

Title: Capturing children and young people’s perspectives to identify the content for a novel vision-related quality of life instrument

Authors:

Jugnoo S Rahi, PhD, FRCOphth^{1,2}

Valerie Tadić, MSc, PhD¹

Sarah Keeley, BSc³

Gillian Lewando-Hundt, MPhil, PhD³

on behalf of the Vision-related Quality of Life (VQoL) group¹

Affiliations:

¹ Medical Research Council (MRC) Centre of Epidemiology for Child Health, Institute of Child Health, University College London (UCL), London, United Kingdom

² Institute of Ophthalmology, UCL, London, United Kingdom

³ School of Health and Social Studies, University of Warwick, Coventry, United Kingdom

Correspondence:

Jugnoo S Rahi, MRC Centre of Epidemiology for Child Health, UCL Institute of Child Health, 30 Guilford Street, London WC1N 1EH, UK; Telephone: 44 (0)20 7905 2250; Fax: 44 (0)20 7905 2381; Email: j.rahi@ich.ucl.ac.uk

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¹ The members of the VQoL group are listed in acknowledgments section

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Key words: Vision-related quality of life; visual impairment; child; questionnaire; instrument; child-centered approach; self-report; interviews.

Abbreviations: QoL – quality of life; VQoL – vision-related quality of life; CYP – children and young people

ABSTRACT

Objective: To describe a child-centered approach to identifying content for a novel self-report questionnaire for assessing vision-related quality of life (QoL) of visually impaired (VI) or blind (BL) children and young people.

Design: Questionnaire development.

Participants: A stratified random patient sample of children and young people (CYP) who are VI/BL (visual acuity in the better eye Snellen worse than 6/18; logarithm of the minimum angle of resolution – logMAR - worse than 0.51) due to any visual disorder, but in the absence of any other significant impairment, aged between 10-15 years (N=49); and a convenience school-based sample of CYP with VI/BL aged between 10-17 years (N=29).

Methods: Individual interviews were conducted with a stratified random sample of 32 CYP with VI/BL, age 10-15 years. The interviews followed a topic guide based on vision-related issues identified from a focus group of affected children and young people, combined with a literature review and consultations with professionals. Collaborative qualitative thematic analysis was undertaken and used to derive draft items of the instrument, using the children's own language wherever possible. Items were reduced, rephrased and refined through individual consultation, as well as an expert reference group of children and young people with VI/BL, and supplemented by the research team's consensus.

Main outcome measure: A draft 47-item instrument.

Results: 874 potential questionnaire items were initially generated spanning the following domains: *Social relations, acceptance and participation; Independence and autonomy; Psychological and emotional well being; Future - aspirations and fears; Functioning - home, school and leisure* and *Treatment of eye condition*.

This was eventually reduced to a 47-item instrument with each item presented as a vignette describing a QoL issue from an 'illustrative' child's perspective. Thus, the responding child

reports on how much they are presently like, and also how much they wish to be like that child, using a 4-point Likert-type scale.

Conclusions: We demonstrate that a child-centered approach to identifying the content for a self-report vision-related QoL questionnaire is feasible. We suggest this approach is critical to capturing accurately children and young people's subjective perspectives on the impact of living with impaired vision.

INTRODUCTION

Health-related quality of life (HR QoL) has been increasingly recognized as an important patient-reported outcome measure in clinical practice and in health services research involving children and young people. Recent views are that the QoL of children is a multidimensional psychological construct encompassing the physical, mental, social, emotional and functional aspects of health and well-being, all considered essentially from the child's perspective.^{1,2} Eiser and colleagues^{3,4} in particular have advocated a child-centered approach to measuring pediatric QoL, arguing that measures of children's QoL should aim to assess the subjective gap between the children's current experiences and their hopes and expectations, following the 'self-discrepancy' theory of QoL.⁵

