Patients' and Parents' Needs, Attitudes, and Perceptions About Early Palliative Care Integration in Pediatric Oncology

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IMPORTANCE Early palliative care integration for cancer patients is now touted as the optimal care model, yet significant barriers often prevent its implementation. A perceived barrier, especially for pediatric oncology patients, is the notion that patients and their families may not need or want palliative care involvement early in the disease trajectory.

OBJECTIVE To determine the perception of symptom burden early in treatment and assess attitudes toward early integration of palliative care in pediatric oncology patient-parent pairs.

DESIGN, SETTING, AND PARTICIPANTS Novel but pretested survey tools were administered to 129 patient-parent dyads of hospital-based pediatric oncology ambulatory clinics and inpatient units between September 2011 and January 2015. All patient participants were aged between 10 and 17 years and were diagnosed as having an oncologic condition 1 month to 1 year before enrollment. Both the patient and the parent in the dyad spoke English, and all participating parents provided written informed consent. A convenience sample was used for selection, with participants screened when otherwise presenting at a participating site. A total of 280 eligible participants were approached for study inclusion, 258 of whom were enrolled in the study (92.1% positive response-rate).

MAIN OUTCOMES AND MEASURES Degree of perceived suffering from early symptom-related causes, attitudes toward early palliative care integration, and patient-parent concordance. Statistical analysis included descriptive statistics, calculation of concordance, McNemar test results, and Cochran-Armitage trend test results.

RESULTS Of the 129 patients in the dyads, 68 were boys, and 61 girls; of the 129 parents, 15 were men, and 114 women. Patients reported the following symptoms in the first month of cancer therapy: nausea (n = 109; 84.5%), loss of appetite (n = 97; 75.2%), pain (n = 96; 74.4%), anxiety (n = 77; 59.7%), constipation (n = 69; 53.5%), depression (n = 64; 49.6%), and diarrhea (n = 52; 40.3%). A large proportion of those reporting suffering indicated substantial suffering severity from specific symptoms (ie, a great deal or a lot) including nausea, 52.3% (57 of 109), loss of appetite, 50.5% (49 of 97), constipation 30.4% (21 of 69), pain 30.2% (29 of 96), anxiety 28.6% (22 of 77), depression 28.1% (18 of 64), and diarrhea 23.1% (12 of 52). Few children and parents expressed opposition to early palliative care involvement (2 [1.6%] and 8 [6.2%]) or perceived any detrimental effects on their relationship with their oncologist (6 [4.7%] and 5 [3.9%]), loss of hope (3 [2.3%] and 10 [7.8%]), or therapy interference (3 [2.3%] and 2 [1.6%], respectively). Intradyad concordance was low overall: 26% to 29% for exact concordance and 40% to 69% for agreement within 1 response category. Significant differences in patient-parent attitudes toward aspects of early palliative care included child participants being more likely than their parents (40.3% [n = 52] vs 17.8% [n = 23]) to indicate that palliative care would have been helpful for treating their symptoms (P < .001).

CONCLUSIONS AND RELEVANCE Pediatric oncology patients experience a high degree of symptom-related suffering early in cancer therapy, and very few patients or parents in this study expressed negative attitudes toward early palliative care. Our findings suggest that pediatric oncology patients and families might benefit from, and are not a barrier to, early palliative care integration in oncology.

Published online March 9, 2017.

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Invited Commentary page 1220
Supplemental content
Approximately 12,400 children are diagnosed with cancer each year in the United States. Regardless of the outcome, cancer and its treatment often take a significant toll on the quality of life of pediatric oncology patients. While the need for palliative care (PC) in this patient population at the end of life, as well as its benefits, have been well established, availability and utilization remain low. Experts in the field have made the recommendation that PC should be introduced at diagnosis and incorporated into care throughout the course of cancer care, but in most practices, this does not occur. The American Academy of Pediatrics recommends “an integrated model of PC ‘in which the components of PC are offered at diagnosis and continued throughout the course of illness, whether the outcome ends in cure or death’” for all children with life-limiting illness. Early integration of PC with oncology care for pediatric patients has the potential to improve patients’ physical and emotional well-being as well as their quality of life throughout the course of therapy and beyond.

