Impact of research participation on parents of seriously ill children*

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Abstract

Background: There is a paucity of research evidence to guide health care providers' practice in pediatric palliative care. At the same time, some clinicians and IRBs are reluctant to approve such studies because of concerns about further burdening families. Yet, there is some evidence that research participation can have positive effects for families.

Objective: To obtain parents' perceptions about their experience of participating in 1 of 2 research studies.

Design: Descriptive, quantitative survey.

Setting/Subjects: Caregivers of children with life-threatening conditions (n=323) who were caring for the child at home.

Measurements: Researcher-designed Impact of Participation questionnaire.

Results: Few differences between the 2 groups were found on Impact responses. Not a single parent reported regretting participating in their study and almost all (96.3%) reported that conducting research about family's experiences in pediatric palliative care had value. Just over three quarters (76.2%) did not find participation at all painful, and 73.7% reported that participation was about as painful as expected, with 23.2% reporting less painful. About half (50.5%) said that participation had at least some positive effect and only 3 parents reported any negative effect. An overwhelming majority (93.4%) would recommend participation to other parents in a similar situation.

Conclusions: Participation in research for families with children who have a life-threatening condition is not only acceptable to parents, but may in fact have a positive effect. Though clinicians and IRBs may be hesitant to fully support such research, it is clear that conducting research in the field of pediatric palliative care is important.

Introduction

Palliative care clinicians from all disciplines value evidence-informed practice. However, palliative care is a relatively new field and, particularly in pediatric palliative care, the scarcity of research upon which to base practice is frequently noted.[1-3] One reason for this lack of evidence may be the challenges to conducting research that, in turn, limit the development of a strong evidence base.[3,4-6] For example, the relatively small number of children with specific conditions often necessitates multi-site studies that are complex and time-consuming.[4] Similarly, patient vulnerability and the burden of research on participants are often cited as concerns about research related to palliative care.[7-10]

Yet, contrary to the frequently expressed concerns by clinicians and Institutional Review Boards (IRBs) that research will be too onerous or troubling for families in palliative situations, there is evidence that participation can have positive effects in altruistic and therapeutic realms.[9,11,12] In a critical synthesis of palliative care research, most participants in all studies experienced positive effects; only a small minority reported distress.[13] Reported benefits include a sense of helping and improving care for others, increased self-awareness, catharsis and healing, empowerment, and autonomy.[13] There also is some indication that writing or talking about traumatic experiences can reduce physical stress responses.[14,15] At the same time, it is recognized that many people find it at least somewhat painful to talk about the death of their child.[11]

In an extensive review of the literature on pediatric palliative care research, Rapoport[16] concluded that participation in research during the illness, and after a child's death, can be a positive experience for participants and that such research should continue. Studies with bereaved parents have highlighted parents' eagerness to share their child's illness and death story and to provide input to help other families.[14,17] While some parents report that discussions about

diagnosis and/or their child's death may be difficult,[11] the majority deny any negative impact or distress from participating in research[17,18]. Indeed, interviews have been reported as therapeutic and a positive experience.[14]

Another concern noted about palliative care research is that it takes time that could be spent in other ways, such as being with family.[19] However, given the potential benefits of participating in research and the practice of family-centred care that is prevalent in pediatrics, it seems somewhat paradoxical that clinicians and IRBs might prevent families from choosing how they wish to spend their time. If this population is consistently denied access to research then not only may the potential benefits of such research be lost, but the autonomy of patients and families may be subverted.[7,8]

Undoubtedly, parents caring for a child with a life-threatening condition (LTC) have a significant burden of care,[20] and they deal with ambiguity about treatments, trajectories, and outcomes.[21] Often the parent's role in caregiving intensifies as the child's condition progresses and the need to navigate support systems for their child increases. Parents' needs and distress may be high in palliative situations and so interventions to address those needs and reduce distress are warranted.[19] Developing such interventions necessitates undertaking research with families to provide a suitable evidence base.[22] But given the demands on these parents, there is often reluctance from clinicians and IRBs to perhaps further burden them with research participation.

