

Differences in self-rated versus parent proxy-rated vision-related quality of life and functional vision of visually impaired children

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PII: S0002-9394(21)00310-X
DOI: <https://doi.org/10.1016/j.ajo.2021.05.017>
Reference: AJOPHT 11874

To appear in: *American Journal of Ophthalmology*

Received date: February 12, 2021
Revised date: May 17, 2021
Accepted date: May 26, 2021

Please cite this article as: Alexandra O Robertson , Valerija Tadić , Lisanne A. Horvat-Gitsels , Mario Cortina-Borja , Jugnoo S Rahi , for the Child Vision PROMs group, Differences in self-rated versus parent proxy-rated vision-related quality of life and functional vision of visually impaired children, *American Journal of Ophthalmology* (2021), doi: <https://doi.org/10.1016/j.ajo.2021.05.017>

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HIGHLIGHTS

- Visually impaired children and their parents disagree on the child's vision-related outcomes.
- Parents consistently under-estimate their child's functional vision.
- Parents can both under- and over-estimate their child's vision-related quality of life.
- Child-parent discrepancy is greatest in older children.

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Title:

Differences in self-rated versus parent proxy-rated vision-related quality of life and functional vision of visually impaired children

Short title:

Disagreement in child self-report and parent-proxy reports.

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ABSTRACT

Purpose: To investigate disagreement between children's self-reported vision-related quality of life (VQoL) and functional vision (FV), and their parents' proxy-reports.

Design: Cross-sectional study.

Methods: 152 children aged 7-18 years with visual impairment (VI) (defined by the World Health Organization), and their parents, were recruited from 22 National Health Service (NHS) Ophthalmology Departments in the United Kingdom.

Age-appropriate versions of 2 vision-specific instruments capturing VQoL and FV, were administered to children alongside modified versions for completion by parents on behalf of their child (i.e. parent proxy-report). Disagreement between self- and parent proxy-report was examined using the Bland-Altman (BA) method, and a threshold of disagreement based on 0.5 standard deviation. Disagreement was analysed according to participants' age, gender and clinical characteristics, using logistic regression analyses.

Results: Children rated themselves as having better outcomes than their parents did, although parents both under- and over-estimated their child's VQoL (mean score difference = 7.7). With each year of increasing age, there was a 1.18 (1.04 – 1.35) higher odds of children self-rating their VQoL better than their parents ($p = 0.013$).

Although parents consistently under-estimated their child's FV (mean score difference = -4.7), no characteristics were significantly associated with differences in disagreement.

Conclusions: Disagreement between child self-report on the impact of VI, and their parents' proxy-reports varies by age. This implies that self-report from children must remain the gold standard. Where self-reporting is not possible, parent proxy-reports may provide useful insights, but must be interpreted with caution.

INTRODUCTION

Health-related quality of life (HRQoL) describes the subjective experience of living with a health condition and its associated impact on everyday life,¹ and is captured using validated patient-reported outcome measures (PROMs). PROMs are now widely used as part of routine clinical practice, and as primary outcomes in clinical trials of new therapies.² Using age-appropriate PROMs, children as young as 5 years can be reliable informants of their HRQoL, as well as other aspects of their physical and mental health.³

When a child is unable to self-report, for example due to physical or cognitive limitations, parent proxy-reports are sometimes used, as parents are considered to be able to understand and report on the impact of the impairment from their child's perspective. A robust evidence-base, however, shows that children and their parents often – and unpredictably – disagree in their assessment of the same subjective outcomes.⁴⁻⁸ This disagreement becomes particularly relevant when decisions are made that determine the child's healthcare.⁹

Parents of children with chronic health conditions or disabilities have been shown to rate their children's HRQoL worse than their children do themselves.^{5,8,10,11} However, the direction and magnitude of disagreement varies by the nature of outcomes measured.⁷ For example, parents and children tend to agree when rating observable

outcomes such as symptoms or physical functioning, and tend to disagree when rating non-observable, psychosocial outcomes.^{12,13} The degree of parent-child agreement/disagreement also varies greatly across different conditions, the type, nature, and severity of the health condition, and the child's sex.^{5,6}

