

Psychological Distress in Parents of Children With Advanced Cancer

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Importance: Parent psychological distress can impact the well-being of childhood cancer patients and other children in the home. Recognizing and alleviating factors of parent distress may improve overall family survivorship experiences following childhood cancer.

Objectives: To describe the prevalence and factors of psychological distress (PD) among parents of children with advanced cancer.

Design: Cohort study embedded within a randomized clinical trial (Pediatric Quality of Life and Evaluation of Symptoms Technology [PediQUEST] study).

Setting: Multicenter study conducted at 3 children's hospitals (Boston Children's Hospital, Children's Hospital of Philadelphia, and Seattle Children's Hospital).

Participants: Parents of children with advanced (progressive, recurrent, or refractory) cancer.

Main Outcome Measure: Parental PD, as measured by the Kessler-6 Psychological Distress Scale.

Results: Eighty-six of 104 parents completed the Survey About Caring for Children With Cancer (83% par-

ticipation); 81 parents had complete Kessler-6 Psychological Distress Scale data. More than 50% of parents reported high PD and 16% met criteria for serious PD (compared with US prevalence of 2%-3%). Parent perceptions of prognosis, goals of therapy, child symptoms/suffering, and financial hardship were associated with PD. In multivariate analyses, average parent Kessler-6 Psychological Distress Scale scores were higher among parents who believed their child was suffering highly and who reported great economic hardship. Conversely, PD was significantly lower among parents whose prognostic understanding was aligned with concrete goals of care.

Conclusions and Relevance: Parenting a child with advanced cancer is strongly associated with high to severe levels of PD. Interventions aimed at aligning prognostic understanding with concrete care goals and easing child suffering and financial hardship may mitigate parental PD.

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CARING FOR A CHILD WITH cancer can be profoundly distressing to parents,^{1,2} and in turn, parental psychological distress (PD) can affect child³⁻⁹ and sibling well-being.^{10,11} Caring for a child with advanced cancer is likely to be even more distressing, yet very few prospective studies have evaluated parent PD among these families. Not only do parents witness their child's protracted physical and emotional suffering,¹²⁻¹⁴ they also must contend with the ultimate threat to their child's life,¹⁵ which is heightened among children with more advanced stages of disease. Prolonged changes in employment and financial challenges^{1,2,12,13,16,17} may also exacerbate parental PD.

We have previously reviewed the psychosocial morbidities of bereaved parents

of children with cancer¹⁸; however, only 2 studies have been conducted among parents of children with advanced or progressive cancer.^{19,20} One study assessed parent emotional adjustment¹⁹ and the other, global levels of parent health during the end-of-life period.²⁰ Neither study used standardized instruments nor did they report on factors contributing to PD, such as degree of prognosis acceptance, goals of therapy, child suffering, or financial hardship.

Understanding the degree of PD and contributing factors among parents with children with advanced cancer has the potential to better enable clinicians to identify higher-risk families and optimize outcomes. In addition, better insight may enable the design and implementation of parental supportive interventions. Accordingly, in this report, we describe the preva-

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lence of PD among parents of children with advanced cancer and explore whether parental PD is associated with known “medical,” “child,” and “parent” factors, including parent perceptions about the child’s experience.

METHODS

Data were collected within the context of the Pediatric Quality of Life and Evaluation of Symptoms Technology (PediQUEST) study, a pilot randomized trial of a supportive care intervention in children with advanced cancer. The primary goal of the PediQUEST study was to assess the PediQUEST system, a software that allows electronic collection of patient-reported outcomes and generation of feedback reports for providers and families. After their parents provided informed consent, consecutive children were enrolled if they were at least 2 years of age; they had at least a 2-week history of progressive, recurrent, or nonresponsive cancer; they received cancer care at 1 of the 3 participating sites (Dana-Farber Children’s Hospital Cancer Center, Children’s Hospital of Philadelphia, or Seattle Children’s Hospital); and their parents or legal guardians had written command of English and were able to understand and complete self-administered surveys. As part of the study, we also surveyed parents or legal guardians of patients at the time of enrollment to assess their views on their child’s illness. Parents were mailed or handed the Survey About Caring for Children With Cancer (SCCC) along with a self-addressed, stamped return envelope and a \$5 coffee-card incentive. Two biweekly reminders were done (by telephone or face to face if the patient came to the clinic) for parents who did not respond. Families selected 1 parent per enrolled child to complete the survey. One hundred four children were enrolled from December 2004 to June 2009.

