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Quality of Life and Visual Function in Children with Intellectual Disabilities

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Quality of Life and Visual Function in Children with Intellectual Disabilities

A thesis submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy

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March, 2013

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ABSTRACT

Questionnaires have been developed to allow assessment of latent traits in patients, such as self-perception of health-related quality of life (HQoL) and vision-related quality of life (VQoL), which have not been investigated in a population of children with intellectual disability (ID). This thesis aims to address this knowledge gap by conducting a study on developing an instrument to investigate HQoL and VQoL in children with ID.

Since the instruments applied in the present study were previously validated in children with normal development, the first part of the thesis re-evaluates the psychometric properties of the adapted instruments used for self-perceived HQoL and VQoL and proxy HQoL and VQoL. The construct of subjective perceptions of quality of life is established by identifying the domains of HQoL and VQoL in children with ID. As part of the validation process, Rasch analysis is used to reduce redundant items and to refine the scaling, thus, increasing the validity and internal consistency of the instruments.

A validated instrument is then applied to the subgroups of the study and revealed that there is significantly lower value in the perception of HQoL in children with ID than those without ID, but no difference identified in VQoL. The objective assessment of vision screening demonstrates the fact that children with ID have a higher risk of visual abnormalities. However, visual abnormality does not indicate significant impact on HQoL and VQoL as evidenced in the survey in children with ID. The proxy responses from parental carers are compared to children's subjective views of their HQoL and VQoL. As predicted, the results from parents and children are not strongly associated.

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Finally, I wish to dedicate this thesis to my adorable daughter Danielle Zhiyu.

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CHAPTER 1 LITERATURE REVIEW

Intellectual disability (ID) is classified as a 'metasyndrome' that is categorised by deficits in intellectual functioning and adaptive behaviour. These deficits emerge during childhood, before the development of skills through learning, and influence an individual's ability to participate in daily activities. The group of clinical conditions that comprise ID range from genetic to nutritional, infectious, metabolic or neurotoxic conditions (Salvador-Carulla & Bertelli, 2008). The diagnostic criteria for ID include an intelligence quotient (IQ) below 70, an impairment in adaptive behavioural skills (conceptual, social and practical skills), and an age of onset before 18 (American Association on Mental Retardation (AAMR), 2002). Although the causes of ID can be genetic, physical and/or social, in 60% of people with ID a direct cause cannot be identified (Schalock et al., 2007). The prevalence of ID is around 1.5% in developed countries and may be up to 4% in low income countries, primarily because of insufficient health care services (Durkin, 2002).

A significant proportion of individuals with ID suffer from multiple disabilities and other medical conditions. These disabilities require ongoing access to healthcare services, have life-long consequences for the affected individual and impose a considerable burden on families and caregivers (Salvador-Carulla & Bertelli, 2008). Thus, epidemiological data in children with ID are needed in order to raise awareness among healthcare providers of the impact of early or developmental cognitive impairment, and to provide a sound basis for setting priorities and designing efficient interventions.

Visual impairment is a common disability in children with ID (see Section 1.2). However, the impact of visual impairment on health-related quality of life (HQoL) and vision-related quality of life (VQoL) in children with ID has not been explored. Though there have been some studies on the relationship between visual function and HQoL in children with normal development (ND) (Boulton, Haines, Smyth, & Fielder, 2006; Cochrane, Marella, Keeffe, & Lamoureux, 2011; Felius et al., 2004; Gothwal,

Lovie-Kitchin, & Nutheti, 2003; Quinn et al., 2004), it is not known whether the relationship between visual impairment and HQoL is similar for children with ID.

There are several reasons for the lack of evidence-based information on the impact of visual impairment on HQoL or VQoL in children with ID. One reason is that HQoL assessment is likely to be challenging in this population. The measurement difficulties encountered when assessing subjective reports by questionnaires (also known as instruments) in children with ID include the reliance on self-reporting of functional assessment, which may raise issues such as the validity and reliability of responses (Cremeens, Eiser, & Blades, 2006a), and whether questions are within the cognitive capacity of the individual (Connolly & Johnson, 1999; Cummins, 1997b, 2001; Eiser, Mohay, & Morse, 2000; Hensel, Rose, Kroese, & Banks-Smith, 2002). Another issue is whether proxy responses, which are widely applied in paediatric HQoL studies (Cremeens, Eiser, & Blades, 2006b; Donohue, 2002; Eiser & Morse, 2001a; Quinn, et al., 2004; Saigal et al., 2000), are valid in the assessment of more subjective aspects of functional status (Hatton & Ager, 2002).

This chapter, firstly, clarifies the definition and dimensions of quality of life used in the thesis, reviews previous assessments of quality of life and practical considerations in children with ID. Secondly, it summarises common vision abnormalities and their prevalence in children with ID, and thirdly, reviews VQoL in children.

1.1 Quality of Life and Health-related quality of life

1.1.1 Concepts and dimensions

Quality of life (QoL) is a term that is used frequently both colloquially and as an outcome in clinical research. As a study outcome, QoL is described as a multidimensional construct and an assessment of wellbeing across various domains (Bjornson & McLaughlin, 2001). One of the most widely accepted definitions is that given by the World Health Organization (WHO): "[QoL is defined as] an individual's perception of their position in life in the context of the culture and value system including the psychological and social and value system in which they live and in

relation to their goals, expectations, standards and concerns. It is a broad ranging concept affected in a complex way by the person's physical health, psychological state, level of independence, social relationships, and their relationship to salient features of their environment" (World Health Organization, 2003). This definition has been referred to in subsequent studies (Bonomi, Patrick, Bushnell, & Martin, 2000; Skevington, Lotfy, & O'Connell, 2004).

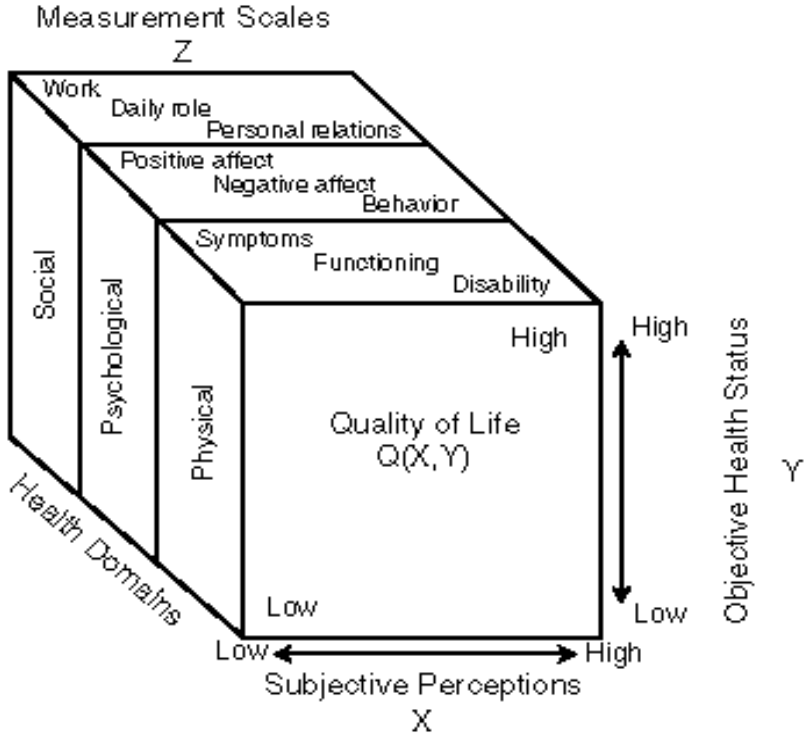
Although HQoL is a sub-domain of QoL, this measure is not well defined (Sherman et al., 2002) and varies from one study to another. HQoL extends across diverse domains and is open to interpretation of personal feelings and satisfaction (Bottomley, Efficace, & Fayers, 2002). Fallowfield (2002) discusses both QoL and HQoL, and has identified four essential domains for HQoL, i.e, physical, psychological/emotional, social and occupational well-being (see Table 1.1), while Testa and Simonson (1996) have described the concept of HQoL using a broader and widely accepted scheme. In Testa and Simonson's scheme (see Figure 1.1), subjective perceptions of health and objective health status are indicated by the X and Y axis respectively. HQoL (Q) is not only dominated by the objective measures of health status (Y) but also by subjective perceptions (X). In other words, people with identical health status might have significantly different views of their HQoL, given individual experience and perceptions of well-being. Q (X,Y) may be measured within several domains as plotted. The measurement scales are designed to cover diverse areas such as work and disability. Within this framework, HQoL consists of several parts, namely physical, psychological and social domains, which are similar to those in Fallowfield's definition. Each domain has a specific focus, for instance in the social domain, the focus will be on people's work, daily life, family relations, and other such areas.

Table 1.1 Primary domains of interest in HQoL

Domain	**Typical items**
Physical and functional well-being	Pain, nausea, weight loss, alopecia
Emotional well-being	Anxiety, depression, body image
Social functioning	Ability to enjoy life, social isolation, engagement with family and friends
Occupational well-being	Ability to work in paid and unpaid employment, ability to perform household duties

Source: Fallowfield(2002)

Figure 1.1 Conceptual scheme of the domains and variables involved in HQoL assessment



Source: Figure 1 Assessment of Quality-of-life Outcome (Testa & Simonson, 1996)

Since neither the domains nor the methods of measurement of HQoL are standardized (Brunner & Giannini, 2003), the meanings and dimensions of HQoL can vary from study to study. A sound HQoL instrument should provide an explicit concept of intention, together with thoughtfully constructed domains for specific study populations.

1.1.2 Measurement of HQoL in children

For those involved in the health care of children, Bradlyn et al (1996) provides a description of HQoL used in paediatric oncology research, which states that "HQoL includes, but is not limited to, the social, physical, and emotional functioning of the child and adolescent, and when indicated, his/her family. Measurement of HQoL must be from the perspective of the child, adolescent, and family, and it must be sensitive to the changes that occur throughout development". This definition is comparable to the one applied in adult populations, except that the impact of developmental changes needs to be considered in the design of the instruments.

Eiser and Morse (2001b) have described key aspects of HQoL in children, emphasizing the factors of subjective perception, multidimensional concepts and comparison of subjective and objective measures in each domain. However, the application of HQoL concepts in paediatric practice in previous studies had identified several challenges. These include a lack of description of domains, confusion about the definition of HQoL, cross-cultural issues, the limited availability of disease-specific measures, discrepancies between the views expressed by children and parents, and other people. Bradlyn and Pollock (1996) agree that a number of barriers exist, and that extra precautions need to be taken when assessing HQoL in children. First, it is difficult to isolate the changes attributed to medical intervention, given children's constant physical and cognitive development. Hence, developmental effects must be accounted for in the case of a large group study with a wide range of ages amongst children. Second, since there are differences between parents, clinical practitioners and children in rating HQoL, misunderstanding of perceptions can affect the results of HQoL investigations when proxy responses are used on behalf of children. Thus, it is essential to implement an instrument based on children's life perspectives, as well as using adults' insights (White-Koning et al., 2005). In addition, there are other concerns with the application of health-related questionnaires in children. For example, children do not have a full understanding of morbidity; there may be diverse interpretation of items; language comprehension may vary and there may be difficulty in the completion of a lengthy instrument (Eiser & Morse, 2001c).

Measurement of HQoL in children has been carried out using both disease-specific and generic instruments. Disease-specific instruments can help the investigators identify subtle changes in patients with certain conditions, while generic instruments allow more broad applications to a variety of groups with different clinical features (Connolly & Johnson, 1999). In addition, the instruments should be applicable to those with as well as those without health problems (Donohue, 2002). A comparison between individuals with and without a disorder (i.e., control group vs. study group) can help investigators to understand whether perspectives across domains of HQoL are general in the population or are different across the subgroups (Felce, 1997).

As previously explained, HQoL is described both objectively and subjectively in domains (Testa & Simonson, 1996). Some researchers prefer to use an objective definition in reflecting social standards and to focus on what a child can do according to his/her health condition (Gothwal, et al., 2003). On the other hand, a subjective assessment will consider children's individual perspectives of their own HQoL. Individuals with identical health status could report differently in subjective assessments (Eiser & Morse, 2001c). An interesting case is that of children with significant physical disabilities, which can adversely influence their lives(Chow, Lo, & Cummins, 2005). Their subjective HQoL has been found to be similar to that of children without a medical condition. Therefore, since the relationship between objective and subjective HQoL is unclear, a thoughtful approach that integrates both indicators across a range of domains seems important in the assessment of HQoL in children.

1.1.3 Quality of life in adults and children with intellectual disabilities

The International Association for the Scientific Study of Intellectual Disabilities uses the term intellectual disabilities to represent a range of conditions known as: mental retardation, developmental disabilities, cognitive disabilities and mental handicap (Schalock et al., 2002). People with ID have commonly been isolated from society in the past (Smith, 1985). In recent years there has been a paradigm shift in attitudes and policies relating to people with ID. This shift incorporates a move from the statutory provision of care to a commitment by the community to provide support for opportunities to work, which requires a closer relationship between people with ID and others in the community. Based on a review of community participation, people with ID are less likely to be employed and involved in community groups and activities than those with other disabilities (Parker & Clarke, 1995). However, even with lower participation level in people with ID, those living in a community setting are more actively involved in the community than those being institutionalized or segregated (Verdonschot, de Witte, Reichrath, Buntinx, & Curfs, 2009a, 2009b). Obviously, people with ID are confronted with obstacles when they contribute to society. Skills set, ability to socialise and education levels are factors that may impose limits on the scope for community engagement in any population. In particular, the high prevalence of visual

impairment in the population of people with ID may impose additional disability to those already with impaired daily functioning (Evenhuis, Sjoukes, Koot, & Kooijman, 2009; Isralowitz, Madar, Lifshitz, & Assa, 2003; Isralowitz, Madar, & Reznik, 2005). As an initial step towards investigating competence in people with ID, previous studies have attempted to explore the health status, quality of life and social involvement of individuals with ID (Bertelli & Brown, 2006; Cummins, 1997b; Hensel, et al., 2002). Generally, it is accepted that people with ID share similar concepts and domains of HQoL as those with normal development (ND)(Schalock, 2004). However, the validity of self-rated measures for HQoL in people with ID is under debate (Cummins, 1997b, 2001; Hatton & Ager, 2002).

In the discipline of special education for children with ID, parents and educational professionals are strongly encouraged to teach children with ID speech and other necessary communication skills at an early developmental stage, with the aim of facilitating their future engagement in the community (Warren, Fey, & Yoder, 2007; Yoder & Warren, 2002, 2004). Innovative educational tools have been used in special inclusive education environments to improve communication skills as early intervention for children with special needs (Panerai, Ferrante, & Zingale, 2002; Panerai et al., 2009). In order to provide timely educational and physical intervention, it is important to study the HQoL of children with ID, and its association with physical anomalies, because such an association would imply that competencies may be further developed if appropriate intervention is implemented at an early stage, and their HQoL may consequently be improved. Nevertheless, several issues remain unresolved in this area.

First, there is a lack of measures of subjective HQoL for children. Most studies have focused on objective assessments of health status, rather than focusing on the child's view of his or her HQoL(Hodgkinson, d'Anjou, Dazord, & Berard, 2002; Liptak et al., 2001). In addition, information about children is often obtained from proxy reports (Fekkes et al., 2000; Schneider, Gurucharri, Gutierrez, & Gaebler-Spira, 2001). The perspectives drawn from experts or parents may not fully match the child's own view. Guyatt et al. discovered that for children under 11 years, it was useful to collect

measures from both children and parents. However for those older than 11, children's responses about their health status are in better agreement with clinical findings than parents' responses about their child's health status (Guyatt, Juniper, Griffith, Feeny, & Ferrie, 1997). Second, it is unclear whether children's self-reported perspectives are repeatable. This is not only because of the possibility of limited comprehension, but is sometimes due to the limitations of the questionnaire development methods themselves.[1] Third, it may be questionable whether the dimensions of HQoL are well balanced and appropriately addressed, and whether they take into consideration the broad background of the children's lives and well-being, in areas such as family impact and social and personal interactions (Young, Rice, Dixon-Woods, Colver, & Parkinson, 2007). Since previous research has shown that children with ID are capable of reliably communicating their own views of HQoL(Dekker, Nunn, Einfeld, Tonge, & Koot, 2002; Ramirez & Kratochwill, 1997), the collection of children's subjective assessment of aspects of their HQoL, where achievable, is highly desirable.

