Running head: Functional vision of children and young people

Title: Development of the Functional Vision Questionnaire for Children and Young People with visual impairment – the FVQ_CYP

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Key words: Functional vision; visual impairment; child; questionnaire; instrument; child-centered approach; self-report; interviews.

Abbreviations: FV – functional vision, VF – visual function, QoL – quality of life; VQoL – vision-related quality of life; CYP – children and young people, LogMAR – the Logarithm of the Minimum Angle of Resolution, VI – Visually Impaired, BL – Blind

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The following material should appear online only: Figure 1, Figure 2 and Figure 3.

ABSTRACT

Objective: To develop a novel age-appropriate measure of Functional Vision (FV) for self-reporting by visually impaired (VI) children and young people. ***Design:*** Questionnaire development. ***Participants:*** A representative patient sample of VI children and young people aged 10-15 years, visual acuity of LogMAR (the Logarithm of the Minimum Angle of Resolution) worse than .48, and a school-based (non-random) expert group sample of VI pupils aged 12-17 years. ***Methods:*** 32 qualitative semi-structured interviews supplemented by narrative feedback from 15 eligible VI children and young people were used to generate draft instrument items. Seventeen VI pupils were consulted individually on item relevance and comprehensibility, instrument instructions, format and administration methods. The resulting draft instrument was piloted with 101 VI children and young people comprising a nationally representative sample, drawn from 21 hospitals in United Kingdom. Initial item reduction was informed by presence of missing data and individual item response pattern. Exploratory Factor Analysis (FA) and Parallel Analysis (PA), and Rasch Analysis (RA) were applied to test the instrument's psychometric properties. ***Main outcome measures:*** Psychometric indices and validity assessment of the FVQ-CYP instrument. ***Results:*** 712 qualitative statements became a 56-item draft scale, capturing the level of difficulty in performing vision-dependent activities. Following piloting, items were removed iteratively as follows: 11 for high percentage of missing data, 4 for skewness, 1 for inadequate item infit and outfit values in RA, 3 having shown differential item functioning across age groups and one across gender in RA. The remaining 36 items showed item fit values within acceptable limits, good measurement precision and targeting, and ordered response categories. The reduced scale has a clear unidimensional structure, with all items having a high factor loading on the single factor in FA and PA. The summary scores correlated significantly with visual acuity. ***Conclusions:*** We have developed a novel, psychometrically robust self-report questionnaire for children and young people - the FVQ_CYP - that captures the functional impact of visual disability from their perspective. The 36-item, 4-point unidimensional scale has potential as a

complementary adjunct to objective clinical assessments in routine pediatric ophthalmology practice as well as in research.

INTRODUCTION

Capturing patients' perspectives of their health outcomes is key to patient-centered health care and is a high priority of health services internationally. [1-3] Accordingly, the development and application of patient-reported outcome measures (PROMs) is increasingly advocated for monitoring a range of diverse and distinct outcomes, including health-related quality of life, functional status and symptom severity.[4, 5] However, there has been limited progress in development and application of such measures in pediatric ophthalmology.

In response to the need for age-appropriate self-report PROMs in pediatric ophthalmology, we have recently developed an instrument to assess self-reported vision-related quality of life (VQoL) of children and young people (for brevity, we refer to children and young people as children in the remainder of the paper) with visual impairment (VI) aged 10-15 years, based on in-depth individual interviews with VI children (the VQoL_CYP instrument[6]). This research and its conceptual framework, based on the extant literature, indicated that a separate measure of functional vision (FV) in pediatric ophthalmology was needed. Whilst our VQoL_CYP instrument was designed to capture the child's view of their position in life in their societal context (e.g. social and emotional impact of living with a visual disability) the FV measure would provide a means for assessing the child's self-reported ability to complete tasks for which vision is required. As such, it would complement objective clinical measures of visual function (VF) such as acuity (for example, it would serve as a tool for evaluating the effectiveness of treatment or low vision rehabilitation) as well as to the VQoL_CYP instrument.

