

ORIGINAL ARTICLE

What factors are important to parents making decisions about neonatal research?

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Background: Although parents of neonates with congenital heart disease are often asked permission for their neonates to participate in research studies, little is known about the factors parents consider when making these decisions.

Objective: To determine the reasons for parents' decisions about participation in research studies.

Methods: Qualitative analysis of the unsolicited comments of 34 parents regarding reasons for agreeing or declining to participate in research studies. Parents' comments were offered spontaneously during interviews about clinical care decisions for neonates with congenital heart disease.

Results: Parents cited five types of reason for or against permitting their newborn to participate in research studies: societal benefit (n = 18), individual benefit for their infant (n = 16), risk of study participation (n = 10), perception that participation posed no harm (n = 9), and anti-experimentation views (n = 4).

Conclusion: Addressing parental decision making in the light of these reasons could enhance the parental permission process for parents of critically ill neonates.

What factors do parents consider when they are asked to enrol their critically ill neonates in research studies? Although these complex decisions are common in many neonatal intensive care units, little is known about how parents conceive these decisions.¹

Retrospective studies of parental permission in neonatal randomised controlled trials have found potential deficiencies in consent in 70% of parents interviewed.² Parental duress associated with having a sick neonate may preclude informed permission.³ Evidence addressing this issue is mixed. One study found that 18 months after participation in neonatal research, 12% of the parents did not remember that they had been asked to consent to a study.⁴ In contrast, when parents whose newborns had participated in a clinical trial and then had died were asked permission to have a post mortem examination performed, they reported not feeling pressured when making this decision.⁵ A study of parents' perspectives, using both retrospectively and prospectively collected data, found that 34–43% of parents had chosen to participate in a study because they believed that their neonate would get better care in the study.⁶ Personal benefit was also given as a reason to choose participation by 64% of parents in another large retrospective study; other reasons parents cited included societal benefit (48.6%) and no risk to their child (39.2%).²

We report here our analysis of a collection of unsolicited comments about neonatal research participation that were provided by parents in the context of a qualitative study on the effect of either prenatal or postnatal diagnosis of congenital heart malformation on parental permission for neonatal cardiac surgery.⁷ Given the paucity of prospectively collected data on parental decision making with regard to neonatal research, these unprompted reasons offered by the parents provide a fortuitous opportunity to examine factors that parents consider when contemplating whether to have their newborn child participate in research.

METHODS

After institutional review board approval, parents of neonates having cardiothoracic surgery were approached from 1 November 2001 through 1 May 2002 and invited to

participate in a study evaluating the impact of prenatal diagnosis on parental permission for neonatal cardiac surgery.⁷ Semistructured qualitative interviews were performed between postoperative day 1 and 7 (mean 3). During these interviews, 34 of 49 parents mentioned, unsolicited, their reasons for or against participation in neonatal research. All of these parents were asked, as a follow up question, if they felt any pressure to participate in the studies.

This report represents a qualitative analysis of these unsolicited, and originally unanticipated, parental comments about research participation. The specific research studies presented to the parents (and their decisions about participation) were obtained from the principle investigator for each study and are presented in table 1. The studies offered to the parents for their neonate's participation depended on diagnosis and type of planned surgery. The number of studies in which the parents were invited to participate ranged from one to four and varied over the course of data collection.^{8,9}

All the interviews were audio-taped, transcribed, and analysed using N-Vivo qualitative software.¹⁰ Three independent reviewers of the transcripts identified parental reasons in the interviews. Through dialogue, a consensus set of reasons was defined—that is, societal benefit, personal benefit for their neonate, perception of risk, perception of no harmful consequences, and anti-experimentation. The transcripts were then reanalysed in the light of the reasons.

To gain further insight into the experience and self report of this sample, we conducted a limited statistical exploratory data analysis. The existence of significant bivariate associations was determined using the χ^2 test. The binomial distribution was used to estimate confidence intervals for proportions. The association between the expression of certain reasons for research participation and declining to participate in research was estimated using the cross table odds ratio. These analyses were conducted using Stata 8.2 (Stata Statistical Software 8.2, College Station, Texas, USA).

RESULTS

Thirty four parents of 24 neonates spontaneously offered their reasons for or against research participation. Of these

Table 1 Participation in interviews stratified by other concurrent study participation

Concurrent studies	Current interview study		
	Participated	Declined	Total
Genetic aetiology of CHD	25	2	27
EEG/long term development	28	2	30
New anaesthetic	11	5	16
Anti-hypertensive	1	2	3
Phosphodiesterase inhibitor	3	0	3
Total	68	11	79

CHD, Congenital heart disease; EEG, electroencephalogram.

parents, 29 were white, four were African American, and one was Hispanic. Fourteen were fathers. Eighteen received a prenatal diagnosis of congenital heart disease, and 16 had neonates who were diagnosed postnatally. The diagnoses of the neonates included hypoplastic left heart syndrome (13), transposition of the great arteries (4), total anomalous pulmonary venous return (3), coarctation of the aorta (3), and tetralogy of Fallot with absent pulmonary valve (1).

