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# 'Silent Voices' in Health Services Research: Ethnicity and Socioeconomic Variation in Participation in Studies of Quality of Life in Childhood Visual Disability

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## Abstract

**Purpose.:** To investigate patterns of participation of visually impaired (VI) children and their families in health services research.

**Methods.:** The authors compared clinical and sociodemographic characteristics of children and their families who participated with those who did not participate in two studies of quality of life (QoL) of VI children. In Study 1, the authors interviewed VI children and adolescents, aged 10 to 15 years, about their vision-related quality of life

(VRQoL) as the first phase of a program to develop a VRQoL instrument for this population. One hundred seven children with visual impairment (visual acuity in the better eye LogMar worse than 0.51) were invited to participate in the interviews. Study 2 investigated health-related quality of life (HRQoL) of VI children using an existing generic instrument, administered in a postal survey. 151 VI children and adolescents, aged 2 to 16 years, with hereditary retinal disorders were invited to participate in the survey.

**Results.:** The overall participation level was below 50%. In both studies, participants from white ethnic and more affluent socioeconomic backgrounds were overrepresented. Participation did not vary by age, sex, or clinical characteristics.

**Conclusions.:** The authors suggest that there are barriers to participation in child- and family-centered research on childhood visual disability for children from socioeconomically deprived or ethnic minority groups. They urge assessment and reporting of participation patterns in further health services research on childhood visual disability. Failure to recognize that there are “silent voices” is likely to have important implications for equitable and appropriate service planning and provision for VI children.

Engaging persons who use health services in research to inform their planning and provision is a well-established principle,<sup>1-3</sup> but achieving high levels of their participation is an ongoing challenge. Despite the efforts to overcome them, participants' sociocultural beliefs and socioeconomic circumstances have been reported to be important barriers in research with adult participants.<sup>2-4</sup> Eliciting the voices of those subgroups who would otherwise be underrepresented is critical to ensuring that their needs are addressed.

There has been limited investigation of participation bias in research in childhood disability, particularly in studies of health-related quality of life (QoL). QoL is viewed as subjective perceptions of how status, condition, and disability affects people's daily lives.<sup>5</sup> Here, we investigate the influence of both clinical and sociodemographic characteristics on participation rates in two *distinct* studies of QoL involving children and adolescents who are visually impaired.

## Methods

### Participants and Design

Two groups of participants were drawn from two larger ongoing programs of work. The aim of the first program was to develop a novel self-report vision-related quality of life (VRQoL) instrument specifically for visually impaired (VI) children and adolescents. In the first phase of this program 32 children with visual impairment were interviewed individually in depth about their QoL, with a view to capturing their experiences of living with visual disability (Study 1). The overall aim of the second program of work was to understand the clinical and genetic characteristics of early-onset hereditary retinal disorders, which most commonly occur in Asian populations.<sup>6,7</sup> We examined the QoL of 44 children and adolescents with hereditary retinal disorders, which enrolled in the parent study using a generic multidimensional pediatric tool for assessing children's health-related (HR) QoL (Pediatric Quality of Life Inventory [PedsQL 4.0]).<sup>8</sup> The two studies were conceived independently, made use of different methodologies in recruitment and procedure, and drew on different populations.

## Identification of Eligible Children and Adolescents

In both studies, the participants were patients in the Department of Ophthalmology or the Developmental Vision Clinic at Great Ormond Street Hospital and in the Pediatric Glaucoma Service or Genetic Eye Disease Service at Moorfields Eye Hospital in London, United Kingdom.

Children and adolescents who participated in Study 1 were drawn from an existing sampling frame of eligible patients ( $N = 375$ ) in the VRQoL program comprising a database that included information on clinical data, ethnicity, and contact details. They were eligible if they were visually impaired (visual acuity [VA] in the better eye: Snellen worse than 6/18 and LogMar worse than 0.51) because of any visual disorder without any other significant impairment and if they were aged between 10 and 15 years. The sampling frame was stratified by age and VA, and children were invited by random selection from each stratum to ensure the sample was representative with respect to those variables. As recruitment proceeded, each nonparticipating child was replaced by another of comparable age and VA. Wherever possible, replacements were also children from an ethnic minority, based on our prior concern about potential underrepresentation of this group in childhood visual disability research.<sup>2</sup> Overall, 107 children and adolescents were invited to participate in interviews. Before establishing contact with each family, the family doctor was contacted and informed of the aims and the design of the study.

Children and adolescents who participated in Study 2 were drawn from an existing cohort of patients already enrolled for clinical and molecular genetic investigation of childhood retinal dystrophies at Moorfields Eye Hospital and Great Ormond Street Hospital. From this cohort, all 151 patients aged 2 to 16 years were eligible and invited, irrespective of level of visual function and of whether the condition was isolated or was part of a systemic disorder.