Ingram and Tyack have demonstrated that children understand dichotomous questions ("yes-no") and multiple options in a familiar context, when they are appropriate to their level of cognitive development (Ingram & Tyack, 1979). The ability to respond to a question is highly dependent on the IQ of the respondent and on the question content (Sigelman, Budd, Winer, Schoenrock, & Martin, 1982). It is essential that both the design of the instrument format and its administration are appropriate. To attract children's attention and to make the assessment easier, a pictorial or analogy response category can be successfully integrated into questionnaires used in children with ID (Manificat, Dazord, Cochat, & Nicolas, 1997). The pictorial design can also deter a possibly premature preference of the first choice by simultaneously displaying a number of options. Young children can provide meaningful responses when there are interesting picture designs, and when these designs increase understanding (Harter & Pike, 1984). Hence it is desirable to use pictorial "yes-no" options to ask children with ID questions via non-verbal pointing. The practical principles involved in assessing children with ID

[1] *See Chapter 2 for a discussion of questionnaire development methods*

have been clearly summarized as follows by White-Koning et al (2005).

> "Questions for children with or without ID should be kept as basic as possible, using simple terms and syntax and active sentence constructions. Comprehension is further facilitated by keeping to a single core idea per question, rather than asking children directly to provide comparisons. In terms of readability, factors such as paragraph length, print size, clarity of instructions, and attractiveness of layout are important. QoL instruments should be administered in a familiar environment where the child feels at ease, is not distracted or under pressure, and is allowed ample completion time".

1.1.4 Proxy reports and children's subjective perception

As described above, questionnaires are widely used in paediatric HQoL assessment, offers an efficient means to gather an individual's perspective of their functional status and QoL(Davis et al., 2006). However, previous research raises questions on the reliability of children's subjective responses due to their limited language skills, comprehension and logical judgment (Fekkes, et al., 2000; Vogels et al., 1998). Moreover, interpretation of the subjective responses of children with ID needs to take into account the different perspectives that children with ID have compared with their peers without ID (Vignes, Coley, Grandjean, Godeau, & Arnaud, 2008; White-Koning, et al., 2005) and the changes in these perspectives with different development stages (Jozefiak, Larsson, & Wichstrom, 2009; Jozefiak, Larsson, Wichstrom, Mattejat, & Ravens-Sieberer, 2008). In an effort to overcome some of these limitations, proxy reports from parents or carers have been used as an additional method to assess a child's functional status or QoL.

In some cases, especially in children with ID, the use of proxy responses from parents or carers may be unavoidable and is essential for children with a severe communication deficiency, or a lack of comprehension in interpreting questions or pictorial categories (Schwartz & Rabinovitz, 2003). In addition, proxy responses provide complementary

information that allows comparison between a child's and parents' perspectives of the child's HQoL. However, proxy perspectives may differ substantially from a child's perspective, as shown by a number of studies reporting significantly higher proxy HQoL scores from parents than scores from the children themselves (Jozefiak, et al., 2008; Theunissen et al., 1998), particularly in children with medical conditions. (Upton, Lawford, & Eiser, 2008).

1.2 Prevalence of visual abnormalities

The high prevalence of visual abnormality in children with ID has been well documented (Cregg et al., 2001; Cregg et al., 2003; Maino, M. E. Rado, & W. J. Pizzi, 1996; D. M. Maino, M. E. Rado, & W. J. Pizzi, 1996). In Denmark, Nielsen et al. (2007b) reported a 10.5% prevalence of low vision in children with ID, compared to 0.16% for children in general. For those with an IQ lower than 50, the prevalence is 22.4%. The most common causes of visual impairment are cerebral visual anomalies, optic atrophy, retinal dystrophies and congenital nystagmus. Studies of children with ID conducted in other countries have also been reviewed and the prevalence of visual impairment and blindness is shown in Table 1.2.

Nielsen et al. (2007b) also recorded the prevalence of refractive errors and strabismus in children with ID. Of the 923 children who were examined: significant hyperopia (≥ + 3 DS) occurred in 15.3%; myopia (≥ -0.5 DS) in 10.8%, and astigmatism (≥1 D C) in 20.6%; strabismus in 26.8% (esotropia in 14.9%, exotropia in 10.3%, and other forms, including mixed types, in 1.6%). The prevalence of refractive errors and strabismus is given in Table 1.3.

Table 1.2 Prevalence and diagnoses of blindness and visual impairmentin children with ID

Country (Author, Year)	Number of subjects	Age	Prevalence(%) of blindness (B) and/or visual impairment (VI)	Most common diagnoses
Netherlands (Copper & Schappert-Kimmijser, 1970)	156	5-17	N/A	Congenital cataract, Optic atrophy, Retinal dystrophies
Denmark (Warburg, 1975)	201	1-21 (IQ<70)	N/A	Optic atrophy, CVI, Cataract, ROP
Denmark (Warburg, 1979)	7700	1-21 (IQ<70)	3.9 (B)	Optic atrophy, CVI, Cataract, ROP
Finland (Tuppurainen, 1983)	149	9 – 10 (IQ≤ 70)	7.4 (VI)	Optic atrophy, CVI, Cataract
Germany (Haussler, Bartels, & Strassburg, 1996)	239	4-18	N/A	Optic atrophy, CVI, Posterior ocular diseases
Hong Kong (Kwok, Ho, Chan, Gandhi, & Lam, 1996)	260	0 – 25 (IQ< 25)	25(B)	Optic atrophy, CVI, Retinal degeneration
Taiwan (Chang, Shih, Tseng, Cheng, & Teng, 2005)	68	15 – 23 (IQ ≤ 70)	4.9 (VI)	N/A
Denmark (Nielsen, Skov, et al., 2007b)	923	4-15 (IQ ≤ 80)	9 (B) and 22 (VI)	CVI, Optic atrophy, Nystagmus, Retinal dystrophies

Source: Nielsen, Skov, et al. (2007b)
Blindness (visual acuity ≤ 6/60 (LogMAR:1) each eye)
Visual impairment (visual acuity ≤ 6/18(LogMAR:0.48)each eye)
ROP = retinopathy of prematurity; CVI = cerebral visual impairment

Table 1.3 Refractive error and strabismus in children with intellectual disabilities

Country (Author, Year)	Number of subjects	Age (IQ)	Prevalence (%)	
			Refractive Error	Strabismus
U.S. (Fletcher & Thompson, 1961)	102	<18	29 [1]	24
U.S. (Byron, 1962)	162		17 [1]	12
U.K. (Bankes, 1974)	137		53 [2]	43
Canada (Woodruff, 1977)	168	5-18	54 [3]	21
Finland (Tuppurainen, 1983)	149	9 – 10 (IQ ≤ 70)	42 [3]	26
Saudi Arabia (McQuaid & Arvidsson, 1992)	58	4-18	41 [4]	35
Hong Kong (Kwok, et al., 1996)	260	0 – 25 (IQ < 25)	24 [3]	10
Taiwan (Chang, et al., 2005)	68	15 – 23 (IQ ≤ 70)	57 [3]	27
Denmark (Lisbeth Sandfeld Nielsen, Liselotte Skov, & Hanne Jensen, 2007)	923	4-15 (IQ ≤ 70)	39 [4]	27

Source: Nielsen, Jensen, et al. (2007)
DD = developmental delay
[1] *Level unknown.*
[2] *Hyperopia ≥ + 1.25 DS, myopia ≤ - 1.00 DS, astigmatism ≥ -0.75 DC.*
[3] *Hyperopia> + 2.00 DS, myopia ≤ 0.50 DS, astigmatism ≥ -1.00 DC.*
[4] *Hyperopia ≥ 3.00 DS, myopia ≤ 0.50DS, astigmatism ≥ -1.00 DC.*

A study in Canada, which screened 180 subjects with ID, aged from 9 to 50 years (mean = 24.5 years), found that refractive error was the most common visual anomaly occurring in 58 out of 166 subjects (Karadag et al., 2007). Table 1.4 shows the detailed findings from the study.

Table 1.4 Distribution of refractive error in individuals with intellectual disabilities

Refractive error (dioptres)	Number of subjects	Prevalence (%)
Emmetropia (-1.00 to +0.90)	110	66.3
Spherical error		
Mild hyperopia (+1.00 to +2.90)	20	12.1
Moderate hyperopia (+3.00 to +5.90)	9	5.4
High hyperopia (+6.00 and over)	1	0.6
Mild myopia (-1.10 to -3.00)	20	12.1
Moderate myopia (-3.10 to -6.00)	4	2.4
High myopia (-6.00 and over)	2	1.2
Astigmatism		
Insignificant astigmatism (0 to 1.00)	131	78.9
Mild astigmatism (1.25 to 3.00)	31	18.7
High astigmatism (3.00 and over)	4	2.4

Source: Karadag, et al. (2007)

Kwok et al. (1996) found that the prevalence of visual impairment in children and adolescents with severe ID in Hong Kong was also higher than in the general population (visual impairment: 25%; refractive error: 24%; strabismus: 8% and other ocular diseases: 8%).[2]In Taiwan, Chang et al. (2005) conducted ophthalmic examinations on 68 high school students (15-23 years old) with ID. The visual anomalies included astigmatism (74.4%), myopia (53.7%), amblyopia (29.3%), exodeviation (23.5%), anisometropia (22.0%), blepharoconjunctivitis (20.6%), hyperopia (18.2%), cataract (13.2%), and suspected glaucoma (11.8%). It has been reported that there is a high prevalence of colour vision defects (13/72) in children with Down syndrome (Perez-Carpinell, de Fez, & Climent, 1994), which is higher than children with ND (Cosstick, Robaei, Rose, Rochtchina, & Mitchell, 2005), whilst the findings are not always consistent in children with ID (Woodhouse et al., 2003).

It is well established that ocular and visual abnormalities occur far more commonly in children with ID than in the normal population (Boulton, et al., 2006; Bromham, Woodhouse, Cregg, Webb, & Fraser, 2002; Cregg, et al., 2001; Cregg, et al., 2003;

[2] *Blind patients were excluded, and refractive errors were identified using the criteria that hyperopia ≥ +2.0 D, myopia ≥ - 0.5 D or astigmatism ≥ 1.0 D.*

Maino, 1995; Rahi & Cable, 2003; Stewart, Woodhouse, Cregg, & Pakeman, 2007; Woodhouse et al., 1997). Thus, the question of whether visual abnormality has a significant impact on quality of life is particularly important in the intellectually disabled population. Of course, vision assessment, diagnosis and screening are essential in all children (for example, to prevent amblyopia), but if visual abnormality does impact significantly on HQoL in children with ID, the importance of timely, regular vision assessment in people with ID becomes more apparent (Cochrane, du Toit, & Le Mesurier, 2010). Further, given the high prevalence of visual abnormality in children with ID, timely vision screening (i) can identify treatable ocular disorders, such as refractive error, congenital cataract, etc, and (ii) can identify an immediate rehabilitation service for those with untreatable conditions. Early identification of appropriate rehabilitation services can help children achieve a better quality of life and relatively satisfactory development. Thus, it is important to understand the impact of visual impairment on children's HQoL to inform the design of special education and visual rehabilitation services (Gilbert, Anderton, Dandona, & Foster, 1999; Gilbert & Ellwein, 2008).

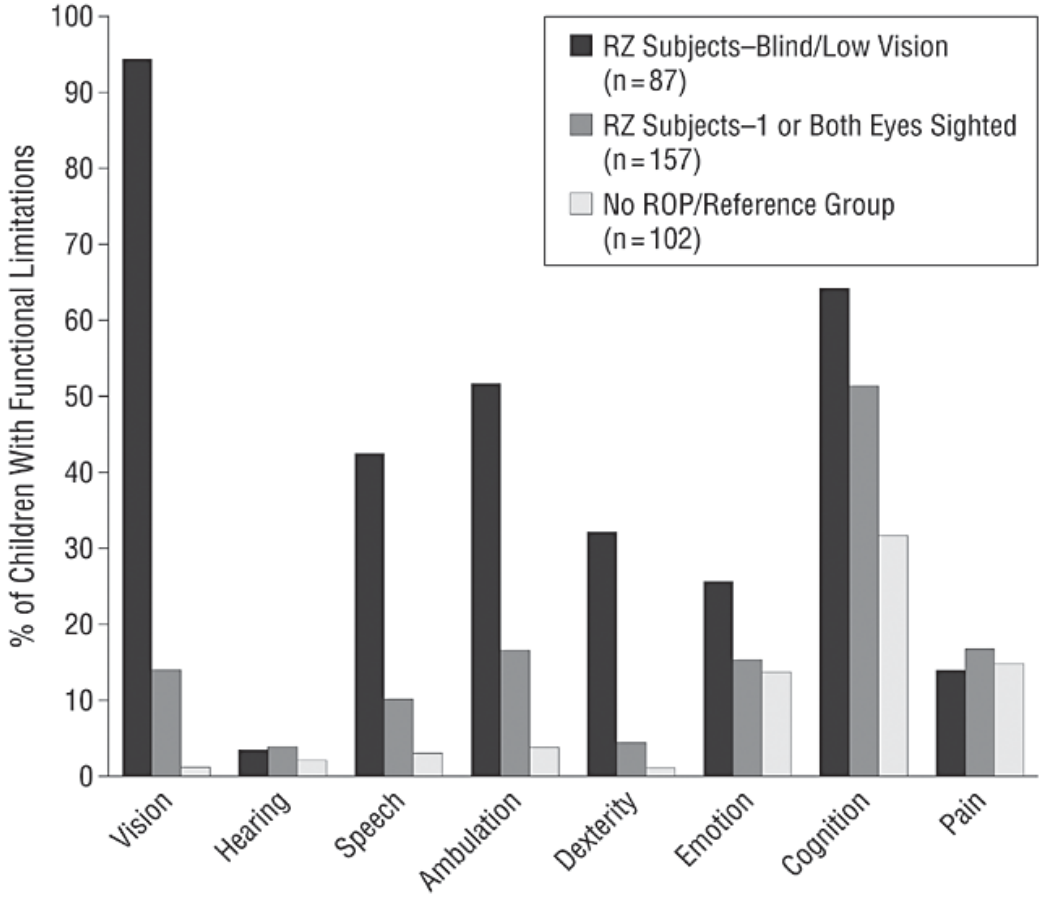
1.3 Impact of visual abnormality on quality of life

The high incidence of visual abnormalities in intellectually disabled children raises questions on the impact of these abnormalities on development. In the non-disabled population, children's early development is adversely affected by visual impairment and its influence on areas of neurodevelopment, learning and social interaction (Kasamatsu, 1982; Singer, 1982). Children with visual impairment are developmentally delayed compared to their peers with normal vision (Reynell, 1978; Sonksen, 1979). Some blind children even show developmental regression and autistic behaviours (Cass, Sonksen, & McConachie, 1994; Rogers & Newhart-Larson, 1989). In children with periventricular leucomalacia, visual impairment has been shown to be a more important indicator in neurodevelopment than motor disability and the visualisation of lesions by magnetic resonance imaging (Cioni et al., 2000). Sonksen and Dale (2002) demonstrated that children's lives and their family functioning are negatively influenced by vision impairment in early childhood.

Boulton et al. (2006) surveyed 79 children aged 3-8 with visual impairment or blindness, and found that half of the subjects with visual problems also had other functional limitations. Thus, visual function might act as an indicator for those at risk of developmental delay or other functional defects. Also, visual impairment limits children's functioning in terms of self-care, mobility and communication. In order to investigate whether these limitations affect children's academic performance or social interactions, Msall et al. (2004) followed up children in special schools with visual impairment due to retinopathy of prematurity. Children with preserved vision were found on the whole to achieve higher academic performance than those without.