A number of instruments assessing QoL in children are currently available, either generic instruments used with children with any chronic disorder^{6,7} or disorder-specific tools.⁸⁻¹⁰ To date, generic QoL instruments have largely been used for assessing QoL of children with visual impairment (VI).¹¹⁻¹⁵ However, it is recognized that these do not enable a comprehensive or detailed assessment of the impact of living with a VI, nor do they detect small changes in an individual's condition or discriminate adequately between children with VI. However, few vision-specific QoL measures for children are currently available. The Child Visual Function Questionnaire, a recently reported instrument still under development, is a parent-report measure that is only suitable for young VI children (≤ 7 years).^{16,17} More recently, Cochrane et al. reported the first phase of developing a self-report scale, the Impact of Vision Impairment on Children - IVI_C, for VI children and young people aged 8-18 years, promoting an amalgamated perspective of young people, their parents and VI professionals - elicited through focus groups - in defining the questionnaire content.¹⁸

We identified the need for a measure that is grounded in children and young people's own perspectives of their lives. Here, we report the process of content identification and item generation for a novel self-report vision related QoL questionnaire for children and young people (CYP) with VI, age 10-15 years (VQoL_CYP). Specifically, we describe a child-centered and interdisciplinary methodological approach for eliciting children and young people's voices about the impact of living with impaired vision. Like Eiser et al.,^{3,4} the objective is to use the instrument to capture the 'self-discrepancy' between the children's actual and desired status in the areas of life affected by their VI. Thus, its development differs substantially from many other QoL instruments^{4,16,18-20} by being grounded on children and young people's views in all the early stages of development. Here, we report this process to demonstrate the feasibility and value of this approach.

METHOD

The study was approved by the National Health Service (NHS) Research Ethics Committee for University College London (UCL) Institute of Child Health and Great Ormond Street Hospital, London, UK, and followed the tenets of the Declaration of Helsinki.

Sample

Identification of eligible children

The participants were drawn from patient databases at the Department of Ophthalmology and the Developmental Vision Clinic at Great Ormond Street Hospital, and the Pediatric Glaucoma Service and Genetic Eye Disease Service at Moorfields Eye Hospital, London UK. The sampling frame of patients had the following inclusion criteria: *i*) they were visually impaired, severely

visually impaired or blind (VI/BL)ⁱⁱ (visual acuity - VA - in the better eye Snellen worse than 6/18 or logarithm of the minimum angle of resolution – LogMAR - worse than 0.51) due to any visual disorder, but without any other significant impairment (i.e., learning, sensory or motor); and *ii*) they were aged 10-15 years. Children and young people were selected for different phases using a stratified random sampling approach to ensure the sample was representative of the total eligible population of interest with respect to characteristics such as level of VI and age at onset of VI (i.e., early/from birth or later) in childhood. The family physician was contacted and informed of the aims and the design of the study and then written consent was sought from the parents and the children.

Additionally, a convenience sample of children and young people with VI, aged between 10-17 years, was drawn from 2 specialist community/VI schools in England, UK after parental consent and head teacher agreement.

Development of the interview topic-guide

We developed an interview topic guide through firstly, a detailed literature review covering visual impairment, childhood disability, and pediatric and adult QoL, and secondly, through observations of consultations at pediatric ophthalmology clinics and discussion with professionals. We then conducted a focus group discussion with 11 children and young people (12-17 years old) with VI at a community special school and at the same school, piloted the topic guide in semi-structured interviews with 4 children and young people (two boys and two girls) aged 12-16 years. This resulted in some refinement and the inclusion of an ‘ice-breaker’ activity to ease the participants into the interview. The six areas of the final topic guide were: 1) School, 2) Home Life, 3) Activities and Socialising, 4) Life Skills and Independent Living, 5) Eye Problems

ⁱⁱ For brevity, we consider term VI in the remainder of the paper.

and the Eye Clinic, and 6) the Future. Each topic was explored in general terms and then specifically in relation to how eyesight affected activities and concerns.

Interview procedure and analysis

Of 107 children and young people invited from the sampling frame, 32 participated in the interviews, of whom 83% had an early onset VI, and 69% had severe VI or blindness (described elsewhere²¹). In-depth individual interviews lasting 30-60 minutes were conducted - 25 at the child's home, 5 at school and 2 at the hospital. 20 interviews were conducted without a parent present, and in the remaining 12, the parent was requested to be a passive observer and completed a questionnaire to report their views as supplementary information that was not included in the formal analyses.

