

Palliative Care of Children With Brain Tumors

A Parental Perspective

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Objective: To explore the end-of-life experience of children with brain tumors and their families.

Design: Qualitative analysis of focus group interviews.

Setting: Children's Hospital, London Health Sciences Center.

Participants: Twenty-five parents of 17 children who had died of brain tumors.

Intervention: Parents participated in 3 semistructured focus group interviews.

Main Outcome Measures: Themes identified through thematic analysis of interview transcripts.

Results: Qualitative analysis identified 3 primary themes. (1) Parents described the dying trajectory of their child as characterized by progressive neurologic deterioration, with the loss of the ability to communicate as a turn-

ing point. Parental coping mechanisms included striving to maintain normality and finding spiritual strength through maintaining hope and in the resilience of their child. (2) Parental struggles during this phase included balancing competing responsibilities and speaking with their child about death. (3) Barriers to achieving a home death included suboptimal symptom management, financial and practical hardships, and inadequate community support. A fourth, secondary theme concerned the therapeutic benefits of the interview.

Conclusion: The neurologic deterioration that characterizes the dying trajectory of children with brain tumors may create significant challenges for health care professionals and the children's parents, supporting the need for increased awareness of the distinct issues in the palliative care of children with brain tumors and for early anticipatory guidance provided for families.

Arch Pediatr Adolesc Med. 2010;164(3):225-230

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DURING THE PAST DECADE, many studies have documented substantial suffering among children dying of cancer.¹⁻²³ Gaps in the palliative care of these children and their families have been identified, including inadequate relief of pain and other symptoms,² communication barriers between the family and the health care team,^{2,4} and stress and regret^{5,6} experienced by parents after the child dies. In the existing literature, the end-of-life (EOL) experience in each disease group has not yet been studied as a unique entity, although research has suggested that there are differences.^{1,7-9} Brain tumors have the highest disease-related mortality rate of all pediatric malignant neoplasms, and these children often have a distinct constellation of symptoms including paralysis, cognitive impairment, and speech and swallowing difficulties.¹ Caring for this

population may pose a unique set of challenges. A better understanding of what these children and their families go through at the end of life would help us more successfully address their needs.

The goal of our study was to explore the EOL experience of children with brain tumors and their families. We chose to use qualitative methods to capture the richness of the data and in turn, to have a greater understanding of the illness than that gleaned from a medical record review or responses to quantitative questionnaires. "Experts in the field," that is, parents of children who had died of a central nervous system malignant neoplasm, were approached to participate in 1 of 3 focus group interviews. This article describes the themes identified by the participants. This research constitutes a preliminary phase of a large multicenter study to develop and validate the Pediatric Supportive Care Scale, an instrument to mea-

Table 1. Semistructured Questions Used in the Focus Group Interview

1. Please describe your child's and family's experience while caring for your child in the last stages of his or her life.
2. What aspects of this experience were most difficult for you?
3. What aspects of your child's care were positive or helpful?
4. What areas of your child's care were in need of improvement?
5. How would you educate future physicians or health care team members about your experience?
6. Are there any other issues that have not been mentioned that anyone feels are important to discuss?

Table 2. Demographic Characteristics of Participants and Children

Characteristic	Total No.
Participants	25
Sex	
Male	9
Female	16
Age range, y	
30-39	6
40-49	10
50-59	8
≥60	1
White	25
Children	17
Sex	
Male	12
Female	5
Age at the time of death, y	
1-5	3
8-11	3
12-19	11
Length of illness ^a	
<12 mo	9
1-5 y	6
>5 y	2
Underlying diagnoses	
Brainstem glioma	4
Supratentorial primitive neuroectodermal tumor	4
Low-grade astrocytoma	2
High-grade astrocytoma	3
Medulloblastoma	2
Ependymoma	1
Choroid plexus carcinoma	1

^aTime from diagnosis to death.

sure the quality of life and health care satisfaction in children with terminal cancer.

