Measuring the impact of visual impairment during childhood and adolescence: Development of vision-specific patient-reported outcome measures for children and young people living with visual impairment.

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I, Alexandra Robertson confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Abstract

Patient-reported outcome measures (PROMs) are increasingly becoming the gold standard for reporting on the quality of healthcare and are used to capture information about the subjective impact of disability or impairment. In the UK, it is estimated that 2 per 1000 children are visually impaired. Despite this, there is a dearth of psychometrically robust and age-appropriate PROMs designed for children and young people.

To address this, a pre-established theoretical and methodological framework was applied to explore the day-to-day impact of visual impairment during childhood and adolescence and, in doing so, develop a suite of age-appropriate, vision-specific PROMs. Specifically, this thesis documents the development of PROMs designed for young people aged older than 15 years. The parallel work comprising final psychometric evaluation of PROMs designed for children aged younger than 10 years is also reported demonstrating calibration of the instruments to allow for their use longitudinally in paediatric ophthalmology contexts.

One hundred and twenty-nine young people (aged 13-19 years) living with visual impairment took part in four phases of instrument development, comprising semi-structured interviews, cognitive interviews, and national postal surveys. A further 86 children (aged 7-13 years) took part in the final phase of psychometric validation for the child-instrument versions.

Qualitative analysis revealed fluctuation in the self-reported impact of visual impairment during childhood and adolescence, and results have implications for developing and administering interventions to promote self-reported outcomes. The final suite of age-appropriate instruments is grounded in the perspectives of children and young people living with visual impairment and psychometrically robust for use independently, and simultaneously, within clinical practice.

Follow-up studies are needed to generate the evidence required to understand children's, young peoples', parents' and clinicians' attitudes towards using the PROMs, as well as the feasibility of implementing the developed instruments within ophthalmology contexts, and with attention to novel, vision-specific approaches.

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Abbreviations

BL – Blind

CTT – Classical test theory

DDA – Disability Discrimination Act

DIF – Differential item functioning

FA – Factor analysis

FP – Fractional polynomial

FV – Functional vision

GP – General Practitioner

HRQoL – Health-related quality of life

ICF – International Classification of Functioning, Disease, and Health

IMD – Index of multiple deprivation

IQR – Interquartile range

IRT – Item response theory

LogMAR - Logarithm of the minimum angle of resolution

LV – Low vision

MNSQ – Mean-square

NHS – National Health Service

PCA – Principal component analysis

PIC – Patient identification centre

PID – Participant identifier

PREM – Patient-reported experience measure

PROM – Patient-reported outcome measure

QoL – Quality of life

RCT – Randomised control trial

RMSE – Root-mean-square standard error

RMT – Rasch model theory

SE – Rasch model-based standard error of the measure, not adjusted for misfit

SES – Socio-economic status

SVI – Severe visual impairment

ToM – Theory of mind

UCL – University College London

UK – United Kingdom

VA – Visual acuity

VI – Visual impairment

VQoL – Vision-related quality of life

WHO – World Health Organisation

YP – Young people

ZSTD – Standardised z-score

Chapter 1 Introduction

Childhood and adolescence are critical periods of life spanning birth to the age of 19 years[1] and characterised by learning, development, and maturation. When a child is visually impaired, childhood is likely to be disrupted in many ways. Within clinical practice and research concerning visually impaired children, there is growing interest in understanding the broader impact of visual impairment in the hope of improving vision-related quality of life (VQoL). This interest is reflected in the recent emergence of patient-reported outcome measures (PROMs) which are designed to capture self-reported and assessed burden. Despite a growth in the number of vision-specific PROMs designed for adults,[2-4] there are few robust vision-specific PROMs which have been designed, and are suitable, for children and young people (YP) with visual impairment. In 2011, Rahi and colleagues[5-7] developed a conceptual and methodological framework for the development of vision-specific PROMs suitable for children and YP, resulting in development of two complementary, age-appropriate, self-report measures: one assessing VQoL and the other functional vision (FV) in children aged 10-15 years.

The framework established by Rahi et al.[5-7] provides a methodological and theoretical starting point in which to develop age-appropriate, vision-specific PROMs capturing VQoL and FV which are suitable for use by a wider age range of children and YP with visual impairment. This thesis reports the extension and adaptation of these PROMs for use by children and YP aged 8-17 years, with particular focus on the development of PROMs designed for YP aged 15 years and older living with visual impairment. Adopting a life course perspective to capture VQoL and FV among YP living with visual impairment affords the opportunity to explore, in depth, YP's experiences of visual impairment, the everyday impact of visual impairment during adolescence, and the experience of growing up with visual impairment, as reported in this thesis.

This thesis begins with a discussion of relevant literature identifying the dynamic impact of visual impairment during childhood, the role of PROMs within generic paediatric

healthcare and, later, within paediatric ophthalmology services. A brief introduction to psychometric measurement within healthcare, classical test theory (CTT) and modern test theory as popular methodologies follows (Chapter 3). The 4-phase method used in the current project is detailed in Chapter 4. Both Chapters 5 and 6 report the results from the analysis; Chapter 5 details the qualitative findings from the first phase of instrument development and Chapter 6 presents the results from the psychometric analysis of PROMs, including the outcome of unidimensionality assessments, and Rasch analysis. The findings (both qualitative and psychometric) are discussed in Chapter 7, and finally in relation to future work needed to assess the acceptability and feasibility of implementing the developed PROMs within routine paediatric ophthalmology services (Chapter 8).

Chapter 2 Background

2.1 Childhood and adolescence

Childhood is a transient period of early life course defined as the period between birth and adolescence.[1] Growth and development during childhood takes place at a rate that exceeds that of any other period of life course, as important neurological changes occur[8, 9] in parallel and association with biological and behavioural changes such as motor[10, 11] and muscular development.[12] Despite aspects of growth and development which are universal and occur within all humans regardless of gender and ethnicity, definitions of childhood are largely reliant on cultural perspective and constructed in a wider social world in which assumptions are made and shared.[13] For example, among Western economically developed societies, childhood is accepted as a time for leisure and learning, which is free from major responsibility and commitment.[14] However, Robson[15] argues that the notion of children as dependent in this context fails to acknowledge the work children do in contributing to household survival and well-being. Indeed, children in developing communities play an important role in contributing to household production, for example by performing labour-intensive agricultural tasks. Within these cultures, childhood is conceptualised as a brief period of life characterised by biological and physical growth before the onset of formal employment and financial responsibility.

Throughout history, scientists have tried to make sense of growth during childhood by developing theories describing and explaining change. Most early theories were centred upon competing forces of nature and nurture. Greek philosophers such as Plato and Aristotle, for example, believed that parents should nurture their children carefully to avoid the negative force of human nature from dominating, and causing children to become rebellious and unruly.[16, 17] Today, perspectives span many aspects of development, and are often focused upon providing details of specific aspects of development which take place in a broader social and cultural context. In parallel to

physical growth, children are expected to develop cognitive capacities allowing them to think critically about the world that they live in, regulate and control their behaviour, and the behaviour of others; language skills for comprehension and speech production; and social skills through which they can establish and maintain meaningful relationships with others. There are several dominant theories within the field of childhood development, each emphasising factors such as biological and sexual drives,[18] rewards and punishments[19, 20] and, socialization.[21] Many of these perspectives describe *stages* of development, viewing childhood as multi-phasic, and thus propose that children of a given age show broad similarities across many situations. Piaget's theory of cognitive development[22, 23] remains particularly influential and has many real-life applications in modern day society, for example, within the field of education. Despite evolution of developmental theories over time, competing forces of nature and nurture remain theoretically influential, viewing children as evolving bio-psychological human organisms in the context of external environments – both immediate and remote. Developmental milestones during childhood are heavily influenced by the culture in which an individual grows up, and the wider social world in which they are nurtured.

At its simplest, adolescence is a term used to describe the *pinnacle* of childhood; a time when most developmental milestones have been achieved, but adulthood has not yet begun.[24] Similar to definitions of childhood, adolescence is a period which is socially constructed and the roles or duties of adolescents within society are largely dependent on cultural perspective. The timing of onset of adolescence is variable and difficult to define in precise terms; based upon multiple factors such as timing of physical and biological growth, growth of abstract thinking,[25-27] increased autonomy and independence,[28, 29] and growth of understanding about the community and broader world in which one lives.[30, 31]

One of the most transparent ways to identify the timing of adolescence is through reference to puberty. Regardless of the culture in which they live, all individuals face a

significant degree of biological change during adolescence as a result of puberty. These changes are essential for reproduction in the future. The timing of onset and duration of puberty is variable, and largely reliant on genetic factors, body mass, nutritional state, and general health.[32, 33] Puberty also occurs at significantly different points for girls and boys.[34-36] Despite occurring at different times, the experience of puberty is new and unusual for both males and females, triggering events such as the onset of pubic hair growth and growth spurts. It is not a single event but a process which takes time and, like the majority of theories of childhood and adolescent development, can be broken down into multiple phases.

Another factor complicating identification of the timing of onset of adolescence is cultural variation due to differences between national laws which set the minimum age thresholds for participating in activities traditionally considered to be reserved for adults. These activities include voting, marriage, property ownership and alcohol consumption. In most Western, developed societies, the age at which individuals gain the right to these activities is 18 years but in other countries this threshold varies. For example, girls in Iran are recognised as adults when they are just 9 years old.[36]

Definitions of adolescence are also influenced by the historical context in which they are developed and the dominant features of society at a specific time point. Early or emerging adulthood has recently been discussed as an extension of adolescence in modern day society as individuals increasingly struggle to achieve and maintain economic independence and security, and thus, delay the onset of adulthood. Greater frequency of college enrolment, time spent exploring future career choices, and later age at marriage and childbearing are currently trending.[37, 38] This is in stark contrast to definitions of adolescence which were used during the 17th and 18th centuries as children became skilled in specialist trades during childhood, and went straight into adulthood.[39] In modern day society, YP aged 18-25 increasingly pursue multiple paths and relations before pursuing serious, long-term stable relationships.[40] The term

'millennial' is increasingly used to describe the role of YP in modern day Western society.[37]

Whilst there is profound variations in the timing of adolescence, the United Nations define adolescents as individuals aged 10-19 years old.[36] For clarity this wide definition of adolescence, which is based on contemporary Western perspectives, is endorsed throughout the remainder of this thesis.

2.1.1 Adolescence as a key transitional life stage

Although puberty and development of reproductive function is often considered a physiological accomplishment occurring at the level of sexual organs, specialised neurons in the brain and hormones secreted by the pituitary gland play a critical role in governing reproductive function.[41, 42] A complex interplay between neurotransmitters such as norepinephrine, dopamine, serotonin, glutamate, neuropeptide Y, galanin, GABA, and corticotropin-releasing hormone during adolescence is required to trigger physical reproductive growth. Thus, hormonal changes are robust during adolescence and may be as great as eighteen-fold increase in males and eight-fold in females.[43] From a physiological perspective, hormones are thought to act on the brain to affect behaviour through their impact upon peripheral and neural-based processes.[44] These physiological interactions are likely to play a role in subjective, self-reported well-being, and are linked to emotional and behavioural characteristics of adolescence such as mood swings, changes in energy levels, and restlessness.[45-47] Some evidence suggests associations between hormonal changes and increased family conflict,[48] and risky behaviour[49] particularly when it results in high rewards.[50, 51] Impulsive, irrational behaviour gradually decreases in parallel with increased grey matter in the occipital lobe.[8, 52]

Adolescence can also be described as a period succeeding the stages of growth outlined by theories of childhood development. For example, from the perspective of cognitive development, at the age of 12, children are thought to enter the stage of

formal operations, in which they are able to think abstractly and hypothetically, taking into account the influence of multiple variables.[26] From a different perspective, children the same age are thought to develop understanding of other people's perspectives, by comparing themselves and others to a 'generalized other', assessing whether the view of one person is shared between others.[21] Completion of the stages outlined by theories of development has likely led to the definition of adolescence as a 'summit' or final stage of development: a prerequisite for adulthood.

In addition to change during adolescence and progression through the stages outlined by theories of development, an adolescent living in the developed Western world will likely experience a number of formal, policy driven transitions. These primarily take place within education. Children who are raised in the UK and enter mainstream education will experience at least two formal transitions during their childhood and early adolescence: one when they enter primary education at the start of term following their 5th birthday[53] and another when they enter secondary education at the age of 11 years.[54] Those who wish to extend their education beyond the statutory minimum requirement of completing secondary education at the age of 16 years[53] may experience numerous further transitions as they enter sixth form, college, or University. With each transition in education, there will be substantial change in environment (both physical and social).

It is also likely that individuals will experience a number of informal transitions during adolescence which, in comparison to formal transitions in education, are a result of changes driven primarily by growth, development and psychological maturation, and may occur at flexible time points which are driven by individual differences. Specifically, transitions in social interactions and the nature of friendships have been highlighted as characteristic of adolescence. For example, during adolescence, friendships with peers and family members have been shown to aid preparation for future independence, socialisation and intimate or romantic interactions,[55-57] and be important to the development of autonomy and a sense of identity as an adult.[58]

One major feature of adolescence is the experience of challenges, difficulties and distress. This may be the consequence of a combination of the biological, physiological, and policy-driven changes which occur during this period. Aristotle stated that during adolescence, individuals 'are heated by Nature as drunken men by wine'. Similarly, Socrates characterised adolescents as inclined to 'contradict their parents' and 'tyrannize their teachers'. Hall (1904) was the first to use the term 'storm and stress' to characterise adolescence[59] and subsequently paved the way for scientific research exploring the experience and cause of the apparent developmental 'crisis' which occurs during adolescence. Stress during adolescence was traditionally conceptualised in terms of conflict with parents, mood disruptions, and increased risky behaviour.[59] A greater number of negative life events related to peers, school and family are encountered during this period and co-occurrence of developmental events make it hard to pinpoint the cause of behavioural change. Negative emotions occur when there is discrepancy between what is expected and what really happens[60, 61] and may be a result of changes in perceived social role during adolescence, challenging ones view of themselves and their value in the lived world.[62] Despite use of terminology depicting adolescence as turbulent, it is increasingly recognised that not all adolescents face such challenges and stresses.[63] From this perspective, storm and stress is not something written into the human life course, but attributable to individual differences in the experience of adolescence. Research documenting the age-related challenges of adolescence in relation to visual impairment is discussed in Section 2.5.2 (pg. 46).

In summary, childhood and adolescence constitutes a critical period for profound growth and development. Adolescence is a period of life course occupied by growth which is unique from that which takes place during childhood. In addition to biological, physiological, and neurological development, adolescents are faced with a number of transitions or changes which inevitably make life more difficult. The social construction

of modern-day Western society means that YP may face a number of economic and financial challenges in addition to social, emotional, and psychological disruption.

2.2 Childhood disability

When a child grows up with a disability or illness, childhood and adolescence is likely to be qualitatively different from that of healthy and non-disabled children. The Disability Discrimination Act (DDA)[64] defines disabled children as those with a 'physical or mental impairment which has substantial and long-term adverse effects on their ability to carry out normal day-to-day activities'. In this definition disability is described as a broad category, stemming from no particular origin. In contrast, other definitions of childhood disability take checklist-type approaches: using umbrella terms such as learning difficulties, traumatic brain injury and autism, and those who need special education and related services.[65] Thus the population of children with disabilities is both substantial and complex in terms of their health-related needs, and may be difficult to characterise.

The social model of disability regards disability as a social construct, existing only as a result of the broader society's failure to accommodate differences brought by disability.[66, 67] This provides stark contrast to early research which tended to ignore the disabled child, and focus on the views and perceptions of those living with the child (e.g. their parents and families). Whilst portraying disabled children as vulnerable and inferior, this type of research focused upon the negative impact of disabled children upon their families and broader society, incurring economic difficulties and strained family relationships.[68] The voices of disabled children themselves were frequently ignored.[66, 69, 70] More recently, and with help from the social model of disability, society and research have begun to recognise the views and opinions of disabled children as valuable in their own right. It is increasingly accepted that disabled children are treated the same as their non-disabled counterparts, and that any adjustments or adaptations they require are put in place to allow for participation. This attitude is legally enforced

by definitions of childhood disability, such as the DDA's definition which specifies individuals' ability to carry out 'normal day-to-day activities'.

A second model demonstrating potential to change the conceptualisation of disability is the International Classification of Functioning, Disease and Health (ICF).[71] This model lists a number of environmental factors which account for the broader social context in which disability occurs. Instead of labelling individuals as 'disabled', this model views disability as a universal attribute, as every human being will experience a decrement in health at some point during their life course.[72] The ICF framework is based on interplay between two models of disability which are thought to be only partially valid: the medical model and the social model. Neither model is thought to be adequate in itself, based on narrow conceptualisations of disability confined to physical health (the medical model) and social construction (the social model). As a result, the ICF distinguishes between three levels of human functioning; bodily functions & structure, activity and participation, and environmental and personal factors, providing a mid-point between medical and social perspectives.

From social perspectives, it is not merely the presence of disease or impairment which renders a child 'disabled' but rather the interaction between physical afflictions and day-to-day culture which is socially constructed. Disability is something which is imposed, whereas impairment is a private, internalised attribute.[73] Some evidence shows how social frameworks have potential for improving disabled children's participation in everyday life, for example, by increased awareness, and removal, of barriers towards participation in education.[74] Thus, a new landscape in which to conceptualise disability is beginning to show potential for improving disabled children's participation in everyday life. Whilst still in development and undergoing modification, social models of disability are the first to allow the 'voices' of disabled children to be heard and thus led to recognition of the broader impact of childhood disability.

2.3 Measuring the impact of disability or illness in healthcare

Information has always been fundamental for healthcare services to ensure good outcomes for patients, and provide high quality care. Traditionally, the impact of disease and illness is measured using assessments and observations of physical function. These include impairment-based measures such as clinical examinations and functional capacity evaluations. Using these assessments, health professionals can gain insight as to the immediate impact of disease or illness on physical health and functional ability, and make inferences about the impact at an individual level and upon a patient's daily life. For example, a patient who reports shoulder pain may likely experience some form of difficulty when brushing their hair or taking a shower. Functional assessments of health also play a critical role in the formal diagnosis, and treatment of disease.

It is only in the last 10 years and with advances in information technology that measurement in health care has exploded in terms of quantity, quality and accessibility. It is now easier than ever before for health professionals to collect, analyse, and report data relating to multiple outcomes. Alongside this growth, it is increasingly apparent that functional assessments of health are not sufficient to capture the impact of disability which, by definition, considers restricted functional ability in 'the manner considered normal for a person'.[75] This is in line with social models of disability such as the social model and ICF which describe individuals living with disability as active, valuable members of society as opposed to oppressed and inferior.

In 1948, the World Health Organisation (WHO) defined health as 'a state of complete physical, social, and mental well-being, and not merely the absence of disease or infirmity'.[76(p1)] At the time, this definition was ground-breaking in terms of its breadth and ambition, overcoming restrictions to disease and including broader domains of physical, mental and social well-being. Subsequently, and likely as a result of the broad definition of health, Quality of Life (QoL) emerged as a multi-dimensional construct which can be used to measure the broader impact of disability or illness. Definitions of

QoL have varied over time and are difficult to establish due to the notion that each individual has their own unique QoL which depends on their experiences, lifestyles, hopes, attitudes and beliefs. It is not surprising that definitions of QoL also vary between contexts and perspectives.[77, 78] The range of factors included under the umbrella term 'QoL' can be placed upon a continuum, ranging from purely subjective, such as an individual's experience or appraisal of their daily lifestyle, to objective appraisals of physical function.[79] The 1995 WHO Quality of Life Group defined QoL as:

> *'an individual's perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, standards, and concerns.'* [80(p1)]

This definition is perhaps the most encompassing and accepted definition within existing literature and has therefore been adopted for the purpose of this thesis.

Interest in measuring QoL within healthcare has emerged over time and in response to recognition of the need to provide patients with treatment that at least makes them feel better when no cure is available. A substantial level of research highlighting the broader impact of disability, disease and illness upon psychosocial issues such as psychological well-being, and social relationships has supported the WHO definition of health and been influential in that there have been notable changes in clinical practice. For example, instead of focusing upon treating, and providing care for the purely physical impact of disease, specialist nurses have been trained in order to provide social support,[81] and psychological therapies such as cognitive behavioural therapy have been designed to enhance adaptation in those diagnosed with terminal illnesses.[82] Psychosocial support for individuals facing long-term disability and/or illness is increasingly emphasised by a number of health-related charities, such as Macmillan Cancer Support,[83] and Samaritans.[84]

When measured among patient populations, a common assumption is that good QoL is associated with good health[85] and if an individual is unable to perform a normative task, QoL is compromised.[86] However, this is discordant with empirical evidence which suggests that, in practice, perceived health, well-being and life satisfaction are often

different from objective health status and disability.[87] In response to this discordance, the 'disability paradox' was developed to explain variance between health, disability and quality of life.[88] Instead of being dependent upon interactions between subjective and objective health outcomes, this theory emphasises a balance between the body, mind and spirit as essential for high levels of QoL among all individuals regardless of disability. Thus capturing QoL within healthcare is conceptually difficult as it is underpinned by a number of subjective evaluations, each involving some degree of individual difference. This difficulty has led to the development of a number of conceptual models and measures of QoL but with very little uniform consensus on what is included and what should be measured.[89, 90] In response to this ambiguity, it has been suggested that three different aspects of health should be assessed within routine healthcare: objective measures such as clinical indices which patients would not necessarily use or be aware of (such as blood pressure), functional performance (the ability to perform certain activities which are part of daily lifestyles), and patients' own evaluation of the experience of living with illness or disability.[77] In relation to the latter, health-related quality of life (HRQoL) has emerged as an adjunct to measure QoL in health and has evolved from a loosely integrated body of research on health status, functional performance and social well-being. Despite ongoing evolution and development, to date, experts have disagreed on the specific definition of HRQoL. For example, in a systematic review, a large number of HRQoL models were found, however more than one-quarter of authors did not define HRQoL using the main concepts which are used by others, and some did not define HRQoL at all.[91] Similarly, the extent to which HRQoL is distinct from QoL has been debated, as some researchers have concluded that aspects of health and functioning are only some of the life domains that may impact QoL.[92]

Despite disagreement which has, to date, impeded the development of a universal definition of HRQoL, there is some agreement that HRQoL includes the same factors included in QoL definitions: subjective factors such as health perception, and emotional

state, and objective factors such as physical function and symptoms, but the factors are framed by the experience of disease, illness or disability in the broader social and cultural context.[93] Insertion of HRQoL into the ICF model of disability has been recommended in the view that disability is not a fixed state, but is fluid and changeable and therefore a general feature of the human experience.[94]

2.3.1 Patient-reported outcome measures (PROMs) and patient-reported experience measures (PREMs)

As life expectancy increases worldwide,[95] more individuals are surviving into old age with chronic and co-morbid conditions. As a result, policies within healthcare increasingly prioritise patients' *quality*, as opposed to *quantity*, of life. Concurrently, decision-making within healthcare is increasingly based upon patient satisfaction and preference. In 2008, Lord Darzi[96] stated three components to quality of care: patient safety, patient experience, and effectiveness of care. Patient-reported outcome measures (PROMs) can be used to measure the final component: effectiveness of care, by providing a platform for patients to report on the subjective impact of clinical care/treatment/interventions. As a consequence of this report, PROMs have played a mandatory role in healthcare in the UK since 2009. Hospitals providing four specific surgical procedures; hip or knee replacement, varicose vein surgery and groin hernia repair have been legally required to collect data from patients using PROMs before and after surgery to reflect the impact upon patients' subjective well-being.[97-100]

The second component of Lord Darzi's statement, that is, patient experience, can be measured using patient-reported experience measures (PREMs), which are distinct from PROMs because of their focus on the process of, as opposed to the *impact* of healthcare. Lord Darzi's report stated that payments to hospitals should be based on the quality of care they provide as opposed to the quantity of patients who are treated. Thus, PREMs are important to improving quality in healthcare: a requirement which was put forward by the NHS Operating Framework in 2012.[101]

Patient-reported outcome measures (PROMs) come in the format of questionnaire instruments which are used within clinical practice and research and allow individuals with disability or illness to self-report on the everyday impact of their condition, including their health status, preferences, condition-specific symptoms, pain intensity, and HRQoL. Self-report on the impact of disability or illness is the defining feature of PROMs and using PROMs, patients can communicate this impact to others, primarily clinical professionals.

The majority of PROMs incorporate some form of HRQoL as the primary outcome and response formats such as Likert-type scales which require the user to indicate, using numbers, images or diagrams their feelings, attitudes and beliefs about their health. Each category or image is assigned a value and summary scores can be calculated to represent a subjective evaluation of disease impact. Thus, PROMs incorporating assessment of HRQoL can be used to quantify the complex and subjective burden of living with a disease, illness or disability by reducing patients' self-report experience into a single, or series, of numeric values (see Chapter 3, pg. 68 for more background information on measurement in healthcare). There are a number of benefits of using PROMs in this way to quantify subjective outcomes, namely that outcomes can be captured quickly and with minimal support from healthcare professionals. Outcomes can also be compared between patients living with different degrees, or forms of disability. In this way, PROMs aid decision making based on prioritisation of interventions, treatment, or medication. In some cases, users are required to reflect on their subjective state within a specific time frame e.g. over the last week/month, giving healthcare professionals insight as to the ongoing, longitudinal nature of disease impact. PROMs are also inexpensive, and flexible instruments as they can be administered both manually using pen and paper and electronically, using computers or tablets, with patients completing measures in their own time outside of clinical settings.

PROMs which are currently available for use in healthcare can be divided into generic- and disease-specific measures. Generic PROMs are used to compare outcomes across different health conditions and interventions,[102] and are particularly beneficial when assessing cost-effectiveness within healthcare.[103, 104] However, the development of these measures has undoubtedly been affected by ambiguity surrounding the definition of HRQoL which is discussed in Section 2.3 (pg. 28). For example, health status has frequently been placed under the umbrella term of HRQoL[105] when it has previously been defined as reflecting an individual's relative level of wellness and illness which does not take into account individuals' perspectives of disease or treatment; a domain covered by the majority of HRQoL definitions.[106, 107] When a health status measure is used to quantify HRQoL, misleading conclusions can be made which may have serious implications such as misinterpretation of the value of an intervention upon HRQoL.

Alternatively, condition-specific PROMs have been developed within specific fields of health such as cancer,[108-110] heart disease,[111-113] and liver disease.[114, 115] These measures can be used to detect the condition-specific impact of a disease or disability which may not necessarily be covered by generic measures and thus discriminate more sensitively between patients with the same condition. Disease-specific measures can be particularly valuable when comparing the subjective well-being of patients living with varying manifestations of the same disease or when assessing the burden of disabilities with unique physical symptoms.

To date, PROMs have primarily been used in clinical trials,[116-118] national audits,[119] and clinical registers[120] but are increasingly widespread – at the level of both primary and secondary healthcare.[121, 122] In a systematic review of evidence supporting the use of PROMs, improvements were demonstrated in processes which took place in primary care (such as advice, education and counselling, and diagnoses given by consultants).[123] PROMs have also been shown to increase patient-provider communication about aspects of HRQoL,[124] enhance consultants' ability to detect

psychological and functional problems,[125] increase emotional support offered by consultants[124] and enhance 'patient centricity' within clinical care: an asset which is highly valued by patients.[103] However, the direct impact of PROMs upon health and well-being has been less convincingly demonstrated, with the majority of randomised control trials suffering from methodological limitations, such as use of multiple instruments and lack of consensus on the relevance of a single PROM.[123]

Although evidence supports that validity and reliability of PROMs are largely comparable to routinely used clinical measures, there are a number of barriers towards using PROMs routinely. These include, for example, the need for clinicians to be able to receive and interpret data promptly and communicate findings in a way which is understandable by patients.[126, 127] Clinicians may also be sceptical about using PROMs in the view that information gathered can force discussions into areas that consultants have little control over.[123] To date, this type of literature has been largely confined to adult populations, and little is known about the benefits of and barriers towards using PROMs within paediatric (and adult) ophthalmology services.

2.3.2 Development and application of PROMs for paediatric settings

To date, a number of PROMs have been developed for use by children living with a specific form of disability or illness.[128-133] In addition to containing items designed to capture the condition-specific symptoms and impact, PROMs which are designed for use by children need to include items which capture factors that contribute to QoL during childhood as these will likely differ from those attributable to QoL during adulthood. Definitions of QoL emphasise experiences, lifestyles, attitudes and beliefs contributing to overall well-being or happiness. It is therefore incorrect to assume a child, who has only a fraction of the experience, and a very different lifestyle from an adult, can use an adult-centred PROM to report on the impact of disease or disability.[134] Nevertheless, in the past, when child-specific PROMs have been unavailable, HRQoL measures for adults have been used in paediatric populations.[135] This approach is largely problematic because instruments not only contain items which

do not sensitively reflect children's priorities, beliefs, and HRQoL, but they are also framed in a way which is difficult for children to understand and interpret[79] (see Section 2.1, pg. 20 for a brief discussion of cognitive and intellectual development during childhood).

In the past, when age-appropriate PROMs have been unavailable for children, and PROMs developed for adults have been deemed inappropriate, measures of HRQoL have been completed by adults who have a substantial amount of everyday contact with the child, with the assumption that such 'proxy' reports provide reliable and valid inferences as to the HRQoL of the child. The most obvious proxy, or informant, used within clinical practice is a parent or guardian given the amount of contact they have with the child, and subsequent insight as to the child's subjective appraisal of HRQoL in multiple contexts e.g. at different times of the day, in different locations, and when playing or engaging with other people. However, any adult who has everyday contact with a child and/or sufficient knowledge of the potential impact of disease or illness can provide a proxy report. In some cases, proxies can be teachers and clinicians.[136] The value of proxy reports provided by adults with varying relationships to the child has been discussed in extant literature, as although mothers may be more involved with their child's healthcare, teachers and clinicians may be more impartial, and have views based upon comparisons of children with varying degrees of disability.[136]

Proxy reports can be particularly useful when a child is unable to self-report due to disability or age, or may be simply unwilling. For example, the youngest age at which children have been shown to reliably and validly self-report on complex outcomes such as HRQoL is 5 years[137] but this may be higher, depending on the nature of impact of disease or illness upon cognitive capacities, such as reading or interpreting items. Moreover, the true reliability and validity of outcomes provided by children as young as 5 years is questionable given that higher reliability is associated with increasing age.[137]

Importantly, when results from self- and proxy-reports are compared, outcomes are likely to be quantitatively and qualitatively different. Proxy reports represent an adult's

perception of the child's experience and are influenced by demographic characteristics such as the age, sex and socio-economic status of the respondent as well as their relationship to the child, and any emotional impact of disease or illness upon their own subjective well-being.[138] Empirical evidence demonstrates parents often underestimate their child's HRQoL[139] and are better at rating externalizing problems such as physical symptoms and somatic distress[140, 141] compared to internalizing problems such as social or emotional issues.[142-144] Thus, when both self- and proxy-reports are available, they must be used as complementary outcomes which provide unique perspectives. Outcomes should never be aggregated and one cannot rely solely on proxy-reports in the absence of PROMs which are suitable for children and sensitive to the impact of disability during childhood.[145] As a last resort, the International Society for Pharmacoeconomics and Outcomes Research recommend that, when an age-appropriate self-report measure is truly unavailable, a proxy-report measure based upon observable content such as behaviour may be used to give insight as to HRQoL.[146]

Identification of the need for child-specific PROMs is implicitly recommended by contemporary perspectives on disability during childhood. Instead of treating the purely physical, functional impact of disability, age-appropriate PROMs can be used to give children and YP a 'voice' and active role within healthcare e.g. in decision making regarding the management of their disability or disease.

2.4 PROM content, design and development

Methods used to develop PROMs and questionnaires in general need to be high quality, controlled, and rigorous if users of these instruments are to be certain that outcomes truly reflect the measurement constructs and that they are reliable. If questionnaires are poorly designed, data will be misinterpreted, and irrelevant and/or incorrect conclusions will be made.

Questionnaire development usually involves a series of standard stages in which ideas about content and format are formulated, and tested and analysed. There are many factors to consider when creating a questionnaire, including question type, wording and order, response options, and method of administration. The early stages of questionnaire design are usually exploratory and qualitative in nature as researchers need to gain information about the measurement outcome, and, in the case of PROM development, the construct of interest, the patient population and their perception of the impact of disability or disease. As discussed in Section 2.3 (pg. 28), PROMs can comprise a number of varying features. Factors such as patients' physical and psychological capability to complete PROMs independently and with ease need to be considered during the development.

The most common way to conceptualise the patient population, their self-reporting needs and the impact of disease or disability is to conduct lengthy, in-depth and often unstructured interviews with a sample of representative patients.[147] Interviews should give a rough shape of the enquiry and inform the content of the new PROM.[148, 149]

Once a questionnaire has been drafted, a quantitative approach is usually taken to ensure that the measure is reliable, valid and psychometrically robust, that is, capable of capturing the intended construct accurately and consistently over time. In 2011, the consensus-based standards for the selection of health measurement instruments (COSMIN) committee developed a checklist which can be used to calculate the quality of PROMs,[150, 151] thereby advocating a systematic, rigorous approach to instrument development which is grounded in psychometric evaluation. Types of validity featured in the checklist include *content* (i.e. that the measure provides coverage of all aspects of the outcome), and *criterion* (i.e. how well the instrument can be used to predict an outcome for another measure) validity. *Face* validity (i.e. that the measure appears to be, at face value, a good measurement tool) and *construct* validity (i.e. the instruments ability to measure the intended outcome) are psychometric properties which can be used as evidence of whether an instrument measures what it purports to measure, and

internal consistency reliability (i.e. the certainty that all items/questions in a questionnaire that are intended to probe the same area will produce similar results) indicates homogeneity among items and the instruments capacity for measurement.[152]

Classical Test Theory (CTT) approaches such as factor analysis (FA) and factor rotation[153] or modern psychometric approaches such as Item Response Theory (IRT)[154, 155] and Rasch analysis[156] are usually applied in the later stages of questionnaire development[157] in order to test the psychometric properties of the questionnaire and ensure that the measure is accurate and precise. The underlying assumption of these approaches is that the outcome of interest is not directly observable (e.g. wellbeing), and therefore one must collect data using mechanisms through which the outcome may be assessed, such as self-report. These mechanisms give us conclusions which can be made about unobservable factors. Using the principles provided by these approaches, one can determine how successful the mechanisms are when estimating unobservable variables.[158] A detailed description of these psychometric approaches, their strengths, weaknesses and current consensus on which approach is optimal is presented in Chapter 3 (pg. 68).

2.4.1 Involving children in questionnaire development

Unlike research conducted in the 1970s which focuses on Piagetian models of development through experiments *with* children, today, researchers are beginning to draw on sociological theories such as theories of childhood and disability.[14] This is evidenced, for example, by direct participation by children in the traditional stages of questionnaire development. In contrast to being the *object* of study, children are increasingly viewed as competent and worthy of a voice in an adult-ist world.[159] Information stemming from children's opinions, attitudes and beliefs is increasingly valued. As a consequence, methodologies such as those used to develop questionnaires, are increasingly designed in a way that allows for children's active participation through, for example, in-depth interviews. Historically, researchers would

have turned to the parents, teachers or paediatricians to represent children within research, and the views of children themselves would have been ignored.

Involving children in the development of questionnaires is important to ensure the questionnaire has content validity (see Section 2.4, pg. 36) by containing questions or items which are meaningful and relevant for the child and, in the context of PROMs, shed light on the subjective experience of living with an illness or disability. It is particularly important for children who are living with illness or disability to take part in instrument development given that outcomes from proxy measures are often discordant from children's own self-report.[139, 160]

When conducting research, all researchers are required to protect their participants from harm. This is a legal requirement which is enforced by ethical guidelines and regulations outlined by the National Institutes of Health[161] and World Medical Association.[162] Whilst the standards outlined by these institutes are applicable to research involving all human subjects, regardless of age, when research involves children, researchers must follow an additional subset of guidelines which are developed with the aim of protecting children. For example, researchers are required to gather informed assent from both the child and at least one parent when a child is under the age of 16 years.[161, 163]

Researchers must also consider ethical standards and research guidelines in relation to the child's developmental capacity and ability to take part in the research. Involving children in interviews and focus groups, essential first steps in the development of questionnaires, may be challenging. Children might not understand what an interview or focus group is, or what is expected of them[164] and may conform to their perception of the interviewer's expectations, resulting in unusually high levels of agreeableness.[164, 165] High levels of imagination during childhood may also cause researchers difficulty when interpreting children's true feelings or beliefs.[164] Children's voices may be interpreted by adult researchers in a way which conforms to the expectations or

understanding of adults, and researchers may add meaning to the perceptions and views of children.[166]

Particular ethical considerations arise when children are disabled, ill or in some way vulnerable. Gathering true self-reported burden of a disease or disability during childhood is likely to be difficult as the child may require some form of assistance from a parent or caregiver, such as reading questions aloud, writing down answers or physical manipulation of a questionnaire. As a consequence, children and YP may avoid disclosing sensitive or confidential information for fear that a parent or guardian may find out.

Institutional research ethics committees working alongside researchers have a duty to protect participants from potential harm, even if this means that modifications have to be made to the research design. This is particularly prominent when involving adolescents in research, given the developmental characteristics of adolescence. Ongoing changes in hormonal, neuroanatomical, and psychosocial well-being[167] make adolescents more susceptible to stress and negative emotions such as anger and worry.[168, 169] Distress could occur if the adolescent is triggered to remember a negative event, or asked about activities or behaviours which they are ashamed about or trying to keep secret. Discussions of sensitive issues such as the functional impact of disease or disability upon, for example, personal hygiene may cause distress and embarrassment.

Ensuring confidentiality is another important factor consistently highlighted in ethical regulations and standards. This is a legal requirement,[161, 162] and if researchers are unable to assure participants that the information they provide will be kept confidential, they may be reluctant to participate.[170] However, ensuring confidentiality is somewhat problematic as researchers also have a duty to pass on information if they feel an individual is ‘at risk’.[171] A common way to overcome this is to ensure that participants are informed prior to participation of the potential need to link sensitive and identifiable data. However, ensuring children or adolescents living with some form of disability are

informed of this possibility may be particularly difficult if, for example, they have specific learning difficulties which impact their understanding of the questions in assent or consent forms. To ensure data remain confidential throughout the course of a research project, most researchers will anonymise the data early to avoid data linkage. However, if data cannot be linked to the participant, researchers are unable to disclose sensitive information when a participant has revealed that they are at risk of abuse or harm.

2.5 Visual Impairment

Visual impairment is a form of childhood disability characterised by reduced visual function commonly measured by visual acuity (VA). VA has been traditionally measured using a Snellen notation, but the modern gold standard is a chart using the logarithm of the minimum angle of resolution (logMAR) notation to quantify the distance at which an individual is capable of reading printed text of various sizes. Globally, 285 million people are estimated to be visually impaired, of whom 39 million are blind (the latter classified according to the WHO taxonomy of severity of visual impairment as logMAR worse than 1.3).[172] In the UK, it is estimated that 2 in every 1000 children are visually impaired.[173, 174] The majority of children and YP with severe visual impairment and blindness (SVI/BL) have very early onset impairment, and grow up without useful vision. Severity of visual impairment can range from moderately reduced VA to complete blindness (see Table 1, pg. 42).[175-178] The most common causes of visual impairment in childhood are cerebral visual impairment, disorders of the optic nerve, and disorders of the retina.[179]

Table 1. Classification of severity of visual impairment.

Category	Presenting distance VA	
	Worse than:	Equal or better than:
0 Mild or no visual impairment		0.46 6/18
1 Moderate visual impairment	0.46 6/18	1.00 6/60
2 Severe visual impairment	1.00 6/60	1.30 3/60
3 Blindness	1.30 3/60	1.77 1/60*
4 Blindness	1.77 1/60*	Light perception
5 Blindness	No light perception	
9	Undetermined or unspecified	

* or counts fingers at 1 metre.

Whilst onset of SVI/BL is likely to be early in life, a number of disorders such as Stargardt's disease and neurological disorders[180] can cause onset of visual impairment or SVI/BL later during adolescence. Impaired vision can either be stable or progressive in nature at presentation and can manifest in a number of ways, affecting VA, visual field function, and contrast sensitivity in addition to oculomotor performance and visual perceptual abilities.[181]

Three quarters of children with SVI/BL have conditions which are neither preventable, nor treatable.[173]

2.5.1 Impact of visual impairment upon development during childhood

From a developmental perspective, the impact of reduced visual sensory input during childhood and early life course is significant, and likely to impact in a number of ways.

A substantial amount of literature supports the notion that communication between individuals begins long before an infant begins to speak. In the 1970s, researchers coined two types of intersubjectivity: primary and secondary. In the first 9 months after birth, infants are thought to be capable of primary intersubjectivity; that is, communicating to others about their immediate desires and emotions. This type of communication is largely driven by expressive behaviour such as crying which is intentionally used to get some kind of reward e.g. food or comfort.[182, 183] After the age of 9 months, infants are thought to be capable of secondary intersubjectivity: the ability to communicate with others about aspects of the wider environment such as surrounding objects, people and events.[184] In this dominant theory, vision is essential for communicating that an object exists and which qualities are particularly interesting.[22, 185]

Children who have visual impairment or blindness have been shown to compensate for the lack of visual stimuli in the development of primary and secondary intersubjectivity, by using auditory clues such as verbal instructions which are provided by others[186, 187] or tactile or sonorous stimulation.[187-190] For example, mothers have been shown to communicate effectively with their visually impaired infants through stroking, blowing or imitating the sounds they made.[191] Others have suggested that spacing plays a critical role among children with visual impairment[192] and these children are able to use their own bodies and the space they take up, as reference for the location, size, and physical attributes of objects.[192, 193]

In contrast to these findings, development of social communication, including joint attention, in children with visual impairment has been likened in the past to the development of children diagnosed with autism in that delay is often seen in specific difficulties such as social avoidance, social approach, and anxiety during social interactions.[194] Theory of mind (ToM) is an important developmental milestone during infancy which is defined as the ability to conceive mental states; that is, understand about the connections between other people's knowledge, desires, feelings and their

actions.[195] ToM is a prerequisite to meaningful communication with others. Critical to the development of ToM is the observation of other people's behaviour and evaluation of the intention of the behaviour. When observed in sighted infants, this milestone is thought to emerge at the age of 2 years, and is enhanced by pretend/or imaginative play.[196] When assessed in children with visual impairment, research has suggested that the development of ToM is universally delayed to some degree until the age of 12 years.[197-199]

Language development is another developmental capacity thought to be delayed in infants with visual impairment. Again, this form of delay is not surprising given that in sighted populations, vocabulary growth is associated with secondary intersubjectivity, and reliant upon observing objects in the wider physical environment.[200] A degree of social communication is also reliant upon non-verbal cues, such as facial expressions, body postures and gestures, which convey important information about how an individual uses language e.g. whether they are lying. Without vision, a child will miss out on these important non-verbal cues. Research documenting this delay, however, has mixed conclusions. Some researchers have shown that children with visual impairment develop language in the same way as sighted children[201] and others have shown delay in relation to limited experience with other people and the environment.[202, 203] It is also likely that children with differing degrees of visual impairment severity experience differences in terms of language development.[194] The origin of this form of delay is difficult to pinpoint due to the complex interface between biological, environmental, and social influences. For example, research documenting developmental delay in language acquisition may be related to the responses of others, such as parents, who may view their child as helpless and therefore incapable of communicating for themselves.[204, 205]

Despite the possibility that visually impaired infants may be capable of compensating somewhat in some sensory domains, it is likely that there is some degree of developmental delay among most children living with visual impairment. The extent to

which this prevails into adolescence and early adulthood is debateable as some literature suggests that visually impaired children eventually catch up with their sighted peers in many aspects of development,[198] and have some cognitive strengths in relation to age-matched controls.[206] When discussed in relation to ToM, for example, developmental gain may be a result of greater opportunity for conversational interactions as visually impaired children get older.[207, 208] In a study of high-functioning adolescents aged 10-16 years living with SVI/BL (versus visual impairment), participants were found to score significantly higher on a battery of neuropsychological tests than age-matched expectations based on test norms, with particular strengths in auditory and working memory.[206] These findings may be explained well by superior cortical capacities, including neural plasticity,[209] and a greater allocation of resources for auditory processing,[210] leading to specific cognitive strengths.[211] Thus, conflicting evidence for the nature and magnitude of the relationship between visual impairment and developmental delay has, to date, prevented development of a strong empirical foundation.

The impact of visual impairment at the level of activity and participation, the latter components of disability outlined by the ICF model, can be examined through self-reported experiences of children and YP living with visual impairment. Despite favouring the same activities as sighted children, children with visual impairment have reported being unable to participate in some activities, such as reading, watching live sports or concerts, or playing outdoor games[212] and subsequently unable to pursue the activities that they enjoy the most. However, self-reported accounts of the impact of visual impairment are often placed within a broader context in which experience of society, cultural norms, other people, and their reactions to visual impairment are influential. For example, children with visual impairment have reported difficulty socialising freely with other children, travelling independently, and choosing academic and sporting activities without input from a parent or guardian.[213] Maintaining a healthy social life may be difficult for children and YP with visual impairment as a consequence

of the functional impact of visual impairment upon social skills such as assertiveness skills or establishing eye contact.[214-216]

Determining the impact of visual impairment during childhood is further complicated by variation in the manifestation of impairment, nature of visual deterioration and onset of impairment. Some research suggests that individuals who are blind from birth have an advantage over those who develop visual impairment at a later stage during childhood in relation to arithmetic skills,[217] working memory,[217] and auditory spatial processing.[218] This may be attributable to these children's ability to compensate for the lack of visual input through re-organisations of neural networks[219, 220] as previously discussed (pg. 42). In comparison, individuals with later onset, acquired visual impairment may have missed the critical period for re-organisation of neural networks. Despite a wealth of empirical attention paid towards investigating differences in brain activity, particularly in the visual cortex, among those with early onset or congenital visual impairment and late onset deterioration in vision,[219-226] differences between the two populations in relation to the self-reported experience of visual impairment are yet to be formally investigated. Given that a substantial amount of learning comes from vision and takes place during early childhood, it is likely that differences between the populations extend beyond these variations.

2.5.2 Impact of visual impairment during adolescence

As previously discussed (see Section 2.1.1, pg. 23), adolescence is widely recognised as a time for growth and development in which individuals experience a number of formal transitions. There are also a number of informal changes that occur during adolescence, as children develop the cognitive and social skills necessary for progression in education and the nature of social interactions change. Development of independence is widely recognised as a defining characteristic of adolescence as individuals learn to perform tasks associated with everyday living such as travelling, cooking, or cleaning, without help from a parent or guardian. Adolescence is a period of life course recognised as qualitatively different from both childhood and adulthood,

when viewed in terms of healthy and sighted individuals. Thus, it is important to consider the impact of visual impairment as equally unique during this period. As children develop into adolescents, the negative impact of visual impairment can be viewed as dynamic and changing: increasing as individuals encounter new environments and increased opportunities for independence, and decreasing as individuals develop effective coping strategies, and adapt to their disability.

At the level of psychological well-being, over time individuals who are living with a chronic illness or disability, such as visual impairment, may become increasingly familiar, and subsequently better able to cope with the everyday impact or burden of their condition. Resilience and coping are intrinsic processes which are often studied in children living with long-term impairment or disability with a view to explain positive adjustment.[227] Resilience is defined as the process of identifying or developing resources and strengths to manage stressors flexibly and gain positive outcomes.[228] Thus, it is likely that an adolescent who has been living with visual impairment since an early age will undergo behavioural-motivational, cognitive and emotional adjustments[229] during childhood and adolescence, which may reduce the negative impact of visual impairment.

Despite the possibility for positive adaptation over time, a number of environmental barriers likely increase the impact of visual impairment during adolescence. As YP develop, they will likely encounter new situations and face a range of new environments. At the same time, they may experience a desire to become more independent and, fitting with the expectations, norms and attitudes of their peer group, resist functional support from others. Evidence documents a large proportion of YP with visual impairment aged 16-25 are not likely to be in employment, education or training (NEET) and are twice as likely to be NEET as the general population of 16-25 year olds.[230] During adolescence, individuals with visual impairment will be faced with realisations of the impact of visual impairment in the future, and may be worried about gaining a job in light of biased appraisals by future employers, and their increased

independence and responsibility in the future.[231] Thus, evidence suggests that as a child develops into adolescence, the impact of visual impairment is increasingly related to external environmental factors such as participation in everyday life events, in contrast to intrinsic, personality-related factors such as adjusting and coping with the functional impact of reduced vision.

Similar to their sighted peers, adolescents with visual impairment will likely experience a number of transitions or changes which will need to be overcome in light of living with visual impairment. Such changes may alter the impact of visual impairment, introducing new restrictions, or the need for new adaptations. To date, the existing literature documents only one normative transition experienced by children with visual impairment as they develop into early adulthood: from full-time education into formal employment. This empirical focus is likely to reflect knowledge of poor post-educational outcomes among YP with visual impairment[230] and the subsequent need to support YP as they transition into competitive economic and occupational climates. Similar to sighted YP, good educational outcomes among YP living with visual impairment are associated with greater rates of employment[232, 233] and social support from parents and broader social networks.[233, 234] Several transition-related programmes have been developed specifically for use by adolescents with visual impairment. These place heavy emphasis upon independence training,[235, 236] work experience,[237] transition planning,[237, 238] and early tuition of career-related skills.[239] Despite a number of informal changes documented among sighted adolescence, namely related to the value of social interactions and friendships, the range and experience of these informal changes during adolescence of YP living with visual impairment is yet to be formally investigated. Although independence is a key characteristic of adolescence, literature exploring the development of independence among YP with visual impairment is largely confined to improving post-educational outcomes. The impact of limited functional ability upon independence has received limited empirical attention and, when it does exist, is primarily discussed in developments of vision-related quality of life

outcome measures,[231, 240] and as a component of the broader, daily impact of visual impairment.

2.6 Measuring the impact of visual impairment

In ophthalmology, quantifications of visual impairment traditionally consist of purely objective, functional assessments such as VA. These measures provide necessary insight as to visual function and are critical for clinical management. However, these measures do not capture the *subjective* impact of visual impairment and are thus not sufficient to capture the interaction between levels and forms of impact of visual impairment as specified by social models of disability such as the ICF.

Attempts have been made to overcome the shortfall of measurement in ophthalmology, by incorporating generic HRQoL measures into both adult,[241, 242] and paediatric ophthalmology practice.[243] However, the full impact of vision-related problems cannot be adequately described, or measured, using generic HRQoL measures, as some QoL issues reported by visually impaired individuals are likely to be specific to the experience of living with impaired vision,[244] hence the need for measuring vision-specific QoL issues i.e. vision-related Quality of Life (VQoL).

Measurement of the impact of visual impairment upon daily functioning, i.e. the individual's ability to participate in activities which are meaningful and enjoyable, is a second outcome which measures of VA do not adequately address. Functional vision (FV) is defined as vision that can be used to perform tasks requiring vision and differs from VA in that it comprises a self-reported evaluation of the difficulty of completing everyday tasks in real everyday environments (e.g. navigating around an unfamiliar room, boiling a pan of water, or locating a bus stop). As with most questionnaires, items included in measures of FV should be developed with a particular population in mind, referencing activities which may only be relevant and meaningful for individuals of the same age or living within the same culture. Thus, functional vision incorporates elements of VA into a broader physical, social, and cultural context.

In the past, and perhaps as a product of issues surrounding conflation of HRQoL and functional status and ability, items relating to FV have been included in measures designed to capture VQoL among the population of adults living with visual impairment.[245, 246] For example, the Refractive Status and Vision Profile (RSVP) contains 8 subscales which include items relevant to both symptoms and physical-social functioning, and expectations and concerns.[247] Similarly, the Impact of Vision Impairment (IVI) measures 'handicap' which is defined as an interaction between limitation or activity and an individual's needs. This measure incorporates items which can be categorized into five domains: emotion, leisure, mobility, social, and household.[248] The multi-dimensional nature of these approaches is potentially problematic as condensing multiple outcomes relating to both VQoL and FV into one summary score, clinicians risk making inaccurate inferences about the effectiveness of treatment and intervention upon different aspects of the impact of visual impairment. To comprehensively capture the impact of visual impairment in light of individual differences such as needs, emotions, and preferences for example, would involve development and administration of VQoL and FV PROMs as complementary, yet distinct measures.

Similar to use of generic HRQoL PROMs (see Section 2.3.2, pg. 34), in the past, when child-appropriate vision-specific PROMs have been unavailable, vision-specific PROMs which were designed for adult populations have been used as alternatives.[249, 250] Whilst these contain items which may be more specific to the impact of visual impairment than items contained in generic HRQoL measures designed for children, a number of factors render them inappropriate for use in paediatric ophthalmology services. Firstly, the items included in these instruments are not developmentally sensitive, meaningful or valuable to the aspects of everyday life which may comprise VQoL and FV among paediatric populations.[107, 251, 252] For example, they contain items that are irrelevant to activities of childhood and adolescence, such as driving or occupation.[253, 254] Secondly, these instruments may contain language or response-categories which are difficult for

children or YP to understand.[255, 256] Finally, there are a number of ophthalmic conditions which present during childhood for which vision-specific adult PROMs may not be applicable.[254] Thus, PROMs which are both age- and vision-specific are needed for use within paediatric ophthalmology, to capture the dynamic impact of visual impairment during childhood and adolescence.

2.6.1 Paediatric vision-specific PROMs

The impact of visual impairment during early life course is complex and dynamic, impacting an individual at the level of bodily functions, activity and participation as outlined by the ICF. The impact may also be prone to qualitative change as children develop physical capacities to adapt to the functional restrictions of visual impairment and psychological maturity to cope with visual impairment. Transitions or changes during childhood and adolescence mean that YP frequently have to adapt to the impact of visual impairment in varying social and cultural contexts, for example, when facing the transition from education to employment. Vision-specific PROMs which are suitable for use by children and YP are therefore important if ophthalmic professionals want to pick up on the dynamic and changing impact, and provide the most appropriate treatment for children and YP living with visual impairment. Despite the need for age-appropriate vision-specific PROMs, to date there are few vision-specific PROMs which are both age-appropriate and suitable for detecting developmental change, and psychometrically robust for use by children and YP.

There are several more reasons why PROMs capturing the broader impact of visual impairment are needed. Firstly, vision-specific PROMs constitute an important methodological tool in the development and evaluation of new treatments or interventions. For instance, outcomes can be measured before and after administration of a particular intervention to evaluate changes which are attributable to the intervention. For example, both VQoL and FV may be valuable outcomes when measured before and after low vision rehabilitation services and clinical interventions as subjective outcomes which complement traditional measures of VA. PROMs may

also be valuable in randomised control trials designed to assess the impact of clinical treatment upon subjective outcomes.

Vision-specific PROMs can also be useful in assessing the longitudinal impact or disease course of an ophthalmic disorder by assessing the progressive impact of visual impairment across multiple developmental milestones and time-points. This measurement allows practitioners and health professionals to assess temporal and societal factors associated with change in the impact of visual impairment such as a change in education.

To date, several vision-specific PROMs have been developed for children with visual impairment. In a recent systematic review of PROMs specifically designed for use by children with visual impairment, 17 eligible instruments were identified.[257] However, the development of these instruments has been influenced substantially by methodological challenges such as defining VQoL and FV, and ensuring the measures conform to psychometric standards. The identified instruments differ substantially in the outcomes that they capture. Six measures were generic, capturing the impact of visual impairment in children irrespective of eye condition/disease and 11 were ophthalmic-condition specific. Seven instruments were designed to measure VQoL and three to measure FV or visual ability. The remaining instruments measured varying outcomes related to visual impairment, including the impact of treatment for amblyopia, symptoms, psychosocial impact, and well-being. Others such as parents, adult stake holders, and sighted children often had input in the item development stage rendering the instrument items potentially inappropriate for children with visual impairment. Only a small number of instruments identified had shown sufficient psychometric properties such as evidence of different types of validity and reliability[151] (see Section 2.4, pg. 36).

Since that review was published, five new vision-specific instruments have been developed for use by children. Table 2 (pg. 54) shows an updated description of all-cause generic visual impairment and eye disorder-specific instruments. Three of these more recent instruments are generic and report to capture activity and participation,[258]

vision-related difficulties[259] and functional vision.[6] However, only one of these conforms to psychometric standards. This instrument was developed during the foundation research, and designed to capture FV among children aged 10-15 years. The remaining two instruments are eye disorder-specific (designed for use by children with amblyopia[260] and refractive error[261]) and one does not differentiate between VQoL and FV.[260] Thus, development of PROMs which are both vision-specific and age-appropriate is an emerging, yet methodologically challenging field of research.

Table 2. Updated description of all-cause generic visual impairment and eye disorder-specific PROMs.

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
Cardiff Visual Ability Questionnaire for Children (CVAQC)[262-264]	2010, UK	English, Chinese, Turkish	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)[265-267]	2004, USA	English, German	Visual impairment	0-7 years	Proxy	Researchers	35 (age <3); 40 (age 3-7)	Vision-related quality of life	FA 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)[213, 268]	2011, Australia	English	Visual impairment	8-18 years	Child	Children, parents and teachers	24	Vision-related quality of life	Rasch-derived unidimensional scale capturing the impact of visual impairment on participation in daily activities
LV Prasad-Functional Vision Questionnaire (LVP-FVQ)[269]	2003, India	English, Hindi, Telugu	Visual impairment	8-18 years	Child	Children, parents, clinicians and researchers	20	Functional vision	Rasch-derived unidimensional scale capturing visual ability/ functional vision

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
LV Prasad-Functional Vision Questionnaire Second Version (LVP-FVQ II)[270]	2012, India	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 and Young People (VQoL_CYP)[5, 7]	2011, UK	English	Visual impairment	10-15 years	Child	Children	35	Vision-related quality of life	Rasch derived scale capturing vision-related quality of life
Amblyopia Treatment Index (ATI)[271]	2001, USA	English	Amblyopia	3-13 years	Child and Proxy	Parents, clinicians and researchers	20 (proxy version, age 3-6) 19 (child version, age 7-13)	Impact of amblyopia treatment	Parent version: FA derived multi-dimensional scale with the following subscales relating to treatment of amblyopia: (1) Adverse effects, (2) Treatment compliance and (3) Social stigma Child version: FA derived multi-dimensional scale with following subscales relating to treatment of amblyopia: (1) Adverse effects, (2) Treatment compliance and

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
									The Amblyopia Treatment Index (ATI) continued… (3) Functioning at near
Children's Amblyopia Treatment Quality of Life Questionnaire (CAT-QoL)[272, 273]	2011, UK	English	Amblyopia	5-7 years	Child	Children	3 items (across 7 different scenarios)	Quality of life	Impact of amblyopia upon daily life (not derived psychometrically)
Convergence Insufficiency Symptom Survey (CISS)[274]	1999, USA	English	Convergence insufficiency	9-18 years	Child	Children and researchers	15	Symptoms	Unidimensional scale capturing symptom severity (not derived psychometrically)
Effects of Youngsters' Eyesight on Quality of Life (EYE-Q)[275-277]	2010, USA	English	Juvenile idiopathic arthritis-associated uveitis	8-18 years	Child	Children and 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)[278]	2004, UK	English	Amblyopia	<67 months	Proxy	Parents and clinicians	15	Impact of amblyopia treatment	Multi-dimensional scale capturing the impact of amblyopia treatment in form of following subscales (not derived psychometrically):

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
									Emotional Impact of Amblyopia Questionnaire (EIAQ) continued… (1) Child's experience, (2) Family's experience and (3) Child's well-being
Intermittent Exotropia Questionnaire (IXTQ)[279-281]	2010, USA	English	Intermittent exotropia	5-17 years	Child and Proxy	Children and parents	12	Health-related quality of life	Child version: Unidimensional scale capturing quality of life of children (not derived psychometrically) Parent version: FA derived multi-dimensional scale with following subscales: (1) Function, (2) Psychosocial effects and (3) Surgery
Nasolacrimal Duct Obstruction Questionnaire (NLDO)[282]	2006, USA	English	Nasolacrimal duct obstruction	6-48 months	Proxy	Parents and clinicians	29	Symptoms and quality of life	A priory determined two-dimensional scale with the following subscales: (1) Symptoms and (2) Child's health-related quality of life

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
Pediatric Refractive Error Profile (PREP)[283, 284]	2006, USA	English	Refractive error	8-18 years	Child	Not reported	26	Vision-related functional and well-being	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)[285, 286]	2002, UK	English	Amblyopia	3-5 years	Proxy	Parents and clinicians	10	Perceived psychosocial well-being of child	Item details not available
Psychological Impact Questionnaire (PIQ)[287]	2006, UK	English	Amblyopia, strabismus, or refractive errors	16-18 years	Child	Children and researchers	8 items (across 4 scenarios)	Psychological impact	Unidimensional scale capturing psychosocial impact of strabismus (not derived psychometrically)
Quality of Life in Children with Vernal Keratoconjunctivitis (QUICK)[288]	2007, Italy	Italian	Allergic conjunctivitis	5-12 years	Child	Children, parents, clinicians and researchers	16	Quality of life	FA derived two-dimensional scale with following subscales: (1) Symptoms and

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
									Quality of Life in Children with Vernal Keratoconjunctivitis (QUICK) continued… (2) Daily activities
Children's Vision for Living Scale (CVLS)[260]*	2013, Saudi Arabia	English	Amblyopia	5-12 years	Child	Children, paediatric eye care professionals and researchers	28	Vision-related quality of life	Rasch-derived unidimensional scale capturing: (1) Mood, (2) Self-esteem, (3) Social relations, (4) Functional vision, (5) Visuo-motor function and, (6) Academic performance
Student Refractive Error and Eyeglasses Questionnaire (SREEQ)[261]*	2014, USA	English	Refractive error	10-20 years	Child	Multi-disciplinary research team	38	Vision-related quality of life	Rasch-derived unidimensional scale capturing the impact of uncorrected and corrected refractive error on vision-related quality of life

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
Participation and Activity Inventory for Children and Youth (PAI-CY) and Young Adults (PAI-YA)[258]*	2017, The Netherlands	Dutch	Visual impairment	0-25 years	Proxy (ages <7 years) and Child/Young adult (ages 7-25 years)	Professionals of low vision rehabilitation centres, parents, young adults, children	44 (age 0-2); 62 (age 3-6); 55 (age 7-12); 58 (age 13-17); 141 (age 18-25)	Activity and participation	Instruments with the following sub-scales: 0-2 years: (1) Bonding, (2) Incentive processing, (3) Visual attention, (4) Sensorial functioning, (5) Orientation, (6) Play, (7) Mobility, (8) Communication (not derived psychometrically) 3-6 years: (1) Bonding, (2) Incentive processing, (3) Visual attention, (4) Sensorial functioning, (5) Orientation, (6) Motor functioning, (7) Reading and writing, (8) Play, (9) Self-reliance, (10) Mobility, (11) Communication, (12) Social relationships, (13) Day-care/school/study (not derived psychometrically) 7-12 years: (1) Play, (2) Self-reliance, (3) Finances, (4) Mobility,

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
									Participation and Activity Inventory for Children and Youth (PAI-CY) and Young Adults (PAI-YA) continued… (5) Communication, (6) Social relationships, (7) Day-care/school/study, (8) Leisure time, (9) Acceptance/self-consciousness (not derived psychometrically) 13-17 years: (1) Self-reliance, (2) Finances, (3) Mobility, (4) Communication, (5) Social relationships, (6) Day-care/school/study, (7) Leisure time, (8) Acceptance/self-consciousness (not derived psychometrically) 18-25 years: (1) Mobility, (2) Communication, (3) Social relationships, (4) Day-care/school/study, (5) Leisure time,

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
									Participation and Activity Inventory for Children and Youth (PAI-CY) and Young Adults (PAI-YA) continued… (6) Acceptance/self-consciousness, (7) Reading and visual aids, (8) Household living, (9) Finances, (10) Self-care, (11) Computer skills, (12) Intimate/romantic relationships, (13) Peer contact, (14) Holiday and going out, (15) Information/regulations, (16) Applying, (17) Work (not derived psychometrically)
Vision-related symptom and performance checklist for children (VSPCL)[259]*	2013, Japan	Japanese	Visual dysfunction	Details not available	Child and proxy	Details not available	39	Vision-related difficulties	Details not available

Instrument	Year and country	Languages	Eye conditions	Age range	Respondent	Input into items	Number of items	Intended construct and/or domains	Captured construct and/or domains
Functional Vision Questionnaire for Children and Young People with Visual Impairment (FVQ_CYP)[6]*	2013, UK	English	Visual impairment	10-15 years	Child	Children	36	Functional vision	Unidimensional Rasch-derived scale capturing Functional Vision

*new, vision-specific instruments developed for children published since the original review

PROMs which are developed for use by paediatric populations need to be accessible in a format which can be accessed by children with disability, reflecting the emphasis on participation in the ICF. Within the context of visual impairment, for example, PROM development needs to consider different modes of administration with consideration of patients' limited VA for reading printed text, and subsequent participation in this kind of measurement. The same principles apply when children and YP with visual impairment take part in the development stages of PROMs as, by law, this population may require specialist assistance or adaptations such as information sheets and consent forms to be enlarged or printed in Braille, which can add further resource demands on researchers. Thus, ethical considerations applicable to the participation of healthy, sighted children in research (see Section 2.4.1, pg. 38) are somewhat magnified when children with visual impairment are required to take part.