The interviews were digitally recorded and transcribed. Four researchers undertook collaborative thematic analysis of the interview data²² by reviewing the same interviews independently and then comparing identified themes to develop an agreed coding template. Data were coded into these low-order descriptive categories using NVivo7 software.²³ Transcripts for children age 10-12 were considered separately to those of young people age 13-15 in order to check if there were age-related themes. Finally, overarching higher-order themes that brought together related areas of children's concerns, and which could be notionally considered 'domains' in a QoL instrument, were identified to provide an architecture for exploring the data (Figure 1, illustrating the sequence of the thematic analysis undertaken, is available at <http://aaojournal.org>).

Item development and reduction

The domains provided the structure for undertaking a child-centered approach firstly, to generate the items from verbatim interview statements, and secondly, to reduce the generated item list.

Item development: Five of the research group (one expert user – a VI parent of a VI child) independently scrutinized the data organized in individual domains, and identified verbatim statements deemed relevant to QoL to file as draft items in the VQoL_CYP ‘item bank’. Statements expressing a single idea (e.g., ‘*I don’t let my eyesight stop me from doing what my friends do*’) were filed as ‘draft items’. More extensive elaborations (e.g., ‘*when we’re in the car, and when my mum gets angry about cars swerving and sometimes she gets really, really angry and she asks me to read out the number plate, she forgets that I’ve got an eye problem. So I have to get my phone out and zoom in and take a picture!*’) were re-phrased into sentences expressing a single idea, keeping close to the children’s language (e.g., ‘*Sometimes people forget that I have an eyesight problem*’), to be filed in the item bank.

Item reduction: Initially, following a Delphi consensus approach,²⁴ five of the team independently rated all the items in the item bank, based on whether they were important and relevant to vision-related QoL (i.e., coding them as ‘keep’ or ‘remove’), and whether they could be answered in terms of ‘self-discrepancy’ between actual and ideal status. Then, the items were discussed and only those with consensus of at least 4 of the 5 team members were retained. This reduced list was then re-scrutinized for repetitions and similar or overlapping items were grouped together. Finally, two of the team independently reviewed the list to select the preferred versions of similar item, giving preference to those most closely derived from verbatim statements. Disagreements regarding the remaining items were resolved by discussion.

The remaining ‘long list’ of items was then evaluated by expert user groups of children and young people with VI to a) gauge importance, relevance and comprehensibility of the items, b) to assess the suitability of a 5-point Likert response scale, and c) to identify suitable administration formats. This exercise included firstly, individually consulting 4 pupils with VI, aged 15-16, at a residential specialist VI school, secondly, conducting an expert reference group discussion with 10 children and young people with VI, aged 10-16 years, at a community specialist school, and

thirdly, consulting individually 4 children and young people with VI, aged 10-16 from the sampling. As a result, the item list was further reduced and refined before we administered in person the reduced VQoL_CYP draft to 13 children and young people, age 10-15, from the study sampling frame.

RESULTS

Questionnaire instrument content

Table 1 shows the final hierarchy of the child-centered data structure that emerged from the topics in the interview guide, first in the form of lower-order descriptive categories and then as the overarching domains that grouped related areas of child-expressed issues.

Three domains were quite congruent with some topics in the interview guide such as 1) Future concerns, 2) Treatment of eye condition, and 3) Independence and autonomy. Three other domains involved cross cutting themes that captured the central concerns of interviewees when talking about their lives. These were 1) Psychological and Emotional well being, 2) Social relationships, participation and acceptance, and 3) Functioning at home, school and leisure. (We describe the content of each domain, in terms of the related, child-expressed issues in Table 2, which is available at <http://aaojournal.org>. Here, to illustrate some of these issues, we also list qualitative data examples from which verbatim statements were sampled to create an item bank).

