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

Alexandra O Robertson ,   Valerija Tadić ,
Lisanne A. Horvat-Gitsels ,   Mario Cortina-Borja ,   Jugnoo S Rahi ,
for the Child Vision PROMs group

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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.

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.

Authors:

Alexandra O Robertson*[1]

Valerija Tadić*[1,2]

Lisanne A. Horvat-Gitsels[1,5]

Mario Cortina-Borja[1]

Jugnoo S Rahi[1, 3, 4, 5] *for the Child Vision PROMs group***

*Dr Robertson and Dr Tadić contributed equally as co-first authors.

**Members of the Child Vision PROMs group are listed in the Acknowledgements.

Professor Rahi is the study chair.

Affiliations:

1 UCL Great Ormond Street Institute of Child Health, UK

2 School of Human Sciences/Institute for Lifecourse Development, University of Greenwich, UK.

3 Great Ormond Street Hospital NHS Foundation Trust, UK

4 National Institute for Health Research (NIHR) Biomedical Research Centre at Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology,

UK

5 Ulverscroft Vision Research Group, UK

Corresponding author (and address for reprints):

Jugnoo S Rahi, UCL Great Ormond Street (GOS) Institute of Child Health, Life Course Epidemiology and Biostatistics Section, Population, Policy and Practice Research and Teaching Department, 30 Guilford Street, London WC1N 1EH, UK; Telephone: 44 (0)20 7905 2250; Email: j.rahi@ucl.ac.uk

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

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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_CYP[19] 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.

Data analysis

Data were entered manually into SPSS version 26[22] and Excel datasets independently by two researchers (AR and VT), to afford cross-checking and correction of data entry errors. Missing data were assessed per individual (i.e. parent or child). As per standard guidelines,[23] parent-child dyads including one or both individual participant with ≥ 20% data missing on either the VQoL_CYP or FVQ_CYP were excluded from the dataset for that instrument. Logistic regression models for remaining missing data (< 20%) in the child and parent datasets were fitted to investigate associations between missingness (as the binary response variable) with child participants' socio-demographic and clinical characteristics. As no significant associations were found, the remaining missing data (< 20% of participants FVQ_CYP and VQoL_CYP scores per participant) were imputed using the mean item score for the given responses of the participant. Scores were calculated separately for children and their parents, and transformed to Rasch-scaled scores using the published scoring instructions for each instrument.[18,19]

The assumption of normality for continuous variables was assessed using *z*-tests of skewness and kurtosis, and screening of histograms.[24] Paired-samples *t*-tests compared the mean scores for children and their parents. The direction of child-parent disagreement was examined using a) the Bland-Altman method of limits of agreement (LOAs),[25] and b) half a SD (as the threshold for a minimally important difference in outcome measures[26,27]). Agreement was coded when the absolute difference between child and parent scores was less than or equal to half a SD of the score with the largest variability (i.e. parent or child score). Disagreement was coded when the absolute difference between scores exceeded this value, and further categorised according to whether the child score was higher (Child High) or lower

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(Child Low) than the parent score. The extent of disagreement was classified into four levels: from 0.5 to 1 SD (minor), from 1 to 1.5 SD (intermediate), from 1.5 to 2 SD (major), and higher than 2 SD (substantial).

Logistic regression models were fitted to investigate associations between disagreement and multiple variables. We did not adjust for specific clinical diagnosis as level of VI, timing of onset of VI, and whether VI was stable or progressive are the variables that both reflect the underlying diagnosis and might be expected to correlate with VQoL or FV and disagreement between children and their parents. To aid interpretation, the age of the youngest participant (7 years) was used as the baseline in the regression models. Dichotomous variables were coded as 0 (*Male, VI (visual impairment), Early,* and *Stable*) or 1 (*Female, SVI/BL (severe visual impairment or blindness), Late,* and *Progressive*), meaning that unstandardized coefficients can be interpreted as the change in score between categories. Goodness-of-fit was evaluated using adjusted R^2 for linear, and Nagelkerke's R^2 for logistic regression models.

Post-hoc analyses to explore, in more detail, the driver of any disagreement between children and their parents included fitting four quantile regression models of children's and parents' VQoL_CYP and FVQ_CYP scores adjusted for multiple variables. The resulting conditional models refer to the outcome's median and avoid transformations for the non-normal distribution of VQoL_CYP and FVQ_CYP scores.

