumor, and \$10,376 for neuroblasmoma Ongoing weekly costs amount for the largest share of expenses	Sufficient sample size Data collection appropriate to study method Analysis appropriate (descriptive) Good response rates Missing data not well described	6

(*Continued*)

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Standards of Psychosocial Care for Parents of Children With Cancer

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Parents and caregivers of children with cancer are both resilient and deeply affected by the child's cancer. A systematic review of published research since 1995 identified 138 studies of moderate quality indicating that parent distress increases around diagnosis, then returns to normal levels. Post-traumatic symptoms are common. Distress may be impairing for vulnerable parents and may impact a child's coping and adjustment. Moderate quality evidence and expert consensus informed a strong recommendation for parents and caregivers to receive early and ongoing assessment of their mental health needs with access to appropriate interventions facilitated to optimize parent, child, and family well being. Pediatr Blood Cancer 2015;62:S632–S683. © 2015 Wiley Periodicals, Inc.

Key words: anxiety; childhood cancer; depression; distress; intervention; mental health; parents; pediatric oncology; psychosocial

INTRODUCTION

Parents are profoundly affected by a child's cancer diagnosis. Many parents are resilient and well functioning; for them pediatric cancer is an extreme stressor which causes transient, marked distress, slowly returning to a new, changed "normal" that includes the reality of the illness. For parents already struggling to cope or with pre-existing mental health problems, a child's diagnosis of cancer can be overwhelming. A parent's emotional issues may disrupt the ill child's cancer treatment, impact parenting and support for the ill child and well siblings, and threaten family functioning and stability over time.[1–4]

Consistent with growing literature on the interconnectedness of parent and child mental health,[5–7] family centered psychosocial care has been long considered essential in pediatric oncology.[8–10] This paper reviews the literature for evidence of a need for parent mental health support, to determine which parent-directed supports should be considered "essential" for pediatric oncology centers to provide, when they should be offered, and what barriers exist to providing this care within diverse healthcare settings. The evidence on psychological impact of childhood cancer on parents of children with cancer (PCC) as well as literature on the development of parent-specific interventions will be reviewed.

METHODS

This review was performed as part of the collaborative Psychosocial Standards of Care Project for Childhood Cancer (PSCPCC) which was created to develop evidence- and consensus-based standards for psychosocial care in pediatric oncology. PSCPCC consists of a group of pediatric oncology psychosocial professionals in collaboration with a larger interdisciplinary group of experts. For a full description of the methods used to develop each standard, please refer to Wiener et al.[11]

After review of published search strategies,[12] and iterative refinement of the strategy, the final search (Supplemental Table SI) was carried out (March 2015) in PubMed, OVID, and PsycINFO databases. Studies were identified which contained four main concepts: (i) psychosocial adjustment; (ii) parents/caregivers; (iii) pediatric/child; and (iv) cancer. Papers were limited to the English language and restricted by publication date range March 1995–2015, resulting in 4,580 citations. Through title, abstract and full text review, they were narrowed by the authors to 138 citations with these inclusion criteria: (1) subjects are parents of pediatric cancer patients, ages 0–18 years old, on treatment (not in survivorship or bereavement) and (2) studies report parent outcomes measuring distress, coping or psychopathology (Supplemental Fig. S1). The three authors received extensive training in GRADE methodology[13,14] as part of the PSCPCC; each author reviewed a selection of the citations for inclusion in the final review. Regular discussion was used for clarification and consistency of the application of both inclusion criteria and assignment of study rigor. High-quality qualitative research on this subject was identified, but given the large number of articles, qualitative studies were excluded except where they added new or clarifying data. International studies were included if judged to be culturally relevant to the United States health system. A detailed summary of each study's results and study rigor is in Supplemental Table SII. This standard was developed using the GRADE methodology[13,14] for both qualitative and quantitative research. The evidence was reviewed throughout its development by several national professional committees

Psychosocial Standard of Care

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.

Abbreviations: PCC, parents of children with cancer; PTSD, post-traumatic stress disorder; PTSS, post-traumatic stress symptoms; PSST, problem solving skills training; RCT, randomized controlled trial

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including experts from multiple disciplines in psycho-oncology and pediatric oncology.

RESULTS

This body of literature is based on empirically supported theoretical frameworks that initially stemmed from developmental literature and have been refined to account for the unique experiences and needs of children with cancer and their families. Three main themes were identified as interwoven in the literature: (i) adjustment trajectories of parents of children with cancer (PCC); (ii) identification of psychopathology in PCC (primarily anxiety, depression and posttraumatic stress symptoms [PTSS]); and (iii) development of psychological interventions for PCC. The 138 articles included 102 observational studies (92 quantitative cohort, case control, or cross-sectional descriptive designs, 10 qualitative); 14 systematic reviews; 18 intervention studies (mostly pilot, randomized controlled trial (RCT) studies) and four critical analysis/expert commentary articles. The evidence consistently indicates that attention to parental mental health in pediatric oncology populations is imperative.

Trajectories of Parental Adjustment and Patterns of Risk for Psychopathology

The first two themes discovered in the literature are found throughout all 138 articles included here. PCC must engage internal and external resources to effectively cope with the tremendous stress of their child's diagnosis and treatment. A typical pattern of adjustment is one of elevations in distress around the time of diagnosis, with decreases over time to normal functioning, 3–6 months post-diagnosis,[3,15–22] but a cluster analysis showed there may be more than one trajectory of distress.[23] Most PCC and families adjust well, and report growth, increased closeness, and good family functioning. Potential resiliency factors include higher socio-economic status[24,25] higher levels of social support[26–30] and higher family cohesion and functioning.[31] Parent attributes such as optimism[32,33] and use of problem-solving coping strategies (defined as "sets about solving problems purposefully", compared to "emotion-focused coping" and other described coping styles)[26,34–36] have also been associated with decreased parental distress.

Parental distress has a negative impact on parental quality of life, mental and physical health factors, family functioning,[3,37–39] and marital distress.[40] Parental psychosocial functioning at 6 months post-diagnosis has been found to predict long-term psychosocial outcomes,[2] with a significant number of PCC (27% of PCC vs. 15% in control group of parents without children with cancer) still reporting elevated distress up to 5 years post-diagnosis.[15] Although most data on parental adjustment is based on mothers, both parents have been shown to exhibit significant distress.[41–45] Gender differences in parent adjustment and coping should be considered in intervention development.[19,34,46–48]

Measurement of the impact of parent distress on child adjustment and distress has been methodologically challenging. Previously, parent reporting bias (distressed parents report more child distress) seemed to explain much of the link between these two outcomes.[18,20,23,31] However, a recent longitudinal study showed a lagged, downstream relationship between maternal distress and child internalizing symptoms, one that may not be completely explained by methodology.[49] Furthermore, adolescents' self-reports of distress have been found to correlate with higher parent distress.[50] More research, using controlled, longitudinal designs and multiple reporters, is needed to delineate this relationship.

Data reflecting elevations in distress are based primarily on self-report questionnaires compared to either measure norms or control groups comprised of parents of healthy children[3,51–54] or parents of children with other acute or chronic conditions.[1] Data from these studies indicate that a subset of PCC is at risk for marked or prolonged distress or psychopathology. Rates of reported depression, anxiety, and PTSD vary amongst samples and methodologies with one paper reporting no clinically significant elevations on standardized measures,[51] and others reporting rates up to 43%.[29,42,52,55–59] Others, using questionnaires based on diagnostic criteria, have shown that 11% of mothers and 9% of fathers have PTSD.[55] Virtue et al.[60] reported 27% of mothers of children undergoing stem cell transplant met criteria for specific depressive or anxiety disorder diagnosis.

Post-traumatic stress symptoms (PTSS) and PTSD have received significant attention. Models of pediatric medical traumatic stress have informed this research and show that a pediatric cancer diagnosis and subsequent events can be potentially traumatic.[9] PCC have more PTSS than ill children themselves—nearly all PCC report at least one PTSS symptom in the first 2–4 weeks after diagnosis and, in one study, 51% of mothers and 40% of fathers met criteria for acute stress disorder (ASD) in the first 2 weeks.[61] Beyond the first month, studies have varied with some groups finding PTSD/PTSS rates similar to those of the general population of adults (without children with cancer), reinforcing the findings that in general PCCs as a group are resilient.[62] Assessment of subclinical levels of PTSS and associations with other outcomes may be most applicable as a framework to inform research and intervention.[63]

Several potential factors have emerged as indicators of risk for parental maladjustment. Socioeconomic factors like lower household income,[55,56] lower level of education, lack of employment;[55,64] pediatric disease factors such as relapse, treatment severity/risk;[65,66] or poorer child's functional impairment or physical symptoms[67] prior traumatic life events and prior parent psychiatric treatment have all been associated with parental caregiving burden and distress and poorer adjustment in different studies.[68]

Interventions

Psychological interventions to reduce distress and improve adjustment in PCCs are emerging. Most are family systems informed cognitive and behavioral therapies that are delivered individually. Twenty-one articles focused on interventions were identified (Supplemental Table SII), of which three were reviews;[69–71] the other 18 described various aspects of development of ten distinct intervention programs.[10,49,72–86] Unifying this group of studies are strong theoretical frameworks based in social ecological psychology, resiliency and illness-specific coping models, as well as overall good tolerability and feasibility. Problem solving skills training (PSST), which has now been implemented with over 800 mothers, was efficacious in improving problem solving skills and reducing negative affect in mothers of children newly diagnosed with cancer.[78–81,87] It may be most effective for young, single mothers, but is effective for all groups, with lasting benefits beyond the timing of the intervention.[78] In addition, Fedele et al.[49,72] showed parent and child benefits after a 12-session intervention targeting mothers' coping with illness uncertainty, which supports the parent-targeted intervention model. Critiques of the literature overall, have cited methodological problems with small populations of generally psychologically healthy parents, difficulties with recruitment when families are stressed, inclusion of mostly mothers, timing of interventions and lack of appropriate controls.[71,88] Future studies should focus on dissemination, and further refining interventions to target patients at risk for poor adjustment.

DISCUSSION

Based on this review a recommendation was developed and circulated to colleagues in pediatric oncology, as well as discussed at a "think tank" of pediatric psycho-oncology professionals from various clinical settings.[11] Structured feedback (received through rating forms) and unstructured comments from the Standards Committee supported the recommendation but helped refine the language, resulting in a final standard that was applicable in a wide range of pediatric oncology centers. We strongly recommend the following standards of psychosocial care for PCC:

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. Table I details a summary of the results and the basis of the recommendation based on GRADE criteria. Family centered care should include standard systematic screening for psychosocial risk,[89] including assessment of parent mental health and coping.[9,90] Assessment of children referred for distress or psychiatric disturbance should also include assessment of parent functioning.

Evidence-based interventions for families facing childhood cancer are not yet routinely available, given the recent and developing nature of this literature, lags in dissemination, and site-specific resource limitations.[91] Appropriate interventions for PCC are those provided by mental health providers who are knowledgeable about childhood illness and its effects on the family. Providers should be familiar with established theoretical frameworks (e.g., pediatric medical traumatic stress, models of resiliency and family systems theory),[92] aware of the normal trajectory of resilience so they do not miss persistent distress or psychopathology in PCC,[93] prepared to address cancer-specific parenting concerns, parental traumatic symptoms, difficulties coping with illness uncertainty, need for support for problem solving, and stress management.

Practical barriers and recommended responses to parent treatment, discussed in Supplemental Table SIII, should be systematically and individually assessed. Importantly, the parents who are most impaired will struggle to overcome these common obstacles to referral-based mental health care and it is important to partner with them to prioritize and facilitate their own mental health care even during a child's active cancer treatment.

Several gaps are identified as targets for research. Specific risk factors for parent maladjustment such as need for stem cell transplant, presence of hereditary cancer risk, and prior child loss will likely require tailored interventions. The impact of co-existing parent mental illness (e.g., major depression, bipolar disorder, substance abuse) on parent and child adjustment in cancer and suitability of these PCC for participation in emerging evidence-based treatments for PCCs is not well understood. Finally, research is urgently needed to guide best practice for the delivery of

TABLE I. Summary of Results (GRADE)[9,10]

Standard	Evidence summary	Methodology[1]	Quality of evidence[2]	Strength of recommendation[3]
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	Most parents experience a trajectory of significant transient distress, but ultimately good coping and resiliency Subset of at-risk parents, 25–30%, will have increased or prolonged distress or psychopathology Parent distress to due childhood cancer has a broad and deep impact on parent, child and family functioning over the long term (at least 5 years)	Mixed-methods, qualitative quantitative studies, and literature reviews Majority cross-sectional survey and in-depth interviews Randomized controlled trials and pilot intervention trials Replication of findings evident Think tank/expert consensus	Moderate quality given consistent findings from lower to moderate level evidence studies.	Strong recommendation based on moderately well documented impact of parent mental health on child mental health and family functioning; consistent replication of findings in moderate quality studies on levels of parent distress and wish for support; flexibility and tolerability (low risk) of the recommended intervention to fit individual parent and family values, address their needs, and overcome barriers to care and limited resources

[1] Types of studies: for example, RCT, cross-sectional, longitudinal; consensus; systematic review article; [2] Quality of evidence: high, moderate, low, or very low (based on GRADE criteria); [3] Strength of recommendation: strong or weak (based on GRADE quality criteria).

integrated medical and mental healthcare that is accessible and effective.[94]

A significant body of literature now supports a family systems approach to pediatric cancer care with special attention to the mental health needs of PCC. Pediatric oncology and psycho-oncology clinicians must address the barriers unique to each setting and case to achieve this essential integrated, flexible, expert care for PCC.

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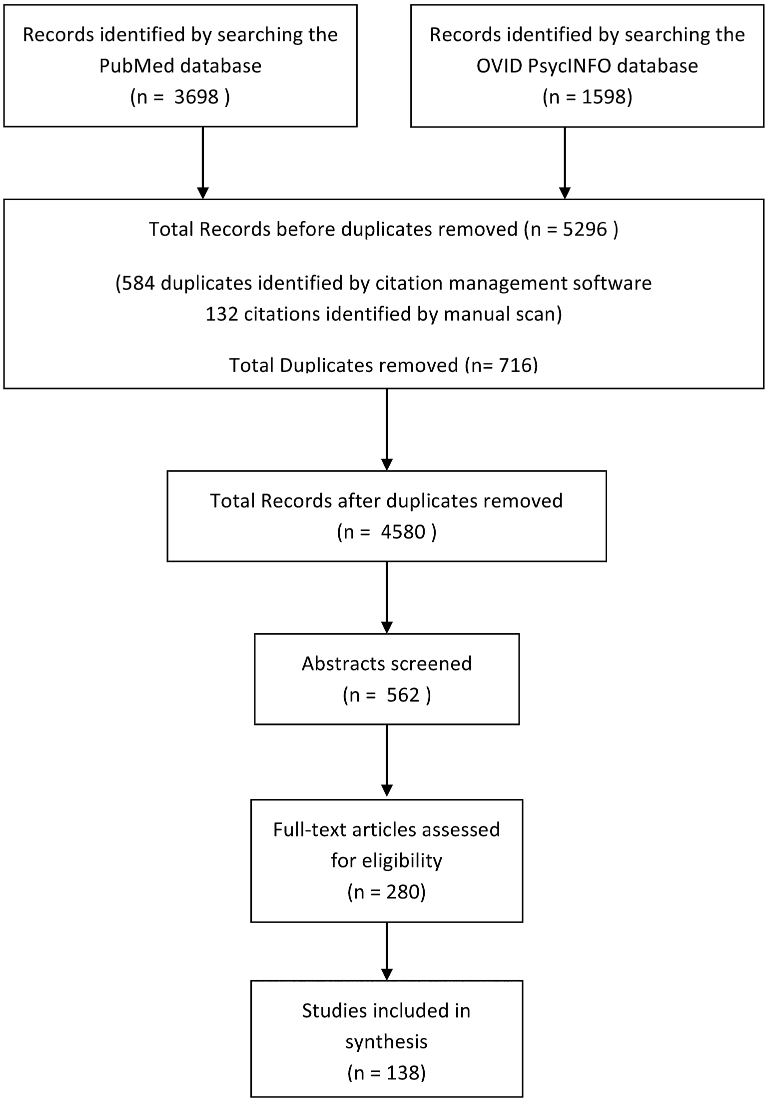
SUPPLEMENTARY INFORMATION



SUPPLEMENTAL FIGURE 1: Flow Diagram of Literature Review for Psychosocial Standard of Care for Parents of Children with Cancer.

Pediatr Blood Cancer 2015;62:S684–S693

Anticipatory Guidance and Psychoeducation as a Standard of Care in Pediatric Oncology

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The aim of this review was to critically evaluate the literature on anticipatory guidance and psychoeducation for youth with cancer and their caregivers. Twenty-one publications were identified. Overall, psychoeducation efforts and interventions were well-liked and accepted by patients and caregivers, improved patient and family knowledge about childhood cancer, and increased patient's health locus of control. A number of modalities are effective in giving families anticipatory guidance, provided the content and delivery are matched to the needs and preferences of individual patients and caregivers. Evidence supports a strong recommendation for psychoeducation for youth with cancer and their families. Pediatr Blood Cancer 2015;62:S684–S693. © 2015 Wiley Periodicals, Inc.

Key words: anticipatory guidance; childhood cancer; psychoeducation; psychosocial

INTRODUCTION

When a child or adolescent is diagnosed with cancer, patients, parents, and other family members are challenged to learn about evolving diagnostic data, treatment and treatment-related side effects, prolonged hospitalizations, tests and procedures, navigating the hospital system, and overall uncertainty about prognosis and outcome.[1] Because the unfamiliar nature of the hospital can be distressing,[2] providing patients and families with anticipatory guidance, information, or psychoeducation about what to expect at points along the disease trajectory (e.g., diagnosis, throughout treatment, during survivorship, at end-of-life) is believed to be an important aspect of care.[3–5] It has been argued that educating children and their parents about the diagnosis and treatment plan helps to reduce uncertainty and decrease associated distress,[6] establish trust with health care providers, and enhance adjustment to illness.[7,8]

Although it was standard practice in the 1960s to withhold information about disease and treatment from children with cancer, it is now widely accepted that patients should be offered a developmentally appropriate education about their disease in a timely fashion, as this understanding contributes to better psychological outcomes.[7–10] Evidence supports that children and adolescents want more information about their bodies, their illnesses, and their treatment,[11,12] and that without such information, they may form beliefs and attitudes that are inaccurate, provoke fear and anxiety, and/or adversely affect compliance.[7,13] Alternatively, children who know more about their illness may feel more in control of their health overall, which can lead to reduced distress and better outcomes.[7,14,15]

Although clinicians agree that providing guidance to and educating patients and families is important, pediatric patients and caregivers continue to report unmet informational needs along the cancer trajectory, i.e., from diagnosis,[16,17] through to end of treatment,[18–20] and into survivorship[21,22] or end-of-life.[23] Retrospectively, caregivers and pediatric patients indicate that they needed additional guidance and information regarding illness and treatment; treatment decisions; side effects; late effects; tests and procedures; fertility; diet/nutrition; caring for their child; physical and emotional impact; available services and resources for support; appropriate follow-up care; and impact on the family.[1–22,24–26] As such, a standard of care for providing guidance and education to patients with cancer and their families is overdue.