METHODS

Our study was a qualitative thematic analysis of the experiences of families of children who died of a brain tumor. We chose the focus group interview as our method of data collection. This method has been used extensively in palliative care research.^{7,10-12} Focus group interviews are facilitator-guided interactive discussions.^{7,13} The interaction of the participants is seen as key in that discussants build on each other to come to a consensus that as individuals they would not have articulated on their own. The use of a semistructured format provides an opportunity for participants to tell their personal nar-

atives while ensuring that they respond to a particular set of questions.⁷

Parents of children who died of a brain tumor while under the care of the Children's Hospital, London Health Sciences Center, between January 1, 1996, and December 31, 2006, were eligible to participate. Exclusion criteria included the death of a child within the past year or families identified by their primary oncologist as having the potential to experience significant psychological repercussions from participating. Approval was obtained from our institution's review and ethics board. Parents were contacted initially by an introductory letter, followed by a telephone call from a member of the research team. Three separate focus groups were conducted with 8 to 10 participants in each group interview. Each interview was facilitated by a member of a research team (S.Z.) and followed a series of methodologically developed semistructured questions (**Table 1**). The questions were developed by members of the research team and reviewed by a parent representative whose child had died of a brain tumor. Each group interview lasted approximately 2 hours. The focus groups were conducted in a private, confidential, and neutral environment outside the hospital at a state-of-the-art focus group facility in London (Acumen Research Group; <http://www.acumenresearch.com>). The interviews were audiotaped and videotaped, and detailed notes were taken by other members of the research team behind a 1-way mirror (D.C. and S.L.B.). A feedback form was completed by all parents at the end of the interview. A follow-up telephone call was made by 2 of the research team members (S.Z. and D.C.) within 2 days after the focus group. A thank-you letter was also sent to all participants.

DATA ANALYSIS

Thematic analysis identified common recurring themes in the transcripts. Thematic analysis is an established qualitative research method that allows the researcher to identify patterned response or meaning within the data set (themes) through a 6-step process outlined by Braun and Clarke,¹⁴ which includes familiarizing oneself with the data, generating initial codes, searching for themes, reviewing themes, defining and naming themes, and producing the report.¹⁵⁻¹⁷ Data analysis was conducted simultaneously by 2 members of the research team (S.Z. and D.C.). To ensure interrater reliability, each researcher reviewed the other's work. Analysis was facilitated using NVIVO 7 software (QSR International, Doncaster, Australia).

RESULTS

DEMOGRAPHIC DATA

Of the 47 families who were eligible to participate, 1 was excluded because of travel time and 4 were excluded because their treating oncologist deemed them as not suitable for approach. Another family was excluded because it included the parent representative for the study. We attempted contact with 41 families by an introductory letter sent in the mail, followed by a telephone call. Eighteen families could not be reached owing to a change in their address or telephone number. Of the 23 families who could be contacted, 19 agreed to participate. Two families agreed to participate but then did not attend the interview.

A total of 25 parents of 17 deceased children participated in the interview. The median time since their child's death was 5.5 (range, 3-12) years. The demographic characteristics of the participants and deceased

Table 3. Themes Characterizing the End-of-Life Experience for Children With Brain Tumors and Their Families