STUDY INSTRUMENT

The SCCC was previously developed by our team based on expert interviews and parent focus groups.²¹ This comprehensive paper-and-pencil, self-administered survey evaluates parents’ perceptions about the child’s illness (current treatment, prognosis, treatment goals, and suffering from treatment or the illness) and financial impact of the illness. In addition, the SCCC includes validated measures of PD and social support and collects parent-reported sociodemographic information. Child’s age, sex, diagnosis, and dates of disease progression were abstracted from the medical records.

MAIN DEPENDENT VARIABLE

Parent PD was the main dependent variable and was measured by the Kessler-6 Psychological Distress Scale (K6).²² This 6-item screening tool was developed for the US National Health Interview Survey and is used by the World Health Organization World Mental Health Survey Initiative to assess population-level mental health. The K6 was selected for this study because it is relatively quick and has been widely validated showing high discriminative properties, including among parents of ill children.^{22,23} The instrument asks: “During the past 30 days, how often did you feel (a) nervous? (b) hopeless? (c) restless or fidgety? (d) so depressed that nothing could cheer you up? (e) that everything was an effort? (f) worthless?” Possible responses are “none of the time,” “a little of the time,” “some of the time,” “most of the time,” and “all of the time.” Responses are scored on a 5-point Likert scale and summed to generate a total symptom score between 0 and 24; scores of 7 or more are indicative of high PD and scores of 13 or greater meet the criteria of serious psychological distress (SPD).²⁴ In the United States, mean K6 scores are approximately 2.5 for well adults.²⁵ Serious, debilitating PD is

rare, with an estimated prevalence of 2% to 3% among well adults²⁶ and higher prevalence among adults with chronic or life-threatening illness.²⁷ Parental PD is reported as mean K6 scores and proportion of parents with SPD.

INDEPENDENT VARIABLES

Child and Medical Factors

Child-related variables included the child’s sex, age, cancer type, current or ongoing receipt of cancer-directed therapy, time from first disease progression to survey date, and child suffering during past month due to symptoms and treatment. Child age was stratified into 3 groups: preschool aged (ages 2-6 years), school aged (ages 7-12 years), and adolescent (≥ 13 years). Whether the child was receiving cancer-directed therapy at the time of PediQUEST enrollment was reported by parents on the SCCC. All other variables were abstracted from the medical record.

Parent Factors

All parent factors were obtained from the SCCC. Sociodemographic variables included parents’ sex, age, race/ethnicity, marital status, level of education, annual income, and social support. Parent age and social support scores were dichotomized at the 25th percentile because we postulated a priori that younger or less supported parents would have greater distress. Social support was assessed using an adapted, 9-item, validated version of the social support subscale of the Medical Outcomes Study, which measures self-reported available support.²⁸ Medical Outcomes Study social support subscale raw scores (ie, simple algebraic sums) were transformed into a 0- to 100-point scale; the higher the score, the better the perception of social support. Parents were also asked to share: (1) their understanding of their child’s current prognosis, (2) primary goal pursued by providing the current medical therapy to their child, (3) degree of child suffering, and (4) degree of family economic hardship due to their child’s cancer. Responses were elicited using 4- and 5-point Likert-type scales and then collapsed for analysis as delineated in the text and tables. Parent perception of prognosis was collapsed into 2 categories: likely to be cured (cure likely/very likely) and unlikely to be cured (unlikely/very unlikely).

Primary goal of current medical treatment held 5 response options (to cure, to keep hoping, to make sure everything has been done, to extend life without hope of cure, and to lessen suffering) developed and validated through qualitative research and used in prior studies.²¹ Parents were asked to select only 1 goal. Prognostic understanding was considered aligned with a concrete goal if parents reported the child was likely to be cured and endorsed the goal of cure or if parents reported the child was unlikely to be cured and endorsed the goal of extending life or lessening suffering.

