Title: Patient Reported Outcome Measures (PROMs) in Paediatric Ophthalmology: A Systematic Review

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Key words (MeSH terms): Questionnaires, Child, Ophthalmology, Quality of Life, Psychometrics

Abbreviations: PROM - Patient Reported Outcome Measure, QoL – quality of life; VQoL – vision-related quality of life, HRQoL – health-related quality of life, VI – visual impairment

Word Count (excluding title page, abstract, references, figures and tables): 2, 998

Abstract

Purpose: To identify patient-reported outcome measures (PROMs) specifically developed and used to assess the impact of ophthalmic disorders in children and to systematically assess their quality as a basis for recommendations about their use in clinical and research settings.

Methods: A systematic review of the literature was performed in MEDLINE, EMBASE, PsychINFO, CINAHL and AMED, supplemented by a grey literature search. Papers reporting development and validation of questionnaire instruments for assessing patient-reported outcomes of an ophthalmic disorder in patients aged 2-18 years were included. Quality was assessed by examining the purpose and psychometric properties of the instruments. Strengths and limitations were summarised with recommendations regarding use.

Results: Search identified 17 instruments. Of these, 11 were condition specific and 6 were intended for a broader population of children and young people with visual impairment regardless of the ophthalmic condition. Three were developed for use in a specific trial and two are still in development.

Conclusions: Paediatric ophthalmology PROM development and application is a developing field and new instruments are needed. There is scope for improvement in this area through a) clarity of definitions of the underlying constructs intended to be measured at the onset of development of new instruments, b) application of child-centred approaches and c) adherence to extant guidance and best practice in questionnaire instrument development.

Introduction

Increasing international emphasis on patient-led assessment of the impact of illness and healthcare[1-3] has resulted in a proliferation of questionnaires intended to measure the impact of illness or disability and treatment from the patient's perspective (i.e. patient-reported outcome measures – PROMs)[4]. PROMs include outcomes such as 'health-related quality of life (HRQoL)', 'activities of daily living 'or' health status', for example in trials of clinical, educational and social interventions[5].

A surge in vision or eye specific PROMs for adults[6-13] contrasts with a hitherto dearth of conceptually grounded, psychometrically robust, self-report questionnaires (typically referred to as instruments in the literature, as they represent measurement tools) suitable for use with children and young people. This paucity reflects the present lack of a clear and established conceptual framework defining the underlying vision/eye related constructs intended to be measured, combined with the challenges of child-centred methodology necessary to drive development of such measures.[14] A particular problem in both the adult and paediatric literature is the frequent (albeit inadvertent) merging of concepts such as vision–related QoL (VQoL), visual function and functional vision to describe 'the impact of visual disability'. This is further complicated by the lack of a universally adopted definition of QoL[15]. Superimposed on this are the challenges inherent in studying populations of children with visual impairment (VI) affected by disorders which are individually uncommon, rendering development of an appropriate PROM methodologically labour-intensive and time-consuming [16]. Thus, to date, generic health-related, rather than vision or eye specific, PROMs have been used in paediatric ophthalmology[17-24]. These can distinguish children with visual disorders from fully sighted or unaffected children, but they have limited value in discriminating between children with the same ophthalmic condition but of varying severity or in detecting clinically significant changes over time[25].

Sometimes, as an alternative to using generic paediatric PROMs, vision/eye-specific measures for adults have been used in paediatric population[26-32]. This approach is recognised as

problematic, because these instruments are not developmentally sensitive and their content and formats do not reflect what children understand or value[14, 33, 34]. For example, they contain items that are irrelevant to children (e.g. driving, financial outcomes)[10, 12] or use language and response categories that are not age-appropriate[9, 11]. Additionally, disease-specific adult PROMs[12, 31] are not applicable across a population of children with different ophthalmic disorders.

Thus, there is a need for child-appropriate vision and eye specific outcome measures that can be *self-reported*, can *differentiate* between children or young people with specific eye/vision conditions and are *sensitive to changes* over time as a result of natural history and/or treatments. As part of our on-going programme of research to develop such VQoL and functional vision instruments[35] we have undertaken and report here a systematic literature review in which we aimed to: a) identify all vision and eye specific PROMs specifically developed and used to assess the impact of eye disease and treatment in children, b) assess the quality of the identified instruments against conventional criteria, and c) provide recommendations about the use of these PROMs in paediatric ophthalmology clinical practice or research.

Methods

Search Strategy. MEDLINE, EMBASE, PsychINFO, CINAHL and AMED databases were searched systematically without any restrictions on publication date or language, with the main concepts being vision and/or eyes, experience, measurement and child (Appendix 1 provides search and MeSH terms). These citations were exported to EndNote citation manager and duplicates removed. A separate citation search was performed on all subsequently included original papers and reviews, supplemented by 'grey' literature search (i.e. conference abstracts, Google search and personal communication). The initial search was carried out in March 2012 and updated in July 2012 and February 2013.

Inclusion/Exclusion Criteria and Process. Abstracts were reviewed independently by 3 researchers (AH, VT, NS) to determine eligibility based on the criteria shown in Table 1. The full

paper was obtained where there was insufficient information in the abstract to determine eligibility. At this stage the papers where abstracts were not in English were excluded as we did not have the resources to ensure robust translation to address cross-cultural validity issues.

(Insert Table 1)

Data Extraction and Quality Assessment. Information was extracted by two researchers (AH, NS) and checked by a third (VT), using an agreed data extraction template, focussing on instrument description and quality (Appendix 2). Different versions of the same instrument reported in different publications were counted as one.