To develop our novel FV instrument we used the qualitative data from our VQoL research programme that described children's own perspectives of what it was like to live with a visual impairment. This was to ensure a robust child-centered method of achieving content validity, as well as direct complementarity of the two instruments by virtue of being grounded in the same population of children.

Here, we report development and piloting of our novel FV instrument, the Functional Vision Questionnaire for Children and Young People (FVQ_CYP). It is designed to capture the self-reported level of difficulty of performing vision-dependent activities and intended for children with a visual impairment, severe visual impairment or blindness (i.e. acuity of Logarithm of the Minimum Angle of Resolution, LogMAR, of worse than 0.48 in better eye).

METHOD

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

Development of the FVQ_CYP followed 3 distinct phases. Item generation for the instrument (Phase 1) was based on the rich qualitative interview data, followed by pre-testing (Phase-2) and piloting of the FVQ_CYP (Phase 3) with VI children. Participating children gave informed individual assent and parents gave informed consent to their child's participation.

Subject identification and recruitment

Children were eligible if *i)* they were visually impaired, severely visually impaired or blind (VI/BL - for brevity, we consider term VI in the remainder of the paper) [visual acuity (VA) in the better eye Snellen worse than 6/18 or of LogMAR worse than 0.48) 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. They were drawn from 3 sources across the 3 phases of the study as follows:

Source 1. Databases of eligible patients attending Department of Ophthalmology at GOSH, and the Pediatric Glaucoma Service and Genetic Eye Disease Service at Moorfields Eye Hospital, London UK were used for recruitment in Phase 1 and Phase 3. In Phase 1, 32 eligible patients (invited using a stratified sampling approach) participated in qualitative interviews and a

further 15 provided additional qualitative feedback about how VI affected them, as part of the VQoL questionnaire completion in the VQoL programme. In Phase 3, 52 patients participated in the postal survey to pilot the FVQ_CYP.

Source 2. 17 available children aged 12-17 years with VI from 2 specialist schools in England, UK for pupils with VI participated in Phase 2, as an 'expert user reference group'.

Source 3. Forty nine patients attending 19 additional hospitals across England and Wales, UK participated in Phase 3 through a postal survey to pilot the FVQ_CYP.

Procedures

Phase 1: Item generation. The content for the FVQ_CYP was derived from qualitative semi-structured interviews with 32 children, supplemented by qualitative narrative feedback from an additional 15 children who participated in our parallel programme (developing and piloting our VQoL_CYP instrument, as described elsewhere[6]).

Questionnaire items were developed by deriving a qualitative data category (using the '*Functioning: home, school and leisure'* theme from our VQoL_CYP instrument[6] as a conceptual starting point) to group qualitative statements relating to general activities, VI-related activities (e.g. adapted sports and technologies), level of functioning, restrictions and limitations in activities and mobility (Figure 1, available at http://aaojournal.org). Two researchers independently coded 3 interviews using NVivo9 software,[7, 8] grouping together all FV relevant statements. As agreement was high, the interviews were coded independently to form a general 'Functional Vision NVivo Category/Node' comprising 712 statements. These statements were reviewed independently by two other study team members (one an ophthalmic services user for herself and her child) who rated all the statements pertaining to a particular activity to be a potential item for the FVQ_CYP. Comparison of independent ratings by 4 researchers resulted in an agreed item pool. Iterative item reduction using a Delphi expert consensus[9] involved five researchers judging relevance and importance of items.

