TITLE

Distribution and associations of vision-related quality of life (VQoL) and functional vision (FV) of children with visual impairment

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All authors met the ICMJE criteria for authorship:

AR contributed to the design of the study, and was accountable for data acquisition and interpretation, data analysis, preparation of the manuscript and final manuscript approval. LH-G and MC-B contributed to the data analysis and interpretation, and critical revision of the manuscript. JSR was accountable for the design of the study, data interpretation, and critical revision of the manuscript. All authors share accountability for all aspects of the work and have approved for the final version to be published.

CONFLICT OF INTEREST

No conflicting relationship exists for any author.

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SYNOPSIS

Self-rated vision-related quality of life of visually impaired children cannot be predicted using clinical characteristics. Self-rated functional vision complements clinical assessments. This study provides a reference for future interpretation of VQoL_CYP and FVQ_CYP scores.

ABSTRACT

Background:

Patient-reported outcome measures (PROMs) are increasingly used in paediatric ophthalmology. However, little is known about the distribution of PROM scores among children and young people with visual impairment.

Aim:

To investigate the distributions and predictors of scores on the VQoL_CYP (*measuring vision-related quality of life*) and FVQ_CYP (*measuring functional vision*).

Methods:

Children and young people aged 8 to 18 years, with visual impairment/blindness (logarithm of the minimum angle of resolution (LogMAR) worse than 0.48 in the better eye, and/or eligible visual field restriction) completed the VQoL_CYP and FVQ_CYP at home or Great Ormond Street Hospital, London, UK. Associations between VQoL_CYP and FVQ_CYP scores and socio-demographic and clinical factors were analysed using multiple linear regression models.

Results:

Among 93 participants, VQoL_CYP scores ranged from 36.55–78.16 (mean=57.86, SD=8.12). FVQ_CYP scores ranged from 23.52–70.29 (mean=48.32, SD=10.10). Only 0.4% of the variation in VQoL_CYP scores was explained, with no associations with the variables of interest. By contrast, 21.6% of the variation in FVQ_CYP scores was explained, with a gradient of worse acuity ($p<0.001$) and female gender ($p=0.04$) associated with worse self-rated functional vision. Age, ethnicity, time of onset and stability/progression of visual impairment were not associated.

Discussion:

Self-rated vision-related quality of life and functional vision are not readily predicted from socio-demographic or clinical characteristics that ophthalmologists measure/record. Routine use of PROMs in clinical practice can offer important insights. Use in research can provide valuable measures of effectiveness of interventions. The reference values provided will aid interpretation in both settings.

INTRODUCTION

Patient-reported outcome measures (PROMs) are described as the 'gold standard' for measuring patients' perspectives of the impact of a disease, impairment or disability, and any related treatment.[1, 2] The benefits of using PROMs are well-recognised by patients, their managing clinicians, and institutions and include improved clinician-patient communication,[3, 4] increased patient satisfaction,[4] meaningful comparisons of treatments,[5] and assessment of quality of healthcare.[6]

Recognising that children as young as 8 years can accurately reflect on their own health-related outcomes,[7] *generic* PROMs capturing health-related quality of life (HRQoL) have been developed for use in paediatrics.[8] As these are intended for use by all children/young people, and lack items (questions) specific to a particular impairment, they are developed with mixed populations and the 'normative' datasets include children both with and without any health conditions or disabilities.[9-11] Such normative datasets describe the range of scores for a given PROM that are expected in the absence of disease or impairment, and for comparisons between different patient groups for example by country, age or gender.

To address the lack of specific items included in *generic* PROMs, disorder/condition-specific PROMs have been developed for use *only* by those with the relevant impairment. As a result, it is conceptually inappropriate to use these instruments with individuals who do not have the relevant impairment, and the concept of normative datasets is not relevant. However, a 'reference' range of scores, derived from a representative population, affords the context for interpreting the scores for individual patients or comparing average scores for different groups of patients. Until recently,

in the area of childhood visual impairment (VI), there have been few psychometrically robust child vision PROMs. Consequently there is currently a lack of information about a) the distribution of the vision-related quality of life and functional vision of children and young people with all-cause VI, and b) variations between different groups of children.

