JOURNAL OF PALLIATIVE MEDICINE
Volume 15, Number 3, 2012

DOI: 10.1089/jpm.2011.0304

Research Participation Experiences of Parents of Children with Cancer Who Were Asked about their Child's Prognosis

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Abstract

Background: In questionnaire-based research, human subject protection committees must assess the emotional impact of the study on participants. Without clear data about the risks and benefits of participating in such studies, however, review board members must use personal judgment to assess emotional harm.
Objective: To examine experiences of distress and value of participation in a study of prognosis communication among parents of children with cancer, and to identify factors associated with predominantly distressing research experiences.
Methods: We surveyed 194 parents of children with cancer (overall response rate, 70%), treated at the Dana-Farber Cancer Institute and Children's Hospital, Boston, Mass, in the first year after the child's cancer diagnosis. The survey focused on the child's prognosis and parent-physician communication; at the end, we asked parents how distressing and how useful completing the survey had been to them personally.
Results: Only 1% of parents found research participation to be "very" distressing. The majority of parents were "not at all" distressed by participating (62%), and most reported that the questionnaire was at least "a little" useful to them personally (69%). Overall, 18% of parents gave higher ratings for distress than for utility. Parents were more likely to experience research participation as predominantly distressing when they found prognostic information to be upsetting (odds ratio [OR] 5.38, $p = 0.005$).
Conclusion: Most participating parents were able to respond to questions about their child's prognosis with little or no distress. Even when distress was present, it was often accompanied by a perception that participating was of value.

Introduction

Human subject protection committees are charged with gauging and mitigating potential harm to research participants, including those who participate in psychosocial research, where risks may be psychological in nature.[1] However, assessing emotional harm to the participant often takes place in the absence of clear evidence. Review boards tend to be more conservative in judging harm than researchers and even participants,[2] who offer a more nuanced perspective. Although participants may experience distress and anxiety due to studies about medical experiences,[3–5] research participation is also perceived as therapeutic or empowering[6–11] and an opportunity to help others in the same situation.[7,12,13] Even participants who express feelings of distress often state that they would take part in a study again.[7,14,15]

Despite previous literature on research participation experiences, data are limited on which participants are most likely to experience distress. In addition, most literature focuses on risks and benefits of study participation separately, even though the combination of the two factors may matter most to individual participants. We conducted a questionnaire-based study about prognosis communication among parents of children with cancer.[16,17] In doing so, we touched on a subject that even many physicians prefer not to address due to concerns about causing parents distress.[18–21] We therefore evaluated parents' experiences as study participants.

Methods

We surveyed parents and physicians of children in the first year of cancer treatment at the Dana-Farber Cancer Institute

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Accepted October 19, 2011.

and Children's Hospital, Boston, between April 2004 and September 2005. Details of the study and survey development have been previously described.[16,17] One parent per family was eligible to participate; parents could decide which parent wished to do so. Of 276 eligible parents, 194 (70%) completed the survey, and 20 of 21 physicians completed surveys, corresponding to 193 of 194 parents.

The parent questionnaire focused on experiences with communication with the physician and expectations for the child's likelihood of cure. Communication quality was evaluated[22] using items on physician sensitivity, clarity, listening, and time for questions. Information quality comprised parent reports on the quality of information they had received about diagnosis, treatment, prognosis, the child's future, causation, and how treatment is working. Parents were asked "how upsetting" it is "to know information about your child's prognosis" and how they feel "about the amount of information you know about your child's prognosis." The questionnaire assessed parents' coping styles,[23] sense of peace,[24] relationship to the child, gender, age, education, marital status, and race/ethnicity. At the end of the questionnaire we asked, "How distressing did you find the experience of completing this questionnaire?" and "How useful to you personally did you find the experience of completing this questionnaire?" Response options were "not at all," "a little," "somewhat," and "very."

The physician questionnaire asked "How likely you think it is that this child will be cured of cancer," with response categories of: "extremely likely (more than 90% chance of cure)"; "very likely (75%–90%)"; "moderately likely (50%–74%)"; "somewhat likely (25%–49%)"; "unlikely (10%–24%)"; "very unlikely (less than 10%)."[25,26] The physician questionnaire also measured type of cancer, treatment received, and time since diagnosis.

The Institutional Review Board of the Dana-Farber Cancer Institute approved this study.

Statistical methods

Responses for questions about distress and utility were compared. Parents who gave higher ratings for distress than utility were defined as parents for whom the distress associated with participating in the study outweighed its utility, also termed parents whose research experience was predominantly distressing. This served as the primary outcome variable. We evaluated relationships between a predominantly distressing research participation experience and attributes of the parent, child, disease, and communication process.