Using extant criteria for assessing PROMs[36, 37] we reviewed the following:

a) *Purpose of the instrument:* Whether a clear definition of the construct being measured was provided, as well as a clear specification of the instrument aim and the target population.

b) *Content, face and construct validity:* *Content validity* is the extent to which questionnaire items reflect all relevant domains of the construct that the instrument aims to measure. To evaluate this we examined whether instrument content and item generation and reduction had used firstly, appropriate qualitative research techniques (e.g. through interviews, focus groups and consultation with patients, supplemented with a literature review and consultations with professionals) and secondly, appropriate psychometric approaches (e.g. removing mis-fitting items through Rasch or Factor analysis to provide a statistically justified summary score). *Face validity* refers to the extent to which the content of the instrument appears 'at face value' to measure what is intended to measure e.g. whether an instrument purportedly measuring QoL reflects different domains that QoL, as a multi-dimensional construct, is meant to capture (e.g. social, emotional and functional impact). We examined this by looking at the actual content area (items and domains) of the instrument. *Construct validity* refers to the extent to which the instrument truly captures the construct under investigation. We examined this by looking at the degree to which the instrument i) correlated with another scale intended to measure related or

unrelated constructs (i.e. *convergent and discriminant validity* respectively) or ii) discriminated between groups expected to differ (i.e. *known groups validity* – treatment vs. control group).

c) *Reliability:* Reliability refers to the consistency of measurement (e.g., over time or different conditions). We investigated this by looking at the different types of reliability reported e.g. *internal consistency* (e.g. whether all items within an instrument or instrument domain are related to each other, reflecting a common latent construct, using Cronbach's alpha) and *test-retest reliability*. Other types of reliability were also considered (e.g. inter-rater reliability or person and item separation reliability using Rasch analysis).

After quality assessment (based on the recommendations by Pesudovs et al 2007[37], Appendix 2), for each instrument extracted data were summarised qualitatively in terms of the main strengths and limitations of the instruments, supplemented with recommendations regarding use.

Cross-cultural validation criterion was not included in the formal quality assessment as the identified instruments were not reviewed for their appropriateness for use in a particular cultural setting, but any cross-cultural adaptation attempts have been considered qualitatively with respect to instrument recommendations.

Results

Search Results. Figure 1 illustrates the process of study and instrument identification. Sixteen eligible instruments developed between 1999 and 2012 were identified (15 through database search and 1 through the grey literature[38]). The second search update identified through grey literature one more article reporting instrument development[39] [(1)]. Thus, 17 instruments were identified in total.

[(1)] This recently published instrument was developed as a distinct (rather than revised) second version of an existing instrument[40] and was therefore subsequently assessed as a stand-alone instrument.

Fifty two articles were excluded following paper review and data extraction (Appendix 3). Two articles[41, 42] were excluded as the full text was not available in English and we did not have resources for a full English translation for the purpose of this review.

(Insert Figure 1)

Instrument description. Of 17 identified instruments, all but one[43] were developed and validated using the English language (although in different versions). Six are intended for the broader population of children and young people with VI, irrespective of the eye condition (referred to as 'all-cause generic VI instruments') (Table 2a) and 11 for children with specific ophthalmic conditions (referred to as 'eye-disorder specific instruments') (Table 2b).

Seven instruments were intended as measures of quality of life (IVI_C[44, 45], CVFQ[46, 47], VQoL_CYP[35], IXTQ[48-50], QUICK[43], PREP[51-54] and CAT-QoL[38]), 3 of functional vision or visual ability (CVAQC[55], LVP-FVQ[40], LVP-FVQ II[39]) , 2 of the impact of treatment for the condition (ATI[56-64] and EIAQ[65]), 1 of symptoms (CISS-V15[66-75]), 1 of both symptoms and quality of life (NLDO[76-79]), 1 of psychosocial impact of the condition (PIQ[80]) and 1 of well-being (PPQ[81, 82]). The EYE-Q[83, 84] is interchangeably referred to as a measure of visual function and quality of life. Tables 2a and 2b provide details of the comparison of intended and achieved measured constructs and/or domains targeted by these instruments.

At the time of writing, 2 of the 17 instruments were still in early development stages: a QoL questionnaire for children with amblyopia (CAT-QoL[38]) and a self-report vision-related QoL instrument for children and young people with visual impairment (VQoL_CYP[35]). A further 3 instruments were developed for specific trials (PREP[51-54], EIAQ[65] and PPQ[81, 82]). Thus, these 5 instruments have not been included in the full quality assessment presented here, but the publications cited describe their development and/or application to date.

(Insert Table 2a)

(Insert Table 2b)

Instrument quality assessment. The 12 instruments assessed for psychometric quality (i.e., Appendix 2) are presented in Tables 3a, 3b and 3c, which summarise their purpose, main strengths and limitations, as well as conclusions and recommendations regarding their use. Within the groupings of 'all-cause generic VI instruments' (Table 3a) and 'eye-disorder specific instruments' (Tables 3b and 3c), the instruments are listed alphabetically, rather than in order of psychometric 'quality value' (from highest to lowest based on the quality criteria, as presented in Appendix 2) because their applicability to the target clinical population or age group of interest, as well as an appropriate cross-cultural adaptation, should always be considered first if deciding on their use for clinical or research purpose.