Psychosocial Standard of Care

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.

The scope of this work is focused on youth ages 0–18 diagnosed with any malignancy and their caregivers and includes both anticipatory guidance and psychoeducation. Anticipatory guidance, a longstanding practice in general pediatrics literature,[27–30] is a proactive, developmentally based education approach that prepares parents for what they should expect in the coming months and years and focuses on the needs of a child at each developmental stage. Psychoeducation is a related concept, used in the context of chronic illnesses to refer to the process of providing information to patients and/or family members in order to empower them, assist with day-to-day management of the disease and decision-making, relieve uncertainty, and enhance psychosocial adaptation to the illness.[31] Guidance and psychoeducation can take many forms, including formal or informal, individual, dyadic, or group models where a

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Abbreviations: QoL, quality-of-life; RCT, randomized control trial

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provider gives information to the patient and/or family members via verbal instructions/consultation, written/printed materials, or computer-mediated methods such as video, computer programs, video games, the Internet, and tutorial CD-ROMS.[32]

METHODS

To develop this standard, we used methods described by Wiener et al.[33] in this special issue for the Standards for Psychosocial Care of Children with Cancer and Their Families project. Computerized literature searchers of PubMed, PsychInfo, and CINHAL were performed. Search inclusion criteria included English-language literature published from March 1, 1995 to March 1, 2015 in peer-reviewed journals with participants aged 0–18 years and with a history of any malignancy. Exclusion criteria included non-cancer diagnoses, patients over age 18, foreign language publications, and literature that was not empirical research. Specific search terms included “psychoeducation,” “anticipatory guidance,” “information intervention,” “didactic intervention,” “patient education (as topic)” OR “family education” AND cancer-related terms AND “Child” OR “Adolescent” OR “Pediatric” OR “Pediatric” OR “Youth” OR “Children” (using indexed MeSH terms). Results of database searches were supplemented with hand-searching of the reference lists of all included studies. Searches revealed a total of 1,168 citations. Authors followed PRISMA guidelines for systematic reviews[34] and excluded 1,147 articles for the following reasons: 1) duplicate articles, 2) articles not related to childhood cancer, 3) articles not about patients or parents of patients age 0–18, and 4) articles not related to the topic of anticipatory guidance/psychoeducation. Articles were restricted to informational interventions, excluding skills-based interventions (i.e., cognitive-behavioral), except in cases where informational interventions were included with skills-based interventions and study designs precluded teasing apart unique effects of intervention components. Articles specific to siblings and to providing procedural support were excluded, as they are more relevant to and are covered in separate standards.[35,36] Finally, articles describing education about research were excluded. These exclusions left 21 articles for inclusion in the synthesis of evidence.

External reviews were conducted by representatives of child life and nursing education, as well as physicians, several parents of children with cancer and childhood cancer survivors. Revisions were made according to feedback received from these reviewers. No members of the study team had any conflicts of interest with the development of this standard.

RESULTS

The search strategy identified 21 peer-reviewed articles, including four systematic reviews, one meta-analysis, and three randomized controlled trials (RCTs). A summary of the evidence is provided in Table I. Supplemental Table I briefly summarizes each

TABLE I. Anticipatory Guidance and Psychoeducation Standard Summary of Evidence Table

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
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.	Patients and caregivers report unmet informational needs along the cancer trajectory. Psychoeducation appears to have the most consistent effects on improving patient/caregiver knowledge about disease and treatment and on increasing health locus of control. A number of modalities are effective in providing families anticipatory guidance, provided the content and delivery is matched to the needs and preferences of the particular patient and caregiver. Existing studies had methodological and conceptual weaknesses, including lack of an organizational model, lack of support for the ideal type of education, and inconsistent assessment of health-related or psychological outcomes.	Systematic reviews, RCTS, Pre/post-test designs, and individual qualitative and quantitative studies. Consistent findings evident, although literature is quite scattered.	Moderate quality given consistent findings from moderate-level evidence studies.	Strong recommendation given risk-benefit ratio (i.e., minimal risk to patients and caregivers and potential benefits of meeting unmet informational needs, improving disease-related knowledge, and increasing health locus of control).

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[1] Based on summary of evidence table for that standard. [2] Types of studies: e.g., RCT, cross-sectional, longitudinal; consensus; systematic review articles. [3] Quality of evidence: high, moderate, low, and very low. [4] Strength of recommendation: strong or weak (based on GRADE quality criteria).

of the 21 studies with regard to study design, sample characteristics, main findings related to this standard, study rigor, and level of evidence.[37] In general, multiple reports support psychoeducation efforts as helpful to, well accepted, and appreciated by patient and family stakeholders.[38–44]

Five articles, including one systematic review and two RCTs, indicated that psychoeducational interventions improve patient and caregiver knowledge about disease and treatment.[45–49] Bradlyn et al.[45] concluded that acquisition of health-related knowledge in children is best facilitated by modalities that are highly interactive and individualized. In a large, multi-site RCT of adolescents and young adults with cancer, a psychoeducational video game improved cancer knowledge and was found to be an effective method of delivery for disease education.[48,49] A small intervention study reported that parents participating in a four session intervention demonstrated better cancer knowledge compared to waitlist controls,[47] and an evaluation of a survivorship education program reported that adolescent and young adult survivors and their caregivers perceived an increase in knowledge of survivorship topics and resources after program attendance.[46]

Three articles found that psychoeducation increased patient's health locus of control.[38,39,42] Children with leukemia,[38] and solid tumors[42] randomized to receive an interactive CD-ROM about disease and treatment showed increased feelings of control over their health, compared to children who received written materials. Data on other health-related outcomes were less consistent, with some studies reporting improvements in outcomes like anxiety,[2,8] depression,[2] pain,[40] disease-related skills,[50] caregiver well-being,[44] self-efficacy,[48] and adherence.[48] Others reported no differences or changes in nausea,[51] stress,[47,48] psychological problems,[47] or quality of life (QoL).[48]

Of note, the systematic review of published research on psychoeducation by Bradlyn et al.[45] concluded that psychoeducation is most effective and impactful when it is tailored to the individual patient/family, rather than based on assumptions about the patient's needs or preferences. Factors suggested to influence the effectiveness of informational interventions include sociodemographics (e.g., education and literacy level of the patient or caregiver), coping styles/preferences (e.g., information seekers vs. information avoiders), learning styles (e.g., auditory, visual), and culture. Similarly, in a study of parents over the course of the first year of diagnosis, qualitative analysis supported that information should be adjusted to caregiver needs and that providers should consider what information is provided when and in what format, depending on family preferences.[52]

Several studies described development, feasibility, and acceptability of computer, video, or web-based formats for providing guidance and information to patients and families.[2,38,39,41,42,45,48,49,53,54] These formats appear to be well-received by families and may be promising, but additional research is needed, as some studies showed low utilization of information web-based resources[53] and no differences in knowledge gains, QoL, self-efficacy, or coping when compared to less technological/handbook formats.[38,42]

DISCUSSION

Overall, review of data from 21 studies indicates that psychoeducation appears to have the most consistent effects on improving patient/caregiver knowledge about disease and treatment and increasing health locus of control. A number of modalities are potentially effective in providing families anticipatory guidance, provided the content and delivery is matched to the needs and preferences of the particular patient and caregiver. The benefits of anticipatory guidance as an intervention per se are limited in the literature; however, information needs at critical times during the cancer trajectory have been described[16,18,19,21,22] and, consistent with the adult cancer literature, patients and families want and benefit from information.[55] Although the Children's Oncology Group's Survivorship Guidelines are currently being used as an anticipatory guidance tool for patients and families,[56] standardized, evidence-based tools for other points along the trajectory are lacking or have not been widely disseminated.

The current evidence base is somewhat scattered and lacks an organizational or theoretical model. In general, studies were rated as moderate quality because of small sample sizes, lack of control groups, and limited RCTs. Many studies combine psychoeducational/informational interventions with skills-based interventions (e.g., cognitive behavioral therapy), making it difficult to tease apart unique effects of intervention components. Furthermore, the terminology used in the cancer literature to refer to psychoeducational efforts (e.g., information, knowledge, education, training, didactics) is inconsistent and frequently used without qualification or definition,[45] which makes it difficult to draw generalizable conclusions across studies.

Psychoeducational or informational interventions for increasing patients' knowledge, self-help skills, and attitudes are already well established for other chronic diseases of childhood such as asthma and diabetes.[57,58] Similarly, the adult cancer literature shows that such interventions can reduce distress, depression, anxiety, improve a variety of health-related outcomes, and increase satisfaction with care.[59,60] Comparatively, studies evaluating the impact of psychoeducation and anticipatory guidance on health outcomes in children with cancer and their families are relatively limited. As such, more research is needed to guide best practice. Important future directions include evaluating effects of informational interventions on health-related (e.g., adherence, illness-self management) or psychological outcomes (e.g., anxiety, depression), determining components of psychoeducation that are most helpful, the appropriate depth and timing of info to give children and families undergoing treatment, and preferred and most efficacious models and methods of delivery. Finally, little is known about how educational needs change at different points along the illness trajectory and across a patient's development or how to tailor education to families of different compositions, cultures, and learning styles.

Having sufficient personnel resources may be an organizational barrier to implementation of psychoeducation and anticipatory guidance for patients with families and their caregivers. This is especially true if providers make efforts to tailor education to the individual needs and preferences of specific patients and families, as the research recommends. Providing education in a group format or through the use of standard curriculum/platforms may assist with reaching multiple patients and families with less expenditure of personnel resources, but the consequence may be a lack of personalization to patient needs and preferences. Lack of role definition of providers may present additional challenges to effective education of patients and caregivers, as professionals across a wide-range of disciplines (e.g., nurses, physicians, social workers, psychologists, child life specialists) have experience with and expertise in providing psychoeducation and anticipatory guidance to patients and families; this may result in duplication of effort and inefficient use of resources. Communication and coordination of services may be accomplished through clear documentation of efforts and through multidisciplinary rounds, where providers can discuss patient and family education needs and delineate what guidance will be provided by whom along the trajectory of cancer care.

With some consistent findings from moderate-level evidence studies, current evidence for this psychoeducation standard is of moderate quality overall; notably, there are no data regarding essential elements of services or consistent evidence for improved health-related outcomes. This standard is an overall strong recommendation given the minimal risk to patients and families and the potential benefits of meeting unmet informational needs, improving disease-related knowledge, and increasing health-locus of control (Table I).

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Procedural Preparation and Support as a Standard of Care in Pediatric Oncology

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Youth with cancer undergo many repeated and invasive medical procedures that are often painful and highly distressing. A systematic review of published research since 1995 identified 65 papers (11 review articles and 54 empirical studies) that investigated preparatory information and psychological interventions for a variety of medical procedures in pediatric cancer. Distraction, combined cognitive–behavioral strategies, and hypnosis were identified as effective for reducing child pain and increasing child coping. Low- to high-quality evidence informed strong recommendations for all youth with cancer to receive developmentally appropriate preparatory information and psychological intervention for invasive medical procedures. Pediatr Blood Cancer 2015;62:S668-S673 © 2015 Wiley Periodicals, Inc.

Key words: anxiety; cognitive–behavioral; distraction; distress; hypnosis; nonpharmacological; pain; pediatric oncology; preparation; procedure; psychosocial; psychological intervention; standards of care; support

INTRODUCTION

Pediatric patients who are diagnosed and treated for cancer undergo many repeated, invasive, and painful medical procedures. Untreated or undertreated procedural-related pain can create significant changes in pain processing, and increased anxiety and distress.[1–3] These sequelae may occur before, during, and even weeks after the procedure [1,4–6] and may contribute to longer lasting psychological symptoms such as posttraumatic stress and anxiety.[7–9] Efforts to reduce these symptoms are critical as unmanaged pain, anxiety, or distress during prior painful procedures has been found to be predictive of difficulty at future procedures [3,10,11] and may result in physiological changes or conditioned responses that impact the way pain is processed.[12–15]

An extensive line of research has been devoted to the provision of information and development of interventions to help youth cope during painful procedures.[see Refs. 16–32] However, over the past few decades, pharmacological interventions, including local and general anesthesia, are being increasingly utilized and creating a paradigm shift in the way youth experience procedures. Therefore, what is stressful to children currently diagnosed and treated for pediatric cancer is different than in previous decades. Despite availability and advances in effective pharmacological approaches, their use remains underutilized and at times ineffective.[33,34] The most effective pain management approaches are multifaceted and combine pharmacological approaches with psychosocial procedural preparation and intervention.[20,35–40]

A review of the literature on procedural preparation has established that special care is required to prepare children to undergo painful medical procedures.[41–44] Preparatory information provides children and adolescents with a sense of predictability and control.[45] Foundational research in pediatric psychology established the importance of providing children with information about procedures through modeling, rehearsal, books, puppets, or medical play,[46–51] focusing on sensory information,[16,52] or combining procedural-sensory information.[17,53–56] It represents the beginning efforts to help children cope with painful and invasive medical procedures. Additional research helped to establish that preparation should be well timed, developmentally appropriate, and include descriptions of the sequence of events that will occur, as well as accurate and honest information about what pain and sensations to expect.[57–63]

Psychosocial Standard of Care

All youth with cancer should receive developmentally appropriate preparatory information about invasive medical procedures. All youth should receive psychological intervention for invasive medical procedures.

There is a significant body of empirical evidence to suggest that children and adolescents benefit from psychological interventions when undergoing painful medical procedures.[64–67] Psychological interventions allow youth to learn, rehearse, and incorporate coping strategies, which often lead to increased self-efficacy. Overall, psychological interventions have been shown to lower self-, parent-, and observer-reported pain, anxiety, and distress, as well as lower physiological arousal in children and adolescents during medical procedures. Empirical evidence and multiple consensus statements agree that providing children information about procedures and implementing psychological intervention can ameliorate many of the deleterious effects from invasive medical procedures, increase child coping and cooperation, and prevent increased pain and distress with subsequent procedures.[10,11,68–76]

Abbreviations: BMA, bone marrow aspiration; LP, lumbar puncture; PSCPCC, Psychosocial Standards of Care Project for Childhood Cancer; RCT, randomized controlled trial

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Assessment is a crucial component of procedural preparation and intervention. Evaluation of a child's developmental and cognitive abilities as well as their preference for provision of information is paramount. Attaining child preferences allows providers to deliver the type and amount of information needed. Some youth prefer detailed information and others prefer more general descriptions as too much information can increase anxiety. Child preferences and abilities may also inform subsequent approach to intervention.[61]

The primary objective of this review was to assess the existing literature on procedural support to inform evidence-based standards for incorporating procedural preparation and support for all youth with cancer.

METHODS

This review was performed as a part of the collaborative *Psychosocial Standards of Care Project for Childhood Cancer* (PSCPCC) effort.[77] Studies were identified by conducting computerized literature searches of Ovid (PubMed, MEDLINE, Cochrane), PsycINFO, and CINAHL. The searches combined "child," "neoplasm," "procedure," and "intervention" terms and follow-up-related terms and MeSH headings (See Supplementary Table I for full list of search terms). Results of the standardized database searches were supplemented with hand searches to ensure that systematic reviews, randomized clinical trials, and other seminal articles were included. Search criteria were standardized across PSCPCC standards in an effort to maintain consistency and provide readers the most useful and updated overview of the current state of the field and its future directions. Inclusion criteria included youth (up to the age of 18 years); English language; all malignancies; dates of publication (March 1, 1995–March 1, 2015); and the following study designs: clinical trial, comparative study, evaluation studies, guideline, meta-analysis, multicenter study, observational study, practice guideline, and systematic review. Exclusion criteria were as follows: patients over the age of 18 years, foreign language only, noncancer diagnoses, case studies, and commentaries. Systematic reviews published after 1995 were excluded if they reviewed individual studies published prior to 1995.

A multidisciplinary group of providers including pediatric oncologists, pediatric psychologists, and a child life specialist reviewed the recommendations to ensure validity and feasibility for practice. Results of their review impacted the inclusion of adult–patient communication and interactions. Grading of Recommendations Assessment, Development and Evaluation methodology was used to appraise the body of evidence. For a full description of the methods used to develop each standard, the reader can refer to Wiener et al. in this special issue.[78]

RESULTS

Database searches identified over 7,000 articles, of which 65 papers (11 review articles and 54 empirical studies) were included. A PRISMA flowchart of study screening, study identification, and reasons for exclusion is provided in Supplementary Figure 1. Of the empirical studies, 28 were randomized controlled trials (RCTs), 25 were quantitative (e.g., longitudinal, cohort, and observational), one was qualitative,[79] and one used mixed methods.[80] Children aged 2–19 years were represented. Psychological interventions were most often compared to one another or to standard care, and less frequently with pharmacological interventions.[35,36,40,81,82] Medical procedures were most often bone marrow aspirations (BMAs) and lumbar punctures (LPs), as well as venipuncture/venous access, port access, intravenous insertion, radiation therapy, chemotherapy, dressing change, and stem cell bone marrow transplantation. Most studies included children undergoing repeated medical procedures, with few studies focused on a first procedure [80,83] or coping with hospitalization.[84] Child pain, anxiety, fear, and/or distress were primary outcomes as rated by the child, parent, or nurse, or behavioral observation. Less common outcomes were child coping, mood, compliance/cooperativeness, symptom severity, physiological arousal, need for sedation, adult–child communication, and parent anxiety.