Component	Illustrative Quotations ^a
Theme 1: Dying Trajectory	
Neurologic deterioration	“He knew he was dying. We kept trying to be positive, like ‘we’ll do this and do that,’ but then the next week he couldn’t play PlayStation anymore and the week after, now he was in a wheelchair, and then he needed help to go to the washroom.” (FG1; L239-L242) “Everything started to go in the last month, and he struggled with that.” (FG1; L78) “I would say that was the hardest on him, that he couldn’t read or play the games he liked to play or watch television and watch movies.” (FG1; L98)
Loss of ability to communicate	“Her sentences were choppy, like we would catch 1 word and she would get frustrated, and then we had to be patient with her because sometimes I didn’t even understand her.” (FG1; L161-L163)
Striving to maintain normality	“Even with the size of his tumor, he went to school and graduated high school. He had great support from his peers and family and friends.” (FG3; L160-L161) “We lived with his brain tumor and we didn’t put him in a bubble; he would carry a cell phone when he would go out with his friends.” (FG3; L270-L271)
Sources of spiritual strength	
Maintaining hope	“You always have that hope that this is going to be the one that solves everything; you don’t want to give that up.” (FG2; L109-L110)
Resilience	“We were surrounded with love. He knew that; he was so good, he directed his own care and directed us and he had good quality of life.” (FG3; L1280-L1281)
Theme 2: Parental Struggles	
Competing responsibilities	“Because you do go into nurse mode, it is a survival mode of meeting their clinical needs and yet you are their parent so trying to do the emotional part too, and you are being the strong one constantly.” (FG2; L333-L334)
Conversations about death and dying	“Because one of the things that I regret, looking back, was never having that talk with your child; like, how do you talk to your child about the fact that he or she is going to die, and I would have liked to know more about what was going on in his mind.” (FG2; L114-L115) “My son, I think he realized sooner than I did that he was not going to make it. For instance, he was giving his belongings to some of his friends and stuff. I was against that; I was like, you are going to need those.” (FG2; L97-L98)
Theme 3: Dying at Home	
Dying at home	“Also asking you what is your request; do you want your child to die at home or in the hospital? We chose home, but to get him home was extremely difficult and there were costs that we had to figure out to get him home.” (FG2; L64-L66) “And the biggest thing I felt, I felt a little abandoned, not when we were in; this was when we were at home.” (FG1; L1006-L1007)

^a Illustrative quotations have been slightly paraphrased to increase readability. Following each quotation is the focus group (FG) number (1, 2, or 3) and the transcript lines (L) from which the passage was taken.

children are outlined in **Table 2**. One parent stated that her child had been identified as having a familial cancer syndrome, and one of her own siblings had died many years before of a brain tumor.

THEMES

The following 3 major themes that characterized the EOL experience were identified: (1) the dying trajectory, (2) parental struggles, and (3) dying at home. A fourth, secondary theme included parental descriptions of the therapeutic benefits of the interview. These themes are described herein, along with the specific components within each domain. **Table 3** presents selected direct quotations that are representative of each of the themes.

THEME 1: THE DYING TRAJECTORY

The *dying trajectory* is the term we applied to parental narratives of what it was like to live through and bear witness to their child actively dying. Mechanisms by which they tried to cope as a family, including striving to maintain normality and finding spiritual strength, were iterated.

Neurologic Deterioration

This component encompassed the progressive functional impairment these children experienced as a result of growth of their tumor. Parents recalled their child’s loss of gross motor function, with resulting paralysis and poor mobility, loss of continence, loss of fine motor function, speech and swallowing difficulties, and visual and auditory impairment. Many children were described as aware of their deterioration and were frustrated or saddened and/or depressed by their inability to play or partake in the activities they most enjoyed. It was this aspect of the entire experience that many parents stated was the hardest on their child psychologically.

Loss of the Ability to Communicate

The loss of the ability to communicate with their child was described as a significant turning point for the parent and child. This not only included the child’s loss of the ability to speak and be understood but also the inability to communicate by writing or other nonverbal means. Anger and frustration were emotions experienced by the child and the family at not being able to understand the child’s wants and needs.

Striving to Maintain Normality

The importance of maintaining life as normally as possible appeared consistently throughout all focus group transcripts and across all age groups of the deceased children. This theme captured the importance of the children continuing to “live their lives” despite the hardships they were enduring. Schooling in particular was mentioned frequently, and the ability to complete a grade or to simply continue attending was referred to with a sense of pride and accomplishment. The maintenance of childhood friendships and the involvement and support of peers was of significance in the adolescent population. Parents encouraged the involvement of friends even in the final stages of their teenager’s illness.