The VQoL_CYP[12] and FVQ_CYP[13] are complementary psychometrically robust PROMs which capture respectively *vision-related quality of life* (VQoL) and *functional vision* (FV) in children and young people (*CYP*) aged from 8 up to 18 years. Two age-appropriate versions exist for each instrument, one for children (aged 8-12 years) and the other for young people (13 up to 18 years) but these have been designed to be used longitudinally, enabling scores from the different versions to be compared directly, or children of different ages to be included in the same analysis.

From a broader programme of research on feasibility of *routine* use of the VQoL_CYP and FVQ_CYP in paediatric ophthalmology practice, we report here the distribution of scores for each instrument as well as associations with potential predictive clinical and sociodemographic factors routinely recorded in clinical records, to inform future implementation of the VQoL_CYP and FVQ_CYP in clinical practice and research.

METHODS

This study was approved prospectively by the National Health Service Research Ethics Committee for UCL Great Ormond Street Institute of Child Health and Great Ormond Street Hospital, London, UK (REC reference: 17/LO/1484) and followed tenets of the Declaration of Helsinki. Participants aged >16 years gave individual

informed consent and those aged <16 years assented whilst their parents gave informed consent to participate.

Participants

All children and young people who were i) visually impaired (VI), severely visually impaired, or blind (corrected visual acuity in the better-seeing eye of logarithm of the minimum angle of resolution (logMAR)[14] 0.48 or worse, or logMAR 0.3 or worse with an NHS certification of VI or fluctuating acuity); ii) aged from 8 to 18 years; and iii) scheduled to attend a follow-up appointment at Great Ormond Street Hospital in the six month period between October 2018 and April 2019 were eligible for inclusion. In keeping with the population for whom the instruments were developed,[12, 13] and the fact the instruments are intended for self-completion, not for proxy (parent or clinician) assessment, children with significant additional impairments that impacted on the ability to self-report were not eligible for inclusion in the study.

Materials

The VQoL_CYP comprises 20 (child version) and 22 (young person version) self-report items asking about the experience of living with VI[12] e.g. *I feel different from other children/young people because of my eyesight.* The respondent indicates how true each statement is about their own life, using a 4-point Likert-type scale, ranging from *1. Not at all true*, to *4. Completely true*. The FVQ_CYP contains 28 (child version) and 38 (young person version) items about age-appropriate everyday activities requiring vision[13] e.g. *Because of my eyesight I find watching TV...*. Respondents indicate ease of completing the activity using a 4-point Likert-type scale, ranging from *1. Very easy,* to *4. Very difficult or impossible.* Possible scores in

both instruments range from 0 to 100, with a higher VQoL_CYP score indicating ***better*** VQoL, and a higher FVQ_CYP score indicating ***worse*** FV. An equating model was applied in the development of the VQoL_CYP[12] and FVQ_CYP[13] which allows scores from either age-appropriate version of the instruments to be compared, on the same measurement scale, despite variation in the number of, and wording of individual items. The instruments and user manuals are available (last accessed February 2021) from https://xip.uclb.com/i/healthcare_tools/VQoL_CYP_V2.html and https://xip.uclb.com/i/healthcare_tools/FVQ_CYP_V2.html.

Both a paper booklet and an electronic version of each age-version of both instruments were developed. The paper booklets contained both the VQoL_CYP and FVQ_CYP in large-print. The electronic format was presented using Qualtrics survey development software,[15] and resembled the paper format as closely as possible with regard to layout and presentation of individual items. The electronic version included quick and easy enabling of text-to-speech software. Both formats were tested for accessibility through consultations with a member of the clinical team who is visually impaired and has extensive expertise in adapting written material for children and young people with VI.

Procedure

All eligible children and young people (as described above) were sent an invitation to participate in the study one month before the date of their next scheduled hospital appointment in the Department of Ophthalmology at Great Ormond Street Hospital NHS Foundation Trust, London, UK.

Participants were invited to complete the VQoL_CYP and FVQ_CYP either at home or during their visit to the hospital for their appointment i.e. two 'real-world' PROM completion settings. Participants were given a choice of completion format and asked to complete the appropriate age-version of both instruments.