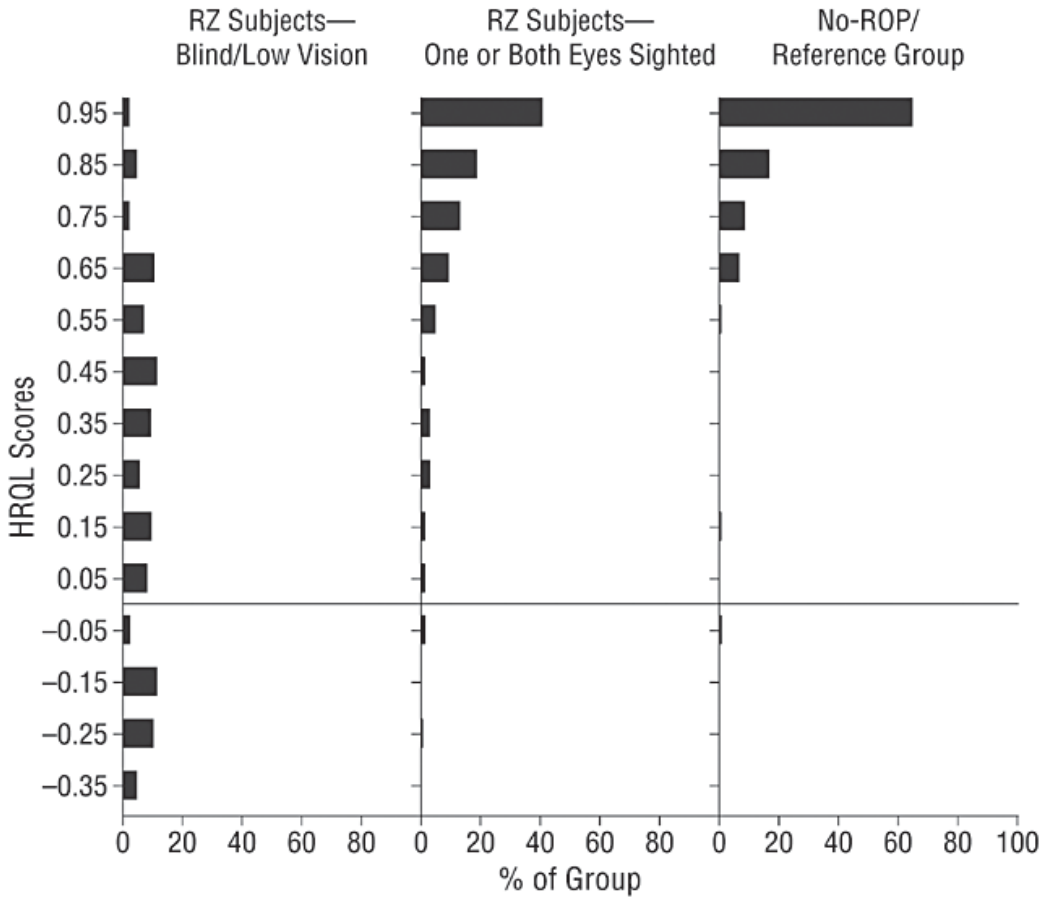
The study *Cryotherapy for Retinopathy of Prematurity(ROP)* has documented the impact of this vision-related condition on children's lives (Quinn, et al., 2004). The majority of children with such visual impairment scored relatively poorly in domains of academic performance, social interactions and independence. HQoL and health status were measured by the Health Utilities Index, which covers elements of health in vision, hearing, speech, ambulation, dexterity, emotion, cognition and pain. The median score for children with ROP (0.72) was lower than that for those without (0.97: $p<0.01$). The median score for the subgroup of children with ROP who were blind or visually impaired was significantly lower (0.27) than the median score for the subgroup of children with ROP who had relatively normal vision (0.87: $p<0.01$). The distribution of functional limitations scores between children with and without ROP is shown in Figure 1.2 and Figure 1.3.

Figure 1.2 Distribution of functional limitations scores between children with and without ROP



Source: HQoL at age 10 years in very low-birth-weight children with and without threshold retinopathy of prematurity (Quinn, et al., 2004).
The vertical axis indicates the percentage of children with functional limitations across the domains of the Health Utilities Index Mark. Children are all 10 years of age. Visual acuity in the randomized (RZ) group - Blind/Low Vision is 20/200 or worse, the RZ group –one or both eyes sighted - is better than 20/200, and the reference group consisted of children who did not develop ROP.

Figure 1.3HQoL scores for children with and without visual impairment in the randomized (RZ) group and reference groups



Source: HQoL at age 10 years in very low-birth-weight children with and without threshold retinopathy of prematurity (Quinn, et al., 2004).
RZ subjects with one or both eyes sighted showed a similar pattern of score distribution to the reference group, whereas RZ subjects with blind/low vision had significantly lower scores.

On the basis of previous studies, severe visual impairments have a negative impact on children's VQoL(Birch, Cheng, & Felius, 2007; Chak & Rahi, 2007; Felius, et al., 2004; Gothwal, et al., 2003; Nirmalan et al., 2004). However, unlike in children with normal intelligence, there has been a lack of research directed towards children with ID.

Visual function has been assessed, however, in a small group of children with neurobehavioral disorder (n=11) using a proxy visual function questionnaire that was designed for children with cerebral palsy, including some with ID. The study found that visual function was better after surgical treatment for ametropia (Tychsen, Hoekel,

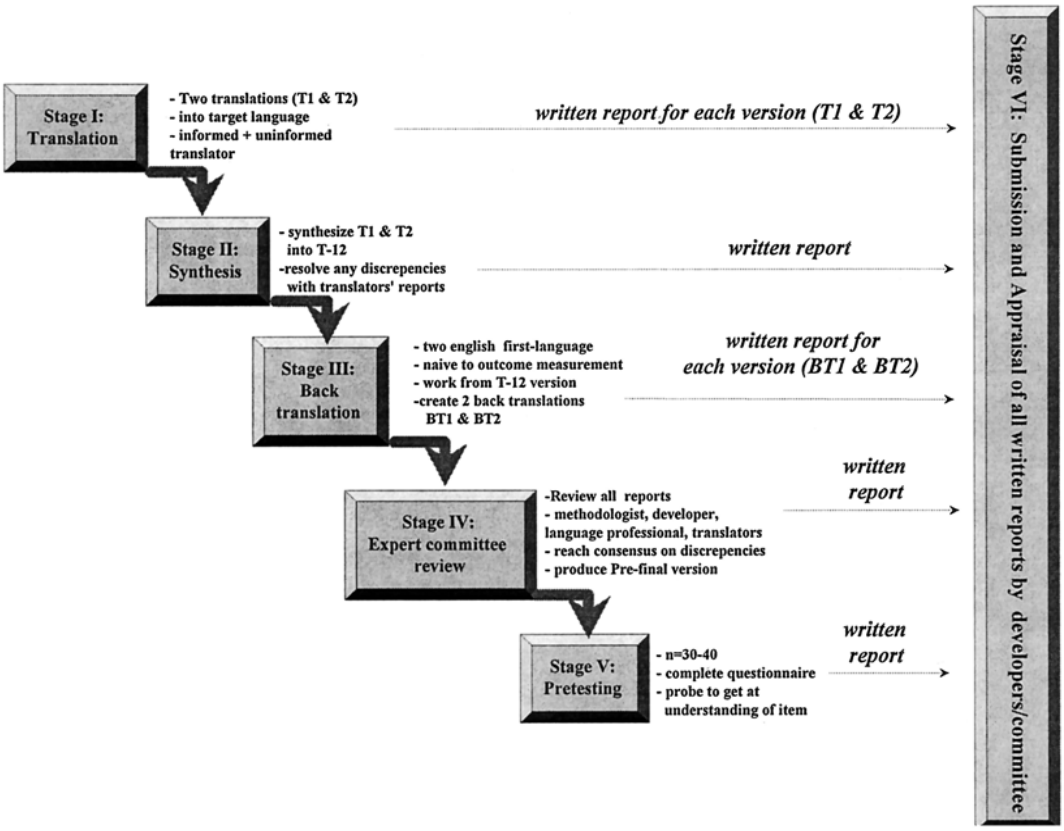
Ghasia, & Yoon-Huang, 2008). Similarly, McCulloch et, al. reported a visual skill inventory to evaluate quality of vision in children with ID (McCulloch et al., 2007).However, there are no instruments specifically designed for assessment of the impact of functional vision on HQoL in children with ID. Therefore, it is not known whether the visual abnormalities that commonly occur in this population present the children with particular difficulties on a daily basis. This is surprising, in view of the relatively high incidence of ocular abnormalities in children with ID. The availability of an appropriate instrument would allow exploration of HQoL in children with ID (Hatton & Ager, 2002). Appropriate instruments should reflect the child's own awareness of any impediment. Ultimately, instruments of this kind may prove valuable clinically, as indicators of the child's level of visual function-related difficulty. It seems likely that visual abnormality might have a different impact on the population of children with ID than in children with ND, since children with ID also commonly have a range of impairments that may impact on their quality of life. For example, communication difficulties, hyperactivity and gaze avoidance have a significant impact on social interaction and on success in education and future career prospects (Koller, Richardson, Katz, & McLaren, 1983; Linna et al., 1999). In addition, people with Down syndrome commonly have defects of the heart and gastro-intestinal tract, among other abnormalities, and in fragile X syndrome, orthopaedic problems are common. Visual abnormality is, therefore, one of a number of factors that may have a negative impact on quality of life in people with ID. It should be noted, however, that visual abnormality differs from many of the difficulties encountered in ID populations, in that its major causes (e.g., refractive errors and strabismus) are treatable, at least during childhood (Cregg, et al., 2001; Cregg, et al., 2003; Lanners, Piccioni, Fea, & Goergen, 1999). Thus, it is possible to improve visual function to normal or near normal levels in many visually impaired children with ID, and it is important to aim for this.

1.4 Instrument adaptation and research methods in a cross-cultural context

Most HQoL instruments have been developed in English and for application in a particular cultural framework, which may limit generalisability. First, simple translation

is not able to replace the functions and objectives of the original instruments given cultural and language differences. Second, the perception of HQoL could vary across cultures. Guidance on the use of HQoL instruments in an international context has been provided in the literature (Herdman, Fox-Rushby, & Badia, 1997, 1998). One of the guidelines suggested by Guillemin (1993) includes the following steps that need to be undertaken: "1) translations; 2) back-translations by qualified people; 3) committee review of those translations and back-translations;4) presenting for equivalence using adequate techniques (with bilingual or monolingual individuals) and 5) re-examination of the weighting of scores. The equivalence of the cross cultural translations should consist of the following semantic, idiomatic, experiential and conceptual equivalence. These guidelines were further developed and adopted by the American Association of Orthopaedic Surgeons (AAOS) Outcomes Committee in translation of outcomes assessment tools (Figure 1.4).

Figure 1.4 Graphic representation of the recommended stages of cross-cultural adaptation of self-report measures



Source: Guidelines for the process of cross-cultural adaptation of self-report measures (Beaton, Bombardier, Guillemin, & Ferraz, 2000)
The AAOS guideline included five stages of instrument adaptation. Stage one of "Initial Translation" is to conduct forward translations by different qualified translators and identify ambiguous wording in the translation process. Stage two of "Synthesis of the Translations" is to gain consensus on the results of the translations and to develop a synthesized version. Stage three of "Back Translation" is to check the validity of the translations by comparing the translated back version of items with the original items included in the instrument. Stage four of the "Expert Committee Review" is to involve a group of healthcare, language and surveyor expert to consolidate the objectives, domain and meaning of each item to reach cross-cultural equivalence.

In addition to the proposed translation process, instruments need to undergo validation to investigate the psychometric criteria, in addition to the solid translation process. It is possible to provide normative scales for a particular population using a valid instrument (Bullinger et al., 1998). Many instruments that have been applied in cross-cultural settings are deemed to require re-evaluation of the psychometric properties using a series of qualitative approaches that include focus groups or an advisory panel, in order to define the construct of the scale and interpretation of the items. In addition,

quantitative approaches, attributed to classical test theory or Rasch analysis, are needed to confirm the reliability and validity of the scales (Fukuhara, Bito, Green, Hsiao, & Kurokawa, 1998; E. L. Lamoureux, Pallant, Pesudovs, Hassell, & Keeffe, 2006; E. L. Lamoureux et al., 2007; E. L. Lamoureux, Pesudovs, Thumboo, Saw, & Wong, 2009; L. Li, Wang, & Shen, 2003; Lin & Chie, 2009; Pesudovs, 2005; Pesudovs, Burr, Harley, & Elliott, 2007; Schmidt et al., 2006; Schmidt, Muhlan, & Power, 2006).

1.5 Classical test theory and item response theory

Modelling and theory have been widely applied in social science to assist with the interpretation and explanation of certain phenomena or mechanisms of underlying traits, assessed by psychological test or questionnaire. In the development of questionnaires, classical test theory (CTT) has been successfully implemented and was the dominant approach until the emergence of item response theory (IRT) (Hambleton, 2000). Currently, both CTT and IRT can be found in the literature as the framework for analysing and developing instruments (Felius, et al., 2004; E. L. Lamoureux, Saw, et al., 2009; Vianya-Estopa, Elliott, & Barrett, 2010).

1.5.1 Classical Test Theory (DeVellis, 2006)

In order to gain insight into the underlying traits of a concept that is not directly observable (such as life satisfaction or HQoL), CTT determines a set of principles to evaluate the extent to which observable information (such as a score in a questionnaire) can estimate the concept that is not directly observable (de Boer et al., 2004).

A fundamental principle of CTT is an assumption that an observed score is determined by the actual state of the underlying trait plus error attributed to all other influences on the observed information. Hence, scores obtained from a valid questionnaire could be reflective of the true underlying trait possessed by respondents, the knowledge of which is critical to answer the research question that a questionnaire is designed for(Hays et al., 2003). There are many opportunities for error when a series of questions is used to interpret a respondent's experience or feelings accurately. In CTT, the error is deemed

to be random, thus, while the error increases item variability, the mean of the error tends to be zero and has a limited effect on the mean score of an item. Item reliability provides an indication of the extent to which the measured score is a true reflection of the trait.

CTT assumes that the items that make up a score are strictly independent of each other (i.e. parallel items). Hence, the items within a domain are assumed to be indicative of the same underlying variable (i.e. unidimensional), errors associated with the items are independent of the true score (i.e.: random error), item covariance with the true score must be equal across items, and each item is determined to the same degree as any other item by the variable (the underlying trait to be tested). The estimate of item reliability can be derived from correlations between items. The scale properties and procedures for validating the questionnaire under classical test theory will be discussed in Chapter 2 (Section 2.4.3 and Section3.1.3.4).

CTT has remained popular despite the emergence of newer measurement approaches. However, the disadvantages of CTT are notable. First, given that the fundamental basis of CTT is the strength of correlations among items, scales appear lengthy with redundant items. Second, CTT doesn't allow scrutiny of item characteristics. As such, the differentiation between items among diverse populations or subject groups is concealed. These disadvantages of CTT may limit the application of CTT in outcome research.

1.5.2 Measurement and raw scores (Mallinson, 2007)

The traditional psychometric validation process emphasizes internal consistency, reproducibility and validity, and focuses less on unidimensionality, hierarchical order and equal interval scaling, which are essential features of a measurement in outcome research. For example, many visual function questionnaires, which are designed using the Likert scale to arbitrarily record subjective responses and create a total score (raw score), may not exhibit the property of a measurement. The questionnaire raw score does not always reflect the trait or the change in trait. Thus, alternative measurement

scales for outcome research should encompass the following essentials.

Hierarchical Order Like physical measurements such as temperature, length, and weight, the measurement should be expressed as “more” or “less”. The underlying trait needs to be addressed in a similar way so that the items designed reflect the different levels of a certain trait.

Equal interval is often referred to as a “unit” in measurement research. Rating scales that assign numbers to response categories such as Very happy, coded as (1), Happy (2), Not happy (3), or Not happy at all (4) will need some transformation before a difference in the rating scale in response to a trait or a function can be claimed. When using units to compare the amount of a trait,it is critical thatan appropriate origin (zero-point) is set as a benchmark for the measure.