RESULTS

A total of 152 parent-child dyads participated, comprising an unbiased sample of the overall UK population of children and young people with VI, with respect to socio-

demographic and clinical characteristics, i.e. the target population for whom the VQoL_CYP and FVQ_CYP are intended (Table 1).[28]

In total, 4 parent-child dyads were excluded from analyses of VQoL_CYP scores, and 3 from analyses of FVQ_CYP scores, due to missing data ≥ 20%. Using the VQoL_CYP, 7 individual children and 7 individual parents (5% of the full sample) had < 20% missing data. Using the FVQ_CYP, 28 individual children and 28 individual parents (18% of the full sample) had < 20% missing data; no characteristics were significantly associated with missing data (Supplemental Table 1).

Histograms containing the mean difference between child and parent scores were screened for normality and considered acceptable. Using the critical z-value of ± 3.29 (and approximate alpha level of 0.05), z-skewness for the mean difference between child and parent scores on the VQoL_CYP and FVQ_CYP indicated a normal distribution ($z = 2.41$ and $z = 0.5$ respectively).

Cronbach's α for parent proxy scores on both instruments exceeded the reliability criteria (> 0.8), indicating good internal consistency in the context of this study.

VQoL

On average, children self-rated their VQoL as significantly better (higher scores) than their parents rated it to be ($t = 3.582$, $p < .001$) (Table 2), although parents both under- and over-estimated their child's VQoL (Figure 1a). Based on the definition of a minimally important difference (i.e. 0.5 SD of the score with the largest variability), the threshold for agreement on the VQoL_CYP was 5 points. Figure 2 shows the distribution of disagreement between child self- and parent proxy-reports, with 41% of children and their parents disagreeing on VQoL. In total, 56% of disagreement was classified as minor (Figure 3a).

Variation in disagreement on VQoL scores by child characteristics

The fully adjusted regression analysis showed that age was significantly associated with children reporting higher (i.e. *better*) VQoL scores than their parents; for each one year increase in age of the child, there was 1.18 (1.04 – 1.35) higher odds of children and parents disagreeing (Table 3). Post-hoc analyses showed that parents were the driver of disagreement; for each one-year increase in age of the child, parents scored their children's VQoL 0.76 (0.23-1.13) points lower, whereas there was no significant association between age of the child and children's self-reported VQoL score (p = 0.81).

In contrast, none of the socio-demographic and clinical characteristics were associated with the odds of children reporting lower (i.e. *worse*) VQoL scores than their parents.

FV

Children's self-reported FV was significantly lower (i.e. *better*) (t = -7.314, p < .001) than their parents' proxy ratings (Table 2), as parents consistently under-estimated their child's FV i.e. gave higher scores than their children (Figure 1b).

The threshold for agreement on the FVQ_CYP was 6 points, meaning that 34% of children and parents disagreed on the child's FV (Figure 2). In total, 67% of disagreement was classified as minor (Figure 3b).

Variation in disagreement on FV scores by child characteristics

The fully adjusted regression analysis showed that none of the socio-demographic and clinical characteristics were associated with the odds of children reporting lower (i.e. *better*) FVQ scores than their parents.

No regression model was fitted for children giving higher (i.e. *worse*) FVQ_CYP scores than their parents, as this was a rare event with only 5 (3%) occurrences.

DISCUSSION

From a cross-sectional study of a large representative sample of children with VI for whom the VQoL_CYP and FVQ_CYP are intended, we report the existence, nature of, and factors associated with meaningful disagreement between visually impaired children's self-reports, and their parent proxy-reports. We found disagreement between parent-child dyads to be most prevalent when reporting VQoL i.e. an outcome encompassing the subjective impact of VI on social and emotional wellbeing. Nevertheless, disagreement was observed among parent-child dyads reporting the impact of VI on FV; an outcome which may be more readily predicted by parents based on observations of their child's daily activities.

The strengths of our study lie in the representativeness of the participant sample with respect to the UK population of visually impaired children and young people,[29] the use of robust child-appropriate vision-specific PROMs, the examination of disagreement in relation to both observed and inferred outcomes, and the size of the study sample (large for a study of the rare outcome of childhood VI), enabling analysis of the assessment of key factors so as to advance understanding of disagreement. Due to limited resources, the study was conducted as a postal survey, which precluded observation of completion and, despite the explicit instructions, there may have been some discussion within families or parental involvement in completion of instruments by their children, which could have produced an erroneously high level of agreement.[30,31] As such, we report minimum estimates of disagreement.