(Insert Table 3a)

(Insert Table 3b)

(Insert Table 3c)

Discussion

Our review demonstrates that, currently, the field of paediatric ophthalmology PROMs is a limited, but dynamically developing area with most reported instruments still subject to further development. Only a small number of instruments that we identified (e.g., LVP-FVQ II[39], IVI_C[44] and CVAQC[55]) are currently in a sufficiently advanced stage in psychometric terms to be recommended for use in clinical care. However, even these instruments, like others, would benefit from further psychometric improvements, such as testing in larger samples and application in trials or other settings to demonstrate their responsiveness. Equally, they may not be suitable for all paediatric ophthalmology populations as appropriateness depends on

applicability to the target clinical population, the cultural context in which instruments are to be used and the age group of the subjects.

Our review highlights the challenges of developing robust child-appropriate PROMs. In keeping with practices set out in the adult PROM literature[37], available instruments should not be employed without first considering carefully their theoretical and psychometric strengths and limitations. Nevertheless, for the majority of the instruments identified by our search the focus of the papers reporting their development is the measurement aspect, largely neglecting the definition of the construct to be measured. Measurement properties of an instrument, including different forms of validity and reliability, form the backbone of any tool intended to place the severity of an outcome on a reliable and valid scale. However, we argue that the quality of an instrument cannot be judged by these properties alone, which in any case, are strongly influenced by the conceptual definition and the theoretical framework underpinning the construct under measurement.

We found that only some instruments provided a definition of the construct measured (PIQ[80], ATI[56, 58], CISS-V15[66-68, 73], LVP-FVQ[40], LVP-FVQ II [39], CVFQ[46, 47] and VQoL_CYP[35]). Despite lacking an explicit definition, the instruments intended to capture functional outcomes, such as visual ability (e.g., CVAQC [55]), may not be as susceptible to misconceptions, as they are by definition ability and function related, and are therefore likely to correlate closely with functional outcomes assessed objectively (e.g. acuity). Failing to define QoL has greater implications. The consensus is that QoL is a subjective, psychological construct encompassing multiple domains of life (i.e. emotional, social, independence)[99]. Translating QoL into ophthalmology, therefore, should involve capturing the impact of having a vision or eye disorder across *all* QoL domains. Nevertheless, we found that some investigators appear to assume that vision or eye condition related QoL involves measurement of the restriction in eye or vision function. Thus, certain measures apparently assessing vision- or eye condition-specific QoL in

fact comprise scales assessing symptoms or functional ability associated with those conditions (e.g. QUICK[43] and EYE_Q[83, 84]).

This apparent 'construct conflation' is not specific to the ophthalmic literature. It has been a general side-effect of the World Health Organisation's broadening of the definition of health as 'a *state of complete physical, mental and social wellbeing and not merely the absence of disease or infirmity'* (p.100)[100] and the subsequent emergence of HRQoL as a patient-reported outcome that is distinct from but complementary to objective clinical measures. Thus, researchers have identified a 'disability paradox' whereby people with severe disabilities and restricted functioning are found to report high QoL, which reaffirms the notion that functioning and/or health are only some of the facets that affect a person's perception of the quality of their life[101, 102]. Thus, although vision-related functional and disability outcomes of visual function (e.g. visual acuity) and functional vision (e.g. mobility capacity given the available visual functions), or symptoms associated with ophthalmic conditions, may be intrinsically related to how a child living with a VI or eye disorder feels about their life (i.e., their QoL), these are conceptually distinct aspects of the impact of a vision/eye condition and require differential assessment.

The danger of construct conflation is of potentially misleading findings. For example, if an instrument purportedly measuring QoL, but actually measuring function, detects 'improvement' over time as a result of low vision rehabilitation, it would be misleading to suggest that the child's QoL had improved as other QoL domains had not been considered. In other words, the effectiveness of this intervention may be incorrectly estimated if it had not been assessed whether the child, although more functional, was still unhappy, dependent and socially isolated. Similarly, if a measure with items tapping parental perception of the impact of a particular condition and associated treatment fails to detect 'improvement' following an intervention, we cannot assume that the intervention itself is not beneficial for the child if we did not ask child-relevant questions of the child in the first place.

The lack of an explicit construct definition for many instruments made our assessment of validity challenging, particularly where there was a mismatch between the instrument intention (including the title) and actual content area. Instruments measuring functional vision (construed to capture the level of difficulty in performing a vision-specific task, e.g. CVAQC[55], LVP-FVQ[40], LVP-FVQ II[39]) would be expected to correlate highly with an objective measure of visual function (e.g. acuity), both being on a ruler-like scale of 'difficulty' related to functioning. However, in keeping with the 'disability paradox' [101],we argue that QoL measures should not necessarily converge with functional outcomes. Instead, it is expected that they should correlate with outcomes on other generic HRQoL and psychosocial measures. Despite this, our review found that evaluation of construct validity of QoL instruments more usually relies on a comparison with a clinical measure such as acuity and contrast sensitivity or solely on observing differences between patients with varying severity of a disorder. Similarly, symptom-driven QoL measures may diverge from HRQoL and converge with clinical symptom assessment (e.g. QUICK[43]).