Statements relating to use of adaptive technologies and assistive devices to maximise FV were identified and used to form a short questionnaire (applied before proceeding to FVQ_CYP completion) to ascertain qualitative information about their use by participants as context for subsequent reports of FV (Figure 2, available at http://aaojournal.org). The visual function domains (e.g. near and distance vision, contrast sensitivity) that may affect FV were also considered. Any statements relating to feelings, social impact, sense of independence and autonomy were excluded from the qualitative FV category as inappropriate, having already been included in item generation for our VQoL_CYP instrument. Statements relating to symptoms (e.g. tired eyes, headaches) were also excluded, in keeping with the literature.[10]

Phase 2: Pre-testing. The draft instrument was evaluated by the expert reference group of 17 pupils with VI (Source 2) who were consulted individually to a) gauge importance, relevance and comprehensibility of the items, b) to review the scale response options, c) to assess the children's understanding of instructions for the instrument and d) to evaluate the need for and understanding of the scale time frame.

Phase 3: Piloting. This was undertaken as a postal survey of a nationally representative (UK) sample of 101 children with VI. The study pack included an invitation letter, information sheet for children and parents, consent and assent forms and large print and electronic versions (on a CD) of the draft instrument, together with postage prepaid envelopes for return of completed documents.

Double data entry (Excel database) of 16% of questionnaires enabled errors to be identified and corrected. The remaining data were entered and checked independently by two researchers.

Formal psychometric item reduction was guided by a) presence of missing data and individual item response pattern, and b) application of exploratory Factor Analysis (FA), including Parallel Analysis (PA)[11], and then Rasch Analysis (RA).[12] FA and PA determine the number of common factors (i.e. underlying latent traits) accounting for the magnitude of

observed correlations between the items and were applied to examine the factor structure of the initial item pool of the FVQ_CYP (i.e. the item loadings on and the variance are explained by the first factor) and thus assess whether the items fit a single underlying construct. RA is a modern psychometric approach which transforms ordinal questionnaire data into an interval scale, allowing derivation of a scale summary score (as extensively described previously[13-16]).

The Rasch Rating Scale Model (RSM)[17] was applied to assess: a) *item fit* (which confirms that the summary score produced by the items in the scale represent a single underlying construct that is FV, substantiating the scale *unidimensionality* and supplementing the results of FA and PA) by examining item infit and outfit statistics, which indicate how well the items fit the underlying construct and which are measured as mean square standardized residuals (MNSQ), with the 0.5-1.5 range being considered acceptable for productive measurement.[18]; b) *differential item functioning (DIF)* (which shows whether subgroups of children with the same latent trait or 'ability' respond differently to items), by stratifying the participants by age group and gender and used the cut off of > 1.0 logit for identifying notable DIF.[19]; c) *response scale ordering*, by examining Rasch category probability curves, to demonstrate the likelihood of each response category on our 4-point scale being selected over the range of the scale. For good ordering the category thresholds should increase by at least 1.4 logits.[20]; d) *targeting*, by examining the item-person map, illustrating a relative position of item difficulty to person ability. For good targeting, the difference of person and item means of up to 1 logit is acceptable.[16]; and e) *measurement precision* (the ability of the instrument to discriminate between different groups of respondents on the measured variable), by observing the person separation index and reliability (≥2.00 and >.80 the minimum accepted levels respectively).[15]

To assess *construct validity* of the FVQ_CYP (i.e. whether the instrument measures what is intended to measure), Pearson correlation coefficients were calculated between FVQ_CYP summary score and the objectively measured visual acuity. A correlation coefficient between 0.3

and 0.9 is considered acceptable[15]. In order to derive summary scores for these analyses, missing item data was imputed using multiple-pattern regression imputation.

For the exploratory FA and PA the data were analysed using MPlus software (version 7)[21] and for Rasch into 'Winsteps' software (version 3.75.0).[22] The remaining analyses were completed using the SPSS (version 21).[23]