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Our study is unable, by design, to examine change in disagreement over time, and this would be an interesting focus for future longitudinal studies, given our finding of a divergence in child and parent agreement with increasing age of the child. Whilst our study raises interesting hypotheses about the influence of socio-demographic and clinical factors, further research is needed to better understand the variables that shape different perspectives.[32] Finally, to examine direction and magnitude of disagreement between parents and children, we used the criteria of 0.5 SD as indicative of a *minimally* important difference in score in statistical terms. A minimal *clinically* important difference (i.e. the smallest difference in score perceived as beneficial to patients and clinicians[33]), will vary by context, for example, a small improvement in either VQoL or FV could be tangibly important to the child and family even if it falls below the *a priori* threshold for outcomes in a clinical trial.

Overall, we found meaningful disagreement among 41% and 34% of parent-child dyads reporting VQoL and FV, respectively. On average, and in keeping with extant literature in other clinical areas, parents in this study had a tendency to under-estimate their visually impaired child's self-reported outcomes (i.e. parents reported *worse* outcomes than children themselves).[7,27,31,34] Specifically, we found that parents more consistently under-estimate their child's FV (i.e. give higher difficulty ratings), whilst the pattern of disagreement on VQoL was bi-directional.[17] One explanation is that parents of children with VI are particularly sensitive to their child's FV and the associated practical limitations, observing on a regular basis, and from an early stage (as most visually impairing conditions are present at birth or in the first year of life), the activities that their child cannot complete independently. When disagreement occurs, therefore, they may be more prone to *under*-estimating their child's FV ability (i.e. reporting *worse* FV). This is echoed by research showing that

parents of children with hearing impairment showed similar patterns of over-estimating the adverse consequences of deafness in their children.[35] Nevertheless, we found substantial disagreement on both outcomes, indicating that the observable nature of FV does not necessarily promote parents' accurate judgements of their child's self-reported FV.

We demonstrate, for the first time in the population of children and young people living with VI, a complex interaction between direction of parent-child disagreement and the age of the child. With regards to VQoL, we found some indication that, on average, the older the child, the greater the odds of disagreement i.e. the child self-reporting *better* VQoL than their parent's assessment. There could be various explanations for this within each dyad, one of which could be related to a dynamic change in the nature of parent-child communication over time, with older children transitioning away from instances of self-disclosure and shared experiences, and towards desires for privacy.[36] Specifically, however, we found that the difference in disagreement between dyads was driven by variation in parent's proxy-reports of VQoL: a finding which suggests a more complex interaction between the age of the child and nature of disagreement. It is possible that, if acceptance of a health condition takes time and effort, [37-39] growing maturity is likely to promote better internal and psychological adjustment to VI. Thus, as they develop, children may be increasingly well-equipped to adjust to their expanding physical and social environments. At the same time, it is possible that parent's perceptions of their child's growing independence and responsibilities trigger changes in their perceptions of their child's VQoL. Although timing of onset of VI was not found to be significant in the adjusted analyses, there was interesting consistency in terms of parents reporting worse FV than their children in those with early onset VI but better

in those with late onset. However, future, longitudinal analysis tracking dyads is essential to unravel the likely complex interaction between direction of disagreement and the age, and vision-specific clinical characteristics of the child. Importantly, our findings show that it is not possible to extrapolate the likelihood or direction of disagreement between parental and child self-rating on one PROM from measured disagreement on another. This underscores the importance of considering very carefully the scale and nature of likely disagreement before attempting to use proxies to complete instruments designed for self-completion by children and young people. They also demonstrate that children and young people are well able to meaningfully self-rate distinct, albeit conceptually related, outcomes.

In conclusion, our study shows that children living with VI and their parents disagree when reporting two complementary, but distinct vision-specific outcomes, and disagreement varies meaningfully based on the age of the child, and potentially in relation to the timing of onset of VI. Because the views of parents are likely to influence the child's ophthalmic care, our findings emphasise the need for greater understanding by both clinicians and parents that disagreement does exist, and is meaningful, even when reporting potentially 'easily' observable outcomes, such as FV. Differences between child and parent proxy-reports are sufficient to advocate that self-reporting by children themselves remain the 'gold standard' in clinical settings. However, where self-reporting by affected children is not possible, and some assessment of the patient's perspective is necessary, parent proxy-reports can potentially add value to the clinical assessment. Further research is needed to elaborate the least error-prone scenarios.

TABLE OF CONTENTS STATEMENT

Visually impaired children and their parents disagree on the child's vision-related outcomes. Whilst parents consistently under-estimated their child's functional vision, parents both under- and over-estimated the child's vision-related quality of life. Additionally, disagreement between child self-report and their parents' proxy-reports varies by age. This implies that self-report from children must remain the gold-standard.

CRediT Statement

Alexandra O. Robertson: Methodology, Validation, Formal analysis, Investigation, Data curation, Writing – original draft, Writing – review & editing, Visualization, Project administration.