The underlying principle of patient-reported outcomes such as QoL and emotional wellbeing is that they are subjectively defined. Therefore, the starting point for every instrument should be discussions with patients affected by the condition of interest so that the issues relevant to their lives can be captured accurately. Instruments that do not adopt this method may lack content validity and, as argued above, can be misleading. Where item generation through discussions with affected individuals (e.g. very young children) is not possible, we recognise the role of consulting parents on the impact of the child's disorder. In this scenario, however, such an approach may result in measures that capture (in addition or solely) parental stress and impact on the family (e.g. NLDO[76-79]). One instrument that we identified (IXTQ[48, 50]) provides an example of how the assessment of parental anxieties and QoL can be helpfully separated from the child's with concurrent development of two complementary but distinct, respondent-targeted measures.

Lack of engagement with the target population in the development process has been a limitation of a number of identified instruments, but there is encouraging evidence of changing trends. Our review identified on-going programmes of research advocating and demonstrating a child-centred approach, involving children and young people at all stages of instrument development. The CAT-QoL[38] is intended as a self-report measure of HRQoL in children undergoing treatment for amblyopia aged 5-7 years, and is grounded in individual interviews with young children. Our own research to develop the VQoL_CYP[35] uses a theoretical framework of 'self-discrepancy between actual experience and expectations' for capturing the quality of life of visually impaired children aged 10-15 years. More recently, DeCarlo et al[103] reported the methodology for identifying child-related concerns, through focus groups with children and their parents, as a basis of a future non-condition-specific vision-targeted HRQoL instrument for use with visually impaired children aged 6-12 years. These programmes and others identified in this review demonstrate the evolving nature of the paediatric ophthalmology PROM field and highlight new approaches to the development of child-appropriate instruments, heralding a coming of age for this important scientific area.

A lack of robust, conceptually grounded instruments renders the routine use of paediatric ophthalmology PROMs in clinical practice very difficult at present, despite the high profile of this approach in health service planning and policy. The ophthalmic community needs to work together to address this challenge. We would encourage clinicians to work with qualitative and quantitative scientists and their paediatric patient populations to develop robust, reliable and easy to use PROMs. Without this, clinical practice and health policy in paediatric ophthalmology cannot be shaped by what matters most to children with visual impairment and/or eye conditions.

Funding: This review was funded by the Medical Research Council (MRC) through a Public Engagement (Public and Patient Involvement) Award and a Small Grant from the MRC Centre of Epidemiology for Child Health. Additional support was received from: Guide Dogs Association for the Blind, Fight for Sight Charity, National Institute of Health Research (NIHR) Biomedical Research Centre at Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology, and NIHR Central and East London Comprehensive Research Network. The review was undertaken at UCL Institute of Child Health/ Great Ormond Street Hospital and Moorfields Eye Hospital / UCL Institute of Ophthalmology, both of which receive a proportion of funding from the Department of Health's NIHR Biomedical Research Centres funding scheme. Members of the team are supported by the Ulverscroft Foundation. The Centre for Paediatric Epidemiology and Biostatistics at Institute of Child Health (ICH) also benefited from funding support from MRC in its capacity as the MRC Centre of Epidemiology for Child Health.

Competing interest: The authors have no competing interests.

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Table 1: Inclusion/exclusion criteria for selection of papers to be considered for review

Inclusion	*Exclusion*
Children with visual impairment and/or eye disorders, paediatric ophthalmology patients/subjects	Children with non-ophthalmic conditions
Age 2-18 years	
Questionnaires, scales, instruments, checklists	
Patient oriented measures i.e., patient-reported and parent proxy reported outcome measures (PROMs)	Objective parameters and tests, clinical indicators and those restricted to demographic or environmental indicators
Measures of impact of living with visual impairment or an ophthalmic condition including quality of life (QoL), wellbeing, visual ability, functional vision, symptoms, functional status	Parental reports of child's behaviours (e.g. autism checklists, strength and difficulties questionnaire and other checklists of behaviours that are external to the child's experience of impact of living with a visual/eye disability/disorder)
	Clinician proxy measures (e.g., clinician's questionnaire report on symptoms, QoL etc)
	Visuo-spatial objective behavioural tasks for sighted children with specific cognitive delays (e.g. William's Syndrome)
Measures designed specifically for ophthalmic patients, eye-specific	Generic measures only, not developed for vision/eyes
Qualitative studies only if qualitative research was carried out as part of a PROM development	Qualitative studies describing impact of eye/vision problem without an objective to develop a patient-reported outcome measure (PROM)
	Review articles, letters, comments or articles without an abstract
	Abstract not in English

Table 2a: Instrument description – 'All-Cause Generic Visual Impairment (VI)' Instruments

Instrument	*Year and Country*	*Languages*	*Eye Condition*	*Age Range*	*Respondent*	*Input into items*	*Number of Items*	*Intended construct and/or domains*	*Captured construct and/or domains*
Cardiff Visual Ability Questionnaire (CVAQC)[55]	2010, UK	English	Visual Impairment	5-18 years	Child*	Children	25	Visual Ability	Rasch-derived unidimensional scale capturing visual ability/functional vision
Children's Visual Function Questionnaire (CVFQ)[46, 47] †	2004, USA	English	Visual Impairment	0-7 years	Proxy	Researchers	35 (age<3); 40 (age 3-7)	Vision-related Quality of Life	Factor Analysis derived multi-dimensional instrument with following subscales: 1) General Health, 2) General Vision, 3) Competence, 4) Personality, 5) Family Impact and 6) Treatment
The impact of vision impairment on children (IVI_C)[44, 45]	2011, Australia	English	Visual Impairment	8-18 years	Child*	Children, parents & teachers	24	Vision-related Quality of Life	Rasch-derived unidimensional scale capturing the impact of VI on participation in daily activities
LV Prasad-Functional Vision Questionnaire (LVP-FVQ)[40] ‡	2003, India	Indian English, Hindi, Telugu	Visual Impairment	8-18 years	Child *	Children, parents, clinicians & researchers	20 §	Functional Vision	Rasch-derived unidimensional scale capturing visual ability/functional vision
LV Prasad – Functional Vision Questionnaire Second Version (LVP-FVQ II) [39] **	2012, India	Indian English, Hindi, Telugu	Visual Impairment	8-16 years	Child *	Children and parents	23	Functional Vision	Rasch-derived unidimensional scale capturing visual ability/functional vision
Vision-related Quality of Life of Children & Young People (VQoL_CYP)[35]	2011, UK	English	Visual Impairment	10-15 years	Child	Children	Under development	Vision-related Quality of Life	Instrument in development