2.6.2 Conceptual and theoretical framework for the development of paediatric vision-specific PROMs

To address the scarcity of psychometrically robust vision-specific PROMs available for use by children and YP with visual impairment, Rahi et al. recently reported research using a child-centred approach to developing two novel PROMs, one assessing VQoL and the other FV (see Section 2.6.1, pg. 51) in children and YP with visual impairment (as per the WHO criteria shown in Table 1, pg. 42).[5-7] The instruments were designed as distinct but complementary measures for capturing different aspects of living with visual impairment: the impact upon functional ability (FV) and the psychosocial-emotional impact (VQoL). In the absence hitherto of an established conceptual framework and child-centred methodology in this area, these measures were developed in the first instance for use by visually impaired children aged 10-15 years. This age group was chosen to ensure children were developmentally capable of self-reporting on the complex issues included in definitions of QoL, HRQoL and VQoL.

This ‘foundation’ research programme demonstrated the feasibility and benefits of involving children with visual impairment in all standard stages of questionnaire design, an approach which is concordant with the values outlined by the ICF. The researchers ensured the items in the PROMs reflected meaning and value for the paediatric population by conducting individual in-depth interviews with children and YP with visual impairment. During interviews, participants were asked to discuss their everyday life in relation to 6 topics: school, home life, activities and socializing, life skills and independent living, eye problems and the eye clinic, and the future. In keeping with definitions of QoL as dependent on individual differences, experiences, lifestyles, hopes, attitudes and beliefs (see Section 2.3, pg. 28) the content of interview discussions were largely child/young person-centred. The second phase involved pre-testing the draft instruments by consulting an expert-reference group of visually impaired children/YP, which was supplemented by an expert consensus meeting of the professionals involved in the development. During consultations, children and YP were asked to reflect on item relevance and comprehensibility, layout of the questions, and administration methods, thus ensuring that completion of the PROMs would be both possible and feasible. The developed instruments were then piloted with an aim of identifying any immediate problems with the questionnaires (e.g. in terms of ceiling or floor effects on particular questions), and finally validated with a nationally representative sample of children and YP with visual impairment through a postal survey. This research demonstrated the feasibility and value of involving visually impaired children and YP in the process of instrument development by grounding the conceptual and theoretical framework for the instruments in their voices and lived experience.

2.7 Research aims

The overarching objective of the research reported in this thesis is to explore the day-to-day impact (both immediate and broader) of visual impairment during childhood and

adolescence and use this knowledge to develop a suite of age-appropriate, vision-specific, complementary PROMs of VQoL and FV for use by adolescents with visual impairment. This research builds directly upon the foundation research by Rahi et al. with 10-15 year olds[5-7] and addresses the gap in the VQoL and FV instrument provision for YP older than 15 years. The reported research uses a conceptual and theoretical framework which is grounded in the voices of YP living with visual impairment. Concordant with social theories of disability, participation of YP with visual impairment is integral to the instrument development, and vital for ensuring the instruments are psychometrically robust for use by YP with visual impairment within ophthalmology services and potentially in the future as outcome measures within clinical trials. The research stages outlined in Section 2.6.2 (pg. 64) afford the opportunity to explore, in detail, the experience of growing up with visual impairment, adopting a life course perspective and drawing on the views of visually impaired adolescents (through in-depth individual qualitative interviews) who are well-placed to retrospectively reflect on their experience of visual impairment. In parallel to the current research project, instruments designed for children younger than 10 years were developed with a view to address the entire spectrum of children and YP living with visual impairment for which age-appropriate, vision-specific PROMs are not readily available.

2.8 Summary

Biological and psychological perspectives demonstrate the dynamic nature of growth, development, learning, and change during childhood and adolescence. Specifically, adolescence is a period associated with a number of age-specific issues as YP experience changes; both intrinsic (related to psychosocial and emotional well-being), and extrinsic (related to the broader social and cultural world in which they develop). When a child or adolescent is living with some form of disability, such as visual impairment, everyday life is likely to be influenced in a number of ways. Despite evidence for the dynamic impact of visual impairment during childhood and

adolescence, there are limited, vision-specific, age-appropriate and psychometrically robust PROMs which are readily available for improving outcomes in paediatric ophthalmology services. In particular, adolescence is a period of life course described as qualitatively unique from both childhood and adulthood. Thus, age-appropriate PROMs that are grounded in the experiences, lifestyles, attitudes, and beliefs of YP living with visual impairment are needed to accurately capture the impact of visual impairment during this period. The aim of the work reported in this thesis is to address this gap, adding to the broader understanding of the impact of visual impairment during childhood and adolescence, and enhancing the conceptualisation and measurement of the impact of visual impairment within paediatric ophthalmology.

Chapter 3 Psychometric measurement in healthcare

As discussed in Section 2.3 (pg. 28), measurement has always been a fundamental component of healthcare and is important for improving the delivery of healthcare, and allowing policy makers to assess major health problems and set goals. The aim of this chapter is to outline the underlying principles of measurement which must be considered when developing PROMs as measures of the subjective impact of disability. The strengths and weaknesses of each approach will be highlighted as a basis for explaining the psychometric approach used in this research.

The earliest evidence of measures of population health in England and Wales date back to mortality rates which were collected during the 17th century as a result of the plague.[289, 290] However, as previously discussed, alongside and likely as a result of growth and development in information technology, measurement in healthcare has exploded in terms of the quantity of measurement instruments available, the ways measurement can be implemented, and the outcomes which can be captured. Today, electronic health records are increasingly implemented within both primary and secondary healthcare settings worldwide.[291, 292] Electronic databases such as the Hospital Episode Statistics[293] contain vast amounts of clinical data collected from the population and are free to access via the internet. Although delivered electronically, with vast improvements in the documentation and indexing of data, the content of these databases (i.e. numeric quantifiable data relating to mortality rates and hospital admissions) is largely comparable to data which were collected in the 17th century. Using electronic databases containing this type of data, scientists and statisticians can use sophisticated statistical methods to make inferences and associations which are essential for monitoring health at the level of the population. Today, these methods can be performed quickly and with ease, using statistical software such as SPSS, STATA or R.

3.1 Change in the nature of measurement

In the past 172 years (since the first statistical representation of life expectancy was constructed[294]), life expectancy has increased in all countries and in 2011, reached almost double of that in 1841.[295] As a result of this increase, data conceptualising aspects of health, such as QoL (versus length of life) are increasingly valued, and there is growing appreciation within healthcare, that treatment methods and interventions should be aimed at enhancing QoL among patients suffering from disease or illness. To measure QoL, we need to capture psychological outcomes which extend beyond numeric, quantifiable data used for the management of public health. This approach represents changing focus from the population to the individual, and in doing so, views patients as the gatekeepers to the *experience* of disease or illness. To systematically capture the experience of disease or illness, one might use a self-report questionnaire.

The purpose of such self-report questionnaires within healthcare has changed over time. Their earliest use focused upon exploring patients' attitudes and analysing how a range of attitudes was distributed within a population.[296] These questionnaires placed little value on scoring or quantitative analyses, and instead focused on capturing patients' verbal answers to open-ended questions, analysing the meaning behind their responses, and assessing their linguistic choices. Whilst these measures are essential for gaining rich insight as to the experience of disease or illness, statistical analyses are necessary to make comparisons between individuals or populations, and cannot be used to analyse the data provided by them. Thus, to make self-report outcomes, such as PROMs, optimal for use within healthcare, researchers involved in instrument development must resolve two incompatible notions: a) that data captured should represent complex, multidimensional, and subjective outcomes, such as HRQoL, which are theoretically underpinned by individual experience (see Section 2.3, pg. 28) and b) that the data should be captured in a way which is clinically beneficial, allowing practitioners and researchers to quantify outcomes, make associations, and compare individuals and populations.

The question ‘Is psychological measurement possible?’ is one which has been debated since the 1930s by scientists such as Norman Campbell[297] and Stanley Stevens[298] who argue as to whether measurement, which is a key component of physical science, can be applied to the domains of social science. Additive measurement, specifically, is a term used to describe measurement which is based upon equal units. For example, a scientist can easily apply additive measurement when measuring a patient’s height or weight. Here, addition of one inch or one pound clearly involves adding one to the measurement number e.g. the patient’s height in inches or weight in pounds. However, adding one unit in physical science (using height, weight, temperature or distance, for example) is conceptually different from adding the same amount in different disciplines e.g. social science.[297] Adding the same amount within social science involves first defining what the constant amount is. When measuring attitudes or experience of illness or disease, scientists and researchers must define what one unit of attitude or experience is. This was a major challenge for most social scientists in the 1930s, and attempts to answer the question ‘Is psychological measurement possible?’ subsequently failed.

Since the debate in the 1930s, science has progressed to the point to which we now understand more about how to measure psychological outcomes such as QoL. Today, PROMs (as described in Section 2.3.1, pg. 31) contain response formats which convert subjective attitudes or emotions into quantifiable outcomes. Likert-type scales are often used, in which verbal response categories are assigned numeric values (e.g. strongly disagree = 1, disagree = 2, agree = 3 etc.). Responses given by patients to single items are usually added to produce a summary score or summary index. Measurement of subjective self-report in this way has a number of benefits in healthcare which are outlined in Section 2.3.1 (pg. 31). The most important benefit is that outcomes derived from subjective self-report, can be collected, handled and analysed as though they are numeric, quantifiable data. This confers a number of statistical benefits, namely the use

of stronger inferential statistics and more powerful analyses to assess trends between individuals and populations.

However, in order for PROMs to be used in this way, they must be developed using a controlled, systematic approach, following the criteria outlined by Classical Test Theory (CTT),[299] Item Response Theory (IRT),[300] or Rasch Model Theory (RMT).[301] If the criteria outlined by these approaches are met, researchers can be sure that the questionnaire is reliable, valid (see Section 2.4, pg. 36),[158] and responsive to change in the unobservable characteristic (e.g. attitude or experience). Most importantly, researchers can use these approaches to define the constant amount or, in the context of social science, what one unit of attitude or experience is, and thus ensure that the questionnaire conforms to additive measurement.

3.2 Classical Test Theory

Classical Test Theory (CTT) is a traditional approach to questionnaire development which was first developed in the early 1900s,[158, 302] and is used to ensure instruments such as questionnaires are high quality (i.e. valid, reliable and responsive to change). There are several key features of CTT but the most important feature is the ability to detect scales which exist within a questionnaire by grouping items. Items are grouped by assessment of the correlation between pairs of items. As a result of item grouping, scale reliability is either confirmed or refuted.

FA[303-306] is the primary statistical method associated with CTT to assess the degree to which groups of items share a common core of information about the true score.[153] In doing so, FA indicates how many scales (i.e. groups of items correlating highly with each other) exist within a single questionnaire and thus indicates dimensionality of the instrument. In order to make inferences about a unit of measurement on an unobservable, psychological outcome, such as HRQoL, the items which make up the instrument must be unidimensional (i.e. they must all measure the same underlying construct). FA works by identifying the characteristics along which items differ

substantially and therefore identifies similarity between items. Maximum likelihood is a statistical method that fits FA models to multivariate observations to determine how many factors (or scales) exist within one instrument. Despite there being some form of statistical criteria for identifying the number of factors present in one instrument, researchers usually interpret outcomes from FA subjectively and iteratively, i.e. judge whether the number of factors determined by FA is conceptually valid and in line with their pre-specified hypotheses.

Throughout the history of questionnaire development, CTT has been a very popular approach to measurement theory and thus, the development of PROMs. This may be because it is relatively easy to understand by those who have some basic understanding of statistics. For example, the coefficient alpha which is used to represent reliability in FA is widely encountered in other statistical methods and may therefore be easy for researchers to interpret in a meaningful way. There are also many readily accessible statistical packages which can be used to perform FA.

Despite CTT being a popular, traditional approach to instrument development, validation, or reduction, it is increasingly recognised that this approach does not address the underlying issue with measurement in psychology; that is, FA *assumes* that the outcome can be measured using an additive model and that responses are linear and merely assesses the similarity *between* items.[307] Thus, FA gives little indication as to whether the items or responses follow a hierarchical structure. Some researchers have proposed that popularity of CTT peaked at a time when researchers focused more on improving the quality of questionnaires, and addressing properties such as validity and reliability, than on the measurement construct itself.[296] Additionally, several authors have argued that, rather than representing true reliability, the Cronbach's alpha value used in CTT for internal consistency reliability, which simply reveals the average degree of 'interrelatedness' between items, leaves little indication of the true unidimensionality of the measure, that is whether the items are similar in terms of the factors they are assigned to.[308]

Perhaps one of the most well documented shortcomings of CTT, is use of Cronbach's alpha as an indicator of the instrument's strength. The underlying assumption in CTT is that the activities/constructs which are captured by individual items in a questionnaire are of equal difficulty. As a consequence, variance between outcome 'summary' scores or indexes are attributed solely to variations between levels of ability among respondents.[307, 309] In reality, PROMs are often designed to capture outcomes such as self-report functional ability and consequently include items which differ in how easy or hard they are for respondents of different ability. This is necessary to ensure PROMs are applicable to patients who have a range of functional abilities, including those who are severely limited, and also those who have only minor difficulties. Thus, variation in respondent ability is unlikely to be the single cause of variation in responses. Assuming that the items are of equal difficulty introduces some measurement bias and may lead to incorrect conclusions within clinical practice.

3.3 Modern Test Theory

Identification of the shortcomings of CTT, and enduring debate surrounding the theoretical application of measurement in social science, has triggered application of a new approach to measurement, representing growth in understanding of theory and methods underlying measurement. Two important developments in test theory appeared in the 1960s. One was set forward by Rasch[156, 301] and the other by Lord and Novick.[300] The latter is termed Item Response Theory (IRT), and the former Rasch Measurement Theory (RMT). Within current literature, there is a broad understanding that CTT, IRT and RMT are fundamentally different from each other, each embodying different paradigms in terms of theory of measurement and techniques used to ensure an instrument truly measures an unobservable construct, such as QoL.

Unlike CTT which *assumes* linearity, modern test theory (both IRT and RMT) articulates the conditions under which interval level data can be estimated using the measurement instrument,[310] and can therefore be used to *construct* linearity and

subsequently ensure that the developed instruments can be used for additive measurement. Additionally, modern test theory specifies a probabilistic model which, unlike the assumption made by Cronbach's alpha in CTT, is based on the understanding that not all items included in a questionnaire are of equal difficulty.[311] In this way, modern test theory addresses the shortcoming of CTT when summarising performance on a measure and attributing it exclusively to variation in person ability.

When used for the development of PROMs, modern test theory has been shown to provide more information about the abilities of the full range of the population (with CTT only showing floor and ceiling effects), the suitability of the response options (for which CTT does not provide any formal analysis), and validity of the instrument, showing fit statistics, expected responses and local independence i.e. the item scoring bias resulting from similar items being included in the same instrument (with CTT merely showing inter-item correlations).[312] Thus, modern test theory (including both IRT and RMT) is broadly accepted as a theoretical and methodological extension of CTT which is increasingly applied within PROM developments.[309, 313]

3.3.1 Different approaches in modern test theory

Whilst similar in terms of contribution to current understanding surrounding test theory, and sharing many of the benefits they bring to instrument development, the techniques which fall under the umbrella term of modern test theory, must be considered as two distinct approaches with several pertinent differences.

The primary difference between IRT and RMT is in their underlying paradigm.[311, 314] When using IRT, researchers search for a model which best describes the pre-defined data. Often, a model with a greater number of parameters is chosen first (as the best fitting model). Statistical checks are then performed to try and reduce the model parsimoniously to have a smaller number of parameters and a comparable predictive power.[314] This paradigm is often taken for granted as the most appropriate approach within any form of statistical analysis but is increasingly endorsed within the context of

instrument development. When used to validate an instrument however, this paradigm may encourage researchers to neglect post-hoc analyses to modify the data (i.e. improve the instrument by re-wording or removing items).[314]

In contrast, the paradigm underlying RMT is that individual responses to the ratings included in PROMs, that is, the data collected, are analysed according to the model of measurement. The theory or model of measurement was first defined by Rasch[311] as allowing for the separation of person and item parameters and thus sufficiency is a key characteristic. Using the notion of sufficiency, Rasch developed models which could be used to provide a probability distribution of responses given by an individual of a specific ability which is dependent only upon the difficulty of the item[301] i.e. the model of measurement. In practice, if data do not fit the model of measurement, post hoc adjustments are usually made iteratively to explore the measurement construct in more detail, and to assess which items need to be removed or amended, thus improving the instrument.

3.3.2 Characteristics of Rasch analysis

In practice, an additional quality of RMT or Rasch analysis is that it can be used to perform assessments of the quality of measurement instruments which are beyond those which can be explored using CTT or IRT.[314] This makes Rasch analysis increasingly favoured within the domain of instrument development. For example, using Rasch analysis, researchers can analyse the response-scales, -thresholds, and -categories to ensure they are optimal for capturing outcomes which are sensitive to the needs of respondents. In doing so, researchers can be sure that the ordering of the response categories is empirical. Additionally, Rasch analysis allows for assessment of differential item functioning (DIF): an indication that respondents with comparable levels of ability are responding differently to an item, and that a particular item should be removed. Thus, Rasch analysis can be used as a robust measure of construct validity, reliability (person, and item) and responsiveness[313] (see Section 2.4, pg. 36)

which is grounded in strong foundations from measurement, test, and item-response theory.

There are two models which can be used in Rasch analysis, offering additional flexibility for researchers developing questionnaires which differ in the nature of response options. The Andrich rating scale model is used when instruments contain items for which patients respond using the same response scale across all items.[311] The partial credit model differs in that it can be used when items vary in terms of their response format.[315] Similarly, there are dichotomous and polytomous versions of Rasch analysis which can be used when instruments contain items with only two response options (dichotomous) and items which contain more than two response options (polytomous). Several statistical packages exist for performing Rasch analysis (e.g. RUMM, Winsteps, R), allowing researchers to tailor the analysis to the needs of the research project. There are also a number of guidelines which can be used by researchers when improving the quantitative functioning and measurement performance of the rating scale. The most pertinent guideline refers to the amount of numerical information which indicates to what extent the data produce coherent raw scores e.g. raw scores that support the Rasch model of measurement. At least 10 observations of each response category is recommended to ensure the step calibration is stable when using the Andrich Rating Scale model with polytomous items.[316, 317]

3.4 Current consensus on the optimal approach

Currently, there is broad acceptance by those who specialise in measurement as a scientific discipline that a direct comparison of the techniques involved in CTT, IRT and RMT, such as FA and Rasch analysis, for the purpose of examining unidimensionality is not very useful. This is because these comparisons encourage researchers to ignore the theoretical differences underpinning the three approaches i.e. the conceptualisation of measurement, and instead search for a method which best describes their data. This approach is often termed the ‘explain the data’ paradigm, which is in contrast to

modifying the data or the measurement instrument to fit the philosophy of measurement.[314] Current consensus is that both IRT and RMT approaches provide more information than using CTT alone,[312] however, when selecting an approach, researchers should carefully address the aims and objectives of the research study, as well as the theoretical underpinnings of CTT, IRT and RMT and the inferences which can be made when using each technique.

The method used in the current development of vision-specific, age-appropriate PROMs suitable for YP is underpinned by a combination of both CTT and RMT, with the former being used as purely an indication of the feasibility of applying Rasch analysis in the later stages of evaluation to assess the psychometric properties of the instruments. The exact statistical techniques which are applied are discussed in more detail in Section 4.9.4 (pg. 114). In summary, FA will be performed first to give an indication as to whether unidimensionality exists. If unidimensionality does exist, Rasch analysis will be performed as a stronger psychometric evaluation of the instruments. If unidimensionality is not demonstrated using FA, items showing floor or ceiling effects will be iteratively removed with the aim of preparing the instrument for Rasch analysis. This approach is concordant with the theoretical underpinnings of modern test theory, and the iterative process in which measurement instruments such as PROMs are tailored for optimal use.

Chapter 4 Method

4.1 Ethics approval

This research project was approved by the NHS Research Ethics Committee for Essex and East of England (ref: 12/EE/0455) and adhered to the tenets of the Declaration of Helsinki.

4.2 Phases of instrument development

The research was undertaken in 4 standard stages of instrument development (as outlined in Section 2.6.2, pg. 64) comprising in-depth interviews conducted with the aim of identifying the content of instrument items (phase 1), expert consultations to explore participants' understanding and interpretation of the draft instruments (phase 2), a small-scale postal survey to identify any immediate problems with the instruments (phase 3), and a large-scale postal survey to validate the final instruments (phase 4).

4.3 Eligibility criteria, patient identification, and recruitment

4.3.1 Eligibility criteria

Disorders leading to visual impairment during childhood can be classified using the following characteristics:

- Degree of visual impairment e.g. severity (VI, SVI, or Blind)[178]
- Timing of onset of impaired sight (early onset at ≤ 2 years of age, late onset at ≥ 3 years of age)
- Stability of visual deterioration (stable impairment or progressive)

The overarching aim of this study was to understand the *experience* of children and adolescents living with visual impairment, and quantify the impact of all-cause visual impairment using vision-specific PROMs. This is in contrast to disease-specific PROMs

which can be used to quantify the impact of a specific, or range of specific, ophthalmic disorder(s). Additionally, patients were required to be capable (developmentally, cognitively, and physically) of self-reporting on VQoL and FV. Therefore, patients were eligible if they met the following criteria:

a) Visually impaired, severely visually impaired, or blind (VA in the better-seeing eye logMAR equal to or worse than 0.5 or Snellen worse than 6/18)

b) No other significant sensory, learning or motor impairment which should impact upon ability to self-report

c) Aged between 13 and 18 years on date of recruitment.

Given the probability of some developmental delay in this population (see Section 2.5, pg. 41), a 'stage'- as opposed to 'age'-approach may be more suitable for children and YP living with long-term disability or impairment. Thus, precise age at the time of data collection was subject to discretionary flexibility. Flexibility was also deemed appropriate with regard to manifestation of visual impairment, and medical histories and nature of visual impairment were evaluated in each individual patient identified to determine suitability for inclusion in the study. Thus, some patients who had visual impairment classified as mild visual impairment based on acuity (see Table 1, pg. 42) but with other manifestations of visual impairment (such as severely reduced visual fields) were invited to take part. This was with a view that YP living with visual impairment in the UK, and fulfilling the eligibility criteria, are a numerically small population and the developed instruments may be used flexibly within clinical contexts.

Before participants were invited to take part in the research, the family GP was contacted to ask if there were any reasons why the research team should not contact the family (see Appendix II, pg. 348).

4.3.2 Sampling sources

The research was carried out in 4 standard phases of questionnaire development (see Section 2.6.2, pg. 64) and the potential participants were sampled across the phases from two main sources described below.

Source 1:

The Department of Ophthalmology at Great Ormond Street Hospital, and the Paediatric Glaucoma Service and Genetic Eye Disease Service at Moorfields Eye Hospital, London, UK were the primary source of eligible patients used across all research phases. Patient attendance lists containing the hospital numbers of all patients who had attended relevant paediatric ophthalmology clinics were scrutinised at the outset to identify patients as per the eligibility criteria outlined in Section 4.3.1 (pg. 78).

Source 2:

During the latter stages of the research project (phase 3 and phase 4), a number of external NHS Trusts were included in the research as patient identification centres (PICs). A total of 20 PICs were identified and recruited through the Paediatric Ophthalmology network and NIHR Clinical Local Research Networks in the UK (see Appendix I, pg. 347). The centres represented an even geographic spread of England, Ireland and Wales. Every centre was required to obtain local research governance approval prior to their participation as a PIC. The purpose of involvement of these centres in the identification and recruitment of eligible patients for phases 3 and 4, where the largest sample sizes were anticipated, was both to ensure a nationally representative sample as well as a sufficient sample size.

All consenting patients identified through these external centres were reported as NIHR patient accrual for the identifying PIC.

4.3.3 Sampling framework

One sampling framework was developed comprising patients from source 1.

Source 1: The clinical and demographic details of eligible patients were collated into a sampling framework for the study. The patient identification from source 1 was continuous throughout the project with the relevant details systematically updated using electronic health records from Great Ormond Street Hospital and Moorfields Eye Hospital, London, UK. Particular attention was paid to recording the outcome of the most recent assessment of VA. Demographic details such as date of birth, and contact details such as current/most recent address and contact telephone number were also recorded. Each entry in the framework was assigned a random number using the random number generator function in Excel, and the entire sampling framework was organised in order of random number (ranging from smallest to greatest). When organised in this way, each entry was assigned a participant study ID or 'PID', and the sampling framework was then re-organised in terms of PID (from smallest to largest). The PID was obtained throughout the course of the research project, and used to identify patients in documents such as consent and assent forms. This was compliant with data protection regulations and necessary to ensure no identifiable information was used when the patients and their families were discussed within the research team.

Source 2: In phases 3 and 4, lists of eligible patients identified at each PIC were collated by the collaborating local clinical team and kept on site as per research governance regulations.

Recruitment procedures across these phases are outlined in more detail in Sections 4.6.1, (pg. 83 (phase 1)), 4.7.1, (pg. 104 (phase 2)), 4.8.1, (pg. 107 (phase 3)), and 4.9.1, (pg. 111 (phase 4)).

4.4 Sample size

An anticipated participation rate of YP with visual impairment based on prior similar studies in which children and YP with visual impairment participated,[5, 240] of approximately 30% was applied to inform the number of patients invited to participate

in each stage of instrument development. To ensure that results were representative of the wider population of visually impaired adolescents, sample sizes in phases 1 and 2 were calculated based on the principles of data saturation during qualitative research methods.[318] Throughout the course of the research project and 4 phases, caution was taken to avoid inviting too many YP to take part in each individual phase in light of the numerically small population, moderate participation rates and with a view to conserve the majority of the sample for the final validation phases.

4.5 Participation analysis

A logistic regression model was fitted to examine the characteristics associated with participation in the 4-phases of instrument development using age, gender, severity of visual impairment, ethnicity, and index of multiple deprivation (IMD) as potential predictors. Severity of visual impairment was categorised based on the WHO classification (see Table 1, pg. 42) but included 'low vision' to account for patients who had VA better than logMAR 0.46 but with other manifestation of visual impairment (e.g. significantly reduced visual fields)(see Table 3, pg. 82). Univariable analyses were performed on each predictor variable individually. Variables which were shown to be significant (p <.05) were then entered into multivariable analyses.

Table 3. Categories used to rank severity of visual impairment.

Visual impairment category	**Presenting corrected distance VA (logMAR) in the better eye**
Low vision (LV)	Better than logMAR 0.48
Visual impairment 1 (VI1)	0.48 – 0.70
Visual impairment 2 (VI2)	0.72 – 1.00
Severe visual impairment (SVI)	1.02 – 1.30
Blind	Worse than logMAR 1.30

4.6 Phase 1: Item development and adaptation

The aim of this phase was to explore the lived experience of visual impairment during childhood and adolescence and identify the issues relevant to YP aged >15 years with a view to develop age- and vision-specific questionnaire items suitable for inclusion in the measures of VQoL and FV. Concordant with the most recent, psychometrically robust, conceptual and theoretical development of vision-specific PROMs, a series of in-depth interviews was conducted with adolescents.

4.6.1 Participants and recruitment method

Participants were recruited for participation using source 1 (see Section 4.3.2, pg. 80). A required sample size of approximately 15-20 participants was calculated based on principles of data saturation in qualitative research.

Thus, 44 eligible patients were identified using a systematic random sampling approach. Participants aged 16 years or above were selected from the sampling framework based on the premise that interviews with YP aged 13-15 years had already been conducted during the 'foundation' research[5, 6, 240] and the interview transcripts from these interviews were already available. A stratified random sampling approach was used to ensure an even spread of patients were invited to take part in this phase with regard to age and gender. When ordered in terms of PID (ranging from smallest to largest), the first patient was reviewed in terms of age and gender for inclusion in the sample. The same method was used to identify subsequent patients, with each patient who fulfilled the pre-established sample characteristics for the phase (in terms of age and gender) recorded. If the patient in the sampling framework did not fulfil the sample requirements for the phase (e.g. violated the even spread of age and gender), the next patient was assessed for suitability, and so on.

Identified patients were sent invitation packs by post inviting them to take part only in this research phase. Each invitation pack was addressed to the parent of the young

person, and included a separate individually sealed letter addressed to the young person themselves. Both the parent and the young person invitation pack contained:

a) an invitation letter
b) a study information sheet
c) a consent form
d) a pre-paid envelope

Parent invitation packs also included a family background questionnaire to collect data about the family context including parent background, occupation, and details of the family structure e.g. single/two-parent family, and any other siblings (see Appendix IX, pg. 371).

All invited families were followed-up by telephone 2 weeks later. If patients were willing to be interviewed arrangements were made first over the phone with a parent or guardian, and then confirmed in writing. A reminder telephone call was made the day before each interview to ensure that each family was aware they would receive a visit from a researcher the following day.

The families of all patients whom the researcher had been unable to contact via telephone were sent a postal reminder 4 weeks following the initial invitation. No additional reminders were sent, in accordance with the ethics approval.

4.6.2 Identifying the issues relevant to YP with visual impairment

4.6.2.1 Topic guide development

The objective of phase 1 was to identify the issues relevant to adolescents living with visual impairment with a view to inform the development of age- and vision-specific items for the VQoL and FV instruments. Thus a flexible topic guide for interviewing YP to capture their needs and concerns was developed. This drew on a systematic review of the literature with the research question ‘What factors contribute to the broader impact of living with a visual impairment during adolescence?’ This review was

supplemented by consultations with professionals working within paediatric ophthalmology.

4.6.2.1.1 Identification of literature

A literature review was conducted using the matrix method to collect and organise findings.[319] PubMed, PsycINFO and EMBASE databases were searched systematically with the main concepts being a) vision, visual impairment or eye disease, b) adolescents/young adults and c) Quality of Life, daily life activity or life satisfaction (see Appendix X, pg. 375 for the search strategy and MeSH terms). The following limits were applied to each search:

- Articles available in English language only
- Humans only

In addition to the main concepts, the limit 'age group' (Child: 6-12 years, Adolescent: 13-18 years, and Young Adult: 19-24 years) was applied in PubMed, and 'human age groups' (School Child <7 to 12 years>, and Adolescent <13 to 17 years>) in EMBASE. This is in keeping with the WHO definition of adolescence as the period between the ages of 10-19 years (see Section 2.1, pg. 20). No age-related limits were applied in PsycINFO due to the small number of results.

The title and abstract of each reference was reviewed for relevance to YP living with visual impairment and relevant citations were exported to EndNote citation manager. Duplicates were removed.

The search was carried out in March 2015.

4.6.2.1.2 Screening of literature

The full text of 284 articles was screened to assess the empirical value of the results. Three systematic reviews were excluded from the search (2 because findings were not applicable to the topic guide[257, 320] and 1 because the author failed to identify any relevant papers[321]) and 3 editorials were excluded.[322-324] One study protocol was

identified and excluded.[325] A grey literature search was conducted using the reference lists of all remaining eligible articles, including the excluded systematic reviews, editorials and study protocol. Thirteen articles were identified through grey literature, and 5 additional articles referenced in the study protocol were added.

4.6.2.1.3 Eligibility of literature

The aim of the systematic review was to identify, as broadly as possible, factors related to QoL and everyday life of adolescents living with visual impairment. Thus, the review focused upon literature which is both vision- and age-specific. This is in line with using the topic guide for interviewing YP aged 10-15 years[5, 6] as a foundation for developing new questions and probes for YP aged older than 15 years. Despite including adolescents/young adults as a main concept in the review (see Appendix X, pg. 375), and using the search limits to focus upon age-range, a substantial number of articles were removed during the eligibility stage because they did not focus specifically upon individuals within the specified age range of 10-19 years. The majority of these included adolescents or YP who were classified as 'adults' and placed within the age range of 18 years +. In these cases, attention was paid to any analyses between age-groups. Those which did not discriminate between age-groups were excluded. After assessing eligibility in terms of the age of participants, three articles were excluded because participants did not have a visual impairment.

During the second stage of eligibility assessment, attention was paid to identifying papers which used qualitative techniques, such as in-depth interviews or focus groups, which yielded data based on self-report. Some papers used measurement instruments which provided quantitative summary scores indicating overall QoL, or subscales of QoL, which were used to compare groups of YP with varying demographic characteristics (e.g. age, gender, severity of visual impairment). In this case, each article was assessed in terms of the contribution it made towards distinguishing specific aspects of VQoL or FV which could be probed during interviews with YP. Since probing

QoL itself (e.g. “Tell me about your QoL”) would likely be confusing for YP, 29 were excluded as shown in Figure 1 (pg. 88).

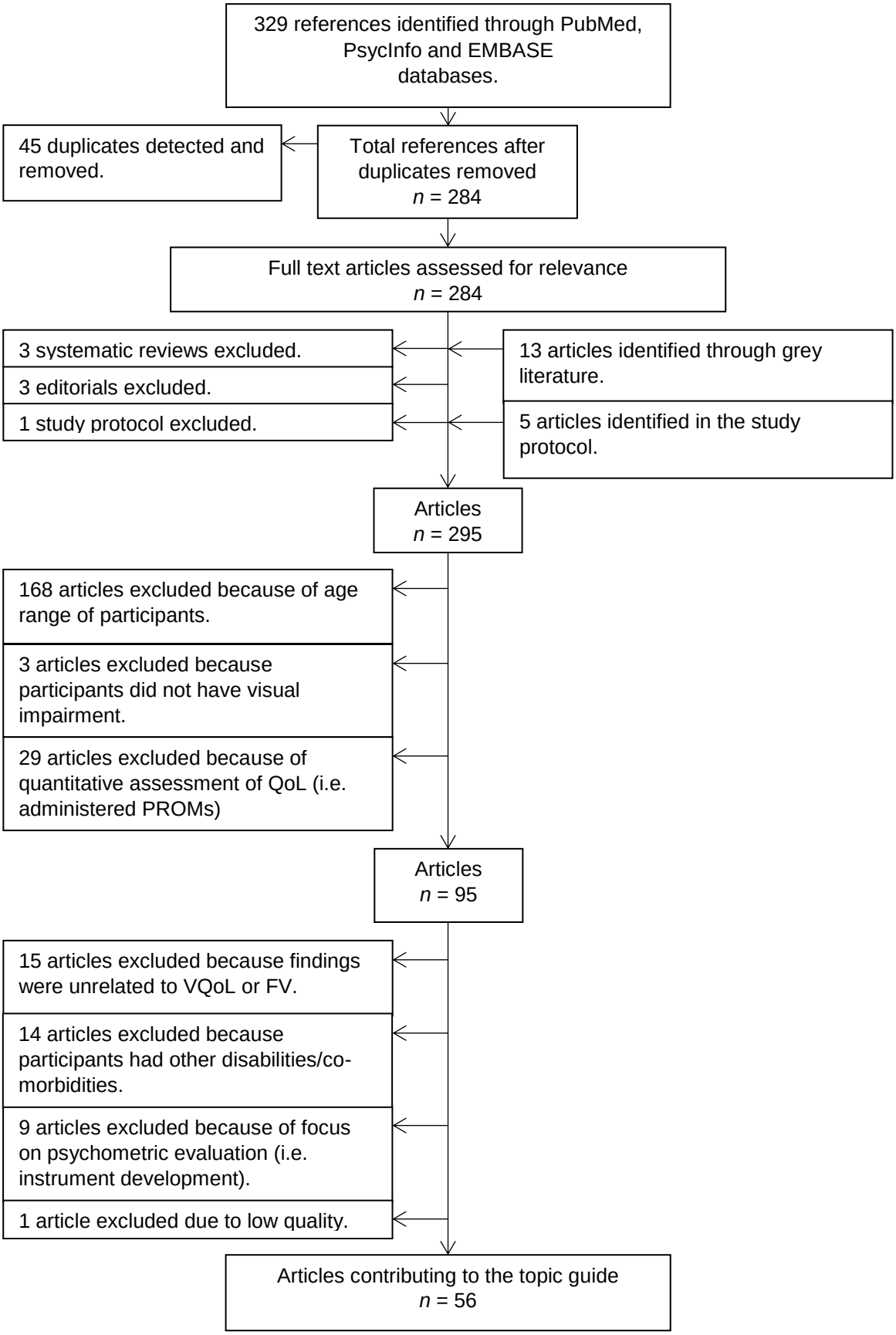
Articles were assessed in terms of the relevance of findings to VQoL and FV as outcomes, aspects of everyday life for YP living with visual impairment, and suitability for contributing to questions and probes in the topic guide. As a result of being unrelated to these factors, 15 articles were excluded. A further 14 articles were excluded because participants had co-morbidities which would have likely impacted upon their perspectives of QoL and experience of everyday life.

A number of articles documenting the development of instruments designed to measure QoL in varying populations were identified. In these cases, attention was paid to any articles which focused upon the content of the items included in the instruments. Those which focused purely upon the psychometric evaluation of these instruments were excluded.

Formal quality assessment of the remaining articles was not deemed appropriate. This was because the aim of the review was to identify, as broadly as possible, all the issues potentially relevant to the lives of adolescents with visual impairment in order to inform the content of the topic guide. Since the content of individual interviews was intended to be largely patient-driven, the purpose of the systematic review was to guide the interviews and ensure all relevant areas were covered, rather than to develop an evidence base to apply the findings e.g. inform policy and decision-making per se. Nevertheless, quality of the identified papers was assessed in part by judging the sample size and outcomes. One article was excluded due to methodological weaknesses indicating low quality.

Due to the inclusive nature of the systematic review, no further exclusion criteria were applied.

Figure 1. PRISMA flowchart showing systematic review.



4.6.2.1.4 Synthesis of findings and application to the topic guide

The articles were categorized according to VQoL or FV domain using the six domains included in the topic guide used to develop PROMs for children aged 10-15 years[5] as a foundation (see Table 4, pg. 91). Attention was paid to identifying any new domains, and modifying the content of existing domains to make the topic guide sensitive to the attitudes, preferences, and everyday lifestyles of YP with visual impairment aged older than 15 years. As a result, the following 6 domains were further developed: home life, education (school/college/university), leisure life, independence, social life, and future – aspirations and fears. The domain 'clinical care' was added. Based on the overarching aim of the research project, that is, exploring the experience of growing up with visual impairment, adopting a life course perspective which encourages retrospective reflection on experience during childhood, a battery of probes was developed to encourage participants to reflect upon the differences between their attitudes, beliefs and lifestyles at the time of the interview and those during early childhood.

Several articles contributed to the development of questions or probes within multiple domains. These articles provided descriptive accounts of the everyday challenges and experiences of YP living with visual impairment.

Literature relating to functional difficulties during adolescence was initially included as a separate domain, but later incorporated into the topic guide in the form of probes which were used within each domain. This ensured participants reflected upon their FV in a number of different contexts e.g. home, school, leisure etc. Similarly, literature relating to psychological and emotional difficulties during adolescence was incorporated into the 6 domains of the topic guide as probes (e.g. How does that make you feel?) rather than as a distinct domain. Literature contributing to the topic guide is shown in Table 4 (pg. 91).

As a result of the systematic literature review, several changes were made to the foundation topic guide that had been developed for YP aged 10-15 years. In the foundation topic guide, only one question about social life (Where do you like to

go/what do you like to do with your friends?) was included under the umbrella term of 'activities and socialising'. However, due to a substantial amount of literature demonstrating age-specific difficulties in social communication in YP with visual impairment, a number of new questions were developed and the domain 'social life' was separated from 'leisure life'. New probes touched upon aspects such as friendship groups/dynamics and feelings of exclusion or fitting in. Development of romantic relationships was also probed sensitively.

The majority of identified literature supported the core questions included in each domain of the existing topic guide, but led to the development of a number of new probes. Most pertinent additions were related to experiences of vision-specialist education, attitudes towards teaching assistants, and experience of transitions within healthcare. Probes about exercise were added with the aim of providing participants a platform in which to discuss psychological aspects of living with visual impairment, such as self-image and self-esteem.

Table 4. Domains of QoL, literature which supports the inclusion of each domain in the topic guide, and modifications which were made to the topic guide in response to each paper identified.

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
Home life[5, 6, 240, 268, 280, 326-340]	Relationships with parents, relationships with other family members (siblings and extended family), practical and emotional support from family members, over-protection from family members, functional ability when completing domestic tasks e.g. feeding a pet dog, taking care of personal hygiene, chores, mobility at home, privacy from parents, me-time, family members understanding, having a role-model in the family, activities at home (watching TV, playing games, using the computer, doing homework, listening to music, reading for fun).	**Who do you live with?** Do you have any siblings? What do you like to do with your siblings? What is your relationship like with your parents and siblings? **Can you tell me about the sorts of things you do when you're at home?** What did you do when you were at home yesterday/last weekend? Did you enjoy doing that? **Is there anything in particular you find difficult doing at home because of your eyesight?** Did you find that difficult? Do you have any pets? Do you need help getting ready in the morning? How about finding your way around the house?	Do you feel like your parents/siblings/other family members understand what it's like for you? Do any of your parents/siblings/other family members also have a visual impairment? Do you have to do any kind of chores when you're at home?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
		Home life continued… Do you feel like you get enough privacy/me-time when you're at home?	
Education (school/college/ university)[5, 6, 213, 240, 268, 335, 336, 339, 341-345]	Likes and dislikes, participation in different lessons, barriers to participation in different subjects, choosing which subjects to take, exams, social life at school, peer support, bullying and teasing at school, relationships with teachers, teaching assistants and specialist education (assistive devices/technology, classes, specialist schools), having to miss school because of hospital visits, teacher knowledge, understanding and support, getting around at school, inclusion/exclusion, keeping up with others and workload, getting around school/mobility, getting to and from school, asking for help, changing school/classes/teachers.	**What sort of things do you enjoy about school?** Can you describe to me what you did at school yesterday? What are your favourite lessons/teachers? What are the classrooms like? Who do you spend most time with at school? What do you like to do together? **What things do you not like about school?** **Do you feel your eyesight gets in the way of things you do at school?** Is there anything that you would really like to do at school, but can't because of your eyesight?	**Do you go to school or college or university at the moment?** What kind of school is it (mainstream/specialist)? Is there a visual impairment unit? Do you travel to school by yourself? Do you have to do many exams at school? Do you have a teaching assistant? Do you like having a teaching assistant? Do you use any special devices or technology at school because of your eyesight? Do you feel like your teachers understand what it's like to have a visual impairment? **Have you ever had to change schools/teachers/classes?** Were there any differences? How did things change?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
			Education (school/college/university) continued… **Do you think your life at school has changed as you've got older?**
Leisure life[5, 6, 212, 240, 268, 327, 330, 333, 335-337, 339, 340, 342, 345-349]	Likes and dislike, physical activity/exercise, meeting friends outside of school, doing sports, walking, running, getting around outdoors, travelling, shopping, visiting restaurants/coffee shops, getting around new places, commitment to leisure activities, adjusting to new environments and activities, extra-curricular activities, playing musical instruments.	**What do you like to do in your free time?** Where do you go? **What activities do you enjoy?** Do you ever go shopping? **Are there any activities that you would like to do but can't because of your eyesight?** **What would be the main way you feel your eyesight affects what you can and can't do in your free-time?** How about travelling places?	What do you enjoy about […]? Do you enjoy exercise? How often do you exercise and where do you do it? Do you use any vision devices/technology to help when you do things outside the house? How about musical instruments?
Independence[5, 6, 212, 240, 268, 327, 336-340, 342, 349]	Responsibilities at home, responsibilities at school, basic economic transactions (e.g. using cash machines and handling money), using public transport when alone,	**Apart from chores, do you have any particular responsibilities when you're at home?** Do you ever do that by yourself? **Do you ever go outside on your own?**	Do you ever go outside in the dark? Have you ever had any mobility training? Do you think you can manage your own time well?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
	Independence continued… getting around outdoors when daylight/night time, using VI-assistive devices such as canes, independence from parents, restraints on physical activity e.g. walking in public, perceived reliance on parents, mobility training, managing time independently, travelling, barriers and wishes, writing and sending applications, work experience.	*Independence continued…* Who comes with you when you go shopping/on the bus/on the train etc.? Do you like them to come with you or would you prefer to be able to go on your own? How do you feel about…coming with you? **Do you ever travel by yourself?** Are there any things you feel you can't do because…?	*Independence continued…* **Do you think your independence has changed as you've got older?**
Social life[6, 240, 268, 280, 327-329, 331, 333-337, 339, 341, 345, 347, 350-363]	Best friends, friends, peers, enemies, initiating and maintaining friendships, self-esteem, developing romantic relationships, dating, barriers to developing romantic relationships, falling in love, eye contact, visual cues, missing body gestures, facial expressions, social support from friends and peers, making friends despite difficulties e.g. parental over-protection,	**Where do you like to go/what do you like to do with your friends?**	**Do you have a best friend or group of best friends?** Do they go to the same college as you? How did you meet? **Are all your friends girls/boys?** **Do any of your friends have a visual impairment?** What are the like? Can you tell me a bit more about him/her? How/when did you meet?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
	Social life continued… arguments with friends, meeting friends outside of school, prejudice from others (friends and members of the public), public perceptions, overprotection from peers, shyness in social situations, other people understanding, other people helping, recognising friends in public, inclusion in social activities, social networks, not telling other people about visual impairment and functional difficulties, reaction of others.		*Social life continued…* What do you like to do together? Would you like to have a boy/girlfriend? **Is there anything you find difficult to do with your friends because of your eyesight?** Is there anything you can't do because of your eyesight? Do you spend more time with your friends or family? Which do you prefer? **Do you feel like your friends understand what it's like for you?** **Do you feel like your friendships have changed at all as you've got older?** What about when you changed school?
Future – aspirations and fears[5, 6, 240, 336, 339, 340, 345, 362]	Aspirations, preferences, desires, worries and concerns, fears, future education (e.g. going to University, exams, choosing courses), future physical health, future visual function, future employment, making plans for the future, barriers in the future,	**What do you think you might do after you've done your GCSEs/finished school/college/University?** **Have you thought about what kind of job you'd like to do in the future?**	**Have you thought about moving away from home in the future?** How do you feel about that? What do you think it will be like to live by yourself/without your parents in the future?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
	Future – aspirations and fears continued… future independence and autonomy, future family life, finding a romantic partner and building a family, future living environment and location, moving away from home/parents, fear of falling short of expectations of others.	*Future – aspirations and fears continued…* Do you have any worries/concerns about the future? What are you most worried about? **Have you thought of any things you would like to do in the future, but might not be able to do because of your eyesight?** Do you have any concerns about your vision in the future? Have you ever thought about having a family in the future?	
Clinical care[5]	Going to eye clinics, hospital visits, likes and dislikes, preferences.	**Do you still regularly attend clinics?** Do you like/not like going? Why?	Where/which hospital do you go to? How often do you have to go? **Have you changed to adult clinics now or do you still go to the children's ones?** Have you ever had to change consultants? How did you find that process? Would you have done it differently?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
Functional vision[6, 240, 277, 337, 340, 351, 364, 365]	Balance, ocular discomfort (e.g. eye redness, photophobia), pain, environmental factors (e.g. sunshine) and impact upon FV, sleeping, daytime napping.	Do you have any difficulties doing that? Is that easy for you? How does your eyesight affect that? How do you find that?	-
Psychological and emotional well-being[240, 268, 280, 327-329, 333, 335, 337, 339, 345, 351, 353, 358, 360, 362, 363, 366-372]	Self-concept, self-image, self-esteem, anxiety, social anxiety, school anxiety, hostility, worry and concern, identity development, emotions (nervousness, loneliness, desires, frustrations, anger, depression, happiness, embarrassment), hyperactivity/inattention, acceptance of visual impairment, psychological adjustment to visual impairment, resilience, fulfilling ambitions/doing what you want to do, confidence, self-consciousness, (dis)satisfaction with self-appearance and body image, learning that visual impairment will never go away,	How do you feel about that? How does that make you feel? What do you prefer? Do you enjoy that? Does that bother you? Why do you feel like that? Is there anything else that's really important to you that I haven't asked you about?	How important do you think that is? Why do you think that's important? Have you always felt that way?

Domain and supporting references	Potential contributors to QoL during adolescence	Existing questions (bold) and probes taken from the foundation topic guide	New age-appropriate questions (bold) and probes
	Psychological and emotional well-being continued… learning that visual impairment is an evolving process, desires to be different/normal/the same as others, coping strategies, motivation to learn new strategies, feeling different, feelings of freedom and independence.		

4.6.3 Data collection: In-depth semi-structured interviews

In-depth semi-structured interviews were conducted between March and June 2015 by one interviewer (AR). The developed topic guide was used flexibly to explore many areas of everyday life. After re-confirmation of consent, an 'ice-breaker' activity preceded each interview. During interviews, participants were encouraged to speak to the interviewer independently of their parents or family members. Due to the nature of interviews, which took place at participants' family homes, this was not always possible, and parents and/or siblings were present during a number of interviews. Generic questions about specific aspects of everyday life (i.e. home, school, free time) were discussed before the impact of visual impairment was probed specifically. Opportunity was given for the YP to lead in opening the topic guide and raise any relevant issues independently. This approach encouraged participants to relax, and feel as though they were engaged in an informal 'chat' as opposed to an interview. To address the overarching aim of the research, that is, to contribute to the current understanding of the experience of growing up with visual impairment, participants were asked within each domain of the topic guide to reflect upon their experiences during childhood and recall any pertinent memories of times when they experienced specific difficulties, challenges, or changes. Open-ended probes were used consistently throughout interviews to encourage participants to take the lead during discussions and talk about things which they felt were most important.

4.6.4 Qualitative data analysis

Each interview was digitally recorded, transcribed and imported into NVivo 10. Qualitative data analysis was then conducted in two separate stages with different purposes. The first stage was conducted with the aim of exploring the voices of visually impaired adolescents in depth with reference to the lived experience of visual impairment throughout childhood and adolescence; the second stage was conducted with the purpose of applying the qualitative data to generate meaningful and age-appropriate instrument items.

4.6.4.1 First stage data analysis (exploring the impact of visual impairment during childhood and adolescence)

Qualitative analysis based on the tenets of Grounded Theory[373] was conducted to identify key themes related to participants' experience of growing up with visual impairment. This approach was selected based on source data stemming from spontaneous speech in addition to answers which were further probed. Thus, both inductive and deductive methods were incorporated.

Data analysis comprised an ongoing, circular process of open-coding.[374, 375] This was completed independently by two members of the research team (AR and VT) with a view to ensure reliability of the coding process. The first stage of coding comprised both members of the research team coding a third of the transcribed data with a view to identify codes corresponding to experiences and any pertinent transitions and changes during childhood and adolescence. Coding was completed manually by one researcher and by the other using NVivo software. After the first stage of coding, the two researchers met to compare notes and develop an inventory of codes. An initial codebook was used by both researchers to code a further third of the data. Any new codes which had not already been identified were added to the codebook after discussion about their meaning and value. Definitions of codes were refined, and changes to the codebook were tracked.

A final codebook was developed by adding description, examples, and criteria of use and then used by both researchers to code the entire dataset. Ongoing meetings and communication between researchers ensured rigorous and consistent use of the codebook. Using NVivo, the data were organised into individual codes using a reference sheet detailing clinical and demographic details of each participant to allow for comparisons to be made between participants (e.g. in terms of gender, severity of visual impairment, and timing of onset of visual impairment). Finally, the themes emerging from the data by combining the codes were identified and labelled.

4.6.4.2 Second stage data analysis (item generation)

Transcriptions were exported into NVivo 10 and organised into seven nodes reflecting the seven topics included in the topic guide. Each node was searched for data relating to the six higher order themes outlined previously by Rahi et al.[5] with 10-15 year olds, namely: 1) social relationships, acceptance and participation; 2) independence and autonomy; 3) psychological and emotional well-being; 4) future – aspirations and fears; 5) functioning – school, home and leisure; and 6) treatment of eye condition. Using these domains as a starting point for item generation was deemed appropriate due to applicability of the domains to existing instrument versions of VQoL and FV for 10-15 year olds, and with the new age-appropriate instrument versions for older adolescents being an adaptation and extension of these existing instruments. Once the data were organised, each theme was re-read and open-coded.[374, 376]

Each item incorporated into the existing instruments (VQoL and FV) during the foundation research is expressed as a statement with a Likert-type response scale comprising four responses. In the existing VQoL instrument, items are phrased as statements and responses refer to users' endorsement of the statement (i.e. whether, and to what extent, it is true about them). In the FV instrument, items are phrased as partial statements, and users are required to indicate their functional visual ability by choosing the suffix which best refers to the perceived difficulty of completing a task. The design and format of VQoL items is empirically grounded based on consideration of using an 'illustrative' child's perspective during the foundation research.[5] Prior to further analysis, all items developed in the foundation research were modified to remove the 'illustrative' child's perspective, which was shown to lack feasibility when used with children and YP, producing significant ceiling effects[5] (see Table 5, pg. 102 for an example of the modified presentation of VQoL and FV items).

Table 5. Example VQoL and FV item developed in the foundation research[5] and modified version in the current development.

Example VQoL item and response options used in the foundation research	**Example FV item and response options used in the foundation research**
Ben has got some good friends. How much are you like Ben? **'Actual' response options:** 1. Not at all 2. A little bit 3. Quite a lot 4. Exactly How much do you want to be like Ben? **'Ideal' response options:** 1. Not at all 2. A little bit 3. Quite a lot 4. Exactly	How easy do you find…watching TV **Response options:** 1. Very easy 2. Easy 3. Difficult 4. Very difficult or impossible 5. This doesn't apply to me/I don't do this for other reasons
Example VQoL item and response options modified for the current development	**Example FV item and response options modified for the current development**
I have got some good friends. **Response options:** 1. Not at all true 2. A little bit true 3. Mostly true 4. Completely true	Because of my eyesight, I find watching TV **Response options:** 1. Very easy 2. Easy 3. Difficult 4. Very difficult or impossible

Open-codes were mapped onto items included in the existing VQoL and FV measures developed for 10-15 year olds in the foundational research[5-7] which were then entered into an Excel spreadsheet. To ensure comprehensive coverage of the VQoL and FV

issues discussed by YP, codes were also mapped onto items which were developed during the early phases of the foundation research but subsequently excluded. Issues relevant to VQoL (e.g. psychosocial issues) were collated for the VQoL instrument and the items relevant to function, activities and difficulties completing them were compiled for the FV instrument. Issues which did not map onto items included in the development of existing VQoL and FV instruments were considered as new items reflecting age-specific concerns. VQoL and FV instrument versions applicable to adolescents older than 15 years were drafted.

Following this, an expert consensus meeting, including 4 members of the study research group, took place with the aims of: a) gathering feedback from the study research group as to the relevance/suitability of the items included in the new VQoL and FV age-appropriate versions, b) discussing possible age-appropriate modifications of these existing items and c) discussing the need for, and development of new age-appropriate items for each instrument. Prior to this meeting, each member of the study research group was sent a copy of each instrument and asked to reflect on suitability of items independently. Each draft item was then discussed in terms of relevance to the patient population, wording and presentation order.

During the expert consensus meeting, the age boundaries for the older (and younger) instrument versions were discussed and a decision was made to modify these according to the data collected i.e. a data-driven process. This decision was largely based on the emergence of independent living at the age of 18 years. Some participants aged 18 and 19 years old at the time of participation in interviews had begun attending University, and thus were living away from home. The perspectives and daily lives of these individuals were not comparable with those of younger participants. As a result, the data collected during the foundation research were re-visited with a view of determining the correct age boundaries for the new older-age instrument versions. The age-range of 13-17 years was deemed the most appropriate based on the emergence of independent activities away from home, less parental

restriction, and increased awareness about the future at the age of 13 years. In parallel, the age boundary for the younger instrument versions was modified from 6-9 years to 8-12 years.

Following the expert consensus meeting, the drafts of the age-appropriate versions of both the VQoL and FV instruments suitable for older adolescents were finalised. A master item pool was developed which included all items in the first draft instruments and was used to track which stage the items had been developed (e.g. in the foundation research or as a result of the interviews with older adolescents).

4.7 Phase 2: Pre-testing

The second phase of instrument development was conducted to operationalise the draft age-appropriate instrument versions developed in phase 1. Specifically, individual cognitive interviews with YP with visual impairment were conducted to identify a) the importance and relevance of items, b) YP's understanding and interpretation of the instructions, response options and items, c) any ambiguity they experienced in the completion of the instruments and d) YP's preferences regarding the wording of the items and any suggestions for re-phrasing.

4.7.1 Participants and recruitment method

As described above, the expert consensus meeting led to a decision to operationalise the age-appropriate instrument versions of VQoL and FV instruments for adolescents aged 13-17 years.

Patients were identified, selected and recruited for participation in phase 2 using the same random, stratified sampling technique as in phase 1 (and outlined in Section 4.6.1, pg. 83).

A total of 68 eligible patients aged 13-17 years were invited to take part in phase 2 of the instrument development. They were sent invitation packs containing the relevant

recruitment materials as specified in Section 4.6.1 (pg. 83) and shown in Appendix II-IX (pg. 348-371).

The recruitment materials reflected the broader age range of participants invited in this phase, compared to those who were invited in phase 1. Parents/guardians of all older patients (i.e. those aged 16-17 years) were sent invitation packs which were addressed to the parent/guardian of each young person, but included a separate invitation letter addressing the young person. Parents and guardians of younger patients (i.e. aged 13-15 years) were sent invitation packs containing only one invitation letter (addressed to the parent/guardian) and an information sheet, assent, and consent forms suitable for children and parents respectively. The follow-up procedure described in Section 4.6.1 (pg. 83) was used.

4.7.2 Data collection: Cognitive interviews

Individual cognitive interviews were conducted between September 2015 and January 2016 by one interviewer (AR). Similar to the data collection procedure outlined in Section 4.6.3 (pg. 99), YP were encouraged to feedback on the individual instruments and items independently, but due to the nature of interviews which took place at participants' family homes this was not always possible.

The new age-appropriate versions of the draft VQoL and FV instruments were used, alongside a standard list of questions used to gauge participants' perceived importance, relevance, and comprehensibility of the items (see Appendix XI, pg. 376). Prior to interviews, consent was re-confirmed and the researcher made clear the intention of the interview e.g. to gather feedback on individual questionnaire items as opposed to gathering feedback about participants' VQoL or FV as outcome values. The presentation of VQoL and FV instruments was rotated with each interview starting with a different instrument. Interviews began with the researcher reading through the instruction page of an instrument. Participants were asked about their understanding of the instructions, and any recommendations they may have on how to make the

instructions easier to understand. The researcher then read aloud the four response options, including their numeric value and label. Each individual item was presented to each participant, read aloud by the researcher, and participants were asked to indicate which response they deemed appropriate for themselves, why they had chosen a particular value, and suitability of the other response options. Any ambiguity that participants noted in relation to the instrument items, instructions, or response options was probed and responses were noted. The researcher recorded, using pen and paper, participants' responses to each item, and any additional feedback or comments. The procedure was repeated for each instrument.

4.7.3 Data analysis

Data entry was ongoing throughout the course of phase 2. After each interview, the researcher entered participants' responses, including any qualitative verbal feedback, into an Excel spreadsheet which was used to collect and compare participants' feedback. Any difficulties participants had when giving a firm answer to any of the items and reasons why this was difficult were recorded. Participants' recommendations for ways to clarify the instructions or response options, and preferred wording of items, were noted in Excel.

A results document was created using Microsoft Word, showing each item and a summary of participants' responses to the item. All items were evaluated by one researcher (AR) and any action (e.g. change or amendment of an item) which was thought to be necessary was recorded in the same document.

A second expert consensus meeting including three members of the research team was carried out to evaluate the action needed for each item. Consistent with the procedure used in Phase 1, all attendees were asked to reflect on the draft instruments independently before the meeting took place. Times of disagreement between researchers were discussed during the meeting before a mutual decision was reached, taking into account the opinions of all members of the team.