Specifically, disagreement between children's and parent proxy-reports has been identified in specific ophthalmic conditions.¹⁴⁻¹⁶ In a pilot study using two PROMs developed specifically for use with children aged 10-15 years living with visual impairment (VI), one which captures vision-related quality of life (VQoL) – the VQoL_CYP, and the other which captures functional vision (FV) – the FVQ_CYP, we found that parents both over- and under-estimated their child's VQoL (i.e. impact of VI on social and emotional well-being), but consistently under-estimated their child's FV (i.e. difficulty to complete activities requiring vision).¹⁷ There was some variation in disagreement based on participants' socio-demographic (i.e. sex, ethnicity, and socioeconomic status) and clinical (i.e. severity of VI, timing of onset of VI, and rate of deterioration of vision) characteristics.

Since that pilot study,¹⁷ both the VQoL_CYP and FVQ_CYP have been further developed and are now available in their final two age-appropriate versions, applicable to visually impaired children aged from 8 up to 18 years.^{18,19} Here we report a formal investigation of agreement between visually impaired children and their parents, using these instrument in a large participant sample with a wide age-range. Our purpose is to advance understanding of the value and potential pitfalls of using parent proxy-report, when a child is unable to self-report using child-appropriate vision-specific PROMs.

METHODS

This cross-sectional study was approved by the National Health Service Research Ethics Committee for Essex and East of England, United Kingdom (UK), and followed tenets of the Declaration of Helsinki. All participants gave informed individual consent or assent (if younger than 16 years), and parents gave informed consent to their child's participation if they were younger than 16 years.

Participants

Study participants were a clinical sample of children with i) VI or blindness (visual acuity in the better eye of 0.48 logMAR (logarithm of the minimum angle of resolution) or worse, and/or additional visual field defects causing VI) as a result of any visual disorder but without any other significant impairment (i.e. learning, sensory or motor) and ii) aged 8 to 18 years, and their parents. Participants were recruited from two main sites between September 2014 and May 2017, comprising the Department of Ophthalmology at Great Ormond Street Hospital and Moorfields Eye Hospital (Paediatric Glaucoma Service and Genetic Eye Disease Service), supplemented by 20 additional hospitals situated throughout the UK (see Acknowledgements), as part of a larger programme of research developing age-appropriate PROMs for children with VI.^{18,19}

Materials and procedures

Through a cross-sectional postal survey, children and their parents independently completed the relevant age-appropriate versions (one suitable for children aged 8-12 years, and the other suitable for those aged 13-17 years) of both the VQoL_CYP¹⁹ and FVQ_CYP.¹⁸

The VQoL_CYP captures the child's perception of the social and emotional impact of VI, with higher scores indicating *better* VQoL. The VQoL_CYP for children aged 8-12 years contains 20 age-appropriate items, and the VQoL_CYP for 13-17 year olds contains 22 items.