* Interviewer administered.

† Used also outside of the original instrument development group[85-88].

‡ In a subsequent study by Nirmalan et al. [89], an 11-item version was applied and validated in a community sample. It is unclear how the shortened scale was derived, thus we only assessed for quality the original instrument.

§ Summary score based on 19 items, with 1 item providing global rating of vision.

** Second, stand-alone version of LVP-FVQ, see footnote (i).

Table 2b: Instrument description – 'Eye-Disorder Specific' Instruments

Instrument	*Year and Country*	*Language*	*Eye Condition*	*Age Range*	*Respondent*	*Input into items*	*Number of Items*	*Intended construct/domains*	*Captured construct/domains*
Amblyopia Treatment Index (ATI)[56-64]	2001, USA	English	Amblyopia	3-13 years	Child * & Proxy	Parents, clinicians & researchers	20† (proxy version, age 3-6) 19 (child version, age 7-13)	Impact of amblyopia treatment	*Parent version:* Factor Analysis derived multi-dimensional scale with following subscales relating to treatment of amblyopia: 1) Adverse effects, 2) Treatment compliance and 3) Social stigma. *Child version:* Factor Analysis derived multi-dimensional scale with following subscales relating to treatment of amblyopia: 1) Adverse effects, 2) Treatment compliance and 3) Functioning at near.
Child Amblyopia Treatment Questionnaire (CAT-QoL)[38]	2011, UK	English	Amblyopia	5-7 years	Child	Children	In development	Quality of Life	Instrument in development
Convergence Insufficiency Symptom Survey (CISS-V15)[66-75] ‡	1999, USA	English	Convergence Insufficiency	9-18 years	Child *	Clinicians & researchers	15	Symptoms	Unidimensional scale capturing symptom severity (not derived psychometrically)
Effects of Youngsters' Eyesight on Quality of Life (EYE-Q)[83, 84]	2010, USA	English	JIA-Associated Uveitis	8-18 years	Child	Children & clinicians	23 (age 8-15); 26 (age 16-18)	Vision-related Quality of Life and/or Visual Function	Unidimensional scale capturing visual ability/functional vision (not derived psychometrically)
Emotional Impact of Amblyopia Questionnaire (EIAQ)[65]	2004, UK	English	Amblyopia	<67 months	Proxy	Parents & clinicians	15	Impact of amblyopia treatment	Multi-dimensional scale capturing the impact of amblyopia treatment in form of following subscales (not derived psychometrically): 1) Child's Experience, 2) Family's Experience, 3) Child's Well-Being
Intermittent Exotropia Questionnaire (IXTQ)[48-50]	2010, USA	English	Intermittent Exotropia	5-17 years	Child § & Proxy	Children & parents	12	Health-Related Quality of Life [73]	*Child version:* Unidimensional scale capturing quality of life of children (not derived psychometrically) *Parent version:* Factor Analysis derived

									multi-dimensional scale with following subscales: 1) Function, 2) Psychosocial effects and 3) Surgery
Nasolacrimal Duct Obstruction (NLDO) Questionnaire[76-79]	2006, USA	English	Nasolacrimal Duct Obstruction	6-48 months	Proxy	Parents & clinicians	29 **	Symptoms and Quality of Life	A priory determined two-dimensional scale with following subscales: 1) Symptoms and 2) Child's Health-Related Quality of Life
Pediatric Refractive Error Profile (PREP)[51-54] ††	2006, USA	English	Refractive Error	8-18 years	Child	Not Reported	26	Vision related functioning and wellbeing	Multidimensional scale with following subscales (not derived psychometrically): 1) Overall Vision, 2) Near Vision, 3) Far Vision, 4) Symptoms, 5) Appearance, 6) Satisfaction, 7) Activities, 8) Academics, 9) Handling and 10) Peer perception
Perceived Psychosocial Questionnaire (PPQ)[81, 82]	2002, UK	English	Amblyopia	3-5 years	Proxy	Parents & clinicians	10	Perceived Psychosocial Well-being of Child	Item details not available
Psychological Impact Questionnaire (PIQ)[80]	2006, UK	English	Amblyopia, strabismus or refractive errors	16-18 years	Child	Children & researchers	8 items (across 4 scenarios)	Psychological Impact	Unidimensional scale capturing psycho-social impact of strabismus (not derived psychometrically)
Quality of Life in Children with Vernal Keratoconjunctivitis (QUICK)[43]	2007, Italy	Italian	Allergic Conjunctivitis	5-12 years	Child	Children, parents, clinicians & researchers	16	Quality of life	Factor Analysis derived two-dimensional scale with following subscales: 1) Symptoms and 2) Daily Activities

* Interviewer administered.
† 18 questions, plus 2 items with a non-applicable option.
‡ Used also outside of the original instrument development group[90, 91].
§ Interviewer administered for children 5-7 years.
** 26 items, plus 3 non-scorable questions on the symptom scale.
†† Used also outside of the original instrument group development[92].