The items included in the second draft of each instrument were entered into the master item pool as the second draft of both the VQoL and FV instrument versions for 13-17 year olds.

4.8 Phase 3: Piloting

The aim of phase 3 was to pilot the draft instruments with a small sample of participants in order to identify any immediate difficulties with the instruments or any of the items, such as ambiguity, false interpretation, patterns of missing data and distribution of responses (e.g. ceiling and floor effects). Thus, a small sample of approximately 15-20 participants was deemed adequate, with respect to conserving the majority of the remaining sampling framework for use during the formal psychometric evaluation phase (phase 4) where the largest sample size was required.

4.8.1 Participants and recruitment method

Participants were identified, selected and recruited for participation in phase 3 of the instrument development using the same random, stratified sampling technique as in previous phases (see Section 4.6.1, pg. 83). At this stage, recruitment of participants from source 1 was supplemented by recruitment from source 2 (see Section 4.3.2, pg. 80). PICs were contacted to confirm an estimated number of patients they were able to identify who fulfilled the eligibility criteria (see Section 4.3.1, pg. 78). Invitation packs were then sent to PICs, along with a recruitment log in which to record the date of invitation of individual patients. On-going communication between the research team and PICs ensured PICs were also sent reminder invitation packs, which were distributed to invited patients 2 weeks following initial invitation, as well as ‘Thank You’ letters which were distributed after a patient had participated.

Sixty-seven eligible patients from source 1 were invited to take part in phase 3 of the instrument development. An additional 55 eligible patients from source 2 were invited to take part. Identified patients were sent invitation packs by post inviting them to take

part in phase 3 of the instrument development. The invitation packs for children aged 13-15 years differed from those for YP aged 16-17 years. Each invitation pack was addressed to the parent of the young person and contained:

a) a parent invitation letter
b) a parent study information sheet
c) a parent consent form
d) a family background questionnaire
e) a young person study information sheet
f) a young person assent form

Invitation packs sent directly to YP aged 16-17 years included an invitation letter addressing the young person, consent form and information sheet designed specifically for older participants (see Appendix VI-VIII, pg. 363-366, 369). The parents/guardians of participants aged 16-17 years received an invitation letter, information sheet, consent form and family background questionnaire as described in Section 4.6.1 (pg. 83) and shown in Appendix III-V, IX (pg. 354-358, 361, 371).

The second draft of the new age-appropriate instrument versions (VQoL and FV) were printed in the format of an A4 questionnaire booklet in large print (N18). These were printed separately for the young person and the parent and labelled accordingly. Items in parent booklets were identical to those in the young person booklet, but were re-phrased to encourage parents to reflect upon their child's vision-specific outcomes by adding the prefix '*My child'* before each item in the VQoL instrument and '*Because of his/her eyesight my child finds'* before each item in the FV instrument (see Table 6, pg. 109). Both parent and young person booklets included a page for qualitative feedback on the instruments. Parent booklets were administered with a view that parents' perspectives are often independent from those of their child (see Section 2.3.2, pg. 34), and to encourage independent self-report from YP. Young person booklets also included a number of simple questions at the end to collect data about ease of completion, understanding of the items, and time taken to complete each questionnaire

(see Appendix XII, pg. 378). Each invitation pack included both a parent and young person questionnaire booklet and a large pre-paid envelope for return of the completed questionnaire booklet, parent consent form, family background questionnaire, and young person assent form. Families of patients who were aged 16-17 were sent two pre-paid envelopes, to encourage YP to participate independently of their parent/guardian.

All invited families were followed-up by telephone 2 weeks later with the aim of answering any questions they had. The families of all patients for whom the researcher had been unable to contact via telephone were sent a single postal reminder 4 weeks following the initial invitation.

Table 6. Example of item adaptation for Parent booklets.

YP-VQoL item	**Parent-VQoL item**
I have got some good friends.	My child has got some good friends.
YP-FV item	**Parent-FV item**
Because of my eyesight, I find watching TV…	Because of his/her eyesight my child finds watching TV…

4.8.2 Data collection: Postal survey

Data collection and entry were ongoing throughout the course of phase 3. Upon receiving a completed questionnaire booklet, and consent forms, the researcher documented important clinical and demographic details into both an Excel spreadsheet and SPSS data set. All quantitative data were entered into each spreadsheet at two different time points to enable identification of errors in data entry (no errors were identified). Any qualitative annotations made to the questionnaire booklet were noted in the Excel spreadsheet. Overall qualitative feedback and answers to the feasibility questions were recorded in a separate document in Excel and SPSS.

4.8.3 Data analysis

Exploratory data analysis used both graphical and non-graphical methods. Feasibility items included at the end of each questionnaire booklet were analysed using simple descriptive statistics (Median, interquartile range (IQR), and percentages) calculated in SPSS. The following descriptive statistics were calculated for each item:

a) percent of missing data (per person and per item)
b) z-score for skewness
c) z-score for kurtosis

Bar graphs were created in SPSS to view the frequency of participants' responses to each item. Qualitative feedback given in response to individual items was assessed as complementary to quantitative results. Each item was screened individually. Items were flagged as problematic if they had > 20% missing data, z-score for skewness and z-score for kurtosis values outside the acceptable limits (-2.00 to +2.00)[313] or a response category(ies) which were not endorsed by any participants, or alternatively by all participants (i.e. floor or ceiling effects). Participants with > 25% missing responses were excluded based on criteria used in the foundation research.[6]

A third expert consensus meeting took place following the procedure outlined in Section 4.7.3 (pg. 106) in which three members of the research team met to discuss the findings. Decisions to amend, or remove items in light of the feedback obtained were made with reference to the master item pool, at which stage the items were developed (e.g. during the foundation or current development), and feedback given in phase 2 of the instrument development. As a result of this phase, items were modified and entered into the master item pool for use in phase 4.

4.9 Phase 4: Formal Validation

The final phase of instrument development aimed to assess the psychometric properties of the new age-appropriate versions of the VQoL and FV instruments. This

was done through a postal administration of the final draft instruments to a larger, nationally representative sample of YP living with visual impairment.

An intended important feature of the developed instrument suite was that the child- (8-12 year olds) and YP- (13-17 year olds) instruments could be used sequentially within ophthalmic practice to detect longitudinal changes in self-reported outcomes. Thus, at the formal validation stage, both the younger (child) and older (YP) versions were simultaneously piloted with both datasets being managed (separately) by AR. To this end, the younger instrument development was conducted by a different member of the research team (VT).

4.9.1 Participants and recruitment method

Children (aged 8-12 years) and YP (aged 13-17 years) were identified, selected and recruited for participation in phase 4 of the instrument development using both primary and secondary recruitment sources (see Section 4.3.2, pg. 80). Due to the minimum sample size needed for formal psychometric evaluation, including Rasch analysis (see Section 3.3.2, pg. 75), all remaining patients in the sampling framework from source 1 (*n* =194) were considered for participation, as well as any patients identified from source 2.

All eligible patients were invited to take part in phase 4 only and as per the same recruitment procedure as in earlier phases.

The questionnaire booklets administered in this phase included the final age-appropriate versions of the two instruments alongside the Pediatric Quality of Life Inventory – Child and Teen versions (PedsQL)[377] (see Appendix XIII, pg. 379 and XIV, pg. 381) which was used to assess construct validity of the VQoL instrument (i.e. whether the VQoL instrument measures what it is intended to measure). A brief, 4-item survey regarding the feasibility of completing the instruments within clinical contexts, as well as participants' preferred method of administration was included at the end of each booklet (results are analysed in a separate research project) (see Appendix XV, pg.

383). Two formats of questionnaire booklet were developed whereby the order of presentation of VQoL and FV was counterbalanced. PedsQL and feedback questions were included at the end. As in phase 3, booklets were also developed for parents to complete independently, and to encourage children/YP to independently self-report. Parent booklets included VQoL and FV questionnaire items which were modified in the same way as in phase 3 (see Section 4.8.1, pg. 107) and presented in the same counterbalanced order as for YP, and Child and Teen – versions of the PedsQL Parent-Proxy report.[377]

Electronic versions of Child, YP and parent booklets were developed by a member of the wider research group (MC) as part of a separate research project exploring the feasibility and design of electronic health records in Ophthalmology. The web-link and instructions for completing the online instruments were provided through the invitation letters sent to participants and their parents/guardians.

Each invitation pack included both a parent and child/YP questionnaire booklet and a large pre-paid envelope for returning the completed questionnaire booklet, parent consent form, family background questionnaire, and child/YP assent form. As in phase 3, families of patients who were aged 16-17 were sent two pre-paid envelopes.

All invited families were followed-up by telephone 2 weeks later with the aim of addressing any queries or concerns they had. The families of all patients for whom the researcher had been unable to contact via telephone were sent a single postal reminder 4 weeks following the initial invitation.

4.9.2 Data collection: Postal survey

Data collection and entry were ongoing throughout the course of phase 4. Excel was used to document all data coming from the questionnaire booklets: that is, quantitative responses to individual items and any qualitative annotations which were made on the questionnaire booklets, overall qualitative feedback to the instruments, answers to the ease and feasibility questions, and participants' clinical and demographic details. All

quantitative responses to the instrument items were input into SPSS. If a participant indicated more than one answer to any of the VQoL or FV items, both responses were documented in Excel. In SPSS, items were allocated a missing data value shown in Table 7 (VQoL) and 8 (FV) (below) which were developed to reduce missing data and over-estimated the child's VQoL/FV. Since the two mid-scale response options represent contrast outcomes (i.e. easy/true and difficult/not true) opting for one response option over the other by over-estimating the child's VQoL/FV is conceptually problematic, and may lead to data misrepresenting the sample. Thus, responses were recorded as missing. Responses assigned a value of 9 were treated as missing data from this point onwards.

Table 7. Missing data values assigned to VQoL responses in phase 4 data analysis.

Response	Value assigned in SPSS for positively-phrased items	Value assigned in SPSS for negatively-phrased items
No response	9	9
1 (Not at all true) and 2 (A little bit true)	2	1
2 (A little bit true) and 3 (Mostly true)	9	9
3 (Mostly true) and 4 (Completely true)	4	3

Table 8. Missing data values assigned to FV responses in phase 4 data analysis.

Response	Value assigned in SPSS
No response	9
1 (Very easy) and 2 (Easy)	1
2 (Easy) and 3 (Difficult)	9
3 (Difficult) and 4 (Very difficult or impossible)	3

All missing data in the PedsQL instrument (child and teen versions), were assigned a 'missing data' value of 9.

4.9.3 Data verification

Data entered into the Excel and SPSS databases were cross-referenced using the subtract command in Excel to examine any errors when entering the data. Ten percent of the data were also entered independently into the Excel spreadsheets by a second researcher, and cross-referenced to identify errors. Any discrepancies were noted, and the original questionnaire booklets were checked to ensure the correct response was recorded.

4.9.4 Data analysis

Participation rates of children (aged 8-12 years) and YP (aged 13-17 years) were calculated.

4.9.4.1 Preliminary item reduction

To optimise sample size, percentage of missing data per person and item were calculated per instrument. Consistent with phase 3, participants were excluded from psychometric evaluation of the VQoL and/or FV development if >25% of item responses in a single instrument were missing. Items were excluded if >50% of participant responses were missing, in keeping with published criteria to guide item removal[313] and understanding that items with large amounts of missing data are likely to be ambiguous, or not applicable to respondents.

Preliminary item reduction was guided by the distribution of the individual item responses. Items with z-score skewness and z-score kurtosis values outside the range of -2.00 to +2.00 were flagged as problematic, but only removed if ceiling/floor effects were also found (e.g. >60% or <1% responses in an item end category). These thresholds were developed based on the understanding that Rasch analysis requires at least one case within each response option for each item, and more than 60% responses in an extreme category would indicate substantial skew in an item.[313]

4.9.4.2 Testing for unidimensionality

For the sake of testing unidimensionality, responses given to negatively worded (e.g. I feel lonely because of my eyesight) versus positively worded (e.g. I feel confident) VQoL items in both child and YP instruments were re-coded so that all responses aligned in the correct direction of the latent trait.

To preserve the sample size whilst testing for unidimensionality a missing data analysis was conducted on the raw datasets after removing items due to distribution of responses. Missing data were imputed using a multiple-pattern regression method,[378, 379] producing 5 imputed datasets for each instrument. Tests of unidimensionality following this imputation were conducted using each imputed dataset, and the results were averaged to derive a final outcome incorporating all imputed missing data values.

The first stage of unidimensionality assessment was conducted using formal exploratory FA with a view to provide evidence of the suitability of the data for Rasch analysis which assumes unidimensionality (see Section 3.3.2, pg. 75). This analysis was conducted using SPSS. The method of principal components was selected with standardised variables and no rotation. The eigenvalue for each factor was analysed in relation to the percent of variance explained. The component matrix was analysed as best evidence indicating unidimensionality in each instrument. Item loadings were scrutinised for values less than 0.4 and items which loaded onto more than one factor. Because the aim of FA was to assess for unidimensionality, no items were removed at this stage.

Parallel analysis is a technique which involves extracting eigenvalues from random data sets that parallel the actual data set with regard to number of cases and variables, and can aid decision making regarding the number of factors to retain in PCA and FA.[380] Parallel analysis was conducted in addition to exploratory FA with a view to provide further evidence for unidimensionality in the instruments. This analysis was conducted using SPSS, and using the syntax provided by O'Connor.[380] The number of

parallel datasets was specified as 1000, and the ‘permutations of the raw data set’ function was specified to account for imputations of missing data. Both the matrix and scree plot were used to determine the number of existing constructs within each instrument.

4.9.4.3 Formal psychometric item reduction and validation (Rasch analysis)

Prior to conducting Rasch analysis, the original datasets (containing missing data, and items which had not yet been reverse scored) were prepared by re-scoring observed responses in both the VQoL and FV instruments ranging from 1-4 to values ranging from 0-3, to reflect the true magnitude of each score in terms of additive measurement. This was performed using the subtract command in Excel. The original raw datasets containing missing values were used for Rasch analysis based on the premise that Rasch analysis conducted using Winsteps software is capable of making estimations and imputing missing data based on observed scores.

The Rasch rating scale model was applied to assess the true measurement capacity of the instruments, providing evidence for item removal. The process of item removal was iterative, meaning the items were removed one at a time until the criteria of Rasch analysis were met, and all measurement difficulties were resolved. Rasch analysis was conducted using Winsteps 4.0.1.[381] The measurement properties of each instrument (after each iteration of Rasch analysis) were documented using Winsteps outputs in NotePad and compared to produce the optimum measurement instrument. The following syntax was used to ensure missing data values in the FV instruments were recognised:

CODES = 01239; matches the data

NEWSCORE = 0123*; rescore 9 as missing

In the VQoL datasets, items were coded based on the direction of the measurement scale (e.g. forward or reverse scored) using the following syntax (where N represents a forward scored item, and R a reverse scored item):

IVALUEN = 0123*; scoring for forward scored items

IVALUER = 3210*; scoring for reverse scored items

The following syntax was used to ensure the discrete missing data values in the VQoL datasets were detected by Winsteps, and treated as missing:

MISSING-SCORED = -1; missing data treated as non-administered

Item fit can be used to confirm that the summary scores provided by the instruments represent a single underlying construct, indicating unidimensionality and supplementing the findings from FA and parallel analysis. INFIT and OUTFIT statistics indicate how well the items contribute to the measurement outcome (e.g. VQoL or FV) and are measured as mean squared standardized residuals, with the 0.5 to 1.5 range being considered acceptable for productive measurement.[382] OUTFIT is based on a sum of squared standardized residuals modelled to approximate a unit normal distribution and INFIT is an information-weighted form of OUTFIT which reduces the influence of less informative, off-target responses.[383] Low values of the fit statistics indicate observations are too predictable (e.g. the data overfit the measurement model) and high values of these statistics indicate unpredictability (e.g. the data underfit the model). Thus, high values of the fit statistics are conventionally inferred as more harmful to measurement than low values which indicate some redundancy in the responses, but do no harm to the instruments capacity for measurement. Items with values of the fit statistics outside the range of 0.5-1.5 were removed iteratively until all items resulted in fit statistics within the acceptable range.

A fundamental component of Rasch analysis, making it distinct from classical test theory approaches such as FA, is that both persons and items differ in terms of ability on the latent trait. Thus, using Rasch analysis, one can assess the *targeting* of the

items in a particular instrument. In this sense, targeting refers to the ability to examine the relative position of item difficulty to person difficulty. The items in an instrument are deemed to target the sample well if the difference between item and person means is less than 1 logit.[384] The item-person map was used as an indicator of the difference between item and person means for each instrument and the difference is also reported in terms of logits.

The direction of alignment of items on the latent trait was assessed using the item polarity command in Winsteps to ensure that higher person abilities corresponded with higher ratings on each item. Observed point-correlations were screened for negative values and average abilities were screened for abilities which were out of order in terms of person ability.

Measurement precision was assessed in each iteration of Rasch analysis for each instrument version using the summary statistics outputs in Winsteps. The person separation index and reliability values were recorded from each analysis using a separate Excel document which enabled the comparison of values across iterations. The threshold of separation index values ≥2.00 and separation reliability values >0.80 were used as the criteria for indicating accurate measurement precision.[313]

Response scale ordering was examined using the Rasch category probability curves which indicate the likelihood of each response scale being selected over the range of the scale. The increase in logits between response options is considered to be good if it increases by at least 1.4 logits,[317] indicating distinct categories, however an increase of more than 5 logits was interpreted as indicating gaps between the response categories. The rating (partial credit) scale function displays the fit mean squared of each response category. Values greater than 1.5 indicate problems with the response options in the same way as the item fit statistics, using the average of the fit mean-square (MNSQ) values associated with the responses in each category. Participants' use of response categories was also assessed using the position of response options

in relation to the ability of the sample. In each case, the ordering of response options was analysed for linear patterns.

Analysis of differential item functioning (DIF) indicates whether subgroups of respondents who have the same person ability respond differently to items. DIF analyses were used to compare participants stratified by age and gender within each analysis. These demographic variables were chosen based on the assumption that the items in the final instrument versions should not produce different results for participants of different gender or age. In other words, items should not be biased in any way to gender or age group. DIF analyses using participants' categorised level of vision (or ability) were not deemed necessary given the developed instruments are intended to detect these differences. All DIF analyses were conducted after each instrument had seemingly fulfilled the multiple criteria of Rasch analysis, and all problematic items had been removed. The threshold of >1.0 was used as an indication of notable DIF between participants.[385] The probability of both Rasch-Welch and Mantel-Hanzel *t*-statistics for each item were analysed in parallel to DIF contrast values, indicating the probability of observing the DIF contrast when no real DIF is present. Theoretically, if the data fit the Rasch model of measurement, the Mantel-Hanzel *t*-statistic should be related to the Rasch-Welch statistic.[386] However, DIF contrasts were used as the primary indicator of DIF. As a result of the relatively small sample size used in phase 4, when conducting DIF analyses by age, participants were stratified to 'Young' and 'Old' age groups within each instrument development (i.e. child and young person) to ensure a large number of participants within each age group, and therefore more accurate indications of DIF.

Items which had DIF values above the threshold of 1.0 were removed iteratively and in order of magnitude (from greatest to smallest). After the removal of an individual item due to DIF, all remaining items were entered into a new iteration which was re-analysed using the full Rasch criteria outlined above.

4.9.4.4 Calibration of the child- and YP-VQoL and FV instruments

An important feature of the developed instruments is that they can be used in parallel to measure VQoL and FV across the broader population of children and YP aged 8-17 years. Thus, when used in collaboration summary scores from individuals of different age groups (and given in response to different instrument age-versions) should be comparable in terms of outcome on the latent variable. To ensure instruments are calibrated in this way, and capable of measuring the same outcome among children and YP of varying ages, a final DIF analysis was conducted using the final (and psychometrically robust) version of each instrument. Overlapping or 'core' items included in both child and YP instruments were assessed for significant DIF (between children and YP) and removed if DIF was above the threshold of 1 logit.

Following calibration using DIF analyses, items included in each instrument (e.g. both 'core' items and corresponding age-appropriate items) were selected individually using the ISELECT command in Winsteps, with a view to convert raw summary scores into scores which are comparable between instrument versions. The complete score-to-measure table was used to convert raw scores into user-friendly scores ranging from 0 to 100 on the latent variable in each instrument, and thus produce scores which can be compared between instrument versions, despite differences in the number and wording of individual items.

An equation is produced automatically as part of the complete score-to-measure tables produced by Winsteps which can be used to convert the raw scores in each instrument into measure (logit) scores which indicate the respondent's ability based on the latent trait i.e. FV or VQoL. This is an important outcome in the current study as it can be used within clinical practice to convert raw scores into scores which can be compared between different time points along the trajectory of childhood and adolescence.

However, the equation provided by Winsteps represents a least-squares fit regression model which produces a straight line model. In practice, this trendline may not always

fit the raw scores in the data accurately,[387] as relationships between two variables are often non-linear. In this analysis, the fit of the linear trendline was assessed in each instrument using the least-squares fit equation provided by Winsteps. If the variables were found to misfit the trendline, an iterative procedure was used to fit fractional polynomials (FPs): an extended family of curved trendlines,[387] with a view to improve the model's goodness-of-fit. R was used to fit the raw to logit scores to 2nd, 3rd and 4th order FP trendlines using the equations presented below.[387]

FPs can be defined in terms of the Box-Tidwell transformation. Let *m* be the order of the FP and **P** = (*p,…,p*$_m$) be a vector of powers, such that $p_1 < \ldots < p_m$; and **ε =** ($\varepsilon_0,\ldots,\varepsilon_m$) be a vector of parameters in the regression model. A FP of order *m* can be written as:

$$\emptyset_m(\boldsymbol{X};\ \varepsilon, \mathbf{p}) = \sum_{j=0}^{m} \varepsilon_j\ H_j\ (X) \qquad \text{(a)}$$

Where **X** is a matrix of a positive-valued covariate:

$$\mathrm{H}_0\ (X) = 1, \mathrm{P}_0\ ; \qquad \text{(b)}$$

And

$$\mathrm{H}_j(X) = \begin{cases} X^{pj} \text{ if } p_j \neq\ p_j - 1 \\ H_{j-1}\ (X) \ln(X) \quad \text{if } p_j =\ p_{j-1} \end{cases} \qquad \text{(c)}$$

With $X^{(pj)}$ defined as the Box-Tidwell transformation

$$X(pj) = \begin{cases} X\ p_j \quad \text{if } p_j\ \neq 0, \\ l\mathrm{n}\ X \quad \text{if } p_j = 0, \end{cases} \qquad \text{(d)}$$

The order of the FP specifies the model's complexity in terms of the number of functions defining it. FPs up to order *m*=4 were fitted and the FP that maximised the gain in goodness-of-fit, that is the difference in deviance between the null model, $\mathrm{measure} =\ \varepsilon_0$, and the model specified by a log and vector of powers, was chosen. The values *p* = [-2, -1, -1/2, 0, 1/2, 1, 2, 3, 4] were considered.

FPs provide very flexible shapes and are adequate to model non-linear shapes. Note that FPs were fitted adding 1 to both the observed scores and the observed measures to transform them to positive values. This doesn't alter the prediction.

4.9.4.5 Assessment of construct validity

Prior to analysis of construct validity, a second missing data analysis was conducted using the raw scores contained in each dataset, after the removal of items according to Rasch analysis. Missing data in both final versions of the VQoL and FV instruments were imputed using the same method as described in Section 4.9.4.2 (pg. 115), using multiple-pattern regression[378, 379] based on data missing at random, and using SPSS. No missing data were imputed in datasets containing raw scores obtained from the PedsQL instruments as per the published scoring instructions for the Child and Teen versions which recommend calculating the sum of items and dividing by the number of items answered.[388]

Summary scores were calculated for each FV and VQoL instrument using each of the 5 imputed datasets. Summary scores were then converted into comparable (logit) scores using the complete score-to-measure tables produced for each instrument, and recorded using SPSS. PedsQL summary scores were calculated using the published scoring instructions for both Child and Teen versions,[388] which recommend removing persons with >50% missing data, and imputing the mean of completed items for remaining persons.

Construct validity refers to an instrument's ability to measure an intended outcome (in this case, VQoL and FV) (see Section 2.4, pg. 36) and can be analysed using a postulated attribute of a population, which is assumed to reflect test performance, as a proxy for the outcome of interest (VQoL or FV). This is a useful and well-established method for determining construct validity.[389] In all cases, bivariate correlations were used. The raw summary scores were transformed to logit scores using the score-to-measure tables (and the process described in Section 4.9.4.4, pg. 120) to analyse

correlations within each suite of age-specific PROMs (e.g. Child-VQoL, Child-FV, YP-VQoL and YP-FV instruments) and also using the Child and YP datasets combined (e.g. VQoL and FV instruments). Consistent with the treatment of missing data in earlier analyses, correlations were conducted using each imputed dataset. Correlation coefficients were then averaged and transformed into z-scores using the following equation.[390]

$$z = (1/2)[\ln(1 + r) - \ln(1 - r)] \quad \text{(e)}$$

Each transformed correlation coefficient was compared to the critical value of 1.96, with a value > 1.96 indicating a significant correlation at 5% level.[391]

Construct validity of the developed suite of age-appropriate VQoL and FV instruments was assessed using correlations between VQoL and FV logit scores, summary scores on the PedsQL instruments (including the psychosocial subscale summary score), and latest recorded VA obtained from participants' clinical records. Correlations between FV logit scores and VA was used as evidence of construct validity of the FV instrument, demonstrating a relationship between logit scores based on self-report and objective clinical assessments. A positive correlation between the two variables was hypothesised, with FV scores predicting functional ability. Thus a 1-tailed significance value was specified.

Correlations between the VQoL logit and PedsQL summary scores were conducted to determine the relationship between the VQoL instrument and a generic measurement tool designed to measure HRQoL. Both overall scale and psychosocial scale PedsQL summary scores were entered into the analysis to determine the similarity between the VQoL and overall HRQoL, and the emotional, social, and school functioning scales of the PedsQL. A 2-tailed significance value was specified due to ambiguity in the current literature regarding definitions of QoL, HRQoL and VQoL and uncertainty as to the similarity between VQoL as captured in the developed instrument, and generic HRQoL as measured by the PedsQL.

Finally, a correlation analysis was conducted to determine the relationship between VQoL logit scores and participants' VA, to assess the presence of the disability paradox (see Section 2.3, pg. 28) within the population of children and YP recruited in the instrument development. A 2-tailed significance value was specified.

All correlations are presented with respect to the age-specific version of each instrument (e.g. child and YP versions) with a view to indicate construct validity of each individual instrument, and also in relation to the entire sample of 8-17 year old children and YP recruited in this study, with a view to indicate construct validity of the collaborative suite of instruments.

4.9.4.6 Analysis of unidimensionality using the final instrument versions

A second final exploratory FA was conducted following item reduction with a view to indicate dimensionality of the final instrument versions. This was conducted using only the final items in each instrument and followed the procedure described in Section 4.9.4.2 (pg. 115). Analysis was conducted using each dataset with missing data imputed and results were averaged to derive a final outcome.

Chapter 5 Growing up with visual impairment: Results from an in-depth qualitative investigation

The theories of childhood development described in Section 2.1 (pg. 20) describe growth during early life course with regard to a specific developmental capacity, and can be used to understand the *nature* of development and learning. Alternatively, theories of life course seek to understand growth and development from the perspective of *progression* through life course, and with respect to lived experience and significant life events.[392] In doing so, these theories place more emphasis on the subjective experience of life course, with respect to individual differences and varying social and environmental structures.[393-395]

As previously discussed, a wealth of literature describes the substantial impact of visual impairment upon multiple aspects of childhood, and can be categorised according to three levels of human functioning. At the level of activity and participation, the subjective components of the ICF model, studies utilising self-report from children living with visual impairment are extremely beneficial when determining the everyday impact of visual impairment, and present a 'snapshot' of the experience at one time point. However, what appears to be missing from this collection of studies is insight as to what it feels like to grow up with visual impairment, with respect to progressing through early life course, encountering significant life events, changes, and overcoming challenges during childhood. Similarly, literature documenting the impact of visual impairment during adolescence is largely focused upon improving educational and occupational outcomes with a view to optimise future occupational and financial success. To date, no literature has documented the progression of these YP through the early stages of adolescence. Understanding the lived experience during childhood and adolescence has important implications for identifying targets for and developing interventions to promote VQoL or FV, and specifically understanding at which time such interventions will be most effective.

The 4-phase method used in the current development of vision-specific PROMs for YP living with visual impairment affords the opportunity to explore, in-depth the experience of visual impairment during childhood and early adolescence, from the perspective of YP who are well placed to reflect upon their experience. Thus, the objective of this sub-component of the research project was to describe the lived experience of visual impairment during childhood and adolescence. The specific aim was to identify 1) key time points during childhood and adolescence when pertinent changes occurred, and 2) specific strategies employed by YP for dealing with the challenges encountered, with a view to inform the future development of interventions that could promote VQoL or FV. Specifically, findings may be applied to ensuring timely and optimal use of the patient-reported outcome instruments developed in this research.

A second opportunity afforded by the 4-phase method is an exploration of participants' views about, and experiences of transition from paediatric- to adult-centred ophthalmology services. This is important to advancing current knowledge which, despite demonstrating the importance of a timely and successful transition from child to adult services[396-398] is yet to explore the experiences, and transition-related needs of YP living with visual impairment. Since the interviews conducted in phase 1 were conducted with YP at either side of the threshold of transition, the opportunity was taken to probe their experience of clinical care during childhood and more recently as they approached the conventional age of transition. Thus, the data collected in phase 1 were analysed with a third aim of exploring the views about, experiences, and transition-related needs of YP with visual impairment so as to develop an evidence-base to inform transition planning and provision in ophthalmology.

5.1 Phase 1 Participation Rate

Seventeen YP aged 16-19 years participated in phase 1 (40.48% response rate). The demographic characteristics of these participants are shown in Table 9 (pg. 146). All participants were recruited from source 1.

Most in-depth interviews were conducted at participants' homes and one interview was conducted in a private room at one of the primary recruitment centres. All participants were encouraged by the researcher to take part in in-depth interviews alone to avoid the influence of other family members however, due to the interviews being conducted at participants' family homes, siblings and parents attended in a number of cases. Nevertheless, efforts were made to address the participant themselves. The duration of in-depth interviews ranged from 40.2-139.36 minutes (Median = 88.09 minutes, IQR = 39.46).

The point of data saturation was judged as the point at which no new themes/issues emerged from interviews and this threshold was judged to have been reached at the 15th interview. To ensure results were comprehensive, a further 2 interviews were conducted, one of which incorporated elements of the first draft of both instruments.

5.2 Findings/themes: Challenges, changes, and coping strategies

Qualitative analysis revealed 5 overarching themes related to the experience of growing up with visual impairment, the challenges, and changes YP recalled. A sixth theme described participants' coping strategies. Finally, key time points of change were identified.

5.2.1 Acceptance of and adaptation to visual impairment

Acceptance of, and adaptation to visual impairment was an aspect of childhood and early life course spontaneously discussed by most participants. Regardless of manifestation of visual impairment, participants viewed their impairment as a significant challenge which had to be overcome. Acceptance was a key component of overcoming the challenge of visual impairment which took time and effort. The study participants were visually impaired due to a range of ophthalmic conditions with different manifestations, and described different stages of the acceptance process.

5.2.1.1 Differences between children/YP with early and late onset visual impairment

Pertinent variations were apparent among those with early and late onset visual impairment. Participants with early onset visual impairment often described visual impairment as a personal attribute, making them unique. This attribute was fixed and had been internalised throughout childhood, as participants had little, or no, previous experience of full vision, and appeared to have endorsed the notion that their vison would not improve in the future. When probed about the impact of visual impairment during childhood, most participants with early onset visual impairment had difficulty pinpointing differences in their experience compared to that of sighted children, describing "not knowing any different" (Male, 16 years, VI, early onset). Those with early onset visual impairment demonstrated high degrees of resilience. Participants tried hard to complete activities in spite of the functional impact of their impairment and often as attempts to prove others wrong. The phrase "I just get on with it" (Female, 16 years, VI, early onset) was often used:

> *"I think I adapted to it from a young age. Cos when you're born with it, it's not really an adaptation. It's more like that's who you are." (Male, 16 years, SVI/BL, early onset).*
>
> *"It's to prove to people that, no matter what disability you've got, you know what I mean? It was my decision to do a blind-folded bungee-jump. I knew how scary it was. Everyone was like 'she's not gonna jump, not gonna jump'. The teacher said 'she's not gonna jump'. I swan dived off!" (Female, 16 years, VI, early onset).*

In comparison, participants with late onset visual deterioration and subsequent prior experience of visual function, described acceptance as a challenging, ongoing process often associated with negative psychological well-being. Late onset visual impairment significantly impacted participants' sense of identity which had been newly established during childhood and adolescence. Participants described coming to terms with their new identity over time and difficulties navigating social encounters in light of this change. Negative responses from others triggered participants' recalibration of expectations and views about themselves. Two participants who had late onset

progressive visual deterioration reported treatment for depression and feelings of grief and loss, signalling that they had not yet reached a point of full acceptance. Ongoing changes in functional vision throughout the course of childhood and adolescence meant that further changes had to be made to participants' choice of physical and leisure activities. Being able to find a substitute for an activity which was no longer possible enhanced psychological well-being, but was not always possible:

> *"You don't want family or friends who you've seen before, and have seen you seeing, noticing that it has got bad." (Male, 16 years, SVI/BL, late onset).*
>
> *"It really upset me. I lost so many friends because I wasn't Pearl. I was Pearl who just found out she has got an eye disability. So I had to mature like that [snaps fingers]. I had to do things for myself […] A lot of people thought I was the person who had changed […] and people didn't like it." (Female, 16 years, VI, late onset).*
>
> *"My eyes had got worse. And then I had to adapt myself again […] So it's like losing someone. Cos you're constantly having to, like, like grieving I suppose." (Female, 19 years, SVI/BL, late onset).*
>
> *"I used to love trampolining, but I had to stop because of my eyes." (Female, 19 years, SVI/BL, late onset).*
>
> *"Like watching TV, cos they [those with early onset VI] haven't done it. And for them, they don't care. But if you've had sight, that's the first thing. It's like teaching me something new." (Male, 16 years, SVI/BL, late onset).*

5.2.1.2 Differences between children/YP with differing severity of visual impairment

Participants who had visual impairment which was classified as severe (SVI/BL) were more resigned to the impact of visual impairment than those who had milder forms of visual impairment. Participants with visual impairment classified as SVI/BL described understanding the restrictive nature of their impairment through experience, and developing realistic expectations of the impact over time. This understanding enhanced acceptance of visual impairment, allowing YP to internalise the functional limitations of visual impairment as an aspect of personality or self-image, and subsequently have positive attitudes towards their limited functional ability.

In comparison, and supporting the presence of the disability paradox (see Section 2.3, pg. 28) in the current sample of YP living with visual impairment, participants with milder forms of visual impairment described hopes that their vision may improve to the extent that they may be able to take part in activities which were currently impossible. Being on the threshold of participation meant that participants were not willing to accept the functional impact of visual impairment in the same way as those with visual impairment classified as SVI/BL. For example, whilst all participants expressed some degree of disappointment or frustration about not being able to drive in the future, those with mild forms of visual impairment described hopes that someday their vision may improve to the level at which they would be legally permitted to drive:

> *"That little niggle in the back of your mind 'oh what happens if my eyesight gets better?' I tried to push myself to try and see a number plate and it wasn't working. I tried my damned hardest to try." (Female, 16 years, VI, early onset).*

5.2.2 Social Environment

Social relationships were dynamic and changeable during the course of childhood. During early childhood, relationships were primarily established within family communities. Siblings and cousins were described as key playmates and valuable friendships were developed with others who attended the same nurseries and pre-schools. Making new friends and losing, or "drifting away" (Male, 16 years, VI, early onset) from old friends is a normative part of childhood for all YP, regardless of visual impairment. As YP with visual impairment developed, appreciation was increasingly paid to 'true' friends who could accept visual impairment as part of their identity and offer functional support, such as guiding in public places, or reading the board at school:

> *"[When I can't see the board at school] I just put my pen down and wait. I'll give my friend 'the look' and he'll be like 'yeah'." (Male, 17 years, VI, early onset).*

As participants developed through childhood, they described increasing awareness of the broader social environment and public perception of disability. Some described experiences of discrimination which impacted willingness to use assistive devices such

as magnifiers or white canes when in public. In particular, YP with visual impairment described difficulties making sure that others were aware of their functional limitation e.g. when crossing the road, but at the same time did not patronise them or embarrass them in public.

5.2.2.1 Relationships with adults

Adults such as teachers and parents were increasingly respected and appreciated as *equals* rather than superiors or carers during the course of childhood and adolescence. Some participants identified teachers who they "could have a laugh with" (Male, 17 years, VI, early onset), or whom they had recently identified as "mates" (Male, 16 years, SVI/BL, early onset). YP with visual impairment valued new and increasingly mature relationships with adults and peers on the basis that these relationships enhanced feelings of safety and support. Instead of playing playground games in which they would likely be excluded or may be at risk of harm, YP discussed preferences of sedentary activities such as chatting with friends in the canteen.

Adults such as teachers and parents were increasingly identified as role models throughout the course of adolescence, and contributed to the development of self-esteem among YP with visual impairment, providing a sense of direction in relation to their future and, in particular, the types of careers or activities which would be possible in light of the functional impact of visual impairment:

> *"When you're at senior school, it's weird cos it changes immediately. You used to play football, rugby, that sort of thing. But now most of the time we chat. Just walk around, have a conversation. Get food, that sort of thing." (Male, 16 years, SVI/BL, early onset).*
>
> *"My uncle's a lawyer. And he's told me stories about people he's dealt with […] I like the idea of it and how he's, even though they've had such a rough background, he's helped them. I would like to do that for other people as well." (Male, 16 years, SVI/BL, early onset).*
>
> *"I did catering in Year 11 and my teacher got me into doing a lot of cooking. And my nan, she used to teach me a lot of cooking as well. So I got it from them really." (Female, 17 years, VI, early onset).*

5.2.2.2 Romantic relationships

Having a boyfriend or girlfriend was described by YP with visual impairment as enjoyable and comforting, and preferable in light of having somebody other than a parent, carer, or teacher, to offer functional support. Romantic partners could offer support in a way which was discreet and subsequently eliminated the psychological burden of having to rely on parents. Despite favouring romantic relationships, YP with visual impairment described challenges maintaining relationships during times of progressive visual deterioration:

> *"I lost quite a few friends when I found out. I was in a relationship with someone and then they ended up ending the relationship cos they didn't think they could cope with someone with an eye problem." (Female, 17 years, VI, early onset).*

5.2.3 Developing and asserting independence and responsibility

All participants described a growing need for autonomy as they grew up, which was signalled by the development of independence and responsibility throughout childhood. Some participants described work experience they had recently undertaken and how they gained increased responsibility for time management and safety of others. Specifically, adolescents with visual impairment described a shift between forced independence, in which parents would encourage participants to 'brave' new situations or environments independently, and self-guided independence in which participants were willing to try new tasks, engage in new activities, and communicate with others without help from a parent, carer, teacher or peer.

5.2.3.1 Barriers to developing independence

Many environmental barriers were perceived as restricting participants' attempts to develop independence in light of visual impairment. These included menus at cafés or fast food restaurants which were positioned behind the counter, glass obstacles in shops, overhanging plants in walkways, price tags which were out of reach, crowded areas, timetables at public transport stations, and inability to obtain a provisional driving license as proof of age. When faced with environmental barriers, most YP with visual

impairment described choosing the ‘safe option’ e.g. when in restaurants/cafés ordering something they had ordered before and which is within a certain (known) price range. Others described the challenges of ‘braving it’ in public which often resulted in negative psychosocial consequences such as feeling stupid, embarrassed or different from others:

> *“[I never ask for help] because sometimes people can be really arse-y towards you. Me and my friend went to SubWay and you know they have the menus on the top bit, neither me or her could see it. My friend asked the lady for help and she said ‘can’t you see the board?!’ and she went ‘I can’t I’m blind!’ Sometimes people don’t react well. We’re like aliens to them.” (Female, 17 years, VI, early onset).*

The majority of participants described the inability to drive, or take driving lessons, as an issue which was increasingly salient as they developed into adolescence and detrimental to their growing independence. The majority of YP relied on parents or siblings to courier them between home, school, and social occasions, but were increasingly aware of public perceptions of this, rendering it inappropriate or “baby-ish” (Male, 16 years, SVI/BL, early onset) in light of their age. Participants were intensely frustrated by the imposed barrier towards independence, which was magnified at the time when they and their friends became legally eligible to drive. In attempts to overcome the barrier, three participants had received mobility-related training in which they were taught strategies to travel independently. Establishing and maintaining mobility training however, was described as difficult and time consuming and participants perceived limited resources available.

> *“I’d rather be able to do it [driving] but not do it, than not be able to do it and want to do it. […] It’s about having that choice. I’d rather have the choice, but I’ll never have it, so, just got to deal with it.” (Female, 17 years, VI, early onset).*

> *“[Driving] is something which has been building up in me over time and it didn’t really affect me till I reached the age of 17 […] my sister passed her test the year before and at that point I wasn’t worried about it. But when my friends started to take it and pass it, it really hit me and I realised that I definitely wouldn’t be able to do that. Before it always seemed like something that was gonna happen in the future.” (Male, 17 years, VI, early onset).*

5.2.4 Future: Developing plans in light of visual impairment

YP with visual impairment were apprehensive about their future. Many were excited about anticipated events such as moving away from home or starting University and had positive expectations. However, there are several challenges that YP discussed in relation to their visual impairment; the most pertinent being realisation of the possibility for future visual deterioration:

> *"I was told by my opticians that I'm gonna reach a point at [age] 40 where it's gonna stay the same and after that it's gonna tail off. Eventually it's gonna get worse. But it's something that I know about now so that by the time I reach that age, hopefully I'll have found ways to work around it." (Male, 17 years, VI, early onset).*

Participants discussed how their plans for the future had developed and, in some cases, been amended, adjusted, or abandoned in light of visual impairment. Coming to terms with the impact of visual impairment upon future occupation was an ongoing process during childhood, which took place regardless of manifestation or nature of visual impairment.

Parents played an important role as advocates for plans in the future which are realistic, providing guidance and giving suggestions, and ensuring YP developed skills related to future independent-living. In some cases, YP had developed ambitious and potentially unrealistic plans for the future and parents played an important role in ensuring plans remained grounded. However, YP with visual impairment often perceived input from parents as overprotective and were frustrated about the imposed limitations.

Safety and familiarity were key considerations for YP when making plans and thinking about the future. The majority of participants discussed preferences for living somewhere close to home in the future and familiarity with the local area was essential to developing further independence:

> *"Then I thought there are a lot of careers [that I can't do]. I can't be a vet […]" (Male, 16 years, SVI/BL, late onset).*

"My dad's a plumber. Later on in life I've got to understand that I can't do manual labour because of my eyesight." (Male, 16 years, VI, late onset).

"My mum's brought me up to be able to cope. […] she's brought me up to develop strategies and be able to be independent." (Male, 16 years, SVI/BL, early onset).

"I want to go and study abroad for a year. I want to go to China but my mum won't let me go. But I want to do it all […] I wanted to study in America, but again, my mum, she's really iffy about that." (Female, 17 years, VI, early onset).

"If my university is somewhere that's really busy, and it's like in central London or something, and my flat is in central London [Dad laughs] I'm just saying!" (Male, 16 years, VI, late onset).

5.2.5 Transitions in Education

Integral to participants' retrospective account of growing up with visual impairment was the progression through education. Periods of childhood were often recalled in terms of key time points in education e.g. "when I was in Year 7 things started to change". (Male, 16 years, SVI/BL, late onset).

At the time of the interview, all participants had experienced a formal transition in education which occurred at the age of 11 years, and is based on the legal educational requirements in the UK. Two participants had experienced transitions between mainstream and specialist education and one had changed schools across separate counties. Five participants had changed school as a result of inadequate VI-support:

"You've got to make a few modifications so that I can have an education […] Well, try living with it! All they had to do is print off a couple of pages." (Female, 17 years, VI, early onset).

Not all transitions in education were formal transitions. Changes were experienced between classes, year groups, classrooms, teachers, and friendship groups. With each transition, YP experienced a range of VI-specific challenges such as navigating new environments, informing others about their VI-specific needs, adapting to new classroom layouts, and managing adaptations in increasingly frequent exams. The impact of visual impairment magnified the degree of change brought by each transition.

Challenges were overcome with varying success and often associated with changes in personality and ability to cope with visual impairment:

> *"I was always shy. I always thought no one will like me because I'm partly blind. But then, I got into secondary school, started doing Performing Arts and then my confidence started getting bigger and bigger and then as soon as I started being able to make people laugh it fuels me and I keep going." (Male, 16 years, SVI/BL, late onset).*

5.2.5.1 Social life at school

Participants described changes in social groups and norms as they progressed through education. Transitions between schools and colleges often resulted in new peer groups. Friendships with others who also had visual impairment were established when YP transitioned from mainstream to specialist education and were valued highly:

> *"I asked one of my friends who's blind how to put something on a coat hanger. I said I lay it on my bed and she goes 'I've got a better idea. If you hold it up with your mouth, and then do the top bit up'. I had never thought to do it like that." (Female, 17 years, VI, early onset).*

5.2.5.2 VI-specific educational support and demands

The level and nature of VI-specific educational support changed dramatically for most participants as they progressed through the education system. Some described variations between schools. Improvements in support enhanced aspects of psychological well-being such as feelings of belonging. Alternatively, deteriorations in VI-specific support made transitions in education particularly challenging, and increased participants' awareness of their unique educational needs and the difference between themselves and their sighted classmates:

> *"I didn't realise how much they could help me. I just assumed it was OK, and it was normal to be happening […] Until it improved I didn't really realise how much better it was." (Male, 17 years, VI, early onset).*

> *"It hadn't been fully set-up for visually impaired children." (Male, 17 years, VI, early onset).*

YP with visual impairment noticed developments in technology which took place throughout the course of their education. These were often discussed with excitement,

positive perceptions of future developments, and ability to cope with visual impairment. Blackboards changed to whiteboards which then became interactive, meaning YP were increasingly able to use devices such as cameras or CCTV devices to access resources easily. As YP progressed through education, they experienced increasing difficulty keeping up with the teacher and their classmates during lessons. Use of VI-specific devices such as magnifiers was associated with pain and fatigue at the end of the school day:

> *"When I concentrate on reading and writing I have neck-ache and it's really bad and I have to carry a lot of books behind my back so it also increases the pain." (Male, 16 years, SVI/BL, early onset).*

> *"We're on the computers all day long and like 5 hours, all the time and it's, um, it does drain you at the end of the day. It's very tiring." (Female, 17 years, VI, early onset).*

5.2.6 Coping strategies

Participants discussed a range of coping strategies that had been developed and used throughout childhood and adolescence. Despite adolescents living with conditions with varying levels of sight impairment, and manifestation of VI, the coping strategies that had been developed were often similar, including both extrinsic (i.e. those based on the availability of assistive devices and functional support) and intrinsic (i.e. those attributable to personality, beliefs and attitudes) strategies.

5.2.6.1 Perseverance

Perseverance was a coping strategy identified among those with early onset visual impairment. Participants discussed attempts to succeed in spite of the impact of visual impairment. YP often described attempts to work around the impact of VI, or find alternatives, implying profound emotional and cognitive investment. Participants with SVI/BL discussed the importance of being organised, knowing where their belongings were, and being well-prepared for new situations:

> *"Try, try, try again and if that fails, just try again! […] And after you fail, go to bed!" (Male, 16 years, SVI/BL, early onset).*

"I try to find ways. Solve problems […] It's a problem solving skill." (Male, 16 years, SVI/BL, early onset).

"I like to put my things where I remember. In the morning, say if I'm in a rush I might be looking in the wrong place. And if I was looking for something small, then I might not notice it." (Male, 16 years, SVI/BL, late onset).

5.2.6.2 Humour

Humour emerged as an important coping strategy which developed throughout childhood and adolescence, and enabled participants to evoke positive reactions from others when they made mistakes or blundered in social contexts. Participants often described 'making fun' of themselves and their impairment in front of others. When initiated by oneself, humour was an important coping strategy. However, when initiated by others, humour was seen as a barrier towards social inclusion:

"I'm quite lucky, if you can get a laugh out of it, you think why not?!" (Male, 16 years, VI, early onset).

"I'm always laughing at myself, always tripping up. Cos that's all you can do." (Female, 17 years, VI, early onset).

"The teachers understood how far was enough. If it was tasteful [then it was fine]. They're not vindictive." (Male, 16 years, SVI/BL, early onset).

5.2.6.3 Functional support from others

Functional support which could be offered by others was an important extrinsic strategy. At a basic level, all participants received functional support from family members which was largely appreciated. As participants developed, however, functional support from family members, particularly parents, became increasingly less desirable based on the perception or experience of stigma from friends and peers. Functional support which could be provided by friends became increasingly salient for YP, enabling them to engage in social activities outside of family communities:

"When I'm at the high street, my school friends are also there and when my mum holds my hand, I don't like it. Because then they'll mug me when I go to school." (Male, 16 years, SVI/BL, early onset).

"The group of friends I've got now, I don't have to ask. They'll come up to me and link arms with me anyway. Even though I don't ask!" (Female, 17 years, VI, early onset).

5.2.7 Key time points of change

Three key time-points were identified, signalling times of change in the nature of the impact of visual impairment during childhood and adolescence. All three were identified in relation to education. As YP grew up, change was increasingly related to internal, intrinsic factors such as self-development, as opposed to change which was driven by formal, policy-related transitions:

5.2.7.1 Transition from Primary to Secondary education at the age of 11 years

The transition from primary to secondary education at the age of 11 years was a mandatory transition imposed by legal educational requirements in the UK. Participants recalled expansions of social environments, and increased awareness of new social norms or conventions as outcomes of the transition. For example, differences were identified in activities during break or lunch times, as methods of social engagement transition from physical to communicative and social group norms matured. Navigation around a new educational environment required time and effort as YP established familiarity with their new surroundings.

5.2.7.2 Mid-way through Secondary education

Mid-way through secondary education was a time-point which YP described in terms of increased awareness, and early development, of independence and autonomy. It is during this time when participants experienced increased choice within education, as subjects were dropped or initiated depending on individual interests. For the majority of YP with early onset, non-progressive sight impairment, this was a time when parents permitted visits to shopping centres, cinemas, local parks, and engagement in social activities outside of school and home increased.

5.2.7.3 Transition from Secondary education into higher education

Change during the transition from secondary education into higher education at the age of 16 was increasingly related to dynamic shifts in internal values and attributes, in contrast to social or physical environments. The transition to an increasingly flexible educational institution allowed participants to experiment with personal style (i.e. in the absence of school uniform restrictions), and identify with adults as equals rather than superiors. During this period independence surged as YP were able to approach vision-related difficulties with maturity, and understand the broader social context in which visual impairment is perceived by others.

> *"I like having independence. A lot of [sixth form] is independent study which I like. You don't have to wear uniform […] It's nicer having a bit more freedom and a bit more choice in everything." (Male, 17 years, VI, early onset).*

5.3 Transition from paediatric to adult ophthalmology services: Views, experiences, and transition-related needs

At the time of the interview, 8 participants had transitioned from paediatric ophthalmic services: 6 into adult services and 2 into dedicated adolescent services. Only 2 (25%) of these participants preferred their prior paediatric service, due to its more child-centre approach to communication, although pros and cons were identified by all. The two participants now in an adolescent service identified significant positive benefits of this specialist service bridging child and adult care. Only 1 subject (14%) still in paediatric services did not want to transition, attributable to a strong relationship with their managing clinician. Two participants were unsure whether they had transitioned: both had stable visual impairment and had not been reviewed for some years.[399]

Fourteen codes emerged from analysis of interview data, identifying two key components relevant to transition '*Communication with professionals within clinical contexts'* and '*Healthcare environment'*. Both were associated with the overarching theme '*Confidence to self-manage healthcare in the future as an adult'*. *'Emotional*

attachments to child-centred care' was a further sub-theme which influenced participants' self-reported willingness to transition.[399]

5.3.1 Confidence to self-manage healthcare in the future as an adult

Participants discussed having increased responsibility for their own healthcare, with most recognising the diminishing role their parents would play once they entered adult services, in some cases describing parents as 'handing over' or encouraging them to take control and build confidence to manage their healthcare independently. They recognised that growing up involved greater maturity and transition into adult care enhanced feelings of autonomy, confidence and control. Nevertheless, attitudes varied, ranging from strong preferences to take control of the transition and subsequent clinical care, to disengagement.

5.3.1.1 Communication with professionals within clinical contexts

Participants who had transitioned described the major differences between paediatric and adult services in relation to communication with their managing clinicians.[399] This reflected, in part, the shorter duration of outpatient appointments and the larger clinical teams in adult services, which meant that participants were not certain of seeing the same clinician(s) at each visit.

> *"[In child-centred care] you'll be treated like a child but in adult clinics you'll be treated like you would be if you were in an interview." (Male, 16 years, transitioned to adult services).*

> *"[Now I've transitioned] they take a bit more time. They used to treat me like a child. They talk to you that way." (Female, 17 years, transitioned to adult services).*

Some participants who had not yet experienced a transition and remained in paediatric services described parents 'taking over' the consultation and communicating on their behalf. Attitudes towards parents 'taking the lead' varied: some felt excluded or embarrassed when parents intervened, whereas others valued their parents' input and disease-specific knowledge.

"My mum will get me involved in the conversation […] and says she'd rather me speak to them cos obviously it's my eyes and not hers but sometimes it feels like my mum and the doctor are having the conversation and I'm like 'hello, I'm here!'" (Female, 18 years, transitioned to adult services).

"I think my mum knows more about my condition than I do! So I'd rather have her there than kick her out and go 'oh I wanna do this by myself'." (Male, 16 years, not transitioned).

5.3.1.2 Environment

Participants who transitioned noted the different environment in adult services: some welcomed this, in particular the reduced sensory 'overload' of paediatric outpatient play areas but for others this was initially unwelcome and surprising, adversely impacting their feelings of belonging, confidence and involvement in healthcare.[399] However those who had not yet transitioned expressed strong dislike of child-centred environments, which was often the primary cause of desire to move into adult-centred care.

"When I had my first appointment I remember it being so different! I thought 'what is this?!' [Laughs]. Cos it's duller. You just sit there and wait and then get called and go." (Female, 18 years, transitioned to adult services).

"I won't play with the toys or watch cartoons. […] I'd rather watch some news or sports on the TV and sit quietly." (Male, 16 years, not transitioned).

"There's loads of little kids. You're the big one and you don't feel like you're in the right place anymore." (Female, 18 years, transitioned to adult services).

"I hate little kids! […] because I'm looking there [gestures at eye level] and they're quite short. Sometimes they're running. And I want to smack them!" (Male, 16 years, not transitioned).

Notably two participants who had transitioned into specific adolescent/young person services valued the new clinical environment, appreciating, in particular, the opportunity for contact with a peer group similar in age, which enhanced their sense of belonging and age-appropriate provision of televisions and computers.

"In the kid's one there were baby things to do there, whereas when you go to the teenage ward there's more grown up things. […] there's a pool table, TV's and computers." (Male, 16 years, transitioned to adolescent services).

5.3.2 Emotional Attachments to Paediatric Ophthalmology Services

Emotional attachment to the managing clinician was cited as a reason to be unwilling to transition by two participants with late onset and/or progressive visual impairment: one participant explained the role of their managing clinician in the process of diagnosis and acceptance of progressive visual deterioration and the desire that this practitioner would be involved in her future health care.[399] The other described losing contact with his paediatric ophthalmologist as causing loss of accessible vision-specific support, which subsequently impacted his acceptance of, and adaptation to late onset visual impairment.

> *"When I was with Doctor J and they told me that I'd eventually go blind, she came over and gave me a hug […] I've stayed in children's' clinics simply because of Doctor J. I have a funny feeling that when I move to adult's clinics I will still see Doctor J." (Female, 17 years, not transitioned).*
>
> *"There was more support. Doctor P was there and she would understand some stuff." (Male, 16 years, transitioned).*
>
> *"I'm a bit sad that I'm leaving my doctor. Most of the doctors I have known for quite a few years, I've got used to them." (Female, 17 years, not transitioned).*

5.4 Summary

Findings from this stream of instrument development indicate that the impact of visual impairment during childhood and adolescence is fluid and dynamic, changing as a result of transitions in school, physical growth, and psychological development. YP with visual impairment identified five core elements spanning intrinsic, personality-based changes to broader social contexts and the influence of others during childhood. Critical to development with visual impairment was a process of adaptation and adjustment, for which variability was seen among those with variations in timing of visual impairment onset and severity.

With regards to transitions from paediatric to adult ophthalmology services, findings indicate some variability in the content and timing of current transition processes in the

UK. Aspects of transition valued by YP, such as age-appropriate communication, suitable physical clinical environments, and an age-appropriate peer group also being served by the service, are likely to be associated with effective transition.[399]

Chapter 6 Results from psychometric evaluation

Results from each phase of instrument development are presented in this chapter including development, modification and validation. After the first phase a master item pool was developed and updated after each subsequent phase.

6.1 Study sample characteristics

A total of 383 patients were identified through source 1. All identified patients were invited to take part in one of the 4 phases of instrument development and a total of 115 YP identified through source 1 participated, rendering the overall participation rate for source 1 30.03%.

During phases 3 and 4, 20 external patient identification centres (PICs) were included as a second source of recruitment. Determining the exact number of potentially eligible patients identified by the PICs is problematic due to an absence of a prior sampling framework but rather rough estimates which were provided by each centre regarding the number of eligible patients they could potentially identify within the time frame of the research project. However, a total of 14 YP identified through source 2 participated.

Thus, in total, 129 YP participated in the instrument development; each contributing to one phase only.

The demographic characteristics of the overall sample are shown in Table 9 (pg. 146). As it was not possible to determine the demographic characteristics of the non-participators identified and invited directly by PICs, this Table shows only those identified through source 1. Notably, the sample size in each phase of development varied according to the specific aims of the phase, the approaches used to collect data, and the nature of the data collected. The majority of patients included in the sampling framework were conserved for the final phase of data collection, where quantitative statistical analyses were required.

Table 9. Demographic and clinical characteristics of the full sample of YP who participated compared to those who did not participate (where known for source 1).

Demographic characteristic	**Participators *n* = 115, *n* (%)**	**Non-participators *n* = 268, *n* (%)**
Age		
13	13 (11.3)	46 (17.16)
14	22 (19.13)	34 (12.69)
15	20 (17.39)	37 (13.81)
16	21 (18.26)	61 (22.76)
17	32 (27.83)	77 (28.73)
18	6 (5.22)	13 (4.85)
19	1 (0.87)	-
Gender		
Male	62 (53.91)	141 (52.61)
Female	53 (46.09)	127 (47.39)
Ethnicity		
White British/other	82 (71.3)	113 (42.16)
Black Caribbean/African/other	4 (3.48)	20 (7.46)
Asian Indian/Pakistani/Bangladeshi/Chinese/other	20 (17.39)	41 (15.3)
Mixed	3 (2.61)	4 (1.49)
Other	4 (3.48)	9 (3.36)
Unknown/not stated	2 (1.74)	81 (30.22)
Severity of visual impairment		
LV: logMAR ≤ 0.46	2 (1.74)	8 (2.99)
VI 1: logMAR 0.48-0.70	46 (40)	91 (33.96)
VI 2: logMAR 0.72-1.00	38 (33.04)	84 (31.34)
SVI: logMAR 1.02-1.30	15 (13.04)	32 (11.94)
Blind: logMAR ≥ 1.32	14 (12.17)	53 (19.78)

Demographic characteristic	Participators *n* = 115, *n* (%)	Non-participators *n* = 268, *n* (%)
Index of multiple deprivation quintile rank		
1: most deprived	17 (14.78)	53 (19.78)
2	18 (15.65)	73 (27.24)
3	22 (19.13)	59 (22.01)
4	26 (22.61)	47 (17.54)
5: least deprived	32 (27.83)	36 (13.43)

The demographic characteristics of the sample represent the population of children living with SVI/BL in the UK well in terms of gender and ethnicity but marginally underrepresent children coming from the most deprived quintile of index of multiple deprivation (14.78% in this study compared to 40% in the UK population[173]). The similarity of the participants included in this sample to the overall population of children and YP living with visual impairment (versus SVI/BL) is unknown due to limited information about the frequency of childhood visual impairment (as opposed to SVI/BL) in the UK and internationally.

Notably, the composition of demographic characteristics of the sample varied in each phase (see Table 11, pg. 151), with phase 4 (the final stage of psychometric evaluation requiring the largest number of participants) containing the greatest spread of characteristics, and being most representative of the population of children living with SVI/BL in the UK.[173]

6.1.1 Relationship between demographic characteristics and participation

Table 9 (pg. 146) shows a substantial number of participants who did not participate and for whom ethnicity was recorded as unknown. This was likely a result of the method used to identify eligible patients using the data stored in electronic health records. Because the families of the YP who did not participate in the research did not

complete a Family Background Questionnaire, these data can be described as missing not at random. Thus, participants with ethnicity recorded as unknown (*n* = 83) were excluded from the logistic regression model. The sample size of some demographic characteristics (namely, ethnic minorities and LV) was small. Thus, ethnicity was re-grouped into 2 categories (White British/other and non-White), and the LV and VI1 categories of severity of visual impairment were merged prior to analysis of the relationship between demographic characteristics and participation in the study.

A test of the full final model against a constant only model was statistically significant (chi-squared = 16.62, *p*=.001, df = 4). Predicted participation overall was 65.3% (87.7% for non-participated and 28.3% for participated). The Wald criterion demonstrated that only IMD and severity of visual impairment made a significant contribution to prediction (*p* <.1). Specifically, a one unit increase in IMD (from more to less deprived) means YP are 1.6% times more likely to participate. Having visual impairment classified as Blind, compared to LV/VI1 means participants are 52.7% times less likely to participate.

Table 10. The coefficients and significance of variables (demographic and clinical characteristics) included in the logistic regression models.