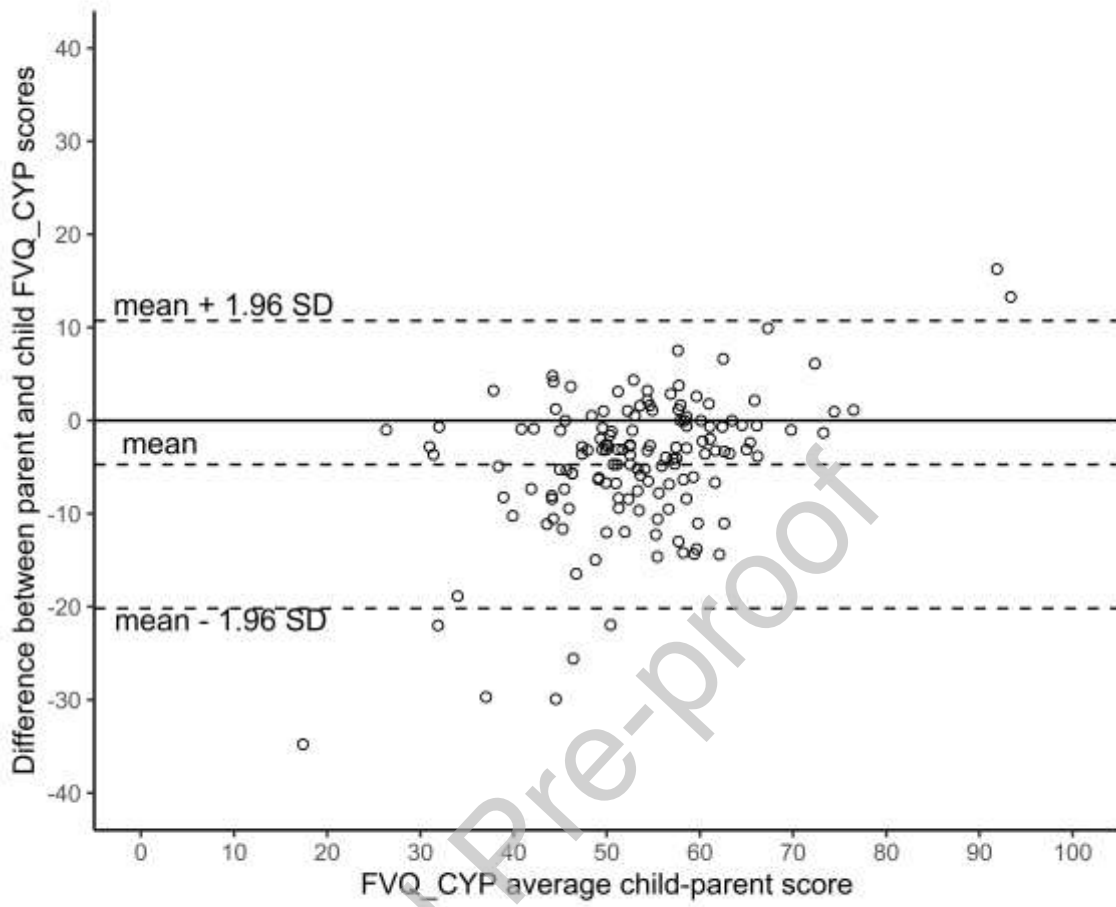
The FVQ_CYP captures self-reported difficulty completing everyday activities requiring vision, with higher scores indicating greater difficulty (i.e. *worse* FV). The FVQ_CYP for children aged 8-12 years contains 28 items specifying everyday activities relevant for children, and the FVQ_CYP for 13-17 year olds contains 38 items. Both age-appropriate versions of the VQoL_CYP and FVQ_CYP are available for use in clinical practice and/or research settings (<https://xip.e-lucid.com>).