STATISTICAL METHODS

Analyses were performed using Stata statistical software version 10.1 (StataCorp). A complete case analysis was performed; missing data were not imputed. Frequencies and means were used to characterize parent, medical, and child variables. To evaluate the association of parental PD and child and parent factors, we ran univariate linear and logistic regression analyses where PD was modeled both as a continuous (increase in mean K6 scores) and dichotomous (likelihood of presenting with SPD) outcome. We ran exploratory multivariate analysis to analyze the independent effect of each of these factors. Variables with a *P* value $< .10$ in the univariate analyses were added in a forward, stepwise fashion into multiple linear regression models.

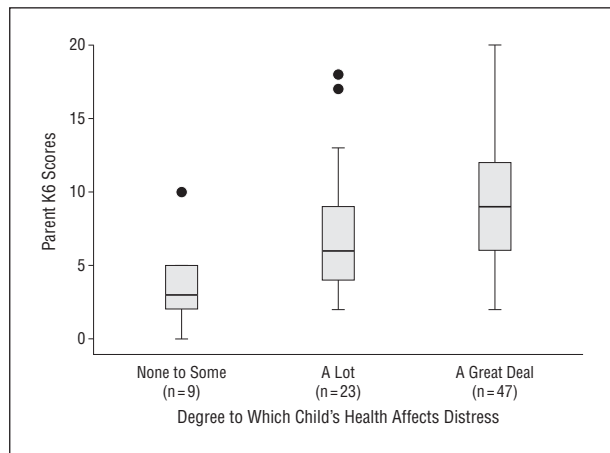


Figure 1. Parent Kessler-6 Psychological Distress Scale (K6) scores stratified by the degree to which child's health affects distress. Distribution of parental distress scores as defined by the K6 among the 79 parents with complete K6 data who also answered the question: "During the last 30 days, to what extent has your child's health contributed to these feelings?" with options "a great deal," "a lot," "somewhat," "a little," or "not at all" ($P < .001$ based on Kruskal-Wallis test).

RESULTS

Seventy-one percent of eligible, approached families enrolled in the PediQUEST study. Of the 104 enrolled families, 86 parents (83%) returned the SCCC. This analysis reports on the 81 parents (78% of the total sample) with complete K6 data. Child's age, type of cancer, and duration of progressive disease were comparable among SCCC respondents, nonrespondents, and respondents with complete and incomplete K6 data.

The mean (SD) parental PD (K6) score was 7.9 (4.4) and 50% of parents had a score of 8 or higher (interquartile range, 5-11). Thirteen parents (16%; 95% CI, 8%-24%) met criteria for serious PD. Fifty-five parents (69%) reported that they felt more distressed than usual, 14 (18%) said they were as distressed as usual, and 10 (13%) reported less distress than usual. Seventy parents (89%) reported that their child's health contributed "a lot or a great deal" to their distress and these parents tended to have higher K6 scores than parents who reported their child's health contributed less to their distress (**Figure 1**).

At the time of enrollment, most children were receiving cancer-directed therapy and 85% had been diagnosed with progressive disease for at least 2 months (**Table 1**). The median duration of progressive disease was 195 days (interquartile range, 86-353 days). Only 3 children (4%) died within 3 months of enrollment. Respondents were predominantly white women who were older than 38 years of age and married. Parents reported high levels of social support (standardized median score, 83%). No child or parental demographic factors were associated with mean K6 scores or risk of parent SPD at the .05 level.

Forty-seven percent of parents believed that cure for their child was likely (**Figure 2**) and the vast majority of these parents endorsed the concrete primary goal of cure (90%). Among the 53% of parents who reported cure was unlikely for their child, 30% endorsed the concrete primary goals to "extend life without hope of cure" (27%) or "lessen suffering" (3%). Parents who believed their child