Psychological interventions involve cognitive and/or behavioral components that focus on modifying children's thoughts and beliefs or teaching behavioral strategies to enhance coping and reduce pain and distress from medical procedures.[64–67,79,85–92] Strategies can be implemented prior (immediately or days/weeks before), during, and/or following completion of the procedure. Reviewed interventions included distraction (21 studies), combined cognitive–behavioral (11 studies), hypnosis (six studies), memory reframing (two studies), breathing (one study), art therapy (one study), behavioral training (one study), and biofeedback/relaxation (one study). Additional studies explored the impact of adult behaviors and adult–child communication on children's coping (10 studies).[93]

Distraction includes all efforts to draw attention away from the medical procedure to something more interesting and engaging.[94] Reviewed RCTs and quantitative studies described music, books, toys, videogames, virtual reality, pet therapy, blowing bubbles, or conversations with parents as distraction techniques.[40,80–82,84,90,95–111]. Review articles and almost all studies found some benefit of distraction, such as reduced pain and distress (anxiety/fear).[64,65,67,84,86,90,91] Even when distraction was no more effective than standard care, children and parents still reported it to be helpful, enjoyable, and that they would like to use it again.[80,98] Despite this evidence, it remains unclear what make an effective distraction intervention, although considerations of adult involvement, child choice of distracter, requirement to interact with a distracter, and/or use of technology may be helpful.[64] Pediatric oncology nurses identified distraction as less effective for children with extreme anxiety or fear, children who are not receptive to distraction, children who have had previous negative experiences, or where there is insufficient time.[79] Higher parent anxiety is also associated with decreased distraction efficacy.[106]

Combined cognitive–behavioral interventions as described in the reviewed studies involved at least two of the following strategies, including imagery, coping self-statements, reframing, relaxation, breathing exercises, modeling, rehearsal, desensitization, positive reinforcement, cognitive or behavioral distraction, parent training, and/or parent/staff coaching. Many interventions included specific procedural preparation, such as information, medical play, in-person rehearsal, and/or filmed modeling of the procedure. These strategies familiarize children with medical apparatus, supplies, and equipment.[112] Several RCTs, quantitative studies, and review articles showed benefits of combined cognitive–behavioral interventions,

TABLE I. Procedure Preparation Standard Summary of Evidence Table

Standard	Evidence summary	Methodology	Quality of evidence	Strength of recommendation
All youth with cancer should receive developmentally appropriate preparatory information about invasive medical procedures.	Empirical research for children and adolescents with cancer indicates benefit of preparatory information for decreasing children's distress and increasing coping and compliance during a variety of medical procedures. Evidence gaps: • Most of the reviewed evidence for preparatory information post-1995 is within the context of broader combined cognitive–behavioral interventions. More research is needed to determine how well children retain and understand the content of preparatory information across the developmental trajectory, as well as ways to make procedure-specific information optimally delivered and understandable for children and their families across procedures that children currently report as the most distressing.	Systematic review articles; randomized controlled trials; repeated measures/ longitudinal studies	Low[1] quality of evidence given inconsistent findings and some indirectness of evidence from several well-designed studies of broader psychological intervention.	Strong recommendation, given the impact on children's coping and distress, respect for children and parents, and potential harms of not informing, as well as the supporting evidence available pre-1995 and with other pediatric populations.
All youth should receive psychological intervention for invasive medical procedures.	Empirical research for children and adolescents with cancer indicates significant benefit of psychological interventions for reducing child pain and distress, in addition to other outcomes, during a variety of medical procedures. Psychological interventions with the most supportive evidence include distraction, hypnosis, and combined cognitive–behavioral interventions. Evidence gaps: • More research is still needed to assess what components of the interventions are the most effective and for whom. • Several interventions have limited, but promising evidence and additional research is needed, including breathing alone, behavioral training alone, and art therapy.	Systematic review articles; randomized controlled trials (single and multisite); nonrandomized/quasi-experimental group comparisons; repeated measures/longitudinal; observational studies; qualitative studies	High quality of evidence given numerous well-designed studies with predominantly consistent findings	Strong recommendation, given the benefits for pain and distress, as well as other outcomes

[1] It is important to note that much of the literature demonstrating the effectiveness of preparatory information fell outside of our targeted literature search criteria (pre-1995) and therefore was not evaluated for this review despite strong evidence to support its effectiveness.

including reduced child pain and/or distress and reduced parental anxiety.[66,83,87,90,91,113–115] Other studies reported no benefits for pain and distress,[116–118] but did find increased coping, lower physiological reactivity, and subjective perceptions of the intervention as helpful.[119–123] Increased practice of intervention strategies at home prior to the procedure appears associated with more benefit.[115]

Individual RCT, quantitative studies, and review articles support hypnosis for reducing child procedural pain and/or distress,[35,64,67,84,86–88,91,124–127] although some inconsistent evidence is also found.[92,104] Hypnosis is a state of heightened awareness and focused attention, often involving relaxation. It can be direct (focused on pain or analgesic suggestion) or indirect (focused on relaxation suggestion), with both showing reduced child pain and distress as compared with standard care.[35,125,127] Hypnosis can be therapist-led or child-led with training, although therapist-led interventions appear more effective.[127] Children with high levels of hypnotizability receive greater treatment benefit.[104]

Reduced pain and/or distress have also been reported for memory reframing,[10,128] supportive touch,[129,130] breathing,[131] behavioral training,[132] and art therapy,[133] but not biofeedback/relaxation alone.[134] Adult (parent, nurse, and physician) communication with children prior to, during, and following medical procedures is also critical.[93,135–141] Criticism, reassurance, empathy, invalidation, or vague commands were typically associated with increased child distress, whereas humor, praise, distraction, specific commands, and talking *with* rather than *at* the child were generally associated with decreased child distress and increased coping.

While the benefits of procedural preparation and support have been clearly demonstrated, limited evidence informs which strategy is best based on situation characteristics (i.e., child age, temperament, coping style, parent anxiety, and type of procedure). Additional challenges are helping healthcare providers, and families, recognize the benefits of preparatory and psychological intervention despite limited time and resources.

An overall summary of the evidence is available in Table I. Details regarding included studies and reviews such as study design, sample, findings, study rigor, and level of evidence are available in Supplementary Table II.

DISCUSSION

This comprehensive literature search found strong empirical support for providing children and adolescents with cancer with preparatory information and psychological intervention when undergoing painful medical procedures. The benefits are well established across the developmental spectrum for a variety of interventions (distraction, combined cognitive–behavioral, hypnosis) and procedures (BMA, LP, needle procedures) with little to no risk.

Further considerations include the importance of child age and developmental level in the appropriateness and implementation of various psychological interventions, as children's coping abilities and preferences develop significantly throughout childhood and adolescence.[142] Not all youth will require ongoing intervention after learning to cope and adapt, although they may continue to benefit from procedure preparation when undergoing any procedure for the first time. Making first procedures as comfortable as possible may prevent a learned response and the related ongoing distress that can ensue.[143] Appropriate procedure preparation and intervention can be implemented by child life specialists, psychologists, or other appropriate staff (e.g., nurses).[144,145] Child life specialists have been essential as primary providers of procedural preparation using many of the approaches developed and tested by psychologists. Psychologists and psychiatrists may be appropriate referrals for additional assistance when routine preparation and intervention have been ineffective.

Additionally, as medical care and treatment advances, there are changes in the way procedures are completed. Children used to endure procedures without appropriate analgesics. Now, more effective approaches to invasive painful procedures, such as BMAs and LPs, can be completed with integrative psychosocial and pharmacological interventions, including local and even general anesthetics. Procedural preparation by medical and psychosocial staff will need to adapt to the ongoing medical and treatment advances to ensure optimal coping for children, adolescents, and their families. Training in child development, assessment of anxiety, pain, and distress allows psychosocial and/or medical staff to appropriately identify who will need ongoing psychological intervention.[146]

CONCLUSIONS

This review sought to evaluate the evidence for providing preparatory information and psychological intervention to youth with cancer undergoing painful medical procedures. Surprisingly, there was low quality of evidence to support providing youth with preparatory *information* given the lack of studies within the reviewed period that exclusively investigated the provision of preparatory information in the absence of other psychological intervention (e.g., cognitive–behavioral strategies), the inconsistent benefits for reduced pain and distress reported in those studies, as well as study design (study limitations and reporting bias). Despite limited evidence within this review, the benefits are well established across the developmental spectrum for the provision of information, with additional supporting evidence available pre-1995 and with other pediatric populations. The recommendation that all youth should receive psychological *intervention* for invasive medical procedures is supported by high-quality evidence, given the numerous well-designed studies. There are strong recommendations for both of these standards given the risk–benefit ratio, including decreasing children's pain and distress and increasing coping and compliance during medical procedures.

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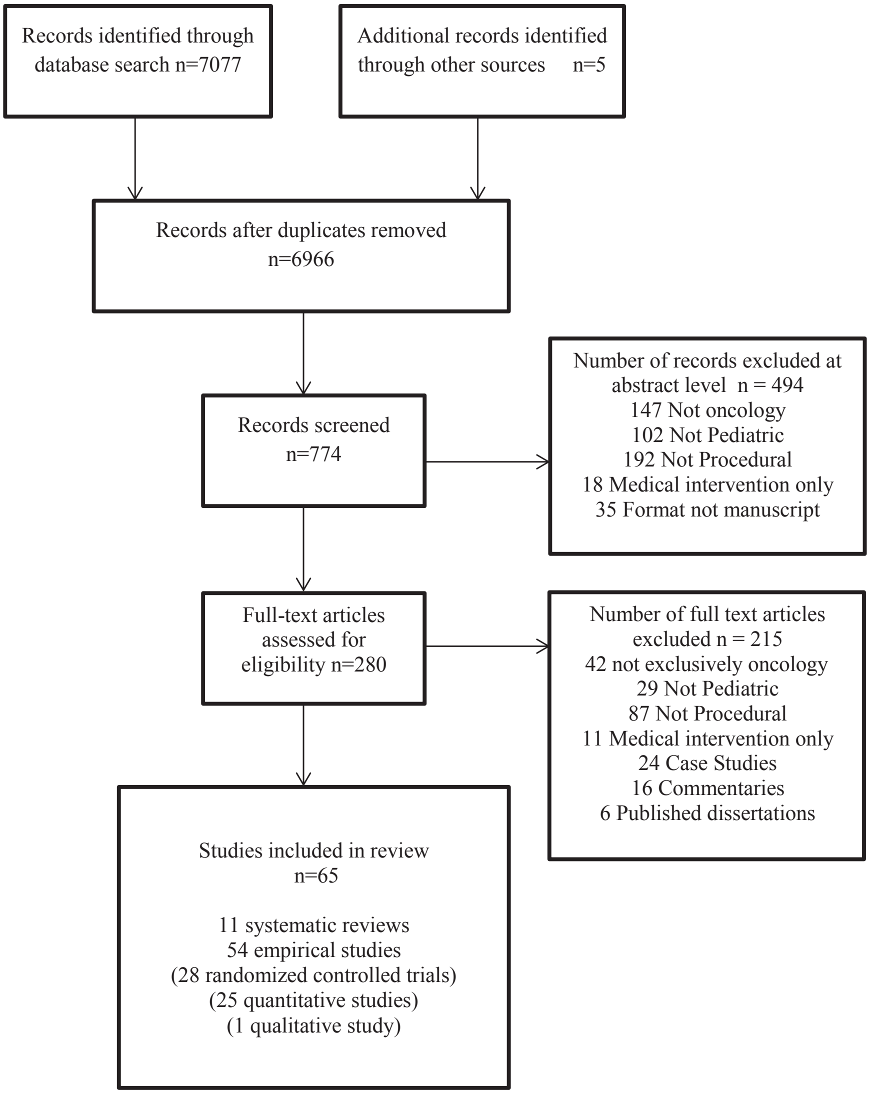
SUPPLEMENTARY INFORMATION



Supplemental Figure I. PRISMA Flowchart

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Providing Children and Adolescents Opportunities for Social Interaction as a Standard of Care in Pediatric Oncology

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Experiences with peers constitute an important aspect of socialization, and children and adolescents with cancer may experience reduced social interaction due to treatment. A literature review was conducted to investigate the evidence to support a standard of care evaluating these experiences. Sixty-four articles were reviewed using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) criteria. Moderate quality of evidence suggest that social interaction can be beneficial to increase knowledge, decrease isolation, and improve adjustment and constitute an important, unmet need. The evidence supports a strong recommendation for youth with cancer to be provided opportunities for social interaction following a careful assessment of their unique characteristics and preferences. Pediatr Blood Cancer 2015;62:S674-S677 © 2015 Wiley Periodicals, Inc.

Key words: pediatric oncology; psychosocial; support care

INTRODUCTION

Starting with the preschool years, experiences with peers can make up a large part of a child's daily life. These experiences can be sources of companionship, stimulation, information, help, rewards, security, joy, and, at times, frustration and harm. For at least seven decades, researchers have been testing hypotheses about the effects of peer interaction.[1] Evidence from population-based longitudinal studies has shown that experiences with peers constitute an important socialization domain for children and adolescents.[2] Specifically, it is known that experiences with peers affect how children and adolescents think about themselves, how they feel, and how they behave. Research evaluating the impact of childhood cancer on social functioning is mixed with some studies showing healthy functioning and other studies identifying children are at risk.[3–7] For example, school-aged children with cancer were found to be similar to peers on measures of emotional functioning [3] and better on multiple measures of social functioning.[3,4] In contrast, survivors who had central nervous system (CNS)-directed treatment and children with a history of a bone marrow transplant have been found to have poorer social functioning overall.[6,7] Also, survivors of childhood brain tumors experience reduced social adjustment.[5,8] Lown et al. (in this special issue) found that a small subset of survivors were more likely to report poor social support and have lower marriage rates compared to peers.[9]

Children and adolescents undergoing cancer treatment and into survivorship experience school absence [10] and subsequent reductions in social activities as well as report social isolation.[11,12] Children who are immunocompromised and socially isolated for long periods of time due to infection risks (e.g., following bone marrow transplant) may be especially vulnerable. Reduced social interactions can be particularly salient for adolescents, for whom development centers on establishing autonomy and self and social identities, as well as the heightened importance of peer relationships.[13]

Psychosocial Standard of Care

- Children and adolescents with cancer should be provided opportunities for social interaction during cancer therapy and into survivorship following careful consideration of the patient's unique characteristics, including developmental level, preferences for social interaction, and health status.
- The patient, parent(s), and a psychosocial team member (e.g., designee from child life, psychology, social work, or nursing) should participate in this evaluation at time of diagnosis, throughout treatment, and when the patient enters survivorship; it may be helpful to include school personnel or additional providers.

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METHODS

This review was performed as part of the collaborative Standards for Psychosocial Care of Children with Cancer and Their Families effort utilizing the Grading of Recommendations Assessment, Development and Evaluation (GRADE) methodology, a system to assess quality of evidence and strength of recommendations. For a full description of the methods used to develop the standard, please refer to Wiener et al. (in this special issue).[14] The literature search utilized three databases: PubMed, OVID, and PsycINFO (March 1, 1995 to March 1, 2015). Due to the limited body of work on the impact of social interaction on adaptation in children with cancer, the search terms were broadened to be inclusive of work that captures the social support needs of children and adolescents with cancer, their social and peer relationships, and interventions that promote social adaptation, including camps and groups. The search utilized the following indexed MeSH terms: "social support" OR "social distance" OR "interpersonal relations" OR "peer group" OR "self-help group" OR "psychotherapy group" OR "hospitalization" OR "camping" AND "neoplasm" OR "cancer." Searches were conducted utilizing the terms "child," "pediatric," "adolescent," and "young adult" to ensure all appropriate studies were captured. Nonresearch articles, with the exception of literature reviews or summaries, consensus, and opinion papers, studies with a primary focus on young adults or family functioning, or whose focus was not relevant to social interactions or peer relations, were excluded. Inclusion criteria included peer-reviewed English language articles. The reference lists of all included studies were hand-searched for additional relevant studies. Searches revealed a total of 710 citations. Authors followed PRISMA guidelines for systematic reviews, leaving 64 articles for inclusion in the synthesis of evidence (Supplementary Fig. I).

The authors of this standard are pediatric psychologists from the field of hematology/oncology. An external team of expert pediatric oncologists, pediatric and developmental psychologists, pediatric oncology social workers, and child life specialists, as well as members of an adolescents and young adult (AYA) panel and family advisors in oncology, reviewed the evidence and recommendation prior to the final draft. Their feedback echoed concerns regarding the limitations of the extant literature, the importance of carefully planned social interactions, and the need to propose specific strategies to overcome barriers; these have been addressed herein.

RESULTS

The search identified 64 peer-reviewed papers, including 26 quantitative studies, 28 qualitative studies, two systematic reviews of the literature, and eight consensus reports evaluating various aspects of social interaction and support. Supplementary Table I includes the studies that met inclusion criteria and briefly summarizes each study with regard to study design, sample characteristics, main findings related to this standard, study rigor, and the level of evidence. Based primarily upon findings from qualitative and descriptive studies, children undergoing cancer treatment often endorse feeling isolated during the treatment [11,15,16] due to the impact on social interactions.[17–21] Patients endorsed concerns that they cannot participate in activities, spend as much time with friends as they prefer,[15] feel different from peers as a result of cancer,[17,18] and sometimes withdraw from peers.[22] Children with cancer reported a desire for opportunities that promote social engagement and activity.[11]

Most adolescents with cancer describe the importance of peer support and the desire or need for social interaction to help them cope with active cancer treatment and survivorship care.[23–31] Some adolescents indicate a decrease in or difficulty with social interactions or lack of peer support, whereas others describe an improvement in social relationships due to cancer. [18,19,23,32–35] Adolescents express feeling socially isolated, disconnected, or different from their same-age peers, as well as more emotionally mature based upon their changed life perspective.[36,37] Importantly, adolescents with cancer report unmet needs in peer interaction and support, including a desire for increased access to cancer support programs (i.e., online or in-person support groups, retreats, and camps). [27,38,39]

Evidence regarding the outcomes of social interaction is limited and mixed. Several descriptive studies report that higher perceived peer support during cancer treatment is related to increased positive affect,[35] decreased anxiety and depression,[40–42] less uncertainty,[43] and increased ease during the transition back to school.[44] On the other hand, two descriptive studies found no significant relationship between peer support and psychological outcomes [45] or health-related quality of life.[46] Findings from qualitative research support that adolescents undergoing treatment find peer support to be a helpful distraction during intense phases of treatment.[25] For youth, connecting with other cancer patients or survivors is described as beneficial.[20,39,47–52] and they rate meeting other survivors as even more important than family or friend connections.[53]

Strategies to increase social interaction for children and adolescents with cancer have focused on traditional face-to-face support groups and camp interventions. Qualitative analyses found that participants in support groups,[34] teen outreach programs,[54] and organized hospital activities [55] obtain increased support and connectedness from these programs. Barriers to successful implementation of such groups include the broad age range of participants and treatment phase, potential death of group members, geography, and cost.[56] However, online forums may reduce access barriers and provide bidirectional emotional support among participants [57] and offer social connection with peers.[58,59] While videoconferencing and online groups and message boards decrease barriers to group participation and engage youth, some may prefer face-to-face groups. [60]

Camps increase interaction of youth with cancer and provide opportunities to share information and support. Literature reviews indicate that camp attendance is associated with improvements in self-confidence, independence, and social contact.[56,59,61] Individual studies evaluating camps are comprised primarily of nonrandomized, postcamp surveys, and interviews that highlight benefits, including camp is enjoyable,[62] increased cancer knowledge through participation,[63] diminished sense of isolation,[21] and improved mood.[64] One adolescent camp study reported reduced depression scores for patients 4–6 months after attending camp,[65] whereas another study did not find differences in adolescent adjustment after

TABLE I. Social Interaction as a Standard of Care in Pediatric Oncology Evidence

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
Children and adolescents with cancer should be provided opportunities for social interaction during cancer therapy and into survivorship following careful assessment of individual needs and social interaction preferences.	• Children and adolescents with cancer request opportunities for social interaction and support due to feelings of isolation. • While adolescents cite peers (at home, with cancer) as playing an important role in coping, they report social support as an unmet need. • Social intervention literature suggests that support groups and camp offer positive opportunities for social connectivity and overall adjustment. • Hospital environments can be structured to facilitate peer interactions and support.	• Mixed-methods, qualitative, quantitative, and literature reviews. • Majority cross-sectional retrospective survey and in-depth interviews. • Intervention trials were primarily single-arm qualitative studies using postintervention surveys or interviews.	Moderate quality given consistent findings without confounding variables from lower level evidence studies.	Strong recommendation given risk–benefit ratio, including need identified by children and adolescents with cancer, evidence that perceived social support is associated with adaptation, and lack of evidence of harm from intervention studies.

