**Title page**

**Title:** 'Silent voices' in health services research: ethnicity and socio-economic variation in participation in studies of quality of life in childhood visual disability

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**Abbreviations:** Visual Impairment (VI), Quality of Life (QoL), Vision Related (VR), Health Related (HR), Office of National Statistics (ONS), Index of Multiple Deprivation (IMD)

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***ABSTRACT***

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

***Methods:*** We compared clinical and socio-demographic 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, we interviewed VI children, age 10-15 years, about their vision-related quality of life (VRQoL) as the first phase of our programme to develop a VRQoL instrument for this population. 107 children with VI (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 visually impaired children and young people (age 2-16) with hereditary retinal disorders were invited to participate in a survey.

***Results:*** The overall participation level was below 50%. In both studies, participants from white ethnic and more affluent socio-economic backgrounds were over-represented. Participation did not vary by age, gender or clinical characteristics.

***Conclusions:*** We suggest that there are barriers to participation in child and family centred research on childhood visual disability for individuals from socio-economically deprived or ethnic minority groups. We urge assessment and reporting of participation patterns in further health services research on childhood visual disability. Failure to recognise that there are 'silent voices' is likely to have important implications for equitable and appropriate service planning and provision for visually impaired children.

## *INTRODUCTION*

Engaging individuals who use health services in research to inform their planning and provision is a well established principle.[1-3] However, achieving high levels of their participation is an ongoing challenge and, despite the efforts to overcome them, participants' socio-cultural beliefs and socio-economic circumstances have been reported to be important barriers in research with adult participants.[2-4] Eliciting the voices of those subgroups which would otherwise be under-represented is critical to ensuring that their needs can be addressed.

There has been limited investigation of participation bias in research in childhood disability, in particular in studies of health related quality of life (QoL). QoL is viewed as a person's subjective perception of how their status, condition or disability affects their daily lives[5]. Here, we investigate the influence of both clinical and socio-demographic characteristics on participation rates in two *distinct* studies of QoL involving children and young people who are visually impaired (VI).

## *METHODS*

### *Participants and design*

Two groups of participants were drawn from two larger ongoing programmes of work. The aim of the first programme is to develop a novel self-report vision-related quality of life (VRQoL) instrument specifically for VI children and young people. In the first phase of this programme 32 children with VI 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 programme of work is to understand the clinical and genetic characteristics of early onset hereditary retinal disorders, which most commonly occur in Asian populations.[6, 7] We examined QoL of 44 children and young people with hereditary retinal disorders enrolled in the parent study using a generic multidimensional paediatric tool for assessing children's health-related (HR) QoL (Pediatric Quality of Life Inventory – PedsQL 4.0).[8]

The two studies were conceived independently, employing different methodologies in recruitment and procedure, and drawing on different populations.

*Identification of eligible children and young people*

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

Children and young people who participated in Study 1 were drawn from an existing sampling frame of eligible patients (N = 375) in the VRQoL programme comprising a database that included information on clinical data, ethnicity and contact details. They were eligible if: *i)* they were visually impaired (visual acuity - VA - in the better eye Snellen worse than 6/18; LogMar worse than 0.51) due to any visual disorder, but in the absence of any other significant impairment; and *ii)* they were aged between 10-15 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 definitely non-participating child was replaced by another comparable in age and VA. Wherever possible 'replacements' were also children from an ethnic minority, based on our prior concern about potential under-representation of this group in childhood visual disability research.[2] Overall, 107 children and young people were invited to participate in interviews. Prior to establishing contact with each family, the family doctor was contacted and informed of the aims and the design of the study.

Children and young people 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 & 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 whether the condition is isolated or 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 socio-economic and demographic information (which they were asked to return whether they were taking part or not). The information sheet contained a detailed description of the content and the purpose of the study, the reasons for why the family was approached, the confidentiality procedure regarding the information that they were asked to provide, and the contact details for further queries and any concerns. All letters were sent out in English. Pre-paid 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 information and whether they had any questions. This allowed for any potential language barriers or misconceptions about research to be resolved. We were unable to conduct the procedure in English only on two occasions: on one we were able to use a translator, and on the second occasion the family we contacted asked us to liaise with a family member who was fluent in English.

If necessary in Study 1, a second phone call and/or mailing were undertaken (e.g., if the invitation letters and forms have been lost in post or at home). By contrast, in Study 2, the families who did not reply were sent a single postal reminder 2-4 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 and Study 2 by Moorfields & Whittingdon Local Research Ethics Committee). Both studies followed the tenets of the Declaration of Helsinki.

***Procedure***

The demands placed upon the participants were different in the two studies. The participants in Study 1 were interviewed individually about their QoL by a research assistant, in the majority of cases at home, but also occasionally in clinic or at school, in a session that lasted generally about an hour. The participants in Study 2 were asked to self-complete the PedsQL 4.0 (or parental completion for children age less than 5 years) and return it by post.