It has been shown that pediatric oncology patients suffer greatly at the end of life and that PC team involvement, especially earlier involvement, has led to substantial improvements in the suffering of patients, better family preparedness, and enhanced advanced care planning. Hospice care has also been reported to be of great benefit to dying children and their families, with barriers noted to include a perception of intrusion and loss of hope, which may be overcome with earlier PC involvement. Pediatric PC consultation has been demonstrated to aid in symptom assessment and management, but often the consult is late in the disease trajectory, thus limiting the benefit of earlier PC interventions. Palliative care services offered to all children with cancer and their families at the time of diagnosis can provide support encompassing medical, social, emotional, and spiritual well-being in a dynamic model that shifts focus based on the needs of the patient at any given time, thereby achieving comprehensive integrated quality care throughout.

Early involvement of PC in adult oncology patients has been demonstrated to improve quality of life, decrease depressive symptoms, and improve patient and caregiver outcomes. Based on strong recent evidence, the American Society of Clinical Oncology recommends early integration of PC for patients with metastatic disease or high symptom burden at presentation. To our knowledge, there are no data on symptom burden and suffering in children during initial cancer therapy or early integration of PC in pediatric patients with cancer. Strategies for identifying barriers to early PC integration and enhancing PC integration have been identified as a high priority for research in the field. In what has been coined the “they are not ready syndrome,” lack of patient and family receptivity to early PC integration is often cited as a barrier to care provision. The aim of the present study was to establish whether patients have unmet needs at the initiation of cancer therapy and whether patient and family attitudes are indeed a barrier to early PC involvement. To this aim, we surveyed pediatric oncology patients and parents to elucidate their perceptions regarding symptoms and suffering in the first month of cancer therapy and their attitudes toward early integration of PC.

**Key Points**

**Question** Are pediatric oncology patients and their parents open to early palliative care integration, and to what extent might it benefit them?

**Findings** This multi-institutional survey study of pediatric oncology patients and their parents revealed a high burden and intensity of symptom-related suffering in the first month of cancer therapy and overall positive attitudes toward early palliative care involvement. Very few respondents opposed early palliative care integration.

**Meaning** The degree of perceived suffering and receptivity to palliative care observed in our study suggest that pediatric oncology patients and their families might benefit from, and are not a barrier to, early palliative care integration in oncology.

**Methods**

**Survey Development**

Two novel survey instruments, a patient survey and a parent survey (see eAppendix in the Supplement), were developed in a 9-part stepwise process: (1) literature review (including review of all relevant available validated survey tools and related published works); (2) draft instrument development; (3) review of the instrument by experts in pediatric oncology and pediatric PC; (4) instrument revision; (5) cognitive pretest through in-person administration to 10 patient-parent paired participants to assess their understanding of the questions (patients in the younger age range were enrolled for pretesting to ensure understanding among the youngest participants) followed by survey instrument revision in light of the information gained; (6) behavioral pretest through in-person administration to an additional 10 patient-parent paired participants with analysis for linguistic precision, requests for clarification, timing, and other details followed by survey revision based on the information gained; (7) comprehension and feasibility assessment; (8) review; and (9) final survey refinement.

**Participant Enrollment**

After obtaining institutional review board approval at the Pediatric Oncology Branch at the National Institutes of Health (NIH), Johns Hopkins Pediatric Oncology (JH), and St Jude Children’s Research Hospital (St Jude), we approached parent-patient dyads for possible participation in the study. Eligibility criteria included patient with an oncologic diagnosis, age at first diagnosis between 10 and 17 years, who were at least 1 month and no more than 1 year from initial diagnosis. Both patient and parent had to be able to read and write in English and be willing and able to participate. Potential participants were approached while in the clinic for otherwise scheduled visits, after approval from their primary teams. Written informed parent consent and patient assent, per institutional guideline, were obtained prior to participant enrollment. Potential participants were assured that declining to participate would not influence their clinical care. The surveys were administered to enrolled child and parent participants simultaneously in different rooms, and a $10 gift card was given to each participant at completion.
Statistical Analysis
Demographic characteristics and questionnaire responses reported by children and their parents were summarized by descriptive statistics. Agreement of children and their parents regarding the degree of suffering from symptoms was assessed using concordance. Concordance was defined as complete agreement in response categories, including non-response. The McNemar test was used to examine differences in questionnaire responses between children and their parents. The association between the level of current quality of life and time since diagnosis with attitudes toward early PC integration was assessed using a Fisher exact test. The association between the level of current quality of life and time since diagnosis with perceived optimal timing of PC involvement was assessed using a Cochran-Armitage trend test. A 2-sided significance level of \( P < .05 \) was used for all statistical tests. All statistical analyses were conducted using SAS software, version 9.3 (SAS Institute Inc).