874 potential questionnaire items were initially generated from verbatim statements in the 6 domains described in Table 2 (available at <http://aaojournal.org>), with some domains resulting in more items than others. The item bank was gradually and systematically reduced to 79 items through research group consensus and finally to 47 items through evaluation by children and

young people with VI (Figure 2, available at <http://aaojournal.org>). As a result of these consultations with children and young people as experts, we removed the items that were considered unimportant or irrelevant, and re-phrased the items that lacked clarity, using their own language.

Questionnaire instrument design

Following consultations with children, we implemented the following changes to the instrument. We decided to present each item as a vignette describing a VQoL issue from an ‘illustrative’ child’s perspective (e.g., *‘Ben feels frustrated because of his eyesight’*) to serve as a standardized anchor for the responding child when considering his/her own Actual and Ideal status, and reduced the Likert-point scale to 4 categories. Thus, in the final version of the draft instrument, the respondent reports on how much they are presently like (Actual Status: *‘How much are you like Ben?’*), and also how much they wish to be like that child (Ideal Status: *‘How much do you want to be like Ben?’*), using one of the following options: 1 = not at all, 2 = a little bit, 3 = quite a lot, and 4 = exactly. We used gender-appropriate names when referring to the ‘illustrative’ child, and adopted the terms ‘children’ and ‘young people’ when addressing participants age range 10-12 years and those age range 13-15 years respectively. The instrument was formatted with a simple uncrowded layout without graphics (e.g., tables or tick boxes), and we considered in-person and electronic presentation (via email or on a CD) for children requiring alternative administration methods to large print. Based on individual in-person administration of the draft VQoL_CYP, the time required for self-completion of the instrument was 15-20 min on average. We are currently undertaking formal piloting of the draft VQoL_CYP on a representative population of children and young people with VI.

DISCUSSION

In this paper, we describe a child-centered and interdisciplinary approach for identifying the content for a novel self-report QoL instrument designed specifically for children and young people with VI age 10-15. Following this approach, we developed a topic guide for individual semi-structured in-depth interviews through consultation with children and young people, and through these interviews we elicited their voices on the impact of living with impaired vision. To develop items from the thematic analysis, we consulted expert reference groups of children and young people with VI, and conducted further individual interviews to shape the content and style of the instrument. This resulted in a 47-item 4-point Likert scale - VQoL_CYP - which we are presently piloting.

Our findings demonstrate that, by using child-centered methods, it is possible to elicit children and young people's hopes, aspirations and concerns, rather than just functional status. In the ophthalmic literature to date, the concepts of functional vision and vision-related quality of life have often been used interchangeably, with instruments being developed to assess in broad terms the 'impact' of vision loss on an individual.²⁵ However, rather than focussing on their ability to perform tasks or activities that require vision (i.e., functional status),²⁶ we explicitly set out to obtain the children and young peoples' perceptions of the impact of living with a visual disability. We believe that conducting individual in-depth interviews enabled us to capture these perceptions and ensured that the view of every participating child and young person was represented. Thus, we demonstrate that a child-centered approach for the development of a QoL instrument that is relevant to children and young people with VI is not only feasible, but also critical for understanding their own views of what is important to them. For example, a frequent source of worry, concern and uncertainty for many children was the unstable nature of their visual prognosis. Related to this was their strong awareness of the challenges and restrictions for their

future education and career, as well as of the practicalities of independent daily living when they were older. Children frequently voiced their disappointments and frustrations with living with a visual disability and the associated stigma, and highlighted the impact this has on their confidence and self-esteem. However, the interviews also revealed their resilience, and coping strategies, developed independently or as a result of support by family and friends.

Although some differences across different age groups were noted, these were not deemed sufficient to warrant designing separate instruments. However, it was recognized that subsequently some items, being generated from these issues, may be more applicable to some children than others depending on their ages (e.g., the transition from primary to secondary school) and the characteristics of their visual condition (e.g., age of onset and stability of the condition).

In line with the child-centered content, we report a rigorous systematic approach to item reduction based on this content rather than exclusively or mainly on psychometric analysis. This child-centered method has a strong internal and external validity, rigorously grounding the VQoL_CYP instrument on children's expressed concerns and using their verbatim statements to develop items.