Table 1. Main Child and Parent Characteristics and Mean Parental K6 Scores

	Total (n = 81 [100%]) ^a	K6 Score, Mean (SD) ^b
Child characteristics		
Sex		
F	43 (53)	7.2 (4.2)
M	38 (47)	8.7 (4.5)
Age, y		
2-6	23 (28)	9.1 (3.5) ^c
7-12	19 (23)	7.6 (5.4)
≥13	39 (48)	7.3 (4.3)
Diagnosis		
Hematological malignancy	27 (33)	7.8 (4.2)
Solid tumor, non-CNS	45 (56)	7.9 (4.6)
Brain tumor	9 (11)	8.1 (4.5)
Currently receiving cancer-directed therapy (n = 80)		
Yes	73 (91)	8.0 (4.3)
No	7 (9)	7.3 (6.0)
Duration of progressive disease, mo		
<2	12 (15)	8.3 (3.9)
≥2	69 (85)	7.8 (4.5)
Parent characteristics		
Sex		
F	70 (86)	8.2 (4.2)
M	11 (14)	5.6 (5.1)
Age, y (n = 80)		
<38	17 (21)	9.6 (4.9) ^d
≥38	63 (79)	7.3 (4.0)
Race/ethnicity (n = 78)		
White/non-Hispanic	72 (90)	7.6 (4.0)
Other race/Hispanic	6 (10)	11.0 (8.2)
Marital status		
Married or living with partner	70 (86)	6.5 (5.3)
Never married, divorced, or separated	11 (14)	8.1 (4.2)
Standardized social support score (n = 78)		
<69%	5 (6)	9.2 (7.5)
≥69%	73 (94)	7.7 (4.1)
Education		
High school, GED, or less	25 (31)	7.6 (4.7)
≥College education	56 (69)	8.0 (4.3)
Annual income, \$ (n = 73)		
<25 000	10 (14)	9.1 (6.5)
25 000-49 999	9 (12)	5.9 (4.5)
≥50 000	54 (74)	7.7 (3.9)

Abbreviations: CNS, central nervous system; GED, General Educational Development; K6, Kessler-6 Psychological Distress Scale.

^an = 81 Indicates the number of parents with complete K6 data. Denominator is indicated when child or parent characteristics had missing data.

^bThe K6 scores had a range of 0 to 24; higher scores suggest greater distress. All P values $> .10$, unless otherwise indicated, based on univariate linear regression models; categorical variables with 2 df used for child's age and child's diagnosis.

^c $P = .08$.

^d $P = .07$.

to be curable had lower distress scores (**Table 2**) and parents whose prognostic understanding was aligned with concrete primary goals also had lower distress scores as well as a lower likelihood of SPD. Further, parents who believed their child benefited from current cancer-directed therapy tended to have lower distress and those who felt their child was suffering from therapy had greater distress. While somatic symptom-related suffering was not related to parental distress, child emotional suffer-