Parents cited five types of reason for or against research participation during the interviews (table 2)

Societal benefit

Benefit to the general population was the most prevalent theme mentioned by parents as a reason to participate in a research study (18/34). "I want other families to benefit from this stuff, too" reported one parent. "I thought we could help other people" reported another parent describing why she chose to participate in the genetics study.

Personal benefit

The second most commonly cited reason (16/34) concerned possible benefit to their neonate, especially with the study that involved postoperative electroencephalogram (EEG) monitoring.⁸ "EEG provides vehicle for feedback" was mentioned by one father. When one parent was asked how they selected studies in which to participate, the answer was "just the ones that are beneficial to the baby."

Risk

A third of parents (10/34) cited the risk of harm as a consideration when deciding whether to have their neonate participate in research studies. Parents distinguished between risks perceived when considering participation from risks appreciated while participating in a study. "After I saw everything they fixed to her head ... I was worried that chunks of hair and skin would be missing off her head" expressed one parent after watching EEG leads being placed on her neonate. For parents who declined research participation because of risk, they often mentioned wanting "standard, normal care." For the antihypertensive study after repair of aortic coarctation, one father reported that he

declined participation because he worried that the baby might suffer harm during the five minute waiting period that the study protocol specified before the dose of medication could be changed. "I didn't want my baby's blood pressure to be like borderline stroke or seize and them be doing, '30 more seconds'."

No harm

Nearly a third of parents (9/34) chose to participate in a research study because they perceived no risk of harm associated with participation. "I just think it was a neutral decision and it wasn't going to hurt her in any way." Others had specific examples of no harm, such as "no pricks, no needles, no chemicals." One mother reported her perception of no harm as it related to the institution, "it's top notch. You're not going to do something that's going to put them at risk."

Anti-experimentation

Several parents (4/34) expressed anti-experimental beliefs. "It's like taking your car in for an experiment basically." Four parents used metaphors comparing neonates participating in research studies to both guinea pigs and frogs. One parent who gave permission for her neonate to participate in two studies and declined one study reported that she declined because "it's more like research, just felt like he was more or less a guinea pig."

Time and pressure

None of the parents interviewed reported having felt any pressure to participate in the research studies. One parent, however, commented that a "half hour before surgery is not the time to ask about a research study." Conversely, other parents described having sufficient "time to ponder." One parent advocated a prenatal approach for parental permission, "that way you'd have a couple of months to come up with questions instead of a couple of hours."

Timing of diagnosis, research participation, and reasons

We categorised the reasons of societal and personal benefit as "pro" reasons, the perception of risk and anti-experimental beliefs as "con" reasons, and the perception of no harm associated with research participation as a "neutral" reason in parental research participation decision making. Five parents expressed neither pro nor con reasons; 18 expressed only pro reasons; five expressed only con reasons; and six expressed both pro and con reasons. Using these aggregated categories of reasons, we found that there was no association between when the neonate's congenital heart malformation had been diagnosed (prenatally or postnatally) and declining to participate in any research study ($p = 0.8$) or in the parents' expression of either pro ($p = 0.8$) or con ($p = 0.5$) reasons. All parents who declined to participate in a research study cited a con reason (8/8; one tailed 97.5% confidence interval (CI) 63% to 100%); conversely, among parents who cited a con reason, 73% declined to participate in a study (8/11; 95% CI 39% to 94%). Parents who expressed a pro reason were less likely to have declined to participate (odds ratio (OR) 0.14; 95% CI 0.02 to 0.98); parents who expressed the neutral reason were not significantly less likely to have declined (OR 0.32; 95% CI 0.03 to 3.29).

DISCUSSION

The 34 parents who during qualitative interviews spontaneously commented on research participation cited a variety of reasons to decide whether their neonate should participate in research studies. From their perspective, these parents felt that they are able to make informed decisions by using

Table 2 Unsolicited reasons from parents for opinions about research participation

Reasons	Parents (n = 34)	
	Number	Percentage
Societal benefit	18	53
Personal benefit	16	47
Perception of risk	10	29
No harm	9	26
Anti-experimentation	4	12

specific reasons of societal benefit, personal benefit, risk perception, perceived lack of harm, and anti-experimentation beliefs. They did not report feeling undue pressure given the critically ill state of their newborn child.

In agreement with other investigations of parental permission, we found that parents who perceived benefit, either personal or societal, were more likely to participate than if they perceived risk.¹¹ Rarely, though, have these investigations studied parents who had recently made decisions about research participation for sick neonates. One study, which administered a questionnaire to parents within "a few days" after considering research participation for their infant, reported that research participation correlated with a composite "risk, benefit, and attitudes" factor.¹² This factor included the probability and magnitude of risk and benefit, altruism, general attitude to research, perceived complexity of decisions, freedom to make decisions, and concerns about reprisal.¹² Our study also collected data within 72 hours of research participation decision, and in contrast with the close ended questionnaire study, analysed unsolicited comments from parents who spontaneously offered as reasons for research participation perceptions of societal and personal benefit, in addition to perceiving risk or lack of harm.