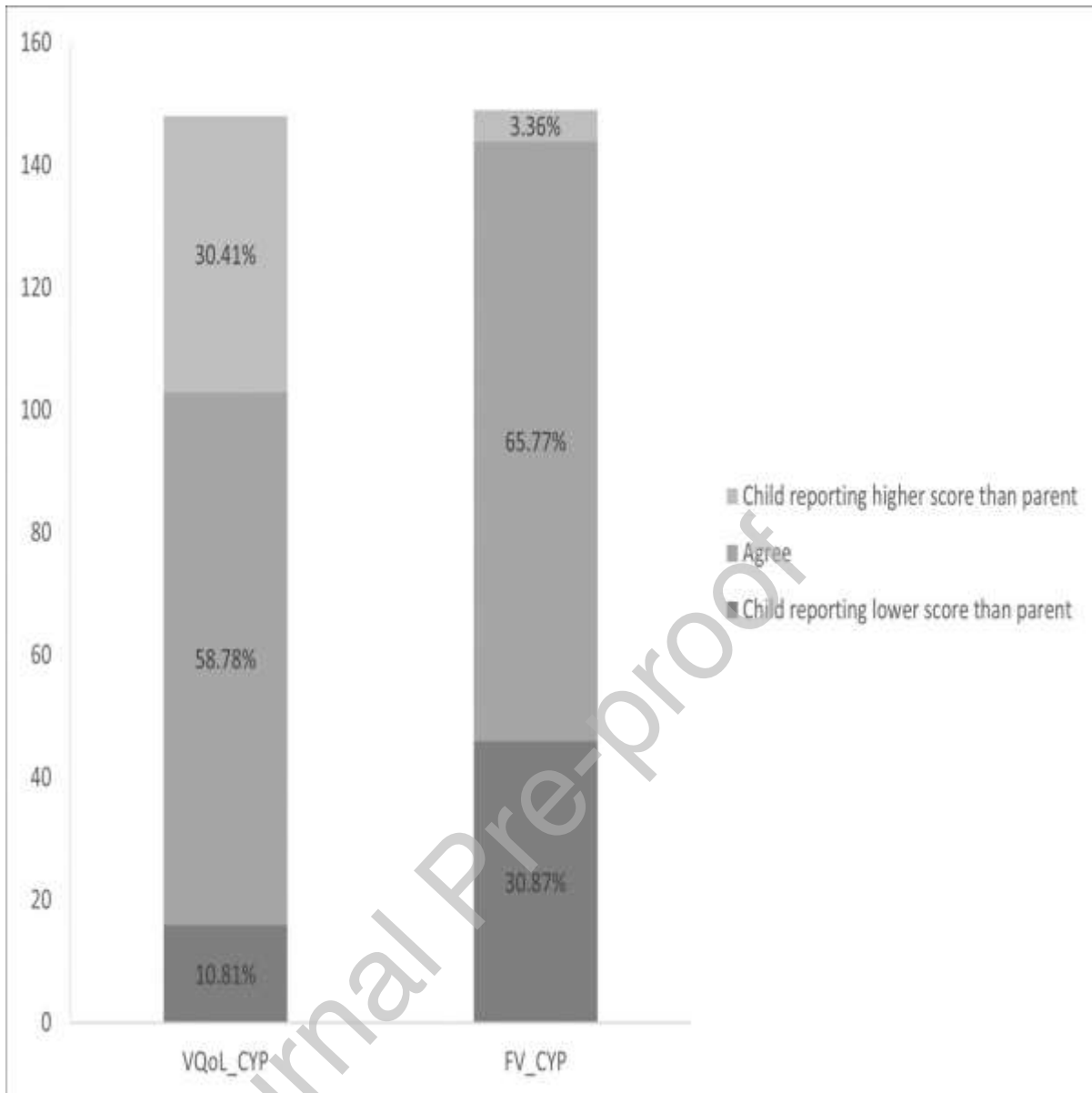
As the age-appropriate versions of the VQoL_CYP and FVQ_CYP have been validated and calibrated using Rasch measurement theory,²⁰ scores from either age-version can be transformed to a scale of 0 to 100, treated as continuous, ratio-level data, and compared, despite variation in the number and wording of items.

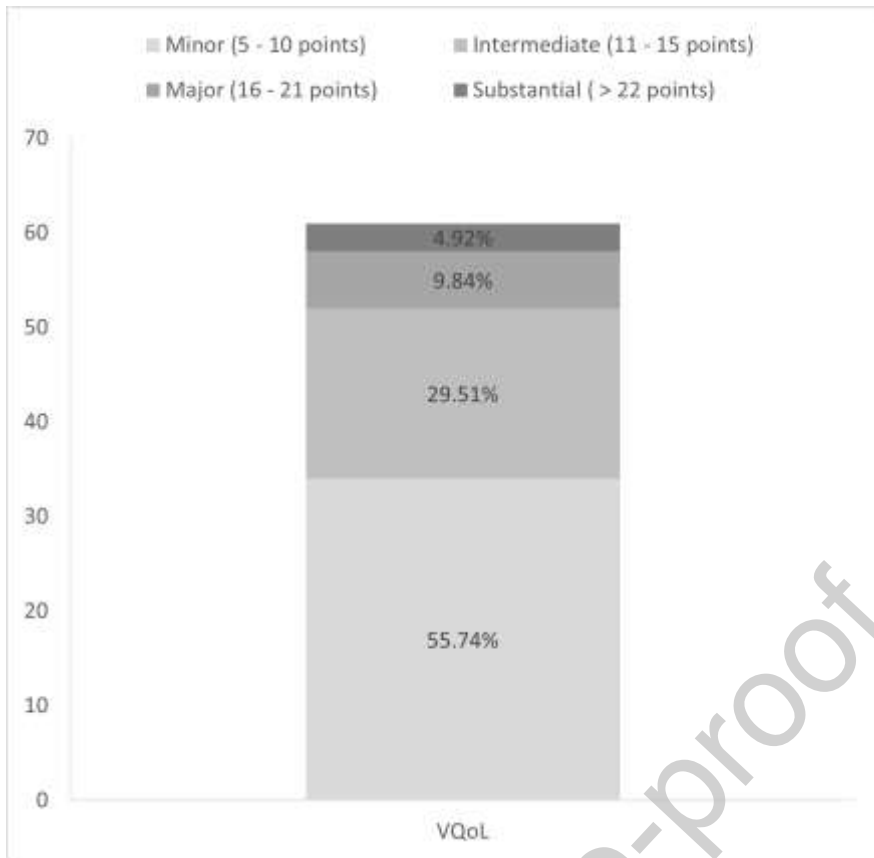
Parent proxy versions of the VQoL_CYP and FVQ_CYP were created for the purpose of this study, containing the same items, but using the item prefix "*My child...*". Items were scored in the same way as the child self-report versions.