	Univariable analyses				**Multivariable analysis**				**Final multivariable analysis**			
Predictor	**Coefficient**	**Wald's χ^2**	***p***	**AOR**	**Coefficient**	**Wald's χ^2**	***p***	**AOR**	**Coefficient**	**Wald's χ^2**	***p***	**AOR**
Age	-0.39	0.24	0.62	0.96	-	-	-	-	-	-	-	-
Gender (Male constant)	0.13	0.3	0.58	1.14	-	-	-	-	-	-	-	-
Ethnicity (White British/other constant)	-0.55	4.52	0.03	0.58	-0.20	0.51	0.48	0.82	-	-	-	-
Severity of visual impairment (LV/VI1 constant)	-	5.22	0.16	-	-	4.11	0.25	-	-	4.29	0.23	-
VI2	-0.14	0.23	0.63	0.87	-0.16	0.29	0.59	0.85	-0.17	0.33	0.57	0.85
SVI	-0.38	1	0.32	0.68	-0.33	0.73	0.39	0.72	-0.34	0.79	0.38	0.71
Blind	-0.8	4.85	0.03	0.45	0.74	3.92	0.05	0.48	-0.75	4.1	0.04	0.47
Index of multiple deprivation (from higher deprivation to lower deprivation)	0.02	14.52	0	1.02	0.02	10.36	0	1.02	0.02	13.57	0	1.02

6.2 Participation rate

Table 11 (pg. 151) shows the demographic characteristics of the YP who participated in each phase of instrument development, including those who were recruited through source 2. Participation ranged from 26.23% (phase 2) to 40.48% (phase 1). Participation in phase 3 and 4 was 31.34% and 26.4% respectively, excluding those who were invited through source 2.

All but one in-depth interview in phase 1 and expert consultations in phase 2 were conducted at participants' family homes. Three YP took part in phase 2 and two YP took part in phase 4 shortly after their 18th birthday and were deemed close enough to the age threshold to be included in analyses.

Table 11. Demographic and clinical characteristics of YP who participated in each phase of instrument development.

Demographic characteristic	**Phase 1 *n* = 17, *n*(%)**	**Phase 2 *n* = 16, *n*(%)**	**Phase 3 *n* = 23, *n*(%)**	**Phase 4 *n* = 73, *n*(%)**
Age				
13	*	3 (18.75)	4 (17.39)	8 (10.96)
14	*	2 (12.5)	6 (26.09)	19 (26.03)
15	*	3 (18.75)	4 (17.39)	15 (20.55)
16	7 (41.18)	2 (12.5)	4 (17.39)	14 (19.18)
17	8 (47.06)	3 (18.75)	5 (21.74)	15 (20.55)
18	1 (5.88)	3 (18.75)	-	2 (2.74)
19	1 (5.88)	-	-	-
Gender				
Male	10 (58.82)	8 (50)	13 (56.52)	39 (53.42)
Female	7 (41.18)	8 (50)	10 (43.48)	34 (46.58)
Ethnicity				
White British/other	11 (64.71)	12 (75)	19 (82.61)	54 (73.97)
Black Caribbean/African/other	-	-	-	3 (4.11)
Asian Indian/Pakistani/Bangladeshi/Chinese/other	4 (23.53)	4 (25)	4 (17.39)	11 (15.07)

Demographic characteristic	Phase 1 *n* = 17, *n*(%)	Phase 2 *n* = 16, *n*(%)	Phase 3 *n* = 23, *n*(%)	Phase 4 *n* = 73, *n*(%)
Ethnicity continued…				
Mixed	1 (5.88)	-	-	2 (2.74)
Other	1 (5.88)	-	-	-
Unknown/not stated	-	-	-	3 (4.11)
Severity of visual impairment				
LV: logMAR ≤ 0.46	1 (5.88)	-	-	1 (1.37)
VI 1: logMAR 0.48-0.70	8 (47.06)	9 (56.25)	9 (39.13)	20 (27.4)
VI 2: logMAR 0.72-1.00	3 (17.65)	5 (31.25)	7 (30.43)	30 (41.1)
SVI: logMAR 1.02-1.30	2 (11.76)	1 (6.25)	4 (17.39)	8 (10.96)
Blind: logMAR ≥ 1.32	3 (17.65)	1 (6.25)	3 (13.04)	14 (19.18)
Timing of onset of visual impairment				
Early (≤2 years)	15 (88.24)	10 (62.5)	21 (91.3)	58 (79.45)
Late	2 (11.76)	6 (37.5)	2 (8.7)	15 (20.55)
Nature of deterioration of visual impairment				
Stable	12 (70.59)	5 (31.25)	21 (91.3)	60 (82.19)
Progressive	5 (29.41)	11 (68.75)	2 (8.7)	13 (17.18)

Demographic characteristic	**Phase 1 *n* = 17, *n*(%)**	**Phase 2 *n* = 16, *n*(%)**	**Phase 3 *n* = 23, *n*(%)**	**Phase 4 *n* = 73, *n*(%)**
Index of multiple deprivation quintile rank				
1: most deprived	1 (5.88)	2 (12.5)	1 (4.35)	17 (23.29)
2	2 (11.76)	-	5 (21.74)	14 (19.18)
3	4 (23.53)	4 (25)	4 (17.39)	11 (15.07)
4	8 (47.06)	3 (18.75)	5 (21.74)	12 (16.44)
5: least deprived	2 (11.76)	7 (43.75)	8 (34.78)	19 (26.03)

* YP aged < 16 years were not recruited to phase 1 (as described in Section 4.6.4.2, pg. 101).

In phase 4, 194 children (aged 8-12 years) were identified as eligible through source 1 and invited to take part. Of these, 60 participated (response rate: 30.93%). Additionally, 26 children who were identified and invited through source 2 participated. Overall, 86 children participated in phase 4. Notably four children completed Child questionnaire booklets: three participated shortly before their 8th birthday, and the other was aged 13 years and 3 days old on the date of participation, and were deemed close enough in age to the threshold to be included in analyses. Their characteristics are shown in Table 12 (pg. 155). This sample was closest (in comparison to the sample in phases 1-4 of YP aged 13-17 years) to being representative of the population of children living with SVI/BL in the UK.[173]

Table 12. Demographic characteristics of children who participated in phase 4.

Demographic characteristic	**Phase 4 *n* = 86, *n*(%)**
Age	
7	3 (3.49)
8	19 (22.1)
9	22 (25.58)
10	9 (10.47)
11	15 (17.44)
12	17 (19.77)
13	1 (1.16)
Gender	
Male	51 (59.3)
Female	35 (40.7)
Ethnicity	
White British/other	53 (61.63)
Black Caribbean/African/other	9 (10.47)
Asian Indian/Pakistani/Bangladeshi/Chinese/other	21 (24.42)
Mixed	3 (3.49)
Other	-
Unknown/not stated	-
Index of multiple deprivation quintile rank	
1: most deprived	21 (24.42)
2	14 (16.28)
3	16 (18.6)
4	15 (17.44)
5: least deprived	17 (19.77)
Missing	3 (3.49)

Demographic characteristic	**Phase 4 *n* = 86, *n*(%)**
Severity of visual impairment	
LV: logMAR ≤ 0.46	5 (5.81)
VI 1: logMAR 0.48-0.70	37 (43.02)
VI 2: logMAR 0.72-1.00	32 (37.21)
SVI: logMAR 1.02-1.30	5 (5.81)
Blind: logMAR ≥ 1.32	7 (8.14)
Timing of onset of visual impairment	
Early (≤2 years)	73 (84.88)
Late	13 (15.12)
Nature of deterioration of visual impairment	
Stable	55 (63.95)
Progressive	31 (36.05)

Data were collected from both children/YP and their parents or guardians in the form of a proxy report administered in phase 4.

Of the 159 families recruited, data were excluded from a) one YP because the parent report was returned in absence of a completed YP report and b) one YP because they failed to return any consent forms even though they returned completed questionnaire booklets. As a result, a total of 86 children and 71 YP were included in phase 4 analyses.

Only two YP used the electronic version of the instruments. However, one of these participants was excluded prior to further analysis because no consent forms were returned. No children participated using the electronic version of either instrument.

6.3 Development of the vision-related quality of life (VQoL) instruments

6.3.1 Phase 1: Item development and adaptation

Table 13 (pg. 158) shows an adaptation of the final hierarchy of child-centred data which was developed in the foundation research.[5-7] This table was used to compare the self-reported impact of visual impairment between children (aged 10-15 years old) and YP (aged 16-19 years old), highlighting areas of overlap and discrepancy between the two age-groups and thus justifying the suitability of existing instrument items (taken from the foundation research) and the need for new, age-appropriate items. Areas of overlap and discrepancy were organised according to the six overarching themes in Rahi et al.'s[5] VQoL thematic framework. In developing the YP-VQoL instrument, aspects of functioning – home, school and leisure which were discussed in relation to psychological and emotional well-being were considered for inclusion as items in the instrument, as opposed to aspects of self-reported ability which were considered for inclusion in the YP-FV instrument (see Section 6.4.1, pg. 227).

Table 13. Areas of overlap and discrepancy between VQoL and FV reported by 16-19 year old YP and 10-15 year old children.

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
Social relationships, acceptance and participation	Relationships with family (parents, siblings, and extended family), Friends (at home and at school), Peers, teachers and general public, Related issues of bullying and teasing, Isolation, fitting in, inclusion and exclusion, Stigma associated with impairment and disability, Current aspirations, Communication.	I've got a few that don't treat me any different to anyone else. (Male, 16, VI, early onset) Someone took my glasses, she stole, took my glasses […] and she hid em. […] and I feel lost without my glasses and I got a bit upset. Apparently they were joking but to me it was bullying. (Female, 17, VI, early onset) Cos they're all talking about different things that I couldn't talk about cos I hadn't experienced it. Like driving, for example.	Sense of burden on others, Unwanted/unhelpful social support and negative attention, Social support in public places/situations, Long-term romantic relationships.	It's nice, because I know that I can rely on him. But at the end of the day, he's only 10. […] it makes me feel a bit guilty at times. (Female, 17, VI, late onset) [*One of my teaching assistants is*] really fussy and there are times when I can be just doing [*my work*] and, and she's looking over my shoulder, peering. Or she'll take my textbook, and give it to someone else. (Male, 16, VI, early onset) My mum came up with a system.

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
		Social relationships, acceptance and participation continued… (Female, 19, SVI/BL, late onset)		*Social relationships, acceptance and participation continued…* In the dark I'd link arms with her, and she'd squeeze my arm (Female, 17, VI, late onset) I was in a relationship with someone and then they ended it cos they didn't think they could cope with someone with an eye problem. (Female, 17, VI, late onset)
Independence and autonomy	Support (e.g. at school, home, leisure, mobility training),	That's irritating but you develop ways around it. (Male, 16, SVI/BL, early onset)	Increased responsibility, Driving as a means to being independent.	I used to get the train with friends. And then, in the last couple of years I've done university trips on my own […]

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
	Independence and autonomy continued… Access to adaptive technologies (at home and school), Having a Learning Support Assistant, Daily independent living activities (e.g. travel, shopping, money, roads, self-care, making food and drinks, housework, telling the time), Mobility issues (e.g. being guided, using a cane),	*Independence and autonomy continued…* I suppose you have responsibility [*to do the cleaning*] but you can't always do it to the best of your ability. I feel quite independent at home. Cos my mum's brought me up to be able to cope […] to develop strategies and be able to, be independent. (Male, 16, SVI/BL, early onset)		*Independence and autonomy continued…* and gone off to see friends on my own and done my own thing. I've become a lot more independent as I've grown up. (Male, 17, VI, early onset) It's not that easy to get about, so I've had to rely on everyone else to drive me about. (Male, 17, VI, early onset) Yeah, it makes me feel a lot less independent, cos I have to rely on other ways of getting around other than driving myself. (Female, 17, VI, early onset)

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
	Independence and autonomy continued… Vulnerability, and communication (e.g. reading text messages and emails, using mobile phones).			*Independence and autonomy continued…* Um, but then when my friends started to take [*their driving test*] and pass it. That's when it really hit me and I realised that I definitely wouldn't be able to drive. (Male, 17, VI, early onset)
Psychological and emotional well-being	Emotions relating to living with visual impairment, Stigma associated with impairment and disability, Coping, Frustrations,	I used to get a bit frustrated when it was something which I couldn't see. (Male, 17, VI, early onset)		

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
	Psychological and emotional well-being continued… Privacy, Acceptance of visual impairment, Uncertainty, Body and self-image, including appearance of the eye, wearing glasses/contact lenses, identifying as impaired or disabled, Self-esteem.	*Psychological and emotional well-being continued…* My eyes had got worse. And then I had to adapt myself again. […] So it's like losing someone. Cos you're constantly having to adjust, like grieving I suppose. (Female, 19, SVI/BL, late onset) I know that eventually it's gonna get worse and worse.		

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
		Psychological and emotional well-being continued… But it's something that I know about now so that by the time I reach that age, hopefully I'll have found ways to work around that. (Male, 17, VI, early onset) I just learn to get on with it. (Male, 16, SVI/BL, late onset)		
Future – aspirations and fears	Challenges, Restrictions, Opportunities for future education and career,	Then I thought, yeah, cos there are a lot of careers I can't do. (Male, 16, SVI/BL, early onset)	Growing acceptance of future impact of visual impairment,	In another life, or if I had vision, I would probably be a Royal Marine. It's one of those things where you can't do it. (Male, 16, SVI/BL, early onset)

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
	Future – aspirations and fears continued… Independent living and mobility (especially driving and reliance on transport), Visual prognosis, Worries, concerns, uncertainties, Future relationships, e.g. dating, marriage and having children.	*Future – aspirations and fears continued…* One day I could just be blind. So I hope for a cure but, at the same time, if it happens, it happens. Nothing I can do about it! (Male, 17, SVI/BL, early onset) I'd like to be a stockbroker or something like that. (Male, 16, VI, early onset)		*Future – aspirations and fears continued…* [*My visual impairment*] narrows what kind of job prospects I could have. I wanted to join the police force. But that had to be knocked on the head pretty quickly cos you need perfect vision for that. (Male, 18, VI, early onset) Bringing [*children*] up. I'd have to do it on my own, in some respect. (Female, 19, SVI/BL, late onset)
Functioning – school, home and leisure	Feelings relating to activities and functioning.	I get a bit upset and disappointed when I can't		

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
		Functioning – school, home and leisure continued… [*play football*]. I used to do tennis a lot with my friends and then realised I couldn't really play competitively with them. (Male, 17, VI, early onset) I used to play a lot of games, like on PlayStation, and stuff but now, I have tried a few times but it just gets, when you hear the sounds and stuff. And I remember some of the controls. But it's not the same. So that gets frustrating.		

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
		Functioning – school, home and leisure continued… Especially if you're at your friends or someone's and they're all playing games and you just sort of, sitting there. (Male, 16, SVI/BL, late onset)		
Treatment of eye condition	Visits to the eye-clinic/hospital (e.g. prolonged waiting times, invasive eye examinations, eye tests, use of eye drops, communication with the doctor),	No I hated going [*to hospital*]. Hated it. (Male, 16, SVI/BL, early onset)	Transition from child- to adult-centred care.	[*My visits to hospital*] slowly tailed off as I got older. I was having an eye test, which then transferred down to just having an eye test at an opticians'. (Male, 17, VI, early onset)

VQoL domain[5]	Domain-pertinent issues identified from interviews previously conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
	Treatment of eye condition continued… Related treatment, operations, and subsequent use of medication, Wider impact, such as missing school and subsequently getting behind with school work.	*Treatment of eye condition continued…* It's annoying cos I'm usually in school and I always have to miss school. I have to go in and out of school. It's annoying cos I miss half the lesson. (Female, 16, VI, early onset) I never used to like [*the eye drops*]. They used to sting. (Male, 16, VI, late onset)		*Treatment of eye condition continued…* I told you I hate children. When they're noisy. When I'm in the waiting room, they're all noisy and I'm 16. (Male, 16, SVI/BL, early onset)

Substantial overlap was found between the self-reported impact of visual impairment, and VQoL issues raised by younger children (aged 10-15 years) in the foundation research and YP (aged 16-19 years) (see Table 13, pg. 158), with both children and YP referencing similar physical and social environments, and daily challenges in light of visual impairment. For example, all children and YP discussed their relationships with friends and family members as valuable aspects of everyday life. However, differences emerged in the way YP described the frequency of these issues and emphasised them as aspects of everyday life and global well-being. For example, it is only during later adolescence (e.g. between the age of 16-19 years) that YP perceived the burden they place upon others when asking for help or needing care. Another example of the differences between child and YP self-report is evident when YP discussed the impact of their vision upon their ability to drive. Many children discussed this as an important barrier in the future. However YP discussed not being able to drive as an aspect of their everyday life which limits independence and inclusion with peers in conversations about driving lessons.

As a result of substantial overlap, the majority of new, age-appropriate codes mapped well onto the existing VQoL items developed in the foundation research. Although most codes reflected some degree of new, age-specific attitude and conceptualisation of visual impairment, the majority of existing VQoL items was deemed suitable for reflecting the underlying issue. In total, codes were mapped onto 31 existing VQoL items. These items covered the VQoL domains: school life, home life, social life, independence, psychological well-being, future aspirations and concerns, and experience of eye clinics. Five items included in the foundation VQoL instrument did not receive any new age-appropriate codes (un-coded) and 12 codes were not mapped onto existing VQoL items, and considered for inclusion as new age-specific items.

Ten VQoL items which had been removed during the foundation instrument development were entered into the first draft instruments based on the relevance of the new, age-appropriate codes. Codes which contributed to the inclusion of these VQoL

items primarily centred upon YP's experience of teasing, mocking, or bullying when at school, and awareness of the negative impact of visual impairment upon their future career development (see Figure 2, below).

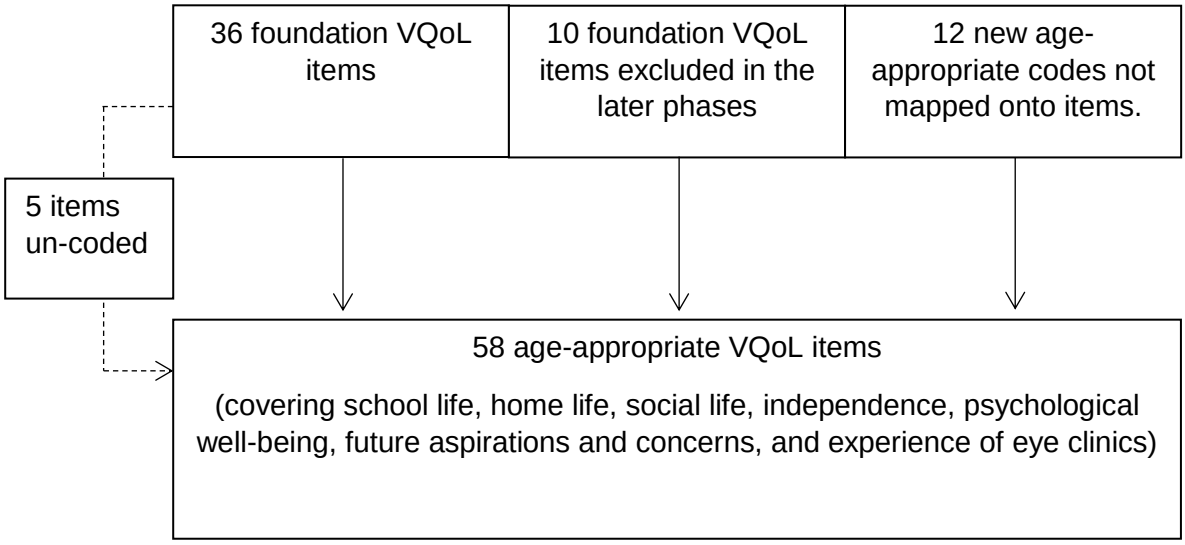


Figure 2. Flowchart showing the origin of items included in the first draft VQoL instrument.

Using the codes, a draft VQoL instrument for YP aged 13-17 years was developed. Table 14 (pg. 170) shows the items included in the first draft instrument.

Table 14. Items included in the first draft VQoL instrument.

Existing items (*n* = 31)	I have got some good friends
	I make new friends easily, despite my eyesight problems
	I get along with my family
	I can stand up for myself if someone picks on me
	My friends understand about my eyes
	I get treated the same as everyone else
	My friends help me at school
	I feel left out because of my eyesight
	I feel like I fit in
	My friends encourage me to join in their activities
	My teachers understand about my eyesight
	I feel different from other children
	In spite of my eyesight, I am independent
	I am comfortable going places on my own
	I can do most things on my own
	People give me a chance to do things on my own
	People overprotect me because of my eyesight
	I am given the freedom to do things on my own
	I am comfortable asking for help
	I have enough time to myself
	I feel frustrated because of my eyesight
	I feel lonely because of my eyesight
	I worry what other people think about my eyes
	I am positive about the future
	I am confident I will be able to look after myself when I'm older
	I worry my eyesight will get worse
	I like to have a go at everything, despite my eyesight
	I can get around on my own
	I like being at school

	I have to work harder at school because of my eyesight
	I am happy with my social life
Existing un-coded items (*n* = 5)	I spend enough time with my friends
	I cope well with my eyesight problems
	I am treated fairly
	I feel confident
	My family encourage me
Foundation items excluded from the later stages of development (*n* = 10)	I get picked on because of my eyesight
	I feel that having extra help at school is embarrassing
	I prefer being with other children who have eyesight problems
	I have friends who don't have eyesight problems
	I have got used to living with my eyesight
	I do not let my eyesight stand in my way
	I worry about what job I will be able to do in the future
	I have plans for the future
	I find going to the eye clinic helpful
	I have a say in what happens to me at the eye clinic
New age-appropriate codes (*n* = 12)	Strangers/public perceptions and other people understanding
	Resilience, having a go despite likelihood of failing
	Embarrassment in public
	Feeling more tired than others
	Dealing with limited choices and restrictions
	Having to take extra time and care
	Accepting, adapting, adjusting, getting used to the impact of VI
	Denial, coming to terms with a diagnosis
	Helplessness/vulnerability
	Guilt
	Pride, overcoming difficulties, succeeding, perseverance
	Missing school/disruption to routine because of eye clinic

Notably, not all of the items included in the foundation VQoL instrument were assigned codes. Despite this, they were entered into the first draft of the new, age-appropriate instruments. This was due to relevant literature and professional opinion indicating that they will likely be relevant items, despite not emerging as highly important for YP during interviews.

6.3.1.1 Outcomes from the expert consensus meeting concerning the VQoL instrument development

The first major outcome of the expert consensus meeting was the decision to modify the age-boundary for the new suite of instruments designed for older adolescents. This is described in more detail in Section 4.6.4.2 (pg. 101).

Following this decision, all items shown in Table 14 (pg. 170) were discussed for suitability and inclusion in the second draft instruments. Minor modifications were made to items to reflect the linguistic preferences of YP who took part in the first phase of instrument development. The term ‘eyes’ as used in the VQoL items ‘My friends understand about my eyes’ and ‘I worry what other people think about my eyes’ was replaced with the term ‘eyesight’ and ‘children’ as used in VQoL items ‘I feel different from other children’ and ‘Other children pick on me because of my eyesight’ was replaced with the term ‘young people’. Other linguistic modifications were made to reflect the content of in-depth interviews (Table 15, pg. 173). For example, the words ‘at school’ in the item ‘My friends help me at school’ were replaced with ‘when I need it’ to reflect times when YP described receiving help from friends outside of school, for example when shopping or visiting the cinema. The majority of linguistic modifications made at this stage reflected the increased maturity of YP when compared to younger children.

One new VQoL item (I feel tired because of my eyesight) was developed during the expert consensus meeting in light of codes representing the physical impact of visual

impairment, triggering achy and sensitive physical sensations. YP often described these types of feelings as fatigue, for example,

"…it's tiring at the end of the day. [I get] so tired." (Female, 17 years, VI, early onset).

Although participants were probed about their experience of clinical care, and codes relating to this experience were developed, items referencing eye clinics or hospital visits were excluded from the second draft instruments based on the premise that these items measure the experience of healthcare as opposed to VQoL or FV, and are therefore better suited for a PREM (see Section 2.3.1, pg. 31).

Table 15. Minor linguistic modifications made to VQoL items during the expert consensus meeting.

Original VQoL item	Modified VQoL item
My friends help me at school	My friends help me when I need it
In spite of my eyesight, I am independent	I am independent
People give me a chance to do things on my own	People give me the opportunity to do things on my own
I worry about what other people think about my eyes	I worry what other people think of me because of my eyesight
I like to have a go at everything, despite my eyesight	I like to have a go at everything
I like being at school	I enjoy school/college

6.3.2 Phase 2: Pre-testing

6.3.2.1 Self-reported feasibility of the VQoL instrument

The majority of participants described the process of VQoL questionnaire completion as 'easy' or 'very easy'. Some participants expressed concerns that the draft instrument took too long to complete, however it was unclear in several circumstances whether this was due to the lengthy expert consultation, and participants' misunderstanding of how they would be expected to complete the instruments in

clinical practice. When probed about the context in which they would prefer to complete the instruments, several participants described preference of completing the VQoL instrument when waiting for their hospital appointments:

> *"There are a lot of questions but I always have to wait a long time before clinics so I have time to do it then." (Female, 18 years, VI, early onset).*

In each of these circumstances, efforts were made to ensure participants understood the nature of the expert consultation, that a phase of item-reduction was due to take place following pre-testing, and that when used in practice, patients would not necessarily be required to justify their answers in the same way as they were during expert consultations. Once this had been clarified, most participants agreed that the instrument was both easy and feasible for completion in clinical contexts.

6.3.2.2 VQoL item modification

Qualitative feedback on individual items ensured participants interpreted each item correctly in relation to their visual impairment. Most participants took the opportunity to elaborate upon each item, relating it to aspects of everyday life and thus confirming content validity. The majority of items were endorsed as meaningful to participants, with participants giving explanations of why they had chosen their answer and factors which influenced their response.

6.3.2.2.1 Linguistic modifications

The majority of modified items shown in Table 16 (pg. 176) reflect the preferred linguistic terminology used by participants during the pre-testing consultations, providing an extension and validating the linguistic changes made as a result of the expert consensus meeting conducted in phase 1 (see Section 6.3.1.1, pg. 172). The term 'tutor' was added in a number of items to reflect participants' labelling of teachers at college. The term 'opportunity' was changed to 'chance' because a number of participants needed help interpreting the meaning of the word 'opportunity' in this context.

6.3.2.2.2 Modifications to aid interpretation and ease of responding

Whilst items were judged as meaningful and relevant to the everyday lives of participants, many YP had difficulty summarising their daily challenges by picking one response option. Frequently, participants asked if they were allowed to choose two response options (e.g. 'A little bit true' and 'Mostly true'), or the mid-point between two response options.

When probed as to why they felt they were unable to choose one response option, participants often indicated that their answers were context and situation dependent, varying in relation to their immediate environment and help provided by other people. For example, when asked about the item 'I have to work harder at school because of my eyesight', one participant responded:

> *"Obviously to a certain degree, yes. It takes me longer. I do extra at home but I like to do extra. It also depends on the subject and place." (Male, 16 years, VI, early onset).*

As a result of these difficulties, a number of items were modified to ensure YP are capable of answering each item using a single response option, whilst at the same time, allowing for some degree of flexibility and application to multiple contexts and settings. This was done in some cases by reducing the breadth of the item e.g. by adding 'by my friends' in the VQoL item 'I am treated fairly'.

One item (I am independent) was modified and separated into two items to account for the different contexts in which participants described their answers (Table 16, pg. 176).

Table 16. Items which were modified as a result of phase 2: pre-testing.

Original YP-VQoL item	Modified YP-VQoL item
My teachers understand how things are for me because of my eyesight	My teachers and tutors understand how things are for me because of my eyesight
I am independent	I am independent at home
	I am independent at school
I can do most things on my own	I can do most activities on my own
People give me the opportunity to do things on my own	People give me the chance to do things on my own
I have enough time to myself	I have enough private time to myself
I am treated fairly	I am treated fairly by my friends
I am confident I will be able to look after myself when I'm older	I am confident I will be able to look after myself in the future

A second theme arising from pre-testing the VQoL instrument was the extent to which participants required reminding that items should be answered in relation to their visual impairment. For example, when asked to justify her answer to the item 'I enjoy school/college', one participant responded:

> *"I hate school. Always hated the people at school but this is not to do with my eyesight." (Female, 15 years, VI, early onset).*

As a result of this difficulty, the phrase 'Remember to say how things are for you **because of your eyesight** and how much they are **true about you**' was added as a header on each page of the VQoL instrument.

6.3.2.2.3 Further item development

Notably, one new VQoL item was added to the item pool during phase 2. This item (I keep new friends easily) was developed initially in light of in-depth interviews with children (aged 6-9 years) which were running in parallel to the interviews with YP. However, this item was also deemed applicable to YP (aged 16-19 years) and suitable

for detecting the difficulties YP discussed when maintaining romantic relationships (see Table 13, pg. 158). Having a boy/girlfriend was discussed as a normative aspect of growing up (see Section 5.2.2.2, pg. 130), however, YP described the difficulties they had when maintaining these relationships in light of deteriorating visual function. When probed about the relevance of this item during phase 2, the majority of participants agreed it should be added to the item pool, and interpreted it with relevance to visual impairment.

6.3.2.3 Changes to response options

During phase 2, a number of participants demonstrated difficulty interpreting and applying the third response option in the VQoL instrument: 'quite a bit true'. This was particularly difficult for some participants to interpret when answering items which had been reverse worded (e.g. Other young people my age pick on me because of my eyesight). When choosing option 3, one participant frequently re-phrased the response option to "mostly true" (Female, 18 years, VI, early onset). The modified wording of this response option was tested with a number participants during phase 2 and deemed an acceptable replacement.

6.3.2.4 Outcomes from the expert consensus meeting concerning the VQoL instrument development

Experts who took part in the consensus meeting confirmed all modifications to existing items, and the development of new items made in phase 2.

During the consensus meeting, changes to the response options of each instrument were discussed, including the modification of the VQoL response option.

6.3.3. Phase 3: Piloting

6.3.3.1 Qualitative feedback

Of the 23 participants, 10 (43.48%) provided qualitative feedback on the instruments using the blank page which was inserted at the end of the questionnaire booklet. Most

of them took the opportunity to describe their experience of living with visual impairment. For example, one participant described their experience of mainstream and specialised-VI school, offering advice for other YP living with visual impairment:

> *"I think it is important to note that my experiences in a school which is specially designed for the blind are significantly better than my experience in mainstream schools due to the far higher understanding of my disabilities and visual impairment. I think blind children would gain much more confidence, teaching and support if they went to a school like mine because in mainstream schools there is simply not enough resources to build these sufficiently." (Male, 14 years, SVI/BL, early onset).*

6.3.3.2 Quantitative feedback

6.3.3.2.1 Missing data

The 'true' missing data (i.e. no response given) per person for the YP-VQoL instrument was ≤10.26% and considered missing completely at random.[378] Similarly, the 'true' missing data per item was ≤13.04% with three responses missing on the VQoL items 'I am independent at home' and 'I am independent at school/college', which were missing at random. This was a result of a modification made to specify context in the original item 'I am independent' after three questionnaire booklets had been sent out.

6.3.3.2.2 Use of response categories

Participants endorsed the response options well, with the majority opting for a single response option to describe their experience of visual impairment. However, one participant demonstrated difficulty opting for a single response option to describe their experience and, on 4 occasions provided the response option 'between 2 and 3' (accounting for 10.26% of this participants' total VQoL responses), and demonstrated difficulty conceptualising the difference between 'A little bit true' and 'Mostly true'. From this point onward, phase 3 data analysis was conducted with these responses coded as missing data. No further analyses were conducted to impute missing data. No participants were removed because of missing data.

5.3.3.2.3 Feasibility

For the VQoL instrument, 17 YP (73.91% of respondents) reported the time it had taken them to complete the instrument. This ranged from 3-60 minutes (Median = 10, IQR = 23.75).

When asked how easy it was for them to complete, 95% of YP rated completion of the VQoL instrument as ‘easy’ or ‘very easy’ and 100% rated the instructions as ‘easy’ or ‘very easy’ to understand.

6.3.3.3 Response distribution in the VQoL instrument

6.3.3.3.1 Skewness and kurtosis in the VQoL instrument

Seven VQoL items were flagged as problematic due to z-score for skewness and/or kurtosis outside the acceptable limits (-2.00 to +2.00).[313] All of these items originated from the early stages of the foundation research (I have got some good friends, Other young people my age pick on me because of my eyesight, I feel left out because of my eyesight, I get along with my family, I have enough private time to myself, I feel lonely because of my eyesight, I like to have a go at everything). One item (Other young people my age pick on me because of my eyesight) was included in the draft instrument having been excluded in the later stages of the foundation research.

6.3.3.3.2 Floor and ceiling effects in the VQoL instrument

When analysed for floor and ceiling effects by calculating the percent of participants who endorsed the extreme categories (i.e. option 1 or 4), 3 VQoL items were flagged as problematic with >60% of participants choosing response option 4 (Completely true). Each item had been flagged as problematic due to high skewness and originated from the foundation research (I have got some good friends, I get along with my family, I have enough private time to myself). No participants endorsed response category 4 when answering one VQoL item (I feel lonely because of my eyesight) (see Figure 3, pg. 181).

One VQoL item was flagged as problematic due to floor effects (i.e. >60% of respondents choosing option 1 (Not at all true)). This item originated from the foundation research (I feel lonely because of my eyesight). No participants endorsed response category 1 when answering 12 VQoL items (I have got some good friends, I can stand up for myself if someone picks on me, My friends help me when I need it, My teachers and tutors understand how things are for me because of my eyesight, I get along with my family, I am independent at home, I am independent at school/college, I have enough private time to myself, I cope well with my eyesight problems, I am treated fairly by my friends, I am positive about the future, I can get around on my own).

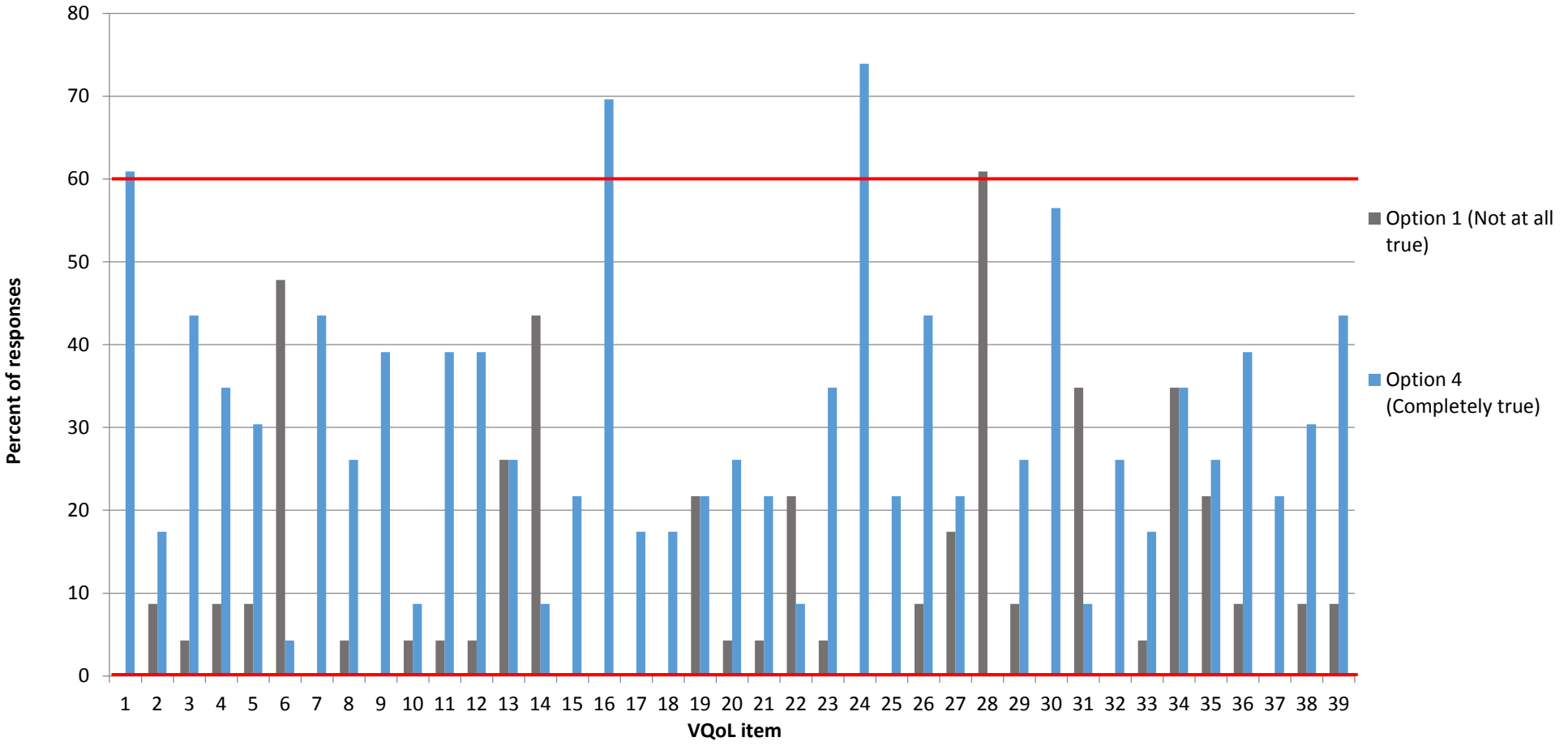


Figure 3. Percent of YP choosing extreme response categories when answering items in the VQoL instrument (1% and 60% thresholds highlighted in red).

Because the aim of phase 3 was to identify any immediate problems with the instruments in preparation for the larger scale postal survey conducted in phase 4, and because of the small sample recruited, no items were removed as a result of response distribution.

6.3.4 Phase 4: Validation

Table 17 (pg. 183) shows the VQoL items entered into the questionnaire booklets which were sent to children (aged 8-12 years) and YP (aged 13-17 years) in the final validation phase. Each item was assigned a value corresponding to the order of presentation within each instrument. Items presented in the same row of Table 17 are overlapping or ‘core’ items which are present in instruments designed for both age groups. Items parallel to empty cells are age-specific items, presented to only one age group.

Table 17. Items included in the Child (8-12 years) and YP (13-17 years) version of the VQoL instruments entered into phase 4 (psychometric validation).

VQoL instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
1	I have got some good friends	1	I have got some good friends
2	I make new friends easily	2	I make new friends easily
3	I keep friends easily	3	I keep friends easily
4	I am happy with how many friends I have	4	I am happy with my social life
5	I spend enough time with my friends	5	I spend enough time with my friends
6	Other children pick on me because of my eyesight	6	Other young people my age pick on me because of my eyesight
7	I can stand up for myself if someone picks on me	7	I can stand up for myself if someone picks on me
8	My friends understand how things are for me because of my eyesight	8	My friends understand how things are for me because of my eyesight
9	My friends help me at school	9	My friends help me when I need it
		10	I get treated the same as everyone else
		11	I feel like I fit in

VQoL instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
10	My friends encourage me to join in their activities	12	My friends encourage me to join in their activities
11	I feel different from other children because of my eyesight	13	I feel different from other young people because of my eyesight
12	I feel left out because of my eyesight	14	I feel left out because of my eyesight
13	My teachers understand how things are for me because of my eyesight	15	My teachers and tutors understand how things are for me because of my eyesight
14	I get along with my family	16	I get along with my family
15	I can decide things for myself		
16	I am independent at home	17	I am independent at home
17	I am independent at school	18	I am independent at school/college
		19	I am comfortable going places on my own
18	People give me a chance to do things for myself	21	People give me the chance to do things on my own
		22	People overprotect me because of my eyesight
19	I am happy asking for help	23	I am comfortable asking for help
		24	I have enough private time to myself

VQoL instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
20	I cope well with my eyesight problems	25	I cope well with my eyesight problems
21	I feel tired because of my eyesight	26	I feel tired because of my eyesight
22	I feel frustrated because of my eyesight	27	I feel frustrated because of my eyesight
23	I feel lonely because of my eyesight	28	I feel lonely because of my eyesight
24	I feel confident	29	I feel confident
25	Other people are fair to me	30	I am treated fairly by my friends
26	I worry what other people think of me because of my eyesight	31	I worry what other people think of me because of my eyesight
		32	I am positive about the future
		33	I am confident I will be able to look after myself in the future
		34	I worry my eyesight will get worse
		35	I worry about what job I will be able to do in the future
27	I like to have a go at everything, although my eyesight isn't perfect	36	I like to have a go at everything
		37	I can get around on my own
28	I like being at school	38	I enjoy school/college

VQoL instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
29	I have to work harder at school because of my eyesight	39	I have to work harder at school/college because of my eyesight
30	I can do most activities on my own	20	I can do most activities on my own

6.3.4.1 Data verification

The full YP-VQoL and PedsQL dataset was verified by the same researcher (AR) to validate the data entry. A total of 4 incorrect data entries were found out of 4,331 individual entries, constituting 0.09% of the dataset. Each incorrect entry was cross-referenced with the original questionnaire booklets and corrected. Ten percent of this dataset was also verified by a second member of the research team (VT) who found 100% of the data to have been entered correctly.

With regards to the Child-VQoL dataset, 43% of data entries were verified by the same researcher who entered the data (VT). No incorrect data entries were found. Accordingly, 10% of the data entries were verified independently by a second member of the research team (AR): 100% were found to be correct.

6.3.4.2 Missing data

6.3.4.2.1 Missing data per person

With regards to missing data, participants were excluded from further analysis if they had >25% missing data, as follows: three children were excluded, leaving 83 included in the Child-VQoL phase 4 analysis. After excluding participants on the basis of not returning completed consent forms and/or booklets (see Section 6.2, pg. 150), no YP were excluded as a result of missing data. Seventy-one YP were included in the YP-VQoL phase 4 analysis.

6.3.4.2.2 Missing data per VQoL item

Missing data was also calculated per item. After removing participants based on missing data, the largest percent of missing data per item was 6.02% for Child-VQoL Item 30 (I can do most activities on my own). This data was missing at random due to addition of the item into the VQoL instrument in the later stages of phase 4 following the recruitment of 4 participants. With regards to the YP instruments, the largest percent of missing data per YP-VQoL item was 2.82% for the item ‘I am independent at

school/college'. All remaining missing data was considered missing completely at random[378] (see Table 18, below).

Table 18. Missing data per item in the VQoL instrument, separated by age-group.

VQoL instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Missing data, *n* (%)**	**Item no.**	**Missing data, *n* (%)**
30*	5 (6.02)	18	2 (2.82)
21	2 (2.41)	39	1 (1.41)
20	2 (2.41)	33	1 (1.41)
16	2 (2.41)	27	1 (1.41)
13	2 (2.41)	19	1 (1.41)
9	2 (2.41)	14	1 (1.41)
6	2 (2.41)	12	1 (1.41)
3	2 (2.41)	9	1 (1.41)
25	1 (1.2)	6	1 (1.41)
24	1 (1.2)		
22	1 (1.2)		
19	1 (1.2)		
18	1 (1.2)		
17	1 (1.2)		
11	1 (1.2)		
8	1 (1.2)		
7	1 (1.2)		
5	1 (1.2)		
4	1 (1.2)		
2	1 (1.2)		
1	1 (1.2)		

* Items with data missing at random (i.e. not included in some questionnaire booklets) versus missing completely at random (i.e. respondent did not answer).

6.3.4.3 Use of response categories

Similar to phase 3 results, participants appeared to endorse response categories well. However, a number of participants indicated on multiple occasions their response to be in between two categories, or a combination of responses. Multiple response options constituted 0.88% of Child-VQoL, and 0.43% of YP-VQoL data. In these cases, responses were re-coded using the values shown in Table 7 (pg. 113).

6.3.4.4 Response distribution

A substantial number of both child- and YP-VQoL (see Table 19, pg. 190) items were shown to have z-score skewness and/or z-score kurtosis values outside the acceptable range of -2 to +2. These items were flagged as problematic.

The distribution of responses to the child- and YP-VQoL items (see Table 19, pg. 190) demonstrated a number of items with floor/ceiling effects and redundant response categories.

Table 19. Z-score skewness, Z-score kurtosis, floor and ceiling effects for each item in the VQoL instrument.

Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4	Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4
VQoL (Child)					VQoL (YP)				
1	-4.02	1.08	2.4	51.8	1	-3.23	-0.56	1.4	53.5
2	-0.62	-1.93	12	25.3	2	-1.27	-1.65	16.9	22.5
3	-2.33	-0.62	2.4	38.6	3	-2.69	0.58	2.8	36.6
4	-5.01	1.41	6	60.2	4	-3.17	0.04	9.9	36.6
5	-2.8	-0.48	4.8	41	5	-2.14	-1.09	12.7	32.4
6	4.63	1.58	49.4	7.2	6	4.79	2.36	54.9	4.2
7	-1.9	-1.92	10.8	39.8	7	-3.15	-0.25	8.5	42.3
8	-2.86	-0.3	6	38.6	8	-2.28	-1.24	9.9	40.8
9	-2.37	-0.26	4.8	32.5	9	-3.19	-0.55	0	57.7
10	-0.99	-1.68	8.4	27.7	10	-2.26	-0.84	7	38
11	-0.11	-2.57	19.3	28.9	11	-2.67	-1.13	9.9	43.7
12	3.37	-0.24	43.4	8.4	12	-3.19	0.13	5.6	42.3
13	-4.05	0.08	3.6	56.6	13	0.59	-1.93	22.5	18.3
14	-6.79	4.73	0	74.7	14	3.43	0.23	43.7	7
15	-3.92	1.74	2.4	48.2	15	-2.14	-1.22	5.6	42.3
16	-2.65	-0.57	2.4	36.1	16	-5.69	1.03	0	80.3
17	-0.9	-0.78	1.2	25.3	17	-2.13	0.17	2.8	32.4
18	-1.61	-0.81	1.2	33.7	18	-1.54	-0.32	4.2	23.9
19	-3.26	0.33	3.6	43.4	19	-0.52	-2.46	25.4	23.9
20	-2.99	-0.76	1.2	49.4	20	-1.36	-1.41	9.9	28.2
21	0.43	-2.37	22.9	21.7	21	-2.43	0.85	4.2	28.2
22	0.82	-1.99	36.5	13.3	22	-0.83	-1.69	22.5	15.5
23	5.7	3.06	57.8	7.2	23	-1.77	-0.99	7	31
24	-2.68	-0.12	4.8	36.1	24	-3.45	-0.5	0	59.2
25	-1.88	-0.49	3.6	30.1	25	-3.28	0.5	5.6	40.8
26	2.08	-2.12	41	16.9	26	0.86	-2.01	22.5	21.1
27	-2.78	-0.82	0	53	27	1.31	-1.57	11.3	21.1

Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4	Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4
VQoL (Child)					VQoL (YP)				
28	-2.25	-0.5	2.4	37.3	28	6.16	4.67	62	5.6
29	-1.89	-1.85	12	37.3	29	-1.30	-1.55	11.3	26.8
30	-1.86	-0.35	3.6	27.7	30	-4.56	2.51	1.4	57.7
					31	1.13	-2.12	28.2	19.7
					32	-1.9	-1.23	11.3	31
					33	-1.68	-1.53	8.5	35.2
					34	1.24	-2.21	28.2	22.5
					35	0.53	-2.02	22.5	18.3
					36	-2.41	-0.9	8.5	38
					37	-1.15	-1.69	8.5	31
					38	-2.26	-1.09	8.5	38
					39	-1.24	-2.15	12.7	38

*VQoL response option 1 = ‘Not at all true’, response option 4 = ‘Completely true’

The percent of respondents endorsing extreme categories in each item was analysed in parallel to the largest, and most problematic, skew and kurtosis values and the child- and YP-VQoL items shown in Table 20 (below) were removed from the subsequent stages of item reduction.

Table 20. Items excluded as a result of response distribution.

VQoL (child)		VQoL (YP)	
Item no.	**Item**	**Item no.**	**Item**
14	I get along with my family	16	I get along with my family
27	I like to have a go at everything	9	My friends help me when I need it
		24	I have enough private time to myself
		28	I feel lonely because of my eyesight

6.3.4.5 Assessment of unidimensionality

6.3.4.5.1 FA

The remaining items in each VQoL instrument version were entered into a formal exploratory FA to assess for unidimensionality. FA was conducted using each of the 5 datasets with imputed missing data for each instrument, and the outcomes reported are the mean values of each dataset (see Table 21, below).

Notably, the mean number of items loading <0.4 on the first/largest factor in each imputed dataset indicates some items which do not contribute to the latent variable within each instrument. Despite this, all items included in the Child-VQoL, loaded positively onto the first factor.

In the YP-VQoL dataset one item (Item 15 (My teachers and tutors understand how things are for me because of my eyesight)) did not load positively onto the first factor in any of the imputed datasets and was therefore outstanding as a potentially problematic item, as shown in Table 21 (below).

Table 21. Outcome from FA conducted to assess dimensionality in the VQoL instruments.

Instrument version	Eigenvalue for the first (largest) factor (% of variance explained)	Variation between eigenvalues in imputed datasets	No. of eigenvalues >1	No. of items loading onto the first (largest) factor <.4	Items not loading positively onto the first (largest) factor
Child-VQoL	8.19 (29.26)	0.06	8.8	3	None
YP-VQoL	12.26 (35.02)	0.15	8.6	2.4	Item 15 (My teachers and tutors understand how things are for me because of my eyesight)

6.3.4.5.2 Parallel analysis

Consistent with the method used to handle multiple imputations of missing data when performing FA, parallel analysis was run using each of the five imputed datasets. The mean eigenvalues for the first five eigenvalues in each dataset are presented in Table 22 (below).

Table 22. Average of the first five eigenvalues produced by parallel analysis for each VQoL dataset.

Actual eigenvalue	Average eigenvalue	95th Percentile eigenvalue
Child-VQoL		
8.19	2.27	2.47
2.15	2.06	2.2
1.87	1.91	2.03
1.68	1.78	1.88
1.48	1.66	1.76
YP-VQoL		
12.26	2.64	2.88
3.27	2.4	2.56
2.28	2.22	2.36
1.89	2.07	2.19
1.5	1.94	2.05

The matrix produced by parallel analysis can be used to assess dimensionality by judging the magnitude of the actual eigenvalue in relation to the average and 95th percentile eigenvalues. If the actual eigenvalue is found to be greater than both the average and 95th percentile eigenvalues, then the eigenvalue is interpreted as a factor. Thus, only one actual eigenvalue greater than the corresponding average and 95th percentile eigenvalues is thought to represent unidimensionality. In the Child-VQoL dataset the average and percentile eigenvalues were discordant in relation to the actual eigenvalue. In this case, the actual eigenvalue was compared to the percentile eigenvalue only as per recommendations in light of PA's tendency to overestimate the number of factors.[400] Thus, the Child-VQoL dataset was deemed unidimensional.

The YP-VQoL dataset was shown to be multidimensional according to the matrix. The largest actual eigenvalue for this dataset was larger than both the average and percentile eigenvalues however, the second largest actual eigenvalue was also

marginally larger than both alternative eigenvalues (see Table 22, pg. 200). Despite this finding, the YP-VQoL dataset was deemed suitable for Rasch analysis based on evidence suggesting unidimensionality in the corresponding Child-VQoL instrument, and aims to develop the instruments in tandem as complementary measures.

The values produced by the averaged matrix (see Table 22, pg. 194) were used to produce a scree plot for each dataset, confirming the results from the matrix (see Figure 4, pg. 196 and 5, pg. 197).

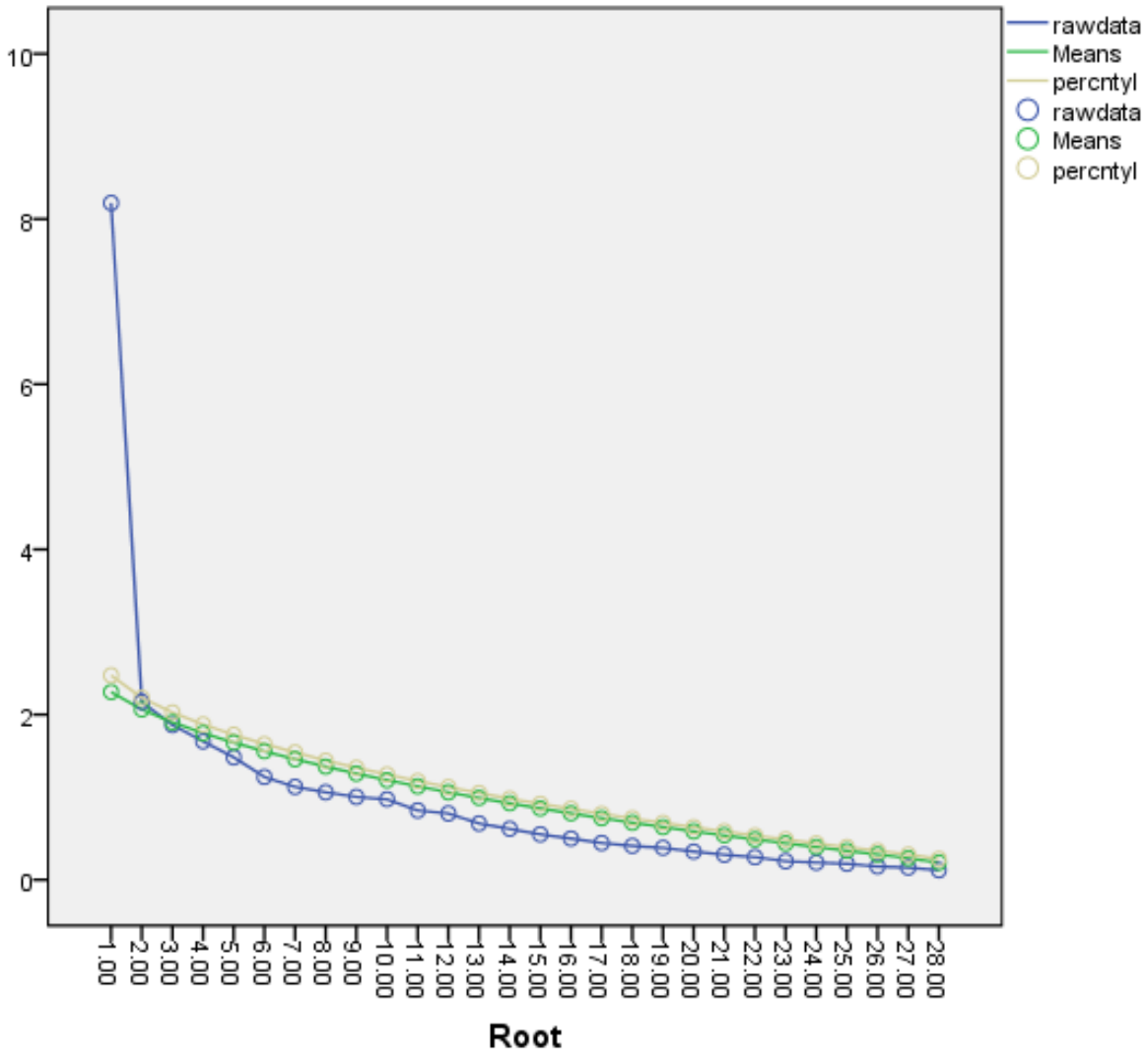


Figure 4. Screeplot showing actual, average and percentile eigenvalues in the Child-VQoL instrument.

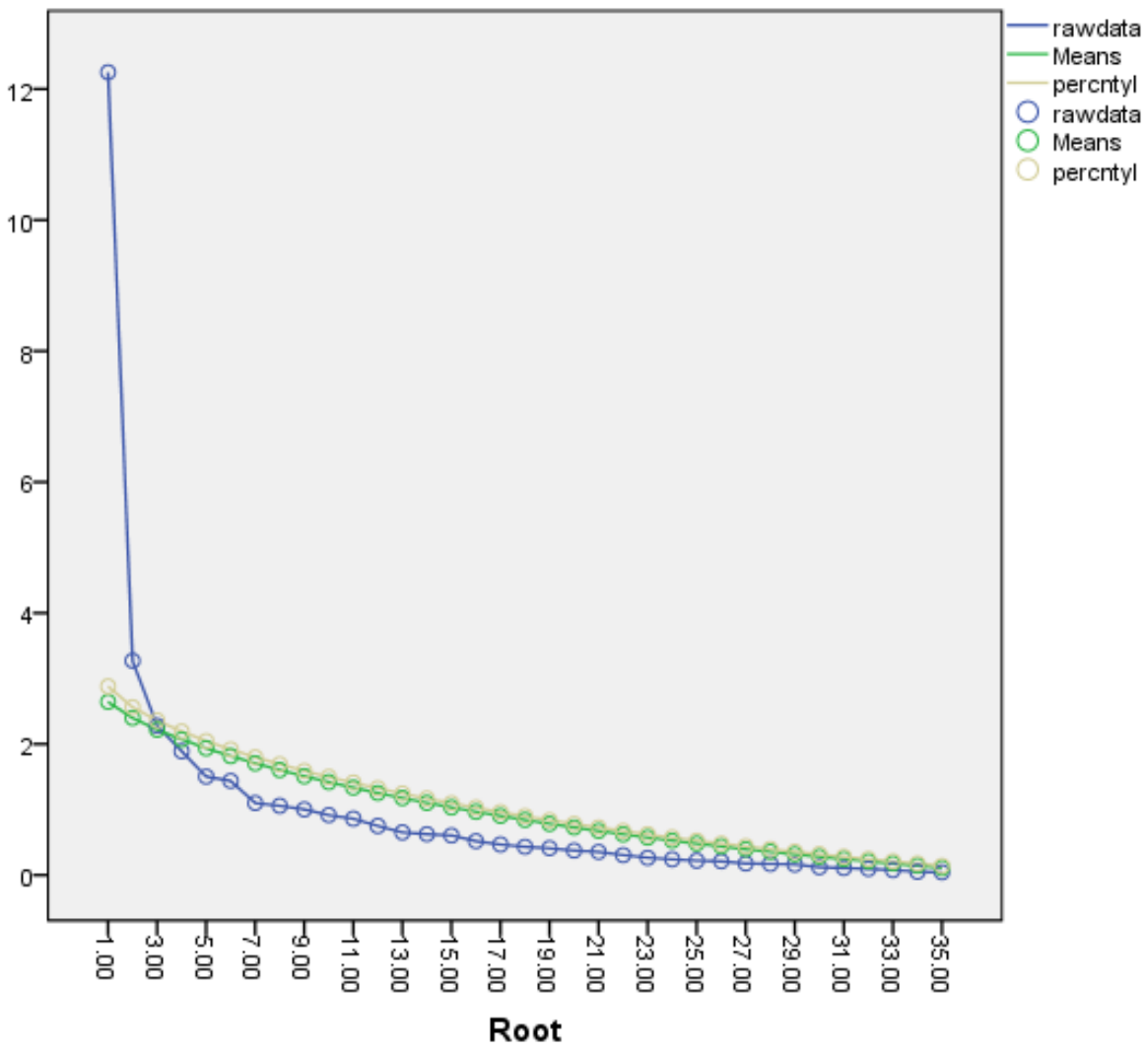


Figure 5. Screeplot showing actual, average and percentile eigenvalues in the YP-VQoL instrument.

6.3.4.6 Rasch analysis

Rasch analysis was conducted using Child- and YP-VQoL datasets independently, and then using combined Child- and YP-VQoL datasets, with the aim of calibrating the instruments. Results are presented in relation to the Child-VQoL, and YP-VQoL instruments separately before combining and calibrating instruments.

6.3.4.7 Rasch analysis using the 28-item Child-VQoL instrument

A total of 4 iterations were required before the Child-VQoL instrument met the functional criteria of Rasch analysis, indicating good measurement capacity as follows:

6.3.4.7.1 Item fit to the Rasch measurement scale

Item 9 was the first item to be removed during this analysis, due to an OUTFIT mean-square value (MNSQ) which was above the threshold of 1.5 (see Table 23, below). Fit statistics for all remaining items fell within the range of 0.5 to 1.5 (see Table 24, pg. 200), demonstrating good fit of the model of measurement to the data.

Table 23. Problematic items in the Child-VQoL instrument due to fit to the Rasch measurement scale.

		INFIT		**OUTFIT**	
Iteration	**Problematic item(s)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**
Iteration 1 (Excluding Item 14 (I get along with my family) and 27 (I like to have a go at everything, although my eyesight isn't perfect))	9 (My friends help me at school)	1.23	1.5	1.72	3.9

6.3.4.7.2 Order of person abilities

Three further items (Item 30 (I can do most activities on my own), 24 (I feel confident) and 23 (I feel lonely because of my eyesight)) emerged as problematic when assessing the observed, sample-dependent, average measure of persons (relative to each item) in this analysis who responded in each category. In each case, the average measure

for a higher score value was lower than for a lower score value. This finding contradicts the assumption that a higher level of VQoL predicts a higher score in the instrument, and therefore refutes the underlying principle of additive measurement in this instrument. As a result, three further iterations were used to assess the measurement properties of the Child-VQoL instrument without these three items. Fit statistics were assessed after the removal of each item, to determine the impact upon the existing good-fitting items.

6.3.4.7.3 Differential item functioning (DIF)

Analysis of DIF by gender and age was performed using participants stratified by age group, using the age categories 'young' (7-9 years) and 'old' (10-13 years). This stratification was necessary to produce two age groups roughly comparable in terms of sample size, and big enough to detect realistic DIF values.

This analysis revealed no DIF contrasts above the threshold of 1.0 (see Table 24, pg. 200). Thus, the final iteration of the Child-VQoL instrument contained 24 items. The measurement properties of the final iteration were checked for conformity to the predefined thresholds, and fit statistics were within acceptable limits (see Table 24, pg. 200). The average measures of persons were ordered in each item in relation to ability.

Table 24. Fit and DIF statistics of items in the final 24-item Child-VQoL scale.

Item code	Child-VQoL item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
1	I have got some good friends	0.84	0.75	-0.39	-0.34
2	I make new friends easily	0.89	0.87	0.23	-0.12
3	I keep friends easily	0.79	0.79	-0.15	-0.05
4	I am happy with how many friends I have	1.29	1.09	0.48	-0.28
5	I spend enough time with my friends	0.90	0.83	0.26	0
6	Other children pick on me because of my eyesight	1.05	1.06	-0.55	0.45
7	I can stand up for myself if someone picks on me	1.23	1.19	0.07	0.27
8	My friends understand how things are for me because of my eyesight	0.98	1.01	-0.25	0.24
10	My friends encourage me to join in their activities	1.22	1.45	-0.06	0.45
11	I feel different from other children because of my eyesight	0.99	1.04	-0.09	0.20
12	I feel left out because of my eyesight	0.71	0.66	0.02	0.15
13	My teachers understand how things are for me because of my eyesight	1.31	1.32	0.41	-0.21
15	I can decide things for myself	1.01	1.03	-0.80	0.28
16	I am independent at home	0.94	0.97	-0.19	0.21
17	I am independent at school	0.94	0.98	-0.05	0.31
18	People give me a chance to do things for myself	0.76	0.73	-0.26	-0.03

Item code	Child-VQoL item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
19	I am happy asking for help	1.08	0.98	0.47	-0.05
20	I cope well with my eyesight problems	0.99	0.93	0.66	-0.67
21	I feel tired because of my eyesight	1.35	1.41	-0.39	-0.56
22	I feel frustrated because of my eyesight	1	1.10	0	-0.06
25	Other people are fair to me	0.65	0.62	-0.09	0.07
26	I worry about what other people think about me because of my eyesight	1.25	1.36	0.51	0.21
28	I like being at school	0.98	0.93	0.27	-0.31
29	I have to work harder at school because of my eyesight	1.14	1.17	0	-0.31

6.3.4.7.4 Targeting of items to persons

The item-person map revealed good targeting of the 24 items in the scale to the responders. The difference between the item and person means were .91 which is within the acceptable limit of 1 (see Figure 6, pg. 202). The person separation and reliability for this iteration were analysed using the real root-mean-square standard error (RMSE) as an indicator of the worst case scenario but were within the acceptable limits (separation = 3.46, reliability = .92), indicating high measurement precision of the Child-VQoL instrument.