Table 3a: Quality assessment and recommendations – 'All-Cause Generic Visual Impairment (VI)' Instruments

Instrument	*Purpose*	*Strengths*	*Limitations*	*Conclusions and recommendations*
CVAQC[55]	Intended as a measure of self-perceived ability to perform vision-related tasks (i.e. visual ability) in children and young people with VI aged 5-18 years.	–Evidence of content and face validity, e.g. the instrument is grounded in the perspectives of visually impaired children and young people, collected through several focus groups. –Evidence of construct validity, internal consistency and statistically justified scoring using Rasch analysis. –Evidence of test-retest reliability 2-3 weeks later.	–Focus groups that informed the instrument development included sighted children. As the measure is intended for visually impaired children, the role of sighted children is unclear and has not been justified. –Instrument not validated for use outside of the country of development (Wales, UK).	Currently one of the most psychometrically robust measures of visual ability (i.e. functional vision) in children and young people with VI. It would serve as a valid and reliable PROM in evaluating the effect of low vision rehabilitation or other vision-specific interventions for this population of children and young people, with appropriate cross-cultural adaptation and validation outside of the country of origin.
CVFQ[46, 47]	Intended as a proxy measure of vision-related QoL for children with VI up to 7 years of age. The measure aims to capture difficulties in performing vision-related tasks, the effect of VI on the child's personality and social behaviour, on the parents or family and on the attitude towards treatment.	–One of the first measures developed to capture the impact of VI on children. –Evidence of construct validity: the scale discriminates between children with bilateral and unilateral cataracts, different levels of vision severity and treatment regimens. –Evidence of internal consistency across scales and age groups. –It has been translated into German and Portuguese[88] (although only the German version was based on recommended cross-cultural translation and validation approaches)[86, 87].	–Low content and questionable face validity: items were adapted from developmental tests and visual function instruments without any input from parents, children or external clinicians. Some items appear irrelevant to children younger than 3 years, regardless of their level of vision (e.g. ability to wash face or help with chores). Many items on the CVFQ address the parent's own QoL, implying a separate 'family impact construct' that should be a separate subscale. –Test-retest reliability time averaged at 4.4 months between the first and second instrument administration, which is longer than recommended.	This is the only available measure for evaluating the impact of VI in younger children. Rather than a measure of the child's QoL, it should be considered as a measure of parent-reported impact of childhood VI on the family and of the child's vision-related competency. Consultations with parents and children where age-appropriate (e.g. 5-7) would enhance the content validity of the scale. Further, appropriate cross-cultural adaptation and validation outside of the country of origin (USA) should also be taken into account if considered for use by researchers and clinicians.
IVI_C[44, 45]	Intended as a vision-specific QoL instrument for visually impaired children and young people aged 8-18 years.	–The first paediatric measure of the impact of VI on children using Rasch analysis measurement model to provide a precise, statistically justified scale. –Evidence of content and face validity e.g. through focus groups with children and young people, as well as professionals. –Evidence of construct validity, internal consistency and statistically justified scoring using Rasch analysis.	–The young people's views (particularly those of children aged 8-11 years) during item generation may have been under-represented compared to those of the adult stake holders' (e.g. 19% vs. 81% in focus groups respectively), contrasting with the established idea of QoL being a subjective construct. –Questionable choice of assessment (Guttman split-half) of test-retest and of mode and inter-observer reliability. –It has not been validated for use outside of the	Currently one of the most psychometrically robust vision-related PROMs available. The scale captures the broader impact of living with VI, with a specific focus on daily activities. It can be a useful tool in evaluating the effectiveness of low vision rehabilitation or treatment intervention in a wider population of children and young people with VI aged 8-18 years, with appropriate cross-cultural adaptation and validation outside of the country of origin.

			country of development (Australia).	
LVP-FVQ[40]	Intended as a measure of functional vision in visually impaired school-aged children in India.	–Evidence of content and face validity i.e. through focus groups and interviews with clinicians, children and parents as well as a literature review informing the instrument's items. –Evidence of convergent validity through Rasch analysis –It has been translated into Arabic for use with Egyptian children (although recommended cross-cultural translation and validation approaches were not reported [93]).	–Low reliability evidenced through Rasch analysis e.g. disordered response categories and inadequate outfit statistics and standardised residuals, as well as low person reliability and person separation statistics. –No evidence of test-retest reliability.	Despite good content validity, it has limited psychometric properties and the second version LVP-FVQ II is recommended instead.
LVP-FVQ II [39]	Intended as a measure of functional vision in visually impaired school-aged children aged 8-16 years in India. Intended as a distinct, psychometrically superior version of the original LVP- FVQ.	–Evidence of content and face validity e.g. through focus groups and interviews with children and parents as well as a literature review of existing questionnaires and adapting items from other similar, validated measures for this population. –Evidence of construct validity, internal consistency and statistically justified scoring using Rasch analysis –Evidence of test-retest reliability 2 months later. –Cross-culturally validated for 3 main languages spoken in India	–Although 'not applicable' response category was included in instrument, its utilisation by responders, nor the presentation of missing responses, has not been reported. –It has not been validated for use outside of the country of development (India).	This 2nd version of the LVP-FVQ is psychometrically superior and is recommended for use instead of the original LVP-FVQ. The instrument would serve as a valid and reliable PROM in evaluating the effect of low vision rehabilitation or other vision-specific interventions for children and young people with VI aged 8-16 years, with appropriate cross-cultural adaptation and validation outside of the country of origin.