***Statistical analyses***

Participation patterns were examined separately for each study. Thus, for each study, the overall participation level was examined and then participants were compared with non-participants with respect to socio-demographic and clinical characteristics: age, gender, severity of vision loss, the time of VI onset, ethnicity and socio-economic status. As the majority of non-participating families failed to return the questionnaires eliciting individual level socio-demographic information, the existing data from the hospital records were used to investigate the variation in ethnicity (classified according to the UK Office for National Statistics classification - ONS[9]) and socio-economic status (based on English postal code used to derive the Index of Multiple Deprivation - IMD[10]). Proportions were compared using the 95% confidence intervals (CI) and a test for statistical differences in proportions.[11]

***RESULTS***

***Participation rates***

Overall participation rates were below 50% in both studies, with participation in Study 1 (Figure 1) being somewhat higher than in Study 2 (Figure 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 due to invalid contact details held within the hospital patient information system (61.5%). Figure 1 provides a breakdown of the reasons why contact could not be established.

(Insert Figure 1)

In Study 1, non-responders were those with whom direct contact by phone had been established, but who failed to provide a definitive response, for instance: 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%, Figure1). Non-

responders in Study 2 were those families who did not reply even after the 2nd mailing (66%, Figure 2). Non-participants thus comprised non-responders plus those who actively declined in each study.

(Insert Figure 2)

***Participation bias***

In each study, a greater proportion of participating compared to non-participating children were of white ethnicity, with Asian participants particularly significantly under-represented in Study 2 (Tables 1 and 2). In both studies, a greater proportion of participants were from families with the most affluent socio-economic status (highest IMD quintile) compared to non-participants. Notably, levels of participation did not vary by age, gender, visual acuity and the time of VI onset in either study.

In Study 1, nearly a half of the families with whom contact could not be established at all were from the most socio-economically deprived group (lowest IMD) and none were from the least deprived group (highest IMD quintile) (Table 1). In addition, more than half of non-contactable children were of non-white ethnicity. There were no differences between non-contactable and participating children with respect to clinical characteristics.

(Insert Table 1)

(Insert Table 2)

***DISCUSSION***

Each of our two studies of quality of life in childhood visual disability aimed to capture the perspective of the affected child and their family, but less than half of invited families participated in each. Participation did not seem to be influenced by the age of the child, level of visual impairment or other clinical features. Rather, participation varied by key socio-cultural characteristics of the children, with those of white majority ethnicity and those from the more socio-economically affluent backgrounds being more likely to take part than those from all other ethnic groups and more socio-economically 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 those 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 VI and parent(s) main language, in order to understand better the patterns of participation and 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 socio-economic status that have important implications for future health services research on childhood visual disability.

It is difficult to directly compare our participation rates to other similar studies of health related quality of life of children, as these are infrequently reported, despite the potential impact of non-participation bias.[12, 13]Our achieved levels of participation were not high, especially when compared to epidemiological surveys of health or disease in adults. However, research that focuses on the 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, whilst supportive

of the research, expressed concerns that their child might find it distressing to talk about their experiences of being visually impaired. Other families, potentially owing to complex cultural factors, may have worried about stigma or about repercussions, as a result of what they may have perceived as 'complaining' about health services.[2] Thus, we suggest that our achieved participation rates, although low, may be a realistic target for similar research in other areas of visual disability in childhood

A somewhat higher participation level 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 had 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 as 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 non-responding families in Study 2 were also non-contactable. 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 socio-economic status or from ethnic minority groups than those invited but subsequently not participating.[2, 14] Every year a significant minority of UK families with children under 15 years of age move home.[15, 16] It is possible that mobility is even greater amongst families of disabled children, especially at key stages such as transitions in education, as well as amongst families from less affluent socio-economic background whose housing may be less stable. Our findings highlight that accurate and regularly updated patient contact information, preferably linked to clinical databases, is a pre-requisite 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. [12, 17] Literature involving adult patients suggests a number of strategies that may be effective in optimising research participation amongst socio-

economically disadvantaged and ethnic minority subgroups: for example community-based recruitment utilising community advisors, suitable patient advocates and having researchers from minority backgrounds.[18-21] However, there are further challenges in research involving children with disabilities, which operates within sensitive ethical constraints.[22-25] Inter-disciplinary collaborations to better understand barriers to participation and develop innovative methods of encouraging participation in childhood visual disability research are needed. 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 socio-culturally sensitive, while allowing the families sufficient time for making an informed decision. [24, 26] Special attention should be given to developing non-coercive approaches to enhancing parental understanding of the importance and the feasibility of their child's participation,[24] 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 centred research on childhood visual disability for individuals from socio-economically deprived or ethnic minority groups. These attributes are inter-related 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 visually impaired children.

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