Sources of Spiritual Strength

Maintaining Hope. This domain encompassed the importance of hope as giving strength to the child and family in the face of adversity. Maintaining hope for a cure was a prominent theme across all focus groups, even at the end stages of life. Families described the ability to hold on to 2 dichotomous beliefs: the realism that their child’s prognosis was poor and the search for a miracle.

Resilience. This theme encompassed parental descriptions of the strength and resilience that their child displayed during the illness. Most of these accounts were by parents of adolescents. These descriptions were in the context of awe of and admiration for their child because many parents believed that they themselves did not have the same fortitude.

THEME 2: PARENTAL STRUGGLE

Parental struggle was another prominent theme. It captures 2 aspects of the parents’ experience with which they struggled physically and emotionally. These included balancing the care of their child with other competing responsibilities and their own internal struggles concerning how to speak with their child about death and dying.

Competing Responsibilities

Parents described the often insurmountable task of meeting their child’s needs while also balancing their own jobs, financial stressors, and care of their other children. The business of their lives often, in retrospect, kept them from enjoying their child and simply spending time with him or her. Many parents were conflicted over performance of nursing duties, such as medication administration. Although they wanted to be the primary caregiver, they often believed that their clinical duties hindered them from being “just a parent.”

Conversations About Death and Dying

This theme encompassed the parental experience in talking about death and dying with their child. Parents ob-

served that their child was aware of and accepted the reality of the advanced stage of illness before they were. Often the child guided the parents through the process of closure and saying goodbye. Some parents who had not discussed death with their child expressed regret in not doing so. Reasons as to why they did not talk to their child about death included being in denial and not wanting to destroy the hope they and their child had for a cure.

THEME 3: DYING AT HOME

This theme encompassed the barriers parents faced in caring for their child at home. Parents who chose to have their child die at home described many reasons for doing so. These included wanting their child to die in his or her own home, surrounded by loved ones, and parents having the desire to be the primary caregiver for their child up to the end. However, 3 prominent barriers to obtaining a home death were identified. The first was that of optimum symptom management—in particular, seizure control—and the belief that symptoms could no longer be adequately managed or controlled at home. A second barrier included the financial and practical hardships of caring for their child at home. Examples of this included the physical difficulties in caring for their child and navigation of a complex health care system to set up the services their child needed. The third barrier was support within their community: families who were linked with a supportive community physician and home care services believed that their needs were met and they were well supported at home. Families who did not have access to such a network often felt lost and abandoned.

THEME 4: THERAPEUTIC BENEFITS OF THE INTERVIEW

Parents were eager to share their stories and participate in the interview. They hoped that their input might assist families in the future. Sharing their story with others who had gone through a similar experience was described as a reassuring and a therapeutic experience. Some parents stated that they were grateful that their child had not been forgotten. Others expressed a sense of cathartic relief after participating.

COMMENT

Our qualitative analysis identified 3 major themes in the parental perspective of the EOL experience of children with brain tumors and their families. As described by the parents in our sample, children with brain tumors have a constellation of neurologic symptoms that predominate their dying trajectory. As their disease advances, these children experience progressive deficits and loss of function. The loss of communication, in particular, was highlighted by parents as a significant turning point in the dying trajectory. This neurologic deterioration in turn has a significant effect on the quality of life of the child and family and implications in their EOL care. For example, the loss of communicative abilities may affect the child’s ability to express emotions, desires, and wishes,

including, in particular, in conversations about death and dying and advanced care planning. To ensure that we as clinicians are aware of our patients' wishes, these issues should be sensitively explored early in the stage of illness when cognition and communication are not impaired.