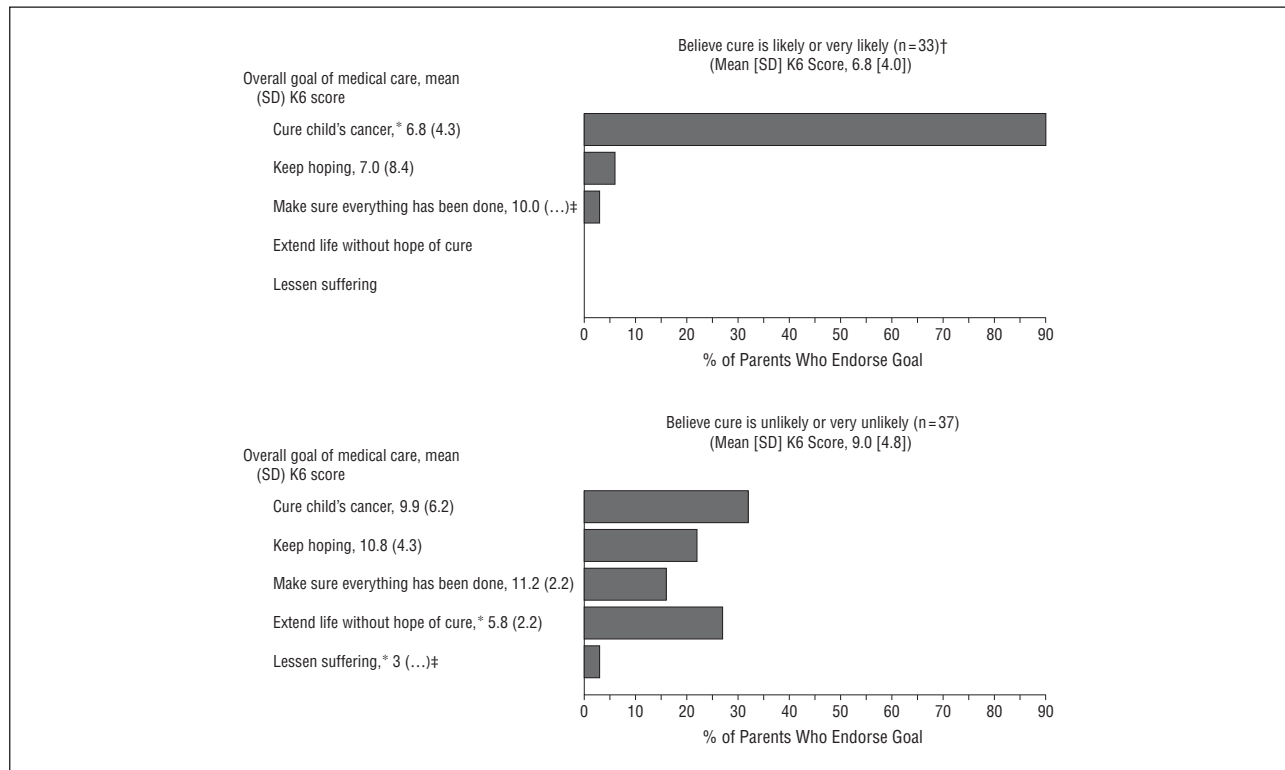


Figure 2. Parent Kessler-6 Psychological Distress Scale (K6) scores associated with perceptions of cure likelihood and corresponding goals of therapy. Distribution of parents' treatment goals according to their understanding of prognosis of their child, with their corresponding mean (SD) K6 scores are presented. *Goals that were concrete and aligned with prognostic understanding. Parents who reported concrete and aligned treatment goals reported lower K6 scores, ie, less distress, than parents who reported other treatment goals. †Two of 33 parents in "likely" or "very likely" group did not report overall goal of medical care. ‡Standard deviation could not be estimated.

ing was. For example, parents who believed their child was sad or anxious had higher distress scores and higher risks of SPD, whereas those who believed their child suffered from pain did not report consistently elevated distress. Parents who perceived that their child was suffering "a lot" overall, however, tended to have higher average distress scores and a higher likelihood of SPD.

Twenty-four families (30%) perceived "great" economic hardship due to their child's cancer and this perception was associated with increased average K6 scores and increased risk of SPD. These trends remained when analyses were adjusted by annual income category.

In multivariate models, average parent K6 scores were 4 points lower among parents whose prognostic understanding was aligned with concrete goals of care, independent of suffering or financial hardship (**Table 3**). Conversely, distress was 3 or more points higher among those who believed their child was suffering highly and who reported great economic hardship. Sensitivity analyses that included adjustments for study site, parent sex, child's age, and the use of 3 categories of alignment of prognostic understanding with concrete goals (cure likely with goal of cure, cure unlikely with palliative goals, and cure unlikely with nonpalliative goals) did not significantly affect these results.

COMMENT

Screening for PD is a critical element of comprehensive, psychosocial care of patients with cancer and their fami-

lies.²⁹ Prior studies have suggested that such screening may improve communication between patients and clinicians and may ultimately improve patient well-being.³⁰ We sought to evaluate PD among parents of children with advanced cancer and found the experience of PD to be highly prevalent. More than half of parents report high levels of PD and approximately 1 in 7 experience SPD. In comparison, parents of children with progressive cancer are 4 times more likely to develop SPD than parents of well children³¹ and more than 2 times more likely to develop SPD than adults with cancer or other serious medical conditions.^{27,32} These findings imply that parenting a child with progressive cancer can profoundly affect mental health.

High PD, as determined by the K6 screen, is highly predictive of serious mental illness (area under the curve, 0.87).²² Adults with high PD have increased risks of poor health outcomes,³³ workplace productivity loss,³⁴ and mortality.³⁵ In addition, PD among parents of well children has been associated with lower attainment of child developmental milestones.³⁶ Among children with cancer, parent PD can ultimately affect patient³⁻⁹ and sibling^{10,11} psychosocial outcomes. Alleviating parent PD may lead to more optimal patient and family health.

Our findings confirm that parental PD is associated with perception of cure likelihood. In this study of patients with advanced cancer, we found strikingly wide variability in the duration of progressive disease duration. However, nearly half of parents still believed cure was likely for their child, and those who did also had lower distress scores.