Data analysis

The instrument responses were entered into Excel and SPSS (version 25)[16] databases. Sociodemographic information included participants' age, gender, ethnicity, and socioeconomic status measured using quintiles of the index of multiple deprivation score, i.e. the conventional area-based index used in the UK (IMD[17]). Clinical characteristics included severity of visual impairment, timing of onset, and stability of vision. Based on the characteristics and size of the sample, ethnicity was re-coded as *White* versus *Any other ethnicity.* Severity of VI was analysed as a continuous variable and later categorised according to participants' better-seeing eye acuity (participants with *Severe VI (logMAR 1.02 – 1.3)* or *Blind (logMAR ≥ 1.32)* were combined. Additionally, for further exploration and confirmation, analysis of acuity in participants' better- and worse-seeing eye (univariable analyses only) was undertaken.

Cross-tabulations, and logistic regression models for participation and missing data were fitted to investigate associations with participants' sociodemographic and clinical characteristics. To aid interpretation, the age of the youngest participant (8 years) was used as the baseline. Participants with ≥ 20% missing responses on either instrument were excluded from the main analysis of that outcome. Remaining missing values (< 20% per participant) were imputed using the mean item score for the given responses of the participant.

Assumptions of data distributions were assessed using *z*-skewness and *z*-kurtosis values for normality, and Levene's tests for homogeneity of variances. Scores were stratified according to the sociodemographic and clinical characteristics (see above) considered, *a priori,* key 'predictors' of VQoL or FV. Independent *t*-tests, Spearman's rank and Pearson's correlations were used to examine whether there were any associations between VQoL_CYP and FVQ_CYP scores and these sociodemographic and clinical characteristics.

Multiple linear regression models, i.e. adjusted for all factors, were used to investigate associations with scores for each instrument. Dummy variables were created for categorical variables containing more than two groups (i.e. socio-economic status and severity of VI), using the following categories as baseline: SES: *5: least deprived,* Severity of VI: *Low vision (logMAR ≤ 0.46*). Dichotomous variables were coded as 0 (*Male, White, Early ≤ 2 years, and Stable*) or 1 (*Female, Any other ethnicity, 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 and Nagelkerke's R^2 for linear and logistic regression models. All significance tests were carried out at the 0.05 level.

RESULTS

In total, 93 children and young people participated, comprising 48% of all those invited. The participant sample did not differ significantly from the eligible non-participating sample with respect to key predictors (i.e. age, ethnicity, socio-economic status, and severity of VI), however there was an over-representation of girls (p = 0.045) (e-Table 1). The sociodemographic and clinical characteristics of the sample who completed the VQoL_CYP and FVQ_CYP are shown in Table 1 and

Table 2, respectively. All participants self-reported that they were able to complete the instruments, either with or without help from a parent/caregiver.

Table 1. Vision-related quality of life (VQoL_CYP) scores stratified by sociodemographic and clinical characteristics.