[1]Based on summary of evidence. [2]Types of studies: for example, RCT, cross-sectional, longitudinal; consensus; systematic review articles. [3]Quality of evidence: High, moderate, low, and very low. [4]Strength of recommendation: Strong or weak based on GRADE quality criteria.[71]

camp attendance.[62] Additional qualitative analyses suggest improved skills making friends,[65] enhanced normalcy,[66,67] and improved adaptation to cancer and its treatment.[67]

DISCUSSION

Although no randomized control trials and few quantitative studies have been conducted to evaluate the impact of social interactions and peer support on adaptation of children and adolescents with cancer, there is a considerable body of qualitative studies and surveys outlining social needs. Existing literature suggests that children and adolescents with cancer request peer support to promote coping, and this is an unmet need. There are limited data with mixed findings on the outcomes of social support, with some pointing to peer support being beneficial to mood and coping, and others finding no significant benefits. Results evaluating the impact of camp and support groups point to benefits of these activities; however, the extant intervention research involves small sample, single arm studies describing response to camp/support group interventions through the use of variables such as knowledge and physical behaviors.[62,65,66] Few account for baseline functioning, which include a comparison group or assess psychosocial functioning.

As such, systematic, controlled evaluation of interventions to support interactions with peers is a critical need. These social interactions may include peers from home or with cancer, or in the context of a therapeutic group or activity program such as camp. Interventions should be tailored to the developmental level and individual social interaction preferences. Finally, the efficacy of group interventions to improve specific skills, such as knowledge, coping and social skills, and self-efficacy/problem solving, needs to be evaluated. Consideration should be given to developing interventions that increase engagement of peers with children with cancer to mitigate social isolation.[47] Barriers to intervention, such as costs and access, may be addressed through the development of web-based/eHealth interventions. Opportunities for children with cancer to engage with peers with cancer and peers from home are preferred, whether it occur in person or electronically (e.g., Face Time and Skype).

CONCLUSION

Children and adolescents with cancer should be provided opportunities for social interaction during cancer therapy and into survivorship following careful consideration of the patient's unique characteristics, including the developmental level, preferences for social interaction, and health status. The patient, parent(s), and a psychosocial team member (e.g., designee from child life, psychology, social work, or nursing) should participate in this evaluation at the time of diagnosis, throughout treatment, and when the patient enters survivorship; it may be helpful to include school personnel or additional providers. Social interaction may be promoted through camps/activity programs, group interventions (e.g., face-to-face, eHealth), or structured hospital environments and activities, facilitated by a psychosocial team member. The hospital environment can be structured to promote social interaction. For example, visitation policies that allow for family and friend visitors when feasible given medical restrictions [68] as well as facilities that include group spaces to allow for peer-to-peer interactions [69] can promote social support.[16,70] Further, inclusion of adolescents in designing hospital spaces that facilitate connection and maintain a focus on social development goals is underscored.[50]

Current evidence for this recommendation is of moderate quality based on consistent evidence across low quality studies, primarily cross-sectional descriptive surveys, and in-depth interviews.[71] Overall, this is a strong recommendation given the risk–benefit ratio that weighs significant implications for mood, coping, adaptation, and health-related quality of life and lack of evidence of significant, negative consequences of interventions (Table I).

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Supporting Siblings as a Standard of Care in Pediatric Oncology

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In this study, evidence is provided for supporting siblings as a standard of care in pediatric oncology. Using Medline, PsycInfo, and CINAHL, a systematic search of articles published over the past two decades about siblings of children with cancer was conducted. A total of 125 articles, which were primarily descriptive studies, were evaluated by the four investigators using Grading of Recommendations Assessment, Development, and Evaluation (GRADE) criteria. There is moderate-quality evidence, as well as support from community stakeholders, to justify a strong recommendation that siblings of children with cancer should be provided with psychosocial services and that parents and professionals are advised about how to meet siblings' needs. Pediatr Blood Cancer 2015;62:S678-S682 © 2015 Wiley Periodicals, Inc.

Key words: adjustment; cancer; intervention; pediatric; sibling

INTRODUCTION

Siblings are exposed to significant stress when a child is diagnosed with cancer. Concern about the ill child, disruptions in family roles and routines, decreased contact with family members, and additional demands for caregiving or other responsibilities in the home are common.[1,2] In some cases, siblings are also called upon to serve as a donor for stem cell transplant, which can introduce other ethical and psychosocial concerns.[3] Taken together, these unique challenges leave siblings of children with cancer at risk for acute and long-term psychosocial difficulties. However, there are no current evidence-based standards for the supportive care of siblings of children with cancer.

Although severe psychopathology is rare, several reviews suggest that some siblings exhibit symptoms of anxiety, depression, posttraumatic stress; lower quality of life and healthcare utilization; and disruption to academic and social functioning.[1,2,4,5] Most difficulties improve over the first year after diagnosis, but they may resurface or worsen with declines in the ill child's health or death.[6] Siblings can also demonstrate resilient outcomes, such as posttraumatic growth.[4,7] This variability in adjustment underscores the need for accurate screening to identify risk and protective factors and to provide appropriate services for siblings vulnerable to difficulties. Unfortunately, siblings have unmet needs and psychosocial support may be limited.[8,9] Further, in a large survey of professionals from three pediatric oncology organizations, only 25% reported that they provide psychosocial services to siblings.[10] Thus, it is critically important to establish evidence-based standards of care for siblings of children with cancer in efforts to encourage the provision of more consistent and comprehensive services for this population.

METHODS

This review was performed as a part of the collaborative effort, Standards for Psychosocial Care of Children with Cancer and Their Families. A full description of the methods used to develop each standard is in the introduction to this special issue.[11] The literature search for this standard used three databases: Medline, PsycInfo, and CINAHL (March 1, 1995–March 1, 2015). Abstract search terms included keywords related to siblings AND childhood AND cancer AND psychosocial outcomes (see Supplementary Table I). The search was limited to peer-reviewed journal articles written in English and involving human subjects. Commentaries, opinion pieces, case studies, dissertations, and unsystematic reviews were excluded.

Psychosocial Standard of Care

Siblings of children with cancer are a psychosocially at-risk group and 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.

After duplicates were removed, the titles and abstracts of 3,205 citations were screened by two authors (see Fig. 1). Empirical studies or reviews, both qualitative and quantitative, that included siblings of children (aged 18 and below) diagnosed with cancer were retained for full text review (N = 278). No research exclusively examined outcomes for sibling donors of children with cancer who received stem cell transplant. Thus, eight articles that were nonspecific to cancer but included sibling donors were retained. The reference lists of retained systematic reviews were also hand-searched, resulting in the addition of six studies. In all, 106 empirical studies (74 quantitative, 32 qualitative), 16 reviews, and three guidelines were included in this report. The preferred reporting items for systematic reviews and meta-analyses (PRISMA) flow diagram is shown in Figure 1. The study team included four doctoral level psychologists (authors of this paper). The founder and current director of *SuperSibs!* provided community stakeholder input, and at least two physicians or psychologists from the larger standards project reviewed

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the manuscript and final recommendation for the standard of care prior to submission for publication.

RESULTS

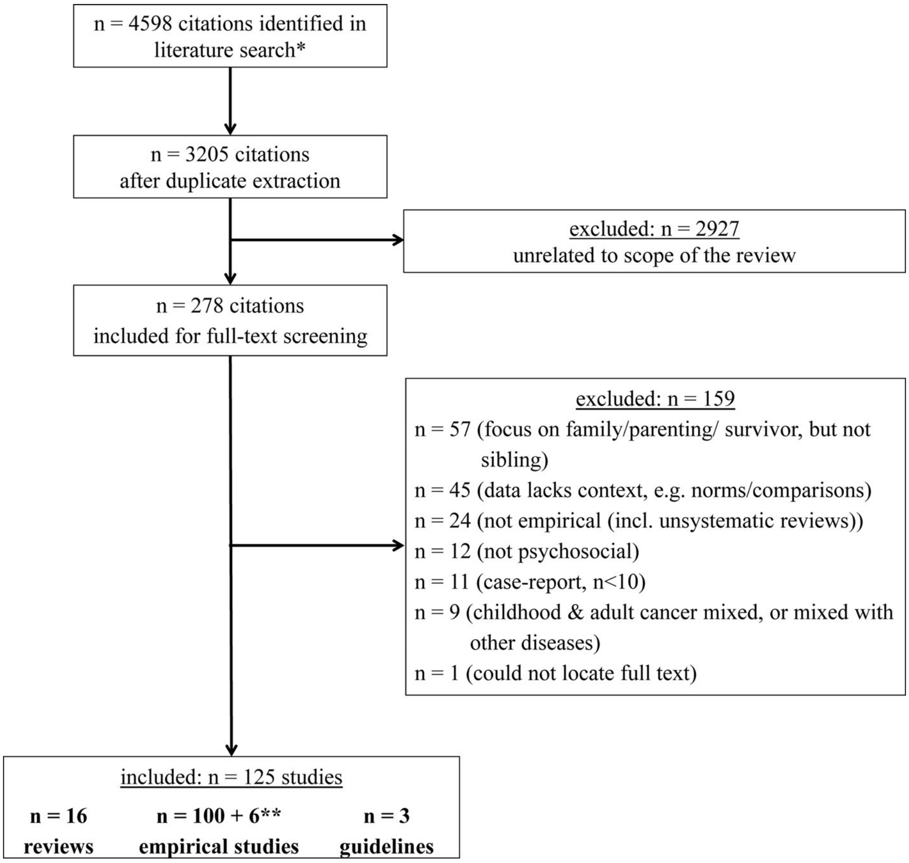
A summary of evidence is presented in Table I, indicating a strong recommendation based on the moderate quality of evidence and the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) system.[12] Detailed results from all included articles can be found in the Supplementary Table II. Several reviews have summarized the issues facing siblings of children with cancer.[1,2,4,5] Thus, selected studies are reported below to highlight examples of support for specific aspects of psychosocial care for siblings.

Sibling Stressors

When a child is diagnosed with cancer, siblings face significant disruption in their lives as evidenced by eight reviews or guidelines, six quantitative, and 11 qualitative studies. This stress includes additional demands for caregiving and other responsibilities at home, as well as experiencing diminished contact with the ill child and less attention from parents.[13–16] Challenges to maintaining normalcy and engaging in typical developmental activities also have been reported.[17,18] For example, siblings may be more likely to miss school compared to peers and fall behind academically,[18,19] although this may improve as treatment subsides.[20,21] They also describe the experience of having a brother or sister with cancer as a loss of their family's way of life and a loss of their sense of self.[1] Thus, these stressors may increase the risk for psychosocial difficulties for siblings of children with cancer.

Communication Needs

Communication with siblings is important over the course of the illness and beyond as indicated by eight reviews or guidelines, four quantitative, and eight qualitative studies. Siblings need information about the child's illness and treatment, as well as opportunities to talk about the impact of the illness on their lives and adjustment.[9,14,22] Psychosocial providers should work with the healthcare team and parents to facilitate their communication with siblings. Siblings should be updated regularly and provided with information about the disease and treatment in a developmentally appropriate manner. Based on one review, three quantitative, and five qualitative studies, siblings who serve as matched donors for stem cell transplant represent a special



* Medline: n=2709, PsycInfo: n=1035, CINAHL: n=854
** identified through bibliography search of included reviews;
(these were not identified in the literature search due to missing cancer-related key terms)

Fig. 1. Preferred reporting items for systematic reviews and meta-analyses (PRISMA) flow diagram demonstrating the article selection process.

TABLE I. Sibling Standard of Care: Summary of Evidence

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
Siblings of children with cancer are a psychosocially at-risk group and 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.	• Descriptive studies indicate family disruption and added stressors in the home increase risk for sibling difficulties. • Descriptive studies and guidelines for care suggest communication and involvement are important for siblings. • Controlled longitudinal research suggests risk for psychopathology is highest early in treatment and in the first 2 years after a child's death. • There is also evidence of resilience from qualitative and quantitative work. • A few intervention studies provide preliminary support for evidence-based practice.	• Mixed methods, qualitative, quantitative, and literature reviews. • Majority cross-sectional retrospective survey and in-depth interviews. • Replication of findings evident for descriptive studies. • Limited intervention trials.	Moderate quality given consistent findings from lower level evidence studies.	Strong recommendation given the risk–benefit ratio including significant psychosocial effects for some siblings and positive outcomes with appropriate intervention.

[1] Based on Supplementary Table II [2] types of studies, for example, RCT, cross-sectional, longitudinal, consensus, and systematic review articles [3] quality of evidence: High, moderate, low, and very low [4] strength of recommendation: Strong or weak based on GRADE criteria.[12]

circumstance in which communication, informed consent, and potential for distress should be evaluated and addressed.[3,23–25] They should be educated about tests and procedures, as well as the potential for success and/or failure of the transplant. The American Academy of Pediatrics advises attention to ethical issues associated with children who serve as donors,[26] and distress should be closely monitored by psychosocial providers during the course of planning for transplant, the procedure itself, and thereafter.

Psychosocial Impact

Evidence for the psychosocial impact of childhood cancer on siblings comes from 11 reviews or guidelines, 59 quantitative, and 12 qualitative studies. Research suggests that siblings of children with cancer are at risk for emotional and behavioral difficulties, such as anxiety, depression, and posttraumatic stress symptoms;[27,28] poorer quality of life and lower healthcare utilization;[29,30] and academic and social disruptions.[30,31] Siblings of children with cancer may also have higher rates of borderline and clinical range scores for internalizing, externalizing, and total problems (23–48%) relative to normative samples (16–17%).[30,32,33] However, psychosocial difficulties are not universal and severe psychopathology is rare.[20,21,34,35] Difficulties tend to be more common in the early phases of the illness and improve over the first year.[30,31] Some siblings can also exhibit resilient outcomes or enhanced functioning,[1,4,7] underscoring the need to identify those at risk in order to triage services. Findings are mixed, but factors such as older age, female gender, premorbid distress, lower social support, and family conflict have been linked to worse sibling outcomes in some cases.[28,35–38]

Bereaved Siblings

One review, five quantitative, and eight qualitative papers focused specifically on bereaved siblings. Siblings report a desire to be involved and informed when their brother or sister is at the end of life.[39,40] During this time, they report both positive and negative changes in themselves (e.g., sad, more mature) and their relationships with others (e.g., closer or more distant from others).[41] Bereaved siblings have been rated by both parents and teachers as having more internalizing and externalizing problems than norms or control groups,[42,43] and they can exhibit difficulties in peer relationships relative to classmates within the first 2 years of the death.[44] These concerns may diminish with time, but bereaved siblings have also reported that long-term outcomes (e.g., educational and career goals) may be affected by the loss.[6] This highlights the need to provide ongoing support to families, especially after a child has died.[45]

Supportive Care

The supportive care of siblings of children with cancer encompasses a broad spectrum of services, including assistance with family communication, psychoeducation, decision making for sibling donors, coping and prevention of psychosocial difficulties, as well as assessment and treatment of psychopathology. The intervention literature is limited but includes various attempts to address sibling needs and difficulties as evidenced by four reviews, 15 quantitative, and three qualitative papers. Most often sibling support groups or camps are described,[46–50] with only a few randomized controlled trials reported.[51,52] One review concluded that camp programs may enhance physical and emotional functioning (e.g., self-esteem),[53] while reviews including other types of interventions indicate improvements in knowledge, mood, and quality of life.[54,55] Given the preliminary nature of intervention research with siblings of children with cancer, reliance on evidence-based strategies derived from work with other populations may be necessary, especially when more severe psychopathology is evident.

BARRIERS

Barriers to provide psychosocial support to siblings include limitations in (i) availability of trained psychosocial staff and community resources, (ii) staff knowledge of issues faced by siblings; (iii) access to standardized screening tools to assess sibling distress and needs, (iv) healthcare providers' access to and communication with siblings, and (vi) intervention research to inform evidence-based care. Institutions should have adequate psychosocial staff (e.g., social work, child life, and psychology) and provide education and training to increase awareness of sibling issues. Periodic assessment and provision of services across the illness spectrum is recommended. Standardized screening tools should be combined with clinical interviews to enhance assessment. Services should be sensitive to the family context and developmental level of the sibling. Partnering with parents, extended family members, and other professionals (e.g., teachers and community-based providers) to anticipate and address sibling psychosocial needs is ideal. Flexibility in location and modality of care is often necessary as contact with siblings may be restricted due to hospital policy or for practical reasons. This is especially true after a child's death. Parent proxy report or phone contact with siblings may be alternatives to in-person meetings. Knowledge of resources for siblings and community referrals are important. In addition, controlled and longitudinal research is needed that includes multiple sites, methods, and informants, particularly in the evaluation of interventions for siblings.

CONCLUSIONS

Siblings of children with cancer are a psychosocially at-risk group and should be provided with appropriate supportive services. Parents and professionals close to the sibling should be advised about ways to anticipate and meet siblings' needs, even when they are at a distance. Data from 74 quantitative, 32 qualitative, and 19 reviews or guidelines affirm this recommendation. Current evidence for this recommendation is of moderate quality given consistent findings from lower level evidence and small-scale studies. Nevertheless, this is an overall strong recommendation given the risk–benefit ratio, including significant psychosocial effects for some siblings and the positive outcomes noted from intervention. Continued research with respect to the identification of groups at risk for psychosocial difficulties and the evaluation of interventions is warranted for siblings of children with cancer.

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Authors' contribution

C.A.G. and M.A.A. were responsible for the conception and design of this standard. All authors were responsible for the collection and assembly of data, data analysis, interpretation and presentation of data, drafting of the manuscript, and critical revisions for important intellectual content. All authors approved the final version of this manuscript and take public responsibility for the content presented in this article.