There is a need to resolve this quandary; direction from parents might lead to a resolution. However, little is known about participants' perceptions regarding the burdens of research,[23] and parents' views around research participation when a child is receiving palliative care are unclear.[3] The purpose of this paper is to describe the impact of participating in a

research study for parents who were concurrently caring for a child with a LTC. We recognize that the terms 'life-limiting' and 'life-threatening' may be used interchangeably by clinicians and in the literature, but there is often controversy over which term should be used; we use 'life-threatening' within this paper.

Methods

Data on impact of participation were obtained from family caregivers in two separate longitudinal, multisite research projects concerning families where a child had a LTC.

Description of studies

The first study, *Stress and growth over time: Caregiving and bereaved parents of children with life-limiting illnesses* (PCG), was conducted with adult family members who were caring or had cared for a child with a LTC; more than one family member could participate. Participants were recruited through clinics and parent support organizations in Canada and the USA. Eligibility included caregivers whose child (0 to 19 years of age at baseline) had any of the physiologically diverse conditions that may pose a threat to the child's life, regardless of the variability in life expectancy or the possibility of cure. Eligibility, therefore, included such conditions as cancer, irreversible organ failure, cystic fibrosis, neurodegenerative diseases, genetic and metabolic disorders, anoxic brain injury, and severe cerebral palsy.

At their convenience, caregivers completed 3 sets of 16-page questionnaire packages, sent and returned by mail at 1-year intervals. Each package included demographic information, short answer questions about caring for their child, and the following scales: 12-item Meaning in Caregiving Scale;[24] 10-item Life Orientation Test - Revised (LOT-R)[25] to measure optimism; 6-item Self Esteem Scale;[26] 22-item Spiritual Involvement and Beliefs Scale - Revised (SIBS-R);[27]

20-item Center for Epidemiologic Studies Depression Scale (CES-D);[28] 4-item Caregiver Burden scale;[24] 21-item Posttraumatic Growth Inventory (PTGI).[29]

The second study, *Charting the Territory: Determining and documenting trajectories for families where a child has a life-threatening condition* (CTT), was a 5-year project involving children 0-19 years diagnosed with progressive neurological, metabolic, or chromosomal conditions; their siblings aged 7-18 years; and at least one, but both parents if possible. Families were followed for a minimum of 18 months and through bereavement if a child died during the study and the family agreed to continue. Recruitment occurred in clinics and hospitals across Canada (n=7) and in the USA (n=2). Eligibility required the child to have a progressive and incurable condition that manifests in neurological (CNS) impairment and is known/likely to have a genetic or metabolic cause. There could be no effective treatment available or treatment was failing, and there had to be a good probability that children with this condition would die before their 20s.

Data collection included monthly parental assessment of the child's symptoms and an annual functional assessment; and completion of established instruments every 6 months for siblings and their parents, including from 6 months after bereavement if appropriate. Similar to PCG, parents in CTT completed the SIBS-R,[27] CES-D,[28] Caregiver Burden,[24] and PTGI.[29] Additional questionnaires included: 20-item Family Adaptability and Cohesion Evaluation Scale;[30] 6-item Norton's Quality of Marriage Index;[31] 12-item SF-12v2™ Health Survey;[32] 20-item State-Trait Anxiety Inventory (State)[33]; 10-item Perceived Stress Scale[34]; 5-item researcher-adapted Grief questionnaire.

Both studies also included an Impact of Participation questionnaire designed within our team for use in our research projects. This questionnaire was completed at Time 3 for both

studies, i.e., as parents completed their third questionnaire package, which was one year post-baseline for CTT and at the final time point for PCG (Table 1). Questions related to perceived value of the research, reasons for participating, expectations about and actual painfulness of participation, and positive and negative effects of participating. Though both studies included bereaved parents, this paper reports on results from non-bereaved participants only, because the low number of bereaved parents was insufficient for a comparison analysis.

Both projects received approval from the IRBs at participating institutions; parents provided written consent for CTT and return of the questionnaires provided assumed consent for PCG.

Analyses were conducted using IBM® SPSS® Version 20; *P* values <.05, two-sided tests, were considered to be statistically significant. Effect sizes were calculated as Cohen's *d* for continuous variables and *Phi coefficient* for categorical variables. Descriptive statistics were used to summarize the demographics and Impact results. Two-sample t-tests and χ^2 tests were used to compare: CTT and PCG on demographics and Impact; responders and non-responders on demographics; and child's condition (PCG only), time since diagnosis, and age on Impact for both male and female caregivers. Bonferroni corrections were made for the comparisons between child and Impact.