Results
A total of 280 eligible patients and parents were approached for inclusion in the study, 258 of whom (129 patient-parent pairs, 92.1% positive response rate) were enrolled in the study from the 3 participating sites from September 2011 to January 2015. Demographic characteristics of the participants did not differ by site, made up a balanced representative sample, and are summarized in the Table. We found no differences among groups in attitudes toward PC when stratified by time from diagnosis.

A large percentage of patients reported suffering from the following symptoms in the first month of cancer therapy: nausea (n = 109; 84.5%), loss of appetite (n = 97; 75.2%), pain (n = 96; 74.4%), anxiety (n = 77; 59.7%), constipation (n = 69; 53.5%), depression (n = 64; 49.6%), and diarrhea (n = 52; 40.3%), (Figure 1A). Among this subset of patients who reported suffering, a substantial proportion reported a high degree of symptom-related suffering (ie, either “a great deal” [the highest level of suffering] or “a lot” [the second highest level of suffering]) from nausea (57 of 109; 52.3%), loss of appetite (49 of 97; 50.5%), constipation (21 of 69; 30.4%), pain (29 of 96; 30.2%), anxiety (22 of 77; 28.6%), depression (18 of 64; 28.1%), and diarrhea (12 of 52; 23.1%). Intradyad patient-parent concordance for exact agreement in reported symptom suffering ranged from 26% to 29%, and concordance for agreement within 1 response category ranged from 40% to 69% (Figure 1B). The majority of both patient (n = 96; 75%) and parent (n = 80; 62%) participants felt that their oncology care teams should focus a great deal or a lot on their quality of life from the beginning of cancer therapy.

The majority of both patients (n = 127; 98.4%) and parents (n = 90; 69.8%) reported that they had never heard the term “palliative care,” and of those familiar with it, none reported a negative attitude toward it. A brief definition of a PC team was given in the survey (see eAppendix in the Supplement): a group of clinicians with expertise in symptom management and a goal of improving quality of life.

Very few children (n = 2; 1.6%) or parents (n = 8; 6.2%) expressed opposition to early PC by indicating that they would definitely not want to meet with a PC team around the time of diagnosis (Figure 2). Very few patient and parent participants demonstrated a perceived detrimental effect of early PC, indicating that it would interfere with their relationship with their oncologist (n = 6 [4.7%] and n = 5 [3.9%], respectively), loss of hope for a cure (n = 3 [2.3%] and n = 10 [7.8%], respectively), or therapy interference (n = 3 [2.3%] and n = 2 [1.6%], respectively) (Figure 3). Children were significantly more likely than parents to endorse that including PC around the time of diagnosis would have been helpful for treating symptoms (40.3% [n = 52] vs 17.8% [n = 23]; \( P < .001 \)) (Figure 3). After learning that, in addition to symptom management and quality-of-life directed care, PC teams also provide end-of-life care, few participants (15.5% of patients [n = 20] and 14.7% of parents [n = 19]) reported that this would have made them less willing to meet with a PC team at the time of diagnosis, while 26.4% (n = 34) of patients and 17.8% of parents (n = 23) indicated it would make them more willing.

Compared with their parents, children were significantly more likely to endorse PC intervention if pain or symptom management was a problem (48.8% [n = 63] vs 34.1% [n = 44];...
Early Palliative Care Integration in Pediatric Oncology

**Discussion**

Perceived barriers to early integration of PC in cancer care include the notion that patients may not have a need for PC services early in the disease trajectory and/or that families, particularly those of young cancer patients, may not be receptive to PC involvement. While previous studies have demonstrated that children with cancer suffer at the end of life, our findings are the first to our knowledge to elucidate a high degree of overall suffering and symptom burden in the first month of cancer therapy. Palliative care intervention has been demonstrated to reduce suffering for dying children, and the high degree of suffering observed in the first month of cancer treatment in this study suggests that early PC integration could offer similar benefit from the initiation of cancer therapy.

