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What's Missing in Missing Data? Omissions in Survey Responses among Parents of Children with Advanced Cancer

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Abstract

Background: Missing data is a common phenomenon with survey-based research; patterns of missing data may elucidate why participants decline to answer certain questions.
Objective: To describe patterns of missing data in the Pediatric Quality of Life and Evaluation of Symptoms Technology (PediQUEST) study, and highlight challenges in asking sensitive research questions.
Design: Cross-sectional, survey-based study embedded within a randomized controlled trial.
Setting: Three large children's hospitals: Dana-Farber/Boston Children's Cancer and Blood Disorders Center (DF/BCCDC); Children's Hospital of Philadelphia (CHOP); and Seattle Children's Hospital (SCH).
Measurements: At the time of their child's enrollment, parents completed the Survey about Caring for Children with Cancer (SCCC), including demographics, perceptions of prognosis, treatment goals, quality of life, and psychological distress.
Results: Eighty-six of 104 parents completed surveys (83% response). The proportion of missing data varied by question type. While 14 parents (16%) left demographic fields blank, over half ($n=48$; 56%) declined to answer at least one question about their child's prognosis, especially life expectancy. The presence of missing data was unrelated to the child's diagnosis, time from progression, time to death, or parent distress ($p>0.3$ for each). Written explanations in survey margins suggested that addressing a child's life expectancy is particularly challenging for parents.
Conclusions and Relevance: Parents of children with cancer commonly refrain from answering questions about their child's prognosis, however, they may be more likely to address general cure likelihood than explicit life expectancy. Understanding acceptability of sensitive questions in survey-based research will foster higher quality palliative care research.

Introduction

Parents of children with advanced cancer have high levels of distress,[1] not only because cancer poses a threat to their child's life, but also because they may feel a sense of failed obligation to protect their child.[2] Parent–physician concordance regarding prognosis is generally poor[3,4]; this lack of agreement may affect patients' quality of life and quality of death.[4] Understanding parents' perspectives about their child's prognosis and the barriers they face in articulating them may lead to improved concordance, and in turn, improved outcomes.

The Pediatric Quality of Life and Evaluation of Symptoms Technology (PediQUEST) Study was a randomized controlled

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trial of a supportive care intervention in children with advanced cancer.[5] Additional aims were to evaluate factors of parent–provider concordance regarding prognostic awareness and goals of care. However, we found a high proportion of missing data when parents were asked to describe their child's prognosis. The objectives of this brief report are to describe patterns of our missing data and highlight challenges in asking sensitive questions while conducting research among parents of children with serious illness.

Methods

Eligible patients were at least 2 years old, had at least a 2-week history of progressive, recurrent, or refractory cancer, and had received cancer therapy at Dana-Farber/Boston Children's Cancer and Blood Disorders Center (DF/BCCDC); Children's Hospital of Philadelphia (CHOP); or Seattle Children's Hospital (SCH). Their parents/legal guardians had written command of the English language. All parents/patients over 17 years of age provided informed consent for participation. The Institutional Review Board at each participating institution approved the study.

Consecutive eligible families were approached and 104 children enrolled between December 2004 and June 2009. At the time of enrollment, all parents were invited to complete the Survey about Caring for Children with Cancer (SCCC). This comprehensive paper-and-pencil, self-administered survey evaluates perceptions of the child's illness. Development has been previously described[1,4,6] using focus groups and interviews with parents of children with advanced cancer, and then pretested for content, response-burden, and cognitive validity. Domains evaluated in the SCCC include:

1) Sociodemographics
2) Prognosis and treatment goals: 32 items query parent perceptions of prognosis, life expectancy, and treatment goals at the time of cancer diagnosis and upon enrollment. Regarding prognosis, parents are asked:
   (a) ''How likely it is that your child will be cured?'' (response options: ''very likely''/''likely''/''unlikely''/''very unlikely''); and,
   (b) ''What is your understanding of how long your child will live?'' (response options: ''days to weeks''/''weeks to months''/''months to years''/''several years''/''normal life expectancy''). Parents are also asked to select a primary treatment goal: ''to cure your child's cancer,'' ''to be able to keep hoping,'' ''to make sure you have done everything,'' ''to extend your child's life as long as possible without expecting a cure,'' ''to lessen suffering,'' and ''to help cancer research.''.
3) Child quality of life: 6 items explore parent perceptions of the child's emotional and physical health-related quality of life in the preceding month.
4) Parent psychological distress: Measured with the Kessler 6-item General Psychological Distress Scale (K6).[7]

Individual responses were defined as ''missing'' when parents left an item blank (no response provided) or ambiguous (more than one response provided). Several parents provided written explanations when omitting a response; these were transcribed verbatim for further analysis. We quantified the prevalence of ''missingness'' by SCCC domain and analyzed the qualitative answers provided.