# Recruitment

Eligible children and their families were initially contacted by a letter including separate information sheets for the parents/guardians and the child, informed consent/assent forms (for parents and children), and a background questionnaire to elicit detailed individual-level socioeconomic and demographic information (which they were asked to return regardless of whether they were taking part). The information sheet contained a detailed description of the content and purpose of the study, the reasons the family was approached, the confidentiality procedure regarding information they were asked to provide, and the contact details for further queries and any concerns. All letters were in English. Prepaid envelopes were provided to facilitate response.

In Study 1 only, families who did not respond to the initial invitation were followed up by a telephone call 2 weeks later to ask whether they received the information and whether they had any questions. This allowed for any potential language barriers or misconceptions about the research to be resolved. We were unable to conduct the procedure in English on only two occasions: on one we were able to use a translator and on the second we were asked to liaise with a family member who was fluent in English.

If necessary in Study 1, a second phone call or mailing was undertaken (e.g., if the invitation letters and forms were lost in the mail or at home). By contrast, in Study 2, the families who did not reply were sent a single postal reminder 2 to 4 weeks later but were not contacted by telephone. Thus, each study adhered to the specific protocols regarding contact with potential participants as approved by respective ethics committees (Study 1 by Great Ormond Street Hospital and UCL Institute of Child Health NHS Research Ethics Committee; Study 2 by Moorfields and Whittingdon Local Research Ethics Committee). Both studies followed the tenets of the Declaration of Helsinki.

# Procedure

Requests made of the participants were different in the two studies. Participants in Study 1 were interviewed individually by a research assistant about their QoL, usually at home but also occasionally in the clinic or at school, in a session that lasted approximately 1 hour.

Participants in Study 2 were asked to self-complete the PedsQL 4.0 (parental completion for children younger than 5 years) and to return it by mail.

# Statistical Analyses

Participation patterns were examined separately for each study. Thus, for each study, after the overall participation level was examined, the participants were compared with nonparticipants with respect to sociodemographic and clinical characteristics: age, sex, severity of vision loss, time of visual impairment onset, ethnicity, and socioeconomic status. Most nonparticipating

families did not return the questionnaires designed to elicit individual level sociodemographic information; therefore, existing data from the hospital records were used to investigate variations in ethnicity (classified according to the UK Office for National Statistics classification<sup>9</sup>) and socioeconomic status (based on English postal code used to derive the Index of Multiple Deprivation [IMD]<sup>10</sup>). Proportions were compared using the 95% confidence interval (CI) and a test for statistical differences in proportions.<sup>11</sup>

## Results

### Participation Rates

Overall participation rates were below 50% in both studies, with participation in Study 1 ([Fig. 1](#)) somewhat higher than in Study 2 ([Fig. 2](#)). Notably, it was not possible to establish contact with 24% of the families considered eligible, and thus invited, for participation in Study 1, largely because of invalid contact details in the hospital patient information system (61.5%). [Figure 1](#) provides a breakdown of the reasons contact could not be established.

#### Figure 1.

[View Original](#) [Download Slide](#)

Flow chart of recruitment and level of participation in Study 1.

#### Figure 2.

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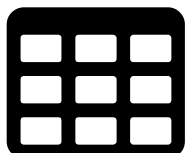
Flow chart of recruitment and level of participation in Study 2.

In Study 1, nonresponders were those with whom direct contact by phone had been established but who failed to provide definitive responses (e.g., have not had time to look at the invitation letter, have not made a decision yet and would respond at a later date, or did not return the consent form after a phone message reminder was left by a researcher (30%; [Figure 1](#)). Nonresponders in Study 2 were those families who did not reply even after the second mailing (66%; [Fig. 2](#)). Nonparticipants thus comprised nonresponders plus those who actively declined in each study.

### Participation Bias

In each study, a greater proportion of participating than nonparticipating children were of white ethnicity, with Asian participants particularly significantly underrepresented in Study 2 ([Tables 1, 2](#)). In both studies, a greater proportion of participants were from families with the most affluent socioeconomic status (highest IMD quintile) compared with nonparticipants. Notably, levels of participation did not vary by age, sex, visual acuity, or time of onset of visual impairment in either study.

## Table 1.

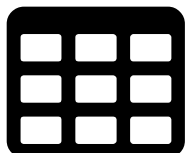


[View Table](#)

Comparison of Characteristics of Participating, Nonparticipating, and Noncontactable Children in Study 1

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## Table 2.



[View Table](#)

Comparison of Characteristics of Participating and Nonparticipating Children in Study 2

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In Study 1, nearly half the families with whom contact could not be established at all were from the most socioeconomically deprived group (lowest IMD), and none were from the least deprived group (highest IMD quintile; [Table 1](#)). In addition, more than half the noncontactable children were of nonwhite ethnicity. There were no differences between noncontactable and participating children with respect to clinical characteristics.