We also pursued a self-report method for completion of the VQoL_CYP, as a means of giving children a voice. The literature supports that if they are cognitively able, children should be encouraged to self-report,^{3,4} as there is good evidence that children may have different views about their QoL from their parents or professionals involved with them.²⁷⁻³¹ Thus, although our approach is in keeping with the current developments in the general field of pediatric QoL,^{1,2} it is at variance with the history of development of pediatric VQoL instruments.

We believe that our final instrument - after piloting and formal implementation - will be complementary to other clinical and proxy instruments measuring VI related concerns in children and young people. The instrument should be a valid tool for repeated assessments, capturing children's current thinking about their lives. It should also enable the professionals working with children and young people with VI to assess the change in their vision-related QoL outcomes across specific developmental stages, such as transition from primary to secondary school, or as a result of treatment interventions. The instrument should be useful in a number of areas. Firstly, it could be used for evaluating the broader benefits of new treatments to preserve and improve sight. Secondly, it should be applicable in planning and provision, and measuring the impact of rehabilitation, education and social services. Thirdly, it should provide a further dimension in clinical settings to aid in managing individual children. Fourthly, it could be applied in population assessments of disease burden. Finally, the instrument would enable prioritization of the agenda for service provision and for future research on visually impairing disorders in childhood. More generally, the use of the instrument will raise awareness of the need for, and constraints to, improving social inclusion of the visually impaired and their families. Through use of the VQoL_CYP in clinical, policy and research settings children's own perspectives of their visual loss will be included directly in decision-making, especially in relation to interventions to preserve or improve vision. This method is clearly potentially transferable to other patient-reported outcome measures.

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References

1. Bullinger M, Ravens-Sieberer U. Health-related QOL assessment in children: a review of the literature. *Eur Rev Appl Psychol* 1995;45:245-54.
2. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. *Health Technol Assess* 2001;5:1-157. Available at: <http://www.hta.ac.uk/fullmono/mon504.pdf>. Accessed July 21, 2010.
3. Eiser C, Cotter I, Oades P, et al. Health-related quality-of-life measures for children. *Int J Cancer Suppl* 1999;12:87-90.
4. Eiser C, Vance YH, Seamark D. The development of a theoretically driven generic measure of quality of life for children aged 6-12 years: a preliminary report. *Child Care Health Dev* 2000;26:445-56.
5. Calman KC. Definitions and dimensions of quality of life. In: Aaronson NK, Beckmann JH, eds. *The Quality of Life of Cancer Patients*. New York: Raven Press; 1987:1-9.
6. Ravens-Sieberer U, Gosch A, Rajmil L, et al. KIDSCREEN-52 quality-of-life measure for children and adolescents. *Expert Rev Pharmacoecon Outcomes Res* 2005;5:353-64.
7. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: Reliability and validity of the Pediatric Quality of Life Inventory version 4.0 Generic Core Scales in healthy and patient populations. *Med Care* 2001;39:800-12.
8. Palmer SN, Meeske KA, Katz ER, et al. The PedsQL Brain Tumor Module: initial reliability and validity. *Pediatr Blood Cancer* 2007;49:287-93.
9. Landgraf JM, Abetz L, Ware JE. *Child Health Questionnaire (CHQ): A user's manual*. Boston, MA: The Health Institute, New England Medical Center; 1996. **AQ: must provide specific inclusive pagination for material being referenced**
10. Baars RM, Atherton CI, Koopman HM, et al, DISABKIDS Group. The European DISABKIDS project: development of seven condition-specific modules to measure health related quality of life in children and adolescents. *Health Qual Life Outcomes* [serial online] 2005;3:70. Available at: <http://www.hqlo.com/content/3/1/70>. Accessed July 21, 2010.
11. Boulton M, Haines L, Smyth D, Fielder A. Health-related quality of life of children with vision impairment or blindness. *Dev Med Child Neurol* 2006;48:656-61.
12. Chak M, Rahi JS, British Congenital Cataract Interest Group. The health-related quality of life of children with congenital cataract: findings of the British Congenital Cataract Study. *Br J Ophthalmol* 2007;91:922-6.