Results

Sample characteristics

In PCG, 123 caregivers responded from 104 families; CTT included 200 parents from 138 families, 8 of whom had 2 affected children. Some differences between PCG and CTT on parental and child demographics were evident. For example, PCG parents were older than those in CTT (*P* = <.001) and the male/female ratio was different, with the percentage of males higher

in CTT (P = .004) (Table 2); children in CTT were younger than those in PCG (P = <.001) and had been diagnosed more recently (P = <.001) (Table 3). No statistically significant differences were found between those who completed the Impact questionnaire, the responders, and those who did not, the non-responders, based on parental (Table 2) or child demographic variables (Table 3).

Impact of participation

Most responses were similar between CTT and PCG parents (Table 4). The vast majority of participants (96.3%, n = 310/322) thought that conducting research about their families' experiences had at least some value. A total of 261 out of 322 parents (81.1%) stated that it was very valuable to conduct research about their affected child's symptoms. Reasons for participating in the research study varied, but the most frequent were for altruistic (to help others; 96.3%, n = 311/323) or therapeutic reasons (to talk about their experiences; 36.8%, n = 119/323). Slightly over three quarters (76.2%, n = 246/323) did not find participation at all painful, and most reported that participation was about as painful as they expected (73.7%, n = 235/319), with 23.2% (n = 74/319) reporting participation being less painful than expected. About half (50.5%, n = 163/323) said that the study had at least some positive effect on them, and almost all the rest (48.6%, n = 157/323) reported neither a positive or negative effect. Only three parents (0.9%) experienced any negative effect. Not a single one of 323 parents reported regretting participating in the research, and 93.4% (n = 300/321) would recommend participation in a similar study to another parent in a similar situation.

Comparisons between the Impact results from male and female responders with the selected child characteristics of condition (PCG only), time since diagnosis, and age were not statistically significant (Table 5).

Discussion

Our findings closely mirror the literature related to research in palliative care.[9,11-13,17,18,35] Similar to parents of children with cancer[36] and to results from a synthesis of palliative care research,[13] the most frequently reported reason for parents in this study to participate in research was to help others. It may be that participation in research helps parents make meaning of their circumstances. Dyregrov[11] found that parents who were bereaved as a result of suicide, SIDS, or accidents felt that interviews facilitated meaning reconstruction and awareness of the bereavement process, even though it was painful to talk about the loss. This meaning making, in turn, may help facilitate growth,[37] growth that can occur in the midst of parents struggling with their loss.[38] Further research needs to be conducted on how clinicians can facilitate meaning making in a variety of ways when parents are caring for a child with a LTC and, thus, assist parents to experience growth despite the challenging circumstances.

Parents also reported that they chose to participate in the research so they could talk about their experiences, a finding that supports previous studies.[3,13] Further, parents may have reported high levels of positive responses to study participation because it helped them with their own physical responses.[14,15] Comparable to what is reported in the literature,[16-18,39,40] parents not only supported the research, but made it clear that participating in this type of research was important.

Although parenting a child with a LTC creates distress for parents and places high demands on their time,[20] the vast majority of parents reported that the research was valuable to them and, for about half, it had at least some positive effect. Some situations might increase distress or decrease positive aspects of research, e.g., repeated research involvement has been associated with less positive reports compared to the first participation.[35] However, participants

in this study had completed measures at three time points when they reported on the impact of participation and their assessments were mostly positive. It will be informative to track the impact of participation over time as the CTT project continues, and ideally through an extension for another 5-year period. Another indication of positive impact is willingness to continue to participate.[15] In addition to participants reporting few negative and some positive effects, both CTT and PCG showed good response and retention rates. Overall, the results reinforce the notion that palliative care research should be available to this population.

While some parents in palliative research report that discussions of their child's condition or death may be difficult, the majority deny any negative impact or distress caused by participation in this type of research.[5,11,12,14,17,18] Somewhat consistent with Dyregrov[11], some of the parents reported that it was 'a little painful' to participate in the current study. However, most did not find it painful at all. Further, for the majority, the pain they expected to feel because of participation in the study matched their expectations. Moreover, most of the others found participation to be less painful than expected. These findings suggest that parents of children with LTCs who choose to participate in studies consider their own tolerance and burden before they agree to participate. Therefore, the decision to participate or not, and to continue or not, may be made only after thoughtful reflection by parents.