Figure 6. Item-person targeting of the final 24-item Child-VQoL scale.

6.3.4.7.5 Response category function

Item category probability plots produced using Winsteps were assessed individually for both the forward and reverse worded items in the 24-item scale and the step calibrations were used to complement findings. The category probability plots for the forward worded items were well ordered and Andrich thresholds increased by at least

1.13 logits across the scale, which is marginally less that the recommended increase of 1.4, showing that despite coherent order of the response categories, the 4 categories may not be optimally defined.

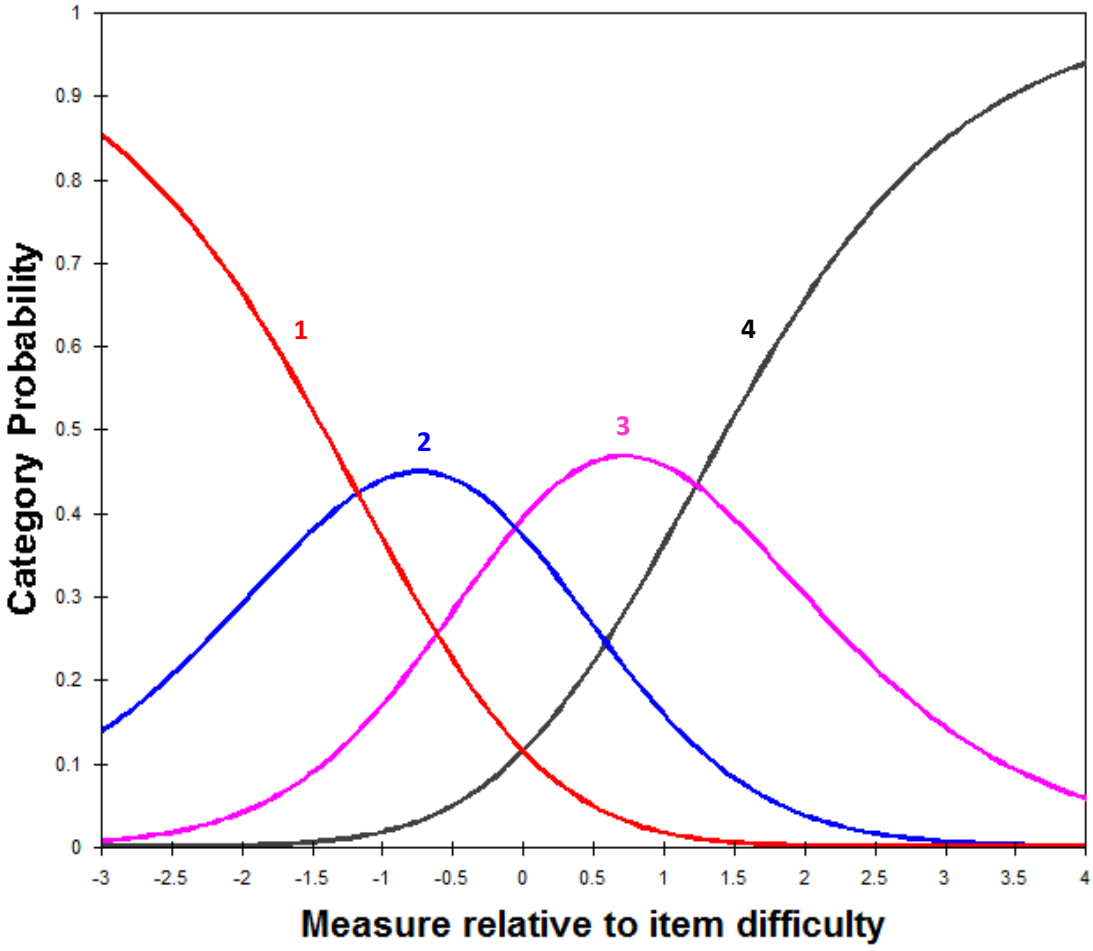


Figure 7. Category probability curves for the 4 response categories in the forward-scored Child-VQoL instrument (1 = Not at all true, 2 = A little bit true, 3 = Mostly true, 4 = Completely true).

When analysed in relation to the reverse worded items, the category probability plots were also well ordered. However, response category 3 (Mostly true) was the least likely response category to be observed, with the majority of the curve remaining submerged beneath the curve for response option 4 (Completely true). Despite the small likelihood of respondents choosing response option 3, the increase in Andrich threshold was .16 between response option 3 and 2, indicating that the response categories are ordered

as expected. The increase in Andrich threshold between response categories 1 and 2 was .45, and between 3 and 4 was 1.03.

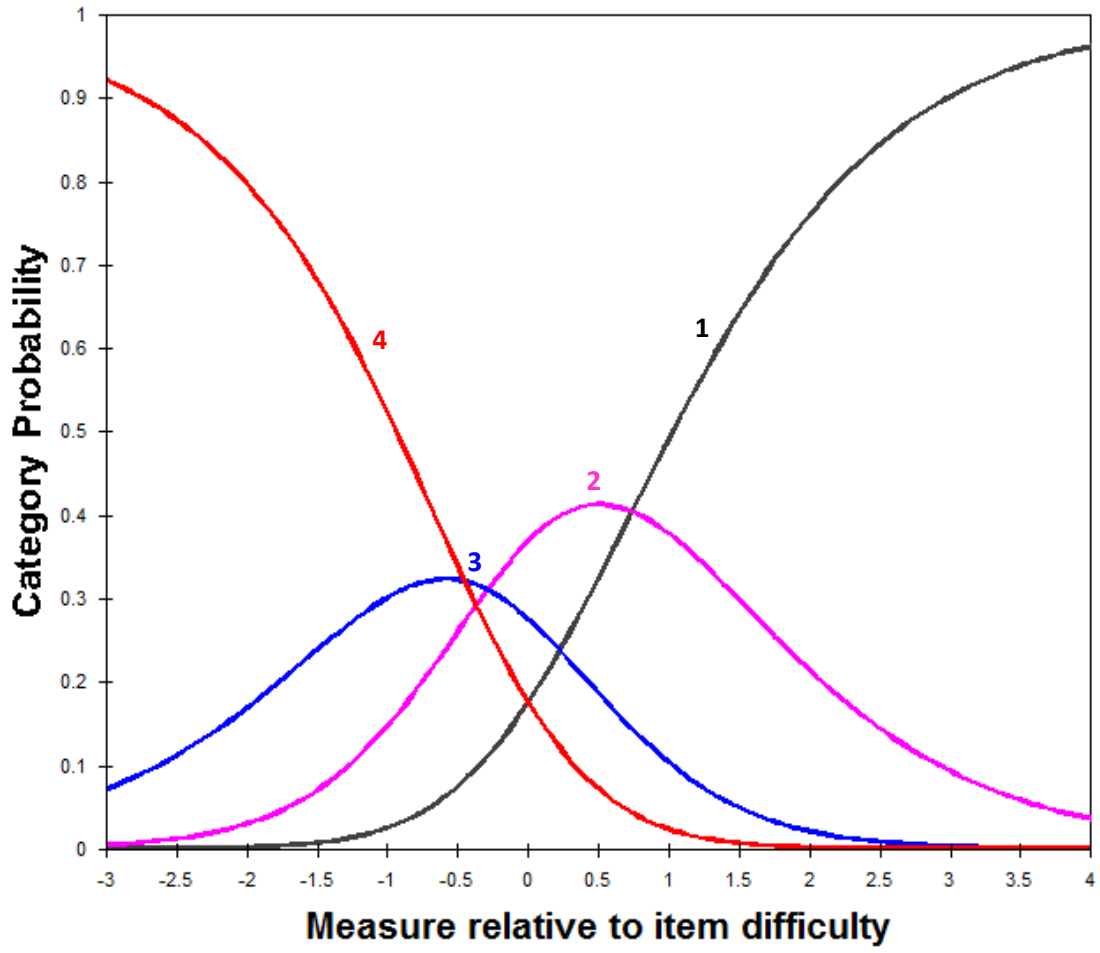


Figure 8. Category probability curves for the 4 response categories in the reverse-scored Child-VQoL items (1 = Not at all true, 2 = A little bit true, 3 = Mostly true, 4 = Completely true).

6.3.4.8 Rasch analysis using the 35-item YP-VQoL instrument

After removing 4 items as a result of preliminary item removal, rendering a total of 35 items in the YP-VQoL instrument, 8 iterations were required before the items conformed to the criteria of Rasch analysis. During these iterations, items were removed sequentially. One iteration (iteration 4) was performed with one previously removed item (Item 22 (People overprotect me because of my eyesight)) retained. This was with the aim of comparing outcomes with and without Items 22 and 26 (I feel tired

because of my eyesight) which were both shown to violate the criteria of good fit to the model.

6.3.4.8.1 Item fit to the Rasch measurement scale

The first items which were removed from the YP-VQoL instrument were Items 15 (My teachers and tutors understand how things are for me because of my eyesight), 22 and 26. These items were removed as a result of large OUTFIT MNSQ statistics. Table 25 (pg. 206) shows the INFIT and OUTFIT statistics for problematic items during each of the early iterations (iterations 1-5). After removing Items 15, 22 and 26, a decision was made to stop removing items based purely upon FIT statistics, as the remaining FIT statistics deviated only by a small percent outside the threshold of 0.5 – 1.5.

Table 25. Problematic items in the YP-VQoL instrument due to fit to the Rasch measurement scale.

		INFIT		OUTFIT	
Iteration	**Problematic item(s)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**	**Mean-square vale (MNSQ)**	**Z-standardised (ZSTD)**
Iteration 1 (Excluding Items 9 (My friends help me when I need it), 16 (I get along with my family), 24 (I have enough private time to myself) and 28 (I feel lonely because of my eyesight))	15 (My teachers and tutors understand how things are for me because of my eyesight)	1.87	4.3	2.26	5.2
	22 (People overprotect me because of my eyesight)	1.36	2.3	2.1	5.1
	26 (I feel tired because of my eyesight)	1.51	3	1.59	3.1
Iteration 2 (Excluding Items 9, 16, 24, 28 and 15)	22 (see above)	1.41	2.5	2.55	6.5
	26 (see above)	1.55	3.2	1.63	3.2
Iteration 3 (Excluding Items 9, 16, 24, 28, 15 and 22)	26 (see above)	1.58	3.3	1.67	3.2
Iteration 4 (Excluding Items 9, 16, 24, 28, 15 and 26)	22 (see above)	1.44	2.6	2.57	6.6
	8 (My friends understand how things are for me	1.29	1.7	1.56	2.7

		INFIT		OUTFIT	
Iteration	**Problematic item(s)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**	**Mean-square vale (MNSQ)**	**Z-standardised (ZSTD)**
	Iteration 4 (Excluding Items 9, 16, 24, 28, 15 and 26 continued… because of my eyesight)				
	34 (I worry about what job I will be able to do in the future)	1.53	3	1.5	2.6
Iteration 5 (Excluding Items 9, 16, 24, 28, 15, 26 and 22)	34 (see above)	1.54	3.1	1.5	2.5
	8 (see above)	1.28	1.6	1.53	2.5

6.3.4.8.2 Order of person abilities

After removing these three items based on FIT statistics, Items 30 (I am treated fairly by my friends), 21 (People give me the chance to do things on my own) and 12 (My friends encourage me to join in their activities) were flagged as problematic and removed due to disordering of the average measures given to participants in relation to participants' average ability. These items were removed iteratively whilst monitoring the impact of removal upon the fit statistics of the remaining items.

6.3.4.8.3 Differential item functioning

The remaining 29 items were entered into a DIF analysis by gender and age.

DIF analysis by gender indicated a moderate to large DIF effect for one item (Item 6 (Other young people my age pick on me because of my eyesight)) for which males

were 1.07 logits less able to endorse as true in this sample and 2 items (Item 19 (I am comfortable going places on my own) and 34 (I worry my eyesight will get worse)) for which females were over 1 logit less able to endorse as true than males.

Before removing any items based on DIF by gender, DIF analysis by age was run using participants stratified to young (13-15 years) and old (16-18 years) age groups. Like the stratification of children in Section 6.3.4.7.3 (pg. 199), this stratification was needed to ensure age-groups were large enough to be compared and produce realistic DIF statistics. The cut off of 15 years was chosen based partly on the size of participants within each age group and producing two groups which are roughly equal in size, and also on the empirical findings presented in Chapter 5 (pg. 125) with regard to changes which may occur at the age of 16 years. DIF analysis by age group indicated one item (Item 7 (I can stand up for myself if someone picks on me)) which was 1.15 logits more difficult for participants in the younger age group to endorse as true.

Three problematic items highlighted by DIF analysis (Item 19, 34 and 7) were removed iteratively and in order of magnitude of DIF contrast (ranging from largest to smallest). FIT statistics were checked after removing each item and DIF analyses were re-run using each iteration. After removing Items 19, 34 and 7, the DIF contrast belonging to Item 6 was reduced to below the threshold of 1 logit. As a result, Item 6 was retained. When checking for DIF by gender using the final iteration, one further item (Item 37 (I can get around on my own)) was flagged as problematic with a DIF contrast of 1.03, indicating greater difficulty for females in this sample. However, this DIF contrast was judged as negligible, and the item was retained in light of preserving the good fit statistics of the remaining items.

The final iteration of the YP-VQoL instrument contained 26 items. The FIT statistics and DIF contrasts of the items included in the final iteration are displayed in Table 26 (pg. 210). Notably, two items (Item 8 (My friends understand how things are for me because of my eyesight) and 39 (I have to work harder at school/college because of

my eyesight)) have been retained despite a small divergence in FIT statistic from the threshold of 0.5-1.5. The suitability for inclusion of these two items, despite high OUTFIT statistics was verified by two further iterations in which both items were removed, and greater fit statistics were produced by the remaining items

Table 26. Fit and DIF statistics of items in the final 26-item YP-VQoL scale.

Item code	YP-VQoL item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
1	I have got some good friends	0.85	0.88	0.09	-0.32
2	I make new friends easily	0.93	0.88	-0.70	0.48
3	I keep friends easily	0.77	0.86	0.03	0.19
4	I am happy with my social life	0.87	0.8	0.45	-0.29
5	I spend enough time with my friends	1.13	1.05	0.49	0.14
6	Other young people my age pick on me because of my eyesight	1.35	1.17	-0.64	-0.99
8	My friends understand how things are for me because of my eyesight	1.33	1.52	-0.38	-0.11
10	I get treated the same as everyone else	1.11	1.08	-0.54	-0.12
11	I feel like I fit in	1.02	0.9	0.10	-0.40
13	I feel different from other young people because of my eyesight	0.84	0.84	0.28	-0.53
14	I feel left out because of my eyesight	0.91	0.78	0.34	-0.31
17	I am independent at home	1.02	1.03	-0.07	-0.20
18	I am independent at school/college	0.74	0.74	0.09	0.19
20	I can do most activities on my own	0.94	0.88	-0.15	0.51
23	I am comfortable asking for help	1.04	1.05	0.08	0.18
25	I cope well with my eyesight problems	0.86	0.78	-0.13	-0.23

Item code	YP-VQoL item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
27	I feel frustrated because of my eyesight	1.21	1.31	0	-0.08
29	I feel confident	0.78	0.77	0.03	0.46
31	I worry what other people think of me because of my eyesight	1.02	0.89	-0.21	0.21
32	I am positive about the future	0.90	0.91	0	-0.12
33	I am confident I will be able to look after myself in the future	0.90	0.79	0	0
35	I worry about what job I will be able to do in the future	0.84	0.82	0	0.46
36	I like to have a go at everything	0.95	0.88	0.47	-0.28
37	I can get around on my own	1.11	1.06	-0.06	1.03
38	I enjoy school/college	1.18	1.36	0.08	-0.66
39	I have to work harder at school/college because of my eyesight	1.4	1.52	0.15	0.15

6.3.4.8.4 Targeting

The person-item map using the items included in the final iteration revealed good targeting of items to the abilities of participants, with a difference of .71 between the mean of participants' ability and the mean difficulty of the remaining items. Additionally, the person separation and reliability for this iteration were within acceptable limits (separation = 2.74, reliability = .88) when analysing the real RMSE. As expected, the RMSE produced by the model, which represents the reliability measures when assuming all misfit in the data is random, were slightly improved (separation = 2.86, reliability = .89).

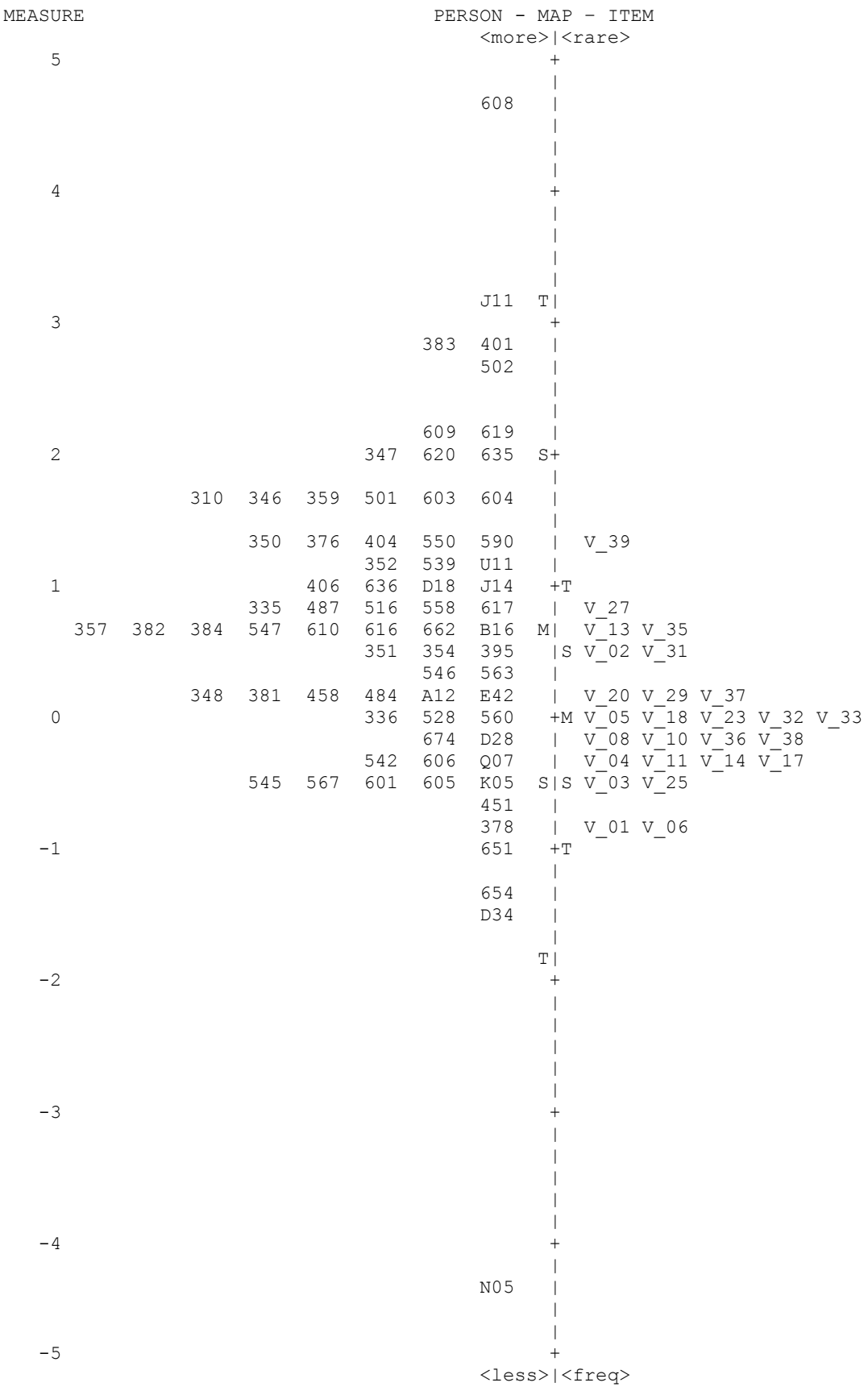


Figure 9. Item-person targeting of the final 26-item YP-VQoL scale.

6.3.4.8.5 Response category function

The Andrich thresholds for the category probability plots for the forward worded items in the final 26-item YP-VQoL scale increased by at least 0.96 logits across the scale (see Figure 10, below), which is marginally less than the increase displayed in the Child-VQoL instrument, and the recommended increase of 1.4. This indicates that the response categories in this measure may not be optimally defined. Despite this, the categories are well-ordered, demonstrating strong potential for accurate measurement.

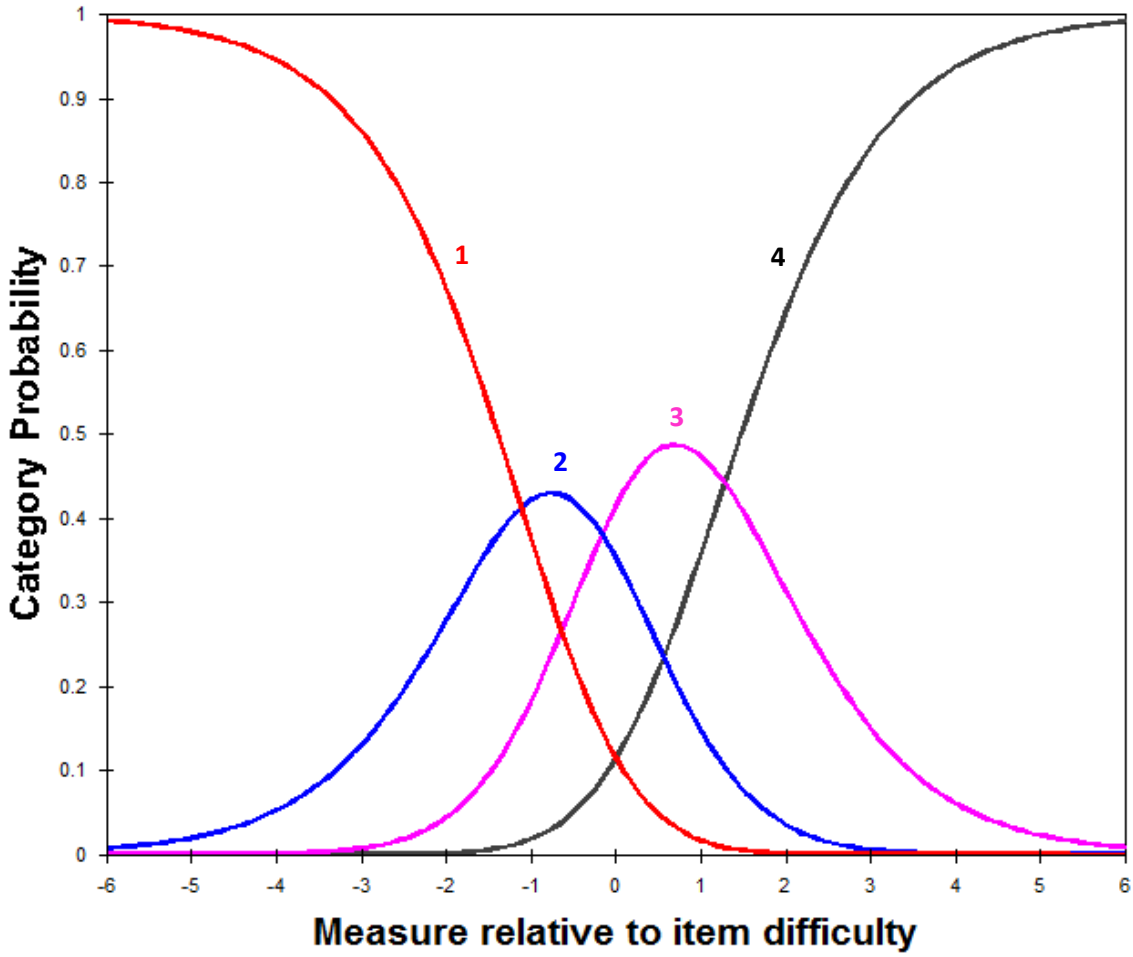


Figure 10. Category probability curves for the 4 response categories in the forward-scored YP-VQoL instrument (1 = Not at all true, 2 = A little bit true, 3 = Mostly true, 4 = Completely true).

As with the Child-VQoL instrument, when analysed in relation to the reverse worded items, the category probability plots were well ordered. However, response category 3 (Mostly true) was again, the least likely response category to be observed, with the majority of the curve remaining submerged beneath the curve for response option 4 (Completely true). The increase in Andrich threshold was .26 between response option 3 and 2, indicating that the response categories are ordered as expected. The increase in Andrich threshold between response categories 1 and 2 was .71, and between 3 and 4 was 1.61 (see Figure 11, below).

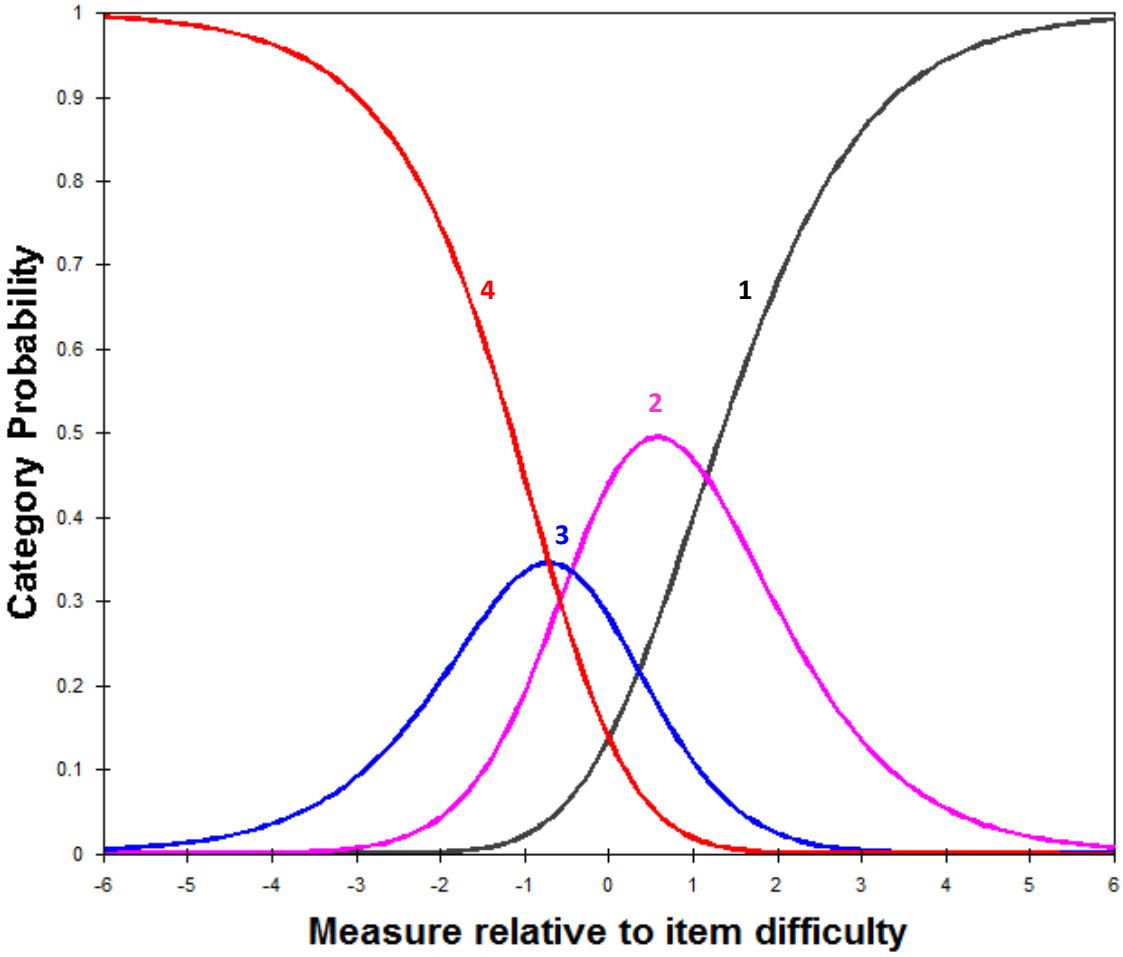


Figure 11. Category probability curves for the 4 response categories in the reverse-scored YP-VQoL instrument (1 = Not at all true, 2 = A little bit true, 3 = Mostly true, 4 = Completely true).

6.3.4.9 Calibration of the Child- and YP-VQoL instruments

Prior to further analyses, the items included in the 24-item Child-VQoL and 26-item YP-VQoL instruments were assigned a new item label corresponding to the VQoL instrument in which they appear (be it Child-VQoL, YP-VQoL or both instruments (see Table 27, pg. 216)). These new item labels were used to identify each item during the process of calibration. Items were assigned a person-factor code (C = child, YP = young people). By combining both child- and YP-VQoL datasets, the sample size used in this phases of analysis was increased to 154 participants aged 7-18 years.

Table 27. Items included in the final 24-item Child- and 26-item YP-VQoL instruments and labels used for calibration**.**

Child		**YP**		**Item label for calibration**
Original item no.	**Label**	**Original item no.**	**Label**	
1	I have got some good friends	1	I have got some good friends	1
2	I make new friends easily	2	I make new friends easily	2
3	I keep friends easily	3	I keep friends easily	3
4	I am happy with how many friends I have	4	I am happy with my social life	4
5	I spend enough time with my friends	5	I spend enough time with my friends	5
6	Other children pick on me because of my eyesight	6	Other young people my age pick on me because of my eyesight	6
7	I can stand up for myself if someone picks on me			7C
8	My friends understand how things are for me because of my eyesight	8	My friends understand how things are for me because of my eyesight	8
		10	I get treated the same as everyone else	9YP
		11	I feel like I fit in	10YP
10	My friends encourage me to join in their activities			11C
11	I feel different from other children because of my eyesight	13	I feel different from other young people because of my eyesight	12

Child		YP		Item label for calibration
Original item no.	**Label**	**Original item no.**	**Label**	
12	I feel left out because of my eyesight	14	I feel left out because of my eyesight	13
13	My teachers understand how things are for me because of my eyesight			14C
15	I can decide things for myself			15C
16	I am independent at home	17	I am independent at home	16
17	I am independent at school	18	I am independent at school/college	17
		20	I can do most activities on my own	18YP
18	People give me a chance to do things on my own			19C
19	I am happy asking for help	23	I am comfortable asking for help	20
20	I cope well with my eyesight problems	25	I cope well with my eyesight problems	21
21	I feel tired because of my eyesight			22C
22	I feel frustrated because of my eyesight	27	I feel frustrated because of my eyesight	23
		29	I feel confident	24YP
25	Other people are fair to me			25C
26	I worry what other people think of me because of my eyesight	31	I worry what other people think of me because of my eyesight	26
		32	I am positive about	27YP

Child		YP		Item label for calibration
Original item no.	**Label**	**Original item no.**	**Label**	
			the future	
		33	I am confident I will be able to look after myself in the future	28YP
		35	I worry about what job I will be able to do in the future	29YP
		36	I like to have a go at everything	30YP
		37	I can get around on my own	31YP
28	I like being at school	38	I enjoy school/college	32
29	I have to work harder at school because of my eyesight	39	I have to work harder at school/college because of my eyesight	33

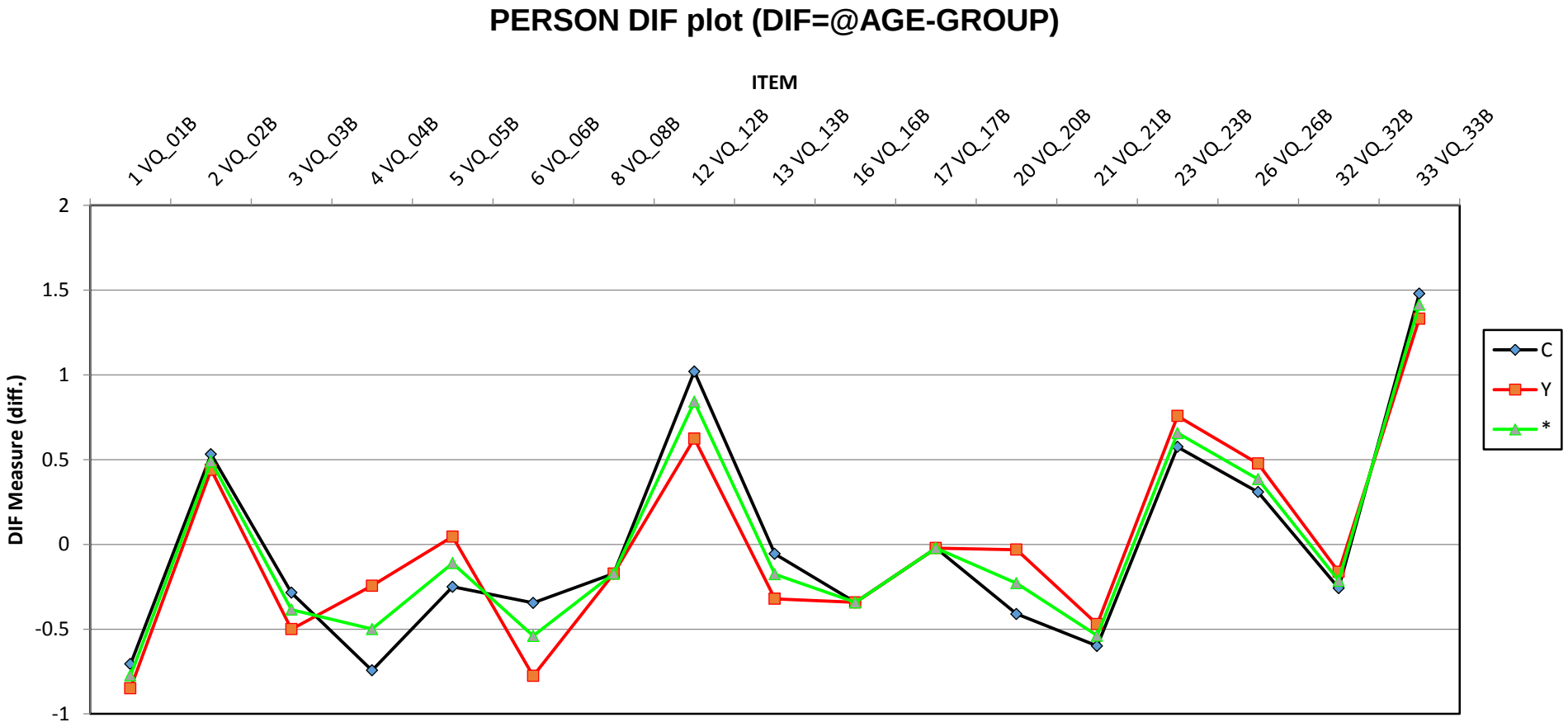
6.3.4.9.1 Item fit to the Rasch measurement model

When analysed as a complete instrument, the overlapping or 'core' Child-VQoL and YP-VQoL items demonstrated good fit with the Rasch measurement model and FIT statistics were all within the range of .62 to 1.48. All observed average measures for persons were ordered in the right direction depending on whether the item was forward or reverse worded.

6.3.4.9.2 Differential item functioning

An analysis was conducted using the overlapping or 'core' items to detect DIF between the Child (8-12 years) and YP (13-17 years) items with a view to detect the stability of the item functioning between instruments and therefore determine whether the core items can be used to fix the scale calibration of the measure in the presence of age-specific items which are presented to only one age-group. Analysis revealed no DIF contrasts greater then 1, indicating that the core items are not biased in any way to

either age group (after adjusting for the overall scores of respondents (see Figure 12, pg. 220). The largest DIF value was observed in Item 4 (I am happy with how many friends I have/I am happy with my social life) which was 0.5 logits more difficult for participants in the older age group (e.g. aged 13-17 years) to endorse as true. This may be a result of the change in wording between the items, with the term ‘social life’ representing complex, broader aspects of QoL than quantity of ‘friends’.



*the baseline measure (no DIF)

Figure 12. DIF in overlapping VQoL items between children (aged 8-12 years) (C) and YP (aged 13-17 years) (Y).

6.3.4.9.3 Fitting FPs to the model

When analysed together, the final 24-item Child-VQoL and 26-item YP-VQoL instrument demonstrated good functional ability, conforming to the numerous criteria of Rasch analysis indicating good fit to the model of measurement, and potential to be used across the full age-range of 8-17 years. The complete score-to-measure tables (see Appendix XVI, pg. 384 and XVII, pg. 385) can be used to convert raw scores on each age-specific instrument into scores which can be compared to converted scores on the opposing instrument, and used to analyse outcomes between children and YP of different age groups with reference to the same measurement scale and latent trait.

The default equations provided by Winsteps software to convert the raw scores into measure (or logit) scores for both Child (f) and YP (g) versions of the VQoL instrument are shown below.

When fitted to FPs, a 4th order polynomial trendline was found to fit the data optimally in both Child (see Figure 13, pg. 222) and YP (see Figure 14, pg. 223) raw scores. The 4 parameters shown in Figures 13 and 14 were entered into the equations presented in Chapter 4, Section 4.9.4.4 (pg. 120) to produce a formula to be used in practice to convert raw scores into logit scores in both the Child and YP instruments (h, i).

$$Child\ VQoL\ Measure = 21.0726 + Score \times .7920 \quad \text{(f)}$$

$$YP\ VQoL\ Measure = 21.5534 + Score \times .7180 \quad \text{(g)}$$

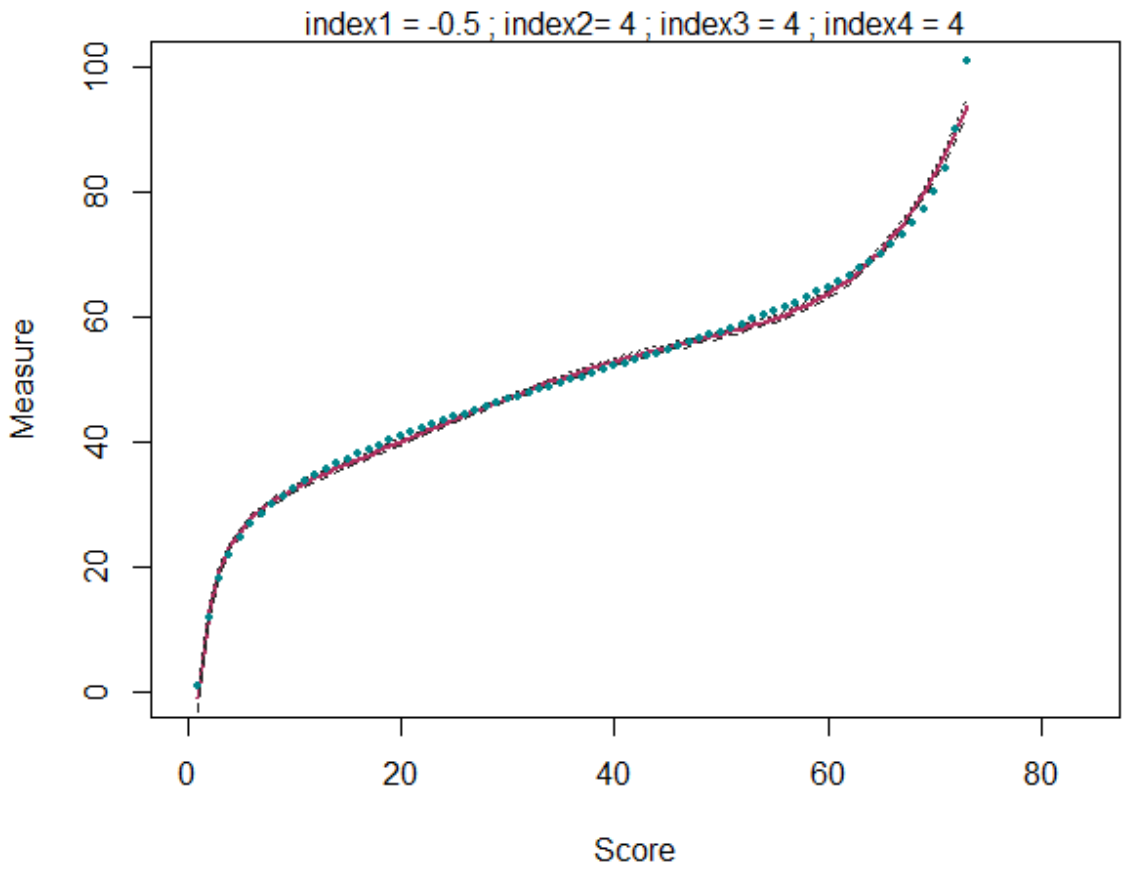


Figure 13. Fit of a 4th order FP trendline to the score-to-measure values in the Child-VQoL instrument.

$$Child\ VQoL\ Measure = 47.31 - 48.44\ \frac{1}{\sqrt{Score}} + 0.0002577\ \times Score^4 - 0.0001226\ \times\ Score^4 \times \ln(Score) + 0.00001467\ \times\ Score^4\ \times \ln(Score) \times\ \ln(Score) \quad \text{(h)}$$

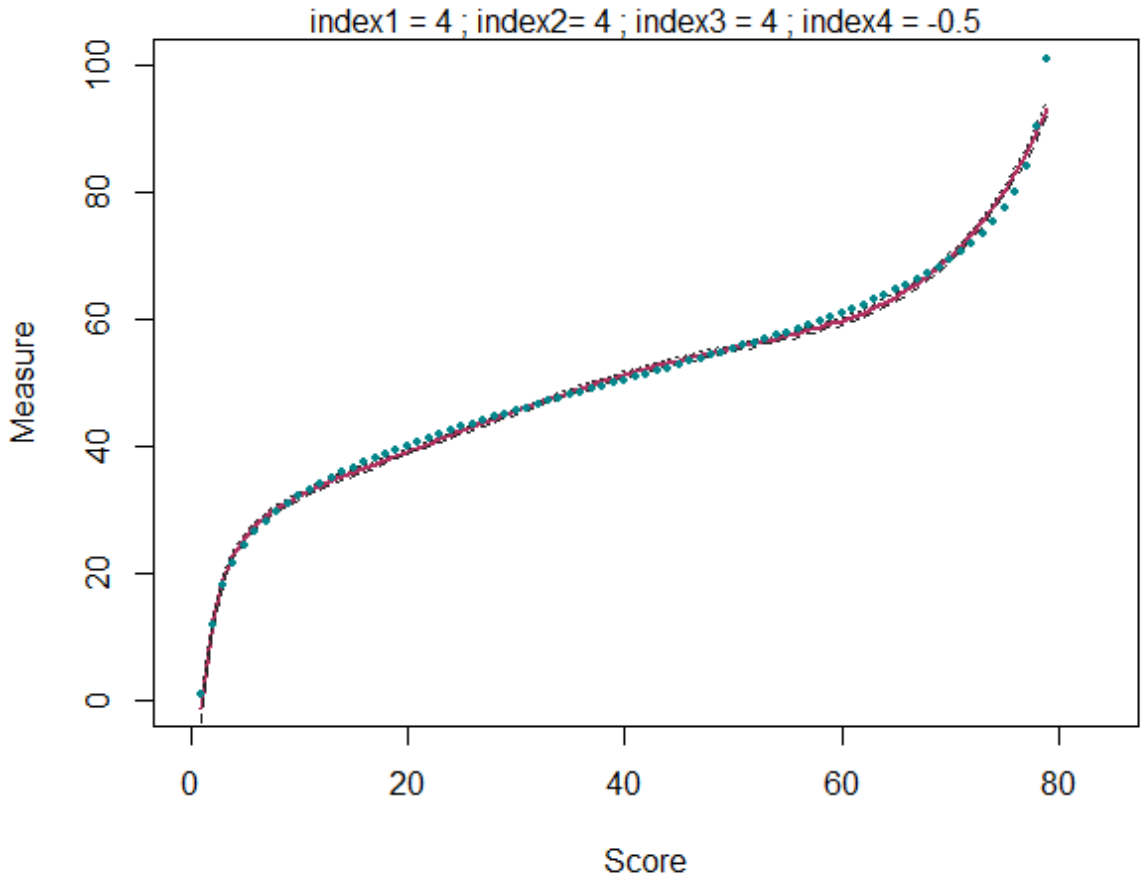


Figure 14. Fit of a 4th order FP trendline to the score-to-measure values in the YP-VQoL instrument.

$$YP-VQoL\ Measure = 47.09 + Score^4 - 0.0000904 \times Score^4 \times \ln(Score) + 0.00001062 \times Score^4 \times \ln(Score) - 48.40 \times \frac{1}{\sqrt{Score}} \quad \text{(i)}$$

Although the model produced by fitting this FP to both Child- and YP-VQoL scores was found to improve upon the linear equation provided by Winsteps, when fitted to raw- and measure-VQoL scores, the model was found to give a poor estimation of extreme scores when fitted to a user-friendly scale of 0-100. Thus, the best way to accurately calculate scores which are comparable between instrument versions is to use the conversion tables produced by Winsteps (see Appendix XVI, pg. 384 and XVII, pg. 385).

6.3.4.10 Testing construct validity

6.3.4.10.1 Demonstrating normality in the logit and summary scores

Skew, kurtosis, and Shapiro-Wilks tests of normality were used to assess construct validity in the developed instruments. Out of a total of 24 variables (comprising VA, VQoL summary logit scores and PedsQL summary and sub-domain scores for children (aged 8-12 years), YP (aged 13-17 years) and the entire sample combined), 17 were calculated as non-normally distributed when judged by the criteria of a) Z-score skewness outside the range of -2.00 to +2.00, b) Z-score kurtosis outside the range of -2.00 to +2.00, c) S-W significance value <.05 or d) a combination of the above. As a result, Spearman's Rank[401] statistics were used to evaluate correlations between all variables.

6.3.4.10.2 Correlation between VQoL logit scores and PedsQL summary scores

To determine construct validity of the instruments designed to measure VQoL, the correlation between VQoL logit scores and PedsQL summary scores was analysed. Seventy-nine children and 69 YP were included in this analysis after removing participants with >50% missing PedsQL scores.[388] When analysed separately, both the Child- and YP-VQoL logit scores correlated positively with the Child- and YP-PedsQL summary scores (r_s = .65, z = 5.71, for Child scores and r_s = .79, z = 6.48, for YP). Child- and YP-VQoL logit scores also correlated positively with the summary scores on the psychosocial subscale of the PedsQL (r_s= .64, z = 5.69, for Child scores and r_s = .81 z = 6.68, for YP).

Analysis also revealed a significant positive correlation between the VQoL logit and PedsQL summary scores when combining the Child and YP datasets (r_s = .72, z = 8.76). This is expected if the VQoL instrument truly detects components of QoL. Similarly, correlation between the VQoL summary scores and summary scores based on the psychosocial subscale of the PedsQL was significant (r_s = .73, z = 8.88), indicating that participants with higher levels of psychosocial well-being as detected by

the PedsQL instrument will also receive higher scores on the VQoL instrument (see Table 28, below).

Table 28. Table showing correlation coefficients between VQoL logit scores, PedsQL summary and sub-scale scores for children and YP aged 8-17 years (two-tailed).

	VQoL logit score	**PedsQL summary score**	**PedsQL psychosocial summary score**
VQoL logit score	1	.72 (8.76)	.73 (8.88)
PedsQL summary score	148	1	-
PedsQL psychosocial summary score	148	148	1

*z-scores shown in parentheses.

6.3.4.10.3 Correlation between VQoL logit scores and VA

Finally a correlation analysis of the relationship between VQoL logit scores and participants' latest recorded VA was undertaken. When analysed individually, neither Child- nor YP-VQoL summary scores correlated with VA (r_s = -.02, z = -0.15 for Child scores and r_s = -.13, z = -0.84 for YP).

After combining the Child and YP datasets, analyses revealed the two variables are non-significantly correlated (r_s = -.1, z = -1.18) indicating that there is no relationship between visual acuity and VQoL in this sample of children and YP (see Table 29, pg. 226).

Table 29. Table showing correlation coefficients between VQoL logit scores and VA for children and YP aged 8-17 years (two-tailed).

	VQoL logit score	VA
VQoL logit score	1	-.1 (-1.18)
VA	154	1

*z-scores shown in parentheses.

6.3.4.11 Final assessment of unidimensionality

Following formal psychometric item reduction, the 24-item Child-VQoL and 26-item YP-VQoL instruments were entered into a final FA to assess unidimensionality.

Notably, the percentage of variance explained by the first (largest) factor in both VQoL instruments increased after psychometric item reduction was performed, demonstrating improvement in the measurement of one construct. The greatest increase in variance explained was observed in the YP-VQoL instrument: 35.02% (see Table 21, pg. 193) to 39.59% (see Table 30, below).

Table 30. Eigenvalues for the largest factor, percent of variance explained, variation between eigenvalues for datasets with imputed missing data, number of eigenvalues and items not loading onto factors in the final FA.

	Eigenvalue for the first (largest) factor (percent of variance explained)	Variation between eigenvalues for the first (largest) factor in imputed datasets	No. of eigenvalues >1	No. of items loading onto Factor 1 <.4	Items not loading positively onto the first (largest) factor
Child-VQoL	7.13 (29.71)	0.12	7.2	2	None
YP-VQoL	10.29 (39.59)	0.12	5	0.2	None

6.4 Development of the functional vision (FV) instruments

6.4.1 Phase 1: Item development and adaptation

The *functioning – school, home and leisure* domain of the final hierarchy of child-centred data which was developed in the foundation research[5-7] was used as the foundation for developing items for the FV instrument (see Table 31, pg. 228).

Table 31. Areas of overlap and discrepancy between FV reported by 16-19 year old YP and 10-15 year old children.

VQoL domain[5]	Domain-pertinent issues identified from interviews conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
Functioning – school, home and leisure	Activities at home (e.g. TV, computers, reading, cooking, music), school (e.g. sport) and leisure (e.g. shopping, cinema), VI-related activities, such as adapted sports and using technology, Level of functioning (at home, at school – lessons and break-times, in leisure, out and about),	Even though I sit at the front, you still do get the odd time when you do need help seeing the board [*at school*]. (Male, 16, SVI/BL, early onset) I couldn't watch the TV from here. (Male, 17, VI, early onset) When I do the washing up, [*my mum*] says 'do it systematically.' (Male, 18, SVI/BL, early onset)	Feeling tired/having aches and pains related to visual impairment, Maintaining physical appearance (e.g. doing hair, putting on make-up), Level of functioning at break/lunchtimes, Age-specific activities (e.g. going to parties/pubs).	I'll get tired quicker and when I'm tired I'll focus less and then I'll get a headache […] And, then I'll come home tired so then I'll have to nap as well. (Female, 17, VI, early onset) Yeah, if I tried to do [*my own hair*], it would either be a guessing game or um, modern art! (Male, 16, SVI/BL, early onset)

VQoL domain[5]	Domain-pertinent issues identified from interviews conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
	Functioning – school, home and leisure continued… Restrictions and limitations, Mobility, Communication (e.g. reading text messages and emails).			*Functioning – school, home and leisure continued…* Oh, yeah, I can't do my eyebrows, make-up, and my toenails […] I have to check with my mum that I've done my make-up right. (Female, 19, SVI/BL, late onset) I just hang around with my friends. Erm, two of my friends, I made friends with this year, we go, most time we sit in this quiet room. Which is really quiet. And we can just relax, then we just chat really. (Female, 17, VI, early onset)

VQoL domain[5]	Domain-pertinent issues identified from interviews conducted with 10-15 year old children[5] and overlap with 16-19 year old YP.	Quotes (16-19 years old)	Areas of discrepancy/novel issues (16-19 years old)	Quotes (16-19 years old)
				Functioning – school, home and leisure continued… Obviously you play football, rugby, that sort of thing. And most of the time we just chat. Just walk around, have a conversation. Get food, that sort of thing. (Male, 16, SVI/BL, early onset) Yeah, sometimes I get lost in clubs […] it's probably because they've got strobe lighting. (Female, 17 years, VI, early onset)

The self-reported functional difficulties associated with visual impairment were comparable to those reported by younger children aged 10-15 years old.[5, 6] Discrepancies emerged when YP discussed functional difficulties associated with new, age-specific activities, such as visiting pubs and going to parties. The majority of new, age-appropriate issues relating to functional ability in YP emerged in relation to independence and autonomy which was largely dependent upon fewer parental restrictions and/or encouragement from parents to try new things, such as making a snack without help from others.

Codes were mapped onto 21 existing FV items. Items covered the domains shown in Table 31 (pg. 228). Fifteen existing items did not receive any new age-appropriate codes. Nineteen items which were excluded in the later stages of the foundation research were re-considered for inclusion. Codes which justified the inclusion of these original items were centred upon the functional consequences of increased independence, for example, cooking food or preparing a snack without help or learning strategies to stay safe when navigating outdoor public places when alone. Three codes that did not map onto existing items were considered for inclusion as new age-specific items (see Figure 15, below).

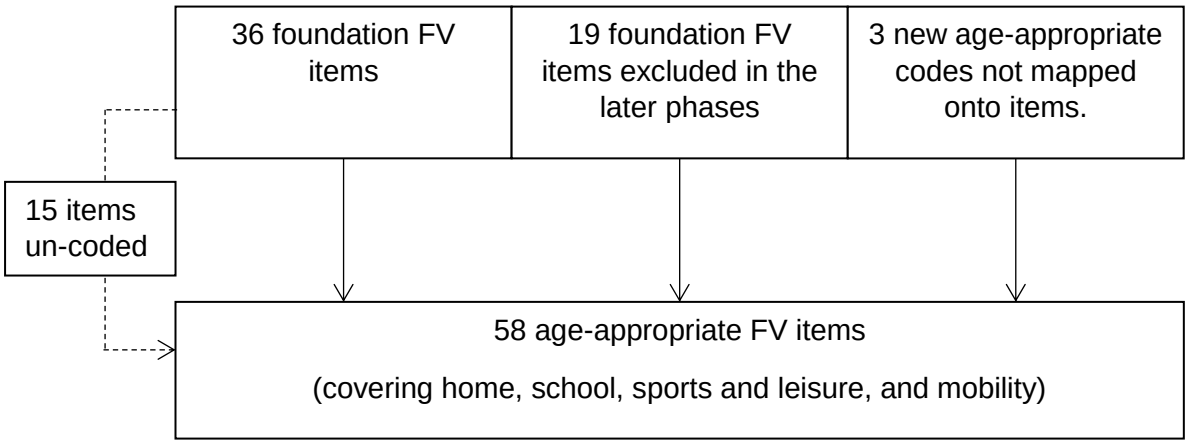


Figure 15. Flowchart showing the origin of items included in the first draft FV instrument.

Using the codes, a draft FV instrument for YP aged 13-17 years was developed. The first draft instrument is displayed in Table 32 (below).

Table 32. Items included in the first draft FV instrument.

Existing items (*n* = 21)	Watching TV
	Playing video and computer games
	Playing other games, e.g. board games or card games
	Using the computer for homework
	Reading food packets, tickets, labels or recipes
	Doing household chores, e.g. washing up
	Reading small print text books, worksheets and exam papers
	Reading enlarged text books, worksheets and exam papers
	Seeing the board in class
	Finding friends in the playground
	Taking part in maths classes
	Taking part in PE
	Taking part in English classes
	Keeping up with the teacher in lessons
	Getting around school by myself
	Getting around outdoors by myself
	Reading signs and posters at stations or shops
	Playing team sports e.g. football, without adaptations
	Watching films in the cinema
	Watching plays and shows in the theatre
	Reading price tags
Existing un-coded items (*n* = 15)	Telling the time on a wrist watch
	Telling the time on a wall clock
	Using the computer for lessons
	Drawing or painting
	Reading hand writing

	Recognising people e.g. in school corridors
	Recognising other people's facial expressions
	Taking part in science classes
	Taking part in geography classes
	Keeping up with other students in class
	Getting around in crowds by myself
	Seeing small moving objects, such as balls
	Seeing large moving objects, such as cars passing
	Using escalators
	Finding correct money to pay
Foundation items excluded from the later stages of development (*n* = 19)	Getting myself a drink
	Making myself a snack
	Cooking
	Finding objects I have dropped or lost
	Getting dressed by myself
	Writing
	Taking part in drama classes
	Going up and down the stairs by myself
	Getting around the house by myself
	Finding my way around an unfamiliar house or a new building
	Crossing the road by myself
	Getting the right bus by myself
	Using other public transport e.g. trains, by myself
	Reading tickets or recipes
	Playing ball sports e.g. football with adaptations, such as brightly coloured or bell balls
	Using mobile phone for texting my friends
	Using mobile phone for phoning people
	Playing musical instruments
	Shopping by myself e.g. for food or clothes

New age-appropriate codes (*n* = 3)	Falling over, tripping, being clumsy, having accidents
	Seeing far-away/distant objects
	Telling what colour something is, colour perception

Consistent with the development of the VQoL instrument, not all of the items included in the foundation FV instrument were assigned codes. Despite this, they were entered into the first draft of the new, age-appropriate instruments, due to relevant literature and professional opinion indicating that they will likely be relevant items.

6.4.1.1 Outcomes from the expert consensus meeting concerning the FV instrument development

In addition to minor modifications made to reflect age-appropriate preferences in linguist terminology (see Table 33, below), two new FV items (Looking after my appearance, for example, doing my hair, shaving, putting on make-up, and Using a mobile phone or tablet for social networking (Facebook, Twitter, MySpace) were discussed for suitability for inclusion in the second draft FV instrument.

Table 33. Minor linguistic modifications made to FV items during the expert consensus meeting.

Original FV item	Modified FV item
Using the computer for lessons	Using the computer at school to do my schoolwork
Finding friends in the playground	Finding friends outdoors at school
Watching plays and shows in the theatre	Watching shows, such as plays, at the theatre

6.4.2 Phase 2: Pre-testing

6.4.2.1 Self-reported feasibility of the FV instrument

Consistent with self-reported feasibility of the YP-VQoL instrument, completion of the YP-FV instrument was described by the majority of participants as ‘easy’ or ‘very easy’ with many participants expressing positive perceptions of completing the instrument, and using it as a means to inform other people about the functional impact of visual impairment. As in pre-testing of the VQoL instrument, some participants expressed concerns that the instrument would take too long to complete. These however, were mitigated when participants were told that they would not necessarily be required to justify their answers when completing the instrument in clinical contexts.

6.4.2.2 FV item modification

6.4.2.2.1 Linguistic modifications

FV items referring to different lessons at school were modified as a result of YP’s experience of flexibility in education, meaning that not all subjects are studied. Modifications made by removing the term ‘lessons’ allowed participants flexibility in answering each item, and reference to their abilities outside of formal education.

6.4.2.2.2 Modifications to aid interpretation and ease of responding

Consistent with the findings from pre-testing the VQoL instrument, some participants experienced difficulty when choosing one response option, and many described the need for a response option which represented the mid-point between response option 2 (Easy) and 3 (Difficult). When probed about their feelings about this, many participants described their functional ability as context dependent, varying in relation to levels of lighting, physical surroundings and social support.

Difficulty participants had when choosing one response option to describe their functional vision was evidenced when participants described their responses to the items ‘Doing household chores, for example, washing up’, and ‘Recognising people, for

example, in school corridors'. Participants flagged these items as dependent upon the specific activity and the VA required, and had difficulty applying the items to a broader range of activities and locations versus, specifically 'washing up' and recognising people in 'school corridors'. As a result of this, items were modified by adding a second example (e.g. '…or tidying my bedroom' and '…in shops') in which to consider functional ability.

One item (Getting around outdoors by myself) was modified and separated by adding an additional item to distinguish between difficulty getting around outdoors in daylight and when it's dark.

Table 34. Items which were modified as a result of phase 2: pre-testing.

Original YP-FV item	Modified YP-FV item
Using the computer at home to do my homework	Using the computer at home to do my homework/coursework
Finding objects I have dropped	Finding objects I have dropped such as coins or glasses on a low contrast surface
Using the computer at school to do schoolwork	Using the computer at school or college to do schoolwork/coursework
Reading handwriting	Reading other people's handwriting
Seeing the board in the classroom	Seeing the board in the classroom when sitting at the front
Recognising people, for example, in school corridors	Recognising people, for example, in school corridors or shops
Recognising other people's facial expressions	Recognising other people's facial expressions when they are close to me/at arm's length
Finding friends outdoors at school	Finding friends in crowded areas
Taking part in Maths lessons	Doing maths
Taking part in English lessons	Doing English or literacy
Taking part in Science lessons	Doing science
Taking part in PE lessons	Doing sports at school/college
Keeping up with the teacher in lessons	Keeping up with the teacher or tutor in lessons
Getting around school by myself	Getting around school/college by myself
Seeing big moving objects, such as bicycles passing	Seeing big moving objects, such as bikes passing in day light.
Getting around outdoors by myself	Getting around outdoors e.g. shops or the park by myself in the daylight
	Getting around outdoors e.g. shops or the park by myself when it's dark

In addition to items requiring greater specificity, a number of participants had difficulty answering items, based on not knowing whether they should respond in relation to their

best corrected, or uncorrected vision. As a result of this, the phrase ‘Remember to tell us how things are for you when wearing your glasses (if you wear them), with your low vision aids and other devices (if you use them for these activities) and with the best lighting and contrast for you’ was added as a header to each page of the FV instrument.

6.4.2.2.3 Outcomes from the expert consensus meeting

The possibility of introducing a 5th response option as a mid-point was discussed during the expert consensus meeting in relation to the difficulty many participants had when choosing one response option and wanting to opt for a mid-option but being unable to do so. A decision was made to retain the 4 response options in light of the psychometric difficulties involved when using a mid-response option indicating neither easy nor difficult. Additionally, the same process of instrument development for younger children (aged <10 years) confirmed the suitability of 4 response options and, since the measures are intended to be used simultaneously in practice, 4 response options were retained.