Societal benefit is an often cited reason for neonatal research participation.^{2 13} In our study, societal benefit was cited by more than 50% of the parents as a reason for their neonate to participate in a clinical trial. In the large Euricon study on the informed consent process for neonatal randomised trials, 49% of the parents chose to participate to benefit future infants.² An altruistic perspective of parents of critically ill neonates to help future babies is positively associated with research participation, in both our study and the literature.¹³

Personal benefit was the next most common reason for parents to choose research participation. Parents perceived the extra monitoring of an EEG portion as a clear and immediate benefit to their neonate. In other studies involving neonatal research, personal benefit was the primary reason to choose research participation.² It has been reported that sick newborn infants may benefit from participation in a randomised controlled trial, as the placebo group had a better outcome than the eligible but non-randomised group.¹⁴ There is no clear explanation for this so called "inclusion benefit" phenomenon.¹³

Our study, with its small sample of parents, has both strengths and weaknesses. The data were collected by interview near to the time when parents had decided whether to participate in research studies, with the mean interview day only three days after their neonate had cardiothoracic surgery. This proximity of decision to data collection minimises potential recall bias, which hampers the interpretation of other retrospective studies of parental permission.^{15 16} Because of the spontaneous nature of parental comments about research participation, our study was not designed to link parental comments to specific research studies, and could thus do so only when parents mentioned a specific identifying feature of a study, such as the EEG leads. We used no means of objectively testing parental understanding of the specific elements of parental permission, nor did we guide them through a structured interview with regard to risks, benefits, and reasons for participation. The spontaneity of the parental comments, though, can also be viewed as an important strength of our study, as the parents

were not prompted nor led to mention societal or personal benefit.

Our findings suggest that systematic, prospective exploration of the reasons parents of critically ill neonates use to make decisions both for and against research participation is warranted. Furthermore, objective testing of the different elements of the parental permission process would enable comparisons between parents' perception of risks and benefits and the actual probability and magnitude of risks and benefits. The parents' perspective should be incorporated into the ongoing discussion of the optimal way to obtain parental permission for neonatal research participation.

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REFERENCES

- 1 **Singhal N, et al.** Parents' perceptions of research with newborns. *J Perinatol* 2002;**22**:57-63.
- 2 **Mason S.** Obtaining informed consent to neonatal randomised controlled trials: interviews with parents and clinicians in the Euricon study. *Lancet* 2000;**356**:2045-51.
- 3 **Mason S.** Obtaining informed consent for neonatal randomised controlled trials: "an elaborate ritual"? *Arch Dis Child Fetal Neonatal Ed* 1997;**76**:F143-5.
- 4 **Stenson BJ, Becher JC, McIntosh N.** Neonatal research: the parental perspective. *Arch Dis Child Fetal Neonatal Ed* 2004;**89**:F321-4.
- 5 **Snowdon C, Elbourne DR, Garcia J.** Perinatal pathology in the context of a clinical trial: attitudes of bereaved parents. *Arch Dis Child Fetal Neonatal Ed* 2004;**89**:F208-11.
- 6 **Burgess E, Singhal N, Amin H, et al.** Consent for clinical research in the neonatal intensive care unit: a retrospective survey and a prospective study *Arch Dis Child Fetal Neonatal Ed* 2003;**88**:F321-3.
- 7 **Hoehn KS, Wernovsky G, Rychik J, et al.** Parental decision-making in congenital heart disease. *Cardiol Young* 2004;**14**:1-6.
- 8 **Gaynor JW, Gerdes M, Zackai E, et al.** Apolipoprotein E genotype and neurodevelopmental sequelae of infant cardiac surgery. *J Thor Cardiovasc Surg* 2003;**126**:1736-45.
- 9 **Zuppa AF, Wernovsky G, Nicolson S, et al.** The effects of cardiopulmonary bypass and modified ultrafiltration on the plasma pharmacokinetics of milrinone in infants with hypoplastic left heart syndrome. *Crit Care Med* 2003;**30**:613.
- 10 **Richards L.** *Using NVivo in qualitative research*, 2nd ed. Melbourne: QSR International, 2000.
- 11 **Tait AR, Voepel-Lewis T, Malviya S.** Factors that influence parents' assessments of the risks and benefits of research involving their children. *Pediatrics* 2004;**113**:727-32.
- 12 **Zupancic JA, Gillie P, Streiner DL, et al.** Determinants of parental authorization for involvement of newborn infants in clinical trials. *Pediatrics* 1997;**99**:E6.
- 13 **Schmidt B, Gillie P, Caco C, et al.** Do sick newborn infants benefit from participation in a randomized clinical trial? *J Pediatr* 1999;**134**:151-5.
- 14 **Lantos JD.** The "inclusion benefit" in clinical trials [editorial]. *J Pediatr* 1999;**134**:130-1.
- 15 **Elbourne DS, Garcia C, Field D.** Trial experience and problems of parental recollection of consent. *BMJ* 2001;**322**:49-50.
- 16 **Smith R.** Babies and consent: yet another NHS scandal. But it should lead to improvements in research governance within the NHS. *BMJ* 2000;**320**:1285-6.



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