Enrolled patient's clinical information was extracted from medical records and included cancer type, age, date of diagnosis, dates of first and subsequent disease progression, and, where relevant, date of death. We categorized patients with ''recent'' progressive disease if they had documented progression within 100 days prior to enrollment. Patients who died within 100 days after enrollment were categorized as having ''early'' death, and all who died within a 3-year period of follow-up were coded as having ''eventual'' death. Descriptive statistics were used to characterize all variables. In order to compare parents with and without missing responses, we used χ^2 and Fisher's exact tests to evaluate associations with child cancer type, progressive disease, and death, and Student's t tests with unequal variance to evaluate the association with average parent distress scores. All analyses were performed with Stata 12.1 statistical software (StataCorp, College Station, TX).

Results

Eighty-six of 104 enrolled parents completed the SCCC (83% response). Parent and child demographics were similar among those who did and did not complete the survey. Most were white mothers with at least a college education and annual household incomes above $75,000 (Table 1). Most parents left at least one item of the SCCC unanswered (Table 2). Fourteen (16%) left individual demographic items blank, most commonly their annual income. By comparison, over half did not complete all items regarding child prognosis and treatment goals. Parents were less likely to respond to questions about life expectancy than overall cure likelihood. Specifically, 15 (17%) and 26 (30%) parents did not provide an answer to items regarding life expectancy at diagnosis or ''now,'' respectively. Eleven (13%) left both questions blank. Fewer parents, 4 (5%) and 12 (14%), respectively, declined to answer questions of cure likelihood, with only one parent leaving both items blank. Most parents reported their goals of therapy at the time of diagnoses, but 15 (17%) did not provide current goals. The majority ($n = 81$, 94%) completed the psychological distress subscale.

There were no differences in average psychological distress scores among those who did and did not respond to prognosis questions ($p = 0.9$). Likewise, there were no strong associations between missing cure-likelihood, life expectancy, and treatment goal answers with either child cancer type ($p = 0.3$–0.9), recent progressive disease ($p = 0.3$–0.6), early ($p = 0.1$–0.3), or eventual death ($p = 0.2$–0.9).

Parents' qualitative comments regarding their child's current life expectancy suggested that this item was perceived distinctly from others in the questionnaire, not only because they provided explanation for their lack of item-response, but also because their comments reflected relative uncertainty (Table 3).

Discussion

Our objectives were to underscore what we can learn from patterns of missing data. We found that parents of children with advanced cancer are less likely to respond to research questions regarding their child's prognosis, particularly current life expectancy, compared with other types of questions, including those regarding race, income, or quality of life.

Table 1. Parent and Child Demographic Characteristics ($N=86$)

Parents	n *(%)*
Gender	
Female	74 (86)
Male	12 (14)
Ethnicity	*(n=85)*
Non-Hispanic/Latino	77 (93)
Hispanic/Latino	6 (7)
Race	*(n=82)*
White	76 (93)
African American	3 (4)
Asian	1 (1)
Other[a]	2 (2)
Education	
High school or less	28 (33)
College	45 (52)
Graduate school or professional degree	13 (15)
Annual family income (before taxes)	*(n=78)*
<$25,000	11 (14)
$25,000–$49,999	11 (14)
$50,000–$74,999	16 (21)
≥$75,000	40 (51)
Mean age (SD): 43.6 years (7.5)	
Children	n *(%)*
Gender	
Female	46 (53)
Male	40 (47)
Diagnosis	
Hematologic malignancy	28 (33)
Non-CNS solid tumor	49 (57)
CNS tumor	9 (10)
Progressive disease within 100 days prior to enrollment	29 (34)
''Early'' death (within 100 days after enrollment)	8 (9)
''Eventual'' death (at any time following study enrollment)	40 (47)
Mean age (SD): 12.1 years (5.8)	

[a]Write-in race responses: ''Hispanic,'' and ''Peruvian.''