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SUPPLEMENTARY INFORMATION

SUPPLEMENTAL TABLE I. Database Search Terms Used in Medline, PsycInfo, and CINAHL

1. sibling* OR sister OR sisters OR brother OR brothers OR family OR families
2. childhood OR child OR children OR adolescen* OR pediatric OR paediatric OR youth
3. cancer OR cancers OR malignan* OR tumor OR tumors OR tumour OR tumours OR neoplasm* OR sarcoma OR sarcomas OR hodgkin* OR leukaemi* OR leukemi* OR lymphom* OR non-hodgkin* OR oncolog* OR hematolo*
4. psychosocial OR psychiatric OR psycholog* OR adjustment OR adaptation OR distress OR stress OR social OR school OR anxiety OR depression OR grief OR grieving OR bereave* OR mourning OR well-being OR "quality of life"
5. 1 AND 2 AND 3 AND 4
6. limit 5 to peer-reviewed journals, published 1995–2015, English language, human, exclude dissertations [PsycInfo, CINAHL]
   limit 5 to published 1995–2015, English language, human [Medline]

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Academic Continuity and School Reentry Support as a Standard of Care in Pediatric Oncology

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Clinicians agree that return to school after diagnosis promotes the positive adjustment of children and adolescents with cancer; however, the school reentry process can present challenges. The aim of this review was to critically evaluate the literature on school reentry support for youth with cancer. Seventeen publications were identified. School reentry services were well-received by families and educators; increased teacher and peer knowledge about childhood cancer; influenced peer and educator attitudes toward the patient; and improved communication and collaboration between patients/families, school, and the healthcare team. Evidence supports a strong recommendation for school reentry support for youth with cancer. Pediatr Blood Cancer 2015;62:S805–S817. © 2015 Wiley Periodicals, Inc.

Key words: childhood cancer; psychosocial; school reentry

INTRODUCTION

Children and adolescents diagnosed with cancer are frequently absent from school because of treatment and treatment-related side effects.[1,2] Absences can be a problem both during and after treatment but are most pronounced in the year after diagnosis.[1] Although empirical support is limited, clinicians agree that a return to the student's community school can facilitate a sense of normalcy, improve health-related quality-of life, and promote positive adjustment, academic progress, and socialization of the child or adolescent with cancer.[3,4]

"School reentry" refers to the process of returning to school after diagnosis and/or treatment for cancer[5] and can present challenges for the healthcare team, patients, classmates, parents, and teachers. Healthcare teams report being unsure how to help parents navigate the school system.[5] Patients may worry about their physical appearance or fear that they would not be able to keep up with activities, while peers may have concerns about catching the disease.[1,3,6] Some parents report concerns about safety and teasing; they are unclear about their role in school reentry[1,3,6] and feel that their children are not receiving all the school services needed.[5] Upon reentry, some parents report that schools are unsupportive toward their child's special needs or, alternatively, are

Psychosocial Standard of Care

- In collaboration with parents, school-aged youth diagnosed with cancer should receive school reentry support that focuses on providing information to school personnel about the patient's diagnosis, treatment, and implications for the school environment and provides recommendations to support the child's school experience.
- Pediatric oncology programs should identify a team member with the requisite knowledge and skills who will coordinate communication between the patient/family, school, and the health care team.

Abbreviations: BASC-2, behavioral assessment system for children second edition; QoL, Quality-of-life

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Authors' Contributions: This work was conducted collaboratively as part of the *Standards for Psychosocial Care of Children with Cancer and their Families* effort. As such, all authors participated in the conception and design of this standard, the collection and assembly of data, data analysis, interpretation and presentation of data, drafting of the manuscript, and critical revisions of content. All authors approved the final version of this manuscript.

Conflict of interest: Author MKI is the current Present of APHOES and author ME is Committee Chair of the APHOES Legislative Committee. ME is also the Vice President of the Division of Physical, Health, and Multiple Disabilities within the Council for Exceptional Children. While their participation in this standard development was critical (given their content expertise), lead authors ALT and RBN took care to reduce any possible bias by strict adherence to evidence based review, wording and GRADE assessment for each standard. MKI and ME participated in review of literature, developing standards and in preparing final manuscript; however, the entire writing team shaped and approved final working of each standard statement such that individual authors could not exert independent influence on how each was worded. Additionally, MKI and ME did not participate in the external reviews conducted by other members of their respective organizations. Communication about these reviews occurred between organization contact and authors RBN and ALT exclusively.

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overly accommodating of the student.[7,8] Despite these concerns, data from teachers and peers suggests that the majority of children return to school and fit in well with their peers.[9]

Given the rarity of childhood cancer, it is not surprising that educators report having little or no training or experience in working with children with cancer.[5,10] As a result, teachers worry about their lack of knowledge about cancer and how other children in the classroom will adjust.[1,3,6] They may feel unprepared to support the educational needs of students with a chronic condition such as cancer.[11,12] Educators desire training and have reported that if they received specific guidance on how to help patients returning to school, they would be more consistent, patient, understanding, and involved in providing support to these students.[7,13]

The Association of Pediatric Hematology Oncology Educational Specialists (APHOES) and the International Society of Paediatric Oncology (SIOP) recommend that school support for students with cancer begin at diagnosis, that school reentry programs be offered, and that clear communication between school and hospital personnel be ongoing.[14,15] Despite these recommendations, an evidence-based standard of care has not yet been established. There are a wide range of school support programs and approaches (e.g., reentry programs, hospital-based schools, homebound instruction, use of videoconferencing technologies) that are designed to mitigate the impact of childhood cancer on the school experience. As most have not been studied systematically in pediatric cancer, this review focuses specifically on school reentry support for school-age youth (ages 4–18) who are returning to a community school after initial diagnosis and treatment for a malignancy. Recommendations for school reentry described here are predicated on the assumption that children with cancer will return to school in the community as soon as they are medically able, although there is considerable variability between individual providers (i.e., pediatric oncologists) and across oncology programs regarding what constitutes a "timely" return to school.[16] In addition, return to school is dependent upon family comfort, which is also quite variable.

METHODS

To develop this standard, we used methods described by Wiener et al.[17] in this special issue for the Standards for Psychosocial Care of Children with Cancer and Their Families project. Our search employed four databases: PubMed, PsychInfo, CINAHL, and ERIC. Search criteria included English-language, peer-reviewed literature published from March 1, 1995 to March 1, 2015, with participants ages of 4–18 and a history of any malignancy. Exclusion criteria eliminated literature that was not empirical research (with the exception of consensus statements from expert panels) and literature about non-cancer diagnoses, patients over age 18, and foreign language publications. Articles were retained that included children with cancer as one disease group among other illnesses. Specific search terms included "school reentry," "school reintegration," "school intervention," "school liaison," OR "schools" AND cancer-related terms AND "child" OR "adolescent" OR "pediatric" OR "paediatric" OR "youth" OR "children" (using indexed MeSH terms). Searches were supplemented with a manual review of the reference lists of included studies and ultimately resulted in a total of 529 citations. Authors followed PRISMA guidelines, leaving 17 articles for inclusion in the synthesis of evidence (Figure 1 in Supplemental Materials).

The study team was comprised of representatives from the fields of psychiatry, psychology, nursing, and education. External reviews were conducted by members of APHOES and the Council for Exceptional Children's Division of Physical, Health, and Multiple Disabilities, an attorney at an Education Law Center, a school administrator, and parents and survivors of childhood cancer.

RESULTS

The search strategy identified 17 peer-reviewed papers, including two meta-analyses and one systematic review of the literature. This literature is summarized in Table I in Supplemental materials. Previous seminal work on school reentry that preceded the selected search timeframe was captured and synthesized in the meta-analyses included in this review.[18,19] Studies indicated that school reentry efforts, in their various formats, were well-received, well-accepted, and deemed helpful by parents and educators.[10,20,21] In general, school reentry programs and approaches varied widely across studies but commonly targeted parents, school personnel, or the patient's classmates, rather than the patients themselves. Programs typically included written, electronic, or in-person communication about diagnosis and treatment, its impact on the school experience, and suggested services of accommodations. For more detailed description of school reentry services in the reviewed studies, please refer to Supplemental Materials, Table II.

Across nine publications, including two meta-analyses,[18,19] one systematic review,[1] two individual quantitative studies,[10,22] and four qualitative studies,[6,23–25] findings consistently indicated that school reentry programs increased educators' knowledge about the medical and psychosocial aspects of cancer, led to more positive teacher attitudes toward the child with cancer, and increased teachers' confidence and comfort levels managing issues encountered by patients with cancer who are returning to school. Of note, one study[6] reported that increased knowledge about pediatric cancer might inadvertently increase worry and concern by teachers regarding side effects and academic achievement (although it should be noted that increased levels of worry, when appropriately directed, might result in more effective school support for the child with cancer). Additionally, two studies found that educators' increased knowledge about diagnosis and treatment improved their ability to provide more comprehensive educational programming suited to students' specific needs.[23,25]

Similarly, four studies, including two meta-analyses,[18,19] a systematic review,[1] and an individual qualitative study,[24] indicated that school reentry programs increased peers' knowledge concerning the medical and psychosocial aspects of cancer and improved peers' attitudes toward and increased interest in interacting with the student with cancer. In a meta-analysis of six intervention studies, increased knowledge among classmates was found to be associated with less fear of and a more positive attitude toward the child with cancer.[18]

Evidence for the impact of school reentry support on the patient is limited, and findings are less consistent than research assessing the impact on school personnel and peers. Helms et al.[18] reported that school reentry support both enhanced the academic achievement of and lowered levels of depression in students with cancer. In small qualitative studies, parents reported decreased peer teasing[6] and improvement in their child's social adjustment and learning.[23] Additionally, a quality improvement study of a school liaison program for pediatric cancer survivors reported that those in the program were more likely to be receiving special education services,[20] which may indicate increased access to noteworthy school supports. In a feasibility

TABLE I. School Reentry Standard Summary of Evidence Table

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
a. In collaboration with parents, school-age youth diagnosed with cancer should receive school reentry support that focuses on providing information to school personnel about the patient's diagnosis, treatment, and implications for the school environment and provides recommendations to support the child's school experience b. Pediatric oncology programs should identify a team member with the requisite knowledge and skills who will coordinate communication between the patient/family, school, and the health care team	School reentry programs and services were well-liked and appreciated by patients, families, and educators; increased teacher and peer knowledge about childhood cancer; influenced peer and educator attitudes toward the patient returning to the classroom; and required significant communication and collaboration between patients/families, school, and the health care team Existing studies had methodological and conceptual weaknesses, including small sample sizes, lack of control groups, lack of randomized controlled trials, and lack of follow-up data regarding effectiveness and impact on patient's adjustment	Pre-post test designs, qualitative, quantitative, meta analyses, and a systematic literature review. No randomized controlled trials. Consistent findings evident	Low quality given consistent findings from lower level evidence studies	Strong recommendation given risk-benefit ratio (i.e., minimal risk to patients, families and educators and potential benefits of improving the child's teachers' and classmates' understanding of the illness and opinions about the child with cancer)

[1]Based on summary of evidence table for that standard; [2]Types of studies: e.g. RCT, cross-sectional, longitudinal; consensus; systematic review articles; [3]Quality of evidence: High, moderate, low, and very low; [4]Strength of recommendation: Strong or weak (based on GRADE quality criteria).

study of a 4-month reentry intervention, parent-report on the Behavioral Assessment System for Children (BASC-2) were normal at pre- and post-testing, but quality-of-life (QoL) decreased over the course of the study;[26] this finding, however, may be due to expected decrements in QoL over the first months of treatment.

Evidence about the impact of school reentry support on parents was also very limited. Three separate qualitative studies reported that school reentry support may strengthen parents' advocacy skills for their child in the school setting[23] and decrease parent concerns related to peer teasing[6] but have no impact on parent concerns regarding their child's safety.[6] Communication and close collaboration among medical staff, school personnel, and families was identified as a critical component of providing effective services to students.[5,7,21,27,28] Stakeholders reported that educators need to keep in touch with children as they recover, that parents and teachers must work together to facilitate a smooth transition back to school, and that support from teachers, tutors, and the hospital staff was instrumental in creating a positive school re-entry experience.[27,28] To support collaboration and address communication challenges, several reviewed studies suggest a designated team member (e.g., NP, school liaison) may be helpful.[5,15,20,23]

DISCUSSION

Our review suggests that school reentry support should be provided to youth diagnosed with cancer by a well-trained, experienced pediatric oncology team member who will coordinate communication between the child/family, school, and health care team and should, at a minimum, focus on providing information to school personnel about the impact of disease and treatment on the school experience. Support may include verbal/written communication with the school, an individualized academic plan, guidance for parents around resources and processes, a school visit to educate peers and school personnel, educator workshops, or formal school liaisons. Two studies documented a positive impact of comprehensive school liaison programs,[20,23] and while these results are promising, further study must be conducted before recommendations can be made about this specific model of support. If resources are available, the use of a hospital-school liaison with expertise in both education and medical systems may help to bridge the gap in communication and increase coordination of efforts across systems and stakeholders.[29]

Methodological and conceptual weaknesses of the current evidence base limit the ability to draw strong conclusions about the impact or effectiveness of school reentry support. In general, studies were rated as low to very low quality evidence because of small sample sizes, lack of control groups, and the lack of randomized clinical trials or between-site comparison trials. Outcomes measures were psychometrically limited and focused on peer knowledge or satisfaction of teachers and/or parents, with little work examining metrics such as numbers of children on 504 plans or Individualized Education Programs (IEPs). Neither study methods nor reentry approaches were informed by a clear theoretical basis or model; as a result, goals and outcomes of school reentry have been unclear to date. There were no standardized approaches to school reentry support (Supplemental Materials, Table II). Programs varied by content, who conducted the program, and to whom the interventions were directed. Finally, there is lack of evidence for improved social or academic outcomes when children receive school reentry services.

Although decisions regarding return to school are dependent on pediatric oncologists and the comfort of caregivers, there is agreement among parents, health care team members, and professional/advocacy organizations (e.g., APHOES, SIOP) that children with cancer can benefit from strategic support to facilitate school reentry.[15,30] Additional research, however, is needed to direct best practice. Future research should address optimal timing and necessary components of support; impact of school reentry support on social or academic outcomes for children with cancer; potential negative effects or unintended consequences on patients and peers; and best practices for providing ongoing educational assessment and support for students with cancer beyond the return to school after diagnosis.

Current research focuses primarily on younger school-age children; research on best practices for students in middle and high-school, when there are unique academic challenges and complexities (e.g., more classes, teachers, and independence), was very limited. Evaluating which components of support are most beneficial to patients will aid in determining allocation of limited financial and personnel resources at childhood cancer centers across the country. Specific focus should be given to patients with brain tumors, who are at risk for significant academic,[31] and social difficulties[32,33] and therefore may require more intensive support in school and interventions that are different in scope, timing, and content than those that may be beneficial for patients with other diagnoses. Research noting social isolation, victimization, and low social acceptance of children surviving brain tumors[32,33] highlights the need for school reentry or liaison programs to mitigate poor outcomes for this vulnerable population.

The most significant organizational barrier to implementation of this standard is cost of programming and personnel. Institutional resources often limit availability of personnel dedicated to school support, as programming is non-revenue generating and thus may be perceived as cost-prohibitive.[34] Another barrier to implementation is large patient volumes and/or centers with large catchment areas that span multiple states and many school districts[3] which may present logistical challenges at the organizational level. Developing procedures and materials to educate school personnel from a distance (i.e., through written, telephone, or electronic communication) may prove helpful in addressing these barriers, but current research in this area is non-existent.

Overall, the current evidence regarding the value of school reentry programs is of low quality based on our assessment of the scientific rigor of the reviewed studies. Findings across studies, however, consistently demonstrated positive endorsement of school reentry programs by parent and education stakeholders and improvements in teacher and classmate understanding of the illness and opinions about the child with cancer. Given these consistently reported benefits of school reentry support, the minimal risk this support poses to the child with cancer, their family, their classmates and school personnel, and the potential harm to the patient in not providing this support, we strongly recommend that children with cancer be provided with school reentry support after diagnosis by a member of the childhood cancer care team (Supplemental Table I). Currently, there is a notable lack of evidence to endorse the essential elements of school reentry support, including the optimal type and timing of interventions and the necessary expertise or qualifications of personnel implementing the interventions and coordinating support.

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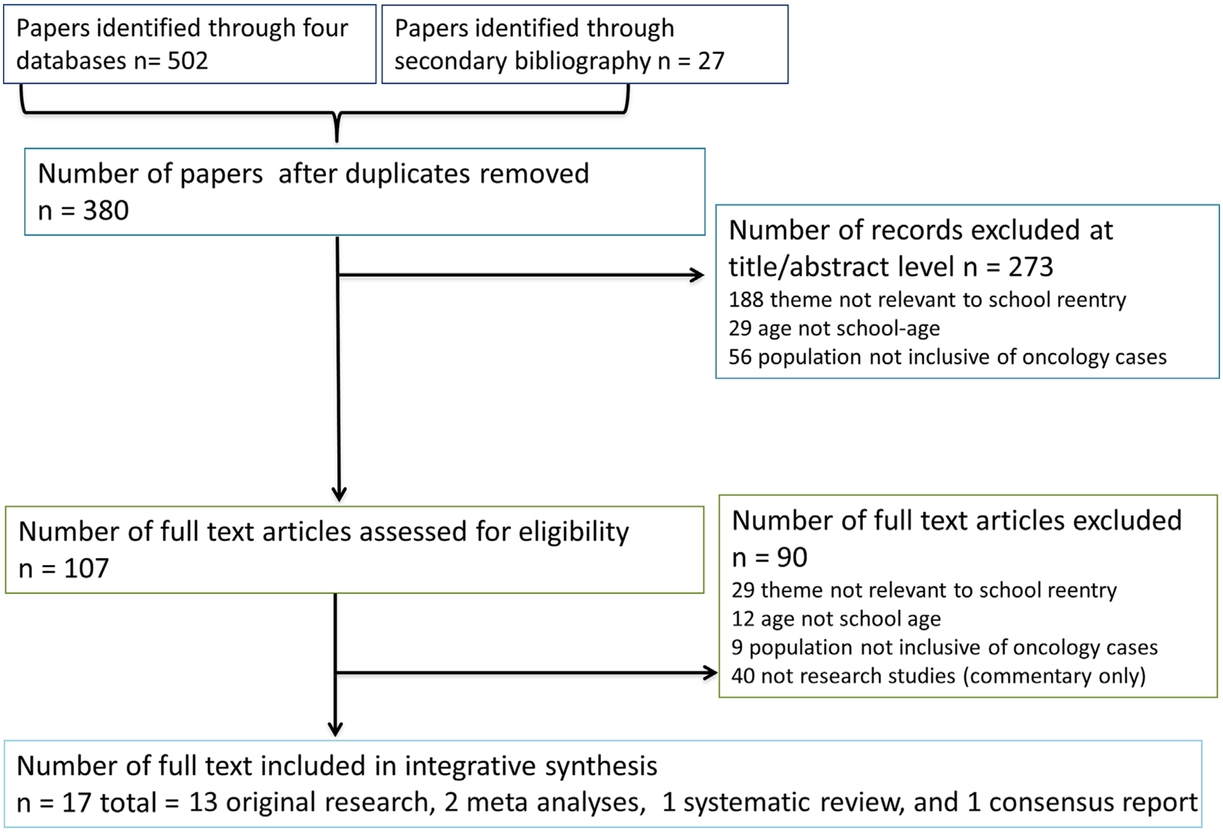
SUPPLEMENTARY INFORMATION

SUPPLEMENTAL FIGURE 1. PRISMA Table: Preferred Reporting for Systematic Reviews and Meta-Analyses



Pediatr Blood Cancer 2015;62:S696–S699

Assessing Medication Adherence as a Standard of Care in Pediatric Oncology

Ahna L. H. Pai, PhD[1,2]* and Meghan E. McGrady, PhD[1,2]

Poor adherence to pediatric cancer treatment protocols may prevent children and adolescents from realizing the potential benefits of therapy. This paper presents the evidence for a standard of care for supporting medication adherence. Databases were reviewed for articles examining adherence and including children and/or adolescents with cancer. Fourteen articles (i.e., qualitative, quantitative, review, and randomized clinical trials) were evaluated for rigor. There is moderate-quality evidence to support a strong recommendation for adherence to be assessed routinely and monitored throughout the treatment. Integrating the proposed clinical procedures into standard clinical care may improve outcomes for children and adolescents with cancer. Pediatr Blood Cancer 2015;62:S696-S699 © 2015 Wiley Periodicals, Inc.