Given that parents of children with LTCs seem to enter studies with some knowledge of the potential impact, it seems that rather than viewing these parents as vulnerable and in need of protection it is incumbent on clinicians and IRBs to recognize parents' autonomy and find ways to support them if they choose to participate in studies. In addition, clinicians and IRBs need to be aware that it is acceptable for a study in pediatric palliative care to potentially cause some psychosocial pain for parents; but it should be left up to the parents to decide for themselves if

the benefits of participating outweigh the risks and to determine if they are willing to tolerate possible pain in order to gain some benefits for themselves and for others. Familiarity with these findings among members of review boards may affect the way that protocols are perceived and reviewed. It seems clear from this study that while IRBs still need to consider the vulnerability of this population, they also need to respect the autonomy of parents and families and not prevent ethically-sound studies from being made available to families in palliative situations because they are worried that a study may be too onerous for these families.

Overall, this study addresses two research gaps by providing new information about research with families of children with LTCs who were mostly from Canada: (1) Research about the impact of participation has tended to focus on palliative patients, carers of adults, or bereaved carers;[13] our population was caregivers of children with LTCs; (2) Most palliative research emerges from the USA, U.K., or Australia;[13] our findings provide evidence from yet another country. This study supports previous contentions that research with these families can be reviewed using the same principles and protocols as other research studies[10,19] and provides guidance for clinicians and IRBs who are deciding about whether or not to support research in this area.

This study has some limitations, though the large sample size for this population lends credence to the results. All the participants self-selected and we do not know how parents who did not participate might react to research participation. However, psychosocial research in palliative care is generally conducted with volunteers; therefore, the self-selection in this study is reflective of most sample selection in palliative research. We do not know much about the experiences of people who withdrew from the study, so it is possible that some participants

withdrew because they found the process too painful. Further examination of the impact of participation is needed with this population.

We have little information about the longer-term effects of participation in research, though one could argue that given parents answered the Impact of Participation questionnaire after completing data collection three times over a period of at least 1 year then the results might hold true for longer-term effects. In CTT, parents will complete the questionnaire again at the end of the study, so at 2-4 years post-baseline. Comparisons of parents' reports over time will add to the evidence about longer-term effects of participation in research.

In some cases, parents might have been inclined to provide more positive responses about research than they actually felt; however, this inclination may have been ameliorated by providing a written, mailed questionnaire rather than face-to-face discussion. The consistency in responses across participants and between the two samples, even though there were some demographic differences, and the wide variety of conditions affecting the children suggest that bias towards answering positively is unlikely. Finally, though there was space for parents to provide some qualitative data, most questions were close-ended and so the tool provided a limited window into the experiences of parents who participate in research. Dyregrov[11] noted the potential for research to help reconstruct meaning and increase awareness of the bereavement process; further investigation of the experiences of caregiving parents might include qualitative interviews to explore the impact of research participation in these areas.

Conclusions

The results of this study suggest that not only is pediatric palliative care research valued by parents, but it also may have a positive impact on participants. Though some clinicians and IRBs may be hesitant to fully support such research, findings from parents is a strong indicator

that research in this field is important to families and these results should be consciously considered when evaluating study applications.

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TABLE 1. RETENTION RATES FOR NON-BEREAVED CAREGIVERS, SEPARATELY BY THE TWO STUDIES

Time-point	*CTT*[a]	*PCG*[b]
Time 1	(Baseline)	(Baseline)
Invited	385 families: 93 ineligible/declined unable to contact 34	367 contacted phoneline 341 eligible
Participated	390 parents in 258 families (67%)	273 usable questionnaires returned (80.1%)
Time 2	(6 months)	(1 year)
Packages sent	338 (227 families)	163
Packages returned	305/338 (203 families) (90.2%)	145/163 (89%)
Time 3 (Impact)	(1 year)	(2 years)
Packages sent	285 (190 families)	145
Packages returned	218/285 (148 families) (76.5%)	123/141 (87.2%)
Impact returned	200/285 parents (70.2%)	123/141 (87.2%)

[a] Families were recruited into the CTT study over a period of approximately 3 years. Not everyone had yet reached the Time 3 mark; therefore, only those non-bereaved caregivers who had done so by the time of analysis for this report were included in the numbers for Times 2 and 3. Also, it should be noted that sometimes families missed a time-point, e.g., because their child's condition needed their attention, they were too busy, or they were away on vacation, but then continued at the next one; numbers at T3 are lower than numbers of families continuing in the study.