Valerija Tadić: Conceptualization, Methodology, Validation, Formal analysis, Investigation, Data curation, Writing – original draft, Writing – review & editing, Visualization, Supervision, Project administration, Funding acquisition.

Lisanne A. Horvat-Gitsels: Validation, Formal analysis, Writing – review & editing.

Mario Cortina-Borja: Software, Validation, Formal analysis, Writing – review & editing.

Jugnoo S. Rahi: Conceptualization, Methodology, Resources, Writing – review & editing, Supervision, Funding acquisition.

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FIGURE LEGENDS

Figure 1a. VQoL_CYP child and parent agreement

Figure 1b. FVQ_CYP child and parent agreement

Figure 2. Agreement and disagreement between children and parents using the

VQoL_CYP* (*n* =148) and FVQ_CYP** (*n* = 149)

*Higher VQoL_CYP score = better outcome

**Higher FVQ_CYP score = worse outcome

Figure 3a. Extent of disagreement between child and parent reports of VQoL (%)

Figure 3b. Extent of disagreement between child and parent reports of FV (%)

Table 1. Participants' socio-demographic and clinical characteristics

Characteristic	*n* (%)
Age (mean, SD)	**12.3 (3.08)**
Sex	
Male	85 (55.9)
Female	67 (44.1)
Ethnicity[d]	
White UK	90 (59.2)
White other	7 (4.6)
Black British	2 (1.3)
Black African/Caribbean	9 (5.9)
Asian Indian	5 (3.3)
Asian Pakistani	12 (7.9)
Asian Bangladeshi	8 (5.3)
Asian other	5 (3.3)
Mixed	6 (3.9)
Socioeconomic status (by Index of Multiple Deprivation quintile)[b, d]	
1: most deprived	37 (24.3)
2	26 (17.1)
3	24 (15.8)
4	27 (17.8)
5: least deprived	32 (21.1)
Severity of VI	
VI (LogMAR 0.48 – 1.00)[c]	120 (78.9)
SVI/Blind (LogMAR ≥ 1.02)	32 (21.1)
Timing of VI onset	

Early (≤2 years)	125 (82.2)
Late (>2 years)	27 (17.8)
Rate of visual deterioration	
Stable	108 (71.1)
Progressive	44 (28.9)
Diagnosis by site of VI[e]	
Whole globe and anterior segment	5 (3.3)
Glaucoma, primary or secondary	15 (9.9)
Cornea (sclerocornea and corneal opacities)	3 (2.0)
Lens (cataract and aphakia)	19 (12.5)
Uvea	11 (7.2)
Retina	101 (66.4)
Optic nerve	14 (9.2)
Cerebral/visual pathways	10 (6.6)
Other (idiopathic nystagmus, high refractive error)	29 (19.1)
Total	152

[a]3 participants who were just outside the age-range of 8-18 years (3 children aged 7.5 years at the time of invitation) were included due to natural developmental variation across the age boundaries.

[b] Index of Multiple Deprivation (IMD) based on English (UK) postal code[28] extracted from medical records.

[c] 2 participants with VA LogMAR 0.07 – 0.46, and additional visual defects that classified them as visually impaired by WHO criteria, were included.

[d] Sample size varies due to missing data; valid percentages shown.

[e] Does not add up to 100% because some children had VI originating in multiple sites.

Table 2. Parent-child disagreement on VQoL and FV.