Table 3b: Quality assessment and recommendations – 'Eye-Disorder Specific' Instruments

Instrument	*Purpose*	*Strengths*	*Limitations*	*Conclusions and recommendations*
ATI [56-64]	Intended to measure the impact of amblyopia treatment as part of study trials of the acceptability of treatment and its impact on the child and family. Initially intended as a proxy measure for children aged 3-7 years[56] (we refer to this as ATI-proxy), it has more recently been adapted for children up to 13 years[59] (we refer to this as ATI-child).	–Evidence of construct validity: a) ATI-proxy discriminates between children receiving different treatment regimens such as patching, atropine drops and Bangerter filters, suggesting it can be a useful scale for evaluating the parent-perceived effect of treatment interventions; b) ATI-child discriminates between children receiving patching and those receiving atropine drops. –ATI-proxy captures three Factor Analysis determined domains, which showed good internal consistency (excluding misfitting items).[57, 58] –It has been translated into Korean (although recommended cross-cultural translation and validation approaches were not reported)[94].	–Questionable internal consistency: poor fitting items identified through psychometric validation remain in the scale in subsequent publications. –Questionable content validity: a) the process of ATI-proxy item generation has largely been led by literature review and professional opinion with minimal input from parents or patients; b) ATI-child was designed *ad hoc* from ATI-proxy without the input of children. –No evidence of test-retest reliability.	If using ATI-proxy, excluding the misfitting items (e.g. 15 and 17) is recommended. ATI-child is not recommended in its current form as its development is not grounded in children's own views, being an adapted version of a parental form. Further construct validity testing against another measure and further development, including consultations with patients with regards to including new and/or revising existing items, are needed to enhance the currently limited psychometric properties of the proxy version and to create a valid and reliable child self-report version of this instrument. Cross-cultural adaptation and validation is also required for use in countries not included in the Pediatric Eye Disease Investigator Group's (PEDIG) randomised control trials sites.
CISS-V15 [66-75]	Developed by the Convergence Insufficiency and Reading Study group specifically for a survey of symptomatic school aged children suffering from convergence insufficiency (CI). Both child and proxy versions of the scale are available.	–The only patient-reported measure available for children with CI. –Evidence of face validity. –Evidence of test-retest reliability 2 weeks later. –Evidence of construct validity: the scale discriminates between children with CI and those with normal vision. –The authors have calculated the cut-off point for symptomatic children and also the minimal difference needed for a clinically significant change, making the tool useful for monitoring therapeutic interventions (although authors caution against using it as a screening tool, as it would not pick up a child who is asymptomatic due to avoidance of symptom-related tasks). –High internal consistency ($\alpha > .90$, although	–Questionable content validity: Symptomatic children and their parents were not included in instrument development. –No formal psychometric item reduction or standard construct validation approaches (e.g. testing against a clinical symptom assessment) have been applied.	Potentially useful for monitoring therapeutic interventions, although it cannot be used for screening purposes. This instrument should be used with some caution as the scale has not been derived through standard psychometric item reduction approaches, hence the summary score is not fully statistically justified. Appropriate cross cultural adaptations should also be considered for use outside of the country of origin (USA).

		such high Cronbach alpha may be indicative of some item redundancy[37]). –Used in Australia[95, 96] and translated into Hebrew [90], although no cross-cultural adaptation methods reported.		
EYE-Q [83, 84]	Intended as a measure of vision-related quality of life (VQoL) and visual function in juvenile idiopathic arthritis (JIA) patients with related uveitis, aged 8-18 years.	–The only available JIA-specific child-appropriate measure. –Evidence of content validity: items were derived from interviews with paediatric rheumatologists, optometrists and children with and without VI. –Evidence of test-retest reliability 10 days later. –Evidence of construct (known groups) validity: the scale discriminates between groups on the basis of the level of vision impairment. –High internal consistency (α>.90, although such Cronbach alpha may be indicative of some item redundancy).	–Questionable face validity: The measure lacks clarity regarding its purpose and underlying construct, being interchangeably referred to as a measure of visual function[83] and VQoL, despite the authors' acknowledgement that the two are distinct constructs[84]. Based on the actual content area, the questionnaire is likely a measure of functional vision. This is further evidenced by EYE-Q scores being weakly associated with HRQoL (measured using generic measure PedsQL[97]), but moderately correlated with visual acuity and contrast sensitivity. –The final scale and summary scoring has not been informed by the standard psychometric item reduction approaches. –No cross-cultural validation undertaken outside of the country of development (USA).	The EYE-Q shows promise as a measure of functional vision, rather than of VQoL in children with JIA. It is currently undergoing further developments and it is recommended that any revisions of the scale are taken into account before considering using this scale. Cross-cultural adaptation and validation are also needed before it can be used outside of the country of origin.