[b] Caregivers in the PCG study were initially in a cross-sectional study that had 273 participants. When funding was obtained for a renewal, caregivers who had indicated an interest in other research (n = 268), but then excluding the 9 who had become bereaved, were contacted for the Times 2 and 3 follow-up study. From the total attempted contacts (n = 259), some could not be contacted (n = 88) and 11 others did not want to participate in the new study, e.g., because 'child now grown', 'not a good time', or no reason given. A further 3 new relatives joined the study. The final sample size for Time 2 non-bereaved, therefore, was 163. A total of 4 parents became bereaved between Times 2 and 3; % for participation at Time 3 reflects the number of non-bereaved rather than the total number of packages sent.

TABLE 2. DEMOGRAPHIC CHARACTERISTICS FOR PARENTS FROM BOTH CTT AND PCG, AND COMPARISONS BETWEEN THE TWO STUDIES

Characteristic	*CTT (N = 200)*	*PCG (N = 123)*	*P values*[a,b,c]
Gender			.002 [.17]
Female	133 (66.5)	101 (82.1)	(.26 [.06]/.75 [.02])
Male	67 (33.5)	22 (17.9)	
Age (years)	39.5 $\pm$ 8.1[d]	45.5 $\pm$ 6.7[d]	<.001 [.81]
20 to 35	59 (29.5)	11 (8.9)	(.36 [.09]/.52 [.08])
36 to 45	94 (47)	44 (35.8)	
46 to 55	40 (20)	57 (46.3)	
Over 55	7 (3.5)	11 (8.9)	
Marital status			.04 [.16]
Married/Living as married	181 (90.5)	98 (79.7)	(.81 [.05]/.24 [.14])
Divorced or separated	12 (6)	18 (14.6)	
Never married	6 (3)	6 (4.9)	
Widowed	1 (0.5)	1 (0.8)	
Employment status		(n=122)	.01 [.22]
Full-time	79 (39.5)	58 (47.5)	(.57 [.12]/.18 [.18])
Not employed	74 (37)	24 (19.7)	
Part-time	30 (15)	27 (22.1)	
Paid leave	9 (4.5)	2 (1.6)	
Self-employed	5 (2.5)	8 (6.6)	
Retired	2 (19)	2 (1.6)	
Unpaid leave	1 (0.5)	1 (0.8)	
Level of education completed			.51 [.04]
High school or less	40 (20)	20 (17.1)	(.8 [.01]/.05 [.12])
Post-secondary	160 (80)	102 (82.9)	
Family household income	(n=196)	(n=120)	.05 [.16]
Under $40,000	48 (24.5)	20 (16.7)	(.19 [.11]/.64 [.08])
$40,000 to < $80,000	74 (37.87)	37 (30.8)	
$80,000 to < 120,000	42 (21.4)	31 (25.8)	
$120,000 and over	32 (16.3)	32 (26.7)	

Numbers are frequency (%) unless otherwise indicated.

[a] χ^2 was used for categorical variables and two-sample t-test was used for the continuous variable to test by study (CTT vs. PCG).

[b] χ^2 was used for categorical variables and two-sample t-test was used for continuous variables to compare parents who completed the Impact questionnaire with those who did not. Comparisons between variables were made using baseline data from all parents at Time 1, and tests were run separately by study (CTT: n = 390 except for income where n = 377; PCG: n = 273 except for income where n = 264). *P* values are reported as (CTT/PCG).

[c] Effect size reported in [], Cohen's *d* for continuous variables, Phi coefficient for categorical variables.

[d] Plus-minus values are means ± SD.