For communication and information quality scales, responses were summed, and sums were dichotomized at the sample median for analysis. Other variables were dichotomized as specified in tables and results, with the exception of physician-rated likelihood of cure, which was treated as an ordinal scale.

Bivariable and multivariable relationships were described using logistic regression with generalized estimating equations to account for clustering of patients within physicians. Multivariable models were devised using a backwards selection technique; we included parent gender, race, educational level, diagnosis, and physician-rated prognosis, regardless of the significance of their coefficients. A sensitivity analysis using any parental distress, without respect to utility, as the outcome provided similar results. Analyses were conducted using the SAS Statistical Package (SAS Institute, Inc., Cary, NC).

Table 1. Parent and Patient Characteristics

Characteristics	*Values* (n = *194*)
Parent	
Female, *n* (%)	153 (79)
Age 30 or older, *n* (%)	172 (89)
College graduate, *n* (%)	115 (60)
Married or living as married, *n* (%)	158 (82)
Race/ethnicity (*n* = 190)	
White, non-Hispanic (%)	85
Black, non-Hispanic (%)	4
Hispanic (%)	7
Asian (%)	2
Other (%)	2
Child	
Female, *n* (%)	92 (49)
Age at diagnosis, median years (range)	6.6 (0.2–17.9)
Days since diagnosis, median (range)	105 (30–552)
Cancer diagnosis (*n* = 194)	
Hematologic malignancy (%)	56
Brain tumor (%)	23
Other solid tumor (%)	22
Received stem cell transplant, *n* (%)	28 (15)

Results

Parents completed the study questionnaire a median of 105 days after the child's cancer diagnosis (Table 1). Most parents reported that they were "not at all" distressed by the questionnaire (62%, Table 2), with the remaining parents reporting feeling "a little" (29%), "somewhat" (8%), or "very" (1%) distressed by their participation. The majority of parents (69%) also found the questionnaire to be at least a little useful to them personally (39% "a little," 25% "somewhat," 5% "very"), with 31% of parents considering participation to be not at all useful. When ratings for distress and utility were compared directly, 18% of parents gave higher ratings for distress than for utility and therefore met our definition of

Table 2. Parent Reports of Distress and Utility of Research Participation

How distressing was the experience of completing this questionnaire?	*How useful to you personally was the experience of completing this questionnaire?*			
	Not at all	*A little*	*Somewhat*	*Very*
Not at all	n = 26	50	31	8
	14%	27%	17%	4%
A little	23	20	10	2
	12%	11%	5%	1%
Somewhat	7	2	5	0
	4%	1%	3%	0%
Very	2	0	0	0
	1%	0%	0%	0%

Parents for whom distress outweighed utility (rating for distress was greater than rating for usefulness): *n* = 34, 18.3%.
Parents for whom distress and utility matched: *n* = 51, 27.4%.
Parents for whom utility outweighed distress: *n* = 101, 54.3%.

Table 3. Factors Associated with a Research Experience that was Predominantly Distressing (Rating for Distress Outweighed Rating for Utility): Bivariate Analyses

	OR (95% CI)	p *value*
Parent attributes		
Parent education, per category of increasing education	1.20 (0.80, 1.79)	0.38
Parent race non-white	1.25 (0.60, 2.60)	0.56
Parent gender female	2.18 (0.65, 7.29)	0.21
Parent age ≥30 years	2.15 (0.87, 5.29)	0.10
Parent coping and adjustment		
Parental coping: active	0.37 (0.12, 0.90)	0.08
Parent has peace of mind about child's illness	0.08 (0.02, 0.37)	0.001
Child and disease attributes		
Child's diagnosis was more than 100 days prior to participation	0.44 (0.23, 0.86)	0.02
Physician-rated prognosis, per category of worsening prognosis	1.16 (0.96, 1.40)	0.13
Communication attributes		
Parent rates physician communication as high quality	0.44 (0.20, 1.00)	0.05
Parent rates information received as high quality	0.56 (0.29, 1.06)	0.07
Parent considers information about prognosis "extremely" or "very" upsetting	5.67 (2.19, 14.69)	0.0004
Parent wishes he or she had additional information about the child's prognosis	2.14 (1.03, 4.47)	0.04

OR, odds ratio; CI, confidence interval.

having had a research experience that was predominantly distressing. Remaining parents either chose the same rating for distress and utility (27%) or provided ratings for utility that were greater than their ratings for distress (54%).