RESULTS

Phase 1

The list of 712 function-relevant statements was systematically reduced to a draft 58-item questionnaire, with some specific features. Firstly, to be contextually meaningful to children, the items were organised into 4 activity categories i.e., Home, School, Sports and Leisure, and Mobility, each introduced by the statement: '*We want to find out how your eyesight affects your activities at [activity category e.g., home]'*. This contrasts with other adult and child instruments organised by VF domains (e.g. Near Vision vs. Distance Vision), which we accounted for during item generation (example items: '*telling the time on a wrist watch'* vs. *'telling the time on a wall clock*'). Secondly, a positive psychology approach was adopted, asking children to report firstly, level of 'ease' before considering the level of 'difficulty' in completing a particular task. Thus, for each activity category the child is asked to consider their optimal visual function (i.e. *'With the best lighting and contrast for you, and with your glasses, low vision aids or other devices, if you use them for these activities'*) and a single question stem (i.e. '*How easy do you find…..?*'), followed by a list of items (e.g., *'Watching TV', 'Using the computer for homework'*). The responding child reports their level of functioning using 5 response options (*1: Very Easy; 2: Easy; 3: Difficult; 4: Very difficult or impossible; and 5: This doesn't apply to me/I don't do this for other reasons*). A higher score indicates greater FV difficulty (excluding the unscorable category 5).

Phase 2

The individual expert user reference consultations took 20 minutes on average. Two items were removed and some were re-phrased for clarity using the children's own language. The time frame reference of 1 month (as in other pediatric questionnaires) was tested and abandoned as children found it confusing and reported reflecting on the last occasion they remembered completing the activity. Two subjects completed the questionnaire independently (though with clarifications as necessary) in less than 10 minutes. All those consulted considered the questionnaire relevant, straightforward and easy to understand, and they all embraced the response options well. However, they found the instructions too lengthy, so these were shortened.

Phase 3

The resulting 56-item FV instrument was piloted with a sample representative of the overall UK population of children with visual impairment or blindness[24] (Table 1).

Data screening and preliminary item reduction. The 'true' missing data (i.e. no response given) on the FVQ_CYP questionnaire were negligible (<3.5%) and missing at random, suggesting all 5 response options were well endorsed. However, as the response category 5 (indicative of 'not applicable – N/A' response) is not scorable, we also treated these as 'missing' data for the purpose of item reduction (the total amount of missing data of 21-54% on a number of items, suggesting these may be irrelevant to a large proportion of respondents). Additionally, endorsement of N/A category on a small number of items was associated with age group. Thus, in total 11 items were removed having over 20% of missing (all types) data: 2 common to both age groups ('*Getting the right bus by yourself*' and '*Using other public transport, e.g. trains, by yourself*'), 6 for the 10-12 year olds ('*Cooking*', '*Reading tickets or receipts'*, *'Using mobile phone for texting your friends',* '*Using mobile phones for phoning people'*, '*Watching films in the cinema* 'and '*Shopping by yourself e.g. for food or*

clothes') and 3 for the 13-15 year olds (*'Taking part in drama classes'*, *'Playing team sports with adaptations'* and '*Playing musical instruments'*).

Three children with over 25% missing responses were excluded (all in the younger age group, two blind and one VI [Vision Level LogMAR 0.48 – 0.70, Table 1]). Skewness and kurtosis for all items were within acceptable limits (-2.00 - + 2.00[15]). However, 2 items showing the response category 4 to be redundant ('*getting dressed by yourself* 'and '*getting around your house by yourself*')and a further 2 with over 60% of responders endorsing the end response category 1[15]('*going up and down the stairs by yourself', 'getting yourself a drink'*) were removed.

Formal psychometric analyses and item reduction. Exploratory FA and PA applied to the remaining 41 items suggested it was most appropriate to extract one factor. The first FA eigenvalue was 19.53 and the second 2.28, which was lower than the second eigenvalue obtained via the PA (2.42), confirming appropriate extraction of a single factor. This factor accounted for 59.85% of the variance in the scale, with all items having a loading greater than 0.55 on the single factor.