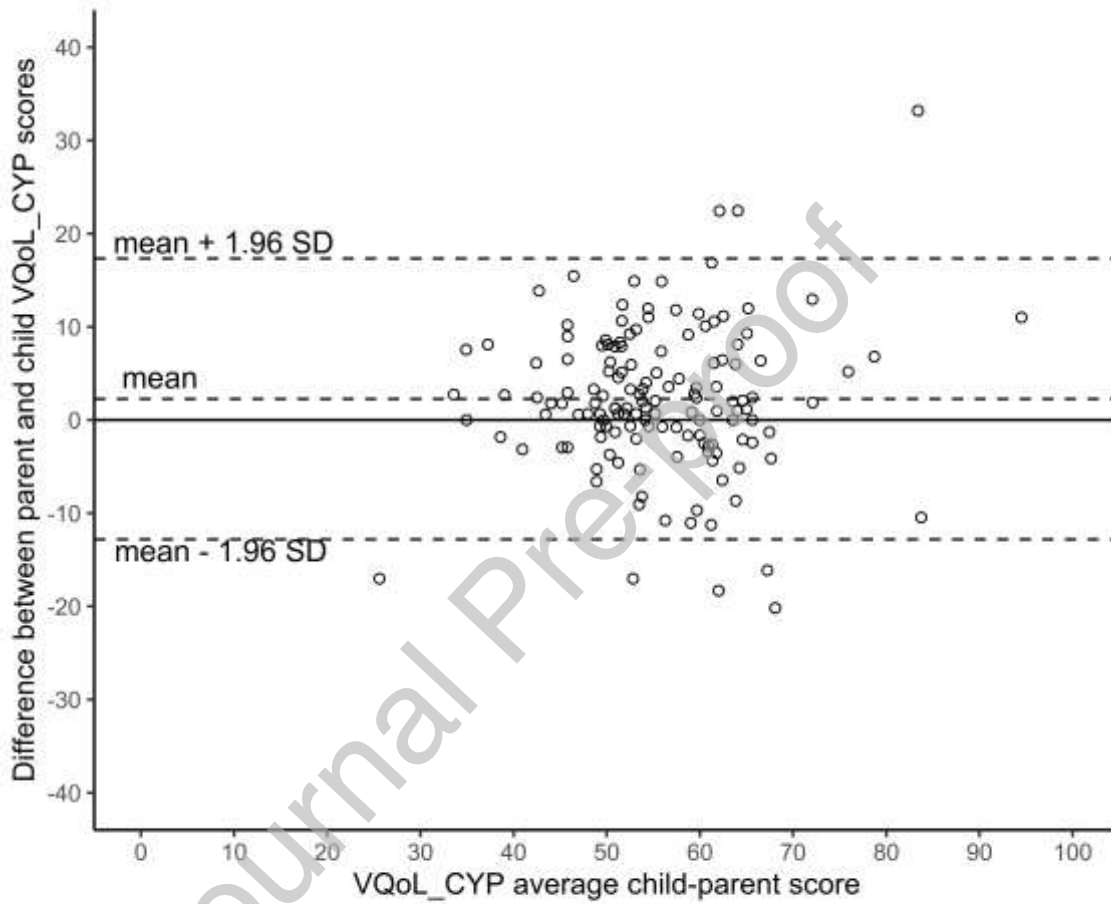| Instrument	Child score (Mean, SD)	Parent score (Mean, SD)	Mean paired difference between scores (SD, 95% CI)	Minimum difference	Maximum difference	Bland-Altman lower-limit of agreement (95% CI)	Bland-Altman upper-limit of agreement (95% CI)
VQoL_CYP	56.7 (10.5)	54.3 (10.0)	2.3 (7.7, 1.0 to 3.5)*	-20.2	33.2	-12.81 (-14.04 to -11.57)	17.33 (16.10 to 18.57)
FVQ_CYP	50.7 (13.0)	55.4 (9.2)	-4.7 (7.9, -6.0 to -3.5)*	-40.0	16.3	-20.18 (-21.45 to -18.92)	10.73 (9.46 to 12.00)