13. Wong HB, Machin D, Tan SB, et al. Visual impairment and its impact on health-related quality of life in adolescents. *Am J Ophthalmol* 2009;147:505-11.
14. CRYO-ROP Cooperative Group. Health-related quality of life at age 10 years in very low-birth-weight children with and without threshold retinopathy of prematurity. *Arch Ophthalmol* 2004;122:1659-66.
15. van Dijk J, Huisman J, Moll AC, et al. Health-related quality of life of child and adolescent retinoblastoma survivors in the Netherlands. *Health Qual Life Outcomes* [serial online] 2007;5:65. Available at: <http://www.hqlo.com/content/5/1/65>. Accessed July 21, 2010.
16. Felius J, Stager DR Sr, Berry PM, et al. Development of an instrument to assess vision-related quality of life in young children. *Am J Ophthalmol* 2004;138:362-72.
17. Birch EE, Cheng CS, Felius J. Validity and reliability of the Children's Visual Function Questionnaire (CVFQ). *J AAPOS* 2007;11:473-9.
18. Cochrane G, Lamoureux E, Keeffe J. Defining the content for a new quality of life questionnaire for students with low vision (the Impact of Vision Impairment on Children: IVI_C). *Ophthalmic Epidemiol* 2008;15:114-20.
19. Cui Y, Stapleton F, Suttle C. Developing an instrument to assess vision-related and subjective quality of life in children with intellectual disability: data collection and preliminary analysis in a Chinese population. *Ophthalmic Physiol Opt* 2008;28:238-46.
20. Watson M, Edwards L, Von Essen L, et al. Development of the Royal Marsden Hospital paediatric oncology quality of life questionnaire. *Int J Cancer Suppl* 2000;12:65-70.
21. Tadic V, Hamblion EL, Keeley S, et al. 'Silent voices' in health services research: ethnicity and socioeconomic variation in participation in studies of quality of life in childhood visual disability. *Invest Ophthalmol Vis Sci* 2010;51:1886-90.
22. Boyatzis RE. *Transforming Qualitative Information: Thematic Analysis and Code Development*. Thousand Oaks, CA: Sage; 1998.
23. Lewins A, Silver C. *Using Software in Qualitative Research: A Step-by-Step Guide*. London: Sage; 2007.
24. Herdman M, Rajmil L, Ravens-Sieberer U, et al, European Kidscreen Group, European Disabkids Group. Expert consensus in the development of a European health-related quality of life measure for children and adolescents: a Delphi study. *Acta Paediatr* 2002;91:1385-90.
25. Massof RW, Rubin GS. Visual function assessment questionnaires. *Surv Ophthalmol* 2001;45:531-48.

26. Gothwal VK, Lovie-Kitchin JE, Nutheti R. The development of the LV Prasad-Functional Vision Questionnaire: a measure of functional vision performance of visually impaired children. *Invest Ophthalmol Vis Sci* 2003;44:4131-9.
27. Janse AJ, Sinnema G, Uiterwaal CS, et al. Quality of life in chronic illness: children, parents and paediatricians have different, but stable perceptions. *Acta Paediatr* 2008;97:1118-24.
28. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. *Qual Life Res* 2001;10:347-57.
29. Davis E, Nicolas C, Waters E, et al. Parent-proxy and child self-reported health-related quality of life: using qualitative methods to explain the discordance. *Qual Life Res* 2007;16:863-71.
30. Cremeens J, Eiser C, Blades M. Factors influencing agreement between child self-report and parent proxy-reports on the Pediatric Quality of Life Inventory 4.0 (PedsQL) generic core scales. *Health Qual Life Outcomes* [serial online] 2006;4:58. Available at: <http://www.hqlo.com/content/4/1/58>. Accessed July 21, 2010.
31. Vance YH, Morse RC, Jenney ME, Eiser C. Issues in measuring quality of life in childhood cancer: measures, proxies, and parental mental health. *J Child Psychol Psychiatry* 2001;42:661-7.