VQoL_CYP	*n*	Mean (M)	Standard deviation (SD)	Minimum	Maximum	*p*-value
All participants	84[a.]	57.9	8.1	36.6	78.2	
Gender						
Male	41	57.9	8.4	40.0	78.2	0.977[c.]
Female	43	57.8	7.9	36.6	75.6	
Age						
8	8	56.6	7.8	42.7	68.1	0.069[d.]
9	9	60.7	4.4	52.2	69.5	
10	18	58.7	7.8	44.9	71.2	
11	11	57.9	9.9	44.9	78.2	
12	5	65.1	4.6	61.7	73.0	
13	10	59.6	10.6	40.0	75.6	
14	11	54.7	6.9	43.1	64.4	
15	8	55.2	5.2	43.7	59.5	
16	3	55.1	6.5	49.7	62.3	
17	1	36.6	-	36.6	36.6	
Ethnicity						
White UK	45	59.3	7.8	40.0	78.2	0.085[c.]
Any other	39	56.2	8.3	36.6	71.4	
Socioeconomic status (index of multiple deprivation quintiles)[b.]						
1: most deprived	13	56.0	8.9	40.0	68.1	0.078[e.]
2	20	57.0	7.6	43.1	71.4	
3	14	57.0	10.4	36.6	73.0	
4	16	58.8	6.2	45.6	71.2	
5: least deprived	19	61.0	7.3	51.5	78.2	
Severity of visual impairment (latest assessment of acuity in the better-seeing eye)						
Low vision (logMAR ≤ 0.46)	35	58.1	7.6	42.7	75.6	0.559[e, f, g.]
Visual impairment (logMAR 0.48 – 0.7)	21	57.8	8.4	40.0	71.4	
Visual impairment (logMAR 0.72 – 1.00)	18	58.7	8.7	43.7	78.2	
Severe visual impairment/Blind (logMAR ≥ 1.02)	10	55.6	9.1	36.6	71.2	
Timing of onset of visual impairment						
Early (≤ 2 years)	72	57.6	7.5	40.0	78.2	0.456[c.]
Late	12	59.5	11.3	36.6	75.6	
Stability of vision						
Stable	59	58.5	7.6	40.0	78.2	0.298[c.]
Progressive	25	56.4	9.3	36.6	75.6	

[a.] 9 participants excluded with ≥ 20% missing data. Seven (78%) were male, 6 (67%) were of White UK ethnicity, 5 (56%) were from the 4th multiple deprivation quintile, and they were aged 8-15 years. Four (44%) had low vision, 9 (100%) had early onset VI and 7 (78%) had non-progressive VI.

[b.] 2 participants with missing index of multiple deprivation.

[c.] Independent samples *t*-test

[d.] Pearson's correlation

[e.] Spearman's rank correlation

[f.] Spearman's rank correlation for better-seeing eye LogMAR (continuous): $p = 0.257$

[g.] Spearman's rank correlation for worse-seeing eye LogMAR (continuous): $p = 0.441$

Table 2. Functional vision (FVQ_CYP) scores stratified by sociodemographic and clinical characteristics.

FVQ_CYP	*n*	Mean (M)	Standard deviation (SD)	Minimum	Maximum	*p*-value
All participants	83[a.]	48.3	10.1	23.5	70.3	
Gender						
Male	42	46.3	10.7	23.5	67.4	0.064[c.]
Female	41	50.4	9.1	28.6	70.3	
Age						
8	8	49.8	6.2	40.0	57.1	0.64[d.]
9	9	40.6	8.6	24.6	52.8	
10	19	50.3	10.3	31.5	70.3	
11	12	50.4	8.5	36.2	68.3	
12	5	49.8	11.1	42.9	69.3	
13	9	45.7	12.1	26.8	65.3	
14	10	46.3	11.1	23.5	63.3	
15	8	52.7	8.3	38.0	66.4	
16	2	40.4	17.9	27.8	53.1	
17	1	60.9	-	60.9	60.9	
Ethnicity						
White UK	46	47.8	9.4	24.6	68.3	0.578[c.]
Any other	37	49.0	11.0	23.5	70.3	
Socioeconomic status (index of multiple deprivation quintiles)[b.]						
1: most deprived	11	53.5	10.8	40.0	67.4	0.159[e.]
2	17	48.0	5.9	38.0	58.8	
3	13	48.7	10.1	26.8	60.9	
4	21	47.1	12.8	23.5	70.3	
5: least deprived	19	46.5	9.8	28.6	69.3	
Severity of visual impairment (latest assessment of acuity in the better-seeing eye)						
Low vision (logMAR ≤ 0.46)	33	44.1	9.6	23.5	63.3	**0.000[e, f, g.]**
Visual impairment (logMAR 0.48 – 0.7)	22	47.2	9.2	27.8	70.3	
Visual impairment (logMAR 0.72 – 1.00)	19	52.6	6.9	41.2	67.4	
Severe visual impairment/Blind (logMAR ≥ 1.02)	9	57.7	11.6	31.5	69.3	
Timing of onset of visual impairment						
Early (≤ 2 years)	73	48.5	10.1	23.5	70.3	0.657[c.]
Late	10	47.0	10.5	28.6	60.9	
Stability of vision						
Stable	60	47.6	10.3	23.5	70.3	0.279[c.]
Progressive	23	50.3	9.6	28.6	68.3	

[a.] 10 participants excluded with ≥ 20% missing data. Six (60%) were male, 5 (50%) were of White UK ethnicity, 6 (60%) were from either the 1st or 2nd multiple deprivation quintile, and they were aged 8 to 16 years. Six (60%) had low vision, 8 (80%) had early onset VI, and 6 (60%) had non-progressive VI.