TABLE 3. DEMOGRAPHIC CHARACTERISTICS FOR CHILDREN FROM BOTH CTT AND PCG, AND COMPARISONS BETWEEN THE TWO STUDIES

Characteristic	*CTT (N = 146)*	*PCG (N = 104)*	*P values*[a,b,c]
Gender			.93 [.01]
Female	75 (51.4)	54 (51.9)	(.77 [.02]/.89 [.01])
Male	71 (48.6)	50 (48.1)	
Age (at Time 3; years)	8.1 ± 5.3[d]	12.8 ± 5[d]	<.001 [.92]
0 to 5	60 (41.1)	5 (4.8)	(.96 [.01]/.64 [.06])
6 to 10	37 (25.3)	29 (27.9)	
11 to 15	31 (21.2)	35 (33.7)	
16 to 20	17 (11.6)	25 (24)	
Over 20	1 (0.7)	10 (9.6)	
Condition		(n=102)	
Group 1 (e.g., cystic fibrosis)	N/A[e]	10 (9.8)	(.16 [.23]/.81 [.06])
Group 2 (e.g., cancer)	N/A[e]	3 (2.9)	
Group 3 total number	146 (100)	84 (80.8)	
Neurodegenerative diseases/Severe epilepsy	31 (21.2)	3 (3.6)	
Multi-organ congenital abnormalities	28 (19.2)	3 (2.9)	
Severe neurological impairment (NYD)	28 (19.2)	1 (1.2)	
Lysosomal/Peroxisomal leukodystrophy	21 (14.4)	3 (3.6)	
Mitochondrial encephalomyopathy	16 (11)	0	
Structural CNS abnormalities	10 (6.8)	1 (1.2)	
Neuromuscular diseases	5 (3.3)	72 (85.7)	
Small molecules disorders	3 (2.1)	0	
Other metabolic diseases	3 (2.1)	1 (1.2)	
Other conditions (NOS)	1 (0.7)	0	
Group 4 (e.g., cerebral palsy)	N/A[d]	5(4.9)	
Time since diagnosis (at Time 3; years)	6.2 ± 4.6[d]	10.3 ± 4.8[d]	<.001 [.88]
0 to 5	54 (37)	15 (14.4)	(.84 [.03]/.39 [.12])
6 to 10	30 (20.5)	40 (38.5)	
11 to 15	15 (10.3)	26 (25)	
16 to 20	7 (4.8)	20 (19.2)	
Over 20	0	2 (1.9)	
Missing/not yet diagnosed	40 (27.4)	1 (1)	

Numbers are frequency (%) unless otherwise indicated.

[a] χ^2 was used for categorical variables and two-sample t-test was used for continuous variables to test by study (CTT vs. PCG), though children's condition was excluded because CTT only included one group.

[b] χ^2 was used for categorical variables and two-sample t-test was used for continuous variables to compare child characteristics for parents who completed the Impact questionnaire with those

who did not. Comparisons between variables were made using baseline data from all children at Time 1, and tests were run separately by study (CTT: n = 275 except for time since diagnosis where n = 206; PCG: n = 228 for gender, n = 235 for age, n = 232 for condition and for time since diagnosis). *P* values are reported as (CTT/PCG).

[c] Effect size reported in [], Cohen's *d* for continuous variables, Phi coefficient for categorical variables.

[d] Plus-minus values are means ± SD.

[e] Only children with group 3 conditions were included in CTT.