Our parents identified ways in which they tried to cope as families. This included "maintaining normality," a theme that has been identified previously in the literature.^{10,18-21} Maintaining normality can preserve a sense of dignity in those who are terminally ill.^{22,23} Our care of these patients should include supporting them in achieving these goals. The second coping mechanism identified by our parents was finding spiritual strength through maintaining hope as well as from the courage and resilience of their child. The parents in our sample described their child as often playing a role in guiding them through the child's dying process. This observation is supported by the literature that has demonstrated that adolescents are cognitively capable of participating in complicated EOL decision making.²⁴ We as clinicians must be aware of the resilience of our patients and must try to include them, when possible, in health care decision making.

Parents were pleased that they had taken part in this study and shared their stories. Consistent with our findings, despite hesitancy in approaching bereaved families for research, parents often report benefit from participation, including the opportunity to share their painful experience and to allow the process of meaningful reconstruction to occur through sharing their narratives.^{25,26} This is despite their account that it is painful to speak about their loss.^{26,27} During our follow-up telephone call, none of our parents had experienced undue levels of distress, and no one requested a referral for counseling. This lack of adverse event reporting is confirmed in many other pediatric oncology EOL studies.^{12,21,26} Expectations that participant harm and burden are inherent in EOL studies is a challenge in pediatric palliative care research and often is a stumbling block in obtaining ethics approval.²⁶ Evidence-based guidelines have been published to aid future researchers in overcoming these barriers in future EOL studies in pediatric oncology.²⁷

Limitations of this study include the fact that we were unable to establish contact with many eligible families. Because eligible families were contacted several years after their child's death, many had relocated, changed telephone numbers, or both. The small sample size, gathered from a single institution, may limit the generalizability of our findings. Finally, our participants were 3 to 12 years removed from the child's death, and their recount of events and emotions was influenced by the coping mechanisms they have used over time. Therefore, the data gathered are not completely reflective of what families experienced during the dying process. However, many parents brought unresolved issues to the table, issues they identified that they had struggled with since the death of their child and about which they wanted to hear feedback from the group. For example, one parent who had regretted not speaking to his teenage son about his fears of dying stated that his motivation in coming to the group was to hear what other parents had done. At the end of the interview, he stated that he had a sense of cathartic relief

in sharing his story and in learning that other families had struggled with similar issues.

To our knowledge, this is the first study to describe the EOL experience of children with brain tumors as a separate and distinct group. We hope this report will increase the awareness of health care professionals concerning the challenges these families face and the need for anticipatory guidance and education of patients and families early in the course of illness.

Accepted for Publication: October 21, 2009.

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Author Contributions: Dr Zelcer had full access to all 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:* Zelcer, Cataudella, Cairney, and Bannister. *Acquisition of data:* Zelcer and Cataudella. *Analysis and interpretation of data:* Zelcer and Cataudella. *Drafting of the manuscript:* Zelcer and Cataudella. *Critical revision of the manuscript for important intellectual content:* Zelcer, Cataudella, Cairney, and Bannister. *Obtained funding:* Zelcer, Cataudella, Cairney, and Bannister. *Study supervision:* Bannister.

Financial Disclosure: None reported.

Funding/Support: This study was supported by a grant from the Pediatric Oncology Group of Ontario.

Role of the Sponsor: The sponsor did not have any role in the design or conduct of the study; in the collection, analysis, or interpretation of the data; or in the manuscript preparation, revision, or approval.

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Announcement

Trial Registration Required. In concert with the International Committee of Medical Journal Editors (ICMJE), *Archives of Pediatrics and Adolescent Medicine* will require, as a condition of consideration for publication, registration of all trials in a public trials registry (such as <http://ClinicalTrials.gov>). Trials must be registered at or before the onset of patient enrollment. This policy applies to any clinical trial starting enrollment after July 1, 2005. The trial registration number should be supplied at the time of submission.

For details about this new policy, and for information on how the ICMJE defines a clinical trial, see the editorials by DeAngelis et al in the September 8, 2004 (2004;292:1363-1364) and June 15, 2005 (2005;293:2927-2929) issues of *JAMA*. Also see the Instructions to Authors on our Web site: www.archpediatrics.com.