Unidimensionality Most types of match/statistic techniques require figures from the same dimension, otherwise the comparison across the subjects appears meaningless. A single domain of a questionnaire is assumed to be unidimensional and, thus, is suitable for mathematic calculation.

Conjoint additivity Cancellation and commutation are the basic principles of conjoint additivity, which is a prerequisite for QoL measurement. The cancellation axiom requires that equal quantities remain equal after the same amount has been subtracted from each. The commutability axiom requires that a series of addition operations always produce the same result, regardless of their sequence. Rasch measurement models indicate whether data comply with these rules. If so, the QoL data can be used for valid measurement. Additive combination is addressed by the joint interaction of observer and observation under the constraints of several quantitative criteria, and is called simultaneous conjoint measurement(Wright & Masters, 1982).

1.5.3 Rasch analysis

Georg Rasch provided a solution to the disadvantages of CTT in 1956 by taking ordinal

level data and translating it into interval measures with the essential features of measurement(Rasch, 1960). His model for dichotomous items that specify the log-odds of success, capitalizes on likelihoods or probabilities, and incorporates the difference between subject ability and item difficulty. Rasch conceptualizes the obtained raw score as the difference between item difficulty (Di) and person ability (Bn). This difference can be shown to be equivalent to the log of the ratio of the probability of being able to do the item, to the probability of not being able to do the item. In his formulation, difficulty and ability are expressed in abstract and interval level units called "logits" (log-odds units). The Rasch model is a measurement abstraction that provides tools to demonstrate the property of the relationship between qualitative observations and quantitative units and supports mathematical formulations(Andrich, 1988). The validation of questionnaires via Rasch analysis is described in Chapter 2 (Section 2.4.4).

In summary, CTT uses a questionnaire score to measure an underlying trait, such as perception of HQoL. However, CTT has been criticized for its assumption that the recipient has similar characteristics regardless of an individual's level of ability or the difficulty of the test. Thus, although CTT is a useful method for identifying instrument domains in questionnaire development and for investigating the relationship between variables included in an HQoL instrument, it may be less useful for monitoring HQoL or comparing progress of a clinical intervention.

Rasch analysis was developed to overcome some of the pitfalls of CTT. Rasch analysis assumes that the latent trait or competence of a respondent is independent of the difficulty of the test. Thus, the probability of selecting an answer to a certain item can be modelled. According to Rasch analysis, a respondent with higher ability is more likely to score correctly in an item according to the common scale shared by both the person and the items.

Since the 1970s, Rasch analysis has been used widely to develop survey instruments and to help guide the validation of instruments. In recent years, this method has become popular for the development of questionnaires in ophthalmic science

(Bezruczko, 2005; E. Lamoureux & Pesudovs, 2011; E. L. Lamoureux, Fenwick, et al., 2009; E. L. Lamoureux, et al., 2006; E. L. Lamoureux, et al., 2007; E. L. Lamoureux, Pesudovs, et al., 2009; Massof & Rubin, 2001; Pesudovs, et al., 2007). Rasch modelling stems from the theory: $log\left(\frac{probablity\ of\ success}{probablity\ of\ failure}\right) = ability - difficulty$, where the probability of success / probability of failure is called "success odds". By this theory, the relationship between person ability and item difficulty can be described on a common interval scale using logit (logarithm of odds) units. When the data are fitted to the model, a common linear scale is established; the person-to-person, item-to-item or item-person differences are represented using logits on the same scale (Wright & Masters, 1982). Fit statistics are used to investigate the degree to which the data from items and people conformed to the expectations of the Rasch model (i.e., easy items are easy for all people) (Pesudovs, Garamendi, Keeves, & Elliott, 2003). Rasch analysis can identify poor targeting of item difficulty to respondents, and allows items to be identified for exclusion. One other essential feature of Rasch analysis in instrument development is the conversion of responses, which are usually in ordinal ranking, into interval data by mathematical transformation (Wright & Masters, 1982). By transforming the data, multivariate analyses, which require data in interval scales, can then be conducted.

Investigation of validity is addressed by three fundamental aspects of Rasch measurement (Smith & Smith, 2004). First, the data should fit the model, as one of the model requirements and measurement properties is unidimensionality, which demonstrates that all items measure a single construct. In addition, the measurement property of additivity, means that data may be converted from the underlying trait as measurement units in an interval scale, called logits. These interval scales can be used for later multivariate analysis. Second, construct validity is assessed on the basis of the order of items and persons on a common scale. This feature helps with the interactive analysis between the participants and the items, which reflects the targeting of the items towards a specific population. Third, fit statistics of the item and person indicates the difference between the observed response and the expected response to each item from

each participant.

Rasch analysis also provides guidelines on the design of rating scales, which influence the quality of data obtained from the scale. The term ‘category’ refers to the response options. When investigating category functioning, category frequencies and average measures may be evaluated. Category frequency provides information about how well each category is used across the items. A redundant category is indicated by a low frequency. Another parameter to consider is the average measure to indicate problematic categories. The average measure of a category indicates the mean ability of the person who chooses the category. The average measures are expected to increase monotonically with the category, in other words, choice of a higher category indicates higher ability/stronger feelings (or to take the AUQUEI example, happiness) related to a concept. If this pattern is violated, redundancy of the category is again considered.

Category threshold, or step calibration, describes how difficult it is to choose one option over another, and is another indication of category functioning. As with the average measures, category threshold is expected to increase monotonically. The probability curve, generated by Rasch analysis, plots the probability of a category being endorsed against the difference between person’s ability and item difficulty. For a large difference, an extreme category is likely to be selected. Each category should have a distinct peak, indicating that at some point of the measured variable, it is the most probable response. If categories were disordered, this would indicate that the range of categories and/or the names of categories were not applicable to the respondents. Disordered categories provide an indication that categories should be collapsed (combined) (E. L. Lamoureux, et al., 2006). Consequently, combining underutilised categories with adjacent ones can often improve category function. For example, a polytomous scale of life satisfaction (very happy, happy, not happy, not happy at all) may have been used while a scale with fewer categories (very happy, happy, not happy) would have been more applicable. This is due to the category names such as “not happy” or “not happy at all” being interpreted differently by the respondents and by the researchers.

The quality of rating scales can be evaluated by another criterion (i.e., fit statistics of the category). According to Bond and Fox (Bond & Fox, 2007), an outfit mean square greater than 2 implies that there could be more misinformation than information in the data, meaning that the particular category is introducing noise into the measurement process. A category of this kind, therefore, should be investigated further and possibly eliminated from the measurement.

In brief, a set of techniques are applicable for rating scale diagnostics, including category frequency, average measures, threshold estimates, probability curves, and category fit. These techniques should be used in combination to revise the rating scale (Bond & Fox, 2007).

1.6 Significance and aims of the study

The questionnaire-based approach, widely used in HQoL assessment, offers an efficient means by which to gather individual perspectives (Hartley & MacLean, 2006). However, hurdles exist in obtaining and understanding individual perceptions surrounding the abstract concept of HQoL in people with ID (Felce, 1997). The usefulness of questionnaires for assessing HQoL in children with ID has been questioned (Shelly et al., 2008; Vignes, et al., 2008). The failure of questionnaires might be due to inappropriate wording and format for children with ID, given their limited capacity for comprehension (White-Koning, et al., 2005). On the other hand, as stated in section 1.1.3, it is generally accepted that people with ID have clear concepts of their HQoL (Schalock, 2004). The perception of HQoL can be accessed, once the relevant concepts have been successfully communicated to people with ID and with reliable responses from them afterwards (Cummins, 1997a, 1997b). Similarly, as a first step towards the study of HQoL and VQoL in children with ID, it is essential to convert the abstract concept into a comprehensible form for those children to understand. This will enable children's individual perceptions of their HQoL and VQoL to be addressed in a set of items and used for further analyses. Thus, the availability of an appropriate instrument would allow exploration of HQoL in children with ID (Hatton & Ager, 2002).

Since the instruments applied in the present study (see Chapter 2) were previously validated in children with normal development, the first aim of this study is to re-evaluate the psychometric properties of the adapted instruments used for self-perceived HQoL and VQoL and proxy HQoL and VQoL in children with ID. The second aim is to investigate the impact of visual abnormality of children with ID on their HQoL and VQoL.

The study reported in this thesis fills an important gap in the understanding of HQoL and VQoL in children with ID. The instruments developed in this study can provide useful measures of subjective visual function alongside more objective clinical measures.

1.7 Research questions and hypotheses

According to the aims of the study, the specific research questions are as follows. The objectives and hypotheses are detailed at the beginning of each chapter.

1. Can a valid measurement of HQoL and VQoL be made in children with ID?
2. What is the difference, if any, in self-perceived HQoL and VQoL between children with and without ID?
3. What is the difference, if any, in proxy-perceived HQoL and VQoL between children with and without ID?
4. What is the impact of visual abnormality on HQoL and VQoL in children with and without ID?

The hypotheses of the study are:

1. HQoL and VQoL can be assessed in children with ID following appropriate instrument development and validation.
2. Children with ID have lower self-perceived HQoL and VQoL than those without ID.
3. Parents of children with ID perceive their children as having lower HQoL and

VQoL than parents of children without ID.

4. Visual abnormalities have a measurable negative impact on HQoL and VQoL in children with and without ID.

CHAPTER 2 GENERAL METHODOLOGY

This chapter consists of five sections: Section 2.1, “Overview and design of the study” provides an outline of the present research design; Section 2.2, “Questionnaire development” describes the composition of the advisory panel for instrument development, the original instruments used for development, how participants (subjects) were selected, recruited and grouped, and how the questionnaire was administered; Section 2.3, “Vision screening”, describes the vision screening procedures used in the study; Section 2.4, “Pilot study - Instrument development and validation” outlines the methods of questionnaire validation used in the pilot study; and Section 2.5, “Main study - Exploration of the impact of visual abnormality on quality of life in children with ID” describes the methods used to assess the performance of the instruments in each cohort group in the main study.

2.1 Overview and design of the study

2.1.1 Background and overview

As reviewed in Section 1.3 “Impact of visual abnormality on quality of life”, there is scope for more research into the effect of visual abnormality on the HQoL and VQoL in children with ID. This area was explored in the present study using instruments (questionnaires) to assess subjective HQoL and VQoL in children with ID and to assess the parents’ view of their children’s HQoL and VQoL (proxy view). The impact of visual abnormality was determined from a range of HQoL and VQoL domains identified in sample groups of the target population.

2.1.2 Focus of the study

Existing instruments for measuring HQoL either do not focus on vision-related problems, or are not specific for children with ID, and thus, are inappropriate for this application. In order to address the concepts of HQoL and VQoL, an appropriate instrument needs to be developed before investigation of the impact of visual abnormality on HQoL and VQoL in children with ID is possible.

The present study aims to develop a valid instrument for children with ID in order to address the research questions as listed in Section 1.6.

2.1.3 Study design

This was a prospective study, conducted by a single investigator at multiple centres in Australia and China. The HQoL and VQoL of children with ID were measured using self-completed questionnaires and a proxy questionnaire completed by their parents. A pilot study was conducted to devise valid instruments for use in the assessment of the impact of visual abnormality on HQoL and VQoL in children with ID. In the main study, questionnaire responses were compared across children with ID and a control group of children with ND. Further, cohort groups in both the study and control groups were categorised by gender, age, and severity of ID (defined by the intelligence quotient, IQ). This categorization allowed investigation of the extent to which these factors contribute, if at all, to the impact of visual abnormality on HQoL and VQoL in participating children. In this way, HQoL and VQoL in children with and without ID were explored. In addition, the association between the presence of visual abnormalities, as indicated by vision screening, and scores from HQoL and VQoL was investigated.

2.2 Instrument adaptation

2.2.1 Advisory panels for instrument adaptation

In order to assist with the adaptation of HQoL and VQoL instruments for application in children with ID, two panels with relevant experience and expertise were formed in Australia and China. For the panel in Sydney, the members included two paediatricians and one speech therapist from the Diagnostic and Developmental Assessment Service, St George Hospital in Kogarah; one teacher of special needs students from Arncliffe Public School, Arncliffe, NSW; one paediatric optometrist from the Optometry Clinic at UNSW, and a mother of a child with Down syndrome, who also works with the Down Syndrome Association, NSW. For the panel in Shanghai, the members included two paediatricians and one paediatric ophthalmologist from Shanghai Children's Medical Centre; two teachers at Lujiazhui Primary School for Special Education and a mother of

a child with cerebral palsy and ID, who also works as a nurse in a paediatric hospital department. A registered translator was involved in the English - Chinese translations of the instruments.

2.2.2 Objectives of the instrument

The instruments used to assess HQoL and VQoL in children with ID in this study were based on the concepts of HQoL and VQoL that were extracted from the literature on HQoL and VQoL in children with ND. Three instruments, described in section 2.2.3, were identified to reflect the concepts of HQoL and VQoL. The domains and original questions from each of these instruments were modified to suit children with ID. After several modifications of the concepts of HQoL and VQoL for children with ID, the advisory panels assisted in refining the domains and questions to be included in the adapted instruments. The domains of HQoL and VQoL are listed in Table 2.1.

Table 2.1 Domains of HQoL, VQoL and proxy VQoL[a]

HQoL[b]	VQoL[c]	Proxy QoL[d]
Health status	Vision quality	General health
Social life	Vision-related activities	General vision
School life	Academic performance	Competence
Family life		Personality
		Family impact
		Treatment

[a]*Definitions are adapted from (Felius, et al., 2004; Gothwal, et al., 2003; Manificat, et al., 1997) the conceptual domains of each quality of life construct are listed in each column.*
[b]*HQoL represents children's satisfaction with aspects of their life that are related to their own health status;*
[c]*VQoL represents children's subjective assessment of their daily functional visual performance*
[d] *Proxy QoL represents the parents' subjective assessment of their children's quality of life and the impact of visual impairment on their lives and their family.*

Based on a review of the current literature (see Section 1.1) and the comments from the advisory panels, the objectives of the questionnaire development were:

1. To describe children's perception of their own HQoL, which covers aspects of awareness of their own health status, daily life at school, home and social life.
2. To describe the children's view of their own visual status or vision quality (clarity of seeing objects), daily activities and academic performance, all of which require a certain level of vision quality.

3. To describe the parents' perception of their children's general health and vision status, competence, ability and personalities related to vision and their impact on family life.

2.2.3 Instruments selection and preparation

The Medline (1980-2005) database was searched combining key words in the following domains: child, vision, questionnaire, health, intellectual disability, quality of life. The references retrieved were reviewed for possible relevant articles and questionnaires for assessment of HQoL and VQoL in children with ID. All abstracts were reviewed to assess whether the articles should be included in the review. Inclusion criteria were:

1. The article should describe questionnaire development for children with ID, or
2. The article should pertain to information regarding the current eye health care in children with ID, or
3. The article should include VQoL or HQoL assessment in children

Articles relating to eye health predominantly covering clinical features (e.g. cataract, conjunctivitis, etc.) were excluded from the review. There were no language limitations in the search. The Medline search yielded 13 abstracts with key words of child and health and intellectual disability and quality of life and questionnaire, 0 abstracts with key words of child and vision and intellectual disability and quality of life and questionnaire, and 26 abstracts with key words of child and vision and quality of life and questionnaire.

The selection of instruments was based on the research questions of the present study, in which views from both children and parents are required to collectively reflect the HQoL and VQoL in children with ID. The instruments with the domains as identified in the discussion with advisory panel were reviewed. The design and format of each instrument were derived from the panel review. In order to help children interpret question response options appropriately, cartoon face design of response options was suggested to be used in the new questionnaire. Thus, three existing instruments for the assessment of HQoL and VQoL in children with ND (Felius, et al., 2004; Gothwal, et al., 2003; Manificat, et al., 1997) were identified to form the basis for instruments to be

used in the present study. The advisory panels helped with the item (question) selection and modification of the instruments using the HQoL and VQoL domains listed in Table 2.1. A brief description of the instruments used in the study is provided below.