Intradyad parent-child concordance for pain and symptom suffering was very low for exact agreement in all of the

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**Figure 1. Patient Perception and Patient-Parent Concordance on Symptom Suffering in the First Month of Cancer Therapy**

(A) Patient perception

- Suffered a little
- Suffered a moderate amount
- Suffered a lot
- Suffered a great deal

(B) Patient–parent concordance

- Exact agreement
- Agreement within 1 category

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**Figure 2. Attitudes Toward Early Palliative Care Integration**

(A) Child

(B) Parent

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P = .01, if the cancer got worse or came back (48.8% [n = 63] vs 31.8% [n = 41]; P = .003), and throughout all of a child’s cancer care (31.8% [n = 41] vs 20.2% [n = 52]; P = .03) (Figure 4). More than half of patient (58.9% [n = 76]) and parent participants (50.4% [n = 65]) indicated that PC teams should be involved in the care of a child with cancer from the beginning of cancer therapy (Figure 4). Patient participants who rated their current quality of life as poor or fair were more likely to indicate that PC teams should be involved from the beginning of cancer therapy (19 of 26; 73.1%) vs those who rated their current quality of life as good (25 of 41; 61.0%) or very good or excellent (32 of 62; 51.6%).

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Attitudes measured via the following question: “Would you want to have met the palliative care team around the time of diagnosis?” Overall percentages may not sum to 100% because missing data are not shown (responses missing for 1 child and 5 parents).
symptoms queried, and when adjusted for agreement within the response category still yielded overall low concordance. While there was consistent lack of agreement within parent-patient dyads in symptom reporting, there was no reliably observed trend toward either overestimation or underestimation of symptom suffering among discordant responses. This suggests that, when possible, symptom assessments should be elicited directly from age-appropriate pediatric patients because proxy reports may not be optimally representative. A recent systematic review of PC literature in children and adolescents found that very few studies included patient voice or patient-reported outcomes and called for increased direct child and adolescent involvement.16 Our finding underscores the importance of pediatric patient inclusion for symptom assessment and confirms the feasibility of such an approach.

Pediatric participants were significantly more likely than their parents to indicate that PC would be helpful for symptom management (40.3% [n = 52] vs 17.8% [n = 23]; P < .001). The high degree of observed symptom-related suffering, the low degree of concordance between parent and child perception of this suffering, and the acknowledgment by young patients themselves that early PC integration would help with symptom management are unique and noteworthy findings that reflect an unmet need for improvement in early symptom management. Pediatric patients were also significantly more likely than parents to recommend that the PC team get involved in their care at multiple time points, including “throughout all of a child’s cancer care” (31.8% [n = 41] vs 20.2% [n = 26]; P = .03). In addition, the observed trend in which child participants who rated their current quality of life as poor or fair were more likely to want PC involvement from the beginning of cancer therapy points toward the potential for improved symptom care for high-risk children. Taken together, these findings validate the importance of pediatric patient inclusion in symptom assessment, substantiate enhanced receptivity to early PC integration with pediatric patient involvement, and support the need for early PC integration.

There is a notion that families are accepting of high symptom burden at the start of therapy because they are focusing exclusively on the pursuit of cure and not quality of life at the time of diagnosis. In contrast to this notion, the majority of patients and parents in our study indicated that teams should...
focus intensely on quality of life from the beginning of treatment. It has been observed that parents of children with cancer nearing the end of life maintain dual goals, comfort and life prolongation, concurrently.¹⁷ Our study is the first to our knowledge to demonstrate that the goals of cure and quality of life are not mutually exclusive and coexist as high priorities among pediatric oncology patients and their parents from the time of diagnosis. In light of the high degree of reported suffering in the first month of therapy and the finding that families place a high priority on quality of life from the beginning of therapy, our study has revealed a new finding that can be mitigated by early PC integration. Palliative care for pediatric oncology patients from the time of treatment initiation has the potential to reduce the severe suffering reported in our study and to focus on optimizing quality of life to help meet observed patient and family priorities.