## Discussion

Each of our two studies of quality of life in childhood visual disability aimed to capture the perspective of the affected child and his or her family, but fewer than half the invited families participated in each. Participation did not seem to be influenced by age of the child, level of visual impairment, or other clinical features. Rather, participation varied by key sociocultural characteristics of the children, with those of white majority ethnicity and those from the more

socioeconomically affluent backgrounds more likely to take part than those from all other ethnic groups and from more socioeconomically deprived groups. Furthermore, there appears to be a gradient of effect with respect to these factors, such that differences in patterns of participation were even greater for eligible families who could not be contacted than for those who were contacted and invited to take part but who did not participate.

It would have been interesting to investigate the patterns of participation by family structure (number of parents living with child), level of parental education and occupation, family history of visual impairment, and parent(s) main language to understand better the patterns of participation and to dissect the possible key causes. However, the size of our sample and the lack of availability of data on these variables within routine clinical records precluded this assessment. Equally, it would have been of interest to compare those who actively declined with those who did not respond, but the small sample prevented statistically meaningful comparisons to be made. Nevertheless, despite the size of our sample, we report consistent findings about variation in participation by ethnicity and socioeconomic status that have important implications for future health services research on childhood visual disability.

It is difficult to directly compare our participation rates with those of similar studies of health-related quality of life of children because these are infrequently reported, despite the potential impact of nonparticipation bias.<sup>12,13</sup> Our achieved levels of participation were not high, especially when compared with epidemiologic surveys of health or disease in adults. However, research that focuses on children's subjective experiences of their disability is likely to be a sensitive issue for families and may affect their willingness to participate. Some families who actively declined to participate in Study 1, though supportive of the research, expressed concerns that their child might find it distressing to talk about their experiences of being visually impaired. Other families, possibly because of complex cultural factors, might have worried about stigma or about repercussions as a result of what they might have perceived as "complaining" about health services.<sup>2</sup> Thus, we suggest that our achieved participation rates, though low, may be a realistic target for similar research in other areas of visual disability in childhood.

A higher level of participation was achieved in Study 1 in which there was direct contact with potential participants by phone. It is possible that, if more than one such contact had been made to follow up on families who indicated an interest but did not subsequently reply, a higher participation rate would have been achieved. Where ethical considerations allow this, we advocate direct contact with invited families during the process of recruitment, especially because it allows potential problems, such as language barriers or any concerns or misconceptions about the research, to be identified and addressed.

Nevertheless, in Study 1, we were unable to establish direct contact with a quarter of invited families using the contact information held as current within the patient information system. It is likely that, unknown to us, a large percentage of nonresponding families in Study 2 were also noncontactable. The effect of this inability to directly contact families was to create attrition in the recruitment process, such that those not contacted (i.e., not invited) were even more likely to be of lower socioeconomic status or from ethnic minority groups than those invited but subsequently not participating.<sup>2,14</sup> Every year a significant minority of families in the United Kingdom with children younger than 15 years move home.<sup>15,16</sup> It is possible that mobility is even greater among families of disabled children, especially at key stages such as transitions in education, and among families from less affluent socioeconomic backgrounds whose housing may be less stable. Our findings highlight that accurate and regularly updated patient contact information, preferably linked to clinical databases, is a prerequisite for effective biomedical research.

Achieving an optimum and representative sample of subjects in health services research is a universal challenge, with evidence of participation declining globally.<sup>12,13</sup> Literature involving adult patients suggests a number of strategies that may be effective in optimizing research participation among socioeconomically disadvantaged and ethnic minority subgroups, among them community-based recruitment using community advisors, suitable patient advocates, and researchers from minority backgrounds.<sup>17-20</sup> However, there are further challenges in research involving children with disabilities that operates within sensitive ethical constraints.<sup>21-24</sup> Interdisciplinary collaborations are needed to better understand barriers to participation and to develop innovative methods of encouraging participation in childhood visual disability research. For instance, these may include ways of improving the content and scope of information about research participation so that it is simple, linguistically accessible, and socioculturally sensitive while it allows families sufficient time to make informed decisions.<sup>23,25</sup> Special attention should be given to developing noncoercive approaches to enhancing parental understanding of the importance and the feasibility of their child's participation,<sup>24</sup> regardless of their disability, as the means of giving their child "a voice."

Our findings add to the emerging body of evidence about ongoing barriers to participation in child- and family-centered research on childhood visual disability for persons from socioeconomically deprived or ethnic minority groups. These attributes are interrelated, and complex interventions will be required to overcome existing barriers. The price of a failure to hear silent voices will be inadequately informed and, thus, potentially inequitable health service planning and provision for VI children.

## Footnotes



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