Key words: adherence; adolescent; cancer; child; oncology; self-management

INTRODUCTION

Children and adolescents are diagnosed with cancer and their families are often required to self-manage a complex treatment regimen including multiple medications administered at varied dosing schedules. In other chronic medical conditions, the complexity and prolonged duration characteristic of many pediatric oncology protocols are associated with high rates of nonadherence.[1] Children and adolescents with cancer, thus, may be at particular risk for medication nonadherence.

Consistent with findings from the World Health Organization citing medication nonadherence as one of the greatest threats to suboptimal health outcomes and treatment failure among patients with a chronic medical condition,[2] poor medication adherence among children and adolescents with cancer is associated with adverse health outcomes (i.e., increased risk of relapse).[3,4] While these findings suggest that anticipating, assessing, and promoting treatment adherence are critical components of comprehensive clinical care,[5] few empirically based guidelines exist for providers seeking to incorporate these procedures into their practice. To address this critical gap, the purpose of this paper was to review the literature examining medication adherence among pediatric cancer patients and develop evidence-based guidelines for supporting medication adherence in clinical care.

METHODS

This review was performed as a part of the collaborative Standards for Psychosocial Care of Children with Cancer and Their Families effort. Grading of Recommendations, Assessment, Development, and Evaluation (GRADE) criteria were used to assess the quality of evidence in the identified studies.[6] For a full description of the methods used to develop each standard, the reader can refer to Wiener and colleagues in this special issue.[7]

Literature Search

PubMed, PsycINFO, Google Scholar, OVID, and EBSCO HOST (i.e., Academic Search Premier, CINAHL, ERIC, MasterFILE Premier, MEDLINE, Psychology and Behavioral Sciences Collection, TOPIC search) were searched for research articles published in English from March 1, 1995 to March 1, 2015. Search strategies included a combination of terms and MeSH headings related to adherence and neoplasm (see Supplementary Table I). Articles obtained via the database searches were supplemented with relevant articles included in the bibliographies of systematic reviews. The inclusion and exclusion criteria for the larger standards project were used with one exception. As predictors and correlates of nonadherence among young adults may have implications for patients under 18 years of age, articles with an age range extending into the young adult period (19–29 years of age) were not excluded as long as the majority of patients were under 18 years of age. Data detailing the study design, sample, and primary findings were extracted by the authors using an abstraction form developed for this study. A total of 14 articles met inclusion criteria and then used to develop clinical standards for supporting medication adherence (see Fig. 1).

Psychosocial Standard of Care

Adherence should be assessed routinely and monitored throughout the treatment.

External Reviews

The proposed standard and supporting evidence were then reviewed by external reviewers through the Second Think Tank for the Development of Psychosocial Care Standards for

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Abbreviations: 6MP, 6-mercaptopurine

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Conflict of Interests: Nothing to declare.

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Children with Cancer and Their Families. Minor editorial changes were recommended and incorporated.

RESULTS

There is an emerging body of evidence supporting routine adherence assessment, monitoring, and intervention into standard clinical care in pediatric oncology ($n = 14$ studies and reviews). A summary of evidence is presented in Table I, indicating a strong recommendation based on the quality of evidence and the GRADE system.[6] Study methodology varied across included studies, with four prospective studies, three cross-sectional studies, one qualitative, one randomized clinical trial, three systematic literature reviews, one narrative review, and one guideline developed by experts. Detailed results from the articles meeting inclusion criteria are presented in Supplementary Table II. Selected studies are reported below to highlight the importance of assessing medication adherence throughout the treatment trajectory.

DISCUSSION

Research findings from quantitative, qualitative, and review studies suggest that best practices for promoting medication adherence among children and adolescents with cancer will likely require a multifaceted approach. To achieve this goal, multidisciplinary teams are encouraged to integrate medication adherence-related assessments, education, anticipatory guidance, and documentation into standard clinical care. Each of these recommendations is outlined below.

Self- and parent-reported assessments of medication adherence should be obtained routinely using standardized language that assesses each specific medication for a specific period of time (e.g., In the last 7 days, how many times have you [has your child] missed a dose of [INSERT MEDICATION NAME] by a designated member of a multidisciplinary team?).[8] The high rates of medication nonadherence (19–53%),[4,9–15] especially among adolescents,[9,11,14] support the implementation of routine and standardized assessment of medication adherence. Assessing nonadherence may identify patients at risk for suboptimal treatment outcomes as nonadherence is associated with an increased risk of relapse and mortality. In pediatric acute lymphoblastic leukemia, 6-mercaptopurine (6MP) adherence rates lower than 95% are associated with a significantly increased risk of relapse.[4] Similarly, adolescents with cancer who are nonadherent to trimethoprim/sulfamethoxazole have lower survival rates than adherent adolescents.[11] As self- and parent-reported rates of nonadherence typically differ,[11] providers are encouraged to administer standardized measures of adherence (e.g., medication adherence measure) [8] to patients who are functioning at the developmental equivalent of 12 years of age or older.

Adherence behaviors occur in the context of daily life and an ongoing developmental course and are thus susceptible to changes in daily routines and family systems. Therefore, mediation adherence should be assessed and monitored routinely

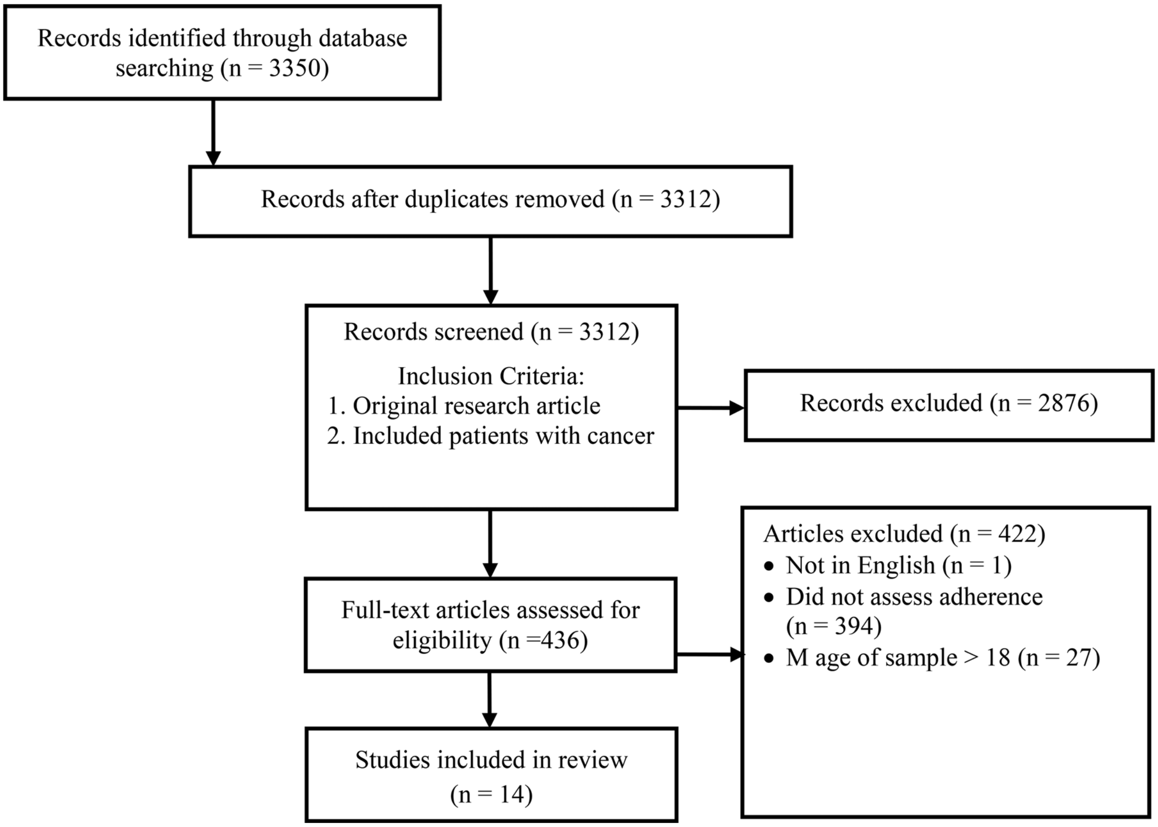

Fig. 1. PRISMA diagram of literature search and article selection.

TABLE I. Summary of Evidence—Adherence

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
Adherence should be assessed routinely and monitored throughout the treatment.	Empirical research for children with cancer indicates prevalent and significant difficulties with adherence to medication regimens. *Evidence gaps*: Additional randomized clinical trials are needed to determine how to best promote adherence to medication regimens among youth with cancer.	Cross-sectional; longitudinal studies; systematic review articles show significant replication of findings; one randomized clinical trial.	Moderate-quality evidence. Evidence from RCTs with important limitations (methodological flaws, indirect evidence, or imprecise results) or unusually strong evidence from unbiased observational studies.	Strong recommendation, given the prevalence of adherence difficulties and relationship between poor adherence and poor disease outcomes. Desirable effects clearly outweigh undesirable effect or vice versa. Recommendation can apply to most patients in most circumstances. Further research (if performed) is likely to have an important effect on our confidence in methods to promote adherence.

[1] Based on the summary of evidence table for that standard; [2] types of studies: for example, randomized clinical trial (RCT), cross-sectional, longitudinal; consensus; systematic review articles; [3] quality of evidence: High, moderate, low, and very low (based on GRADE criteria); [4] strength of recommendation: strong or weak (based on GRADE quality criteria).

throughout the course of treatment. For children and adolescents with leukemia or lymphoma who are prescribed two doses of 6MP per day, adherence decreases as rapidly as 6% per day, with rates falling as much as 40% over time.[15] As children enter adolescence, the variability in the timing of medication administration often increases and nonadherence becomes even more prevalent.[14,15] To capture the anticipated changes in medication adherence over time, providers are encouraged to assess and monitor medication adherence at each follow-up outpatient clinic visit.

Developmentally appropriate education about the purpose, administration, and side effects of each medication and importance of medication adherence should be provided to youth with cancer and/or their family immediately prior to the transfer of self-management responsibilities to the patient and/or family and whenever there is a change in the medication regimen. Adolescents with cancer and their families cite disease knowledge and cancer care skills as the foundation for medication regimen self-management.[16] The critical role of education is further supported by results of a systematic review indicating that patients who receive interventions with an educational component demonstrate higher rates of medication adherence, based on blood tests for metabolites, than patients in control conditions.[17] Therefore, developmentally appropriate regimen education is a core component of adherence care.[18]

Anticipatory guidance including a discussion of common barriers to adherence, previous experiences taking medication, and strategies to improve medication adherence should be provided immediately prior to the transfer of self-management responsibilities to the patient and/or family and whenever there is a change in the medication regimen. Children and adolescents with cancer and their families describe multiple barriers to medication adherence that can be addressed with behavioral intervention including forgetting, being away from home when doses are due, difficulty in swallowing pills, taste of medications, and not feeling well.[19] Observational studies suggest that interventions targeting family support [19] and patient psychosocial functioning (i.e., depressive symptoms) [11] may improve adherence. As behavioral and multicomponent interventions providing such guidance are more effective in improving medication adherence than educational interventions alone [20] and have been shown to improve self-efficacy, cancer-related knowledge, and adherence,[21] clinicians are encouraged to consider partnering with relevant disciplines (i.e., psychology, social work, and child life) to develop procedures for assessing barriers and delivering behavioral interventions as appropriate.

Adherence-related assessments, education, and anticipatory guidance should be documented. Specific and distinct documentation outlining assessment results, the education provided, and interventions to promote medication adherence should occur every visit. This practice facilitates the tracking of patients' adherence, progress toward adherence-related goals, and the supports in place to optimize adherence.[22]

Barriers to integrating adherence care INTO practice. Despite the critical importance of supporting medication adherence, integrating the above recommendations into clinical care has been hindered by several logistical and systemic barriers. Most fundamentally, standardized approaches of obtaining self-reported adherence are rarely used.[2] However, assessing adherence is critical in setting the stage for open dialogue between patients and providers regarding the difficulties inherent to maintaining high levels of adherence.[23] Training providers about the complexity of adherence behaviors, how to assess adherence (e.g., In the last 7 days, how many times have you [has your child] missed a dose of 6MP?), and the importance of routinely assessing adherence can facilitate the delivery of appropriate adherence care.

Another barrier to adherence assessment in pediatric oncology is the concern that assessing adherence behaviors could compromise the patient–provider relationship. One method of addressing this concern is to incorporate standardized adherence assessments into each clinic visit. This practice reduces the stigma associated with discussing medication adherence. When providers acknowledge the common barriers to medication poor adherence and importance of an open dialogue from the onset of treatment, patients and families may be more likely to disclose adherence difficulties and as a result, receive the support and resources they need to address any concerns.

Finally, effective interventions to target barriers to medication nonadherence require predetermined systems for adherence care and well-coordinated care plans. Providers lacking a specific plan for identifying and intervening on the factors contributing to poor adherence may be less likely to assess adherence. To overcome this barrier, providers are encouraged to work with members of the multidisciplinary team (i.e., physicians, nurses, psychology, social workers, and child life specialists) to establish a plan for supporting adherence and intervening when there are adherence difficulties. Plans should include identifying the specific team members responsible for providing interventions for specific adherence barriers. For example, a team could have a nurse or care manager to address patient lack of medication regimen knowledge by providing education, a psychologist to address emotional or behavioral barriers to adherence using cognitive and or behavioral therapies, and child life therapists to address pill-swallowing difficulties contributing to poor adherence. This example is far from comprehensive as each institution will assign roles in adherence care based on local resources and in some cases may need to utilize community agencies to provide support for adherence behaviors.[24]

While this review demonstrates that the existing quality of evidence is moderate, we strongly recommend the integration of routine adherence monitoring into the standard of care in pediatric oncology given the consistencies in evidence between pediatric oncology and the larger pediatric literature to date.[18] Moreover, poor adherence is common and can have adverse and life-threatening outcomes for youth with cancer. Therefore, adherence to oral medication regimens should be assessed routinely and monitored throughout the treatment in youth with cancer. The integration of these recommendations into standard clinical care could optimize treatment adherence and ultimately the health outcomes of youth with cancer.

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Pediatr Blood Cancer 2015;62:S829–S833

Palliative Care as a Standard of Care in Pediatric Oncology

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Lori Wiener, PhD, DCSW,[5,†] **Robert L. Casey**, PhD,[6] **Cynthia J. Bell**, PhD, RN,[7,8,†] **Joanne Wolfe**, MD, MPH,[9]
Amy M. Garee, RN, MS, PNP,[10,†] **Anne Watson**, PhD, MSc, RN,[11,†] and **Pamela S. Hinds**, PhD, RN, FAAN[4,12,†]

The study team conducted a systematic review of pediatric and adolescent palliative cancer care literature from 1995 to 2015 using four databases to inform development of a palliative care psychosocial standard. A total of 209 papers were reviewed with inclusion of 73 papers for final synthesis. Revealed topics of urgent consideration include the following: symptom assessment and intervention, direct patient report, effective communication, and shared decision-making. Standardization of palliative care assessments and interventions in pediatric oncology has the potential to foster improved quality of care across the cancer trajectory for children and adolescents with cancer and their family members. Pediatr Blood Cancer 2015;62:S829–S833. © 2015 Wiley Periodicals, Inc.

Key words: communication; family-centered care; palliative care; psychosocial support; quality of life

INTRODUCTION

The World Health Organization defines palliative care as a comprehensive care approach which “improves the quality of life of patients and their families facing the problems associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual.”[1] A standard of early integration of palliative care for children and adolescents with cancer translates into whole-person, whole-family support regardless of anticipated disease trajectory.[2] The early integration of palliative care as a standard across sites and settings provides meaningful opportunity to care for not only the physical domains impacted by cancer, but to also attend to the full impact of illness on the patient’s psychological, developmental, and spiritual wellness within the social context of each patient’s family and community. The American Academy of Pediatrics advocates for an integrated, interdisciplinary approach to competent and compassionate care: “in which the components of palliative care are offered at diagnosis and continued throughout the course of illness, whether the outcome ends in cure or death.”[3] The Worldwide Palliative Care Alliance recognizes the importance of integrating palliative care as a human right for children even in resource-limited health system settings,[4] requiring global collaboration for a resource-effective, evidence-based approach to best practice standards for early integration of palliative care for children and their families.

A review of the literature suggests that palliative care for pediatric cancer patients and their families varies across settings and resources and has only recently included access to services similar to those offered to adult cancer patients.[5] Empirical research for children and adolescents diagnosed with cancer reveals significant symptom and psychosocial suffering. Specifically, communication between the medical team and the patient and family; ongoing assessment of patient and family needs; developmentally informed interventions; and tangible support during times of inpatient and home care transition are target areas in which comprehensive care could be improved through a standard of palliative care.[6,7] Empirical data have found the quality of life for pediatric cancer patients and their family members can be enhanced through the prevention and alleviation of child and family suffering via the practice of compassionate and honest communication, symptom alleviation, and the psychosocial attentiveness incorporated by quality palliative care services.[5,8] This review was performed to determine whether palliative care concepts should be introduced early in the course of the diagnosis and throughout the trajectory of care.

Psychosocial Standard of Care

- Youth with cancer and their families should be introduced to palliative care concepts to reduce suffering throughout the disease process regardless of disease status.
- When necessary, youth and families should receive developmentally appropriate end of life care (which includes bereavement care after the child’s death).

METHODS

This review was performed as part of the collaborative Standards for Psychosocial Care of Children with Cancer and Their Families Workgroup. For a full description of the methods used to develop

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Conflict of interest: Nothing to declare.