[b.] 2 participants with missing index of multiple deprivation

[c.] Independent samples *t*-test

[d.] Pearson's correlation

[e.] Spearman's rank correlation

[f.] Spearman's rank correlation for better-seeing eye LogMAR (continuous): $p = 0.000$

[g.] Spearman's rank correlation for worse-seeing eye LogMAR (continuous): $p = 0.002$

The data for 9 participants were excluded from the analysis of VQoL_CYP scores and for 10 participants were excluded from analyses of FVQ_CYP scores based on the standard threshold of ≥ 20% missing responses, as shown in e-Table 2 and e-Table 3. The proportion of children and young people with < 20% missing data was 15.5% in the VQoL_CYP dataset and 36.1% in the FVQ_CYP dataset (see e-Table 2 and e-Table 3). No characteristics were significantly associated with < 20% missing data in the VQoL_CYP and FVQ_CYP (e-Table 4), except that participants with VI classified (logMAR 0.48 – 0.7 in the better-seeing eye) were less likely than those with low vision (logMAR 0.46 or better) to have missing data in the FVQ_CYP.

Z-skewness and *z*-kurtosis for VQoL_CYP scores were -0.39 and 0.30 respectively, and -0.73 and 0.43 for FVQ scores, indicating normality. Levene's tests indicated homogeneity of variances across VQoL_CYP scores (p = 0.170 to 0.936) and across FVQ_CYP scores (p = 0.099 to 0.832) for comparisons of all subgroups.

Vision-related quality of life (VQoL_CYP):

The mean VQoL_CYP score (higher score indicates better quality of life) was 57.86 (SD = 8.12) in the total sample (Table 1). Univariable analyses provided no evidence that the distribution of VQoL_CYP scores varied by any of the key characteristics. As shown in Table 3, a non-significant regression equation was found ($F(12, 69) = 1.03$, p = 0.435), with an adjusted R^2 indicating that 0.4% of the variance in VQoL_CYP scores can be explained by participants' characteristics. Unstandardized coefficients revealed that participants with late onset VI scored 6.4 (95% CI of 0.0 to 12.8) points higher (i.e. reported better quality of life) than those with early onset VI. Participants with VI which was progressive scored 4.4 (-0.3 to 9.2) points lower (i.e. reported

worse quality of life) than those with VI which was stable but neither association was significant at the specified level.

Table 3. Multiple linear regression models for change in VQoL_CYP and FVQ_CYP scores.