The scale was then fitted into Rasch RSM and items were removed iteratively using the pre-defined criteria. Only one item (*'Finding objects you have dropped or lost'*) was removed having an outfit value outside of the acceptable range (outfit MNSQ = 1.61). Three items were removed after showing notable DIF across age group (*'Making yourself a snack*', '*Finding your way around an unfamiliar house or a new building*' and '*Crossing the road by yourself*') and one showing notable DIF by gender (*'Writing*'). The remaining 36-item scale showed DIF and infit and outfit values within acceptable limits (Table 2), and a clear unidimensional structure (Figure 3, available at http://aaojournal.org), with all items having a high factor loading on the first factor in FA (> 0.55). A small number of items with infit and/or outfit values outside of the more stringent .7-1.3 range[15] (Table 2) may indicate some potential noise and item redundancy and could be considered for removal in the future.

Item category probability plots for each item on the 36-item scale showed well ordered response categories overall, with an increase of category thresholds by at least 1.4 logits across the scale, showing a good distinction between the 4 response categories (Figure 4). As indicated by the item-person map (Figure 5), the scale showed good targeting of the 36 items to the responders, with the difference between item and person means within acceptable limits (-.33). The person separation (4.60) and reliability (.95) were high, indicating high measurement precision of the instrument.

Following item calibration in Rasch, the 4 response categories were recoded into 0-3 scale, and the scores were added to derive the FVQ_CYP summary score for the reduced 36-item scale. Multiple imputation of missing data was used prior to derivation of the summary scores for the original (unimputed) dataset and individual and pooled imputation iterations. There was a highly significant positive moderate association between FVQ_CYP summary scores and visual acuity with greater severity of visual impairment correlated with greater the self-reported FV difficulty, (imputed datasets pooled $r = .560$, $p > 0.001$), which held true for unimputed and imputed datasets. This supports the construct validity of the FVQ_CYP.

DISCUSSION

We have developed a novel self-report FVQ_CYP instrument for children aged 10-15 years to capture their self-assessed ability to complete vision-dependent tasks, which is psychometrically robust and relatively short and easy to complete. It has good construct validity, with FV summary scores correlating significantly with visual acuity. The FVQ_CYP is a unidimensional scale (i.e. capturing a single latent trait that is FV) with high measurement precision, which is targeted well to children with a range of visual impairment across the 10-15 years age range and gender, and which discriminates between children with different levels of visual acuity.

Currently only two other measures of 'visual ability' are available for children. The two versions of the LV Prasad-Functional Vision Questionnaire[25, 26] were developed for children in the

developing world and some items have limited applicability elsewhere. The Cardiff Visual Ability Questionnaire for Children and Young People [27] is a 25-item instrument that is either interviewer or self-administered, and captures information on very specific activities that children and young people 5-18 years might undertake. To date, its applicability has only been tested with a population drawn from the same specific geographical area in the UK in which the scale was developed.

Our FVQ_CYP is intended to be used both as a stand-alone PROM as well as a module within a comprehensive child-led assessment of the impact of vision loss, which will also include our recent instrument assessing VQoL in children, the VQoL_CYP,[6] and objective clinical measures of VF (e.g., acuity). Distinction between these measures is important but the underlying constructs are frequently conflated in the ophthalmic literature.[28] To report on their FV, the child's focus needs to be on their abilities and levels of functioning e.g., how difficult it is to navigate around the house. By contrast, to report on their VQoL, the child needs to reflect on the balance between his/her current situation and their hopes and expectations in the social context e.g., personal autonomy and social participation. Thus, VQoL and FV outcomes will not necessarily have a straightforward relationship with the objectively measured vision parameters or changes in vision as a result of treatment or visual rehabilitation. Here, we have demonstrated a significant association of child-reported FV, as measured by our novel instrument, with visual acuity, substantiating the instrument's construct and criterion validity. We suggest that the FVQ_CYP should be useful in pediatric ophthalmology care to capture children's own perspectives concerning their daily functioning, as an adjunct to clinical assessments, especially when a more detailed understanding of outcome change is necessary.

As our VQoL_CYP instrument is currently in the final stage of development, we cannot here formally evaluate the complementarity of the two instruments with statistical tests. Their complementarity is partly ensured by their development, being grounded in the same population of children and drawing on a common qualitative dataset capturing children's own views of the impact of visual impairment derived using a child-centered approach (which includes consulting

children as experts, when shaping the instrument so that it is feasible, acceptable and user-friendly).