2.2.3.1 Auto questionnaire enfant image (AUQUEI)

An illustrated children's self-completion questionnaire, the AUQUEI, was used to assess children's subjective views of their HQoL. The original AUQUEI (prior to modifications made in the present study, as discussed in Section 3.3.1) consists of 26 items, which address children's perceived satisfaction with a range of life domains including: autonomy (7 items), leisure activities (6 items), function (6 items) and family life (7 items). The child chooses a picture in response to each question to depict their satisfaction level. Response choices are pictures that are intended to indicate the following states: "very happy", "happy", "not happy", and "not happy at all". The results are treated as a 4-point Likert scale with the rating scores from 1 to 4 (Manificat et al., 2003; Manificat, et al., 1997). The AUQUEI was developed in the French language, validated using CTT and applied in children with human immunodeficiency virus (HIV). The instrument was able to distinguish quality of life differences between healthy children and sick children (HIV positive).

2.2.3.2 LV Prasad-functionalvision questionnaire (LVP-FVQ)

The 19-item LVP-FVQ is used to assess children's subjective views of their VQoL. The original LVP-FVQ covers aspects of visual function and vision-related activities, such as distance vision (6 items), near vision (6 items), colour vision (2 items), and visual field (5 items). Each item was asked in a "Yes" or "No" format. If the answer was "No," the response was recorded as "No difficulty," and the score for that particular question was zero. If the answer was "Yes", then the participants were instructed to select a level to describe the difficulty they experienced in performing the task using a 5-point Likert scale with options such as "1. A little difficulty" to "5.Unable to do the activity due to visual reasons". An additional response of "not applicable" was offered, which would be treated as missing data for the purpose of statistical analysis (Gothwal, et al., 2003; Nirmalan, et al., 2004).In order to simplify the options for the children with ID, a

dichotomous scale, as had been used by Gothwal et al., is applied in the present study. The layout of the response options and their adaptation process is discussed in Section 3.1.4. The LVP-FVQ was validated using Rasch Analysis in Indian school children aged 8-18 years with visual impairment. The instrument provides valid measurement of functional vision.

2.2.3.3 Children's visual function questionnaire (CVFQ)

CVFQ is completed by caregivers and thus provides an important supplement to questionnaires completed by the children themselves. The original instrument consists of 45 items, which covers: general health (1 item), general vision (2 items), competence (19 items), personality (10 items), family impact (8 items) and treatment (5 items). The 5-point Likert scales used for the response options include:

- Quality scale with options of "1. Excellent", "2. Very good", "3. Good", "4. Fair" and "5. Poor";
- Frequency scale with options of "1. Never", "2. Once in a while", "3. Sometimes", "4. Often" and "5. Always";
- Agreement scale with options of "1. Strongly disagree", "2. Disagree", "3. Not sure", "4. Agree" and "5. Strongly agree";
- Difficulty scale with options of "1. No difficulty because of eyesight', "2. A little difficulty because of eyesight", "3. Moderate difficulty because of eyesight", "4. Extreme difficulty because of eyesight" and "5. Cannot do this at all because of eyesight".

The CVFQ includes two versions: for older (from 3 to 7 years) and younger (under 3 years) children in order to consider the impact of a child's development on daily vision-related activities (Felius, et al., 2004). Both versions were used in the present study because the cognitive ability of the children with ID was likely to be less predictable than in the normal population. The instrument was validated using CTT in North American parents, whose children have various eye diseases. The successful applications of the instrument demonstrate the impact of visual abnormalities on

children and their family life.

The development of the original instruments and their applications are described in prior publications (Birch, et al., 2007; Felius, et al., 2004; Gothwal, et al., 2003; Manificat, et al., 2003; Manificat, et al., 1997; Nirmalan, et al., 2004). Three examples of items from each of the three instruments are given in Table 2.2., and further details of the original instruments are provided in Appendix 1. Preliminary instruments were trialled in a group of children with ID and the appropriate modifications were subsequently made (this pilot work is described in full in Section 3.2.1). An information sheet was attached to the questionnaires to collect demographic data from each participant. A set of instructions was given to parents or carers so that they could assist in the completion of each questionnaire. The modified instrument details can be found in Appendix 2.

Table 2.2 Example items from the AUQUEI, LVP-FVQ and CVFQ[a]

AUQUEI	LVP-FVQ	CVFQ
When I am having dinner with my family, I feel…	Can you kick a ball when you play?	Do you worry about your child's eyesight?
My brothers and sisters make me feel…	Can you find food on your plate when eating?	My child can locate a small piece of food and grasp it.
At school, I feel…	Can you see bus numbers clearly?	My child can recognise faces (friends, relatives) across a room.

[a]*Items in the modified AUQUEI, LVP-FVQ with simple wording for children, and CVFQ for parents, were devised to assess children's subjective HQoL and VQoL and proxy VQoL, respectively.*

2.2.4 Ethical approval

This study received approval from the Human Research Ethics Committee of the University of New South Wales, and was formally ratified by the school principals of the Lujiazhui Primary School for Special Education, the Shangnan Primary School for Special Education, the Shanghai Low Vision School, and the Pudong Primary School in Shanghai. Signed informed caregiver consent was obtained before participation (See Appendix 4).

2.2.5 Participants and recruitment

A heterogeneous sample of participants was recruited with various levels of visual function, severity of ID, developmental age and gender. The eligibility criteria are set as follows:

2.2.5.1 Inclusion criteria

Eligible participants with ID were aged from 8 years to 18 years, with an IQ from 40 to 70, based on the Wechsler Intelligence Scale for Children-Revised (Li, Jin, Vandenberg, Zhu, & Tang, 1990). The diagnosis of ID and measurement of IQ level were obtained from school records of the student participants or clinical records of the patient participants. Eligible participants with ND were aged 4 to 9 years with no diagnosed or suspected intellectual disability or developmental delay. In all participants the required best-corrected vision in the better eye was no poorer than 6/120 (LogMAR: 1.3).

2.2.5.2 Exclusion criteria

Participants with best-corrected visual acuity worse than 6/120 (LogMAR; 1.3) in the better eye, may not be able to differentiate the cartoon face options included in the instrument and were therefore excluded from participation in the study. In addition, Children who were unable to understand the questions and instructions were excluded, irrespective of their IQ. The number of children excluded for this reason was not recorded.

2.2.6 Recruitment

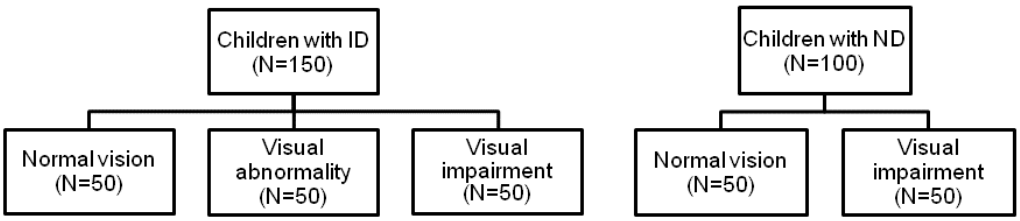
Due to limited resources and access to potential participants in Australia, the present study was conducted at multiple centres to ensure a sufficiently large sample. In the pilot study, participants were recruited from the student cohort of the Lujiazhui Primary School for Special Education in Shanghai, China. Participants were classified by severity of IQ according to the school record. Participants with an IQ of 55 to 70 were classified as having mild ID and those with an IQ of 40 to 54 were classified as having moderate ID (Shalock, Borthwick-Duffy, Buntinx, Coulter, & Craig, 2010).

Following the pilot study, participants with diverse characteristics were recruited from China and Australia for the main study. In Shanghai, children were recruited from the student population of the Lujiazhui Primary School for Special Education and the Shangnan Primary School for Special Education and from the patient population of the Shanghai Children's Medical Centre. In Sydney, participants were recruited from the Diagnostic and Developmental Assessment Service, St George Hospital in Kogarah and the Down Syndrome Association, NSW. As a control group, children with ND were recruited from the Shanghai Low Vision School, Pudong Primary School in Shanghai, China and the Optometry Clinic, UNSW in Sydney, Australia.

2.2.7 Participant grouping

As shown in Figure 2.1, participants in the main study were allocated to either the ID or ND group. Children in each of these groups were of equal number (N=50), mixed gender and similar developmental age ranges. The developmental age range for this study was calculated by multiplying the chronological age by IQ/100 (Mosby, 2009) and was based on the assumption that the average IQ scores for the participants of the ID group was 50. Thus, in the present study, the chronological age range of children with ID (4 to 9 years) was half that of the chronological age range of children with ND (8 to 18 years).

Figure 2.1 Participant grouping in the main study



ID: Intellectual disability

ND: Normal development

The study group comprised children with ID who were allocated according to their visual status into three groups. The control group comprised children with ND who were allocated to two subgroups with normal and low vision (Figure 2.1).

The World Health Organization (WHO) definition of visual impairment (Table 2.3) was used, and adapted to define a subset of visual abnormality for the ID group (Cochrane, et al., 2011; World Health Organization, 2003). In the ND group, children with normal vision (corrected VA equal to or better than 6/9.5 (LogMAR: 0.2)) were allocated to the normal vision subgroup and children with visual impairment (corrected VA worse than 6/19 (LogMAR: 0.5)) were allocated to the visual impairment subgroup.

In children with ID, those with VA better than 6/9.5 (LogMAR; 0.2) were allocated to the Normal Vision group, VA 6/9.5 to 6/19 (LogMAR: 0.2 to 0.5) to the Visual Abnormality group, and those with poorer VA than 6/19 (LogMAR: 0.5 to 1.3) to the Visual Impairment group. The reason for the different numbers of subgroups between ID and ND children is that a large number of children with ID in this study had a best-corrected VA with the range between 6/9.5 to 6/19 (LogMAR: 0.2 to 0.5), which is rarely observed in children of ND.

Table 2.3 Adapted categories of visual abnormality and visual impairment from WHO's proposal

Category	**Presenting binocular distance visual acuity(LogMAR)**	
	Worse than	Equal to or better than
Normal vision	N/A	0.2
Visual abnormality	0.2	0.5
Visual impairment	0.5	1.3

Adapted from World Health Organization (2003)

In order to explore whether a deficiency in stereopsis could impact HQoL and VQoL in children, the participants were also grouped according to their stereoacuity. Reduced stereoacuity was defined as poorer than 100 seconds of arc (Richardson, Wright, Hrisos, Buck, & Clarke, 2005). The visual function for each subgroup will be presented in Chapter 4.

2.2.8 Demographic and clinical information

The demographic data collected included: gender, age, IQ (in children with ID), and postcode. This information was obtained from parents and teachers, where available.

Ocular health information, obtained by vision screening, included: distance and near VA, ocular motility, refractive error, visual alignment, stereopsis, colour vision and ocular health. This information was derived from the vision screening results described in Section 2.3.

2.2.9 Questionnaire administration

The children completed the AUQUEI and LVP-FVQ in their classroom with instructions from a teacher, or in a clinic with the research investigator, in strict accordance with the instructions provided within the questionnaire. If the questionnaire was conducted in the school, the CVFQ was taken home by participants and returned to the teacher the next day following completion by a parent or caregiver. For those who completed the questionnaire in hospital, the CVFQ was returned to the investigator right after completion. To maximise test validity and reliability, the questionnaires were administered in a consistent manner. Requirements for individual guidance were likely to vary across the children within the ID group, due in part to the different types and severity of ID. With this in mind, groups of five to six participants were allocated to a teacher who was familiar with the academic performance and behaviour of members of the group. The teachers read each question to the participants, and helped with the meaning of each question if necessary. All teachers involved went through a group orientation organised by the investigator prior to administering the questionnaire. The purpose of this orientation was to clarify the questionnaire instructions and to emphasise the importance of not guiding participants toward a certain response. While the AUQUEI and LVP-FVQ questionnaires were being completed, the investigator observed the procedure, as a check of teacher and participant compliance. Most of the participants completed the questionnaire in the class, which lasted 30 minutes. Children who were unable to complete the questionnaire due to apparent difficulties with

comprehension of the questions (most of these children had autistic spectrum disorders), were excluded from the study. The method of questionnaire administration was identical to children with ND.

2.2.10 Questionnaire translation

Following initial development, the set of modified instruments was applied in a population of children with ID in China. The instructions and question items in the instruments were translated by a native Mandarin speaker. The translator was instructed to emphasise a conceptual rather than literal translation, so that translated questions conveyed equivalent meanings. The translated questions were reverse-translated by a native English speaker. The original and reverse-translated versions were compared, and if a translated item showed inconsistency, an alternative translation was proposed. The final translated version was presented to the advisory panel in China. Further modifications, such as question rewording, and modifications to topics and terms in the questions were made in response to feedback from the advisory panel in China. These modifications stemmed from issues of cultural difference or socio-economic differences between Australia and China. The interpretation and acceptability of the questionnaires were confirmed by monolingual participants.

2.3 Vision screening

Vision screening, as commonly used in an optometric clinic, was conducted to establish the visual status of the participants. Table 2.4 outlines the tests conducted with the corresponding objective measures. In view of the communication and attention difficulties in children with ID, the tests were chosen to allow efficient completion with minimal cooperation from the participants (Laukkanen, Valente-Caywood, & Nett, 2006; Maino, 1995). In addition, the tests selected were to identify common visual abnormalities, such as refractive errors, strabismus, binocular vision dysfunction and ocular pathology.

Table 2.4 Clinical tests applied in vision screening

Clinical measures	Test selected
Distance visual acuity	Lea Symbol Chart (Precision Vision, Villa Park, IL)
Near visual acuity	Patti Pics™ Logarithmic Visual Acuity Chart (Precision Vision, La Salle, IL)
Refractive status	Static retinoscopy with lens racks
Binocular vision and ocular motility	Hirschberg test, cover test, Krimsky test and versions
Stereopsis	Randot preschool test (Stereo Optical Co, Inc, Chicago, IL)
Colour vision	Waggoner HRR Pseudoisochromatic Plates (Home Vision Care, Gulf Breeze, FL)
Ocular health	Direct ophthalmoscopy and pen torch with 20D lens

Although the selection of clinical measures and tests were adapted from optometric assessment techniques for people with ID, they were also used to assess visual status in the control group of children without ID for consistency in the results. The following sections provide a summary of these tests.

2.3.1 Distance visual acuity

Visual acuity (VA) is commonly used by eye care practitioners as a measure of visual function. In the present study, the Lea Symbols Visual Acuity Test was applied by the presentation of pictures. The Lea Symbols chart presents optotypes with 6/3 to 6/60 (LogMAR: -0.3 to 1.0) in 0.1 log unit steps and the results were recorded as a Snellen equivalent. Before the test, the participants were asked to identify the demonstration symbol at near. With this check, the investigator confirmed the participants' level of comprehension and the reliability of the response. The test was administered from a viewing distance of three meters and proceeded from larger symbols to smaller ones. Participants were encouraged to name, sign, or match identical symbols presented on a hand-held matching card. At least 75% (e.g. three out of four or four out of five if there are five symbols in a line) of responses needed to be correct before proceeding to the next line, otherwise the VA was recorded as the current line. The correctness and speed of the responses were observed. A slow, hesitant response suggested uncertainty on the part of the child and another response was sought. The participants were encouraged to attempt to identify the letters. Testing was conducted monocularly, followed by binocularly. A patch or an occluder was used if tolerated by the participant, otherwise,

binocular VA was recorded. To ensure participants did not peek around the occluder, the participants were monitored and corrected where necessary during the testing (Maino, 1995).

2.3.2 Near visual acuity

The Patti Pics Logarithmic Visual Acuity Chart was used for testing near VA. The chart was held by the participant and measured at 40 cm using a measured length of cord. The procedure used was similar to the one employed for the Lea Symbols Visual Acuity Test. However, at least three out of five symbols, as opposed to three out of four, were to be identified correctly by the participant, otherwise near VA was recorded as the current line.

As explained above, monocular and binocular visual acuities were measured. For children with optical correction, both corrected VA and uncorrected VA were measured. Similar to measuring distance visual acuity, if monocular VA could not be measured, binocular VA only was recorded.