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each standard refer to the introduction in this special issue.[9] The literature search for this paper utilized four databases: PubMed, Cochrane, PsycINFO, and SCOPUS (March 1, 1995 to March 1, 2015). Search terms included "palliative care" OR "palliative" OR "hospice" OR "end of life" OR "bereavement" AND "psychosocial" OR "communication" OR "support" OR "quality of life" AND malignancy-related terms AND "child" OR "adolescent" OR "young adult" OR "family" OR "sibling" (using indexed MeSH terms). Two medical librarians independently approved the search strategies. Due to the cancer-specific nature of this recommendation, palliative care studies including patients with non-malignant diagnoses were included only if data for the cancer population were specifically summarized. The last search was run on March 30, 2015. The reference lists of all included studies were hand-searched for additional studies. GRADE Guidelines framed the approach to this standard.[10]

The study team included geographically diverse representatives from the fields of oncology, palliative care and hospice, psychology, nursing, and social work. Expert opinion from members of the SIOP Pediatric Oncology in Developing Countries Palliative Care Working Group provided the reviewer team with additional global perspective. An external team of expert pediatric oncologists and child psychologists externally reviewed the recommendation prior to final draft. External review resulted in improved awareness of standard feasibility in resource-limited settings and inclusion of 14 additional papers with psychosocial emphasis.

RESULTS

Review of data from seven mixed method, seven qualitative, 21 quantitative, and four review method papers plus 32 editorials and two consensus reports support that children and adolescents with cancer and their family members should be introduced to palliative care concepts early in the course of the diagnosis and throughout the trajectory of care. Twenty-nine of the reviewed papers specifically called for early integration of palliative care starting at diagnosis. Members of the study team previously published a paper depicting the literature review strategy and methods using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRIMSA) reporting guidelines.[6] Due to the expanded search dates used in the current paper, an updated PRISMA flow diagram was generated (Supplemental Figure 1).

Symptom Assessment and Intervention

Comprehensive palliative care includes assessing and intervening on behalf of patients' psychological and physical symptoms. Validated questionnaire studies of bereaved parents and chart reviews of patients suggest that children and adolescents with cancer experience complex psychosocial symptoms during cancer treatment with exponentiation of these symptoms at end of life; to include symptoms of anxiety,[11–13] depression,[11,13,14] distress, worry, sadness, fear of being alone, difficulty talking about feelings, loss of independence, and loss of perspective.[15] These psychosocial symptoms often increase in the setting of disease progression and are often untreated.[16] Pain is reported as a prevalent end of life symptom in children with cancer,[8,11,15,17] a symptom reported as significantly distressing for family members to observe.[18] Over one-third of 141 bereaved parents retrospectively reported that they would have considered hastening their child's death had the child been in uncontrollable pain.[19] Bereaved parental anxiety and quality of life scales have correlated with level of child anxiety and pain at end of life,[19–22] implying early integration of palliative care as a standard of care has potential to improve long-term family wellness outcomes.

Patient Perspectives

A systematic review of empirically based end of life publications in pediatrics found that of 26 publications, only four (15.4%) included patient-reported outcomes, six (23.1%) included parent only-reported outcomes, and five (19.2%) included staff only-reported outcomes.[23] Additional research is required to solicit and document patient reports of their unique experiences to ensure appropriate interventions and care from primary stakeholders.[24] Information regarding palliative care needs should be sought directly from children and adolescents to honor the patient voice while also incorporating perspectives of family members.[25]

Compassionate and Honest Communication

Children and adolescents with cancer may wish to talk about the meaning of being ill, particularly prognosis; and care team involvement in these conversations may be important for individual children[2] and parents.[26] Recognizing that children are often aware of their imminent death, none of the 147 parents who talked with their child about death regretted having these important conversations.[27] Chart reviews investigating physician–family communication about a child or adolescent's end of life or prognostic issues rarely documented the child or adolescent's presence during these important conversations.[12,28] Notably, adolescent cancer patients and survivors describe a need for honest and respectful communication in addition to the provision of psychosocial support.[29,30] Interviews with parents of children with cancer reveal that they perceive accurate, clear, and understandable communication as beneficial.[17,31,32] Cross-sectional surveys with bereaved parents confirm that parent perceptions of clear and compassionate communication are associated with excellent care and improved psychosocial outcomes.[17,33–35] These findings underscore the importance of ongoing supportive communication, particularly as disease progresses.

Decisional Preferences

A longitudinal, multi-institutional adult cohort study revealed that patients with cancer are more likely to receive end of life care that is consistent with their preferences when they have had the opportunity to discuss their wishes regarding end of life care with a clinician.[36] Likewise, family-centered advance care planning increased patients' wishes for families to make appropriate decisions as needed, improved patients' understanding about end of life options, increased likelihood of limiting futile treatment, and increased family's ability to honor wishes of their children.[37] Adolescents with cancer described age-appropriate advance care planning as acceptable, useful, and helpful.[38,39] Effective communication among all parties from the patient and parent to the comprehensive care team (social workers, psychologists, child life specialists, and clinicians) is crucial to successfully capture the child/adolescent's end of life preferences, including the practicalities of location of death.

Relatedly, families whose primary oncologist clearly explained treatment options during their child's end of life care and who involved home-care services were more likely to plan for child's location of death.[19] Perspectives of bereaved parents reveal a need for improved and cohesive care transitions, particularly in the form and feasibility of home care and respite support services knowledgeable on pediatric and adolescent psychosocial and symptom care needs.[17,40–42] Parents were more likely to decide against resuscitation for their child with incurable cancer if an end of life discussion with their inter-disciplinary medical team occurred prior to emergent situations,[13] when both parents and providers may experience intense and conflicting emotions. The opportunity to plan location of death is associated with high quality palliative care and may suggest that communication regarding planning rather than actual location of death may be a more relevant outcome.[19,43]

DISCUSSION

Perceived cost[15,44–51] and lack of sufficient time [33,37,47,51–53] are the most frequently cited barriers to introducing palliative care concepts, thus warranting consideration of reimbursement for palliative care conversations and services. Further barriers include limited access to established support services[43] or psychosocial professionals,[11,52] thus compelling advocacy for improved palliative care education for general health providers.[8,17,54–56] Additional barriers include provider discomfort with palliative care conversations,[2,19,23,38,57] or misconceptions regarding palliative care as only beneficial when treatment is no longer effective[32,58,59] both of which may benefit from clarity in palliative care guidelines.[60]

Early integration of palliative care as a standard approach for children and adolescents diagnosed with cancer carries meaningful opportunities to improve symptom control and quality of life for patients and their families. As a minimum, symptom burden assessment with subsequent intervention and effective communication must be prioritized, as these tenets are associated with improved psychosocial outcomes for pediatric cancer patients and their families. When resources avail, tangible support for transitions between care settings (whether hospital care, ambulatory care, or home care) should support the practical needs and available resources of patients and their families. Future research should

TABLE I. Palliative Care Standard Summary of Evidence Table

Standard	Evidence summary[a]	Methodology[b]	Quality of evidence[c]	Strength of recommendation[d]
Youth with cancer and their families should be introduced to palliative care concepts to reduce suffering throughout the disease process regardless of disease status When necessary youth and families should receive developmentally appropriate end of life care [which includes bereavement care after the child's death]	Uncontrolled symptoms continue to be problematic; well-integrated palliative care has potential to improve high quality care, including decreased physical and psychological symptom burden Family centered communication, to include prioritization of the patient's voice, is critical to reducing child and family suffering Developmentally Informed advanced care planning and decision-making interventions foster honoring patient and family preferences which results in improved long-term family outcomes Tangible support during times of care transition, such as respite or home care service transitions, improves perceived cohesiveness of care	Mixed-methods, qualitative, quantitative, and literature reviews Majority cross sectional retrospective survey and in-depth interviews Limited intervention trials Replication of findings evident	Moderate quality given consistent findings from lower level evidence studies. Few cost-effective studies have been conducted. Sibling and grandparent responses are currently under-recognized as part of the larger family unit	Strong recommendation given risk-benefit ratio including significant health impact on bereaved family members and positive outcomes with earlier integration of palliative care and end of life care

[a]Based on summary of evidence table for that standard; [b]Types of studies; [c]High, moderate, low, very low; [d]Strong or weak (based on GRADE quality criteria).[7]

integrate direct patient reported preferences and prospective, longitudinal, multi-institute investigations with attentiveness to reporting cost, staffing needs, and psychosocial outcomes.

CONCLUSION

Children and adolescents living with cancer and their families should receive early, integrated access to family-centered palliative care concepts (symptom assessment and intervention; direct patient report; effective communication; and shared decision-making) to minimize symptom burden, ease suffering, effectively manage pain, and provide preventative bereavement care. The target population for palliative care includes not only patients but also those individuals identified as formative in the child's social and relational spheres, whether siblings, parents or guardians, grandparents, or other extended relatives.[15,31,40,52,61]

Current evidence for this recommendation is of moderate quality given consistent findings from lower level evidence studies (Table I). This is an overall strong recommendation given risk-benefit ratio including significant health impact on bereaved family members and positive outcomes with earlier integration of palliative care and end of life care.[2,6,51] Youth and their families should be introduced to palliative care concepts to reduce suffering and enhance well-being throughout the disease process regardless of disease status. Ongoing psychosocial assessments and appropriate interventions pertaining to palliative and end of life care are essential components of quality care for youth with cancer and their family members.

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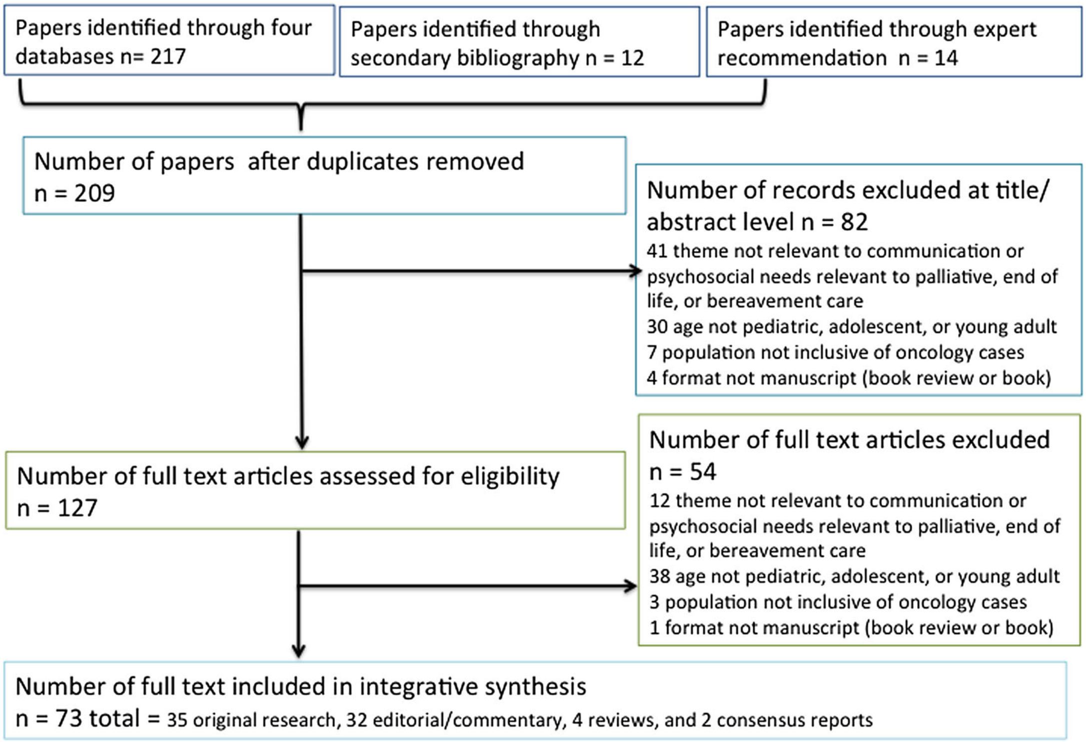

Supplemental Figure I. Preferred Reporting for systematic Reviews and Meta-Analyses (PRISMA) Flow Diagram.

Pediatr Blood Cancer 2015;62:S834–S869

Bereavement Follow-Up After the Death of a Child as a Standard of Care in Pediatric Oncology

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After a child's death to cancer, families commonly want continued connection with the healthcare team that cared for their child, yet bereavement follow-up is often sporadic. A comprehensive literature search found that many bereaved parents experience poor psychological outcomes during bereavement and that parents want follow-up and benefit from continued connection with their child's healthcare providers. Evidence suggests that the standard of care should consist of at least one meaningful contact between the healthcare team and bereaved parents to identify those at risk for negative psychosocial sequelae and to provide resources for bereavement support. Pediatr Blood Cancer 2015;62:S834–S869.

Key words: bereaved parents; bereavement; cancer; death of a child; palliative care; pediatric oncology

INTRODUCTION

The loss of a child is considered by many to be the most devastating type of bereavement.[1] Intense and persistent grief reactions are common and may be debilitating for some bereaved family members.[2–4] Protracted grief reactions, such as prolonged grief disorder, are associated with poor psychological and physical health outcomes, including mortality.[5–8] Grief reactions may be compounded by secondary losses, such as the loss of support from the child's medical team, whom the family has come to trust and rely on for comfort and information.[9] An abrupt end to contact soon after the child's death is experienced by some parents as abandonment.[10,11] This loss may be mitigated by providing continuity of care through bereavement to the families from pediatric cancer care facilities.[12] Unfortunately, however, resources, such as dedicated staff and time, are often limited, and thus, families commonly return to their communities without a sustained connection to the medical team.

Bereavement follow-up through telephone calls, emails, and/or mailed cards or letters is considered to be part of good medical care,[13] and the need to provide bereavement care has been increasingly recognized.[14,15] The American Academy of Pediatrics and the Institute of Medicine each call for care to continue through bereavement.[16,17] Many providers make efforts to support bereaved families, but efforts are highly inconsistent, in part, because there are no existing guidelines advising staff on best practices for bereavement follow-up. Several reports have described the obligation that hospitals have to provide some level of bereavement follow-up to the patient's family.[18–21] A recent article suggests that pediatric palliative care physicians have an ethical duty of "nonabandonment" to care for the families of children with life-threatening conditions through their illness and bereavement.[22] Moreover, the child's primary medical team can be an important source of transitional support for bereaved parents and other family members.[17] The objective of this review was to assess and appraise the literature on bereavement outcomes, follow-up, and needs to determine an evidence-based standard for routine assessment of bereavement needs of parents whose children died from cancer.

Psychosocial Standard of Care

A member of the healthcare team should contact the family after a child's death to assess family needs, to identify those at risk for negative psychosocial sequelae, to continue care, and to provide resources for bereavement support.

METHODS

This review was performed as part of the collaborative *Standards for Psychosocial Care of Children with Cancer and Their Families* effort. For a full description of the methods used to develop each standard, refer to Wiener, Kazak, Noll, Patenaude, and Kupst.[23] A comprehensive electronic literature search for articles in PubMed, PsycINFO, CINAHL, EMBASE, SCOPUS, and Web of Science was performed by a medical librarian at Memorial Sloan Kettering Cancer Center. This was an expansion of an initial preliminary literature search conducted in early 2014.

Abbreviations: PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses

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Recommendations were generated based on the search findings. The search strategy, recommendations, and potential barriers to implementation of the recommendations were sent for external blinded review. The reviewers were identified through the Standards of Pediatric Psychosocial Oncology Care workgroup.[23] The returned comments noted the need for future evaluative research. Reviewers indicated that there was evidence that parents find bereavement contact helpful, and although more research needs to be done, there is sufficient evidence to indicate that some bereavement contact should be part of good psychosocial care. Reviewers offered recommendations for future research directions. We responded to these reviews by expanding the literature search.

The searches of the English-language literature published from March 1, 1995 to March 1, 2015 combined pediatric cancer terms with bereavement and follow-up-related terms and MeSH headings. See Supplemental Table I for a description of the search terms. Studies were eligible for inclusion if they focused on bereaved parents', siblings', or grandparents' experiences after the loss of a child ages 0–18 at the time of cancer diagnosis and at least one of the following areas: bereavement support programs or interventions for parents; parents, clinicians, or siblings reporting need for follow-up; bereavement outcomes after the death of a child; outcomes associated with bereavement aftercare; predictors of negative bereavement outcomes; and end-of-life care factors associated with bereavement outcomes or need for aftercare. Articles were excluded if they were biomedical in nature and if they focused exclusively on the following topics: diseases other than cancer; psychosocial factors associated with active cancer treatment or survivorship rather than bereavement; experiences of the child with cancer; or bereaved parents of adult children.

Supplemental Figure 1 depicts the review process according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.[24] For the full-text review phase, the articles were randomly assigned to six authors (W.L., C.S., K.R., G.C., L.D., and L.W.) to code for exclusion and to extract pre-determined information from each eligible article for the qualitative synthesis. The authors had regular discussions to assure consistent rating of study rigor and to reach consensus about the inclusion and exclusion of all articles.

RESULTS

See Table I for a summary of the evidence extracted from the literature and Supplemental Table II for complete results from the literature review. The review of the evidence demonstrated that bereaved parents experience poor psychological outcomes in bereavement, following up with and supporting bereaved parents is perceived to be helpful, and bereaved families want and need services.

Bereavement Outcomes and Associated Factors

Several studies have demonstrated the psychological challenges parents bereaved by cancer face,[25] with increased rates of depression and anxiety,[26] grief,[27] existential distress, challenges to their sense of identity and meaning-making,[28–30] guilt,[31] and posttraumatic stress disorder.[32] These symptoms commonly persist over time.[26,27] Approximately 10–25% of parents bereaved by cancer experience debilitating levels of grief.[27,33–35] Furthermore, studies have shown that bereaved parents' grief typically persists and often even intensifies after the first year.[34,36] Poorer outcomes have been reported among mothers,[36,37] regardless of cultural background;[38] among parents dissatisfied with their child's medical care;[31] and among those whose children had anxiety or sleep disturbances,[39] a stem cell transplant,[30,40,41] or uncontrolled pain or a difficult death.[31,42]

Families bereaved by cancer also often struggle with isolation due to fear of burdening their support network with their persistent pain

TABLE I. Summary of Evidence Table

Standard	Evidence summary[1]	Methodology[2]	Quality of evidence[3]	Strength of recommendation[4]
A member of the healthcare team should contact the family after a child's death to assess family needs, to identify those at risk for negative psychosocial sequelae, to continue care, and to provide resources for bereavement support	• Many bereaved parents, siblings, and grandparents experience long-lasting negative outcomes, with a subset debilitated by their distress • Parents have generally indicated their wish for and appreciation of follow-up by the healthcare team after the death of their child • Professional support can assist with grief, particularly among those with more severe, debilitating symptoms	• Quantitative, qualitative, and literature reviews • Majority were cross-sectional surveys and in-depth interviews • Limited randomized controlled trials • Replication of some findings evident	Moderate quality; findings from lower level evidence studies were consistent	Strong recommendation given risk–benefit ratio, including significant health impact on bereaved family members and positive outcomes with contact made after the death of the child

[1] Based on summary of evidence table for that standard; [2] Types of studies: for example, randomized controlled trial, cross-sectional, longitudinal; consensus; systematic review articles; [3] Quality of evidence: High, moderate, low, and very low (based on Grading of Recommendations, Assessment, Development and Evaluation [GRADE] criteria [109]); [4] Strength of recommendation: Strong or weak (based on GRADE quality criteria).