6.4.3 Phase 3: Piloting

6.4.3.1 Qualitative feedback

In their qualitative feedback, and consistent with results from phase 2 of the FV instrument development, many participants described the different answers they would give depending on different contexts, and therefore confirmed the need for the added header inserted on each page of the FV instrument reminding participants to respond in relation to ‘the best lighting and contrast for you’ (see above). For example, one participant described the impact of lighting:

> *“Ease of doing things is very much improved if the lighting conditions are right. For instance bright sunlight makes everything practically impossible unless a room is made darker or wearing wrap-around dark glasses over correction glasses.” (Female, 15 years, SVI/BL, early onset).*

6.4.3.2 Quantitative feedback

6.4.3.2.1 Missing data

The ‘true’ missing data (i.e. no response given) per item for the FV instrument were ≤13.04% with three missing responses on the FV item ‘Watching shows, such as play, at the theatre’, and missing completely at random. Four participants had 5.13% missing data as a result of items which had been modified after the questionnaire booklets had been sent out. These items had been modified significantly and responses were coded as ‘no response’ (see Table 8, pg. 113).

6.4.3.2.2 Use of response categories

Similar to results from piloting the VQoL instrument, a small number of participants indicated their favoured response option was missing from the FV instrument by circling two answers. The majority of these answers were reported by the same participants. For example, one participant provided the response option ‘between 2 and 3’ on 13 separate occasions (accounting for 31.71% of this participants’ total FV responses) indicating some difficulty deciding between the response ‘Easy’ and ‘Difficult’. These responses were coded according to the values shown in Table 8 (pg. 113), and results are presented in relation to the number of valid responses after entering these values. As in phase 3 of the VQoL instrument development, no further analyses were conducted to impute missing data and no participants were removed because of missing data.

6.4.3.2.3 Feasibility

Completion time for the FV instrument ranged from 5-60 minutes (median = 14.5, IQR = 10) and 91% of participants rated completion of the YP-FV instrument as ‘easy’ or ‘very easy’. All participants rated the instructions as ‘easy’ or ‘very easy’ to understand.

When asked about their overall opinion of the questionnaire booklet containing both VQoL and FV instruments, 56.52% of YP stated that completion of the questionnaire booklet was useful to them. Nine participants (31.13%) reported needing help from

another person to complete the full booklet. The majority of these (*n* = 7) reported needing help to read the items and response options.

6.4.3.3 Response distribution in the FV instrument

6.4.3.3.1 Skew and kurtosis in the FV instrument

Four FV items were flagged as problematic in terms of skewness and kurtosis outside the acceptable limits: all originated from the foundation research and three referred to reading printed text (Reading food packets, tickets, labels or recipes, Reading enlarged textbooks, worksheets and exam papers, Reading other people's handwriting, Finding friends in crowded areas).

6.4.3.3.2 Floor and ceiling effects in the FV instrument

One FV item was flagged as problematic due to floor effects (e.g. >60% choosing response option 4 (Very difficult or impossible)). This item (Reading small print textbooks, worksheets or exam papers) originated from the foundation research. No participants chose response option 4 when answering 5 FV items (Making myself a snack at home, Doing English or literacy, Getting around school/college by myself, Finding correct money to pay when shopping, Using a mobile phone or tablet for social networking, for example, Facebook, Twitter or MySpace).

No FV items appeared to have ceiling effects (>60% respondents choosing option 1 (Very easy)). However, three FV items (Reading small print textbooks, worksheets and exam papers, Seeing small balls when playing games, such as tennis or cricket, Reading signs and posters at stations or shops) were not allocated response option 1 by any participants, indicating greater functional difficulty (see Figure 16, pg. 241).

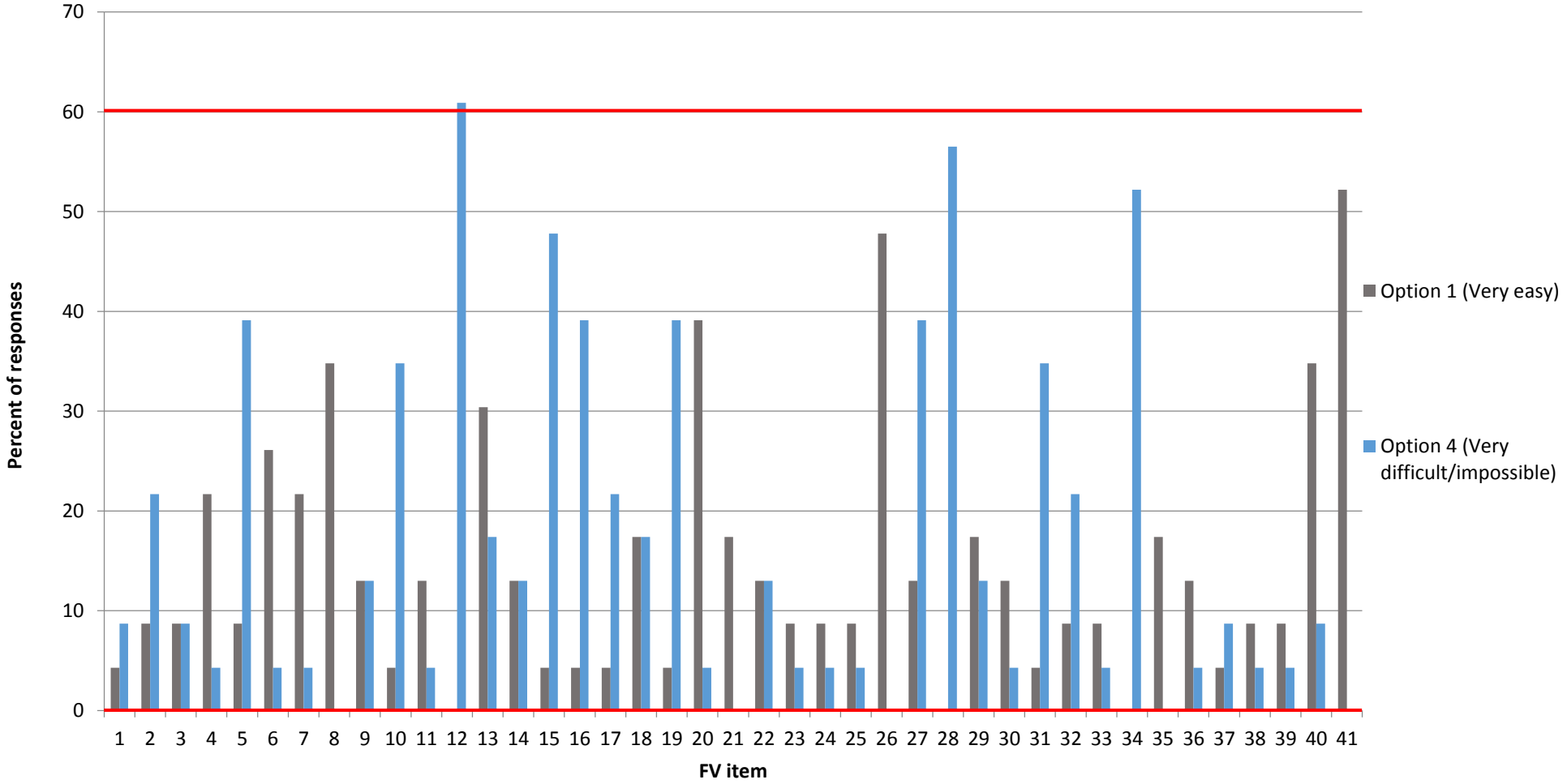


Figure 16. Percent of YP choosing extreme response categories when answering items in the FV instrument (1% and 60% thresholds highlighted in red).

6.4.4 Phase 4: Validation

Table 35 (pg. 243) shows the FV items entered into the questionnaire booklets which were sent to children (aged 8-12 years) and YP (aged 13-17 years) in the final validation phase. As in phase 4 of the VQoL instrument development, each item was assigned a value corresponding to the order of presentation within each instrument. Items presented in the same row of Table 35 are overlapping or 'core' items which are present in instruments designed for both age groups. Items parallel to empty cells are age-specific items, presented to only one age group.

Table 35. Items included in the Child (8-12 years) and YP (13-17 years) version of the FV instrument entered into phase 4 (psychometric validation).

FV instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
1	Watching TV	1	Watching TV
2	Playing video and computer games	2	Playing video and computer games
3	Playing other indoor games, such as board games or card games	3	Playing other indoor games, such as board games or card games
4	Playing outdoor games, such as tag or hide and seek		
5	Using the computer at home to do my school work	4	Using the computer at home to do my homework/coursework
6	Reading small writing such as food packets or instructions for toys	5	Reading food packets, tickets, labels or recipes
7	Doing household jobs, for example, tidying up my toys	6	Doing household chores, for example, washing up or tidying my bedroom
		7	Looking after my appearance, for example, doing my hair, shaving, putting on make-up
		8	Making myself a snack at home
		9	Making myself a meal
		10	Finding objects I have dropped such as coins or glasses on a low contrast surface

FV instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
8	Using the computer in school lessons	11	Using the computer at school or college to do schoolwork/coursework
9	Reading small print worksheets and text books like dictionaries	12	Reading small print textbooks, worksheets and exam papers
10	Reading enlarged worksheets and text books like dictionaries	13	Reading enlarged textbooks, worksheets and exam papers
11	Drawing or painting	14	Drawing or painting
12	Reading other people's handwriting	15	Reading other people's handwriting
13	Seeing the board in the classroom	16	Seeing the board in the classroom when sitting at the front
14	Recognising people, for example in school corridors	17	Recognising people, for example, in corridors at school/college or shops
15	Recognising other people's facial expressions	18	Recognising other people's facial expressions when they are close to me/at arm's length
16	Finding friends in the playground	19	Finding friends in crowded areas
17	Doing maths in lessons	20	Doing maths
18	Doing literacy in lessons	21	Doing English or literacy
		22	Doing science
19	Doing PE	23	Doing sports at school/college

FV instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
20	Keeping up with the teacher in lessons	24	Keeping up with the teacher or tutor in lessons
21	Keeping up with other children in lessons	25	Keeping up with other students in lessons
22	Getting around the school without someone helping me	26	Getting around school/college by myself
23	Playing team sports without special balls	27	Playing team sports, such as football, without adaptations such as special balls
24	Seeing small balls when playing games, such as tennis or cricket	28	Seeing small balls when playing games, such as tennis or cricket
25	Seeing big moving objects, such as bicycles passing by	29	Seeing big moving objects, such as bikes passing, in daylight.
26	Getting around outdoors in daytime	30	Getting around outdoors e.g. shops or the park, by myself when it's daylight
27	Getting around outdoors when it is dark	31	Getting around outdoors e.g. shops or the park, by myself when it's dark
		32	Getting around in crowds by myself
		33	Finding my way around an unfamiliar house or a new building
28	Reading signs and posters at stations or shops	34	Reading signs and posters at stations or shops
		35	Finding correct money to pay when shopping

FV instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Item**	**Item no.**	**Item**
29	Watching films in the cinema	36	Watching films in the cinema
30	Watching shows at the theatre	37	Watching shows, such as plays, at the theatre
		38	Crossing the road by myself
		39	Using public transport, such as trains, buses or the tube by myself
		40	Using a mobile phone to text people
		41	Using a mobile phone or tablet for social networking, for example, Facebook, Twitter or MySpace

6.4.4.1 Data verification

The FV dataset was verified by the same researcher who entered the data in the first instance (AR) to validate the data entry. A total of 12 incorrect data entries were found out of 2,911 individual entries constituting 0.27% of the dataset. Each incorrect entry was cross-referenced with the original data source and corrected. Ten percent of this dataset was verified by a second member of the research team (VT) who found 100% of the data to have been entered correctly.

With regards to the Child-FV dataset, 43% of data entries were verified by the same researcher who entered the data (VT). No incorrect data entries were found. Accordingly, 10% of the data entries were verified independently by a second member of the research team (AR): 100% were found to be correct.

6.4.4.2 Missing data

6.4.4.2.1 Missing data per person

With regards to missing data, four children were excluded from the Child-FV instrument development based on the criteria for exclusion of >25% missing data. Thus, data from 82 children were included in the phase 4 analysis. After excluding participants on the basis of not returning completed consent forms and/or booklets, no YP were excluded as a result of missing data. Seventy-one YP were included in the YP-FV phase 4 analysis.

6.4.4.2.2 Missing data per FV item

After removing participants based on missing data, the largest percent of missing data per item was 4.88% for the Child-FV items ‘Playing team sports without special balls’, and ‘Getting around outdoors when it is dark’. With regards to the YP instrument, the largest percent of missing data was 7.04% for the item ‘Watching shows, such as plays, at the theatre’ (see Table 36, pg. 248). Amount of missing data was negligible, justifying inclusion of all items in the subsequent stages of analysis.

Table 36. Missing data per item in the FV instrument, separated by age-group.

FV instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Missing data, *n* (%)**	**Item no.**	**Missing data, *n* (%)**
27	4 (4.88)	37	5 (7.04)
23	4 (4.88)	38	3 (4.23)
29	3 (3.66)	36	3 (4.23)
25	2 (2.44)	29	3 (4.23)
21	2 (2.44)	16	3 (4.23)
18	2 (2.44)	2	3 (4.23)
17	2 (2.44)	41	2 (2.82)
5	2 (2.44)	26	2 (2.82)
4	2 (2.44)	23	2 (2.82)
1	2 (2.44)	14	2 (2.82)
30	1 (1.22)	4	2 (2.82)
26	1 (1.22)	3	2 (2.82)
20	1 (1.22)	40	1 (1.41)
16	1 (1.22)	39	1 (1.41)
15	1 (1.22)	35	1 (1.41)
14	1 (1.22)	33	1 (1.41)
12	1 (1.22)	32	1 (1.41)
10	1 (1.22)	31	1 (1.41)
9	1 (1.22)	30	1 (1.41)
8	1 (1.22)	28	1 (1.41)
7	1 (1.22)	25	1 (1.41)
6	1 (1.22)	24	1 (1.41)

FV instrument			
Child (8-12 years)		**YP (13-17 years)**	
Item no.	**Missing data, *n* (%)**	**Item no.**	**Missing data, *n* (%)**
3	1 (1.22)	19	1 (1.41)
2	1 (1.22)	13	1 (1.41)
		12	1 (1.41)
		11	1 (1.41)
		10	1 (1.41)
		1	1 (1.41)

6.4.4.3 Use of response categories

The majority of participants appeared to endorse response categories well. However, occasions when children selected multiple response options, or indicated their favoured response option to be missing constituted 1.02% of the Child, and 1.2% of the YP-FV datasets. Responses were re-coded using the values shown in Table 8 (pg. 113).

6.4.4.4 Response distribution

As with the items included in phase 4 of the VQoL instrument development, a substantial number of child-FV and YP-FV items (see Table 37, pg. 250) were shown to have z-score skewness and/or z-score kurtosis values outside the acceptable thresholds of -2 to +2. These items were flagged as problematic.

Table 37. Z-score skewness, z-score kurtosis, floor and ceiling effects for each item in the child- and YP-FV instruments.

Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4	Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4
FV (Child)					FV (YP)				
1	-1.17	-1.6	19.5	9.8	1	1.25	-0.89	14.1	12.7
2	-0.3	-1.52	15.9	13.4	2	-0.07	-1.64	12.7	19.7
3	-0.27	-1.68	20.7	8.5	3	0.48	-0.91	11.3	11.3
4	-0.51	-2.18	18.3	23.2	4	1.5	-1.1	15.5	15.5
5	-0.35	-1.86	19.5	14.6	5	-1.95	-0.89	5.6	35.2
6	-5.56	3.31	3.7	59.8	6	1.29	-0.96	23.9	7
7	1.21	-1.29	31.7	1.2	7	0.59	-1.78	22.5	14.1
8	-0.64	-1.48	13.4	17.1	8	2.52	-0.32	36.6	5.6
9	-4.8	2.01	4.9	53.7	9	0.33	-1.94	23.9	14.1
10	3.61	-0.35	43.9	7.3	10	-2.55	1.03	4.2	29.6
11	1.54	-1.45	30.5	8.5	11	0.65	-1.37	16.9	12.7
12	-3.34	1.47	4.9	34.1	12	-2.92	-0.73	1.4	52.9
13	-4.2	1.16	7.3	46.3	13	2.95	-0.37	36.6	11.3
14	-0.56	-1.89	12.2	24.4	14	-0.09	-2.23	25.4	16.9
15	0.09	-2.04	17.1	20.7	15	-1.55	-0.86	1.4	35.2
16	-3.4	-0.36	9.8	43.9	16	-1.8	-1.06	7	32.4
17	-0.05	-1.67	22	6.1	17	-1.89	-0.85	9.9	28.2
18	0.01	-1.03	15.9	4.9	18	0.50	-2.41	23.9	25.4
19	-0.72	-1.61	14.6	18.3	19	-1.34	-1.5	0	39.4
20	-0.61	-1.68	19.5	11	20	1.63	-0.8	28.2	5.6
21	-0.57	-1.30	13.4	13.4	21	1.13	-1.02	21.1	8.5
22	2.73	-0.6	46.3	1.2	22	0.93	-1.52	19.7	15.5
23	-1.05	-2.01	15.9	25.6	23	-0.62	-1.57	14.1	19.7
24	-3.1	-1.02	12.2	45.1	24	0.73	-0.82	15.5	8.5
25	0.35	-1.49	22	6.1	25	0.66	-1.05	18.3	8.5
26	0.84	-1.36	32.2	7.3	26	1.95	-0.33	31	4.2
27	-3.59	-0.04	9.8	42.7	27	-1.42	-1.8	14.1	32.4

Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4	Item no.	Z-score skew	Z-score kurtosis	% endorsing option 1	% endorsing option 4
FV (Child)					FV (YP)				
28	-2.74	0.02	7.3	31.7	28	-4.15	2.4	1.4	50.7
29	1.96	-1.06	29.3	8.5	29	0.45	-1.87	21.1	15.5
30	-1	-2.19	17.1	28	30	1.97	-0.76	21.1	12.7
					31	-1.05	-1.67	11.3	28.2
					32	-1.66	-1.18	11.3	25.4
					33	-0.63	-1.71	12.7	23.9
					34	-3.10	0.57	4.2	42.3
					35	0.93	-1.18	22.5	8.5
					36	2.05	-0.59	18.3	12.7
					37	0.02	-1.31	2.8	19.7
					38	0.3	-1.83	19.7	15.5
					39	-1.01	-1.5	15.5	19.7
					40	2.52	-0.71	36.6	9.9
					41	2.55	-0.92	36.6	12.7

*FV response option 1 = ‘Very easy’, response option 4 = ‘Very difficult or impossible’.

The percent of respondents endorsing extreme categories in each item was analysed in parallel to the largest, and most problematic, skew and kurtosis values. Only one item (Item 19 (Finding friends in crowded areas)) was removed from the subsequent stages of item reduction of the YP-instrument development, based on no respondents choosing response option 1 (Very easy).

6.4.4.5 Assessment of unidimensionality

6.4.4.5.1 FA

FA was conducted using each of the 5 datasets with imputed missing data for each instrument, and the outcomes shown in Table 38 (pg. 252) are the mean values of each dataset.

Both Child- and YP-FV instruments appeared to have good unidimensionality, with all items loading positively (and >0.4) onto the first factor.

Table 38. Outcome from FA conducted to assess dimensionality in the FV instruments.

Instrument version	Eigenvalue for the first (largest) factor (percent of variance explained)	Variation between the first (largest) eigenvalues in imputed datasets	No. of eigenvalues >1	No. of items loading positive onto Factor 1 <.4	Items not loading positive onto the first (largest) factor
Child-FV	13.49 (44.95)	0.12	6	0	None
YP-FV	20.79 (51.97)	0.17	6	0	None

6.4.4.5.2 Parallel analysis

Parallel analysis was run using each of the 5 imputed datasets. The mean eigenvalues for the largest 5 eigenvalues in each dataset are presented in Table 39 (pg. 253).

Table 39. Average of the first five eigenvalues produced by parallel analysis for each dataset.

Actual eigenvalue	Average eigenvalue	95th Percentile eigenvalue
Child-FV		
13.48	2.36	2.56
1.96	2.14	2.29
1.63	1.98	2.1
1.4	1.85	1.96
1.14	1.73	1.82
YP-FV		
20.79	2.81	3.05
2.35	2.56	2.73
1.92	2.38	2.52
1.42	2.22	2.35
1.29	2.09	2.2

The matrix produced by parallel analysis for each dataset provides evidence to support the finding of unidimensionality in both Child-FV and YP-FV datasets as the second largest eigenvalue is smaller than both the average eigenvalue and percentile eigenvalue in both Child- and YP-FV datasets.

The values produced by the averaged matrix (see Table 39, above) were used to produce a scree plot for each dataset. Both scree plots visually represent unidimensionality in the datasets.

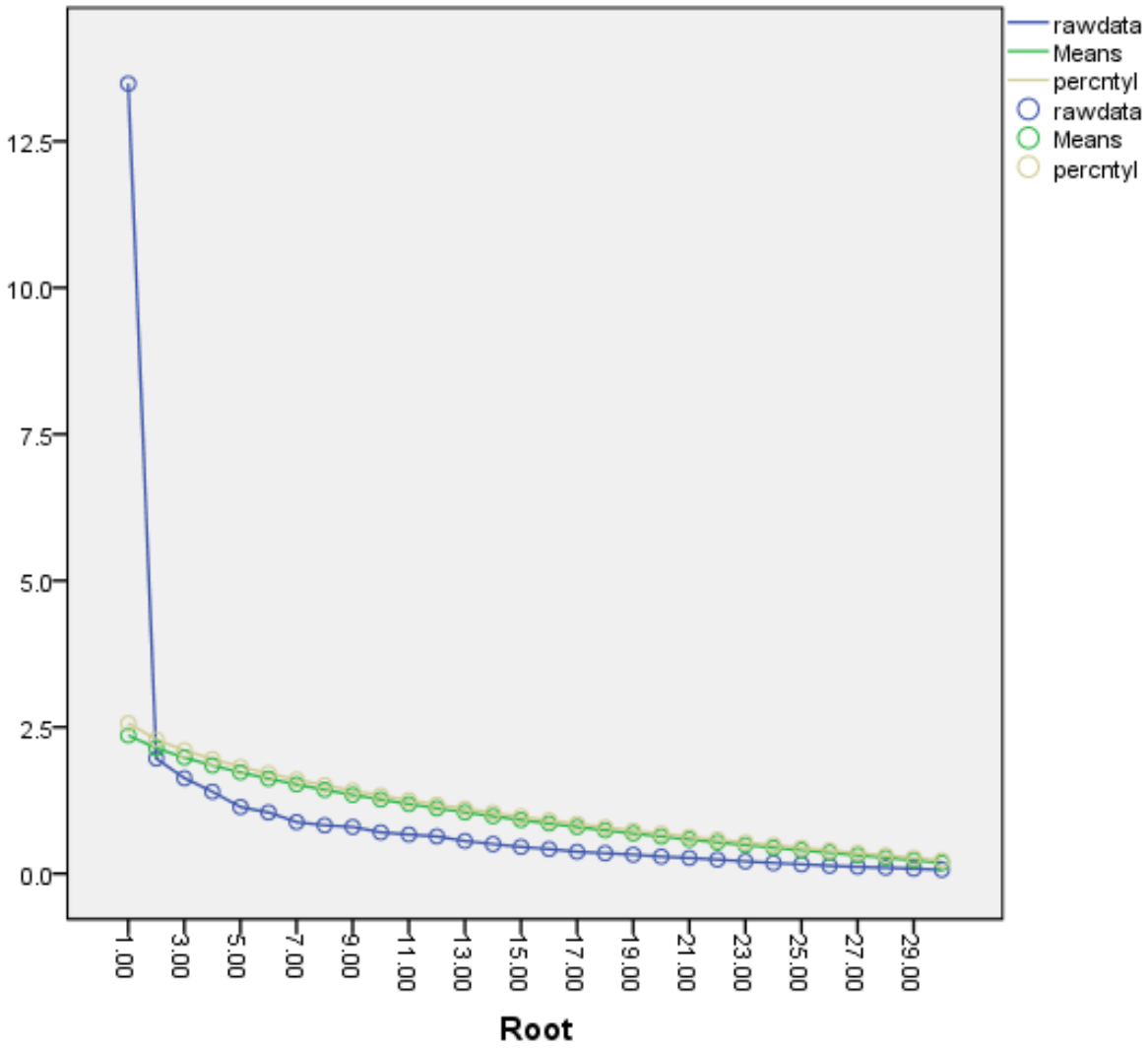


Figure 17. Screeplot showing actual, average and percentile eigenvalues in the Child-FV instrument.

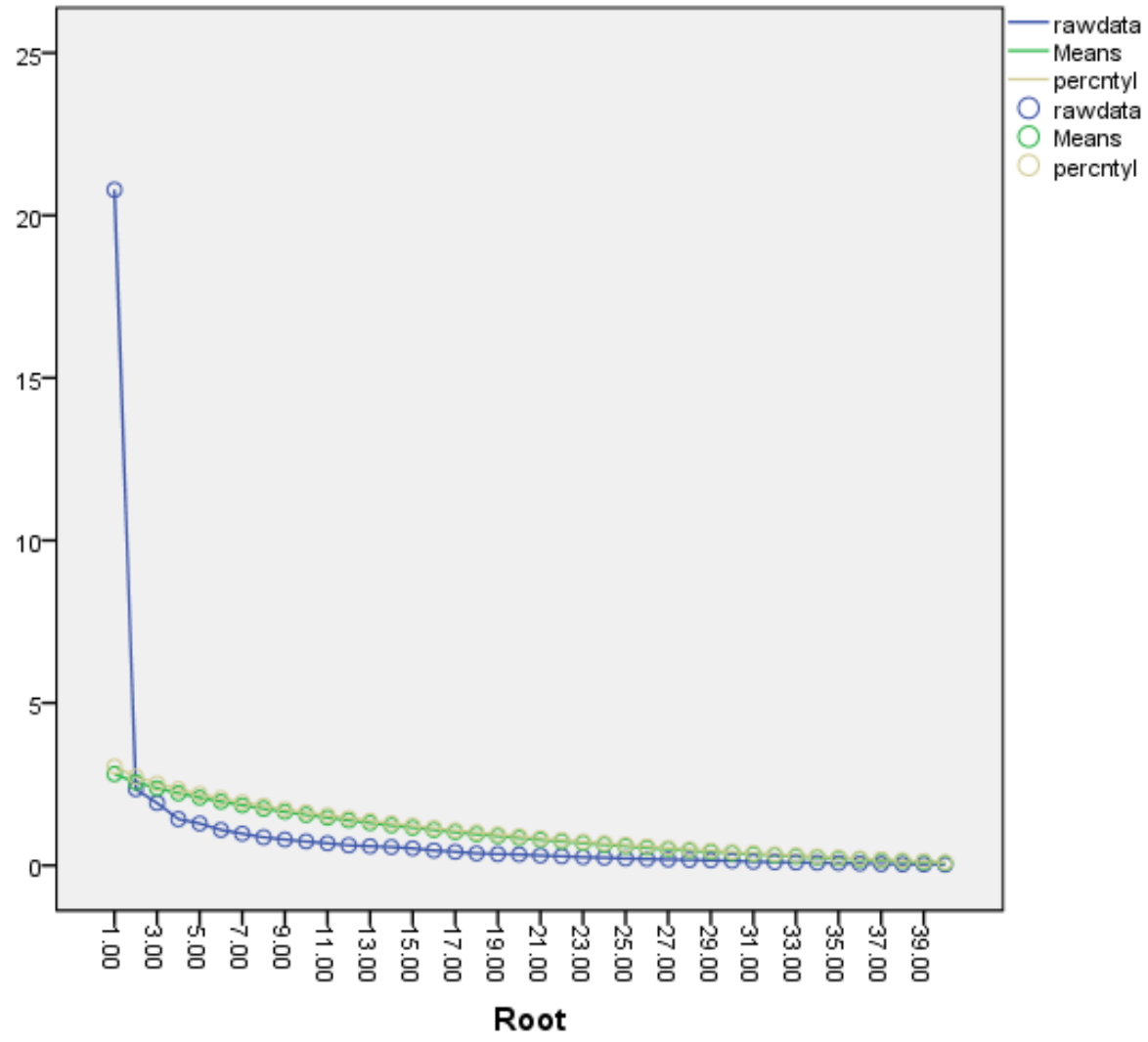


Figure 18. Screeplot showing actual, average and percentile eigenvalues in the YP-FV instrument.

6.4.4.6 Rasch analysis

Rasch analysis was performed on the Child- and YP-FV instrument datasets independently, and then, using the datasets combined with the aim of calibrating the instruments. Results from the Child-FV, YP-FV and calibration of both instruments are presented respectively.

6.4.4.7 Rasch analysis using the 30-item Child-FV instrument

All 30 items included in the draft Child-FV instrument were entered into the Rasch analysis after fulfilling the criteria specified in the preliminary item removal stage. Only

two iterations of Rasch analysis were required before the items included in the instrument conformed to the multiple criteria of good fit to the model of measurement.

6.4.4.7.1 Item fit to the Rasch measurement model

The first item to be removed was as a result of fit statistics outside the threshold of 0.5 to 1.5 (see Table 40, below). Item 27 (Getting around outdoors when it is dark) had a large OUTFIT MNSQ indicating a high level of unexpected observations by persons on this item which is relatively very easy or very hard for them and therefore a threat to validity. After removing Item 27, all fit statistics fell within the required threshold, demonstrating good fit of the data to the Rasch measurement model.

Table 40. Problematic items in the Child-FV instrument due to fit to the Rasch measurement scale.

		INFIT		**OUTFIT**	
Iteration	**Problematic item(s)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**
Iteration 1	27 (Getting around outdoors when it is dark)	1.33	1.9	1.67	3.1

6.4.4.7.2 Order of person abilities

After removing Item 27, all items were analysed for average measures of persons and the direction of ordering of these measures. No items were found to have person abilities which were out of order in relation to the responses on items.

6.4.4.7.3 Differential item functioning

DIF analyses were conducted using gender and the same stratified age groups described in Section 6.3.4.7.3 (pg. 199). Using the 29-item scale, analysis revealed no DIF contrasts above the threshold of 1.0 (see Table 41, pg. 258). The largest DIF

contrast observed between males and females was .73 in Item 11 (Drawing or painting) which was more difficult for females than males in this sample. The largest DIF contrast observed between young and old age groups was Item 23 (Playing team sports without special balls) which was .68 logits more difficult for participants in the older age group (aged 10-13 years).

The final iteration of the Child-FV instrument contained 29 items. The measurement properties of the final iteration were checked for conformity to the predefined thresholds, and fit statistics were within acceptable limits (see Table 41, pg. 258). The average measures of persons were ordered in each item in relation to ability.

Table 41. Fit and DIF statistics of items in the final 29-item Child-FV scale.

Item code	Child-FV item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
1	Watching TV	0.82	0.77	-0.43	0.18
2	Playing video and computer games	0.99	0.96	0	-0.13
3	Playing other indoor games, such as board games or card games	0.74	0.71	0.23	-0.12
4	Playing outdoor games, such as tag or hide and seek	1.03	0.99	0.11	0.02
5	Using the computer at home to do my school work	1.26	1.23	0.16	0.09
6	Reading small writing such as food packets or instructions for toys	1.03	0.94	-0.11	-0.22
7	Doing household jobs, for example tidying my toys	0.94	0.98	-0.19	0.09
8	Using the computer in school lessons	0.91	0.86	0.08	-0.28
9	Reading small print worksheets and textbooks like dictionaries	1	0.94	-0.13	0.63
10	Reading enlarged worksheets and textbooks like dictionaries	1.15	1.37	-0.07	-0.38
11	Drawing or painting	1.11	1.22	-0.35	0.73
12	Reading other people's handwriting	0.64	0.63	-0.16	0.07
13	Seeing the board in the classroom	1.10	1.06	-0.66	0.36
14	Recognising people, for example in school corridors	1.08	1.07	0	0

Item code	Child-FV item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
15	Recognising other people's facial expressions	0.94	0.91	0.09	0.62
16	Finding friends in the playground	1.07	0.96	0	0.11
17	Doing maths in lessons	1.08	1.05	0.34	-0.41
18	Doing literacy in lessons	1.07	1.11	-0.34	-0.21
19	Doing PE	1.15	1.23	0.13	-0.53
20	Keeping up with the teacher in lessons	1.24	1.26	-0.20	-0.15
21	Keeping up with other children in lessons	1	1.07	0.47	-0.08
22	Getting around the school without someone helping me	1.23	1.05	-0.25	-0.37
23	Playing team sports without special balls	1.26	1.16	0.68	-0.44
24	Seeing small balls when playing games such as tennis or cricket	1.25	1.15	0.19	-0.13
25	Seeing big moving objects, such as bicycles passing by	0.73	0.75	0.16	0.13
26	Getting around outdoors in daytime	0.75	0.74	-0.20	-0.13
28	Reading signs and posters at stations and shops	0.59	0.56	-0.25	0
29	Watching films in the cinema	0.91	0.84	0.40	0.20
30	Watching shows at the theatre	1.07	1.05	0	0.47

6.4.4.7.4 Targeting

The person-item map using the final 29-item scale is presented in Figure 19 (pg. 261). The map revealed good targeting of the items to ability of participants with a difference between the two means of .04. The separation and reliability values support the good targeting of this scale and are within acceptable limits (separation = 5.54, reliability = .97) when analysing the real RMSE.

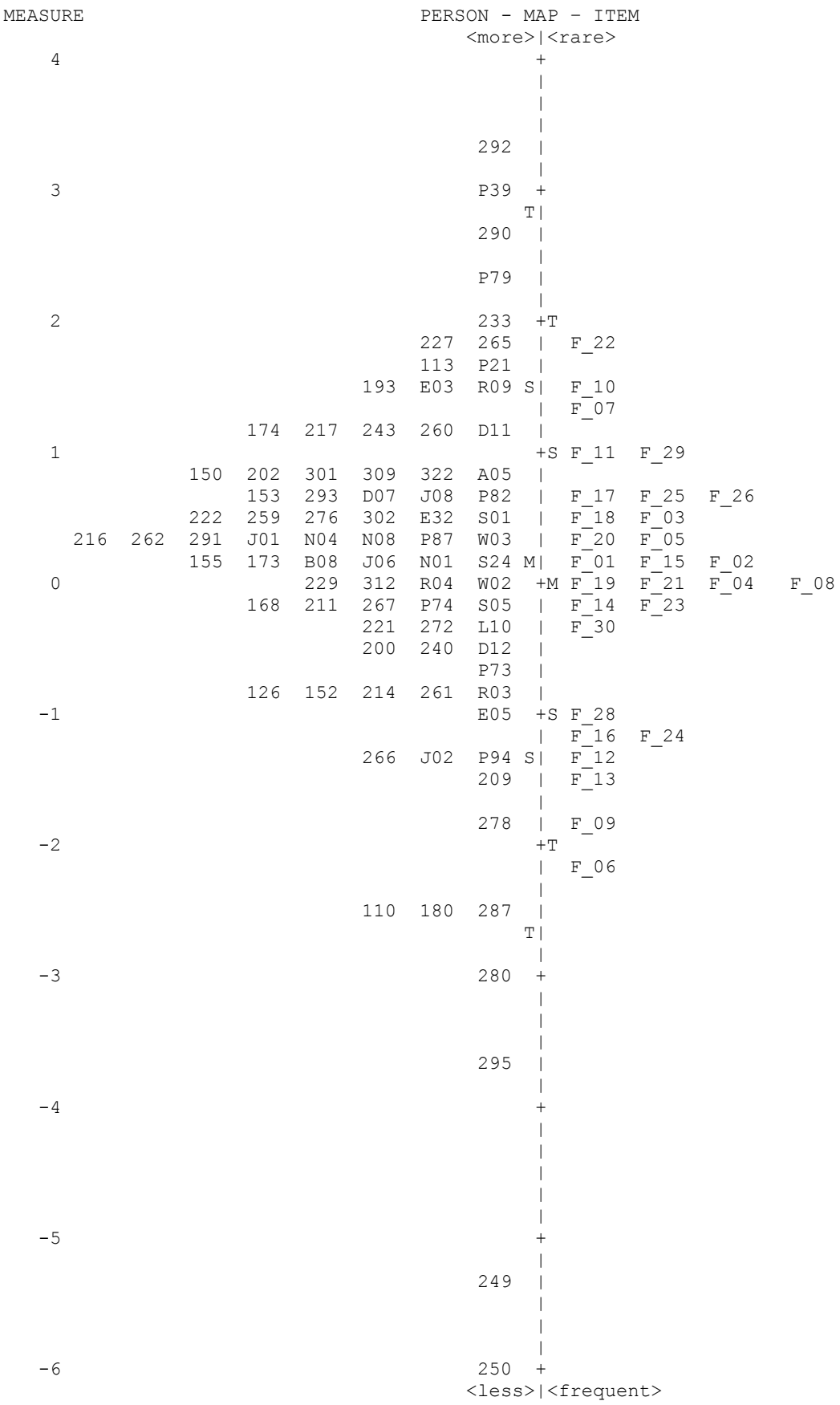


Figure 19. Item-person targeting of the final 29-item Child-FV scale.

6.4.4.7.5 Response category function

Because all items included in the FV instrument are worded in the same direction (with higher response options indicating lower functional ability) only one collection of category probability curves was produced by Rasch analysis. Figure 20 (below) demonstrates a distinct period in terms of the latent variable (displayed on the x axis) when each category is most probable. In terms of the Andrich thresholds for the categories, thresholds increased by at least 1.42 logits across the scale, which meets the criteria of an increase of 1.4. Thus, there is evidence to prove that the response categories in the Child-FV instrument function well in terms of measurement across the abilities of participants in this sample.

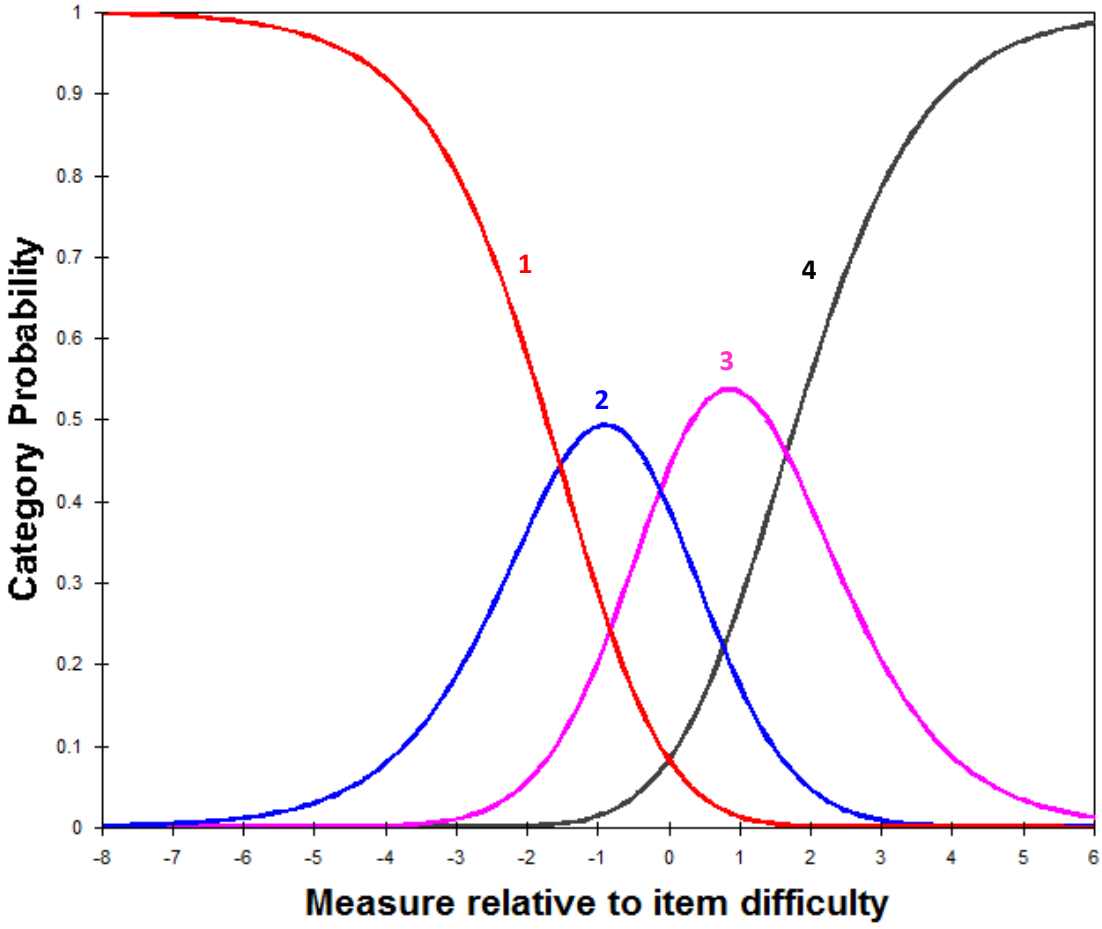


Figure 20. Category probability curves for the 29-item Child-FV instrument (1 = Very easy, 2 = Easy, 3 = Difficult, 4 = Very difficult or impossible).

6.4.4.8 Rasch analysis using the 40-item YP-FV instrument

Forty items were entered into Rasch analysis of the YP-FV instrument, with one item (Item 19 (Finding friends in crowded areas)) having been removed in the preliminary stage of item removal (see Section 6.4.4.4, pg. 249). During the first iteration of Rasch analysis (using the 40-item scale) all items fulfilled the criteria of good measurement in terms of fit to the Rasch measurement model and ordering of person abilities in relation to response categories. Thus, DIF analyses were conducted using the full 40-item scale.

6.4.4.8.1 Differential item functioning

DIF analyses were conducted using groups of participants stratified by gender and the ages specified in Section 6.3.4.8.3 (pg. 207). When stratified by gender, three items were shown to be problematic, with DIF contrasts greater than 1. Two of these items (Items 21 (Doing English or literacy) and 40 (Using a mobile phone to text people)) were shown to be more difficult for females in the sample with DIF contrasts of 1.30 and 1.06 respectively. The remaining item (Item 38 (Crossing the road by myself)) was assigned a DIF contrast of -1.15, being more difficult for males.

In relation to DIF by age group, four items were flagged as problematic. Three items (Items 13 (Reading enlarged textbooks, worksheets and exam papers), 14 (Drawing or painting) and 27 (Playing team sports, such as football, without adaptations such as special balls)) were shown to be more difficult for participants in the younger age group (aged 13-15 years) with DIF contrasts of -1.25, -1.59 and -1.03 respectively, and one item (Item 32 (Getting around in crowds by myself)) was shown to be more difficult for participants classified as ‘old’ (aged 16-18 years) with a DIF contrast of 1.03.

Six problematic items were removed iteratively in the order of magnitude of DIF contrast (ranging from largest to smallest). After removing Item 13 for DIF by age group, the DIF contrast for Item 32 was no longer greater than the threshold of 1, and subsequently retained in the scale. The 34-item scale was then entered into a new

Rasch analysis and assessed for fit to the Rasch measurement scale. As a result of the removal of items due to substantial DIF, the fit statistics of three further items reached the unacceptable threshold of >1.5 and were removed (see Table 42, below).

Table 42. Problematic items in the YP-FV instrument due to fit to the Rasch measurement scale.

		INFIT		OUTFIT	
Iteration	**Problematic item(s)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**
Iteration 7 (Excluding Item 19 (Finding friends in crowded areas), 14 (Drawing or painting), 21 (Doing English or literacy), 13 (Reading enlarged textbooks, worksheets and exam papers), 40 (Using a mobile phone to text people) and 27 (Playing team sports, such as football, without adaptations such as special balls)	23 (Doing sports at school/ college)	1.57	3.0	1.64	3.2
Iteration 8 (Excluding Items 19, 14, 21, 13, 40, 27 and 23)	17 (Recognising people, for example, in corridors at school/college or shops)	1.58	3.1	1.43	2.3

		INFIT		OUTFIT	
Iteration	**Problematic item(s)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**	**Mean-square value (MNSQ)**	**Z-standardised (ZSTD)**
Iteration 8 (Excluding Items 19, 14, 21, 13, 40, 27, 23 and 17)	18 (Recognising other people's facial expressions when they are close to me/at arm's length)	1.59	3.1	1.49	2.6

The results of DIF analyses which were conducted using the revised 31-item scale are presented in Table 43 (pg. 266). No DIF contrasts reached the threshold of >1, and all remaining items were retained.

Table 43. Fit and DIF statistics of items in the final 31-item YP-FV scale.

Item code	YP-FV item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
1	Watching TV	0.96	1.04	-0.11	0.38
2	Playing video and computer games	1.20	1.25	-0.78	0.34
3	Playing other indoor games, such as board games or card games	0.78	0.82	-0.27	0.25
4	Using the computer at home to do my homework/coursework	1.32	1.36	0.81	0.48
5	Reading food packets, tickets, labels or recipes	0.83	0.77	-0.09	0.34
6	Doing household chores, for example, washing up or tidying my bedroom	0.81	0.85	-0.30	0.05
7	Looking after my appearance, for example, doing my hair, shaving or putting on make-up	1.03	1.01	0.45	-0.32
8	Making myself a snack at home	0.74	0.69	0.61	0.36
9	Making myself a meal	0.98	0.97	0.68	0.36
10	Finding objects I have dropped such as coins or glasses on a low contrast surface	1.03	1.30	-0.65	0.33
11	Using the computer at school or college to do schoolwork/coursework	1.04	0.98	0	0
12	Reading small print worksheets, textbooks and exam papers	1.06	1.15	-0.36	-0.32
15	Reading other people's handwriting	0.91	0.92	-0.21	0.24

Item code	YP-FV item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
16	Seeing the board in the classroom when sitting at the front	1.16	1.10	-0.78	0.18
20	Doing maths	1.26	1.26	-0.11	0.16
22	Doing science	1.24	1.23	-0.28	0.52
24	Keeping up with the teacher or tutor in lessons	0.94	0.91	-0.28	0.59
25	Keeping up with other students in lessons	0.78	0.77	-0.27	-0.13
26	Getting around school/college by myself	0.81	0.75	-0.23	0
28	Seeing small balls when playing games, such as tennis or cricket	1.08	1.10	-0.44	0.23
29	Seeing big moving object, such as bikes passing, in daylight	0.97	0.94	-0.30	-0.79
30	Getting around outdoors e.g. shops or the park, by myself when it's daylight	0.58	0.56	0.53	-0.69
31	Getting around outdoors e.g. shops or the park, by myself when it's dark	1.17	1.10	0.51	-0.79
32	Getting around in crowds by myself	1.11	1.02	0.96	0.85
33	Finding my way around an unfamiliar house or a new building	1.04	0.97	0.62	-0.62
34	Reading signs and posters at stations or shops	0.79	0.70	0	0.07
35	Finding correct money to pay when shopping	1.14	1.17	0.25	0.39
36	Watching films in the cinema	0.83	0.81	0.03	-0.42

Item code	YP-FV item	INFIT MNSQ	OUTFIT MNSQ	DIF contrast by age (logits)	DIF contrast by gender (logits)
37	Watching shows, such as plays, at the theatre	0.99	1.05	-0.13	0.42
39	Using public transport, such as trains, buses or the tube by myself	1.10	1.07	0.41	-0.84
41	Using a mobile phone or tablet for social networking, for example, Facebook, Twitter or MySpace	1.17	1.08	-0.58	0.05

6.4.4.8.2 Targeting

The person-item map using the final 31-item scale is presented in Figure 21 (pg. 269). Concordant with the previous three analyses, the difficulty of the final 31 items was well targeted to the functional ability of the participants. The difference between the person and item means was .03 and separation and reliability values were high when using the real RMSE values (5.40 and .97 respectively).

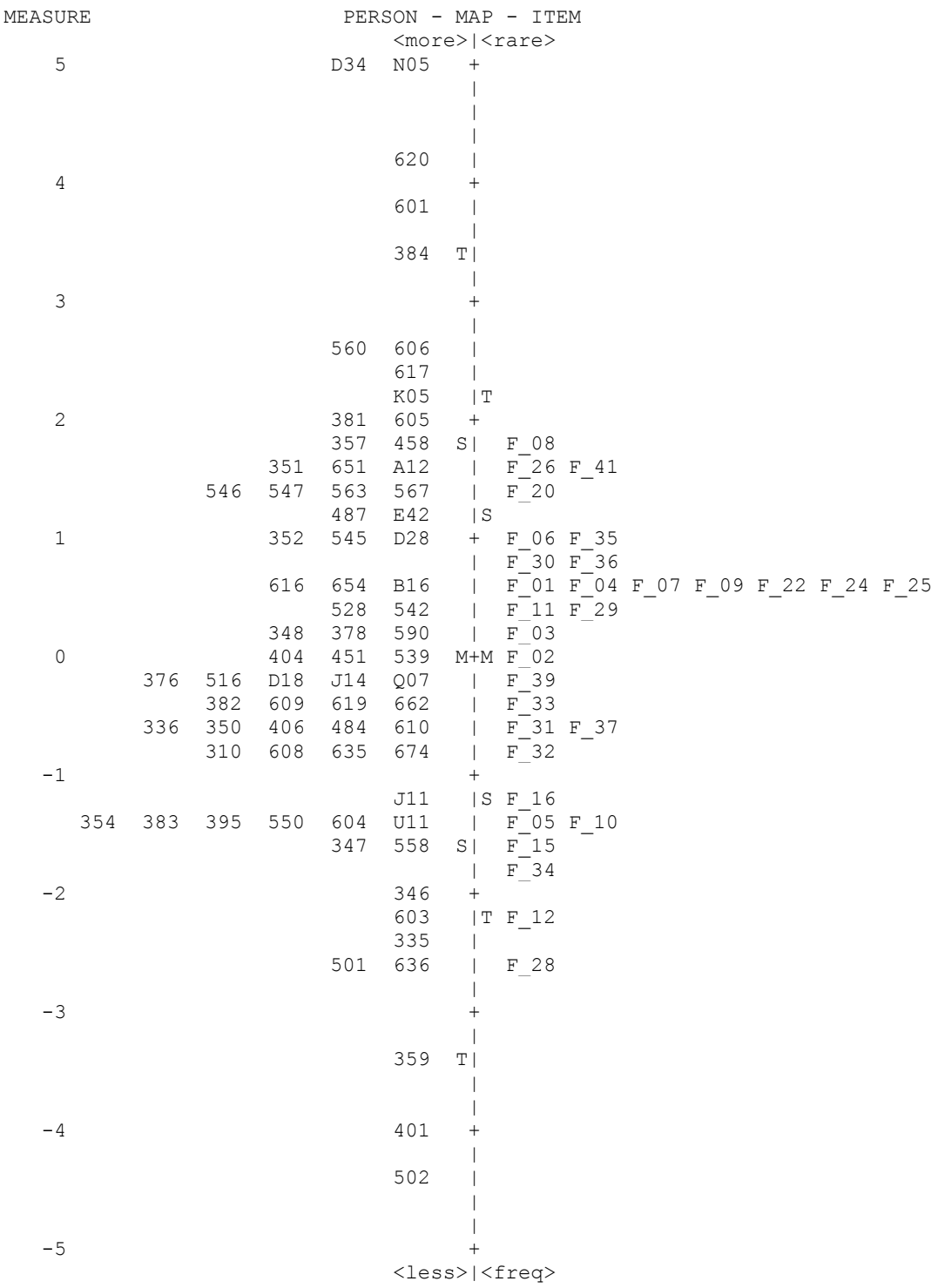


Figure 21. Item-person targeting of the final 31-item YP-FV scale.

6.4.4.8.3 Response category function

Each of the four response categories can be seen to peak, representing a period along the latent trait when each response category is the most likely to be chosen (see Figure 22, pg. 270). The category probability curves are supported by the increase in Andrich

threshold which was ≥2.22 between each response category, indicating that each response category is well defined and likely to be observed in future applications of the instrument.

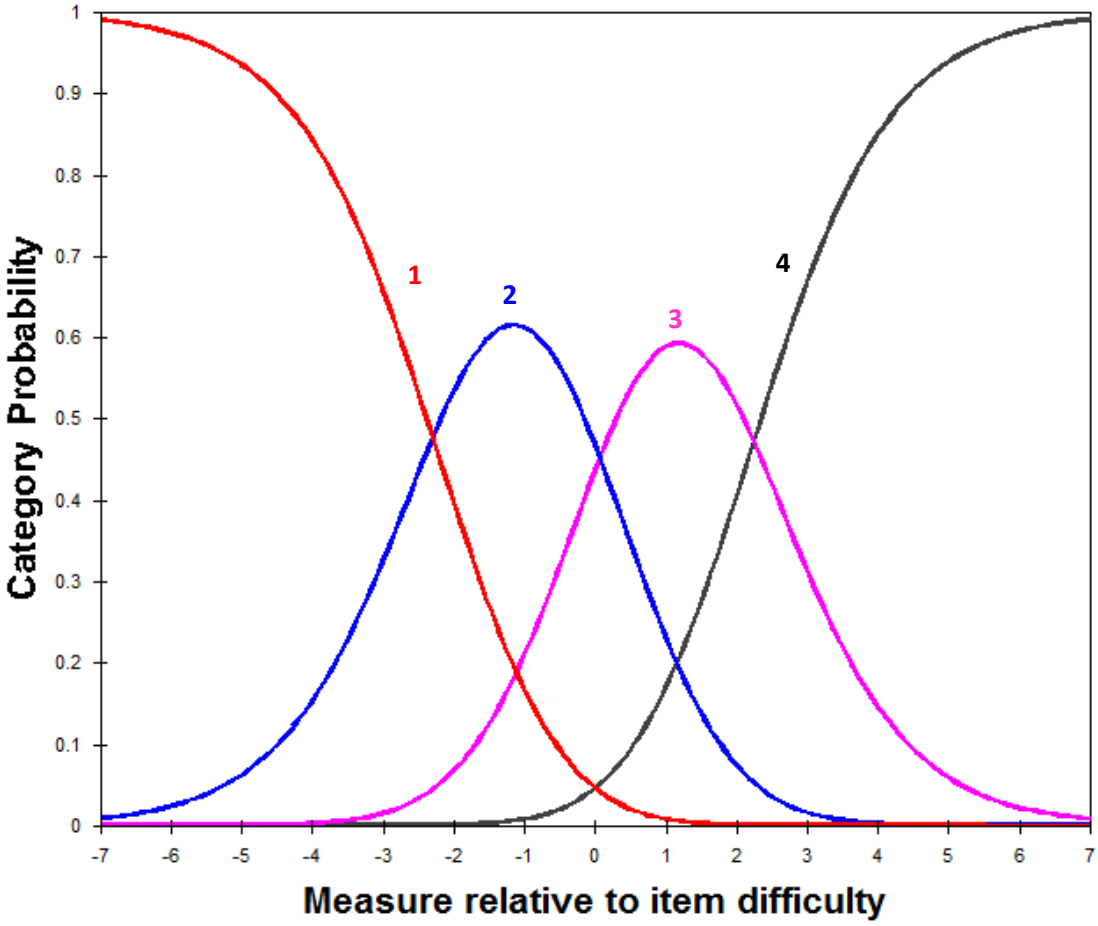


Figure 22. Category probability curves for the 4 response categories in the 31-item YP-FV instrument (1 = Very easy, 2 = Easy, 3 = Difficult, 4 = Very difficult or impossible).

6.4.4.9 Calibration of the Child- and YP-FV instruments

The 29-item Child- and 31-item YP-FV scales were used in the calibration of the instruments. All items were assigned a new item label indicating which instrument they appear in (see Table 44, pg. 271). The sample size of the dataset containing both child and YP responses was *n* = 153.

Table 44. Items included in the final 29-item Child- and 31-Item YP-FV instruments and labels used for calibration of the two instruments.

Child		YP		Item label for calibration
Original item no.	**Label**	**Original item no.**	**Label**	
1	Watching TV	1	Watching TV	1
2	Playing video and computer games	2	Playing video and computer games	2
3	Playing other indoor games, such as board games or card games	3	Playing other indoor games, such as board games or card games	3
4	Playing outdoor games, such as tag or hide and seek			4C
5	Using the computer at home to do my school work	4	Using the computer at home to do my homework/coursework	5
6	Reading small writing such as food packets or instructions for toys	5	Reading food packets, tickets, labels or recipes	6
7	Doing household jobs, for example, tidying up my toys	6	Doing household chores, for example, washing up or tidying my bedroom	7
		7	Looking after my appearance, for example, doing my hair, shaving or putting on make-up	8YP
		8	Making myself a snack at home	9YP
		9	Making myself a meal	10YP
		10	Finding objects I have dropped such as coins or glasses on a low contrast surface	11YP

Child		YP		Item label for calibration
Original item no.	**Label**	**Original item no.**	**Label**	
8	Using the computer in school lessons	11	Using the computer at school or college to do schoolwork/coursework	12
9	Reading small print worksheets and textbooks like dictionaries	12	Reading small print textbooks, worksheets and exam papers	13
10	Reading enlarged worksheets and textbooks like dictionaries			14C
11	Drawing or painting			15C
12	Reading other people's handwriting	15	Reading other people's handwriting	16
13	Seeing the board in the classroom	16	Seeing the board in the classroom when sitting at the front	17
14	Recognising people, for example in school corridors			18C
15	Recognising other people's facial expressions			19C
16	Finding friends in the playground			20C
17	Doing maths in lessons	20	Doing maths	21
18	Doing literacy in lessons			22C
		22	Doing science	23YP
19	Doing PE			24C
20	Keeping up with the teacher in lessons	24	Keeping up with the teacher or tutor in lessons	25

Child		YP		Item label for calibration
Original item no.	**Label**	**Original item no.**	**Label**	
21	Keeping up with other children in lessons	25	Keeping up with other students in lessons	26
22	Getting around the school without someone helping me	26	Getting around school/college by myself	27
23	Playing team sports without special balls			28C
24	Seeing small balls when playing games such as tennis or cricket	28	Seeing small balls when playing games such as tennis or cricket	29
25	Seeing big moving objects, such as bicycles passing by	29	Seeing big moving objects, such as bikes passing, in daylight	30
26	Getting around outdoors in daytime	30	Setting around outdoors e.g. shops or the park, by myself when it's daylight	31
		31	Getting around outdoors e.g. shops or the park, by myself when it's dark	32YP
		32	Getting around in crowds by myself	33YP
		33	Finding my way around an unfamiliar house or a new building	34YP
28	Reading signs and posters at stations or shops	34	Reading signs and posters at stations or shops	35
		35	Finding correct money to pay when shopping	36YP
29	Watching films in the cinema	36	Watching films in the cinema	37

Child		YP		Item label for calibration
Original item no.	**Label**	**Original item no.**	**Label**	
30	Watching shows at the theatre	37	Watching shows, such as plays, at the theatre	38
		39	Using public transport, such as trains, buses or the tube by myself	39YP
		41	Using a mobile phone or tablet for social networking, for example, Facebook, Twitter or MySpace	40YP

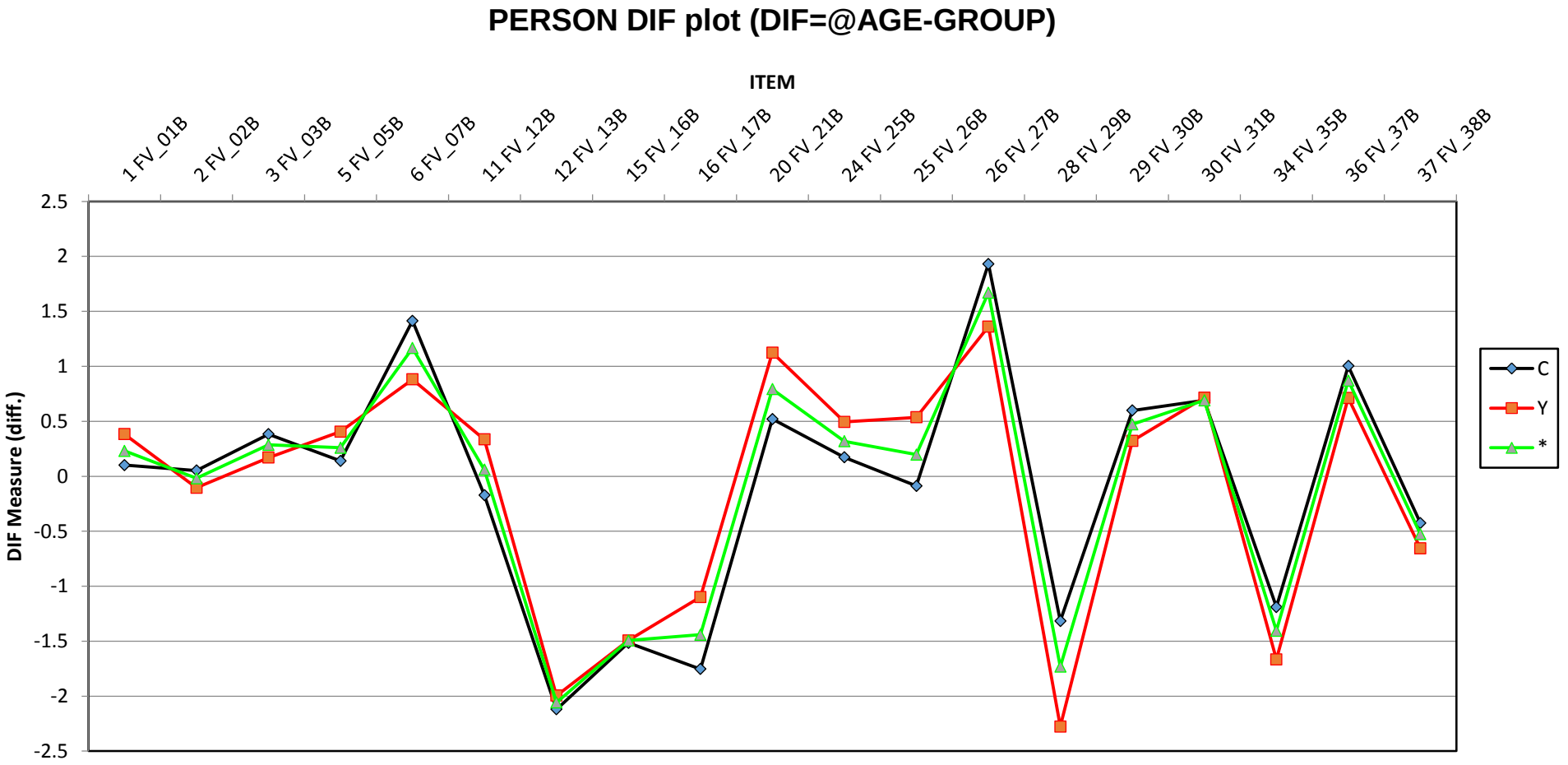
6.4.4.9.1 Item fit to the measurement model

When analysed together, the final 29-item Child-FV and 31-item YP-FV instruments demonstrated good functional ability in terms of fit to the Rasch model of measurement. All item fit statistics were within the range of .61 to 1.44 and all observed average measures for persons were ordered in the right direction in terms of the difficulty of items.