2.3.3 Static retinoscopy

Static retinoscopy was performed to identify the refractive error of the participants. In an attempt to control fixation and accommodation, a cartoon movie was viewed on a television from a viewing distance of 6 meters. The examiner frequently asked the participant what was happening in the movie and, thus, maintained his/her attention. In examining children with ID, a research assistant holding toys sat beside the television to attract the attention of the participant, whenever the movie failed to attract attention. A +1.50 dioptre lens was used at a working distance set at 67 cm. A lens rack was applied to improve the speed of refraction, an important factor when working with subjects of limited attention span such as children with ID. Refractive error was recorded from the results obtained from the lens rack.

2.3.4 Ocular motility

Eye movement was evaluated by the ocular motility test. The participants were instructed to follow a fixation target (a pen torch or a finger puppet) at a 50 cm working distance. To eliminate head movement, a hand was placed on the participant's head when necessary. The target was traced from the top right visual field position and moved in nine positions of gaze directed by a double H. Any asymmetric gaze, lack of smooth movement or inaccurate tracing indicated problems of movement and was recorded.

2.3.5 Binocular alignment

The participant was asked to look at a fixation target, at a viewing distance of 40 cm in primary gaze. Any asymmetric deviation of the corneal reflex was recorded, as this usually indicates strabismus or ocular misalignment. In general, every 1 mm decentration accounts for 15 prism dioptres (Rosenbloom & Morgan, 1990).

A cover test for in distance viewing was conducted to screen for eye alignment. In consideration of time efficiency, no near cover test was conducted. In most cases, an assistant was required to hold the fixation target and thereby attract the child's attention. A prism bar was incorporated with the alternating cover test to measure the deviation, if any, indicated by the unilateral cover test. As with the VA test, speed and efficiency of the eye examination and clearly communicated instructions are important in subjects with limited attention and compliance.

2.3.6 Colour vision test

Waggoner HRR Pseudoisochromatic Plates, under standard classroom/clinic lighting, were used as a quick screening test for colour vision. This test uses a circle, star, and/or square as the test pattern, and the participant was asked to name the shape or match the shape to a demonstration card. After a child's understanding of the test was confirmed, the full colour vision screening test was performed. A preliminary diagnosis of any colour vision disorder was based on the manufacturer's instruction (Waggoner HRR

Pseudoisochromatic Plates; Home Vision Care, Gulf Breeze, FL, USA)[3]

2.3.7 Stereoacuity

Stereoacuity was assessed by the Randot Preschool Stereoacuity Test (Randot Preschool Stereoacuity Test; Stereo Optical Co. Inc, Chicago, IL, USA), which employs random-dot patterns to avoid monocular cues and has a range of disparities from 800 to 40 seconds of arc. The symbols were introduced to the participants before the test started to ensure the participant was able to name or match the symbol to a demonstration card. The participant wore polaroid filters and a 40cm viewing distance was used. The symbols were called out or matched with the demonstration card. If the participant was able to correctly identify at least two of three shapes at a certain disparity level, a smaller disparity was introduced. Stereoacuity was recorded as the smallest disparity at which the child was able to correctly identify at least two of three shapes (Kulp & Mitchell, 2005).

2.3.8 Ocular health

Ocular health was examined using a direct ophthalmoscope for media and fundus viewing and as a light source with a +20 D lens for magnification for the assessment of the anterior segment, included assessment of the lids and lashes, cornea, conjunctiva, anterior chamber, pupil reflex, and iris. The posterior segment test included assessment of lens, vitreous and retina and was examined by the direct ophthalmoscope without the additional lens.

The American Association for Paediatric Ophthalmology and Strabismus (AAPOS) Vision Screening Committee has formulated a set of risk factors that need to be assessed by vision screening. These criteria are outlined in Table 2.5 (Donahue, 2004; Donahue, Arnold, & Ruben, 2003; Donahue, Johnson, & Merin, 2001). During the screening, participants found to have uncorrected visual abnormalities according to these criteria were referred for further assessment as appropriate.

[3] *Colour Vision Testing Made Easy (CVTME) was used instead with identical manner when Waggoner HRR test was not available in vision screening.*

Table 2.5 Definitions of visual abnormalities

Definition
Visual acuity: *> 2 line interocular difference or better eye VA worse than 6/9(LogMAR:0.18)*
Anisometropia: *>1.00 D (cylindrical or spherical)*
Manifest strabismus*: any*
Hyperopia:*> +3.50 D in any meridian*
Myopia: *> -3.00 D in any meridian*
Media opacity*: any*
Astigmatism : *> 1.5 D at 90 or 180; > 1.00 D at oblique axis*
Motility: *restriction in any position of gaze*

Adapted from Donahue (2004)

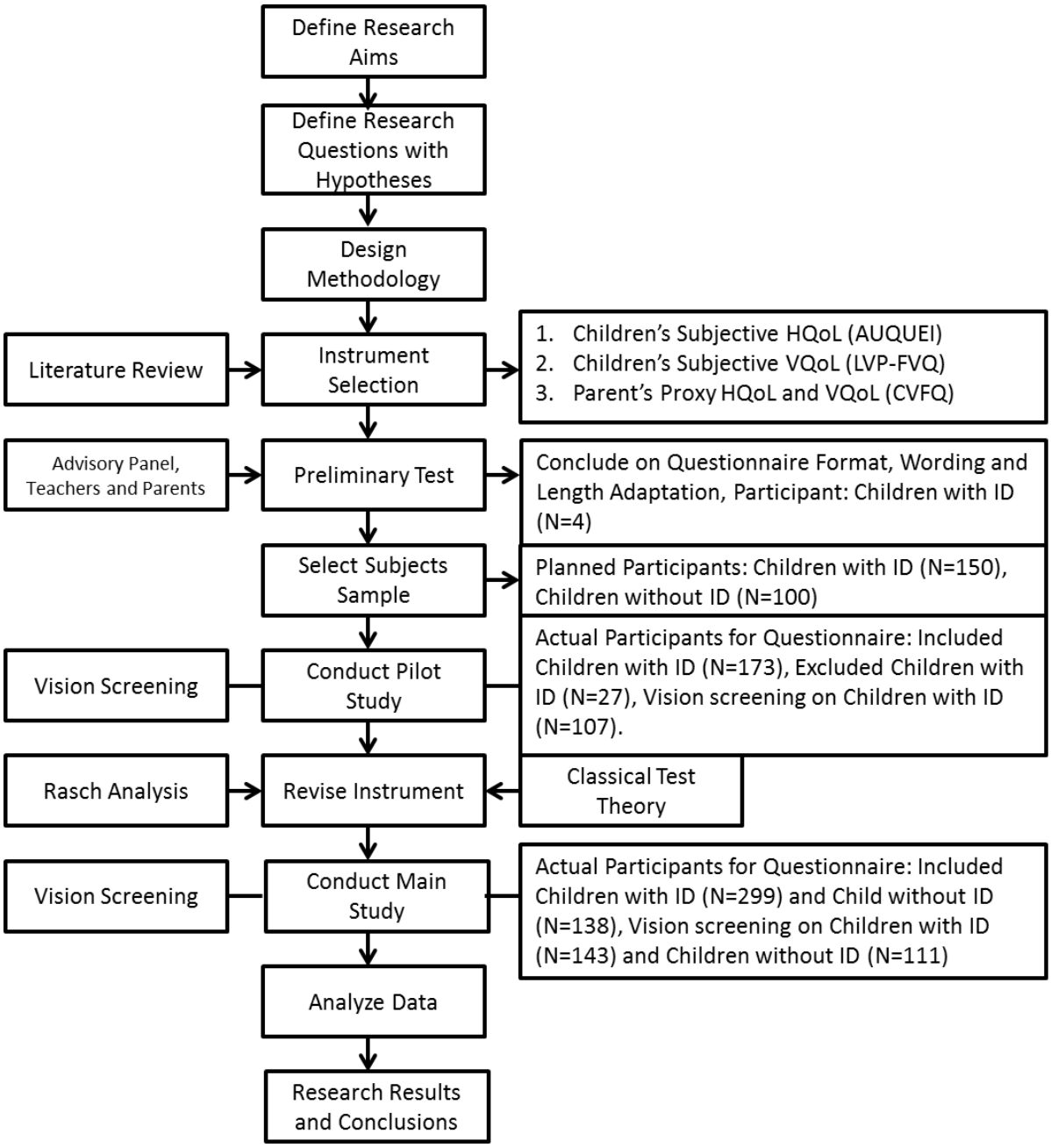
2.4 Pilot study – Instrument development and validation

A pilot study was conducted to develop a valid and reliable instrument for assessment of HQoL and VQoL in children with ID. The specific objectives and process are as follows.

2.4.1 Instrument development process

Figure 2.2 summarises the process of instrument development in this study. After an in-depth literature review, the selected instruments were modified with the aim of gathering subjective and proxy perspectives on HQoL and VQoL in children with ID. Content areas were established by the definitions of the domains included in each instrument. Findings from the literature review and consultation with the advisory panels helped to generate the appropriate items for the instruments, which may or may not have been present in the original instruments. In addition, the instrument layout, format and wording were modified, which helped to obtain good content validity of the instruments, ensuring comprehension of items.

Figure 2.2 Flowchart for the method in the pilot study



2.4.2 Performance of the instruments

The approach described here was adapted from the literature on instrument development and helped to explore the appropriateness of the instruments and the extent to which they were successful when applied to the children with ID. Most of the instruments described in the literature are based on (i) classical test theory (including factor analysis), which is the conventional method to evaluate the psychometric properties of instruments, or more recently on (ii) Rasch analysis, which is deemed a more scientific way of validating a questionnaire and interpreting its results. Both methods were employed in the present study.

The purpose of using classical test theory is to test and refine the domains and items within each instrument, from their original application in children with ND to application in children with ID. Based on the findings of classical test theory, Rasch analysis further tests the validity and reliability of items, and in the present research allows conversion of the ordinal scores into interval scores for application in the main study. In addition to verifying the domains derived from classical test theory, Rasch analysis also confirms the targeting of the instrument items to the capability of participants.

2.4.3 Validation via classical test theory

2.4.3.1 Validity

Content validity refers to the extent to which a group of items reflect a content dimension or domain(R. F. DeVellis, 2003). The items and domains included in the AUQUEI, LVP-FVQ and CVFQ were evaluated by the advisory panel (refer to section 3.1.4.1). The responses and suggested changes to each item were collected in a comments sheet from the panel member and were incorporated into the instrument set to be pretested. This preliminary instrument was introduced to the parents and teachers of children with ID in order to ascertain the appropriateness of the items. Successful completion of the instrument in a small group of children with ID was observed by the investigator, as an indicator of appropriateness of the instrument. Items with content irrelevant to children with ID were excluded.

Construct validity evaluates the underlying structure of the HQoL and VQoL of each instrument. It is expected that included domains reflect the relevant variables (in this case, HQoL and VQoL) (R. F. DeVellis, 2003). Factor analysis was conducted to condense the large set of the items into meaningful domains (factors) and used to confirm the conceptual domains and make it possible to look for associations between these domains. In the present study, principal component analysis (PCA) was used for the factor analysis.

Divergent/convergent validity In each instrument, the Pearson's (Spearsman's for non-parametric) correlation coefficient for continuous variables was used to look for associations between domains. Before being able to claim convergent validity in domains with close connection, the assumptions of closeness (*convergent validity*) needed to be tested and verified by significant statistical findings with strong correlations. For domains not closely connected (*divergent validity*), the opposite applies and weak correlations were expected.

2.4.3.2 Reliability via Classical Analysis

Instrument or scale reliability refers to the ratio of error variance to the true score of a latent variable (R. F. DeVellis, 2003). As a fundamental measure of reliability, *internal consistency* is an indicator of how well the items measure the underlying trait. Internal consistency reliability was measured according to the correlation coefficient between items within domains and Cronbach's α of each domain extracted from the factor analysis. Although Cronbach's α is somewhat dependent on the number of items, a level of 0.7 is considered acceptable (de Boer et al., 2005; R. F. DeVellis, 2003).

2.4.4 Validation via Rasch analysis

Construct validity of the instrument is specified by the data fit and the form of the relationship between respondents and the items. The process of analysis used here began with the evaluation of the compliance of rating categories to a continuum of lower to higher difficulty/ability. Thus, a respondent selecting a lower category response would be rated lower in the respective latent trait, which represents attitudes and

satisfactions towards subjective HQoL and VQoL in this section.

The frequency distributions of categories were examined to identify those that were redundant. The removal of redundant and misfit items can improve the internal consistency and targeting of the instruments and results in greater precision than conventional Likert scoring in discriminating between participants (Smith & Smith, 2004).

Rasch analysis was applied to further validate the instruments. Data normality and the fit of the model were optimised, and the validity of the construct was enhanced by category collapse and item reduction of the initial instruments.

2.4.4.1 Validity

Fit statistics (see Section 1.5.3), including infit and outfit, help to identify which items contribute most to the measurement of a latent trait. For example, if infit and outfit mean squares have an expected value of 1.00, then an infit mean of less than 0.8 represents items that are too predictable (the items have at least 20% less variation than expected). These over fitting items may be redundant or lack enough variance to contribute new information to the measure. Mean outfit values greater than 1.20 represent misfit (at least 20% more variance than was expected) and suggest that the item measures something different from the overall scale. Acceptable values for item inclusion may be between 0.80 and 1.20 for a strict definition (often used for infit) or between 0.70 and 1.30 or higher for a more lenient definition. For maximum effectiveness, the criteria described in the literature should be used to guide item removal and to incorporate various statistical approaches. The suggested infit and outfit ranges for item elimination should be considered as a guide and can depend largely on sample size. In the present study, infit/outfit mean squares outside 0.50 to 1.50 were applied as an indication of potential problematic items and inside 0.70 to 1.30 were used to confirm an appropriate item. For items with borderline infit/outfit values (e.g. 0.6; 1.4), additional criteria such as normality and the reliability index are used to determine appropriateness (Pesudovs, et al., 2007).

2.4.4.2 Reliability via Rasch Analysis

Reliability assessment by Rasch analysis is equivalent to internal consistency in classical test theory. In Rasch analysis, the reliability indices are based on model error variance for each estimate of a person's ability and item difficulty. Therefore, a person's ability and item difficulty are aligned on a common linear scale. The person and item reliability index indicates the reproducibility of the order of the person or items compared with a parallel test on the same construct, and whether persons and items are represented adequately on a continuum (Wright & Masters, 1982). It is worth noting that the person separation index provides an indication of whether the instrument is sensitive enough to differentiate the hierarchy of the ability/attitude of an underlying trait in participants.

Rasch analysis can also indicate the effect of removing an item on overall scale performance. If removal of an item considerably decreases person separation, that item should be retained. Person separation is an indicator of the ability (precision) of the instrument to differentiate between the qualities of life of different people. Person separation is expressed as the ratio of the adjusted standard deviation to the root mean square error. A person separation value of 2.0 or more is indicative that the scores of scale for the participants in this study are significantly different across the measurement distribution (Pesudovs, et al., 2007).

2.5 Main study - Exploration of impact of visual abnormality on HQoL and VQoL in children with ID

This study aimed to investigate the impact of visual abnormality on HQoL and VQoL in children with ID. The instruments involved were validated in the pilot study and applied in the cohort groups in the main study. Rasch calibrated scores were used to measure the performance of the participants in subscales of the instruments that were validated by Rasch analysis. Likert scale scores as validated by classical test theory were used in subscales of instruments that did not meet the Rasch criteria. Thus, a subtotal score was calculated in each subscale across the instruments in the final set, which represents

subjective or proxy perceptions of HQoL and VQoL in children. The domains, as indicated by the score of subscales, perceived by participants with ID and ND were compared.

The associations between objective visual measures (as indicated by binocular VA and stereopsis), and subjective measures of HQoL and VQoL were explored. Correlation of VA and subscale scores, and of stereoacuity and subscale scores were calculated. In addition, scores of subscales for children with and without visual abnormality were compared. The difference in HQoL and VQoL in children with and without ID was also explored.