[43] and experience decreased social support over time.[29,44] Other family members also struggle after the loss, including some siblings [45–48] (see Gerhardt, Lehmann, Long, and Alderfer[49] in this issue) and grandparents.[50] Being reminded of, speaking about, and feeling connected to the deceased child are frequently described as helpful[51–56] and are associated with better outcomes.[57]

Bereavement Follow-Up Efforts and Formal Interventions

Despite families' risk for poor outcomes, few bereavement follow-up programs and formal interventions to support them have been rigorously evaluated. A systematic review of studies on hospital-based bereavement programs concluded that such programs help families feel cared for, reduce their sense of isolation, and improve their coping.[58] Importantly, in qualitative studies, parents have not reported negative effects of follow-up, and most have expressed appreciation.[59–62] Additionally, staff who engage in bereavement follow-up have also reported that they, too, experience benefits from reaching out to families.[59,63–66]

Without an existing standard of care, however, follow-up attempts by a given medical team are inconsistent,[11,62,67] and some families are never contacted. A variety of practices are used when there is follow-up, including making phone calls; sending cards; attending funerals; providing information and resources; connecting parents with other bereaved parents; acknowledging birthdays, holidays, and anniversaries; visiting the family; holding family meetings; and organizing memorial services.[63,68–71] Qualitative studies have found that phone calls and family meetings after the child's death are beneficial, helping parents feel like their child is remembered, facilitating meaning-making, responding to unanswered questions about their child's care, reassuring them they did everything they could, and providing a sense of closure.[11,64–66,72–76] Of note, some follow-up efforts have been perceived as inadequate,[77] suggesting the importance of training staff. In addition, involving bereaved parents in developing and administering bereavement education programs and interventions has been shown to have great value for them,[28,78] further giving meaning to their child's life.[79]

Need for Bereavement Follow-Up and Mental Health Services

One of the most consistent findings in the literature is that most parents want and appreciate continued connection with the medical team[11,66,80–83] and expect follow-up after their child underwent such intense treatment.[70,77] Parents often value ongoing contact with a provider who knew their child and may appreciate providers connecting them with other bereaved parents.[83–85]

Research has shown that some parents would like, in addition to follow-up, bereavement mental health support,[34,86,87] sometimes well beyond the first year of bereavement.[26,27,34] However, parents, especially minorities, often face barriers in accessing such support.[34,88] For example, although parents often appreciate services from the institution at which their child was treated,[11,44,89] studies have shown that it is emotionally difficult for parents to physically return to the place of treatment,[11,70] which may be, in part, why home visits are desirable.[87] Parents are also frequently concerned about the well-being of their surviving children and want formalized bereaved sibling support.[75,83,87,88,90] Evidence suggests that there are benefits to grief interventions focusing on the entire family.[85,91]

There is a need for effective, evidence-based bereavement mental health services. Although randomized controlled trials are limited, descriptive data suggest that bereavement mental health services can be beneficial,[3] particularly for those with more severe symptoms or prolonged grief disorder.[8,28,58,92] Unfortunately, existing services appear underused,[93] and some are perceived as unhelpful.[2,34,94] Healthcare providers are in a position to link families with bereavement services and should, thus, provide referrals when making contact. There is also a need for screening to identify those in greatest need.[14,15,95] Existing bereavement needs assessments have been described as insufficient,[96] but efforts to develop a more valid, reliable screening tool are underway.[97] Once identified, family members may benefit from interventions designed to prevent or reduce debilitating prolonged grief symptoms.[28,92,98]

DISCUSSION

This review demonstrated that families who lose a child to cancer are at risk for poor bereavement outcomes and desire continued connection to their child's healthcare team.[9,99] Bereaved families often feel abandoned when contact is lacking, creating a secondary loss in addition to the excruciating loss of their child.[70,83,99,100] Contact with staff is commonly wanted by parents and may prove very helpful to families as they transition back to their communities,[80,100] wherein support frequently wanes over time.[36,44]

The IOM has emphasized that bereavement care is a public health priority and should be a part of comprehensive emotional and spiritual care for grieving family members.[17,101] Data suggest that what healthcare providers do around the time of a child's death has a lasting impact on families[102,103] and can be important in processing their own grief.[59,63–66,104] Review of the current available literature provides compelling evidence for the benefit of having a member of the healthcare team contact the bereaved family by phone at least one time after a child's death in order to assess the family's needs, to let them know they and their child have not been forgotten, to identify families who are at risk for negative bereavement outcomes, and to link families to resources for bereavement support in their community.[105,106]

Although the literature included rich evidence on the need for bereavement care and recognized bereavement services, many of the studies reviewed, which were mostly descriptive, cross-sectional, and lacking control groups, had substantial methodological and conceptual weaknesses. Additional limitations included healthy selection biases, underrepresentation of fathers, and lack of racial and ethnic diversity.[34] Future studies should address these limitations, using prospective, longitudinal, and randomized controlled designs with more representative, diverse samples. Specifically, a randomized controlled trial examining the effects of the healthcare team following up with families at least one time and exploring the impact of continued follow-up is warranted. In addition, studies should address the important logistical and emotional barriers that may impede follow-up and the development of bereavement programs [68] through evaluation of continuing education[59,107,108] and organizational structure.[65] Research to strengthen the evidence base on bereavement interventions is also needed.

CONCLUSION

Consistency of findings from 94 studies utilizing different study designs and methodologies supports the importance of healthcare providers initiating contact with families after the death of a child to cancer. The data suggest that the standard of care should consist of at least one meaningful contact, such as a call, email, or letter, between the healthcare team and bereaved parents following the death of a child to cancer. Efforts to remember the child through, for example, invitations to memorial services or the sending of special occasion cards, are often greatly appreciated.[11,70] Bereavement support from the psychosocial team, including psychoeducation, risk assessment, and referrals to the community when appropriate, should also be offered to all grieving family members.[22,44–47] Although moderate evidence was found, an overall strong recommendation for this standard is given due to minimal risk to families and the consistent evidence of potential long-term benefits.

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Communication, Documentation, and Training Standards in Pediatric Psychosocial Oncology

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As part of a larger effort to create standards for psychosocial care of children with cancer, we document consensus and evidence-based data on interprofessional communication, documentation, and training for professionals providing psycho-oncology services. Six databases were searched. Sixty-five articles and six guidelines and consensus-based documents were identified; 35 met inclusion criteria. Data support strong recommendations for standards of care in communication/collaboration, documentation of patient information, and training in pediatric psycho-oncology. These are areas where extensive research is unlikely to be conducted; however, professional expectations and qualifications may be further clarified and strengthened with time. Pediatr Blood Cancer 2015;62:S870–S895. © 2015 Wiley Periodicals, Inc.

Key words: documentation; interdisciplinary communication; pediatric oncology; pediatric psycho-oncology; training

INTRODUCTION

This paper addresses the training and professional responsibilities of mental health professionals who provide psychosocial support for children and youth with cancer and for their family members in pediatric cancer clinics, hospitals, and in survivorship clinics. In the absence of comprehensive evidence- and consensus-based standards,[1] these standards were developed as part of the collaborative *Standards for Psychosocial Care of Children with Cancer and Their Families* project to develop a full set of standards to guide essential psychosocial care delivery to all children with cancer and their families. The history of this effort and the methodology used to develop these standards are detailed in Wiener, Kazak, Noll, Patenaude, Kupst.[2]

Although there is limited research on the diverse communications of psychosocial providers within a medical team, especially in pediatric oncology, and lack of full consensus about the teaching of communication skills,[3] it is clear that effective communication is a cornerstone of family-centered care.[4–7] Collaboration requires an intentional partnership between professional disciplines wherein mutual valuing, respect for skills and scope of practice, accountability, and shared goals for the child and family are practiced.[8–10] Psychosocial providers, whose training focuses on development of communication skills in the medical setting, often determine and share with the team the particular stresses, vulnerabilities, and strengths of the child with cancer and their family. This facilitates appropriate communication around consent and treatment, enhancing the experience for families and medical teams. Team conferences, team-patient and family dialogue, as well as patient care rounds are all effective communication strategies.[11–13]

To facilitate coordinated care plans and treatment goals among the interdisciplinary team members providing care to pediatric patients and families, pediatric psychosocial providers should have access to the patient's medical records and document assessments and interventions.[14–16]

Pediatric psychosocial providers require specialized education and training above and beyond their discipline-specific competencies and credentials in mental health care (i.e., psychology, psychiatry, social work, etc.) in order to effectively and sensitively care for pediatric cancer patients and their families.[5,6,15,17–19]

Psychosocial Standard of Care

- Open, respectful communication and collaboration among medical and psychosocial providers, patients and families is essential to effective patient- and family-centered care. Psychosocial professionals should be integrated into pediatric oncology care settings as integral team members and be participants in patient care rounds/meetings.
- Pediatric psychosocial providers should have access to medical records and relevant reports should be shared among care team professionals, with psychological report interpretation provided by psychosocial providers to staff and patients/families for patient care planning. Psychosocial providers should follow documentation policies of the health system where they practice in accordance with ethical requirements of their profession and state/federal laws.
- Pediatric psychosocial providers must have specialized training and education and be credentialed in their discipline to provide developmentally appropriate assessment and treatment for children with cancer and their families. Experience working with children with serious, chronic illness is crucial as well as ongoing relevant supervision/peer support.

Abbreviations: AGREE, appraisal guidelines for research and evaluation; GRADE, grading recommendations assessment, development, and evaluation

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TABLE I. Summary of Evidence Table-Communication, Training and Documentation Standards in Psychosocial Oncology

Standard	Evidence Summary	Methodology	Quality of Evidence	Strength of Recommendation
Open, respectful communication and collaboration among medical and psychosocial providers, patients and families is essential to effective patient- and family-centered care. Psychosocial professionals should be integrated into pediatric oncology programs as integral team members and be participants in patient care rounds/ meetings.	Given the complexities of the provision of care for children with cancer and their families, research and consensus recommendations support the importance of a well-structured, integrated environment for mutually respectful inter-professional communication and collaboration around assessment and treatment.	Systematic review articles; cross-sectional, descriptive, qualitative studies; consensus and opinions of respected authorities and expert committees.	Moderate Important limitation, methodologic flaws, indirect evidence	Strong recommendation. Consensus of expert advice outweighs relatively low level of research evidence.
Pediatric psychosocial providers should have access to medical records and relevant reports should be shared among care team professionals, with psychological report interpretation provided by psychosocial providers to staff and patients/families for patient care planning.	Psychosocial providers must abide by ethical documentation requirements of their professional organizations, recognizing the need to tailor documentation to policies of the health care system in which they practice. Documentation should describe the emotional impact of the patient's diagnosis and treatment as well as providing a summary of psychosocial services and their impact on the patient/family to monitor progress and communicate with other providers.	Opinions of respected authorities and expert committees	Low	Strong recommendation, Recommendations based on respected professional opinion.
Pediatric psychosocial providers must have specialized training and education and be credentialed in their discipline to provide developmentally appropriate assessment and treatment for children with cancer and their families. Experience working with children with serious, chronic illnesses is crucial as well as ongoing, relevant supervision/peer support.	Professional organizations develop core competencies for pediatric psycho- social providers which transcend disciplines.). Some skills are specific to a particular discipline (i.e., prescription of psychotherapeutic medication) and require additional training and certification.	Consensus and opinions of respected authorities and expert committees, one cross-sectional descriptive study.	Low Professional training standards are typically determined by consensus among experienced professionals, not by research.	Strong recommendation. Consensus around domains to be mastered, essential credentials and skills needed outweighs the relatively low level of evidence.

METHODS

Literature Search Strategy

Pubmed, Medline, CINAHL, PsychINFO, Google Scholar, OVID were utilized for the communication, documentation, and training literature searches. For communication, the search used the terms: "inter-professional communication" OR "collaboration" OR "communication" OR "multi-disciplinary teams" OR "mental health professional roles" OR "family-centered care rounds". For documentation, the search used the terms: "documentation" OR "medical record documentation" OR "staff documentation." The training search terms were: "training" OR "standards" OR "competency" OR "guidelines." All of the communication, documentation, and training search terms were combined with the terms: "pediatric oncology" OR "pediatric cancer" OR "psychosocial oncology professionals" OR "psycho-oncology" OR "pediatric psychology" OR "adolescent cancer."

Results were limited to English-language-only publications from March 1, 1995–2015. Studies utilizing any type of methodology and literature reviews and summaries were included. A hand-search of reference lists from relevant review articles was also undertaken. The title and abstract of all citations obtained through the search strategy were reviewed and full texts obtained for more in-depth review of potentially eligible studies. Included were relevant materials from the Canadian Association of Psychosocial Oncology, American Psychological Association, Association of Pediatric Oncology Social Workers, Association of Oncology Social Workers, and National Association of Social Workers.

Sixty-five articles and six guidelines and consensus-based documents about communication, documentation, and training in psychosocial oncology were identified. Thirty-five met inclusion criteria and were incorporated into this review [See Supplemental Evidence Tables]. Articles were excluded which concerned psychosocial adjustment to cancer rather than issues of communication, documentation, or training. Of these, one was a meta-analysis of controlled studies, one was an evidence-based clinical practice guideline, three were systematic reviews of descriptive/qualitative studies, 14 were individual descriptive or qualitative studies, and 16 were opinions of respected authorities and expert committees. Evidence tables were rated for the quality and strength of evidence utilizing the GRADE system [See Summary Evidence Table I].[20]

RESULTS

Communication

Collaborative person-centered practice is an approach to care that involves "the continuous interaction of two or more professionals or disciplines, organized into a common effort, to solve or explore common issues with the best possible participation" of the ill person.[21] Interprofessional communication occurs within the pediatric oncology unit or clinic and between pediatric oncology medical and psychosocial professionals and members of community organizations.[5] In rural areas, telehealth is an increasingly utilized method of communication.[22,23] Four elements that need to exist within a team include a willingness to collaborate, good communication, mutual trust, and respect.[6,9] Members of effective interdisciplinary care teams respect each other's expertise and knowledge base[7,14] and can negotiate differences of professional opinion.[15] Poor communication exacts potentially enormous economic, social, psychological, emotional, and collateral costs to the patient, the patient's support network, the clinicians, the cancer care system, and to the larger society.[24]

Young patients have the right to be fully informed about their illness and treatment and desire empathetic, direct communication with physicians.[25,26] Adolescents with cancer report the importance of being well-informed and included in decision-making about their treatment and end of life care.[27–29] Tailoring of the message and choice of the voice are important because not all want full details of their condition, care, and prognosis[26,30] and preferences vary for preferred source.

Family-centered rounds, an evidence-based intervention with families, are associated with higher parent[31] and staff[12] satisfaction, consistent medical information and care plan discussions, with little or no additional burden of time for involved providers.[11,13,32]

Documentation

Documentation must include reports of patient and/or family impact of illness, of assessments conducted, nature and impact of psychosocial services provided, monitoring of illness status, and treatment plans.[16,25] Psychosocial providers must be familiar with professional codes of ethics[15] and federal and state regulations about protection of the privacy of health information and limits of privacy protection.[4,14] Documentation and communication with other professionals must reflect appropriate understanding of what information is essential for team members and/or family members to know, when specific permission is needed, and what different rules apply under extraordinary conditions of risk of harm.[16] Treatment facilities may impose their own requirements for documentation as long as they comply with state and federal regulations, which represent the final authority.[14] The creation and handling of electronic medical record information should be governed by the same professional rules as other health care communication.[14,16]

Training

Specialized training is needed to work with pediatric populations[33,34] as well as additional professional training in psycho-oncology.[15,18] Mental health professionals working in pediatric psycho-oncology should be licensed in their professional discipline (e.g., psychology, social work) by the state or province in which they practice and credentialed in their health care setting.[5,16] Required skills include developmental, behavioral and psychological assessment, ability to assess and treat psychopathological symptoms, and also to differentiate normal reactions to severe illness from psychopathology. Providers must be experienced in the treatment of and consultation with children with cancer and their family members and knowledgeable about available psychotherapeutic and psycho-educational interventions. Training is also essential in pain management, promotion of positive health behaviors and adherence, support for decision-making, grief and bereavement, and in consultation with a broad range of other professionals.[15,17,33–36] Awareness of the challenges of the hospital environment and familiarity with the organization and workings of the pediatric cancer unit are essential to successful support of families of children with cancer.[17] Knowledge of the ethical issues which can arise in the context of pediatric cancer care is also necessary. Recognition of the interplay between cultural and spiritual beliefs and practices in the context of the family's and patient's reactions to illness and treatment are, similarly, basic aspects of the care psychosocial providers provide.[17–19,35] Some psycho-oncology providers are also experts in psychopharmacologic[37] and neuropsychological[38] assessment.

Research training aids in the understanding of progress in patient care in pediatric oncology, pediatric psychology and psychosocial oncology and of the requirements for ethical care of subjects including, notably, informed consent.[18,19,35] Personal qualities which the work requires include compassion, self-awareness, commitment to teamwork, personal growth, and a belief in the potential for resilience.[35] Learning and skill acquisition are lifelong aspects of work in this field. Because of the intensity of the work environment and of patient needs, supervision, peer support, professional growth activities, and opportunity for discussion of difficult cases are needed to prevent burnout.[18,25]

BARRIERS TO IMPLEMENTATION OF STANDARD

Barriers to interdisciplinary communication and collaboration may include role ambiguity, lack of skill sets, evolving team structures, time restraints, interpersonal interaction issues, conflict, and value differences.[10] Hierarchical organizational structures can produce a sense of inequality between members of different disciplines as well as a lack of understanding of the roles and skills of different disciplines.[7] Barriers to implementation of documentation and training standards include limited financial resources and lack of trained professionals with pediatric psycho-oncology experience as a consequence of the scarcity of training programs.[19]

DISCUSSION

Consensus reports from esteemed experts in the field and reviews by panels of senior members of professional organizations do not establish the same level of evidence as research, which is largely lacking in this area. Although research might be helpful to definitively assess salient components of psychosocial communication, documentation and training and their impact on patient care, these are unlikely areas for major research investigations. Reliance upon professional organizations, credentialing bodies, and sporadic research studies for further evidence in these important areas will be required.

CONCLUSION

Upholding professional standards in the psychosocial care of children with cancer requires hiring of professionals with the training, credentials, and skills to do this complex work. Best accomplished in the context of multi-disciplinary teams, there are both discipline-specific and common skill areas needed to provide and document psychosocial services to children with cancer and their families and to develop and implement interventions and programs of care. Shared communication requires mutual respect, clear documentation of services delivered and needed, medical status and prognosis information, and assessments of patient and family functioning. Because the work is inherently complex and challenging, opportunities for peer or mentored supervision, and/or professional development are recommended at all levels of professional experience.

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