We recognise certain methodological limitations, which reflect the challenges in testing and piloting different aspects of the instrument in small study samples. This issue of 'power' is largely unavoidable when studying an uncommon disability affecting children for whom participation in such research is a challenge.[29] So we adapted our approach, recognising, for example, that adopting conservative criteria for item removal during a tentative stage of instrument development with a small sample could compromise content and face validity and be counterproductive in the long term if potentially informative items were discarded before larger samples were available. Thus, we used a more lenient infit/outfit criteria[18] to inform item removal, but recognise that there may be some redundancy of items that could be addressed in the future.

A less stringent cut off criteria (>50%)[15] than ours (>20%) for exclusion of missing data has been proposed which may explain the issues we have noted in relation to the 'not applicable' response option (which in literature is generally treated as missing data). For instance, its endorsement on a small number of items was associated with age group and we removed these items before psychometric analyses. The small and uneven numbers of participants in the two age groups precluded us from examining age-dependent responses in detail. Further development of the scale may include separate age-appropriate versions. For instance, such items could be 'banked' and used for future testing across other age groups, whilst the currently derived scale is applicable across the 10-15 years age range. Interestingly, other studies developing similar measures for children with visual impairment have used a much broader age range of 5-18 but have not reported on age applicability issues across this wide age range where vision dependent tasks can be anticipated to vary considerably (e.g. using public transport or using mobile phones).

Inclusion of the 'not applicable' option may have caused some data attrition by potentially confounding the endorsement of the 'very difficult or impossible' response category

preceding it. This may have been exaggerated by the 'positive psychology' approach we adopted, which involved cuing children to consider the level of ease before considering the difficulty in completing tasks. Further testing of the scale with larger samples by reversing the order of response-options presentation, starting with 'how difficult' may help resolve the possible ambiguity caused by the proximity of the end category for difficulty and not-applicable category.

Further development of our instrument (including testing of the difference between a low vision intervention group and a control non-intervention group, test-retest reliability and long-term responsiveness over time) is needed to further ensure its validity and reliability. Future studies including multicenter evaluation will be necessary to replicate the present findings regarding reliability and validity as well as to assess the instrument's acceptability and feasibility. However, the psychometric strengths of the instrument demonstrated thus far are sufficient pre-requisites for its formal implementation into routine clinical practice, as planned at our clinical centers. Importantly, this will enable us to test the instrument further in the clinical context for which the instrument has been designed.

In its current form, the FVQ_CYP (available presently on request from the corresponding author) is a short, user-friendly, psychometrically robust instrument that can now be considered for implementation in routine pediatric ophthalmology practices, where its reliability and validity can further be evaluated. The FVQ_CYP is intended a) to be a valid and reliable tool for assessment of the self-perceived impact of visual impairment on children and young people in terms of the level of difficulty of performing vision-dependent activities; b) to be a complementary measure to objective clinical assessments (e.g. acuity or fields) as part of a broader assessment of the functional impact of visual impairment on children; and c) to be a complementary measure to other PROMs, such as our VQoL instrument,[6] to delineate the functional and socio-emotional impact of visual impairment on children. Thus, the FVQ_CYP should be a useful clinical tool for evaluating the effectiveness of low vision rehabilitation or other clinical interventions. It will also enable longitudinal assessment of child-perceived changes in

functional ability in progressive disorders such as retinal dystrophies, in order to inform decisions about technology-based adaptations and psychological support at school and home. Incorporating use of the FVQ_CYP into routine clinical care offers the potential for individualised assessment of impact of vision impairment. . This will permit children's perspectives of their visual disability and its impact on their daily lives to be included in decision-making about treatment, healthcare and resource allocation.

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The Functional Vision Questionnaire for Children and Young People with visual impairment (FVQ_CYP) is available on request from the corresponding author.