The association between parents' proxy views and children's own perception of HQoL and VQoL was examined by comparing similar domains between instruments. The method for each research question is described below and the study is described in detail in Chapter 4.

2.5.1 Children's perception of HQoL and VQoL

After the pilot study, an instrument with valid items was developed for use in the main study, in which sum scores from individual domains were calculated with interval scales by Rasch analysis or with Likert scales by classical test theory, where appropriate. The impact of visual abnormality in HQoL and VQoL was assessed by comparing the scores of subscales of the adapted AUQUEI and LVP-FVQ between children with normal vision and with visual abnormality (Table 2.6).

2.5.2 Parents' perception of HQoL and VQoL

Parents' perceptions of HQoL and VQoL were measured from the sum scores of subscales from the adapted CVFQ. Table 2.7 summarises the research questions and data analysis methods. Scores of the subscales were also compared to the scores of HQoL and VQoL from children's perception.

Table 2.6 Evaluation of subjective HQoL and VQoL in children [a]

Research question	Statistical test	Independent variable	Dependent variable
Are subjective perceptions of HQoL different between children with and without ID?	Independent sample t-test	Intellectual capability: ND and ID	Subscale scores on the adapted AUQUEI
Are subjective perceptions of HQoL different between children with mild ID and with moderate ID?	Independent sample t-test	Intellectual capability: Moderate ID and Mild ID	Subscale scores on the adapted AUQUEI
Are subjective perceptions of HQoL different between intellectual disabled children with normal vision, visual abnormality and visual impairment?	ANOVA with Bonferroni-adjusted post hoc test	Visual status: Normal vision, Visual abnormality and Visual impairment	Subscale scores on the adapted AUQUEI
Are subjective perceptions of VQoL different between children with and without ID?	Independent sample t-test	Intellectual capability: ND and ID	Subscale scores on the adapted LVP-FVQ
Are subjective perceptions of VQoL different between children with mild ID and with moderate ID?	Independent sample t-test	Intellectual capability: Moderate ID and Mild ID	Subscale scores on the adapted LVP-FVQ
Are subjective perceptions of VQoL different between intellectual disabled children with normal vision, visual abnormality and visual impairment?	ANOVA with Bonferroni-adjusted post hoc test	Visual status: Normal vision Visual abnormality and Visual impairment	Subscale scores on the adapted LVP-FVQ
Is visual status different between children with mild ID and with moderate ID?	Independent sample t-test	Intellectual capability: Moderate ID/ Mild ID	Corrected binocular LogMAR VA and Stereopsis
Is visual status different between children with and without ID?	Independent sample t-test	Intellectual capability: Moderate ID/ Mild ID	Corrected binocular LogMAR VA and Stereopsis
Is there an association between HQoL and visual acuity?	Pearson's correlation	Subscale score of the adapted AUQUEI and LogMAR VA	N/A

Table 2.6 Evaluation of subjective HQoL and VQoL in children (continued)

Research question	Statistical test	Independent variable	Dependent variable
Is there an association between HQoL and stereopsis?	Pearson's correlation	Subscale score of the adapted AUQUEI and stereoacuity	N/A
Is there an association between VQoL and visual acuity?	Pearson's correlation	Subscale score of the adapted LVP-FVQ and LogMAR VA	N/A
Is there an association between VQoL and stereopsis?	Pearson's correlation	Subscale score of the adapted LVP-FVQ and stereoacuity	N/A
Is there an association between HQoL and developmental age?	Pearson's correlation	Subscale score of the adapted AUQUEI and age (yr)	N/A
Is there an association between HQoL and IQ?	Pearson's correlation	Subscale score of the adapted AUQUEI and IQ scores	N/A
Is there an association between VQoL and developmental age?	Pearson's correlation	Subscale score of the adapted LVP-FVQ and age (yr)	N/A
Is there an association between VQoL and IQ?	Pearson's correlation	Subscale score of the adapted LVP-FVQ and IQ scores	N/A
Is there a difference in subjective perceptions of HQoL according to gender and age ?	Two-way between groups ANOVA	Age groups, Sex	Total score of the adapted AUQUEI
Is there a difference in subjective perceptions of VQoL according to gender and age?	Two-way between groups ANOVA	Age groups, Sex	Total score of the adapted LVP-FVQ

[a]*Children's HQoL measured using Rasch-scaled scores and VQoL measured using Likert-scales scores were compared in the groups with diverse intellectual capability and visual status. The relationship between HQoL and VQoL, visual status, age and IQ are evaluated by Pearson's correlation, followed by multivariate analysis with relevant variables. Further, comparisons of the subscales of the adapted AUQUEI and LVP-FVQ between subgroups were performed.*

Table 2.7 Evaluation of parents' perception of HQoL and VQoL[a]

Research question	Statistical test	Independent variable	Dependent variable
Is there an association between parents' perception of VQoL and children's objective visual status?	Pearson's correlation	Scores of subscales in CVFQ, Corrected binocular LogMAR VA and stereoacuity	N/A
Is there an association between the perceptions of parents and children on HQoL and VQoL?	Pearson's correlation	Scores of subscale in each relevant domain across adapted AUQUEI, LVP-FVQ and CVFQ	N/A
Is there a difference on proxy perceptions of HQoL and VQoL according to gender and age?	Two-way between groups ANOVA	Age groups, Sex	Scores of subscale in CVFQ
Is there a difference in CVFQ scores for children with different ID severity, LogMAR VA, stereopsis and age?	ANOVA with Bonferroni-adjusted post hoc test	IQ scores, Corrected binocular LogMAR, stereopsis and age	Score of subscales in CVFQ

[a]*The association between HQoL and VQoL domains and visual status (LogMAR and stereopsis) was assessed by correlation analysis. The relationship between the parents' assessment of their children's HQoL and VQoL and the children's assessment of their own HQoL and VQoL were evaluated by correlations of the scores of the relevant AUQUEI and CVFQ domains. A comprehensive evaluation of parents' assessment of HQoL and VQoL was conducted by comparing the CVFQ subscale scores across the cohort groups (ANOVA with Bonferroni-adjusted post hoc test).*

CHAPTER 3 PILOT STUDY: DEVELOPMENT AND VALIDATION OF INSTRUMENTS FOR USE IN CHILDREN WITH ID

3.1 Instrument modification and preliminary validation

In Chapter 1, the use of instruments with items that widely cover the concepts of children's HQoL and VQoL was reviewed. The use of proxy responses is commonly applied in the assessment of HQoL in people with ID(McVilly, Burton-Smith, & Davison, 2000). However, evidence exists that children with ID can communicate their own perceptions of HQoL(Dekker, et al., 2002; Ramirez & Kratochwill, 1997). Thus, given the importance of subjective perceptions of a person's own life (White-Koning, et al., 2005), proxy responses should ideally be used as a complement to children's subjective views on their HQoL or VQoL.

3.1.1 Aims

1 To develop a process for the administration of a questionnaire for use in children with ID.

2 To test the validity of instruments by identifying the HQoL and VQoL domains that are applicable to children with ID.

3 To improve the validity and reliability of instruments by modifying the items in domains of HQoL and VQoL that are applicable to children with ID.

3.1.2 Hypotheses

1 HQoL and VQoL of children with ID are measureable using instruments that are specifically designed and formatted for use in children with ID.

2 HQoL and VQoL domains of children with ID are distinct from those of children with ND.

3.1.3 Methods

3.1.3.1 Preliminary study on instrument development

A prototype instrument for the assessment of HQoL and VQoL in children with ID was constructed after a review of the literature relating to existing instruments (Bader, 1974;

Felius, et al., 2004; Gothwal, et al., 2003; Gupta et al., 2005; Manificat, et al., 1997; Stevenson et al., 2005). An advisory panel was formed to assist in the development of the instrument as described in Chapter 2. An item bank was built in order to investigate (i) children's perspectives of HQoL and VQoL; and (ii) parent's perspectives of their children's HQoL and VQoL. The items were selected from the three questionnaires as described in section 2.2.3. Items for children's subjective HQoL were derived from the AUQUEI**;** items for children's subjective VQoL were derived from the LVP-FVQ; and items for parents' perspectives of their children's HQoL and VQoL were derived from the CVFQ. These instruments have been successfully used and validated in children with ND. The prototype items that were reviewed by the advisory panel are presented in Table 3.1.

The prototype items were sent to the advisory panel in Australia. After feedback from the panel, the preliminary instruments were tested in a small group of children with ID at the optometry clinic at UNSW (n=4, age 8 to 14) and modifications were made according to the panel's feedback and results of the preliminary test.

Table 3.1 Prototype items selected from the AUQUEI, LVP-FVQ and CVFQ instruments

AUQUEI
1. How do you feel when you're having dinner with your family?
2. How do you feel when you go to bed at night?
3. If you have brothers and sisters, how do you feel when you play with them?
4. How do you feel when you're asleep at night?
5. How do you feel when you are at school?
6. How do you feel when you look at a picture of yourself?
7. How do you feel when you go to the doctor's?
8. How do you feel when you think about your father?
9. How do you feel on your birthday?
10. How do you feel when you think about your mother?
11. How do you feel when you stay in hospital?
12. How do you feel when you play alone?
13. How do you feel when your parents are talking about you?
14. How do you feel when you spend the night away from home?
15. How do you feel when people ask you how to show what you're able to do?
16. How do you feel when your friends are talking about you?
17. How do you feel when you take a medicine?
18. How do you feel during the holidays?
19. How do you feel when you make a drawing?
20. How do you feel when you see yourself as a grown-up?
21. How do you feel when you are with your grand-parents?
22. How do you feel when you watch television?
23. How do you feel when you move (walk, run, and jump)?
24. How do you feel when you are eating?
25. Some days you are well, some days you are sick, how do you feel when you think about your health?
26. How do you feel when people tell you what to do?
LVP-FVQ
1. Do you have any difficulty in seeing whether somebody is calling you by waving his or her hand from across the road?
2. Do you have difficulty in walking alone in the corridor at school without bumping into objects or people?
3. Do you have any difficulty in walking home at night (from tuition or a friend's house) without assistance when there are streetlights?
4. Do you have any difficulty in copying from the blackboard while sitting on the first bench in your class?
5. Do you have difficulty in reading the bus numbers?
6. Do you have any difficulty in reading the other details on the bus (such as its destination?)
7. Do you have any difficulty in reading your textbooks at an arm's length?
8. Do you have any difficulty in writing along a straight line?
9. Do you have any difficulty in finding the next line while reading when you take a break and then resume reading?
10. Do you have any difficulty in locating dropped objects (pen, pencil, eraser) within the classroom?
11. Do you have any difficulty in threading a needle?
12. How much difficulty do you have in distinguishing between 1 dollar and 2 dollar coins (without touching)?
13. Do you have difficulty in climbing up or down stairs?
14. Do you have difficulty in lacing your shoes?
15. Do have difficulty in locating a ball while playing in the daylight?
16. Do you have difficulty in applying paste on your toothbrush?
17. Do you have difficulty in locating food on your plate while eating?
18. Do you difficulty in identifying colours (e.g., while colouring)?

Table 3.1 Prototype items selected from the AUQUEI, LVP-FVQ and CVFQ instruments (continued)

CVFQ
1. In general, would you say that your child's overall health is:
2. At the present time, would you say that your child's eyesight is:
3.If your child has an eyesight problem for only one eye, would you say that your child's eyesight in the affected eye is:
4.How much of the time do you worry about your child's eyesight?
5.How much time do you need to spend on treatment for your child's vision problem (eye doctor appointments, patching, eye drops, and therapy)?
6.Does the time you spend on your child's vision problem (eye doctor appointments, patching, eye drops, and therapy) take away from time you would like to spend with your other children or husband/wife?
7. Do you and other family members (your spouse or parents) argue about the medical care your child is getting or about treatment that the doctor has prescribed?
8.I am afraid that my child will never have good vision.
9. I am bothered by other people's comments about my child's vision or eyes when I take him/her to a store or mall.
10. My child likes to try new things.
11. Taking my child to the eye doctor is stressful.
12. I think that my child's vision will improve.
13. My child feels different from other children.
14. My child is happy most of the time.
15. I notice other children looking at my child.
16. My child likes to visit with relatives.
17. My child is teased because of his/her vision problems.
18. My child cries a lot.
19. I worry that my child may not be able to read, watch TV, or drive a car.
20. My child makes new friends easily.
21. My child is affectionate.
22. My child gets along well with our other children and friends.
23. My child gets angry or frustrated because of his vision problem.
24. We stay at home a lot because of my child's vision problem.
25. My child can feed himself/herself.
26. My child plays with toys.
27. My child can recognise faces (friends, relatives) across a room.
28. My child can imitate others (make a face, stick tongue out, play peek-a-boo).
29. My child can dress himself/herself.
30. My child can brush his/her teeth.
31. My child can wash his/her face.
32. My child adjusts to changes in lighting (going out into bright sunlight or entering a dark room or theater.)
33. My child can ride a bicycle.
34. My child can play a sport or active game (for example, tag).
35. My child will track a mobile or a moving toy.
36. My child can locate a small piece of food (a raisin or Cheerio) and grasp it.
37. My child can pour liquid into a cup or glass.
38. My child can dial a telephone.
39. My child helps with chores.
40. My child can tell what time it is.
41. My child can identify coins.
42. My child enjoys looking at books.
43. My child is interested in playing with our pet(s).
44. My child has a regular sleep routine.
45. My child's eyesight makes it difficult for him/her to learn to walk, run, skip, or jump.

Table 3.1 Prototype items selected from the AUQUEI, LVP-FVQ and CVFQ instruments (continued)

CVFQ
46. My child's vision gets in the way of his/her learning.
47. My child's eyesight has made it difficult for him/her to learn to read.
48. My child enjoys watching television, videos, or playing video games.
49. My child likes to travel on family vacations.
50. My child enjoys playing with others (sisters and brothers or friends).
51. My child enjoys drawing, painting or other art activities.
52. My child's eyesight makes it difficult for him/her to find something on a crowded shelf or in a closet.
53. My child makes eye contact with me and smiles.
54. My child bumps into people, walls, or furniture.
55. My child trips over curbs or steps.
56. My child bumps into other people.
57. I have trouble applying treatment (for example, putting on an eye patch or glasses, giving eye drops or other medication).
58. My child is uncomfortable when treated (for example, while wearing a patch or glasses or when you put in eye drops).
59. My child is less active when treated (for example, when wearing a patch or glasses, or when taking eye drops or medication).
60. I worry when my child refuses treatment (for example, pulls off the patch or glasses, or squeezes eye shut when trying to put in eye drops).

3.1.3.2 Pilot questionnaire development

The instrument set (as enclosed in Appendix 2) was applied in children with ID at the Lujiazhui School for Special Education, Shanghai, China. Of the 200 children invited to participate, 173 completed at least one of the questionnaires, and of these, data on age, gender and IQ were available in 111. Participants were classified by severity of ID according to the IQ (see Section 2.2.5.1). Mild ID accounted for 31.5% of participants and moderate ID accounted for 68.5% of participants. The participants were of chronological age 8 to 18 (mean=14.3 years, standard deviation=3.0), and 57% of the respondents were male. The questionnaire administration has been described previously (see Section 2.2.9).

3.1.3.3 Vision screening

An ophthalmic examination was carried out on each of the 107 participants whose parents gave consent for this. The examination consisted of the screening procedure as described in Chapter 2 (see Section 2.3).

3.1.3.4 Data analysis

3.1.3.4.1 Factor analysis (FA)

Statistical analysis was performed using a commercially available software package (SPSS, Chicago, IL, U.S.A.). Several FA methods were used to extract the underlying dimensions of AUQUEI and LVP-FVQ (Hair, Black, Babin, Anderson, & Tatham, 2006; Pallant, 2007). The following sections outline the procedures that were performed with FA.