Cronbach's α was used to establish reliability of the parent proxy versions (accepted threshold > 0.7).²¹

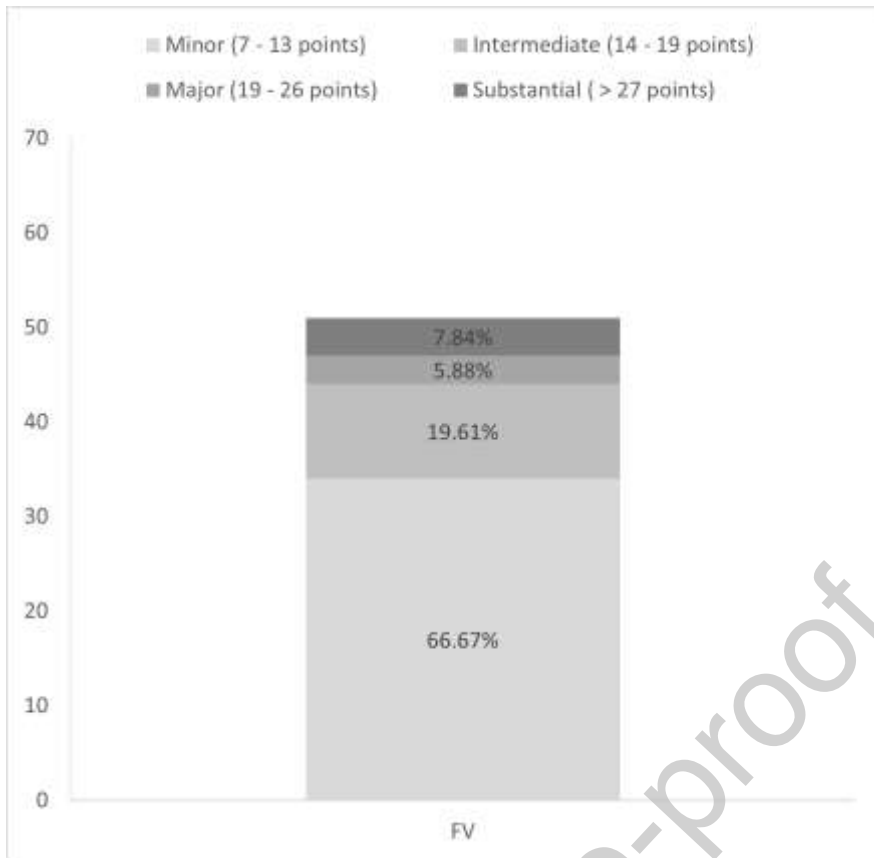
Participants' age, sex and clinical characteristics comprising diagnosis, severity of VI, timing of VI onset, and whether VI was stable or progressive, were collected from hospital electronic records.







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