*Paired *t*-test difference significant at *p* < .001

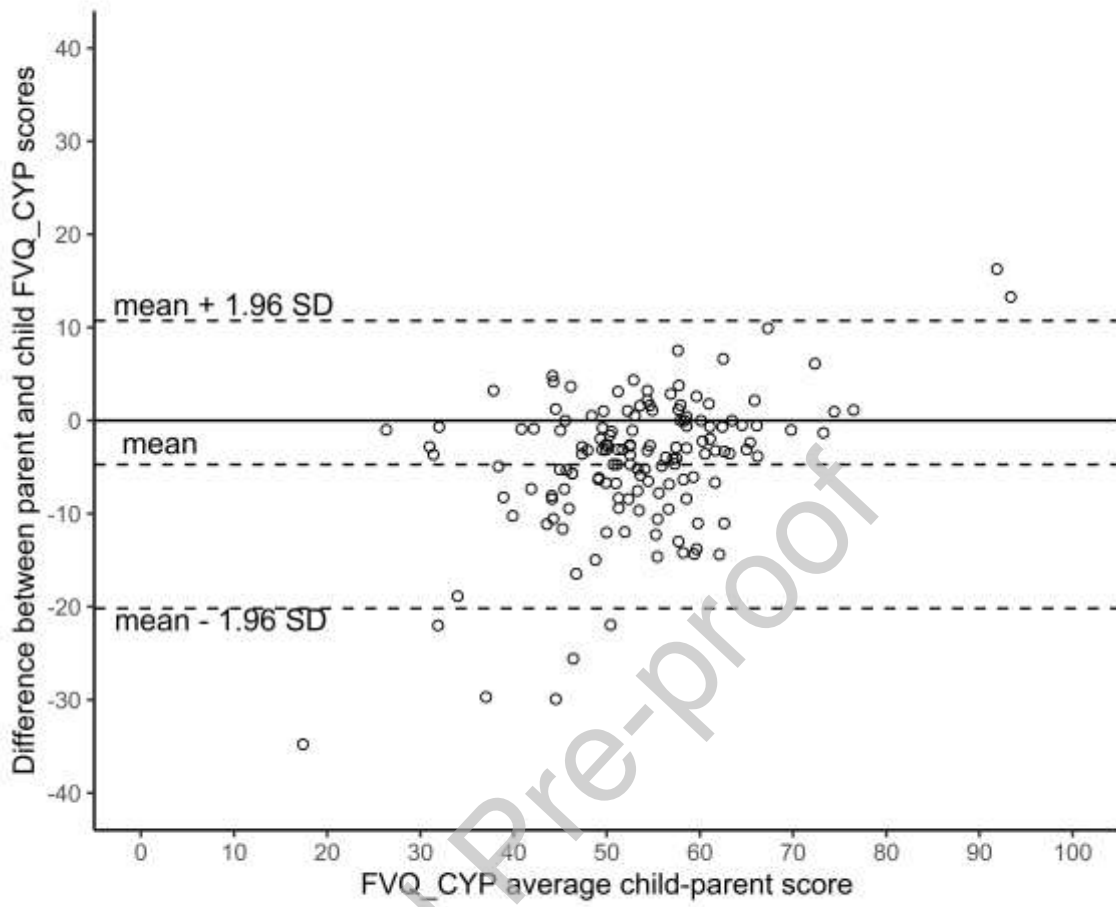**Table 3. Multiple logistic regression models for direction of disagreement on VQoL and FV between children and parents**