Table 3c: Quality assessment and recommendations – 'Eye-Disorder Specific' Instruments (continued)

Instrument	*Purpose*	*Strengths*	*Limitations*	*Conclusions and recommendations*
IXTQ [48-50]	Intended as a measure of QoL in children with IXT, IXTQ comprises 3 versions, 2 of which concern the child's QoL (child and proxy versions) and one addressing the parental QoL (the latter excluded from quality assessment in this review).	–Evidence of content validity: the scale is grounded in the experience of exotropic children and addresses the psychosocial and subjective experience of the child. –Evidence of construct (known groups) validity: the scale discriminates between children with IXT and controls (orthotropic children with a median visual acuity of 20/20). –There are complementary proxy and age-appropriate self-report versions, including a self-report version for children aged 5-7, which in the literature is often substituted by proxy report. –High internal consistency (α >.90, although such high Cronbach alpha may indicate some item redundancy).	–Questionable statistical justification provided for deriving the summary IXTQ score, e.g. subnormal cut-off and 'normative' scores for QoL calculated and based on a small sample. –No evidence of test-retest reliability. –It has not been translated and validated for use outside of the country of development (USA)	The IXTQ has good content grounding, having been developed from the concerns raised by children with IXQT and shows promise as a condition-specific PROM for this population. Both parent and child versions were developed in parallel and are complementary for capturing their distinct perspectives. Further psychometric testing (including comparison of children with IXT receiving different treatments for the condition, test-retest reliability testing and further item reduction) should be considered for this scale as well as appropriate cross-cultural adaptation and validation if used outside of the country of origin.
NLDO [76-79]	Developed as part of research trials by PEDIG, NLDO was intended as a proxy measure of symptoms and health-related QoL (HRQoL) in children with the condition aged 6 to 48 months.	–The only PROM available for this population. –Evidence of content validity: content is based on interviews with parents of children with NLDO, and discussions with clinicians. –Evidence of construct (known groups) validity: the scale discriminates between children with and without the condition –Evidence of responsiveness: the scale shows sensitivity to changes in the condition due to treatment intervention[77-79]. –High internal consistency for both the questionnaire as a whole and the symptoms scale, although not the HRQoL scale (α >.90, although such high Chronbach alpha may be indicative of some item redundancy).	–The summary scoring is based on items on an *a priori* decided subscale (Symptoms and HRQoL) that did not withstand psychometric item reduction. –Questionable face validity: The HRQoL subscale items combine the impact of NLDO on both parent and the child, implying the underlying construct measured is 'the impact on the family' rather than the subjective HRQOL of the child (with only two questions relating specifically to the child's experience). –No evidence of test-retest reliability. –It has not been validated for use outside of the country of origin (USA).	Although potentially useful as a measure of symptoms and family impact of the child's condition when monitoring the progression of NLDO treatment in affected children, NLDO is not recommended as a measure of child's HRQoL. Further psychometric validation with larger samples should be considered for this scale as well as cross-cultural adaptation and validation for use outside of the country of origin.

PIQ [80]	Intended for assessing the psychological impact in general daily life of amblyopia, strabismus and refractive error in teenagers aged 16-18 years.	–Evidence of content validity: item development grounded in the views of amblyopic adolescents. –Evidence of test-retest reliability 2-4 weeks later. –Evidence of construct validity: low correlations with an adult visual function questionnaire (VF-14[12]) and significantly different PIQ scores between cases and controls with normal vision.	–The scale has not been subjected to formal psychometric item reduction and the summary score has not been statistically justified. –Despite enlisting the help of adolescents when developing the instrument it is unclear how items were generated and evaluated for relevance and to what extent adolescents were involved. –It has not been validated for use outside of the country of development (UK).	PIQ could be useful as a starting point for potential items tapping the psychosocial impact of amblyopia, strabismus or refractive error on young people. However, its use as a valid and reliable PROM is limited, as the suitability of the items has not been formally tested through appropriate psychometric approaches, nor has the scale been cross-culturally validated for use outside of the country of origin.
QUICK [43]	Developed in Italy to measure the impact of ocular allergy on HRQoL in children aged 5-12 years.	–Currently the only instrument available specifically for this population. –Evidence of content validity: both user (parents and children) and professional views (literature review as well as clinicians' opinion) were considered when developing instrument items. –The two-dimensional QUICK scale (Symptoms and Daily Activities) possesses adequate internal consistency and construct validity, with both subscales correlating highly with the disease subscale of the KINDL[98] (a generic HRQoL instrument) and the Symptoms subscale correlating with clinical symptom parameters.	–The item reduction approach using Factor Analysis (requiring large samples) was based on very small samples, limiting reliability. –Questionable face validity: prior to Factor Analysis, the authors removed the ten lowest scoring items with little statistical justification, eliminating most of the items tapping psychosocial aspects of QoL. Contrary to its title, the QUICK appears to be largely a symptom scale, with only four items tapping participation in daily activities. This is further evidenced by the significant correlation with the disease subscale of the KINDL[98] and the absence of an association with important psychosocial indicators of HRQoL such as well-being and self-esteem. –No evidence of test-retest reliability. –English translation of the instrument has been undertaken by the authors without recommended cross-cultural translation and validation approaches.	Although the only paediatric PROM available specifically for this population, psychometric approaches to developing QUICK were significantly limited, suggesting it should be used with caution. QUICK can be used as a symptom scale, rather than a measure of QoL, but the broader impact of living with vernal keratoconjunctivitis needs to be incorporated into the scale, as well as appropriate psychometric validation with a larger group of participants. Appropriate cross-cultural adaptation and validation is recommended before use outside of the country of origin.