6.4.4.9.2 Differential item functioning

DIF analyses were conducted between the Child (8-12 years) and YP (13-17 years) age groups and on all the 'core' overlapping items shown in Table 44 (pg. 271). Analyses revealed two items (Item 6 (Reading small writing such as food packets or instructions for toys/Reading food packets, tickets, labels or recipes) and 29 (Seeing small balls when playing games, such as tennis or cricket) with DIF contrasts greater than the threshold of 1 and classified as moderate to large.[402] Item 6 was shown to be 1.21 logits more difficult for YP (aged 13-17 years) included in this analysis to perform. In contrast, Item 29 was shown to be 1.02 logits more difficult for the children (aged 8-12 years) to perform. Both items were significant using the Rasch-Welch statistic (t = 4.25, p = <.001 for Item 6, t = -3.65, p =.004 for Item 29). After removing Item 6, the

DIF contrast for Item 29 decreased to an acceptable value of -0.96 and was therefore retained. Figure 23 (pg. 276) shows the DIF contrasts for the remaining ‘core’ items.



*the baseline measure (no DIF)

Figure 23. DIF in overlapping FV items between children (aged 8-12 years) (C) and YP (aged 13-17 years) (Y).

6.4.4.9.3 Fitting FPs to the model

The complete score-to-measure tables for the FV instruments (see Appendix XVIII, pg. 386 and XIX, pg. 387) displayed equations provided by Winsteps software which can be used to convert the raw scores into measure (or logit) scores for both Child (j) and YP (k) versions of the FV instrument.

$$Child\ FV\ Measure = 19.3516 + Score \times .7353 \quad \text{(j)}$$

$$YP\ FV\ Measure = 19.6981 + Score \times .6791 \quad \text{(k)}$$

When fitted to FPs, using a 4th order polynomial trendline, fit of the raw scores and the logit scores to the model was found to improve in both Child (see Figure 24, pg. 278) and YP (see Figure 25, pg. 279) scores. The 4 parameters used are shown in Figure 24 and 25. The equations produced by fitting the trendlines in Figures 24 and 25 are presented for scores of the Child (l) and YP (m) instrument versions.

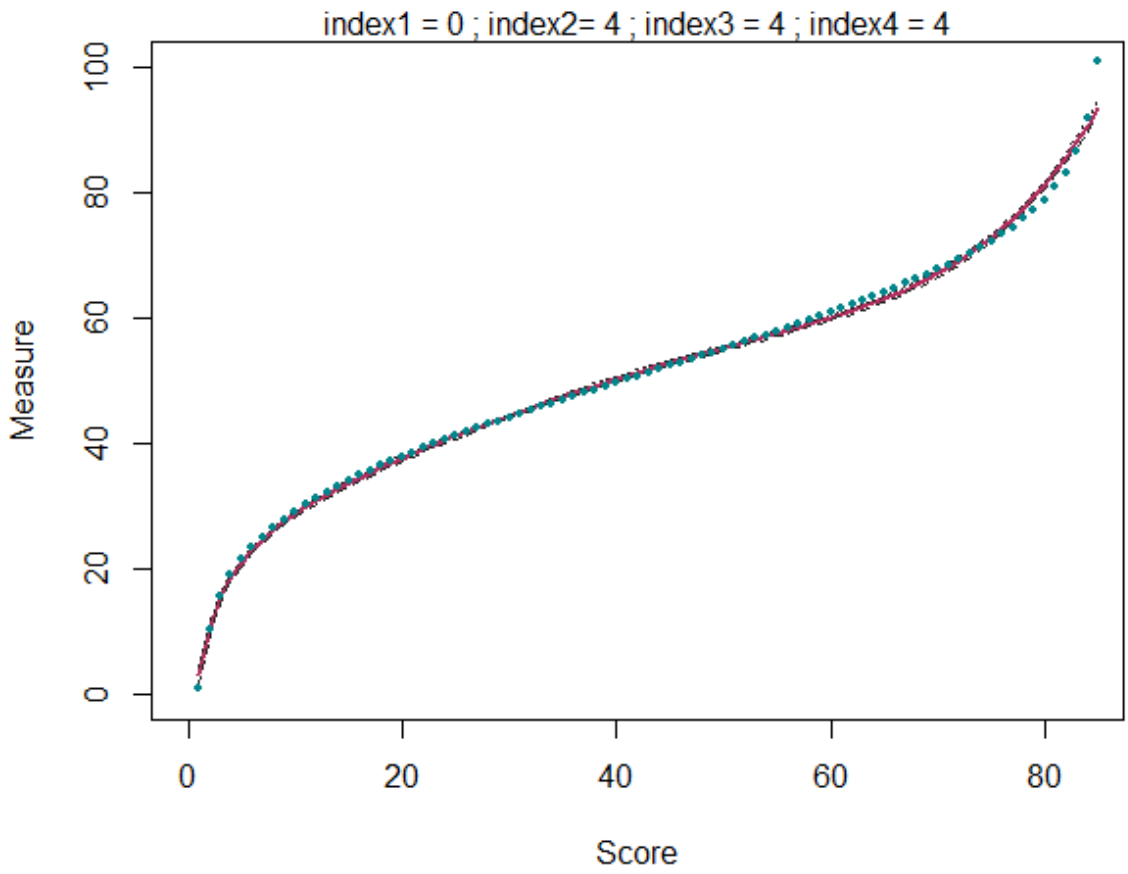


Figure 24. Fit of a 4th order FP trendline to the score-to-measure values in the Child-FV instrument.

$$\begin{aligned} Child\ FV\ Measure &= 2.983 + 11.08 \times \ln(Score) \\ &= 0.00009009 \times Score^4 - 0.00004156 \times Score^4 \times \ln(Score) \\ &+ 0.00000483 \times Score^4 \times \ln(Score) \times \ln(Score) \end{aligned} \quad \text{(l)}$$

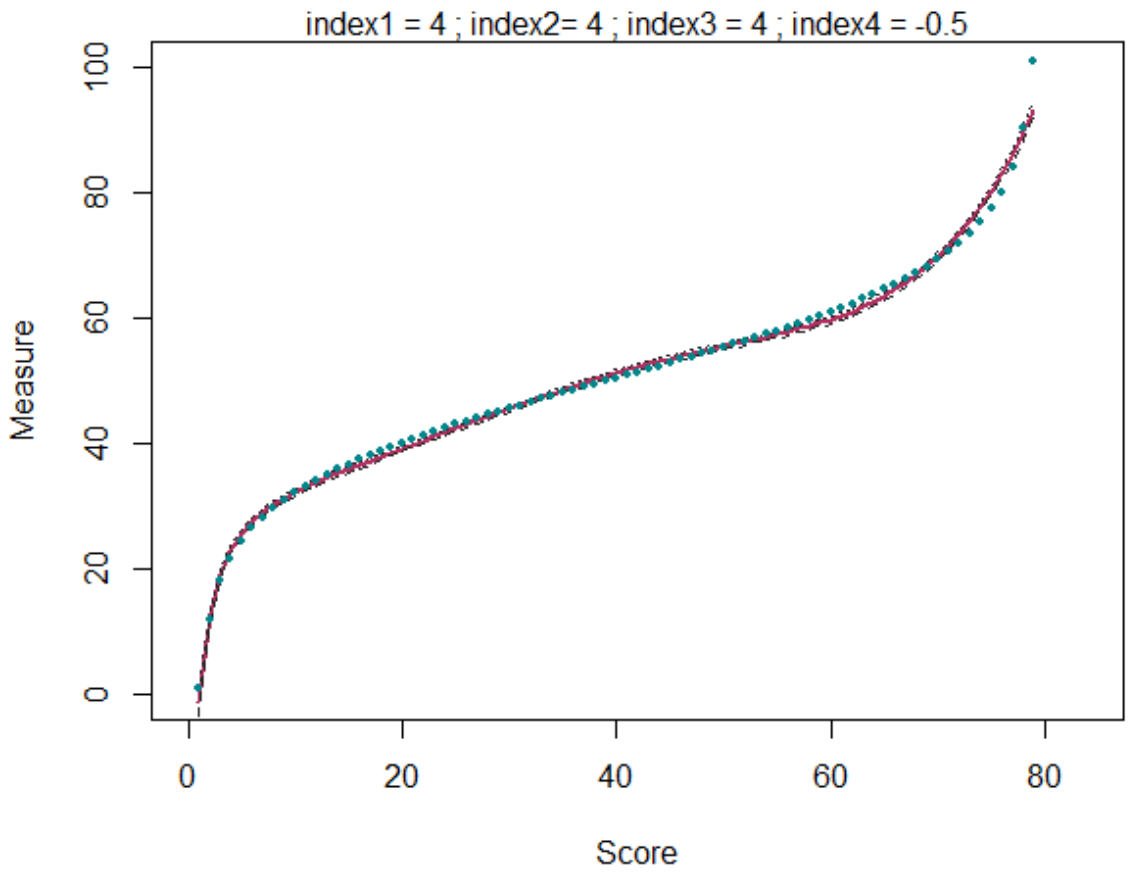


Figure 25. Fit of a 4th order polynomial FP trendline to the score-to-measure values in the YP-FV instrument.

$$YP\ FV\ Measure = 3.042 + 10.87 \times \ln(Score) + 0.000076 \times Score^4 - 0.00003213 \times Score^4 \times \ln(Score) + 0.000003675 \times Score^4 \times \ln(Score) \times \ln(Score) \quad (m)$$

Although the FP model was found to improve upon the linear equation produced automatically by Winsteps, it was also found to inaccurately predict scores at the extreme end of the scale when manipulated to fit the scale of 0-100. Thus, the optimal method for converting raw scores into measure scores which can be compared between age-groups is to use the score-to-measure table produced by Winsteps (see Appendix XVIII, pg. 386 and XIX, pg. 387).

6.4.4.10 Testing construct validity

6.4.4.10.1 Demonstrating normality in the logit scores

Out of a total of 18 variables (comprising VA and FV logit scores for children (aged 8-12 years), YP (aged 13-17 years) and the entire sample combined), 13 were calculated as non-normally distributed judged using the criteria described in Section 6.3.4.10.1 (pg. 224). As a result, Spearman's Rank[401] statistics were used to evaluate correlations between all variables.

6.4.4.10.2 Correlation between FV summary scores and VA

When analysed separately, both the Child- and YP-FV logit scores based on the measures correlated positively with participants' VA (r_s = .49, z = 4.37, (one-tailed) for Child scores and r_s = .42, z = 3.53, (one-tailed) for YP), indicating construct validity of the individual Child- and YP-FV instruments.

A correlation was also found when analysing the relationship between FV logit scores and VA when combining the logit scores of both children and YP. This relationship was significant (r_s = .44, z = 5.42, (one-tailed)), signalling that higher levels of VA are associated with lower FV summary scores based on the measure and demonstrating construct validity of the entire suite of core and age-specific items (see Table 45, below).

Table 45. Correlation coefficients comparing FV logit scores and VA for children and YP aged 8-17 years (one-tailed).

	FV logit score	**VA**
FV logit score	1	.44 (5.42)
VA	153	1

6.4.4.11 Final assessment of unidimensionality

Following Rasch analysis, the 28-item Child-FV and 30-item YP-FV instruments were entered into a final FA to assess unidimensionality. Notably, the percent of variance explained by the first (largest) factor increased in the YP-FV instrument after psychometric item reduction had been performed, demonstrating improvement in the measurement of FV as one construct. However, in the Child-FV instrument, the percent of variance explained decreased marginally: from 44.95 (see Table 38, pg. 252) to 44.69 (see Table 46, below).

Table 46. Eigenvalues for the largest factor, percent of variance explained, variation between eigenvalues for datasets with imputed missing data, number of eigenvalues and items not loading onto factors.

	Eigenvalue for the first (largest) factor (percent of variance explained)	**Variation between eigenvalues for the first (largest) factor in imputed datasets**	**No. of eigenvalues >1**	**No. of items loading onto Factor 1 <.4**	**Items not loading positively onto the first (largest) factor**
Child-FV	12.51 (44.69)	0.17	5.2	0	None
YP-FV	16.28 (54.28)	0.14	6	0	None

6.5 Directions for use of instruments

To ensure the developed instruments can be readily applied to both research and clinical ophthalmology contexts, we have developed guidelines for clinical practitioners and other health professionals. Specifically, a higher summary and measure score on the child- and YP-VQoL instruments denotes higher VQoL, and a higher summary and measure score on both FV instruments denotes a higher level of functional difficulty. Both instruments have been developed so that scores across the entire questionnaire must be added to make up a valid and reliable summary score.

If missing responses are found, it is important that researchers and clinicians explore the reason for missing responses (e.g. by addressing any instances where a respondent has written notes on the instrument, or indicated that some items are not relevant or very difficult to answer). In the context of research, if responses are found to be missing at random, we suggest they are coded using an improbable value (e.g. 999) and estimated or imputed using statistical techniques such as the ones we have used (see Section 4.9.4.2, pg. 115).[378, 379] If used in a clinical setting, the available scores may be used to derive a pro-rated summary score (i.e. by replacing the missing scores for a given patient with their observed mean score on other items on the scale). Specifically, this may be done using the following formula:

Where **S** = the observed summary score,

L = the number of items with complete responses (i.e. with scores not missing) and,

M = the number of items with missing responses:

$$\frac{S}{L-M} \times M \qquad \text{(n)}$$

And adding the resulting value to the summary score. The decimal-score point in the pro-rated summary score may be interpolated for a more accurate estimation. However, we recommend, for ease and consistency, that the final summary score is rounded to the closest integer. This may be used to locate the corresponding measure score when using Child- and YP- instruments simultaneously.

6.6 Summary

Following development which is grounded in the perspectives of children and YP living with visual impairment in the UK, and analysis using robust psychometric methods, the final Child-VQoL, YP-VQoL, Child-FV and YP-FV comprise 24, 26, 28 and 30 items respectively, and may be suitable for application within clinical contexts to capture the self-reported impact of visual impairment during childhood and adolescence. The final Child- and YP-version of each instrument are also valid for use sequentially or in parallel, rendering the instruments suitable for measuring the impact of visual impairment longitudinally throughout the course of childhood and adolescence (i.e. throughout the age range of 8-17 years) or across a population with age-range of 8-17 years, whilst maintaining an age-appropriate approach and capturing age-specific issues and challenges.

Chapter 7 Discussion

Key findings, strengths and limitations of the study are presented in this chapter. The self-reported experience of visual impairment, from the perspectives of children and YP is discussed in relation to the format and implementation of the final instruments and in relation to existing generic- and vision-specific PROMs. Results from both streams of the project (i.e. qualitative investigation and instrument development) are considered in relation to the extant generic literature and the implications for policy and practice within paediatric ophthalmology services in the UK are discussed.

7.1 Strengths

To our knowledge, this study is the first in-depth exploration of the experience of visual impairment during childhood and adolescence applied to the development of an age-appropriate suite of PROMs designed to capture the impact of visual impairment during everyday life. Using a pre-established methodological framework, the instrument development has been consistently grounded in the perspectives of children and YP living with visual impairment. This renders the project an important methodological advance on previous efforts to develop vision-specific PROMs for which input from children and YP has often been supplemented or replaced by input from adults living, or working with children and YP.[261, 265, 266, 271, 274, 278, 282, 285, 286] During each stage of development, we took precautions to encourage independent self-report from participants, by interviewing them directly, engaging them in expert consultations, and providing materials designed for parents and guardians alongside those designed for children and YP. These attempts are reflected in the content and format of the finalised child- and YP-centred instruments.

7.1.1 Instrument calibration

The majority of existing vision-specific PROMs designed for children and YP are designed for use by individuals within a broad age-range i.e. 8-18 years[213, 268-270, 276, 283,

[284, 403-405] and therefore are not sensitive to aspects of developmental change and capturing the impact of issues and challenges which are age-specific. The defining feature of the current development is that two complementary age-appropriate suites of PROMs have been developed for children aged 8-12 years and YP aged 13-17 years. Because these age-appropriate PROMs were developed in parallel, within the same research team and using the same theoretical and methodological approach, they can be used collaboratively within clinical practice to capture the impact of visual impairment across the age-range of 8-17 years, whilst maintaining specificity to the developmental nuances of particular age groups. The method of development we used is comparable to the Wolfe and Chiu approach[406] in that we have ensured that the shared, or overlapping, items in each instrument are stable in terms of measurement function i.e. we have developed a common scale calibration. To our knowledge, this approach has been applied in the past to the development of a PROM designed to capture the impact of spectacle correction on refractive error (i.e. pre- and post-intervention),[261] but not in relation to the calibration of age-specific instruments as we have done.

7.1.2 Age-range of users of the developed PROMs

The finalised instruments were developed for children aged older than 8 years specifically, based on a) our findings from the early stages of instrument development for children younger than 10 years which will not be discussed in detail in this thesis and b) literature stemming from cognitive development and ability to self-report during childhood.[137] This decision echoes the fact that the majority of existing child-centred, vision-specific PROMs have a minimum age-threshold of 8 years: namely the EYE-Q,[276, 403] IVI_C,[213, 268] LVP-FVQ,[269, 404] LVP-FVQ II[270] and the PREP[283, 284, 405] (see Table 2, pg. 54). As previously described (see Section 2.5.1, pg. 42), varying degrees of developmental delay are often reported in samples of children living with visual impairment. Therefore, we would suggest a fluid approach to administering the developed instruments within clinical practice to children within the age-thresholds we

have defined, as some children aged over 8 years may experience difficulty conceptualising some items. Additionally, some children younger than 8 years may be cognitive capable of completing the instruments in a meaningful way.

To date, only one existing vision-specific PROM specifies 17 years as the upper age threshold of respondents,[279, 281, 407] with the majority extending this to 18 years. Our decision to specify 17 years as the upper age threshold is, however, well validated by our findings from the early phases of instrument development. We found that the self-reported impact of visual impairment in relation to everyday life was prone to change after participants had reached their 18th birthday. Changes were often a result of YP entering further education or formal employment and triggered new vision-specific challenges which were difficult to capture using the same questions as those used to capture aspects of VQoL in younger children. For example, asking YP to differentiate between their home life and education was difficult when YP had moved away from their family home, and were living in student accommodation at a college or university. Similarly, YP experienced difficulty answering questions and probes relating to specific aspects of education (such as seeing the board in the classroom) after they had transitioned to further education. However, consistent with our recommendations for the minimum age threshold, we would also recommend flexibility in the implementation of the maximum age threshold when the developed instruments are used within clinical practice and that practitioners consider the contextual circumstances of the patient. This is in keeping with the 'stage'-appropriate approach YP emphasised in our investigation of the experience of transition from paediatric- to adult-ophthalmology services.

7.1.3 Psychometric properties of the final instruments

As previously discussed (see Section 3.3, pg. 73), Rasch analysis can be used for instrument development to construct linearity by fitting the data (in this case the instrument items) to the model of measurement. This is done whilst considering the difficulty of each item in relation to the ability of respondents. Using Rasch analysis,

therefore, we can be sure the final instruments are capable of additive measurement. Calibration of the two age-specific instruments using the method we used confirms that the principles of additive measurement are met when using instruments independently, as well as collaboratively to compare outcomes of individuals at different stages in the trajectory of childhood. This is an important methodological advance on earlier developments of vision-specific PROMs, for which FA has been widely implemented.

The early, exploratory phases confirmed content validity of the final instruments as children and YP had an active role in contributing to the individual items. Efforts were made to incorporate elements of participants' speech in the item development and several items were worded verbatim based on spontaneous speech from children and YP. Qualitative analysis ensured the wording of each item was optimal to capture the underlying theme or experience. Thus, content and face validity in each instrument can be confirmed.

Using Rasch analysis, we were able to validate the instruments in terms of a) item fit, b) targeting of persons to items, c) item-category thresholds, d) response scale functioning and e) item bias, and, in doing so, provide a detailed description of how each instrument functions in relation to the outcomes it is designed to measure as well as the ability of the sample.

7.2 Limitations

7.2.1 Participation rate and sample size

Participation rate of YP living with visual impairment was low (≈30%) throughout the study, but is consistent with previous studies recruiting children and YP living with visual impairment.[5, 240] This was expected at the outset of the study and we addressed this by sampling patients cautiously throughout the 4 phases, inviting only those needed to fulfil the requirements of analyses, and preserving the majority of the sample for the final phase of validation. Efforts were made throughout the study to a) locate all

YP living with visual impairment in England, Ireland and Wales using recruitment from multiple sources, b) appeal to YP and their families using age-appropriate recruitment materials, and c) contact patients at multiple times, whilst sending reminder and confirmation letters. The resulting sample size of the first three phases of development was large enough to fulfil the requirements of the qualitative and early quantitative analyses and represented children and YP with a range of demographic characteristics, living in different communities and with different manifestations of visual impairment. In-depth interviews and expert consultations produced informative and detailed qualitative data, suggesting participants were eager to share their experience of visual impairment and happy to take part in the research study.

Although the participation rate of YP was consistent throughout the phases of development, the obtained sample size of the dataset in the final phase of development was particularly small in light of the planned psychometric development. A relatively small sample size was also attained in the parallel development of instruments for children. This posed a number of difficulties when validating each instrument using Factor and Rasch analyses. With regard to FA, the obtained sample size was well below the advised 'rule of thumb' ratio of at least 10-15 participants per variable.[408] This may have increased the errors in correlation coefficients and hence factor loadings, suggesting a greater contribution of individual items to factors. However, the importance of applying this 'rule of thumb' has been disputed as some authors have found that changes in the ratio of participants to variables make little difference to the stability of the factor solution.[409, 410] In this study, we used FA for the sole purpose of screening the datasets for unidimensionality prior to Rasch analysis, and thus, applied criteria to the first, most dominant factor in each analysis. In the FV instruments, all items loaded well onto the dominant factor, and it was only in the VQoL instruments that we observed some items which did not load heavily onto the dominant factor. Thus it is likely that an increase in sample size would have resulted in stronger evidence to

support unidimensionality in the child- and YP-VQoL instruments rather than alternative findings per se.

With regards to Rasch analysis, it is recommended that sample size should equal at least 10 observations in each response category for each item to ensure that the step calibration between response options is stable.[317] The sample size of the individual child and YP datasets which were used to analyse the final response scale functioning would have been large enough to fulfil this criterion if there had been an even spread of responses. However, in each instrument, responses to some items were skewed, with the majority of participants choosing responses at one end of the scale. This reflected minor variation in participants' well-being and ability at the time of recruitment. The likely impact of this skew would manifest itself in the strength of step calibrations between response options, rendering these less precise and stable than they might have been in a larger scale of development with less skewness. As with any other statistical analyses, the small sample size may have also produced less precise estimates, less powerful fit statistics, and less robust estimates which are sensitive to extreme responses.[316] However, it should be noted that the development of age-appropriate instruments in parallel revealed some consistency in the functioning of response categories between two independent groups and provides some evidence to support the final response scale in this development.

Findings regarding the overall participation rate in the current study has implications for the feasibility of future studies recruiting children and YP with complex health conditions, such as visual impairment, and performing psychometric analyses such as Rasch analysis. For future studies, we would suggest that sample size could be improved by recruiting children and YP during school holidays or other anticipated leisure times. This strategy was impractical to implement in the current study given the time frame of the project but we suggest this may be beneficial for future studies, particularly since many potential participants with more severe manifestations of visual impairment we contacted were attending specialist schools away from their family

homes, and were therefore uncontactable. Given that approximately one third of children and YP living in the UK with visual impairment attend specialist schools for pupils with learning, physical and/or sensory impairments,[411] future studies could address this by gaining support from visual impairment specialist residential institutions for children and YP at the outset. However, variation in the functional, emotional and psychosocial outcomes of children and YP attending these institutions in comparison to those attending mainstream, non-specialist education must be considered.

Contact details of potential participants were collected using electronic patient management systems at the two primary recruitment centres. These systems were not suitable for systematically searching patient records, and this had to be done manually throughout the project and was extremely time-consuming. One of the primary recruitment centres was a children's hospital and may have been the first institution involved in the specialist ophthalmic care of the majority of children and YP who have visual impairment onset before the age of 16 years. Thus the contact details of potential participants aged 13 years and older at the time of the project may have been out-of-date. This was particularly likely for individuals who no longer required routine specialist follow-up care and had early onset visual impairment which was non-progressive. Although the recruitment sources restricted the number of patients we were able to contact, we were able to recruit a good distribution of patients with varying manifestations of visual impairment (i.e. including those with early and late onset visual impairment which was stable and progressive in nature of deterioration). Given that some instrument items were taken from instruments developed in the foundation research which also recruited a good, representative sample of children and YP[6, 7] we are confident that instrument items are meaningful and valid for application in the population of children and YP living with visual impairment in the UK.

A final factor influencing the small sample size used in the current study is the restriction of the study sample to children and YP with visual impairment and no other significant physical, sensory or learning impairments. This was a unique aspect of our

study, and essential to achieve a focus on visual impairment *per se*, whilst ensuring participants were cognitively capable of reflecting on the impact of visual impairment. However, we acknowledge that the majority (77%) of children and YP living with visual impairment which is classified as severe in the UK do have additional sensory, physical or psychological impairments,[412] and were excluded from the sampling framework. Thus, recruitment of children and YP with visual impairment may be more successful in studies which do not require the same focus on visual impairment.

7.2.1.1 Predictors of participation: Index of multiple deprivation and severity of visual impairment

Participation analysis revealed that higher socioeconomic status was associated with a greater likelihood of participation in any of the four phases of instrument development. This finding is consistent with literature demonstrating similar participation trends in children aged 10-15 years[413] and the families of younger children living with a visual impairment[414] who were recruited through the primary recruitment site(s) used in the current study. However, severity of visual impairment was also found to predict participation in the current study: those with worse vision were less likely to participate. Thus, despite efforts to ensure all research materials were accessible e.g. by providing alternative electronic formats and contacting families by telephone to discuss the requirements of participation, it is possible that some YP living with blindness experienced some response burden,[415, 416] and perceived the activities involved in the latter phases of development (i.e. completing consent forms and questionnaire booklets) as too difficult and/or time consuming.

In contrast with literature demonstrating participation trends in similar samples[413, 414] ethnicity was not found to predict participation in the current study. Throughout the study, we recruited YP with a range of ethnic backgrounds. However, only a minority of participants came from Black Caribbean/African/other, 'Other', and 'Mixed' backgrounds. Because ethnicity was found to significantly predict participation in the early, univariate analyses, it is hypothesised that a larger sample size (and wider

distribution of participants with a range of ethnic backgrounds) would have diluted this effect.

Determining the impact of socioeconomic status (SES) upon outcomes in the current study is complicated because there are multiple levels of IMD influences (e.g. community, neighbourhood, and family) which can either be transitory or persistent. However, to date, research has demonstrated an association between SES and cognitive development emerging early in life course.[417] Family income has been shown to be a powerful correlate of IQ at the age of 5 years, and neighbourhood income differences may be determinant of IQ and externalising behaviour.[418] Living in a high-SES neighbourhood has positive benefits for school readiness and achievement.[419] With regards to health, research demonstrating the dynamic impact of SES upon outcomes during childhood[420] has shown disparities in access to resources such as nutrition,[421] cognitively stimulating experiences, and healthcare.[422] Evidence also shows that children from lower SES groups are less likely to see an eye-care specialist.[423, 424] However, we tried to negate any kind of bias caused by the participation trend in the current study (i.e. individuals with lower socioeconomic deprivation being more likely to participate) by grounding the instrument development in the perspectives of YP with a range of socioeconomic and ethnic backgrounds, and manifestations of visual impairment, and using qualitative techniques and expert consensus versus statistical analyses. We are therefore confident that the final instrument items are valid and reliable in terms of measurement precision for the national population of children and YP living with visual impairment.

7.2.2 Influence of others upon self-report

Interviews conducted with YP were lengthy (lasting on average over 1 hour), and the majority of YP were extremely willing to discuss their experience of visual impairment. A one-to-one context in which attempts were made to exclude other family members enhanced perceptions of confidentiality, allowing participants to feel secure and open to describing their true feelings to the researcher. Efforts were also made to ensure

participants would be capable, in terms of visual function, to participate independently, particularly in the latter stages of development when they were required to complete printed questionnaire booklets. We encouraged independent self-report by providing parents and caregivers their own questionnaire booklet to report on their perceptions of their child/young person's VQoL or FV. However, it became apparent in the early phases of development, that it was not always possible to exclude parents, guardians, siblings or other family members from influencing YP's self-reported impact of visual impairment. At times, parents encouraged other family members, such as siblings to attend and contribute to interviews which were conducted with children and YP. Thus, presence of others may have limited the content of the developed instrument items and some items detecting more sensitive components of VQoL or FV may have been omitted.

The dominating role of parents also emerged in a number of everyday domains which were explored in this study: in the way YP formed future ambitions and goals, developed and asserted independence, and perceived the benefits of forming romantic relationships, and can be likened to traditional models of disability in which disabled children are seen as somewhat incapable of communicating or performing activities themselves (see Section 2.2, pg. 26). In the past, research exploring the role of parents in the broader context of childhood disability has linked parents' responses to their children's symptoms to children/YP's own experiences of pain. Incorporating elements of social learning theory[425, 426] such as positive and negative reinforcement, this perspective explains how protective, or dominating parenting can inadvertently result in an increase in the child/YP's self-reported symptoms and functional disability.[427-431] Research has yet to explore this association in the context of visually impaired children and adolescents, however, given that visual impairment is a sensory impairment associated with reduced ability to complete activities independently, it may be that children/YP's perceptions of their functional impairment rely, somewhat, on the responses of parents, or significant others, who view the child/YP as incapable.

Findings from the current study mirror those in a study exploring peer and adult relationships in adolescents with restricted mobility[432] in which adolescents perceived their parents as controlling, and an obstacle to their attainment of independence. Participants described their relationships with parents as asymmetrical where only the parents were involved in decision-making processes.[432]

7.3 The final suite of vision-specific PROMs

One aim of the current project was to apply the knowledge gained through the exploratory, qualitative investigation as to the impact of visual impairment during childhood and adolescence to develop a suite of age-appropriate, vision-specific measurement instruments.

7.3.1 The Child (8-12 years) and YP (13-17 years) versions of the Vision-Related Quality of Life (VQoL) instrument

The finalised child- and YP-centred VQoL instruments contain 24 items and 26 items respectively and are valid for use with children aged 8-12 years and YP aged 13-17 years. The instruments contain 17 overlapping items, 7 items designed specifically for children and 9 items designed for YP. These items span five of the six domains of VQoL specified in the foundation research: 1) social relationships, acceptance and participation, 2) independence and autonomy, 3) psychological and emotional well-being, 4) future – aspirations and fears and 5) functioning – school, home and leisure.[5] Items corresponding to the sixth domain: treatment of eye condition, were excluded in the early stages of development based on the premise that these items would be better suited for a patient-reported experience measure (PREM) and one item related to the YP's concerns that their eyesight might deteriorate (I worry my eyesight will get worse) was excluded during Rasch analysis because a gender bias was detected. Thus, the final instruments cover all domains of VQoL which were expected, and are appropriate, for inclusion in the PROMs. Items relating to the 5th component of VQoL (functioning –

school, home and leisure) were worded carefully and with emphasis on a broader social context, rendering them appropriate for inclusion in the VQoL instrument, as distinct from the FV instrument which focuses on a level of difficulty in performing a particular activity. For example, five items (I feel tired because of my eyesight, I have to work harder at school/college because of my eyesight, I can do most activities on my own, and I can get around on my own) were designed to pick up on the underlying psychosocial burden of visual impairment and effort to complete tasks, as distinct from reality of functional difficulty.

7.3.1.1 Definition of VQoL (as defined by children and YP with visual impairment)

In our study, factors defining VQoL in children and YP were largely similar, comprising values, goals and concerns, with emphasis on social environment and other people's perceptions and understanding of visual impairment. These findings can be described in relation to broader, well-supported definitions of QoL which emphasise the role of experience in constructing a mental representation of current QoL, such as that provided by the WHO Quality of Life Group (see Section 2.3, pg. 28). However, we also found that some aspects of VQoL were age-specific. For example, it is only during adolescence that participants in this study explained concerns about the impact of visual impairment in the future and the consequences this could have upon their future lifestyle and, specifically, what kind of job they might be able to do. Whilst all participants described the impact of their impairment upon their social life, age-specific VQoL items developed for children emphasise social inclusion and fairness, which hold parallel to social models of disability,[433, 434] showing individuals' awareness of social discrimination related to their disability emerging at a young age.

7.3.1.2 Comparison to existing vision-specific PROMs designed for children and YP

The number of items in both the final child- and YP-centred VQoL instruments are comparable to the number of items in existing vision-specific PROMs designed for children which range from 3[272, 435] to 39.[259] In our experience, the numbers of items in PROMs need to reflect a comprehensive coverage of the measurement construct, whilst considering the condition-specific needs of the intended users, such as response burden. During the second phase of development, some participants described the burden of having to complete lengthy self-report measures, particularly since this traditionally takes place within clinical contexts which are often perceived as time-consuming and arduous.[436, 437] Thus, we would conclude that the number of items in each of the developed instruments is optimal for capturing outcomes whilst minimising effort needed from respondents.

With regards to format, the final VQoL instrument (comprising statements for which respondents are required to indicate as true or not true) is comparable to three existing vision-specific PROMs designed for children: namely the IXTQ,[279, 281, 407] SREEQ,[261] and PREP,[283, 284, 405] and two vision-specific PROMs designed for parents i.e. proxy reports (the CVFQ[265, 266] and ATI[271]). With regards to response options, the VQoL instruments feature a 4 Likert-type response scale which is in keeping with the average number of response options featured in existing vision-specific PROMs designed for children (ranging from 2 – 6 response options). Three existing vision-specific PROMs designed for children include a 'not applicable' response option[213, 258, 268, 271]: a feature that a number of YP who took part in the current development favoured. Despite this, we decided to exclude a 5th 'not applicable' response option based on the premise that this would involve adding the response option to the corresponding child instrument, and may exceed the cognitive capacity required of children and YP to complete the final instruments in the absence of full functional vision.[137] We also made efforts throughout the development to include only items which are meaningful and relevant to

children and YP of varying ages. Inclusion of a 'not applicable' response option would confound this, indicating to users of the instruments that not all items may be applicable and/or important. Additionally, a 'not applicable' response option was implemented in the foundation research and led to some methodological difficulties, potentially confounding participants' endorsement of the 'Very difficult or impossible' response category preceding it.[6]

In keeping with the format of the final VQoL instruments, and participants' tendency to refer to factors other than visual impairment, such as personality when answering items, one existing vision-specific PROM designed for children (the IVI_C)[213, 268] contains a statement which is comparable to the one we have implemented with the aim of reminding participants that 'The questions are all about how things are for you because of your eyesight'.

7.3.1.3 Construct validity of the VQoL instrument(s)

We correlated participants' logit scores in each VQoL instrument to other outcomes: namely VA, PedsQL summary scores, and PedsQL psychosocial summary scores with a view to determine convergent validity as a form of criterion validity. Analysis of associations between novel instruments and existing generic instruments, such as the PedsQL is an approach used widely throughout existing developments of novel PROMs and applied to a range of patient populations including children born preterm,[438] living with physical disabilities such as cerebral palsy[439] and cardiac disease,[440] and to measurement of personality-based constructs such as self-stigmatization.[441] Consistent with the majority of existing developments, and as hypothesised, VQoL scores were significantly associated with PedsQL summary and sub-scale scores, indicating that the instruments capture similar constructs. Additionally, and in keeping with findings from the foundation research,[7] VQoL was not associated with VA and therefore we provide further evidence of the disability paradox[88] (see Section 2.3, pg. 28) in the sample. This finding points towards the utility of developing two complementary PROMs distinguishing between aspects of VQoL

which may not be related to each other and/or traditional objective outcomes such as VA.

7.3.2 The Child (8-12 years) and YP (13-17 years) versions of the Functional Vision (FV) instrument

The child- and YP-FV instruments contain 28 and 30 items respectively, of which 19 were overlapping. The final version of the Child-FV instrument contains 9 age-appropriate items, and the YP-FV instrument contains 11 age-appropriate items.

Each item in the final FV instruments is expressed as a verb phrase in the statement 'Because of my eyesight I find…' This format is comparable to that of one existing instrument (the CVAQC)[262] which was designed to capture visual ability and includes a reminder for participants to consider their ability in relation to their best corrected visual function. Our development of FV age-appropriate instruments advances upon the CVAQC in terms of sensitivity of the developed items to age-appropriate activities, such as playing in the playground, getting around outdoors when it's dark, and looking after physical appearances, which were found to be meaningful to only one age-group of children and YP.

7.3.2.1 Definition of FV (as defined by children and YP with visual impairment)

Unlike definitions of QoL, HRQoL and VQoL which are controversial but nevertheless receive empirical attention, FV is yet to be formally defined by extant literature. However, there is a broad understanding that FV occupies the gap between person-outcomes and symptom-outcomes as specified by the ICF model. Thus, FV can be defined as a self-reported evaluation of ability to complete meaningful daily activities in real everyday environments, incorporating elements of VA and broader physical and social surroundings. In this development, FV items which were developed for children largely span activities which are performed at home and in school. Age-appropriate items developed for the YP-version refer to activities which are performed

independently, and reflect a primary outcome from the in-depth exploratory phase of the project: emergence of independence and autonomy during adolescence.

7.3.2.2 Construct validity of the FV instrument(s)

When correlated with VA; an objective clinical measurement of visual function, participants' logit scores on the finalised child- and YP-FV instruments were positively associated, demonstrating good convergent validity in the final instruments, and indicating some consistency between objective and subjective assessments of visual function.

7.4 Experience of visual impairment during childhood and adolescence: Key findings in relation to existing knowledge

Use of in-depth, semi-structured interviews in the early stages of instrument development in this study afforded the opportunity to explore and understand children and YP's experience of growing up with a visual impairment, and thus capture novel data. To date, there has been limited investigation of children and adolescents' lived *experience* of visual impairment, with most accounts focused on describing the everyday difficulties or challenges, and using cross-sectional approaches. This form of research has shown that children and adolescents living with visual impairment want to be independent,[231] and perceive not being able to drive as a significant barrier to this,[212, 231] are worried about the future[231] and may experience embarrassment and isolation in social contexts.[213] We have extended this evidence base to identify when age- and vision-specific challenges and changes were experienced, and how children and YP overcame these.

We found that children develop and refine a number of coping strategies which are used to negate the negative impact of visual impairment in varying contexts, and when faced with varying challenges. Existing literature aimed at identifying the range and effectiveness of coping strategies used by individuals with visual impairment is largely

restricted to adult populations[442-444] and cannot be readily applied to children and YP who have different developmental capacities and needs and, more importantly, grow up without sight as opposed to having had vision earlier. Where literature focusing on children or YP does exist, it is centred upon capturing the burden of caring for children and YP with visual impairment upon their parents and families,[445-448] and ignores the voices of children and YP themselves. To date, one empirical account examines coping strategies developed by YP aged 16-24 years with congenital visual impairment in relation to resisting the impact of visual impairment.[449] Findings are somewhat comparable to the coping strategies identified in the current study. For example, advocacy which was described by authors as 'actions of disempowered people to achieve justice, equity or inclusion'[447(p196)] can be compared to times in the current study when participants described their attempts to prove others wrong about their ability to participate in physical activities, be included in social activities, and develop friendships in which they were viewed as equal to their sighted peers. Similarly, aspects of 'passing' i.e. minimising the impact of visual impairment by refusing to use aids or identify as disabled, were discussed by some participants in their use of humour as an attempt to avoid the negative social stigma related to visual impairment.

A pertinent finding from the early, qualitative stages of instrument development was variation between the experience of those with early onset and late onset visual impairment, particularly with regard to acceptance of and adaptation to visual impairment. This finding supports the 'early timing hypothesis'[450] in the context of childhood visual impairment as those who experienced the impact of visual impairment early in childhood, appeared to better able to cope (both psychologically and functionally) with their impairment at a later stage. To date, existing literature examining differences between individuals with early and late onset visual impairment is centred upon identifying neuroanatomical differences and, in particular, sensitive or critical periods for visual input in light of cortical plasticity.[451, 452] Our findings extend the current evidence base, demonstrating differences in the subjective, self-reported experience of

visual impairment among those with varying manifestations of vision disorders. In the future, these findings can be combined using mixed-method studies which explore possible interactions between physiological (i.e. neuroanatomical) and psychological (i.e. self-reported) impacts of visual impairment during childhood and use techniques such as fMRI imaging alongside self-report. Findings may be used to clarify the psychosocial and emotional impact of visual impairment which manifests in late childhood and early adolescence.

7.4.1 Age- and vision-specific transitions and changes during childhood and early adolescence

A number of formal and informal transitions during childhood and early adolescence were described by participants living with visual impairment and often in relation to intrinsic changes in personality, attitudes, and beliefs, such as emerging perceptions of responsibility, and increased awareness of other people's reactions to impairment and the stigma associated with disability. Transitions which took place within education were described as particularly memorable and meaningful in terms of social contexts, as friendship groups were established, and adults were increasingly appreciated as role models. This finding is supported by literature investigating shifts in social identity among healthy adolescents, showing that social identity effects occur alongside experiences of change.[453] The school to adult-world transition has been viewed as a process involving changes in roles (i.e. socially expected behaviour or patterns of meaningful activities that are expected of individuals in various contexts[454]). These changes are thought to trigger further changes in how adolescents make sense of themselves and their world,[455, 456] suggesting that the changes children and YP described in the current study would have long-term effects upon identity as being visually impaired or disabled.

Whilst many of the transitions YP described were normative, and likely to be experienced by all YP regardless of visual impairment, participants often described

vision-specific challenges, rendering the impact of visual impairment fluid and dynamic during childhood and adolescence. These findings speak to the value of longitudinal use of the developed instruments within paediatric ophthalmology. It may be valuable to monitor changes in the impact of visual impairment which are triggered by transitions in education, for example, as changes to a new educational environment may alter the daily impact of visual impairment, in addition to deeper perceptions about the future. Several YP who took part in the early phases of instrument development described transitioning from specialist to mainstream (or vice versa) schools or colleges, and the changes they experienced in terms of physical environments, vision-specific support and social surroundings. Again, these findings speak to targeting vision-specific support services and treatments to children and YP who are most in need of intervention, which can be done by administering the developed PROMs at critical time points during childhood and adolescence, and in contexts outside of paediatric ophthalmology settings, such as education.

7.4.2 Transition from paediatric- to adult-ophthalmology services: Implications for clinical practice

One transition we took the opportunity to explore in detail was the transition from paediatric to adult ophthalmology services. We designed the age threshold for participation at the outset of the current study to reflect the gap in VQoL and FV instrument provision for YP older than 15 years. However, we also noted that recruitment of YP aged 16-19 years in interviews would be optimal for exploring participants' experience of the transition from paediatric to adult ophthalmology services, since participants were on the conventional transition age-threshold. The importance of ensuring a timely and successful transition from child- to adult services is recognised internationally,[396-398] particularly for those who progress into adulthood with rare childhood onset conditions and complex health needs.[457] A growing literature has identified the impact of a timely and successful transition in terms of secure disease-related knowledge,[458] high self-efficacy and good confidence for self-management of

health,[459] and assessed the effectiveness of technology-based systems and methods such as use of the internet and mobile phones to provide YP with access to transition-related information and increased control over their transition.[459-461] Despite this understanding, there is currently very scant literature to inform transition planning and provision in ophthalmology.[399]

YP who took part in this stage of development described pertinent differences between clinical settings which subsequently impacted their feelings of being in control of their healthcare and confidence to manage their care independently in the future. Emphasis was placed upon provisions which are patient-centred when transitioning from paediatric to adult-centred care, and findings largely support application of the generic NHS transition guidelines[396] to ophthalmology services. The value YP placed upon an appropriate peer group being served by the same services confirms the key importance of considering 'stage' as opposed to 'age' in timing of transition to ensure it occurs after the developmental tasks of adolescence have been completed.[462] Transition that takes place too early in this trajectory risks feelings of insecurity in the new environment.[399]

In 2015, 74.7% (5.2 of 7.07 million) of out-patient appointments in adult ophthalmology services in the UK were attended by patients over the age of 50 years. This is in contrast to 10.5% (764,400), which were attended by infants and children aged 0-15 years and a mere 0.9% (65,200), which were attended by YP aged 16-19 years.[463] Given this skewed age distribution and the specific needs of YP, it is arguable that transition from paediatric ophthalmology should ideally be into specialist adolescent/young adult services.[399] Models of this provision exist in other areas of child health, for example, endocrinology services for the late sequelae of childhood cancer,[464] which have promising patient-reported outcomes.[465] This would address the challenges of the 'no-man's land' that lies between child and adult ophthalmology services.[399]

Good and consistent communication between patients and their families and their managing clinicians lies at the heart of effective paediatric ophthalmology services.

Implementation of vision-specific, age-appropriate PROMs may constitute a means in which to enhance age-appropriate communication, specifically when YP are at either side of the age-threshold of transition in ophthalmology services, by ensuring clinicians address the factors which are most important to patients during clinical consultations. Consistent use of the age-appropriate PROMs within the latter stages of paediatric, and early stages of adult-ophthalmology services may address patients' perceptions of clinicians' knowledge about their specific needs: a factor that we found to influence patients' perceptions of less effective communication after transitioning.

7.6 Application of the finalised PROMs

The value of measuring the impact of a disability or illness over time is reflected in the number of clinical trials which have implemented PROMs in the past with a view to assess the impact of health-related interventions or treatments within a wide range of clinical specialities.[116, 118, 466-469] Most long-term follow-up studies using PROMs have administered the same instruments at different time points with the aim of ensuring measurements are reliable and that change in outcomes reflect true change in the construct of interest.[470-472] However, when children are recruited as participants in these studies, this approach may be conceptually problematic in that fluctuations in the experience of disability or impairment may be related to growth and development, and changing attitudes, values and priorities, as opposed to the intervention of interest. In a review of measures of HRQoL suitable for long-term follow-up in children who have experienced major trauma,[473] authors identified only 3 out of 14 HRQoL measures that met the criteria for psychometric validation and were also suitable for an age-range of at least 10 years: the DISABKIDS,[474, 475] KIDSCREEN 52[476, 477] and PedsQL.[478] Authors speculate that, because the number of items and scoring systems in different age-versions of the instruments are similar, different versions of these instruments can be considered as one measure. Indeed, there are two versions of the DISABKIDS instrument (suitable for children aged 4-7 years and 8-16 years) and four versions of

the PedsQL (suitable for parent proxy report for children aged 2-4 years, and self-report for children aged 5-7 years, 8-12 years, and 13-18 years) and the format of response options and scoring systems are similar in each age-version. However, unlike the approach used in the work reported in this thesis, neither of these instruments were developed using psychometric analyses to validate the function of each age-version independently, and in collaboration with the alternative age-versions. Justification of the suitability for different ages is based on subtle changes in terminology, for example in the PedsQL the term 'kid' is modified to 'teen' and in the DISABKIDS, the Likert scale is presented as series of smiley faces. To our knowledge, the KIDSCREEN, which was described by authors of this review[473] as suitable for an age-range of 10 years, is not available in age-specific formats, but instead short- and long-versions comprising 27 and 52 items respectively, which are both described as suitable for children and YP aged from 8-18 years. Therefore, it is likely that the majority of existing PROMs designed to capture HRQoL among children and YP omit important age-specific contributors to QoL,[479-481] particularly since definitions of QoL emphasise life experience (see Section 7.3.1.1, pg. 295). Despite this, we chose to administer the child (8-12 years) and teen (13-17 years) versions of the PedsQL in the current study because it has been validated, and used widely in a range of paediatric health conditions.[482, 483]

7.7 The role of PROMs within the current NHS: Policy and practice

PROMs have been developed and used within clinical practice over the last 30 years but are increasingly recognised as valuable tools with potential to trigger changes in clinical practice.[123] The current NHS emphasises a patient-centred approach[484] and PROMs can provide valuable insight for provider trusts and clinical commissioning groups and for individual clinicians through their ability to evaluate the impact of clinical interventions upon outcomes which are traditionally non-quantifiable. In parallel to the emergence of psychometric understanding and application using varying statistical

approaches, we have now reached a stage where it is possible to quantify complex psychologically grounded concepts, such as QoL and, in doing so, enable clinical practitioners and trusts to develop patient-centred approaches. PROMs play an important role in NHS England's Five Year Forward View objective which was put forth in 2014 and emphasises patient empowerment through allowing them greater access to information, and greater control over their healthcare.[485] The patients' organisation, National Voice, describes how important it is for statutory services to recognise patients' own life goals, wellbeing and independence as the first steps in developing personalised care,[486] and thereby emphasises the role of PROMs in fulfilling the future goals of the NHS.

Despite an increase in life expectancy over time,[95] and a population which is becoming increasingly healthier, demand for convenient, effective and available NHS services is continuing to increase. The impact of this is largely financial and may be explained partly by the cost of caring for elderly patients.[487] At the same time, funding throughout the NHS has reduced dramatically.[488] A framework for understanding the different ways in which access to high-quality care can be limited by commissioners and providers emphasises six types of rationing: deflection, delay, denial, selection, deterrence and dilution.[489] Rationing by dilution, specifically, refers to the process in which budgets are cut, resources are spread thinly, and the quality of healthcare is reduced. Dilution in the current NHS climate is evidenced by staff shortages[490] which can have negative impacts on patients who may experience greater delays, increased cancellations and appointments needing to be rescheduled as a result.[489, 491, 492] Currently, in NHS adult ophthalmology services an alarming number of patients appear to be losing their sight needlessly due to delayed diagnosis which is caused by capacity problems,[493] such as those associated with rationing by dilution. Although this finding is yet to be applied to paediatric services, it is clear that financial demand and capacity problems in ophthalmology services in the NHS are having a negative impact upon patient outcomes. In these contexts, PROMs can also be used to evaluate quality and

consistency of healthcare over time: outcomes which reflect the impact of budget cuts upon patient-reported outcomes, and the NHS's goals of prioritising patient empowerment.

In the current political climate, the NHS is facing unprecedented financial and operational pressures.[488] Despite an increase of around 8000 new doctors and nurses working in the NHS since 2014,[494] there are some gaps in availability of specialist professionals. In a recent survey, only 52% of current NHS staff self-reported satisfaction with opportunities for flexible working and 36.7% report feeling unwell as a result of work-related stress.[495] Because PROMs can be used flexibly, administered in waiting rooms or at patients' family homes prior to consultations, and come in multiple, cost-effective formats, it is possible that PROMs constitute an affordable means to alleviate some of the demands placed upon NHS staff, by reducing the need for lengthy consultations, and allowing paediatric ophthalmologists to capture, quickly and accurately, patients' primary concerns. Additionally, caring for children and YP who are losing their eyesight is a challenging, and at times emotionally-fuelled task.[496, 497] Conversations can be particularly distressing when they involve discussions about possible future deterioration of vision. The vision-specific PROMs we have developed provide a platform in which clinicians may breach sensitive topics within clinical consultations, and the first step in understanding patients' individual needs.

As previously discussed (see Section 7.2.1, pg. 287), children and YP living with visual impairment and an additional associated non-ophthalmic disorder were excluded from the current development, based on theoretical considerations. However, the prevalence of additional, non-ophthalmic conditions speak to the implementation of the developed PROMs within clinical practice, as children and YP visiting ophthalmic services may likely also attend a range of alternative specialist clinics. The extant generic child health literature indicates that transitions from paediatric to adult services are particularly successful when YP's health needs are predominantly due to one condition,[498] and that successful transition for those with a number of comorbid health

conditions relies on excellent communication and organisation between caregivers, specialities/departments and institutions.[499] Thus, the developed instruments may be used to enhance consistent communication between clinical teams, promoting multidisciplinary approaches to the treatment of childhood visual impairment.

7.8 Summary

We suggest that, following further empirical investigation into feasibility of routine implementation, the developed PROMs should be implemented into routine clinical practice, using an approach which takes account of patients' developmental 'stage' (versus 'age'). Routine application of the instruments at a nationwide level can result in benefits for both patients and clinical professionals, allowing effective and timely targeting of vision-specific interventions, whilst reducing the workload of clinical professionals and addressing economic factors prioritised by higher level commissioning bodies. Having explored the dynamic impact of visual impairment throughout childhood and early adolescence, we recommend that, at a minimum, the instruments are used with patients on either side of the age threshold of transitions which take place within ophthalmology services, but to optimise clinical outcomes, are used regularly as patients progress through childhood and adolescence, and face a number of age-related transitions.

Chapter 8 Future Work

As previously discussed (see Section 2.3.2, pg. 31 and Section 7.7, pg. 305), use of PROMs can facilitate patient-centeredness: a focus of several NHS frameworks. Patient-reported outcomes such as VQoL and FV may be collected and fed back to care providers longitudinally to track patient's outcomes over time, evaluate whether treatments are effective and facilitate treatment modifications as needed.[500]

At a higher level of practices, policies and guidelines, PROMs may improve patient care via two pathways. Firstly, a 'change' pathway in which feedback from patients encourages practices to take steps to change clinical care, and secondly a 'selection' pathway, whereby patients and higher organizations such as commissioning and regulatory bodies choose high-performing providers over low-performing providers.[501-503]

Because a large proportion of children and YP living with visual impairment in the UK also suffer from additional sensory, physical or psychological problems (see Section 7.2.1.1, pg. 291), and in keeping with our recommendations about the timing of transition from paediatric- to adult-ophthalmology services (see Section 7.6, pg. 304) we advocate a 'stage' versus 'age'-approach to implementing the developed instruments with children/YP of varying ages, and that clinicians consider patients' living conditions and developmental capacity to self-report when selecting a PROM. In relation to children and YP living with visual impairment, the 'one size fits all' methodological model which currently prevails in the context of PROM administration may not be optimal.[504] There is also a need to assess the feasibility of using PROMs within routine paediatric ophthalmology services.

8.1 Preferences and attitudes towards using PROMs

There is a difference between collecting data using PROMs as part of a scientific study (as we have done) and as part of routine clinical care. In our study, efforts were made

to engage with children and YP and their families throughout the 4 phases of instrument development, emphasising the value of the research project, using age-appropriate materials, and ongoing communication and contact with patients and their families. However, in routine clinical practice, PROMs must be administered and results collected, scored and interpreted (often prior to doctor-patient interaction) without disrupting the clinical workflow. Successful implementation of PROMs relies on excellent co-operation between patients, their parents/guardians, and clinicians. Assessing these stakeholders' preferences is integral to integrating the developed PROMs into routine practice.

8.1.1 Patient preferences

Patients need to be happy to complete PROMs for them to be successfully incorporated into routine ophthalmology services. Where it does exist, research focused on children and YP's perceptions of using PROMs is largely confined to mental health settings and the extent to which this can be extrapolated to children/YP living with visual impairment is unclear. However, in the past, children and YP aged 8-19 years visiting mental health services have described their support for PROMs, and preference to complete written measures versus talking to a clinician about difficult issues.[505] However, the main objection to using PROMs in this context was patients' perceptions of the ability of instruments to reflect complex and dynamic outcomes such as well-being. For example, some participants described day-to-day fluctuations in outcomes and concerns that this would make self-reporting impossible. Many participants expressed concerns about the practicality of using PROMs within mental health services, indicating that they wanted to be better informed about, and have more control over, the assessment process so that they were comfortable with the timing of the data collection and presentation of results.

In the UK, specifically, YP aged 9-17 years visiting mental health and diabetes services were shown to be worried that, using PROMs, the scope of clinical discussions may be widened, and outcomes may attract unwarranted professional attention to emotional

issues.[506] Many YP found questions to be personal, and felt that the information should be used only with discretion. Some had worries about the confidentiality of their self-report. In these cases, YP did not want information divulged outside of their immediate clinical team.

Findings from these studies point towards the impact of patients' preferences upon obtaining valid and reliable outcomes. If a child/YP has a negative attitude to completing a PROM, they may become tired or bored and, as a result, skip certain questions. Within paediatric ophthalmology settings, specifically, children and YP may be required to have a number of assessments/procedures conducted at each outpatient visit (e.g. imaging which involves pupil dilation) which may affect their ability and willingness to complete the instruments at particular time points during their clinic visits.

In the current study, YP described parents/carers as playing an active role in managing clinical care and, at times, dominating discussions during consultations. The perceived need for parents/carers to dominate within clinical contexts was often magnified by the functional difficulties YP described when attending hospital visits independently and navigating their way around clinical environments, or accessing written information, meaning that they were unable to attend hospital appointments without a parent or carer accompanying them.

Input from parents during clinical consultations when a patient is under the age of 15 years is required to some degree for legal reasons,[507] but may affect children/YP's self-reported outcomes, particularly since the VQoL instruments we designed contain some items which reflect complex psychological states such as coping and life satisfaction. In a study investigating the views of adolescents living with mobility restrictions,[432] participants raised concerns that they might come into conflict with their parents if they did not openly express their appreciation and satisfaction with the help they receive from them. Adolescence is frequently described as a time of heightened awareness of the perceptions of others[21, 508-510] and children/YP living with visual impairment who

took part in the current study described their increasing concerns about the reactions of other people (e.g. siblings, parents, and peers) towards their impairment. Thus, it may be the case that, as children develop into adolescence, it is increasingly difficult to capture true self-report which is not influenced by the presence of others when completing or, specifically, patients' perceptions about how they *should* (versus *want*) to respond.

Consideration must therefore be given to the setting, presence of others, and method in which the developed instruments are administered in clinical ophthalmology contexts, especially if, even after transitioning from paediatric to adult-centred services, YP are likely to be accompanied to their clinical visits, and may have few opportunities for confidential disclosure with clinical professionals.

8.1.2 Parent preferences

At the level of family members such as parents/guardians, implementing PROMs within paediatric ophthalmology services may serve some benefits, namely that responses from child self-report can provide insight as to the subjective impact of visual impairment, and the specific problems their child might be facing.[505] However, parents of children with mental health issues have been shown to be worried of professional scrutiny of the quality of care they are providing for their child when completing PROMs.[506] Parents may also be concerned that PROMs are too restrictive[505] and can replace (versus complement) important discussions about clinical assessments.[506]

The items included in the final suite of vision-specific PROMs we have developed are designed to reflect children/YP's self-reported difficulties, challenges and daily struggles in light of living with visual impairment. Thus, users of the instruments are required to reflect on some sensitive, psychological issues and it is possible that completion of the PROMs may cause some degree of distress for some children/YP. If it is the case that parents/guardians of these users do not understand the value of patient-reported outcomes, or how PROMs will be used within clinical contexts, and

perceive instrument completion to be potentially distressing, they will likely object to their child completing the instrument. Given that consent from someone with parental responsibility is essential before a child under the age of 16 is treated within clinical contexts,[507] it is imperative to promote parent/guardians' perceptions of the utility of PROMs, with emphasis on positive patient outcomes, to ensure they can be implemented successfully in routine ophthalmology services.

8.1.3 Clinician preferences

Clinicians may be reluctant to use PROMs if they do not perceive the data as credible or understand the application to improving quality of care.[503] Extant generic literature reflects findings from adult healthcare settings and populations, and may not be readily applicable to paediatric ophthalmology contexts. However, as a foundation, some considerations may be made.

Consultants may be worried that implementing PROMs will lead to an increase in workload associated with collecting and analysing data, and communicating outcomes to patients.[511-514] This attitude may be reinforced by perceptions that mandatory public reporting programmes initiated by regulators or national governments are driven solely by political motives and do not impact upon routine clinical practice or patient outcomes.[503] Clinicians have also raised concerns that they do not understand the clinical importance of the data collected, and what to do when validity of the measurement is compromised i.e. when participants did not complete the measures accurately.[515] Preferences have been shown to parallel those of parents, as some clinicians have expressed concerns that PROMs may constrain the patient-clinician relationship because they trivialise patients' emotions.[503]

However, in a RCT evaluating improvements in individual patient care in adult oncology services in the UK,[516] PROMs were shown to lead to improvements in patient-doctor communication, with increased discussion of non-specific chronic symptoms. Implementing PROMs in this trial did not significantly increase the duration of

consultations, but instead led to greater patient engagement and empowerment through improved communication about aspects of health which are rated as important by patients. Thus, there may be some dissonance between the evidence-base and the preferences/attitudes of healthcare professionals.

If clinicians maintain negative attitudes towards implementing PROMs within routine clinical practice, or have concerns about their relevance or impact, it is possible that instrument validity may be compromised, and incorrect data captured. For example, clinicians have been found, in the past, to adapt PROMs to make them more suitable for use with individual patients.[503] Although we advocate a 'one size does not fit all' approach,[504] making changes by altering the PROMs to suit individual patients will compromise validity of the data captured.

8.2 Promoting positive attitudes to using PROMs within paediatric ophthalmology contexts

Common barriers towards implementing PROMs within routine clinical practice largely stem from lack of understanding on behalf of all stakeholders as to the utility of PROMs, the confidential nature of the data collected, and the potential benefits at the levels of individuals (i.e. patients and clinicians). Thus, education for both patients and their parents/guardians about how patient-reported outcomes are going to be used and who is going to look at the results is an important first step towards promoting positive attitudes towards using the developed PROMs.

In the past, scientific attention has focused upon improving health literacy: the ability to read, understand and act upon health information, with an aim to empower patients to take control of their healthcare.[517] Patient information materials (printed and electronic) have emerged as practical methods to enhance patients' knowledge and understanding of their condition.[518] The positive impact of these materials has been shown to be greater when information is personalized,[519-521] well targeted to patients

and available at the right time.[522] Therefore, it is possible that patient information materials comprising leaflets, information booklets, or online resources may be used to increase patients' and their parents' understanding about why PROMs are important outcomes. However, the best method to do this is yet to be explored within paediatric ophthalmology contexts and may be complicated by factors such as the nature of visual impairment and developmental delay in cognitive ability.