	VQoL_CYP*		FVQ_CYP**	
	Unstandardized coefficient (95% CI)	*p*-value	Unstandardized coefficient (95% CI)	*p*-value
Constant	63.69 (52.86 to 74.53)	<0.001	39.42 (27.26 to 51.58)	<0.001
Sociodemographic characteristics				
Age (baseline = *8 years*)	-0.44 (-1.23 to 0.35)	0.274	-0.02 (-0.94 to 0.90)	0.969
Gender (baseline = *Male*)	-0.50 (-4.24 to 3.23)	0.789	4.35 (0.17 to 8.53)	0.042
Ethnicity (baseline = *White*)	-1.62 (-5.54 to 2.29)	0.411	0.58 (-3.99 to 5.16)	0.800
Socio-economic status (index of multiple deprivation quintiles) (baseline = *5: least deprived*)				
1: most deprived	-4.42 (-10.45 to 1.62)	0.149	6.39 (-0.74 to 13.51)	0.078
2	-2.47 (-8.05 to 3.11)	0.380	1.16 (-5.40 to 7.73)	0.724
3	-3.49 (-9.36 to 2.38)	0.239	1.33 (-5.35 to 8.00)	0.693
4	-1.09 (-6.81 to 4.64)	0.706	0.30 (-5.65 to 6.25)	0.920
Clinical characteristics				
Severity of visual impairment (latest assessment of acuity in the better-seeing eye) (baseline = *Low vision (logMAR ≤ 0.46*)				
Visual impairment (logMAR 0.48 – 0.7)	-0.76 (-5.50 to 3.98)	0.750	3.71 (-1.57 to 8.99)	0.166
Visual impairment (logMAR 0.72 – 1.00)	0.58 (-4.27 to 5.44)	0.812	9.15 (3.76 to 14.55)	0.001
Severe visual impairment/ Blind (logMAR ≥ 1.02)	-2.12 (-8.13 to 3.88)	0.483	13.11 (6.06 to 20.15)	<0.001
Timing of onset of visual impairment (baseline = *Early ≤ 2 years*)	6.40 (-0.04 to 12.85)	0.051	-7.78 (-15.82 to 0.26)	0.058
Stability of vision (baseline = *Stable*)	-4.44 (-9.24 to 0.35)	0.069	3.28 (-2.42 to 8.98)	0.255

* Adjusted R^2 = .004, higher score = better outcome

** Adjusted R^2 = .216, higher score = worse outcome

Functional vision (FVQ_CYP):

The mean FVQ_CYP score (lower score indicates better functional vision) was 48.3 (SD = 10.1) in the total sample (Table 2). There was a gradient of higher (i.e. worse) self-reported FV with increasing severity of VI (r_s (81) = 0.46, p = <0.001). Multiple regression analysis and an adjusted R^2 showed 21.6% of the variance in FVQ_CYP scores could be explained by participants' characteristics (F(12, 68) = 2.834, p = 0.003). In this model, females scored 4.4 (0.2 to 8.5) points higher (i.e. reported worse FV) than males (p = 0.042). Participants with VI classified as logMAR 0.72 – 1.00 scored 9.2 (3.8 to 14.5) points higher on the FVQ_CYP, and those with the most severe VI (logMAR ≥ 1.02) scored 13.1 (6.1 to 20.2) points higher (i.e. both reported significantly worse FV than those with low vision (logMAR ≤ 0.48), p ≤ 0.001). Visual acuity alone explained 21% of variance in FVQ_CYP scores. There was some indication of an association between FVQ_CYP scores and timing of onset of VI, as participants with late-onset VI scored 7.8 (-0.3 to 15.8) points lower (i.e. reported better FV) than those with early onset VI, though it did not reach statistical significance (p = 0.058).

DISCUSSION

From a study of the target population of children and young people with all-cause visual disability for whom the VQoL and FVQ instruments are intended, we report a Gaussian distribution of scores for each instrument, with children and young people utilising a wide range of the full measurement scale in both instruments. The mean VQoL_CYP score was 7.9 points higher than the midpoint of the range (higher scores signify better VQoL) and the mean FVQ score was 1.7 points lower than the midpoint of the range (higher scores signify worse FV) in this sample. None of the

key sociodemographic or clinical characteristics investigated were found to be associated with VQoL scores ($p \leq 0.05$), and a multiple linear regression model predicted only 0.4% of the variance in the full dataset. By contrast, FVQ_CYP scores were associated with severity of VI and gender.

One strength of this study is the setting of routine PROM administration in clinical practice. To enable a 'real world' assessment and achieve a study sample of children and young people with visual impairment for whom the instruments have been developed, we deliberately embedded recruitment and implementation into routine clinical practice and therefore the schedule of existing clinical appointments. We report elsewhere the feasibility of administering PROMs in two different settings and using two different formats (i.e. an important design feature of this study). There was a high participation rate in comparison with similar research with the same clinical population [12, 19, 20] demonstrating, in part, the willingness of children and young people with visual impairment to use the VQoL_CYP and FVQ_CYP in 'real life' settings.