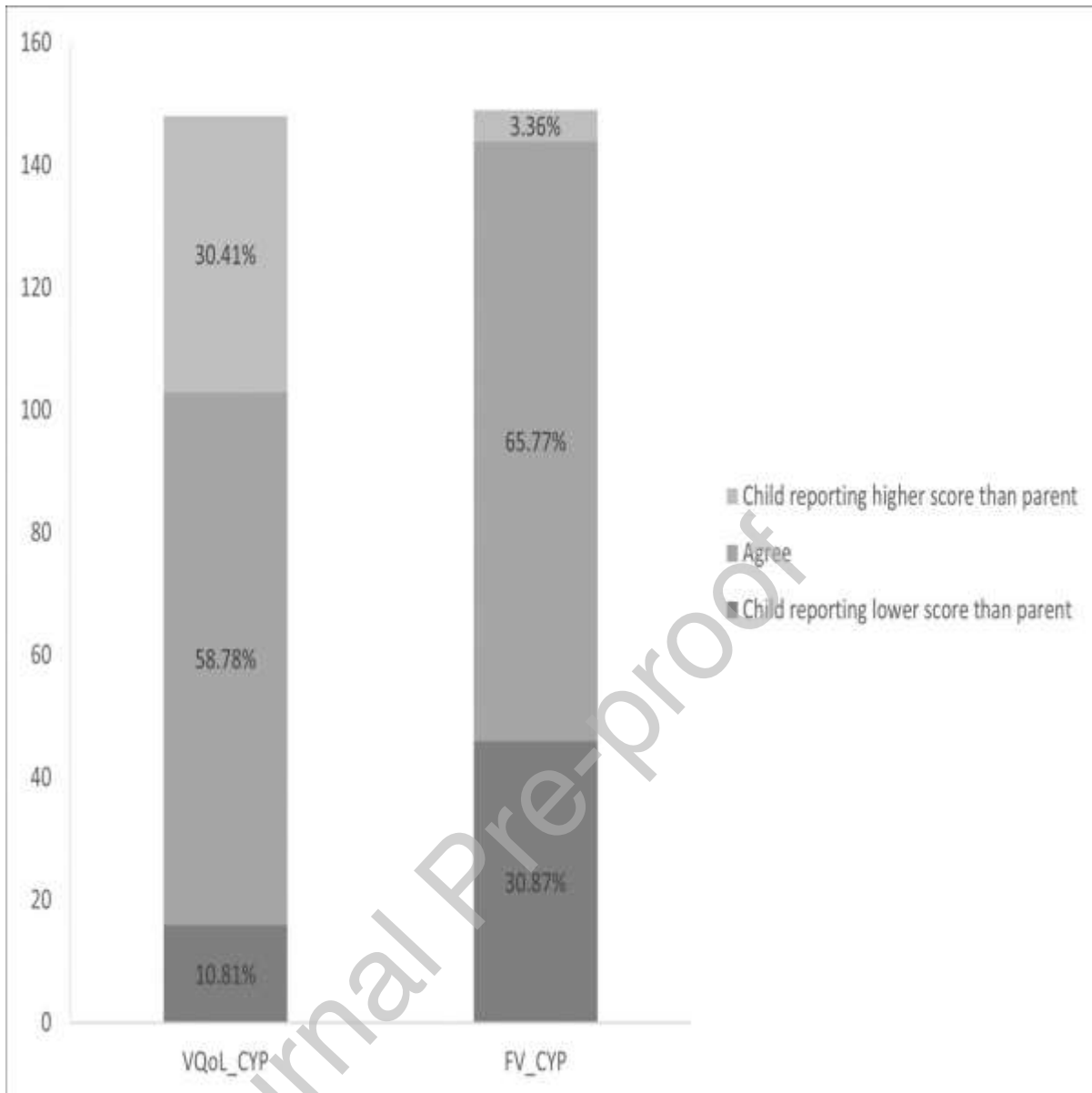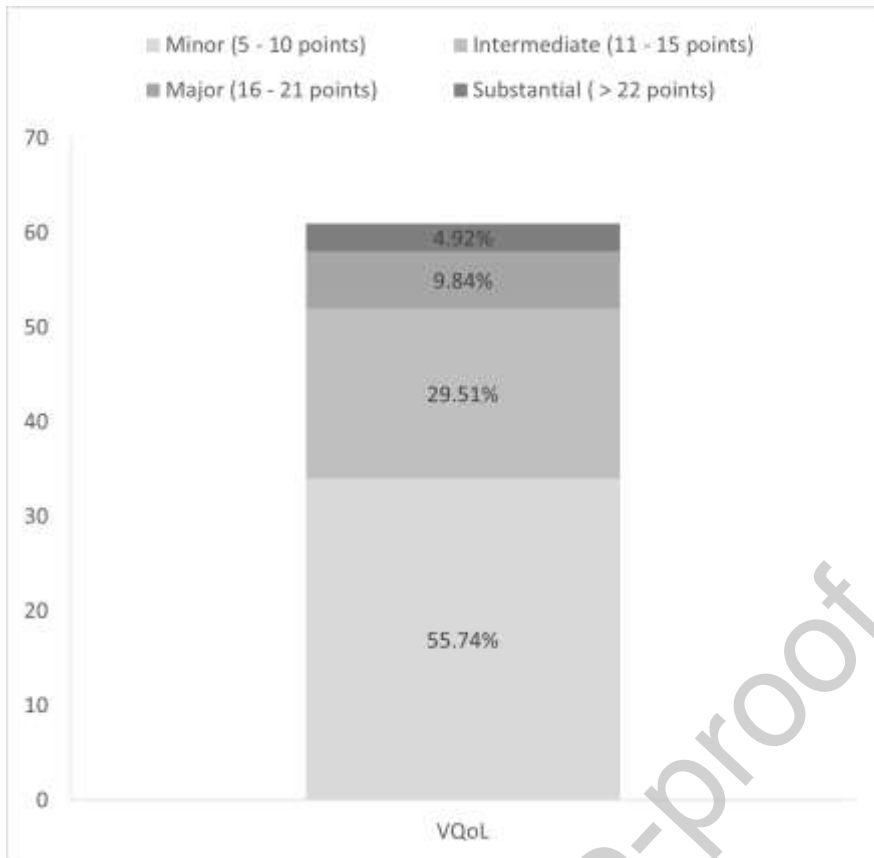| VQoL_CYP				Child LOW vs. Child HIGH/Agree*		Child HIGH vs. Child LOW/Agree**	
	Child LOW [*n*(%)]	AGREE [*n*(%)]	Child HIGH [*n*(%)]	OR (95% CI)	*p*-value	OR (95% CI)	*p*-value
Sex							
Male	12 (75)	45 (51.7)	26 (57.8)	0.38 (0.12 to 1.25)	0.111	0.97 (0.46 to 2.04)	0.940
Female	4 (25)	42 (48.3)	19 (42.2)				
Age (baseline = 7 years)							
7-12 years	10 (62.5)	51 (58.6)	17 (37.8)	0.93 (0.77 to 1.13)	0.462	1.18 (1.04 to 1.35)	0.013
13-17 years	6 (37.5)	36 (41.4)	28 (62.2)				
Severity of VI							
VI	13 (81.3)	73 (83.9)	32 (71.1)	0.96 (0.24 to 3.76)	0.947	1.65 (0.69 to 3.91)	0.260
SVI/BL	3 (18.8)	14 (16.1)	13 (28.9)				
Onset of VI							
Early	13 (81.3)	72 (82.8)	36 (80)	0.95 (0.19 to 4.67)	0.953	0.92 (0.31 to 2.74)	0.879
Late	3 (18.8)	15 (17.2)	9 (20)				
Course of VI							
Stable	10 (62.5)	64 (73.6)	32 (71.1)	1.52 (0.42 to 5.52)	0.523	1.31 (0.50 to 3.44)	0.586
Progressive	6 (37.5)	23 (26.4)	13 (28.9)				
FVQ_CYP*							
Sex							
Male	28 (60.9)	54 (55.1)	2 (40)	0.78 (0.37 to 1.62)	0.500		
Female	18 (39.1)	44 (44.9)	3 (60)				
Age (baseline = 7 years)							
7-12 years	30 (65.2)	49 (50)	0	0.91 (0.80 to 1.03)	0.118		
13-17 years	16 (34.8)	49 (50)	5 (100)				
Severity of VI							
VI	38 (82.6)	77 (78.6)	3 (60)	0.81 (0.32 to 2.07)	0.660		
SVI/BL	8 (17.4)	21 (21.4)	2 (40)				
Onset of VI							
Early	43 (93.5)	77 (78.6)	2 (40)	0.29 (0.07 to 1.16)	0.081		
Late	3 (6.5)	21 (21.4)	3 (60)				
Course of VI							
Stable	36 (78.3)	68 (69.4)	2 (40)	0.78 (0.30 to 1.99)	0.596		
Progressive	10 (21.7)	30 (30.6)	3 (60)				