Despite the notion that families may not be receptive to PC early in the disease trajectory, very few participants expressed opposition to meeting with the PC team around the time of diagnosis or indicated that early PC would have a negative impact on their care. In fact the opposite was observed; the majority of both child and parent participants were open to early integration of PC, and many expressed the perception that early PC integration would be a positive addition to their overall care. While the majority of respondents indicated positive attitudes toward early PC integration, the number of observed unsure responses by participants is notable and underscores the need for clinicians to communicate effectively with patients and families about early PC in oncology. We specifically asked participants about preconceived notions or negative experiences with PC and found that an overwhelming majority had never heard of PC and did not have inherent opposition. We also asked participants in various ways about their attitudes toward PC with respect to intrusion, interference, loss of hope, and association with end-of-life care and found exceedingly few respondents for whom PC was perceived negatively in any of these facets. In fact, early PC integration that is universally applied may mitigate the concerns of the small number of respondents who perceived potential intrusion, interference, or loss of hope.

The observed lack of bias against PC and the overall positive attitude toward early PC integration among pediatric oncology families suggests that families are ready for an integrated model of care. A model of integration recently proposed calls for elements of high-quality PC from the outset of a child’s cancer diagnosis, utilizing PC teams for embedded expertise while maintaining the primary oncology model, systems-based education, and trigger-based consultation for high-risk patients.⁷ Further study is warranted to identify the optimal model of integration as well as to identify improvement in outcomes in such models. It is clear, however, that early PC in oncology has the potential to benefit patients and families and that if families are ready for early PC integration, the medical community should be as well.

Limitations

The high rate of participation in our study is likely owing to the low burden of study participation (a 1-time survey with a maximum duration of 30 minutes), convenience of completion while otherwise in clinic, gift card incentive, and culture of willingness to participate in research. The majority of parent participants were mothers. The abundance of maternal respondents may have affected the parental results and may not be representative of the attitudes of fathers, but as the majority of children’s primary caretakers are mothers, this was an expected phenomenon and is a limitation of unknown significance.

Our study was also limited by its retrospective nature with a potential for recall bias; ie, participants may not have been able to recall the events of 1 to 11 months prior with complete accuracy. The accuracy of recalled information, however, should not affect general participant perception and attitudes, which are primary outcome measures of the study, and thus is not thought to significantly affect the overall findings. Furthermore, when stratified by time from diagnosis (comparison of 1-3, 4-6, 7-9, 10-12-month groupings and 1-6, 7-12-month groupings) we found no differences in attitudes.

The survey tools were developed and tested in English only, so non–English-speaking participants were excluded, which could have led to cultural bias. Age is another study limitation in that children younger than 10 years and their parents were excluded because this young age group was not thought to be cognitively and developmentally capable of participation; thus, the perceptions of this younger population were not assessed. Finally, the survey administered is not a validated instrument, since currently there are no widely acceptable instruments to elicit these data, and our survey could have had questions or wording that led to skewed understanding among participants influencing responses; however, extensive and rigorous survey development procedures and pretesting make this less likely.

Conclusions

Pediatric oncology patients report experiencing a high degree of symptom-related suffering from the beginning of cancer therapy and would likely benefit from early integration of PC. Very few patient or parent participants in this study were opposed to early PC or expressed a negative attitude toward early PC in oncology. Our findings indicate that patient and family attitudes should not be seen as a barrier, but rather a facilitator, to high-quality comprehensive care for pediatric cancer patients and their families through early PC integration in oncology.
Author Contributions: Dr Levine had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Levine, Symons, Wendler. Acquisition, analysis, or interpretation of data: All authors. Drafting of the manuscript: Levine, Mandrell, Sykes, Baker. Critical revision of the manuscript for important intellectual content: Levine, Pritchard, Gibson, Symons, Wendler, Baker. Statistical analysis: Levine, Sykes. Obtained funding: Levine, Baker. Administrative, technical, or material support: Levine, Mandrell, Gibson, Symons, Baker. Supervision: Levine, Symons, Wendler, Baker.

Conflict of Interest Disclosures: None reported.

Funding/Support: This work was funded in part by ALSAC (American Lebanese Associated Charities).

Role of the Funder/Sponsor: ALSAC had no role in the design and conduct of the study; collection, management, analysis, or interpretation of the data; preparation, review, or approval of the manuscript; and decision to submit the manuscript for publication.

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