Table 2. Parent Perceptions of Child's Illness Associated With PD and SPD,^a Univariate Analysis

	Total (n = 81 [100%])	K6 Score, Mean (SD)	P Value ^b	Likelihood of SPD, ^c OR (95% CI)
Prognosis and goals of therapy				
What is your understanding now of how likely it is that your child will be cured? (n = 70)				
Likely/very likely	33 (47)	6.8 (4.0)	.04	0.4 (0.1-1.6)
Unlikely/very unlikely	37 (53)	9.0 (4.8)		1 [Reference]
Prognostic understanding aligned with concrete goals (n = 70)				
Yes	41 (59)	6.3 (3.7)	.001	0.2 (0.1-0.9)
No	29 (41)	10.2 (4.7)		1 [Reference]
Perceived child symptoms and suffering				
Benefit from current cancer-directed treatment (n = 68)				
Not at all/a little	5 (7)	10.0 (1.9)	.01	0 ^d
Somewhat/a lot/a great deal	63 (93)	7.5 (4.4)		1 [Reference]
Suffering from current cancer-directed treatment (n = 76)				
A lot/a great deal	16 (21)	11.0 (3.6)	<.001	3.0 (0.8-10.9)
Not at all/a little/somewhat	60 (79)	7.1 (4.3)		1 [Reference]
Frequency of feeling afraid (n = 75)				
Some/most/all of the time	29 (39)	9.4 (4.6)	.04	2.1 (0.6-7.2)
None/a little of the time	46 (61)	7.1 (4.1)		1 [Reference]
Frequency of feeling calm or peaceful (n = 78)				
None/a little of the time	10 (13)	10.7 (5.0)	.03	3.2 (0.7-15.2)
Some/most/all of the time	68 (87)	7.2 (3.9)		1 [Reference]
Suffering from anxiety (n = 69)				
A lot/a great deal	12 (17)	11.8 (5.9)	.01	7.1 (1.8-28.7)
Not at all/a little/somewhat	57 (83)	7.4 (3.8)		1 [Reference]
Suffering from sadness (n = 69)				
A lot/a great deal	7 (10)	12.4 (6.3)	.04	7.9 (1.5-41.6)
Not at all/a little/somewhat	62 (90)	7.6 (3.9)		1 [Reference]
Suffering from pain (n = 75)				
A lot to great suffering	29 (39)	9.0 (5.1)	.19	2.1 (0.6-7.2)
None to some suffering	46 (61)	7.6 (3.8)		1 [Reference]
Suffering from nausea or vomiting (n = 74)				
A lot to great suffering	17 (23)	9.9 (4.7)	.05	2.5 (1.1-14.0)
None to some suffering	57 (77)	7.5 (4.2)		1 [Reference]
Suffering from sleep disturbances (n = 67)				
A lot to great suffering	17 (25)	7.5 (4.5)	.41	0.5 (0.1-2.4)
None to some suffering	50 (75)	8.5 (4.5)		1 [Reference]
Overall suffering (n = 80)				
A lot/a great deal	12 (15)	10.6 (5.2)	.045	5.4 (1.4-21.9)
Not at all/a little/somewhat	68 (85)	7.4 (4.1)		1 [Reference]
Financial hardship				
Perceived financial hardship due to child's illness (n = 79)				
Great	24 (30)	10.1 (5.3)	.009	5.0 (1.4-17.6)
No/little/moderate	55 (70)	6.9 (3.7)		1 [Reference]

Abbreviations: K6, Kessler-6 Psychological Distress Scale; OR, odds ratio; PD, psychological distress; SPD, serious psychological distress.

^aSerious psychological distress defined as a K6 score of 13 or more.

^bP value for univariate linear regression models comparing PD between groups (measured as mean K6 scores).

^cUnadjusted OR and 95% CI for univariate logistic regression models comparing PD between groups (measured as likelihood of SPD).

^dNo parents met criteria for SPD; 95% CI not calculated.