[*] Nagelkerke's R^2 = 0.055
[**] Nagelkerke's R^2 = 0.089
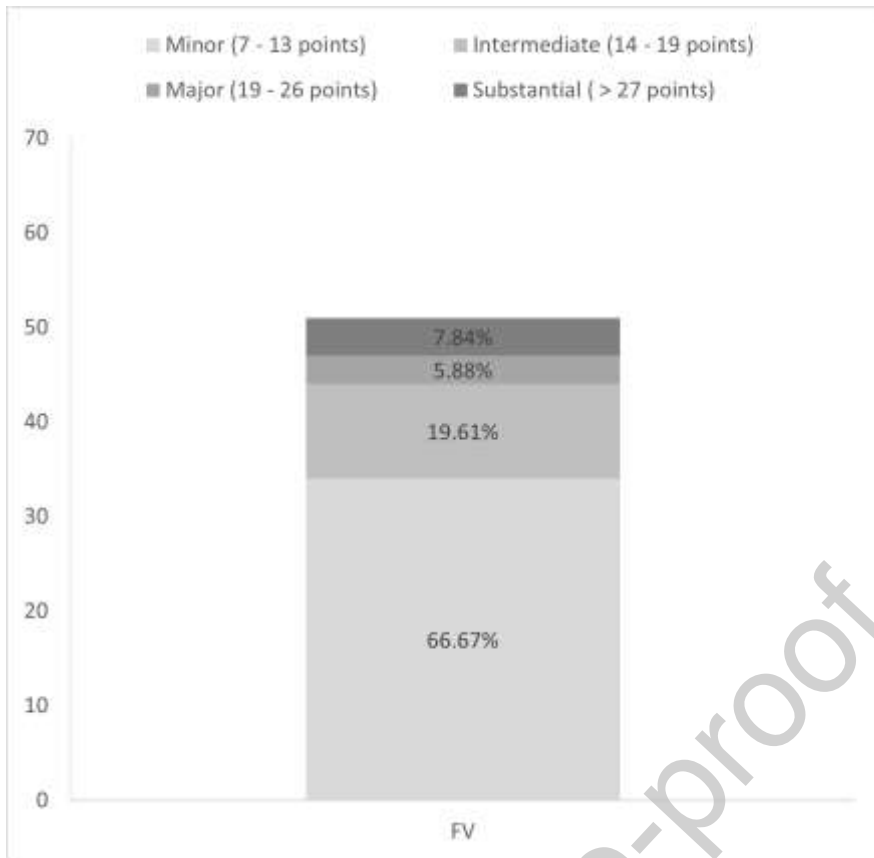[***] Nagelkerke's R^2 = 0.095. Binary logistic regression omitted from analysis of FVQ_CYP scores, due to distribution of sub-groups.