In the past, 2 potential misuses of PROMs have been demonstrated at the level of clinical professionals: in using PROMs to evaluate the value of treatment/intervention upon individual patient outcomes *before* administering it, and considering only short-term outcomes when deciding on competing demands between funding for treatments.[103] Therefore, education is needed to promote awareness among clinicians of the benefits and clinical value (as discussed in Section 8.1.3, pg. 313), and psychometric properties of PROMs.[523]

An intervention incorporating educational, epidemiological, marketing, behavioural, organisational, coercive, and social interaction approaches, involving a multidisciplinary training team, evidence based support for the values of PROMs, familiarization and interaction prior to use, and decision-support[524] has been applied to changing practice within paediatric clinical contexts. PROMs were administered online and paediatricians received background knowledge about the PROMs and their potential benefits prior to data collection.[525] This study highlighted the value of consultants observing (via DVD) others using PROMs, and the positive effect of evidence upon ideas of how to use patient-reported outcomes, and incorporate PROMs into routine clinical care.[526] If the 'one size does not fit all' approach[504] we advocate is used as a foundation for developing ophthalmic-specific guidelines, studies assessing the feasibility of using this type of intervention are needed, as learning through observing others, and empirical evidence-based ideas may be complicated when patients' physical and developmental capacities vary.

8.3 Electronic methods to administer PROMs

In the past, a number of theoretical benefits have been associated with implementing PROMs administered using electronic formats, such as smartphones, tablets and touch screens within generic adult settings.[527] Not only are electronic methods financially optimal (in comparison with pen-and-paper and postal methods which require printing, processes of data entry and postage)[528] but they can be completed by patients in flexible locations without adversely affecting clinic workflow or visit length.[516, 529] Using electronic methods, data can also be entered automatically, and outcomes can be assessed quickly. Fewer missing data have been reported when using electronic (versus pen-and-paper) methods, thereby improving measurement accuracy.[530-532] Objective and subjective outcomes may be integrated into one system, and visual representations, such as graphs, can allow professionals to compare outcomes quickly and accurately.

Another benefit of using electronic methods of administration is the potential for real-time reporting and alerts.[529] Systems capable of real-time reporting have been developed within the field of adult oncology services,[533] and allow patients to enter and monitor their own outcomes and generate longitudinal reports which can be made available to staff. When applied to paediatric ophthalmology contexts, real-time reporting may provide early warning signs about potentially concerning issues and be particularly valuable given the progressive nature of some forms of visual impairment. Integration of PROMs into routine electronic health records could also allow practitioners to identify patients who need to complete PROMs, who can be contacted ahead of their clinic appointment and asked to complete PROMs at home before they come to clinics, and therefore reduce the administrative burden of having to complete PROMs within clinical settings.[534] A method to alert clinicians of issues requiring immediate attention may also be incorporated, and provide information directing clinicians to targeted resources (e.g. teachers or support workers) available to address any self-reported problems which may be resolved outside of clinical settings.[535]

The quantity and quality of electronic software designed specifically for users with visual impairment has expanded in the recent years, comprising screen readers, video magnifiers and note-takers.[536] To date, some studies have evaluated the feasibility of administering electronic questionnaires among child populations, showing that electronic methods are feasible, reliable and valid.[537-539] However, if these methods are going to be integrated into routine ophthalmology practice then software needs to be programmed to allow children and YP living with visual impairment to access the instruments, understand page layouts, save their data when sessions are interrupted and move through items.

Although this approach has immense potential to improve the administration and completion of PROMs within paediatric ophthalmology settings, which are located within an NHS framework which emphasises better use of data and technology in the next 2 years,[540-542] there is currently limited understanding as to the preferences of children/YP living with visual impairment, and their vision-related ability to use electronic methods. In the final phase of the current study, all participants were given the choice to complete the developed instruments using either pen-and-paper or electronic formats designed specifically for individuals with reduced visual function. However, only two YP chose to complete the electronic versions. Thus, electronic methods of PROM administration need to be designed carefully, and with respect to children/YP's preferences and a range of complex and dynamic functional difficulties.

8.3.1 Combining auditory and visual techniques: A feasible method of administering PROMs?

In 1997, Scott and colleagues[543] discussed the feasibility of using auditory devices such as Walkmans to ensure children could respond to questionnaires without needing help from others, and have their answers remain confidential. In that study, Walkmans were shown to have two advantages: no one other than the respondent could hear the questions being asked and, because the answer booklet contained only response

options, no one could tell which answers corresponded to which questions. Additionally, users were able to control the questionnaire administration process by pausing or rewinding the audio tape, and subsequently avoid the potential embarrassment of having to ask an interviewer to repeat certain questions. Although research has yet to explore the suitability of this technique for children/YP living with restricted functional vision, a combination of similar auditory and visual methods may be optimal for capturing true self-report in light of the potential influence of others whilst completing PROMs (see Section 8.1.1, pg. 310).

However, there are functional-related issues which need to be investigated before the Walkman technique can be implemented within paediatric ophthalmology settings, including what voice type, volume, and speed of reading is optimal. One difficulty of administering instruments using Walkmans was children's literal interpretation of questions when read aloud by an automated voice, as some participants tried to guess the state of the script reader rather than consider the items in relation to themselves.[543] This may pose difficulties among the population of children and YP living with visual impairment, especially when completing items included in the finalised FV instruments which are formatted as simple verb phrases. Thus, further multidisciplinary investigation combining input from information technology, paediatric ophthalmology and psychology specialities is required to develop sophisticated, user-friendly devices which can be used for PROM administration and completion.

8.4 Capacity of services to address psychologically-related outcomes

Implementing PROMs within paediatric ophthalmology practices speaks to ensuring vision-specific services are able to address outcomes. Although the primary aim of implementing PROMs is to evaluate the quality of ophthalmic care and treatment, it is likely that, using PROMs as a platform in which to self-report, some children/YP may report that they are experiencing profound psychological/social/emotional problems

associated with their visual impairment, such as depression or anxiety. Using the developed instruments, respondents may report information which they are reluctant to communicate in other ways. This places some responsibility on those gathering, and analysing outcomes, to identify any responses indicating some concern, and how these should be dealt with.

In the past, clinicians have raised concerns that the information gathered using PROMs can interfere with treatment, forcing discussions into areas which are not clinically relevant and for which clinicians have little control over.[123, 503] Therefore it is important to ensure services are in place to respond to outcomes, and alleviate some of the burden placed upon ophthalmic consultants to address aspects of health which may be perceived to be beyond their clinical expertise. The population of children and YP living with visual impairment is complex in terms of its health-related needs and, as a result, may be receiving support from a number of vision-specific services within education, social, and mobility contexts. The capacity of these services for addressing patient-reported outcomes needs to be evaluated (and promoted) before a shared model of decision making (a component of the NHS Five Year Forward View [485]) can be developed.

8.5 Summary

Combining the findings from this study with the future work outlined in this chapter would allow for incorporation of the developed instruments within paediatric ophthalmology clinical services throughout the UK to measure, with accuracy, sensitivity, and meaning, the subjective, self-reported impact of visual impairment during childhood and adolescence. Integration of the instruments into routine ophthalmology services within the NHS would be the first step to incorporating the instruments within a range of support services, including education and social support services, encouraging a multidisciplinary approach to supporting children and YP living with visual impairment, whilst ensuring consistency between institutions and clinical

teams. Once this is complete it will be possible to systematically develop, and evaluate the impact of, a range of clinical and/or non-clinical vision-specific interventions, and target specifically what matters most to children and YP living with visual impairment.

Chapter 9 References

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Chapter 10 Appendices

10.1 Appendix I – External NHS Trusts included as patient identification centres (PICs) in phase 3 and phase 4

East Lancashire Hospitals NHS Trust

University Hospital Southampton NHS Foundation Trust

West Suffolk NHS Foundation Trust

Royal Cornwall Hospitals NHS Trust

Hampshire Hospitals NHS Foundation Trust

Countess of Chester Hospital NHS Foundation Trust

Hinchingbrooke Health Care NHS Trust

Mid Cheshire Hospitals NHS Foundation Trust

University Hospitals of North Midlands NHS Trust

The Queen Elizabeth Hospital, Kings Lynn, NHS Foundation Trust

Royal Devon & Exeter NHS Foundation Trust

University Hospital of Wales, Cardiff and Vale University Health Board

Birmingham Children's Hospital NHS Foundation Trust

Epsom and St Helier University Hospitals NHS Trust

Leeds Teaching Hospitals NHS Trust

Portsmouth Hospitals NHS Trust

Bristol Eye Hospital, University Hospitals Bristol NHS Foundation Trust

Bradford Royal Infirmary, Bradford Teaching Hospitals NHS Foundation Trust

Addenbrooke's Hospital, Cambridge University Hospitals NHS Trust

Manchester Royal Eye Hospital, Central Manchester University Hospitals NHS Foundation Trust

10.2 Appendix II – GP letter

[Printed on UCL headed paper]

Dear *[GP name]*

Re:

Research Project: Functional Vision (FV) and Vision-related Quality of Life (VQoL) of children and young people with visual impairment: development of age-appropriate patient reported outcome measures (PROMs) for routine use in paediatric ophthalmology

Patient: *[patient name, date of birth, address]*

Our research team at UCL Institute of Child Health are working on a project, funded by the Fight for Sight charity, to develop age-appropriate patient reported outcome measures of vision related quality of life and functional vision for children and young people who are visually impaired. The study has been approved by the National Research Ethics Service *[REC reference number: 12-EE-0455].*

We are writing to inform you that we are about to write to the family of your patient [*patient name*] who has previously been treated at [*hospital name*] to ask whether they would like to participate in the study. This will involve *[to be amended as per below in accordance to the relevant study phase:*

- *Letter for phase 1:* discussing with me whether and how his/her eye-sight affects different aspects of his/her life.
- *Letter for phase 2:* discussing with me whether the draft questions we have developed are meaningful and relevant and what we can do (e.g. in terms of format) to improve our draft questionnaires.
- *Letter for phase 3:* completing two self-report questionnaires, one asking questions about his/her functional vision and the other asking questions about his/her quality of life.
- *Letter for phase 4:* completing three self-report questionnaires, one asking questions about his/her functional vision and two asking questions about his/her quality of life.*]*

It would be very helpful if you would indicate on the attached reply slip whether we should or should not contact this family. We have enclosed a project information sheet for your reference but please let us know if you would like any further information regarding the study.

We are very grateful for your help with this study. Please do not hesitate to contact us if you have any questions or comments about it.

Yours sincerely,

Miss Alexandra Robertson (PhD student),

Dr Valerie Tadić (Research Associate),

Professor Jugnoo Rahi (Study Chief Investigator),

Reply Slip

Research Project:
Functional Vision (FV) and Vision-related Quality of Life (VQoL) of children and young people with visual impairment: development of age-appropriate patient reported outcome measures (PROMs) for routine use in paediatric ophthalmology

Name of GP: ***[GP name]***

Name of Child: ***[Patient name]***

Please tick the box that applies:

	Yes	**No**
I would contact this family.	☐	☐

Signature: …….

Date:……

10.3 Appendix III – Parent information sheets

10.3.1 Parents of children aged 13-15 years

Information for parents

Study title: 'Children and young people's views on living with impaired sight'

We would like to ask you and your son/daughter *[to be written as is gender-appropriate and amended for the rest of the document]* to take part in this study.

If you or your child wish to have this document in Braille or have it read to you or him/her by a researcher, please let us know using the contact details at the end of this letter.

1. What is the aim of the study?

The aim of the study is to find out more about the quality of life and visual ability of children and young people who have problems with their eyesight. We know that poor vision can affect children and young people in a number of ways as they develop and grow, especially at school or college. But we do not know what children and young people themselves think about how their eyesight and any treatment they may have for it, affects their day-to-day lives.

2. Why is the study being done?

We are developing a series of age-appropriate questionnaires that children and young people with visual impairment can fill in and which measure their quality of life (e.g. how they feel about their eyesight) and visual functioning (e.g. difficulty reading from the computer or with independent mobility). We hope that these questionnaires will be used in the future by those who provide medical, educational or social services for children and young people with visual loss and those who carry out research into the causes of visual loss. We hope that this will help to give children and young people a say in all these areas.

3. Why are we being asked to take part?

With the help of other children and young people we have already developed questionnaires of quality of life and visual functioning that are suitable for children and young people with visual impairment aged 10-15 years. We now wish to develop questionnaires that are suitable for younger children (aged 8-12 years) and also for older teenagers (aged 13-17 years). We are asking you to take part because we would like to include all children and young people with visual impairment who are in that age range and who have attended an Eye Department in the UK.

4. What will happen if we decide to take part?

This is a large study, which involves different phases, but we would **only** ask you and your son/daughter to take part in the first phase of the study. This would involve the following:

a) If you and your son/daughter agree to take part, we would like you to complete the attached consent forms to let us know. You can use the freepost envelopes provided to post the forms back.

b) We would then like to *[to be amended as per below in accordance to the relevant study phase:*
   - *Letter for phase 2:* ask your son/daughter to talk to a researcher about some questions that we have developed with the help of other children and young people with visual impairment. We will meet your son/daughter in your home at a time convenient for your family. We would ask your son/daughter to give us advice regarding the individual questions, for example, if they are important or easy to understand, and whether the instructions for completing them are clear. We would also show some of these to him/her on a laptop to ask for his/her opinion on the electronic version of our questionnaire. This will help us ensure our questionnaire covers all the important information and is child/young person-friendly. This will take about 1 hour.
   - *Letter for phase 3:* ask you and your son/daughter to complete 2 forms we have developed with the help of other children and young people with visual impairment. One asks about his/her quality of life and the other about his/her visual ability. We will also ask you to provide some feedback about the questionnaire. You and your son/daughter can complete these in print or electronically, using a Word version on a CD or online. This will help us ensure that our questionnaires are working as they should and highlight if there are any problems with the questions (e.g. most children and young people skipping one particular question will tell us that this is not a particularly relevant question for this age group). It will also help us obtain the different perspectives that affected children or young people and their parents may have on all this. This will take between 10 and 45 minutes.
   - *Letter for phase 4:* We would like to ask you and your son/daughter to complete 3 forms asking questions about his/her quality of life and his/ her visual ability. Two of these we have developed with the help of other children and young people with visual impairment. The third is a general questionnaire that asks questions that should be suitable for all children and young people of the same age as your son/daughter, whether or not they have a health condition. You and your son/daughter can complete these in print or electronically, using a Word version on a CD or online version. This will help us ensure that our questionnaires are working as they should and highlight if there are any problems with the questions. It will also help us obtain the different perspectives that affected children or young people and their parents may have on all this. This will take between 15 minutes and 1 hour.*]*

c) We would also like you to complete the enclose short form (see the enclosed Family Background Questionnaire) which asks some questions

about you, your child and family so that we can learn more about your family background. This will take around 3 minutes. This also helps us to make sure that we are including as many different types of families in our study as possible so we can get as representative a point of view as possible.

We hope that taking part in our study should be a positive experience for your son/daughter allowing him/her to tell us in detail what it means for him/her to live with an eyesight problem. However, we recognise that there is the potential for you or your son/daughter to find some aspects of this difficult. We would like to reassure you that our study team is experienced and appropriately trained and able to deal sensitively with any difficulties that might arise. We also remind you that you are free to withdraw from the study at any point.

With your permission, we may also contact your family GP to inform them of any health or care related concerns that arise from your and your child's participation in this study.

5. Who will have access to my child's records?

Only the research team involved in this study and representatives from Regulatory Authorities or from the NHS Trust where it is relevant to you and your child taking part in this research would have access to your child's medical notes and the actual data collected during the study.

6. Who are the researchers involved in this study?

- Professor Jugnoo Rahi, Study Chief Investigator, Professor of Ophthalmic Epidemiology and Honorary Consultant Ophthalmologist, UCL Institute of Child Health and Great Ormond Street Hospital for Children NHS Foundation Trust
- Miss Alexandra Robertson, Primary Researcher, PhD Student, UCL Institute of Child Health
- Dr Val Tadić, Research Associate, UCL Institute of Child Health
- Mrs Phillippa Cumberland, Senior Research Associate in Biostatistics, UCL Institute of Child Health
- Professor Gillian Hundt, Professor of Social Sciences in Health, University of Warwick
- Dr Alki Liasis, Clinical Scientist and Head of Ophthalmology Department, Great Ormond Street Hospital for Children NHS Foundation Trust
- Ms Lisa Davies, Clinical Outcomes Lead, Great Ormond Street Hospital for Children NHS Foundation Trust
- Mrs Corie Brown, a parent of NHS user
- Professor Peng Khaw, the Director of the NIHR Biomedical Research Centre and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology
- Professor Anthony Moore, Duke Elder Professor of Ophthalmology and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology

7. What will happen with the findings of the study?

We plan to publish the findings of the study in scientific journals so that we can let other professionals know what we have learned from this study. However, we will pool information in our reports, so that **it will not be possible to identify any individual person who takes part. Everything you and your son/daughter tell us will be strictly confidential.** We will also write to you at a later date to let you know about the study findings.

8. What are the potential benefits?

This study is unlikely to bring any immediate benefits to your son/daughter, although many children and young people have found it a very positive experience to share their thoughts about their life with us. However, we hope that it will improve our understanding of children and young people's perspectives of their visual loss and our ability to measure their quality of life and vision in order to improve the treatment and services provided for them at this hospital and elsewhere in the country in the future.

9. Do I have to take part in this study?

If you or your son/daughter decide, now or at a later stage, that you do **not** wish to take part in this research project, that is entirely your right. This **will not** affect any present or future treatment of your son/daughter in any way.

10. Who do I speak to if problems arise?

If you have any questions or complaints about the way in which this study has been, or is being conducted, please, in the first instance, discuss them with the researchers named below. For independent advice you can contact the Patient Advice and Liaison Services (PALS) (http://www.pals.nhs.uk).

11. Details of how to contact the researchers:

If you have any questions or concerns that you would like to discuss before agreeing to participate in this study and signing the consent form you can contact any of the main researchers by letter, email or phone below.

Miss Alexandra Robertson (PhD Student),

Dr Valerie Tadić (Study Research Associate),

Professor Jugnoo Rahi (Study Chief Investigator),

10.3.2 Parents of YP aged 16-19 years

Information for parents

Study title: 'Children and young people's views on living with impaired sight'

We would like to ask you and your son/daughter *[to be written as is gender-appropriate and amended for the rest of the document]* to take part in this study.

If you or your child wish to have this document in Braille or have it read to you or him/her by a researcher, please let us know using the contact details at the end of this letter.

1. What is the aim of the study?

The aim of the study is to find out more about the quality of life and visual ability of children and young people who have problems with their eyesight. We know that poor vision can affect children and young people in a number of ways as they develop and grow, especially at school or college. But we do not know what children and young people themselves think about how their eyesight and any treatment they may have for it, affects their day-to-day lives.

2. Why is the study being done?

We are developing a series of age-appropriate questionnaires that children and young people with visual impairment can fill in and which measure their quality of life (e.g. how they feel about their eyesight) and visual functioning (e.g. difficulty reading from the computer or with independent mobility). We hope that these questionnaires will be used in the future by those who provide medical, educational or social services for children and young people with visual loss and those who carry out research into the causes of visual loss. We hope that this will help to give children and young people a say in all these areas.

3. Why are we being asked to take part?

With the help of other children and young people we have already developed questionnaires of quality of life and visual functioning that are suitable for children and young people with visual impairment aged 10-15 years. We now wish to develop questionnaires that are suitable for younger children (aged 6-9 years) and also for older teenagers (aged 16-18 years). We are asking you to take part because we would like to include all children and young people with visual impairment who are in that age range and who have attended an Eye Department in the UK.

4. What will happen if we decide to take part?

This is a large study, which involves different phases, but we would **only** ask you and your son/daughter to take part in one phase of the study. This would involve the following:

a) If you and your son/daughter agree to take part, we would like you to complete the attached consent forms to let us know. You can use the freepost envelopes provided to post the forms back.

b) We would then like to *[to be amended as per below in accordance to the relevant study phase:*
    - *Letter for phase 1:* ask your son/daughter to talk to a researcher about how his/her eyesight affects different aspects of his/her life. For this, we will meet your son/daughter in your home at a time convenient for your family. This could take between 30 min and 1 ½ hours. All the information we gather from these discussions will be pooled together anonymously. This will then be used to decide on the individual questions that will make up the questionnaires we aim to develop. We would also like to be able to quote anonymously comments you or your son/daughter make if they identify important issues in a particularly powerful way. In all our scientific papers or other reports about the study we will ensure that neither you nor your son/daughter can be identified.
    - *Letter for phase 2:* ask your son/daughter to talk to a researcher about some questions that we have developed with the help of other children and young people with visual impairment. We will meet your son/daughter in your home at a time convenient for your family. We would ask your son/daughter to give us advice regarding the individual questions, for example, if they are important or easy to understand, and whether the instructions for completing them are clear. This will help us ensure our questionnaire covers all the important information and is child/young person-friendly. This will take about 1 hour.
    - *Letter for phase 3:* ask you and your son/daughter to complete 2 forms we have developed with the help of other children and young people with visual impairment. One asks about his/her quality of life and the other about his/her visual ability. We will also ask you to provide some feedback about the questionnaire. You and your son/daughter can complete these in print or electronically, using a Word version on a CD or online. This will help us ensure that our questionnaires are working as they should and highlight if there are any problems with the questions (e.g. most children and young people skipping one particular question will tell us that this is not a particularly relevant question for this age group). It will also help us obtain the different perspectives that affected children or young people and their parents may have on all this. This will take between 10 and 45 minutes.
    - *Letter for phase 4:* We would like to ask you and your son/daughter to complete 3 forms asking questions about his/her quality of life and his/her visual ability. Two of these we have developed with the help of other children and young people with visual impairment. The third is a general questionnaire that asks questions that should be suitable for all children and young people of the same age as your son/daughter, whether or not they have a health condition. You and your son/daughter can complete these in print or electronically, using a Word version on a CD or online version. This will help us ensure that our questionnaires are working as they should and highlight if there are any problems with

the questions. It will also help us obtain the different perspectives that affected children or young people and their parents may have on all this. This will take between 15 minutes and 1 hour.*]*

c) We would also like you to complete the enclosed short form (see the enclosed Family Background Questionnaire) which asks some questions about you, your child and family so that we can learn more about your family background. This will take around 3 minutes. This also helps us to make sure that we are including as many different types of families in our study as possible so we can get as representative a point of view as possible.

We hope that taking part in our study should be a positive experience for your son/daughter allowing him/her to tell us in detail what it means for him/her to live with an eyesight problem. However, we recognise that there is the potential for you or your son/daughter to find some aspects of this difficult. We would like to reassure you that our study team is experienced and appropriately trained and able to deal sensitively with any difficulties that might arise. We also remind you that you are free to withdraw from the study at any point.

With your permission, we may also contact your family GP to inform them of any health or care related concerns that arise from your and your child's participation in this study.

5. Who will have access to my child's records?

Only the research team involved in this study and representatives from Regulatory Authorities or from the NHS Trust where it is relevant to you and your child taking part in this research would have access to your child's medical notes and the actual data collected during the study.

6. Who are the researchers involved in this study?

- Professor Jugnoo Rahi, Study Chief Investigator, Professor of Ophthalmic Epidemiology and Honorary Consultant Ophthalmologist, UCL Institute of Child Health and Great Ormond Street Hospital for Children NHS Foundation Trust
- Miss Alexandra Robertson, Primary Researcher, PhD Student, UCL Institute of Child Health
- Dr Val Tadić, Research Associate, UCL Institute of Child Health
- Mrs Phillippa Cumberland, Senior Research Associate in Biostatistics, UCL Institute of Child Health
- Professor Gillian Hundt, Professor of Social Sciences in Health, University of Warwick
- Dr Alki Liasis, Clinical Scientist and Head of Ophthalmology Department, Great Ormond Street Hospital for Children NHS Foundation Trust
- Ms Lisa Davies, Clinical Outcomes Lead, Great Ormond Street Hospital for Children NHS Foundation Trust
- Mrs Corie Brown, a parent of NHS user

- Professor Peng Khaw, the Director of the NIHR Biomedical Research Centre and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology
- Professor Anthony Moore, Duke Elder Professor of Ophthalmology and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology

7. What will happen with the findings of the study?

We plan to publish the findings of the study in scientific journals so that we can let other professionals know what we have learned from this study. However, we will pool information in our reports, so that **it will not be possible to identify any individual person who takes part. Everything you and your son/daughter tell us will be strictly confidential.** We will also write to you at a later date to let you know about the study findings.

8. What are the potential benefits?

This study is unlikely to bring any immediate benefits to your son/daughter, although many children and young people have found it a very positive experience to share their thoughts about their life with us. However, we hope that it will improve our understanding of children and young people's perspectives of their visual loss and our ability to measure their quality of life and vision in order to improve the treatment and services provided for them at this hospital and elsewhere in the country in the future.

9. Do I have to take part in this study?

If you or your son/daughter decide, now or at a later stage, that you do **not** wish to take part in this research project, that is entirely your right. This **will not** affect any present or future treatment of your son/daughter in any way.

10. Who do I speak to if problems arise?

If you have any questions or complaints about the way in which this study has been, or is being conducted, please, in the first instance, discuss them with the researchers named below. For independent advice you can contact the Patient Advice and Liaison Services (PALS) (http://www.pals.nhs.uk).

11. Details of how to contact the researchers:

If you have any questions or concerns that you would like to discuss before agreeing to participate in this study and signing the consent form you can contact any of the main researchers by letter, email or phone below.

Miss Alexandra Robertson (PhD Student),

Dr Valerie Tadić (Study Research Associate),

Professor Jugnoo Rahi (Study Chief Investigator),

10.4 Appendix IV – Parent invitation letter (for children aged 13-15 years and YP aged 16-19 years)

*NOTE: The starred sentence is for the letters to parents of young people aged 16-18 years only, as they will also receive an invitation letter.

[Printed on UCL headed paper]

Dear *[name of parents],*

We are writing to you as the parent of [*child/young person name*] who has attended the Ophthalmology Department of [*hospital name*]. We would like to ask you and [*child/young person name*] to take part in our study. *We have also written to [*young person name*] so that you can discuss this as a family before you decide whether to take part.*

If you or your child wish to have this document in Braille or have it read to you or him/her *[amend as gender appropriate, also for the rest of the document]* by a researcher, please let us know using the contact details at the end of this letter.

The purpose of the study is to try to find out more about the quality of life and visual ability of children and young people who have problems with their eyesight. To do this, we are developing questionnaires for children and young people with visual impairment, which they can fill in themselves and which ask questions about how their eyesight problems affect their daily life. We hope that these questionnaires will be used by people who work with children and young people with visual loss in hospitals, schools or colleges and social services, so that they can take the children's and young people's perspectives into account when providing care and services.

To help us develop these questionnaires we would like to *[to be amended as per below in accordance to the relevant study phase:*

- *Letter for phase 1:* talk to your son/daughter about his/her daily life so that we know what kind of questions we should include in these questionnaires.
- *Letter for phase 2:* ask your son/daughter what he/she thinks about the questionnaires that we have developed to make sure that they are easily understood, meaningful and that we have included the right kind of questions. This will help us improve our questionnaires and make sure that they are easy to complete and comprehensible to children and young people.
- *Letter for phase 3:* ask you and your son/daughter to complete 2 questionnaires that we have already developed with the help of other children and young people with visual impairment, and to let us know his/her thoughts on these. This will help us improve our questionnaires and make sure that they are easy to complete and comprehensible to children and young people.
- *Letter for phase 4:* ask you and your son/daughter to complete 2 draft questionnaires that we have already developed, as well as another general health questionnaire. This will help us make sure that our new questionnaires are working as they should and are user friendly.*]*

The information sheet enclosed with this letter offers a more detailed summary of our study and what is required of you if you decide to participate.

We would be grateful if you would discuss this with your son/daughter and let us know whether you would be willing to take part. You can let us know by completing and signing the enclosed consent form. If you decide not to take part, this will not affect your son/daughter's medical care in any way.

We can send you and your son/daughter all the documents in large font or electronically (via email or on a CD), if you or your son/daughter prefer this.

If we do not hear from you, we will phone you in the next couple of weeks to ensure that you have received this letter. In the meantime, if you need more information or you would like some help in filling in the forms or if you would prefer to have the study explained in another language, please feel free to phone or email us using our contact details below.

Thank you for considering taking part in our study.

Yours sincerely,

Miss Alexandra Robertson (PhD Student),

Dr Valerie Tadić (Research Associate),

Professor Jugnoo Rahi (Professor of Ophthalmic Epidemiology),

On behalf of the research group:

- Miss Alexandra Robertson, Primary Researcher, PhD Student, UCL Institute of Child Health
- Professor Jugnoo Rahi, Study Chief Investigator, Professor of Ophthalmic Epidemiology and Honorary Consultant Ophthalmologist, UCL Institute of Child Health and Great Ormond Street Hospital for Children NHS Foundation Trust
- Dr Valerie Tadić, Research Associate, UCL Institute of Child Health
- Mrs Phillippa Cumberland, Senior Research Associate in Biostatistics, UCL Institute of Child Health
- Professor Gillian Hundt, Professor of Social Sciences in Health, University of Warwick
- Dr Alki Liasis, Clinical Scientist and Head of Ophthalmology Department, Great Ormond Street Hospital for Children NHS Foundation Trust
- Ms Lisa Davies, Clinical Outcomes Lead, Great Ormond Street Hospital for Children NHS Foundation Trust
- Mrs Corie Brown, a parent of NHS user
- Professor Peng Khaw, the Director of the NIHR Biomedical Research Centre and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology
- Professor Anthony Moore, Duke Elder Professor of Ophthalmology and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology

10.5 Appendix V – Parent consent forms

10.5.1 Parents of children aged 13-15 years

[Printed on UCL headed paper]

Consent form for parents

Study title: Children and young people's views on living with impaired sight

Sponsor Protocol No*:* NRES Committee East of England - Essex REC ref: 12-EE-0455

Investigator: Jugnoo Rahi **Contact details:** ☎020 7905 2250 💻: j.rahi@ucl.ac.uk

Person study ID: ***[PID]***

Please circle YES or NO below so we know if you agree with these statements:

1	I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these questions answered satisfactorily.	Yes	No
2	I understand that my participation and that of my child is voluntary and that we are free to withdraw from the study at any time, without giving any reason and without my child's medical care or legal rights being affected.	Yes	No
3	I understand that relevant sections of my child's medical notes and data collected during the study may be looked at by the researchers involved in this study and the representatives from Regulatory Authorities or from the NHS Trust where it is relevant to our participation in this research. I give permission for these individuals to have access to these records.	Yes	No
4	I agree to my family GP being informed of any health or care related concerns that are identified from our participation in this study.	Yes	No
5	I agree to take part in this study.	Yes	No
6	I agree for my child to take part in this study.	Yes	No

……..	…….	
Child's Name	Date	
……..	…….	…….
Parent Name	Date	Signature
……..	…….	…….
Name of Person taking consent	Date	Signature

10.5.2 Parents of YP aged 16-19 years

[Printed on UCL headed paper]

Consent form for parents

Study title: Children and young people's views on living with impaired sight

Sponsor Protocol No*:* NRES Committee East of England - Essex REC ref: 12-EE-0455

Investigator: Jugnoo Rahi **Contact details:** ☎020 7905 2250 💻: j.rahi@ucl.ac.uk

Person study ID: *[PID]*

Please circle YES or NO below so we know if you agree with these statements:

1	I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these questions answered satisfactorily.	Yes	No
2	I understand that my participation and that of my child is voluntary and that we are free to withdraw from the study at any time, without giving any reason and without my child's medical care or legal rights being affected.	Yes	No
3	I understand that relevant sections of my child's medical notes and data collected during the study may be looked at by the researchers involved in this study and the representatives from Regulatory Authorities or from the NHS Trust where it is relevant to our participation in this research. I give permission for these individuals to have access to these records.	Yes	No
4	I agree to my family GP being informed of any health or care related concerns that are identified from our participation in this study.	Yes	No
5	I agree to take part in this study.	Yes	No

……..	…….	
Child's Name	Date	
……..	…….	…….
Parent Name	Date	Signature
……..	…….	…….
Name of Person taking consent	Date	Signature

10.6 Appendix VI – Participant information sheets

10.6.1 Children aged 13-15 years

Information for children and young people

Study title: Children and young people's views on living with impaired sight

We would like to ask you to take part in this study.

We want to learn more about what it's like to be a child or a young person with eyesight problems. We want to make forms that children and young people like you can fill in and which ask questions about your daily life. Doctors and other people who look after you in hospital and in school can then use this to understand more about you and your life.
Once you have talked about the study with your parents, they will let us know if you are happy to take part in it.

We would like you to *[to be amended as per below in accordance to the relevant study phase:*

- *Letter for phase 2:* meet a member of our research team. She will show you 2 forms that we have made with the help of other children and young people like you. She will ask you to fill in these forms. She will also ask you what you think about them, for example, whether the questions are easy to understand or how you could change them so they are more child-friendly. She would come to talk to you about this in your house. This may take up to 1 hour. Your answers will help us to make these forms better and more child- and young person-friendly.
- *Letter for phase 3:* We would like you to fill in 2 forms that have been made with help of other young people your age. They ask questions about how your eyesight affects your everyday life and daily activities. You can either fill in the forms on printed paper or on the computer, and you can let you parents know which one would be easier for you to do. This may take up to 1 hour. Your answers will help us make these forms better and more young person-friendly.
- *Letter for phase 4:* fill in 3 forms that have been made with help of other children and young people your age. They ask questions about how your eyesight affects your everyday life and daily activities. You can either fill in the forms on printed paper or on the computer, and you can let your parents know which one would be easier for you to do. This may take up to 1 hour. Your answers will help us make these forms better and more child and young people friendly.*]*

We cannot promise that taking part in our study will help you now. But it will help us learn how we can give better care to children and young people like you in our hospital and other hospitals in the future. We will not talk about what you tell us with anyone who is not part of our study team.

If you decide that you do **not** want to help with this study, you can just tell your parents and it will not be a problem.

If you want to read this letter or the lists of questions in Braille or have them read to you by the researcher please let you parents know and they will call us to arrange this.

10.6.2 YP aged 16-19 years

Information for young people

Study title: 'Children and young people's views on living with impaired sight'

We would like to ask you to take part in this study.

If you wish to have this document in Braille or have it read to you by a researcher, please let us know using the contact details at the end of this information sheet.

1. What is the aim of the study?

The aim of the study is to find out more about the quality of life and visual ability of children and young people who have problems with their eyesight. We especially want to learn what children and young people themselves think about their eye condition and how it might affect everyday life.

2. Why is the study being done?

We are developing questionnaires that young people can fill in to tell us more about their quality of life (e.g. how they feel about their eyesight) and their vision (e.g. difficulty reading from the computer or completing homework). We want these questionnaires to be used in the future by professionals in hospitals, schools or social services who work with children and young people with visual impairment. This will make sure that they take children and young people's views into account when providing medical care.

3. Why am I being asked to take part?

With the help of other young people we have already developed questionnaires that are suitable for children and young people with visual impairment aged 10-15 years. We now wish to develop questionnaires that are suitable for younger children (aged 6-9 years) and older teenagers (aged 16-18 years). This is why we are asking for your help.

4. What will happen if I decide to take part?

Taking part would involve the following:

a) First, we would like you to discuss the study with your parents and decide whether you wish to take part. If you **do** want to take part, we would like you to complete the attached consent forms and send them back to us using the prepaid return envelope we have given you.

b) We would then like to *[to be amended as per below in accordance with the relevant study phase:*
   - *Letter for phase 1:* talk to you in more detail about how your eyesight affects different aspects of your life. To do this, we will visit your home at a time that suits you. This may take between half an hour and 1 and a half hours. We would need to audio-record your conversation with the researcher. All the information

you give us will be added to what other young people with eyesight problems have told us. This will then be used to design questions for the questionnaire we are developing. We may use what you tell us word-for-word in our future reports but your name will never be used and if we do include anything you have said, we will make sure you remain anonymous.

- *Letter for phase 2:* ask you to talk to a researcher about some questions that we have developed with the help of other young people with visual impairment. For this, we will meet you in your home at a time that suits you. We would ask you to give us advice regarding the individual questions, for example, if they are important or easy to understand and if the instructions for completing them are clear. We would also show you some of these on a laptop to ask for your opinion on the electronic version of our questionnaires. This will take about 1 hour. This will help us ensure our questionnaire covers all the important information and is young person-friendly.
- *Letter for phase 3:* ask you to complete 2 forms that we have developed with the help of other young people with visual impairment. One asks about your quality of life and the other about your vision. We will also ask you to provide some feedback about the questionnaires. You can complete these in print or electronically, using a Word version on a CD. This will help us ensure that our questionnaires are working as they should and highlight if there are any problems with the questions. This will take between 10 and 45 min.
- *Letter for phase 4:* ask you to complete 3 forms asking questions about your quality of life and vision. Two of these we have developed with the help of other young people with visual impairment. The third is a general questionnaire that asks questions about your life and health. You can complete these in print or electronically, using a Word version on a CD or online version. This will help us ensure that our questionnaires are working as they should and highlight if there are any problems with the questions. This will take between 15 minutes and 1 hour.*]*

We hope that taking part in this study will provide a chance for you to talk about what it means for you to live with an eyesight problem. However, we understand if you find some of this a little difficult. We would like you to know that you can drop out of the study at any point if you feel you need to. This would not affect your medical care in any way.

With your permission, we may also contact your GP to let him/her know if any health or care related concerns arise from your participation in this study.

5. Who will be allowed to look at my notes?

The only people allowed to look at your research and medical notes will be the research team who carry out this study. People from the hospital where you have received your eye treatment and those from the ‘research ethics committee’ may

also need to look at these notes as they have to make sure that the study is done safely and properly.

6. Will taking part in this study make a difference to my life?

We cannot promise that the study will make a difference to you now, but we hope it will help us understand more how young people feel about their eyesight problems. This will help us improve how we treat other young people at this hospital and elsewhere in the country in the future.

7. Do I have to take part in this study?

This is entirely up to you. If you decide now or later that you do not want to take part, this would not affect your medical care or your rights in any way.

8. What will happen with the findings of the study?

We plan to write reports and scientific papers from this study so that we can let other professionals know what we have learned from young people like you. However, we will put all the information together in our reports in a way that **it will not be possible for anyone to recognise you or any other young person in this study. We will not share anything you say with anybody outside of our study group.** We will also write to you at a later date to let you know our findings.

9. Who can I talk to if there is a problem?

If you have any questions or complaints about the study you can tell the researcher talking to you straight away. You can also talk about it to your parents and they can take it further with our research team.

10. Details of how to contact the researchers:

If you have any questions or concerns that you would like to discuss before agreeing to participate in this study and signing the consent form you can contact any of the main researchers by letter, email or phone below.

Miss Alexandra Robertson, Primary Researcher (PhD Student),

Dr Valerie Tadić, Study Research Associate,

Professor Jugnoo Rahi, Study Chief Investigator,

10.7 Appendix VII – Invitation letters for YP aged 16-19 years

[Printed on UCL headed paper]

Dear *[Name of young person],*

We are writing to you as a patient who has attended the Eye Department of *[hospital name].* We would like to ask you to take part in our study. We have also written about this to your parents so you can talk it over with them before you decide whether you want to take part.

If you wish to have this document in Braille or have it read to you by a researcher, please let us know using the contact details at the end of this letter.

The purpose of the study is to learn more about what life is like for young people who have problems with their eyesight. To do this, we are developing questionnaires, which young people with impaired vision can fill in to explain how their eyesight problems affect their daily life. We hope that these questionnaires will be used in hospitals, schools and social services, so that the views of young people are taken into account when providing care and services.

To help us develop these questionnaires, we would like to *[to be amended as per below in accordance to the relevant study phase:*

- *Letter for phase 1:* talk to you about your day-to-day life so that we know what kind of questions we should include in these questionnaires.
- *Letter for phase 2:* ask you what you think about the questionnaires that we have developed to make sure that they are easily understood, meaningful and that we have included the right kind of questions.
- *Letter for phase 3:* ask you to fill in two questionnaires that we have developed with the help of other young people with eyesight problems, and to let us know what you think about them. This will help us improve our questionnaires and make sure that they are easy to complete and understand.
- *Letter for phase 4:* ask you to fill in two draft questionnaires that we have developed, as well as another general health questionnaire. This will help us make sure that our new questionnaires are working as they should and that they are user-friendly.*]*

The information sheet enclosed with this letter explains the study in more detail and what we would ask of you if you decide to participate.

Please discuss this with your parents and let us know whether you would be willing to take part. You can let us know by completing and signing the enclosed consent form, which can be posted back to us in the prepaid envelope provided. We have asked your parents to do the same. If you decide <u>not</u> to take part, this will not affect your medical care in any way.

We can send you all the documents in large font or electronically (via email or on a CD), if you prefer this.

If we do not hear from you, we will phone you in the next couple of weeks to ensure that you have received this letter. If you need more information now or you would like some help in filling in the forms or if you would prefer to have the study explained in another language, please feel free to telephone or email us using our contact details below.

Thank you for considering taking part in our study.

Yours sincerely,

Miss Alexandra Robertson (PhD Student),

Dr Valerie Tadić (Study Research Associate),

Professor Jugnoo Rahi (Study Chief Investigator),

On behalf of the research group:

- Miss Alexandra Robertson, Primary Researcher, PhD Student, UCL Institute of Child Health
- Professor Jugnoo Rahi, Study Chief Investigator, Professor of Ophthalmic Epidemiology and Honorary Consultant Ophthalmologist, UCL Institute of Child Health and Great Ormond Street Hospital for Children NHS Foundation Trust
- Dr Valerie Tadić, Research Associate, UCL Institute of Child Health
- Mrs Phillippa Cumberland, Senior Research Associate in Biostatistics, UCL Institute of Child Health
- Professor Gillian Hundt, Professor of Social Sciences in Health, University of Warwick
- Dr Alki Liasis, Clinical Scientist and Head of Ophthalmology Department, Great Ormond Street Hospital for Children NHS Foundation Trust
- Ms Lisa Davies, Clinical Outcomes Lead, Great Ormond Street Hospital for Children NHS Foundation Trust
- Mrs Corie Brown, a parent of NHS user
- Professor Peng Khaw, the Director of the NIHR Biomedical Research Centre and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology
- Professor Anthony Moore, Duke Elder Professor of Ophthalmology and Consultant Ophthalmologist, Moorfields Eye Hospital NHS Foundation Trust and UCL Institute of Ophthalmology

10.8 Appendix VIII – Participant consent forms

10.8.1 Children aged 13-15 years

[Printed on UCL headed paper]

Child and Young Person Assent Form

STUDY TITLE:

Children and young people's views on living with impaired sight

Please circle:

Have your parents or carers talked to you about this study?	**YES / NO**
Do you understand why you are being asked to take part?	**YES / NO**
Have you asked all the questions you want to?	**YES / NO**
Do you understand that it's OK to stop taking part at any time?	**YES / NO**
Are you happy to take part?	**YES / NO**

If you **do** want to take part please write your name below

Your Name…….....

Date…….

Researcher's name: Alexandra Robertson

Signature: ……

Date: ……

Thank you very much for your help!

10.8.2 YP aged 16-19 years

[Printed on UCL headed paper]

Young Person Assent Form

Study title: Children and young people's views on living with impaired sight

Sponsor Protocol No*:* NRES Committee East of England - Essex REC ref: 12-EE-0455

Investigator: Jugnoo Rahi **Contact details:** ☎020 7905 2250 💻: j.rahi@ucl.ac.uk

Person study ID: *[PID]*

Please circle YES or NO below so we know if you agree with these statements:

1	I confirm that I understand the information sheet for this study.	Yes	No
2	I have had the chance to consider the information, ask questions and have had these questions answered.	Yes	No
3	I understand that I am free to choose whether I take part in this study or not, that I am free to withdraw from this study at any time, without giving any reason, and that this will not affect my medical care or my rights.	Yes	No
4	I understand that some sections of my medical notes and information collected during the study may be looked at by researchers doing this study and also the people who are making sure that the study is done safely and properly. I am happy for these people to have access to these records.	Yes	No
5	I agree to my family GP being told of any health or care related concerns that are identified from my participation in this study.	Yes	No
6	I agree for my interview with the researcher to be audio-recorded and for anonymised direct quotes from these interviews to be used *[for participants in phase 1 only]*	Yes	No
7	I agree to take part in this study.	Yes	No

…… Name	…… Date	…….. Signature
…… Name of person taking consent	…… Date	…….. Signature

10.9 Appendix IX – Family Background Questionnaire

FAMILY BACKGROUND QUESTIONNAIRE FOR PARENTS

This form asks for background information about you and your family, which we hope you will be able to provide. This will help us check if a broad range of families have been included in the study.

All the information you provide will be treated in strict confidence.

ABOUT YOU:

What is your relationship to your child with an eyesight problem?

☐ Natural Mother ☐ Stepmother ☐ Adoptive Mother ☐ Foster Mother

☐ Natural Father ☐ Stepfather ☐ Adoptive Father ☐ Foster Father

☐ Other guardian, please specify:

……..

How old are you?

☐ Less than 20 years ☐ 21 to 30 years ☐ 31 to 40 years ☐ 41 to 50 years

☐ More than 51 years

3. Please tell us to which group(s) you feel you belong:

☐ White - English, Scottish, Welsh, Irish ☐ White other, please specify: …….

☐ Black Caribbean ☐Black African ☐ Black other, please specify: ……..

☐ Indian ☐ Pakistani ☐ Bangladeshi

☐ Chinese

☐ Mixed

☐ Asian other, please specify: ……..……..

☐ Other, please specify: ……..……

4. Please describe your education or training:

☐ No qualifications or training

☐ A level

☐ City and Guilds Certificate

☐ NVQs

☐ Degree

☐ Professional qualification

☐ Other, please specify:

……....

5. Please describe your job:

(e.g. housewife, nurse, bus driver, engineer, accounting clerk, or tell us if you are unemployed)

……

6. Is the house/flat where you live:

☐ Owned by you

☐ Privately rented

☐ Rented from a housing association

☐ Rented from the local authority

☐ Other, please specify:

7. How many cars do you have access to?

☐ None

☐ One

☐ Two or more

ABOUT YOUR CHILD WITH AN EYESIGHT PROBLEM:

8. Please tell us to which group(s) your child belongs to:

☐ White - English, Scottish, Welsh, Irish ☐ White other, please specify:……..

☐ Black Caribbean ☐Black African ☐ Black other, please specify: ……....

☐ Indian ☐ Pakistani ☐ Bangladeshi

☐ Chinese

☐ Mixed

☐ Asian other, please specify: …….

☐ Other, please specify: ……...

9. Does your child have any <u>other</u> problems affecting development? ☐ Yes ☐ No

If YES, please tick any boxes which apply.

☐ Movement disorder (e.g. cerebral palsy, spina bifida, muscular dystrophy)

☐ Communication disorder (e.g. autism)

☐ Language disorder (e.g. speech not understandable by others)

☐ Behaviour disorder

☐ Developmental delay

☐ Epilepsy or seizure disorder

☐ Hearing impairment

☐ Eating disorder

☐ Other, please specify:

……..

10. What type of school/college/university does your child attend?

☐ Mainstream

☐ Specialist VI school/college/university

☐ Community specialist school/college/university

☐ Home educated

☐ Other, please specify……

ABOUT YOUR FAMILY:

11. How would you describe your family structure?

☐ Two-parent family

☐ Single parent family

☐ Other, please specify:

……..

12. Does your child with eyesight problem have any siblings? ☐ Yes ☐ No

(please provide the details of their gender and ages, e.g. brother aged 12)

……

……

13. Do any of your *CHILD'S SIBLINGS* have:

an eyesight problem? ☐ No ☐ Yes, please specify: ……

any other serious medical problems? ☐ No ☐ Yes, please specify: ……

14. Do any of your CHILD'S PARENTS have:

an eyesight problem? ☐ No ☐ Yes, please specify: ……

any other serious medical problems? ☐ No ☐ Yes, please specify: ……

15. PLEASE LET US KNOW IF YOU HAVE ANY COMMENTS

……

……

……

THANK YOU VERY MUCH FOR FILLING IN THIS FORM

10.10 Appendix X – Systematic review search terms

(((((("vision disorders"[MeSH Terms] OR ("vision"[All Fields] AND "disorders"[All Fields]) OR "vision disorders"[All Fields] OR ("visual"[All Fields] AND "impairment"[All Fields]) OR "visual impairment"[All Fields])) OR ("eye diseases"[MeSH Terms] OR ("eye"[All Fields] AND "diseases"[All Fields]) OR "eye diseases"[All Fields] OR ("eye"[All Fields] AND "disease"[All Fields]) OR "eye disease"[All Fields])))

((((("adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "teen"[All Fields])) OR (young[All Fields] AND ("persons"[MeSH Terms] OR "persons"[All Fields] OR "people"[All Fields]))) OR ("young adult"[MeSH Terms] OR ("young"[All Fields] AND "adult"[All Fields]) OR "young adult"[All Fields])))

(((((("quality of life"[MeSH Terms] OR ("quality"[All Fields] AND "life"[All Fields]) OR "quality of life"[All Fields])) OR ("activities of daily living"[MeSH Terms] OR ("activities"[All Fields] AND "daily"[All Fields] AND "living"[All Fields]) OR "activities of daily living"[All Fields])) OR “gratification”[All Fields]) OR ("personal satisfaction"[MeSH Terms] OR ("personal"[All Fields] AND "satisfaction"[All Fields]) OR "personal satisfaction"[All Fields] OR "satisfaction"[All Fields]))

10.11 Appendix XI – Feasibility questions and probes used in phase 2 (expert consultations)

Feedback on the instruction page:

1. The instructions for this questionnaire are…

[*Researcher reads instructions aloud*]

2. When we go through these questions, I'd like to remind you that there are no right or wrong answers and we want to know how true each statement is for you.

3. Do you think these instructions are clear?

4. Is there anything you would do to make the instructions more clear?

Feedback on the response options:

5. The response options are…

[*Researcher reads response options aloud*]

6. What do you think about the response choices?

7. How would you make the response options easier to understand?

Feedback on individual items:

[*Researcher reads aloud Item 1*]

8. What answer would you give to this question?

9. Why would you choose that answer?

10. OK, so you would pick [*researcher reads aloud the chosen response option]*? Why not this answer [*researcher reads aloud an alternative response option*] instead?

11. Can you give me an example of why you chose that answer?

12. What does this question mean to you?

13. Was this question easy to understand/Were there any specific words that are difficult to understand?

14. Was this question easy to answer? If not, why not?

[*Researcher repeats steps 8-14 with all subsequent* items*. When all items have been answered, researcher repeats steps 1-14 with all items in the alternative questionnaire*]

Feedback on the overall questionnaire:

15. What did you think about that questionnaire?

16. Is there anything you would change about the questionnaire?

17. If we gave it to you when you came to hospital, how would you feel about filling it in?

18. Would it be best if we gave it to you on paper? [*If not, researcher probes alternative methods of administration*]

19. Are there any things that we forgot to ask you about that you think are important in your life?

10.12 Appendix XII – Feasibility questions included in phase 3 questionnaire booklets (presented at the end of each instrument)

We would also like you to answer some short questions about this form. This will help us understand how easy or difficult you found completing it so that we make it easier for young people to fill in.

How long did it take you to complete this form?

……..

How easy was it?

- Very easy
- Easy
- Difficult
- Very difficult

How easy did you find the instructions?

- Very easy
- Easy
- Difficult
- Very difficult

10.13 Appendix XIII – Paediatric Quality of Life Inventory Version 4.0 – UK English, Child Report (ages 8-12)©

On the following pages is a list of things that might be a problem for you.

Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem

1 if it is **almost never** a problem

2 if it is **sometimes** a problem

3 if it is **often** a problem

4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

*In the **past month,** how much of a problem has this been for you…*

ABOUT MY HEALTH AND ACTIVITIES *(problems with…)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

ABOUT MY FEELINGS *(problems with)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ON WITH OTHERS	**Never**	**Almost**	**Some-**	**Often**	**Almost**

(problems with…)		**Never**	**times**		**Always**
1. I have trouble getting on with other children	0	1	2	3	4
2. Other children do not want to be my friend	0	1	2	3	4
3. Other children tease me	0	1	2	3	4
4. I cannot do things that other children my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other children	0	1	2	3	4

ABOUT SCHOOL *(problems with…)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

10.14 Appendix XIV – Paediatric Quality of Life Inventory Version 4.0 – UK English, Teenager Report (ages 13-17)©

On the following pages is a list of things that might be a problem for you.

Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem

1 if it is **almost never** a problem

2 if it is **sometimes** a problem

3 if it is **often** a problem

4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

*In the **past month,** how much of a problem has this been for you…*

ABOUT MY HEALTH AND ACTIVITIES *(problems with…)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

ABOUT MY FEELINGS *(problems with)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ON WITH OTHERS *(problems with…)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I have trouble getting on with other	0	1	2	3	4

teenagers					
2. Other teenagers do not want to be my friend	0	1	2	3	4
3. Other teenagers tease me	0	1	2	3	4
4. I cannot do things that other teenagers my age can do	0	1	2	3	4
5. It is hard to keep up with other teenagers my age	0	1	2	3	4

ABOUT SCHOOL/COLLEGE *(problems with…)*	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my school/college work	0	1	2	3	4
4. I miss school/college because of not feeling well	0	1	2	3	4
5. I miss school/college to go to the doctor or hospital	0	1	2	3	4

10.15 Appendix XV – Questionnaire assessing participants' preferred method of administration (presented at the end of phase 4 questionnaire booklets)

How easy did you find it to complete the booklet?

1. Very easy
2. Easy
3. A bit difficult
4. Very difficult or impossible

If you were to do this again, how would you prefer to complete the forms?

1. Using pen and paper
2. Using a computer, laptop, tablet or mobile phone

When you are reading or using a computer, do you use any assistive technologies to help you?

YES/NO

Which assistive technologies do you use?

Screen reader e.g. JAWS **YES/NO**

Screen magnifier **YES/NO**

Speech software **YES/NO**

Any other assistive devices?

……..

When do you use assistive technologies?

At home? **YES/NO**

At school? **YES/NO**

Any other times?

……..

10.16 Appendix XVI – Child VQoL scaled score-to-measure table

Score	Measure	SE	Score	Measure	SE	Score	Measure	SE
0	.00	16.34	25	43.49	2.26	50	57.23	2.36
1	10.84	8.99	26	44.05	2.24	51	57.86	2.39
2	17.15	6.40	27	44.61	2.23	52	58.51	2.42
3	20.89	5.26	28	45.16	2.21	53	59.17	2.46
4	23.58	4.59	29	45.71	2.20	54	59.86	2.50
5	25.71	4.14	30	46.25	2.19	55	60.58	2.55
6	27.47	3.81	31	46.79	2.18	56	61.32	2.60
7	28.98	3.55	32	47.32	2.18	57	62.09	2.66
8	30.31	3.35	33	47.85	2.17	58	62.91	2.73
9	31.51	3.19	34	48.38	2.17	59	63.77	2.81
10	32.60	3.05	35	48.90	2.17	60	64.68	2.90
11	33.60	2.94	36	49.43	2.17	61	65.65	3.00
12	34.54	2.84	37	49.96	2.17	62	66.70	3.12
13	35.41	2.75	38	50.49	2.18	63	67.83	3.26
14	36.23	2.68	39	51.02	2.18	64	69.08	3.42
15	37.02	2.61	40	51.55	2.19	65	70.47	3.63
16	37.77	2.56	41	52.09	2.19	66	72.04	3.88
17	38.48	2.51	42	52.63	2.20	67	73.87	4.21
18	39.17	2.46	43	53.18	2.22	68	76.07	4.67
19	39.84	2.42	44	53.73	2.23	69	78.85	5.34
20	40.49	2.39	45	54.29	2.24	70	82.68	6.47
21	41.11	2.35	46	54.86	2.26	71	89.09	9.04
22	41.73	2.33	47	55.43	2.28	72	100.00	16.37
23	42.33	2.30	48	56.02	2.30			
24	42.91	2.28	49	56.62	2.33			

10.17 Appendix XVII – YP VQoL scaled score-to-measure table

Score	Measure	SE	Score	Measure	SE	Score	Measure	SE
0	.00	16.17	27	43.52	2.13	54	56.93	2.23
1	10.71	8.89	28	44.03	2.12	55	57.50	2.26
2	16.94	6.32	29	44.54	2.11	56	58.09	2.29
3	20.63	5.19	30	45.04	2.09	57	58.69	2.32
4	23.28	4.53	31	45.53	2.08	58	59.30	2.36
5	25.36	4.08	32	46.02	2.07	59	59.94	2.40
6	27.09	3.75	33	46.50	2.07	60	60.60	2.44
7	28.57	3.49	34	46.99	2.06	61	61.29	2.49
8	29.87	3.29	35	47.47	2.06	62	62.01	2.54
9	31.03	3.13	36	47.94	2.05	63	62.76	2.61
10	32.09	2.99	37	48.42	2.05	64	63.54	2.67
11	33.06	2.87	38	48.90	2.05	65	64.38	2.75
12	33.96	2.77	39	49.37	2.05	66	65.26	2.84
13	34.80	2.69	40	49.85	2.05	67	66.21	2.94
14	35.60	2.61	41	50.32	2.06	68	67.23	3.06
15	36.35	2.55	42	50.80	2.06	69	68.33	3.20
16	37.06	2.49	43	51.28	2.07	70	69.55	3.37
17	37.75	2.43	44	51.77	2.07	71	70.91	3.57
18	38.41	2.39	45	52.26	2.08	72	72.45	3.82
19	39.04	2.35	46	52.75	2.09	73	74.24	4.15
20	39.65	2.31	47	53.25	2.10	74	76.40	4.60
21	40.25	2.27	48	53.75	2.11	75	79.13	5.26
22	40.82	2.24	49	54.26	2.13	76	82.90	6.38
23	41.39	2.22	50	54.77	2.15	77	89.22	8.94
24	41.94	2.19	51	55.30	2.16	78	100.00	16.20
25	42.47	2.17	52	55.83	2.18			
26	43.00	2.15	53	56.38	2.21			

10.18 Appendix XVIII – Child FV scaled score-to-measure table

Score	Measure	SE	Score	Measure	SE	Score	Measure	SE
0	.00	13.49	29	43.11	2.06	58	59.12	2.11
1	9.10	7.52	30	43.69	2.04	59	59.82	2.12
2	14.56	5.45	31	44.25	2.03	60	60.44	2.14
3	17.91	4.54	32	44.82	2.02	61	61.07	2.16
4	20.39	4.02	33	45.37	2.02	62	61.72	2.18
5	22.39	3.66	34	45.92	2.01	63	62.38	2.21
6	24.09	3.40	35	46.47	2.00	64	63.05	2.24
7	25.57	3.20	36	47.02	2.00	65	63.74	2.27
8	26.90	3.04	37	47.56	1.99	66	64.45	2.30
9	28.11	2.91	38	48.10	1.99	67	65.18	2.33
10	29.22	2.80	39	48.64	1.99	68	65.94	2.38
11	30.25	2.71	40	49.18	1.99	69	66.72	2.42
12	31.22	2.62	41	49.72	1.99	70	67.54	2.47
13	32.13	2.55	42	50.26	1.99	71	68.39	2.53
14	33.00	2.49	43	50.79	1.99	72	69.29	2.60
15	33.83	2.44	44	51.33	1.99	73	70.24	2.67
16	34.62	2.39	45	51.87	1.99	74	71.24	2.76
17	35.38	2.34	46	52.41	1.99	75	72.33	2.87
18	36.12	2.30	47	52.96	2.00	76	73.50	3.00
19	36.83	2.27	48	53.50	2.00	77	74.79	3.16
20	37.52	2.24	49	54.05	2.01	78	76.23	3.35
21	38.20	2.21	50	54.60	2.02	79	77.88	3.61
22	38.86	2.18	51	55.16	2.02	80	79.83	3.97
23	39.50	2.16	52	55.72	2.03	81	82.25	4.50
24	40.13	2.14	53	56.28	2.04	82	85.55	5.40
25	40.74	2.12	54	56.86	2.05	83	90.95	7.49
26	41.35	2.10	55	57.43	2.06	84	100.00	13.47
27	41.95	2.08	56	58.02	2.08			
28	42.53	2.07	57	58.61	2.09			

10.19 Appendix XIX – YP FV scaled score-to-measure table

Score	Measure	SE	Score	Measure	SE	Score	Measure	SE
0	.00	13.35	31	43.17	1.97	62	59.07	2.01
1	8.99	7.44	32	43.70	1.96	63	59.63	2.03
2	14.38	5.38	33	44.23	1.95	64	60.20	2.04
3	17.67	4.48	34	44.75	1.94	65	60.78	2.06
4	20.11	3.96	35	45.26	1.93	66	61.37	2.08
5	22.07	3.60	36	45.78	1.92	67	61.98	2.10
6	23.72	3.34	37	46.29	1.92	68	62.59	2.13
7	25.17	3.15	38	46.79	1.91	69	63.22	2.15
8	26.46	2.99	39	47.29	1.91	70	63.87	2.18
9	27.64	2.86	40	47.80	1.91	71	64.53	2.21
10	28.72	2.75	41	48.30	1.90	72	65.21	2.24
11	29.72	2.65	42	48.79	1.90	73	65.91	2.28
12	30.66	2.57	43	49.29	1.90	74	66.64	2.32
13	31.55	2.50	44	49.79	1.90	75	67.40	2.37
14	32.39	2.44	45	50.28	1.90	76	68.18	2.42
15	33.19	2.38	46	50.78	1.90	77	69.01	2.48
16	33.96	2.34	47	51.28	1.90	78	69.87	2.54
17	34.69	2.29	48	51.78	1.90	79	70.79	2.62
18	35.40	2.25	49	52.28	1.91	80	71.77	2.71
19	36.09	2.22	50	52.78	1.91	81	72.82	2.81
20	36.76	2.18	51	53.28	1.91	82	73.96	2.94
21	37.40	2.15	52	53.79	1.92	83	75.21	3.10
22	38.03	2.13	53	54.29	1.92	84	76.61	3.29
23	38.65	2.10	54	54.81	1.93	85	78.22	3.55
24	39.25	2.08	55	55.32	1.94	86	80.13	3.91
25	39.84	2.06	56	55.84	1.95	87	82.50	4.43
26	40.42	2.04	57	56.36	1.95	88	85.74	5.33
27	40.98	2.02	58	56.89	1.96	89	91.06	7.40
28	41.54	2.01	59	57.43	1.97	90	100.00	13.33
29	42.09	1.99	60	57.97	1.99			
30	42.64	1.98	61	58.51	2.00			

10.20 Appendix XX – Peer-reviewed paper

Robertson, AO; Tadić, V, Rahi JS on behalf of the Child Vision Patient-Reported Outcomes (PROMs) Group. Transition from paediatric to adult ophthalmology services: what matters most to young people with visual impairment. Eye, 2017; 10.1038/eye.2017.203.