Our results also demonstrate that goals of therapy further define PD and that the relationship between these goals and prognostic understanding is key. For example, we found that when prognostic understanding is aligned with concrete treatment goals parents experience lower levels of distress, even if parents understand the child's illness to be incurable. These findings are akin to theories of cognitive dissonance, which explain the psychological effects of conflicting beliefs and behaviors (ie, understanding of prognosis and goals of therapy).³⁷ Studies have shown that the dissonant state may be resolved by changing individual beliefs; however, cognitive dissonance may have lasting, psychologically detrimental effects if the incon-

sistencies endure.³⁸ Because a cancer diagnosis represents the potential death of a child and challenges a parent's basic assumptions, the level of cognitive dissonance, and consequent PD, may be greater in this population.³⁹ These results support the notion that early integration of palliative care strategies as a means of facilitating the alignment of parents' prognostic understanding with concrete goals of care may ease their distress.

Parent perception of child suffering was also related to their experience of PD. Perceived child psychological, but not somatic, symptoms were associated with parental distress, regardless of perceived prognosis. Parents may have greater emotional struggles when they feel that their child

Table 3. Parent Perceptions of Child's Illness Associated with Increasing Psychological Distress, Multivariate Analysis^a

	Increased Parental Distress Scores ^b Average Increased K6 Score β (95% CI)
Prognostic understanding aligned with concrete goals of care	-4.0 (-2.1 to -5.8)
Overall child suffering "a lot" or "a great deal"	3.0 (0.7 to 5.3)
"Great" financial hardship due to child's illness	3.1 (1.0 to 5.2)

Abbreviation: K6, Kessler-6 Psychological Distress Scale.

^an = 70.

^bMultivariate linear regression model showing average change in K6 score, independent of other listed factors.

is unhappy or afraid. They may feel less knowledgeable or more helpless in the face of their child's emotional symptoms than they would with other cancer-related somatic complaints and may feel more responsible for addressing psychological concerns. Likewise, a child's emotional suffering may more strongly impair parental coping. Family functioning and parenting styles also may relate to the development of parent distress. Others have described a relationship between child emotional distress and parent distress⁴⁰ but have been challenged to determine causality.⁴¹ Indeed, parents with their own histories of PD may be more likely to report child psychological symptoms. The nature of our study design precludes inference regarding the direction of any influential relationship between parent mental health and child suffering.

Our findings suggest that younger parents, or those of younger children, tend to have higher levels of PD. This may reflect lower levels of maturity or fewer economic and social resources. Unlike prior studies, however, we did not find a relationship between income and parent distress. Rather, perceived financial hardship was an important factor of parent distress. We have previously described financial hardship among parents of children with cancer and have shown that poor families have greater hardship.¹⁷ The present findings suggest that families who perceive economic stressors are more likely to be psychologically distressed, regardless of their baseline financial resources.

The study has several limitations. First, while our data were collected during the child's illness and not retrospectively, they are not longitudinal. We cannot infer trends in parental distress over time nor can we estimate potential causality in the described associations. For example, there may be a process of adjustment where parents first understand their child has no likely chance for cure and later realign their goals; however, we found no association between duration of progressive disease and PD. Second, since we do not report concurrent child-reported distress or physician opinions of prognosis, we cannot validate parent-reported child experience or perspectives of cure likelihood. Future studies should focus on concordance of perceptions among patients, parents, and physicians and how this relates to parent PD.

In addition, while we had a comparatively large sample in relation to other studies conducted among parents, the

absolute number of subjects was not large, and so, even moderate relationships could not be assessed reliably. For example, despite notable trends, we could not confirm with confidence that child or parent age or parent sex was associated with parent PD. Though our study is multi-institutional, the characteristics of our sample are somewhat narrowly distributed to white women with annual incomes above the poverty level and mainly represent families cared for at large tertiary-level cancer centers, and so, our findings may not be generalizable to families of varying backgrounds cared for in other settings. Finally, the K6 instrument is an excellent screening tool that has been used in parents of other ill children; however, the depth of information regarding PD is limited. To gain a better understanding about parental PD and even about the use of K6 as a screening tool in this population, a study using more specific PD tools would provide valuable data.

CONCLUSIONS

This is one of the first studies to describe the experience of distress in parents of children with advanced cancer. Distress levels in this population are high and are associated with parent experiences during their child's cancer care. Importantly, our findings suggest ways in which providers may ease parental PD by exploring their views on prognosis and treatment goals, perceived child suffering, and economic hardship. Future research should focus on family interventions aimed at easing distress among parents of children with advanced cancer.

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