CNS, central nervous system; IQR, interquartile range; SD, standard deviation.

Those who leave questions blank do not appear to be more (or less) distressed than other parents, nor is their lack of response related to their child's overall condition. Rather, written comments suggest an unwillingness or inability to explicitly document an expected duration of life.

These results are limited in that they represent a cross-sectional sample of parents with narrow diversity (mostly white mothers with higher education). We were unable to assess how racial and cultural perspectives might play into willingness to articulate a child's prognosis. Finally, we only determined ''missingness'' within the sample of parents who returned their larger study surveys, and thus cannot generalize to the 17% of nonresponding parents.

Missing data in survey research is a common and expected phenomenon,[8] and there are established methods for handling item nonresponse.[9,10] Unfortunately, these methods are unable to address why individual survey items are omitted; instead, we are left to make reasonable speculations. For example, we did not identify an association between ''early'' patient death and missing parent response. This may be due to limited power, but reasons for missingness are likely more complex and subjective. Perhaps what parents publicly acknowledge is different from what they privately think. Perhaps formally accepting the possibility of death of one's child is akin to ''giving up'' and undermines parental identity.[11] Perhaps responding in writing (and committing) to research questions regarding prognosis is associated with superstition (''if I say it, will it come true'').

Most parents of children with cancer acknowledge that hearing their child's prognosis is a distressing but necessary element of their medical care.[12] While the majority of those who participate in research regarding prognosis report that it

Table 2. Prevalence of Missing Responses in Selected SCCC items ($N=86$)

	Total number of parents who left unanswered or ambiguous item response (%)[a]
Parent sociodemographics	14 (16)
Age	1 (1)
Gender	0 (0)
Ethnicity or race	5 (6)
Education	0 (0)
Income	8 (9)
Prognosis and treatment goals	48 (56)
Likelihood of cure (at diagnosis)	4 (5)
Likelihood of cure (currently)	12 (14)
Life expectancy (at diagnosis)	15 (17)
Life expectancy (currently)	26 (30)
Treatment goals (at diagnosis)	3 (3)
Treatment goals (currently)	15 (17)
Parent report of child quality of life	9 (11)
Psychological symptoms	7 (8)
Physical symptoms	3 (3)
Parent psychological distress	5 (6)

[a]Missing items defined as any item left blank or with ambiguous answer.

SCCC, Survey about Caring for Children with Cancer.

Table 3. Examples of Parent's Written Explanations for Leaving Formal Responses Blank

SCCC question	*Parent responses*
''What is your understanding now of how long your child will live?''	*''Don't know, day to day, I really don't get into this—What good does this do anyway?''*
	''No one can answer this question.''
	''50% cure rate if bone marrow was possible and without treatment maybe 1 year.''
	''Doctors cannot be sure…there is no way to know for sure. It depends on what we do next.''
	''Don't know yet.''

SCCC, Survey about Caring for Children with Cancer.

is at least ''a little'' useful to them, the odds that they will experience distress are over five times higher when the prognostic information is upsetting.[13] Furthermore, parents of children at the end of life want ''what is best for their child,''[14,15] but often request continued cancer-directed therapy even when there is no chance of cure.[16,17] They tend to acknowledge their child's impending death 100 days later than their health care providers.[4] Meanwhile, health care providers may struggle with what they perceive as unrealistic parent expectations[18] or futile treatments.

Understanding and resolving these conundrums is one of the objectives of pediatric palliative care research, and many studies rely on surveys to identify parent perspectives. We pretested the SCCC and found no evidence of parent response-burden or concern with individual items. Still, our findings suggest that parents selectively choose not to answer certain types of questions and that there are legitimate explanations for their omissions. Future qualitative investigations may focus on why certain data are commonly missing, and continued research may address which questions investigators should (and should not) ask. Doing so will help us better understand the parent experience in survey research, and incorporate methodological modifications to ensure higher quality research.

Conclusion

Parents of children with cancer commonly refrain from answering questions about their child's prognosis. Understanding the patterns of missing data will enable higher quality research which, in turn, may enable improved parent, patient, and whole-family care.

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Author Disclosure Statement

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