We evaluated factors associated with a parental research experience that was predominantly distressing (Tables 3 and 4). In a multivariable analysis, parents were less likely to have a distressing research experience when they participated in the study more than 100 days after the child's diagnosis (odds ratio [OR] 0.45, $p=0.05$). Parents who had a sense of peace of mind about the child's illness (OR 0.11, $p=0.0001$) were also less likely to experience the study as predominantly distressing. A distressing experience was more likely, however, among parents who found prognostic information upsetting (OR 5.38, $p=0.005$) and who wanted additional information about the child's prognosis (OR 2.06, $p=0.04$) beyond what they had already received from the physician.

Discussion

We administered a questionnaire to parents about a delicate and painful issue—the possibility that their child could die of cancer. We were concerned that, by raising this difficult subject, we could cause emotional harm to parents who were already under great stress, and as a result we evaluated parents' experiences with participation. Consistent with previous research, however, distress levels were limited[11,14,15,27,28]; only 1% of participating parents found the experience to be very distressing, and most reported no distress at all. Notably, parents indicated a dual nature to their experiences with the survey; even when parents experienced distress, they also often found personal value in participating in the study.

Not surprisingly, parents' experiences tended to be predominantly distressing when they considered prognostic information upsetting, when they had little peace of mind about the child's illness, and when their children were newly diagnosed. Each of these factors might understandably identify parents with heightened emotions around the child's illness. Unexpectedly, however, parents were also more likely to feel distressed if they wanted additional information about prognosis beyond what they had already received from the physician. This finding is consistent with previous work that suggests that uncertainty often underlies distress, and that information, even when difficult, can allay one's worst fears.[29,30] This situation may best be remedied, not by altering the nature of the research, but by helping parents to access the information they need.

Parents whose children had been diagnosed more recently tended to report greater distress from participation. This raises the question as to whether patterns of distress might have differed if we had recruited parents at the time of the child's diagnosis. Researchers who wish to mitigate distress may wish to consider recruiting parents for similar studies after the most acutely stressful period has passed.

Table 4. Factors Associated with a Research Experience that was Predominantly Distressing (Rating for Distress Outweighed Rating for Utility): Multivariable Model[a]

	OR (95% CI)	p *value*
Parent has peace of mind about child's illness	0.11 (0.04, 0.34)	0.0001
Parent considers information about prognosis "extremely" or "very" upsetting	5.38 (1.66, 17.06)	0.005
Parent wishes he or she had additional information about the child's prognosis	2.06 (1.04, 4.07)	0.04
Child's diagnosis was more than 100 days prior to participation	0.45 (0.20, 1.01)	0.05

[a]Adjusted for parent gender, race, educational level, diagnosis, physician-rated prognosis, and for clustering by physician.
OR, odds ratio; CI confidence interval.

Some limitations are worth considering. Although 70% of eligible parents participated in this study, those who did not may have been those most distressed by participation. In addition, we only interviewed one parent per family. However, the lack of significant distress among participating parents suggests that parents were able to make reasonable decisions about their ability to participate in the study. Self-selection appears to have been effective in identifying parents who could address these issues without major resultant distress.

This was a small study, involving just fewer than 200 parents in total and only 34 parents who found the research experience to be predominantly distressing. Although low rates of distress are reassuring, our interest in identifying parents most likely to be distressed is limited by the small sample size. Future research would be useful to confirm these results and evaluate predictors of distress in more depth.

Finally, we compared parental ratings of distress and utility. However, participants were not asked to compare the two issues directly. We do not know if parents would agree with our assessment that utility often outweighed distress. However, even without a direct comparison, our data suggest that even parents who find a questionnaire distressing may also find some value in participation.

Those charged with the protection of human subjects find themselves faced with difficult decisions about whether studies cause undue risks, including psychological distress, to participants. However, parents may find value in participating in research, and parents may experience participation in both helpful and difficult ways, not only one or the other. Allowing parents to judge their ability to participate appears to be effective in identifying a population of participants who experience limited distress. Distress may also be mitigated by approaching subjects after the period of acute stress has passed, and for parents whose distress is related to a desire for more information, considering ways to accomplish this outside of the research setting. Internal review committees may wish to consider the breadth of parental experiences as they evaluate the ethics of psychosocial research about difficult topics.

Acknowledgments

Dr. Mack was supported by a fellowship from the Agency for Healthcare Research and Quality (T32 HS00063), an American Society of Clinical Oncology Young Investigator Award, and a fellowship from the Glaser Pediatric Research Network.

Dr. Mack had full access to all of the data in the study and takes full responsibility for the integrity of the data and the accuracy of the data analysis.

We are indebted to the parents and physicians who participated in the study, and to Amy Lynch, MPH, for assistance in enrolling participants.

Author Disclosure Statement

No competing financial interests exist.

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