Nevertheless the sample size, although large for studies of childhood VI,[18] was modest in comparison to studies of whole child populations.[9, 11] A formal power calculation was not possible given the lack of prior research in this area, so it is not possible to assess accurately if the study had limited power to identify any true associations. In keeping with best practice, we excluded data from participants with ≥ 20% missing VQoL_CYP and/or FVQ_CYP data since scores containing less than 80% data would be unreliable and skew the measurement construct. We imputed remaining missing data using individual mean imputation which may lead to limited false increased precision with slightly less variation in the constructs. Since the VQoL_CYP and FVQ_CYP instruments are not intended for proxy completion

(parents or clinicians) but rather self-assessment and self-reporting by affected children and young people, and they capture vision-related (rather than generic health-related) issues, our study sample necessarily did not include children with significant additional impairments where these would have precluded self-assessment e.g. significant communication or learning impairment. Thus, our findings are not applicable to children with VI who would be unable to self-assess and self-report.

There are no similar studies that have reported a 'reference' dataset for specific instruments and thus no studies with which we can directly compare our findings. However, the relevance of our findings can be considered in the context of broader literature in child health and paediatrics. The 'disability paradox'[21] is a well-established concept outside ophthalmology. Our findings serve as empirical evidence of this phenomenon in ophthalmology, amplifying findings we previously reported during the development of the VQoL_CYP and FV_CYP.[12, 13] It can be challenging for ophthalmic clinicians to understand how a child or young person with significantly impaired vision might report very high VQoL but our data show that this, and the reverse relationship, are not infrequent i.e. severity of VI does not predict VQoL. Equally, our findings show that VQoL cannot be predicted by other key clinical or sociodemographic factors that may be recorded in ophthalmic practice.

The consequences of incorrectly assuming a relationship between subjective vision-related well-being and visual function are as important to clinical practice as they are to research. For example, important influences on patients' well-being will be overlooked through sole reliance on clinical measures of visual function. Use of a PROM that directly measures well-being is therefore essential for an accurate representation of 'unobservable' outcomes.

Our finding that FVQ_CYP scores were positively correlated with severity of visual impairment is to be expected, given the nature of FVQ_CYP items. The finding that girls reported worse FV is interesting, unexplained and warrants further investigation as there is scant research on gender differences in the daily functional impact of VI. The FVQ_CYP was developed to allow understanding of how and to what degree VI impacts on activities in everyday contexts which are also influenced by issues such as accessibility, and appropriate support. This, in turn, provides granularity and affords a deeper level of understanding of function outside of clinical settings i.e. a more holistic view of functional impact, which is of value in understanding whether and to what extent treatment or other aspects of care improve functioning. It is possible that the gender difference we found, reflect a broader context in which girls with VI have reported lower overall confidence[22] and self-esteem[23] than boys, regarding physical functioning, and place greater value on social means of functional support.[24]

Whilst an association with timing of onset, suggesting that children and young people with late-onset VI have better FV and better VQoL than those with early onset (≤ 2 years), did not reach conventional thresholds of ‘statistical significance’, it is interesting to consider whether the global delay in developmental milestones among children diagnosed with VI during early childhood,[25] may manifest in impaired functional vision. Equally, the association between late-onset VI and VQoL, may benefit from further consideration, given the broader literature on disability documents that acceptance or/and adaptation to late-onset disability takes time and effort.[26, 27]

Our models, assessing the key sociodemographic and clinical characteristics generally measured in ophthalmic practice, predicted 0.4% of the variation in VQoL

scores and 21.6% of the variation in FVQ_CYP scores. These findings indicate the need for primary research, which was outside the scope of our study, to investigate specifically, what shapes these outcomes, with an overarching aim to develop interventions which promote VQoL and FV among children and young people at greater risk of adverse outcomes.

Our study, using the VQoL_CYP and FVQ_CYP instruments completed in a real world setting, demonstrates that both VQoL and FV vary widely among children and young people with all-cause VI and cannot be predicted from the child/young person's sociodemographic or clinical profile. Routine use of these complementary PROMs in clinical practice can provide critical insights for clinicians when evaluating impact of care, and their use in research can provide new insights into effectiveness of treatments. Our findings provide a useful reference for future use of these instruments in the population for whom they are intended.

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