TABLE 4. IMPACT OF PARTICIPATION RESULTS

Impact Question	*CTT (N=200)*	*PCG (N=123)*	*P values*[a]
How valuable is it to conduct research about your affected child's symptoms?	(n=199)		.02 [.15]
Not valuable	9 (4.5)	2 (1.6)	
A little valuable	38 (19.1)	12 (9.8)	
Very valuable	152 (76.4)	109 (88.6)	
How valuable is it to conduct research about you and your family's experiences?	(n=199)		.77 [.13]
Not valuable	10 (5)	2 (1.6)	
A little valuable	46 (23.1)	20 (16.3)	
Very valuable	143 (71.9)	101 (82.1)	
Why did you agree to participate in this research study?[b]			
To help others in a similar situation	193 (96.5)	118 (95.9)	.79 [.02]
So I could talk about the experience I had	78 (39)	41 (33.3)	.3 [.06]
Other (e.g., because research/science in general is beneficial and crucial to overall advancements; to learn something about oneself)	27 (13.5)	12 (9.8)	.32 [.06]
Felt pressured by my family/friend	8 (4)	2 (1.6)	.23 [.07]
Felt pressured by the researcher	1 (0.5)	0 (0)	.43 [.04]
Felt pressured by the clinic physician	1 (0.5)	0 (0)	.43 [.04]
How painful was it to participate in this study?			.57 [.06]
Very painful	1 (0.5)	2 (1.6)	
A little painful	47 (23.5)	27 (22)	
Not painful at all	152 (76)	94 (76.4)	
Compared to what you might have been expecting, were the questions?	(n=198)	(n=121)	.54 [.06]
More painful than expected	5 (2.5)	5 (4.1)	
About what I expected	144 (72.7)	91 (75.2)	
Less painful than expected	49 (24.7)	25 (20.7)	
Overall would you say participating in this study had a positive or a negative effect on you?			.72 [.06]
Very positive	36 (18)	24 (19.5)	
A little positive	60 (30)	43 (35)	
Neither positive or negative	102 (51)	55 (44.7)	
A little negative	2 (1)	1 (0.8)	
Very negative	0 (0)	0 (0)	
Do you regret participating in this study	(n=199)		
No	199 (100)	123 (100)	
Would you recommend to another parent in a similar situation that they participate in a similar study?	(n=198)		.02 [.13]
Yes	180 (90.9)	120 (97.6)	
No	18 (9.1)	3 (2.4)	

Numbers are frequency (%).

[a] χ^2 was used for categorical variables to test by study (CTT vs. PCG), except for if parents regretted participating as no-one had responded 'Yes'; Phi coefficient calculated for effect size, reported in [].
[b] Numbers total more than sample size for relevant study group (CTT & PCG) because parent could select multiple options; % is out of total sample size for relevant study group.

TABLE 5. COMPARISONS BETWEEN SELECTED CHILD CHARACTERISTICS AND SELECTED PARENTAL IMPACT OF PARTICIPATION RESULTS BY PARENT GENDER

Impact Question	*P values Age*[a]	*P values Time*[b]	*P values Condition*[c]
How valuable is it to conduct research about your affected child's symptoms?	.76 [.2]/.89 [.19]; .52 [.33]/.84 [.43]	.22 [.29]/.86 [.23]; .64 [.3]/.58 [.45]	.98 [.1]/.49 [.26]
How valuable is it to conduct research about you and your family's experiences?	.52 [.24]/.77 [.22]; .8 [.26]/.23 [.68]	.04[d] [.36]/.99 [.16]; .81 [.26]/.32 [.55]	.61 [.14]/.89 [.1]
How painful was it to participate in this study?	.78 [.19]/.42 [.29]; .98 [.08]/69 [.5]	.88 [.17]/.4 [.32]; .43 [.24]/.33 [.55]	.43 [.25]/.68 [.19]
Compared to what you might have been expecting, were the questions?	.89 [.17]/.79 [.22]; .91 [.23]/.47 [.4]	.8 [.19]/.75 [.26]; .94 [.21]/.28 [.42]	.42 [.25]/.24 [.26]
Overall would you say participating in this study had a positive or a negative effect on you?	.21 [.35]/.18 [.4]; .63 [.38]/.6 [.53]	.72 [.26]/.17 [.45]; .96 [.27]/.74 [.39]	.71 [.25]/.76 [.16]
Would you recommend to another parent in a similar situation that they participate in a similar study?	.39 [.18]/.97 [.07]; .89 [.13]/N/A[e]	.58 [.15]/.97 [.1]; .14 [.33]/N/A[e]	.88 [.08]/N/A[e]

Phi coefficient calculated for effect size, reported in [].

[a] χ^2 was used to compare child's age in 5 categories with parental responses on selected Impact questions, by gender of parent. Tests were run separately by study and *P* values are reported as CTT female/PCG female; CTT male/PCG male.
[b] χ^2 was used to compare child's time since diagnosis in categories with parental responses on Impact questions, by gender of parent. Tests were run separately by study and *P* values are reported as CTT female/PCG female; CTT male/PCG male.
[c] χ^2 was used to compare child's conditions in 4 groups with parental responses on Impact questions, by gender of parent. Tests were run only for PCG because CTT included only 1 group and *P* values are reported as PCG female/PCG male.
[d] Not statistically significant at Bonferroni-corrected $P < .0008$.
[e] No males in PCG cohort responded 'No', so test could not be run.