3.1.3.4.2 Suitability of the data for FA

The sample size and the strength of the internal relationship among the variables were examined. A sample of at least 150 children, with at least five responses for each item, was considered to be sufficient for the FA as long as the variable loading was significant (Stevens, 1996). Additionally, an inspection of the correlation matrix was conducted to ascertain the strength of relationships between the items. A preliminary examination of item correlation (coefficients greater than 0.3) implies appropriateness of FA as a method for analysis (Tabachnick & Fidell, 2007). Two statistical tests were used to indicate the suitability of the data for FA, i.e., The Bartlett test of sphericity was significant and a Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy greater than 0.7 (Hair, et al., 2006).

3.1.3.4.3 Factor extraction

Factor extraction by principal components analysis (PCA) determined the number of factors in the underlying construct and their relationships. Three techniques, described below, were applied to assist with extraction of factors within each construct (i.e., the dimensions of HQoL measured by the AUQUEI, and VQoL measured by the LVP-FVQ).

3.1.3.4.4 Kaiser's criterion

The Eigenvalue describes the proportion of the variance explained by the factor. Only factors with loading values greater than 1.0 were taken into account for further analysis and investigation.

3.1.3.4.5 Catell's scree test

A scree plot represents Eigenvalue against cumulative variance and indicates which factors are responsible for the majority of the variance in the data. SPSS provides a scree test from the plot of the Eigenvalue of each factor, irrespective of whether it exceeds 1.0. This test detects a point where the plot curve of the Eigenvalue changes direction or becomes flatter. The factors above the point are retained, as those factors account for most of the variance of the data (Catell & Horn, 1963).

3.1.3.4.6 Horn's parallel analysis

Horn's parallel analysis refers to the comparison between the Eigenvalues retained from the scree test, and the value from random data of similar sample size to the study. The Eigenvalues of the retained factors from the study sample should be larger than those generated from random data, thereby ensuring factors are selected using a stringent criterion (Horn, 1965). The computer program used to perform parallel analysis was the Monte Carlo PCA for Parallel Analysis (Watkins, 2000).

3.1.3.4.7 Factor rotation and interpretation

Varimax orthogonal rotation helped with the interpretation of factor patterns. As an initial interpretation, an item with a loading value greater than 0.45 was considered; confirmation of a factor was based on the content of the items. Those items that did not reflect the constructs according to the HQoL or VQoL domain were considered for removal (Hair, et al., 2006).

3.1.3.4.8 Reliability

The factors derived from the FA were treated as a scale for certain domains of the HQoL and VQoL construct. Cronbach's α coefficient was used to indicate internal consistency of the scale. In most cases, the Cronbach's coefficient should be greater than 0.7. However, in short scales with fewer than ten items, the criterion can be lowered to 0.6. In this case, inter-item correlation was reported, as an alternative indicator of internal consistency, with an optimal range from 0.2 to 0.4. Therefore, in this study, a Cronbach's α between 0.60-0.80 was accepted as an indication of homogeneity without

redundancy in the pool of questions (O'Connor, 2004). A significant increase in Cronbach's α after deletion of an item indicated that the item did not belong to the domain it appeared in and, therefore, should be removed.

FA was not conducted on the CVFQ, since the domains were fixed using different scales (e.g., quality, frequency, agreement and difficulty). The internal consistency reliability was calculated to confirm the domains of the construct as adapted from the published literature.

3.1.3.4.9 Inter and intra domain associations

As an indication of construct validity, the associations between domains within and between the adapted AUQUEI and LVP-FVQ (derived after FA), were evaluated using non-parametric analysis (Spearman's rho). The size of the correlation coefficient (r) indicated the strength of the relationship. Cohen (1988) recommends that the strength of the correlation coefficient can be described as low (r=0.1-0.29), medium (r=0.3-0.49), or high (r=0.50-1.0). A two-tailed level of statistical significance was set at $P<0.01$.

3.1.4 Results

3.1.4.1 Preliminary study and questionnaire adaptation

The suggested modifications from the advisory panels to the items selected from the AUQUEI and LVP-FVQ included: a reduction in the page content; changes to the scale category, where appropriate; and adding illustrations and symbols to help convey the response options. A summary from the advisory panel and the findings from the preliminary study in the small group of children with ID and their parents are presented in Table 3.2. The cartoon design of the scale options of AUQUEI and LVP-FVQ in both original forms, and their modified forms, are illustrated in Figures 3.1 to 3.6. In the AUQUEI, a 4-point Likert scale format was retained (Figure 3.3). In the LVP-FVQ, children were required to tick one of two options, each indicated by cartoon faces representing "yes" or "no" (Figure 3.6). These faces were considered by the advisory panel to be more familiar and meaningful for children with ID, as these cartoons were commonly used in school for special education. Modifications to the CVFQ included

changing an item or its scales. Items related to severe visual loss or not applicable to children with ID were excluded from the CVFQ. The modifications to all three questionnaires are summarised in Table 3.3. The instrument used in the pilot study is shown in Appendix 2.

Table 3.2 Qualitative results derived from the preliminary test of the instruments

Advisory panel comments on instrument design	
1.	Too many questionnaires are included in one package, which one is the dominant piece? It is suggested to divide the questionnaire into sections for children and parents.
2.	Too many questions displayed in one page.
3.	Some sentences in the questionnaires are too complicated, which is not suitable for use in children with ID
4.	The four faces option in the AUQUEI look too similar (hard to differentiate) for children with ID
5.	A forced choice question in LVP-FVQ might be more appropriate for children with ID
6.	The picture option should be designed in consultation with a speech specialist, who is familiar with the board makers, symbolic words for special education, that are used by children with ID
7.	A class questionnaire administration in a special school needs to be arranged, rather than in a clinic, as the school environment would be more familiar for children with ID. Thus, making it easier for them to concentrate.
8.	Items included in the questionnaires need modification before they can be applied to children with ID, since the original version is validated for normative children.
Insights derived from the preliminary test of children with ID (n=4) and their parents at an optometry clinic, UNSW	
1.	Three out of the four parents were not aware of any impact of vision on their children's life.
2.	Two of the children could not wear spectacles. The most common reason is that the child does not like to wear spectacles and feels frustrated when wearing spectacles.
3.	Three children liked the shining, colourful pictures.
4.	Two children, who had mild ID could perform visual tracking, letter recognition, number counting, simple geometric drawing and basic arithmetic.
5.	As discussed earlier (sections 1.1.3 and 2.2.7) chronological age is not a reliable method of judging activity performance. Observation was consistent with this, with one child, who was older than the other three participants, failed to complete the questionnaire. This child provided thoughtless answers and was not responsive to the investigator's instruction.
6.	Watching TV was the most common entertainment for children with ID
7.	Most feedback on questionnaire design was derived from the parents, since direct opinions on the questionnaire from children were difficult to obtain. While this observation is in only four children and their parents, it suggests a need to validate questionnaires for children's subjective perceptions of HQoL and VQoL.

Figure 3.1 Response options used in the original AUQUEI

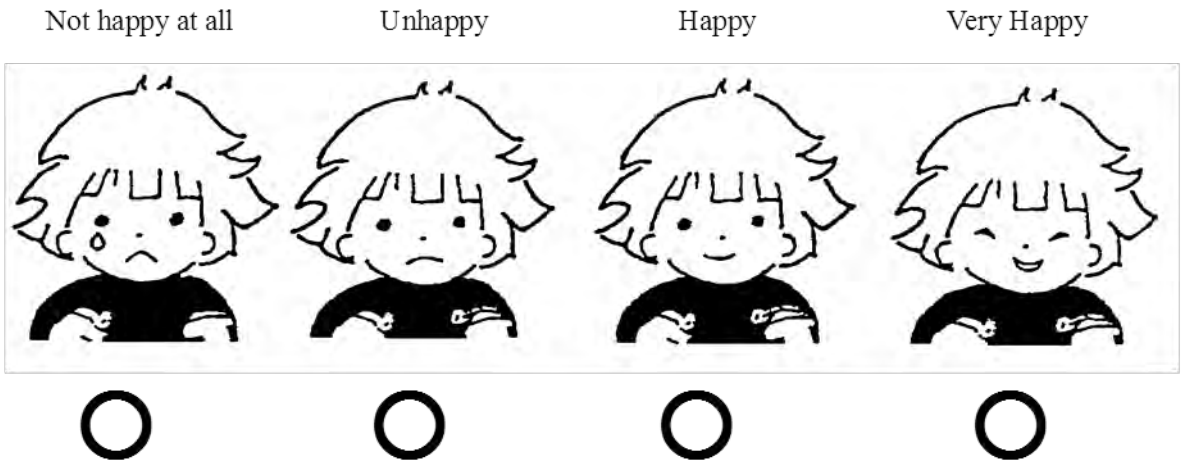


Figure 3.2 Response options used in the revised AUQUEI that were used in the preliminary study

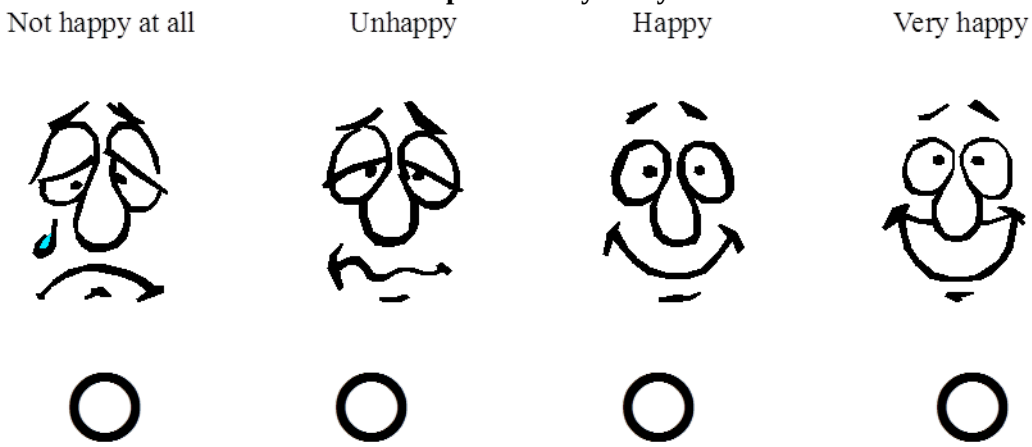


Figure 3.3 Response options used in the AUQUEI after preliminary study and applied in the pilot study

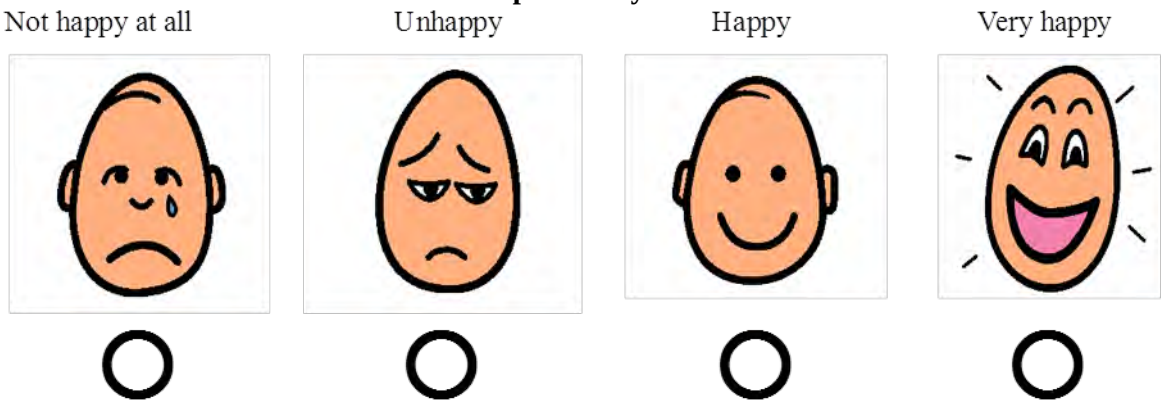


Figure 3.4 Response options used in the LVP-FVQ in the preliminary test

Figure 3.5 Response options suggested by a paediatrician from the Australian advisory panel

Figure 3.6 Response options used in the LVP-FVQ that were suggested by a speech therapist and applied in the pilot study

Both the AUQUEI and LVP-FVQ were designed for self-completion; children were instructed to select one option by circling the answer or pointing to the picture. The domains and items from the prototype instruments that were modified after the preliminary test are listed below (Table 3.4-3.6). These items were used in the pilot study.

Table 3.3 Modifications made to the draft questionnaires after the preliminary test

	AUQUEI		LVP-FVQ		CVFQ	
	Initial	***Final***	***Initial***	***Final***	***Initial***	***Final***
Format	Five questions per page	Two questions per page	Five questions per page	Three questions per page	Mixed format	Tabulated format
Domain	Four	Four	Four	Three	Six	Six
Scale	Four point Likert scale	Four point Likert scale	Five point Likert scale	Dichotomous scale	Five to six point Likert scale	Five point Likert scale
Length	28 items	23 items	23 items	22 items	61 items	45 items
Design	Cartoon face expression	Cartoon face expressions, with clear difference	Black and white Smiley/sad face options.	Colour. Smiley/sad face options.	Text	Text

[a]*These modifications were made based on the feedback from the advisory group and observation of a small sample of children with ID in preliminary study.*

Table 3.4 Items (n=23) allocated to four domains in the adapted AUQUEI

Family life	1. When I am having dinner with my family, I feel 2. When I go to bed at night, I feel 3. My brothers and sisters make me feel 12. When I am with my grandparents, I feel 13. When I watch television, I feel 15. When I am eating, I feel
Health care	5. When I go to the doctor's, I feel 7. When I stay in hospital, I feel 9. When I take medicine, I feel 16. When I am sick, I feel 17. When I am not sick, I feel
Social life	6. On my birthday, I feel 8. When I play alone, I feel 10. When it is holiday, I feel 18. When people tell me what to do, I feel
School Life	4. At school, I feel 11. When I make a drawing, I feel 14. When I move (walk, run, jump), I feel 19. When I am playing outside, I feel 20. When I play a sport, I feel 21. When I do my homework, I feel 22. When I read a book, I feel 23. When my work is marked at school, I feel

Table 3.5 Items (n=22) allocated to three domains in the adapted LVP-FVQ

Vision related activities	1. Can you climb up and down stairs? 2. Do you bump into things? 3. Can you kick a ball when you play? 4. Can you hit the ball when you play? 5. Can you find food on your plate when eating? 6. Can you find your favourite toy at play time? 7. Can you pick up a red pencil from a box of pencils? 8. Can you put toothpaste on your toothbrush by yourself? 9. Can you do (tie) up your shoelaces by yourself?
Vision quality	10. Can you see the TV clearly? 11. Can you see a person's face across the road? 12. Can you see bus numbers clearly? 13. Can you see the pictures in your books clearly?
Academic performance	14. Can you read a book by yourself? 15. Can you draw a straight line on paper without a ruler? 16. Can you copy from the board in class? 17. Can you write the numbers from 1 to 10? 18. Can you write your name? 19. Can you remember words easily? 20. Can you finish your homework on time? 21. Can you do math? 22. Is your handwriting neat?

Table 3.6 Items (n=45) allocated to six domains in the adapted CVFQ

General health	1. In general, would you say that your child's overall health is:
General vision	2. At the present time, would you say that your child's eyesight (using both eyes) is: 3. If your child has an eyesight problem for only one eye, would you say that your child's eyesight in the affected eye is:
Competence	4. My child can feed himself/herself. 5. My child can recognise faces (friends, relatives) across a room. 6. My child can dress himself/herself. 7. My child can brush his/her teeth. 8. My child can wash his/her face. 9. My child can ride a bicycle. 10. My child can play a sport or an active game (for example, football). 11. My child can locate a small piece of food (a raisin or Cheerio) and grasp it. 12. My child can pour liquid into a cup or glass. 13. My child can dial a telephone. 14. My child helps with chores. 15. My child can tell what time it is. 16. My child can identify coins. 17. My child's eyesight makes it difficult for him/her to find something on a crowded shelf or in a closet. 18. My child bumps into people, walls, or furniture. 19. My child trips over curbs or steps.
Personality	20. My child is happy most of the time. 21. My child likes to visit with relatives. 22. My child makes friends easily. 23. My child is affectionate. 24. My child gets along well with our other children and friends. 25. My child enjoys looking at books. 26. My child's eyesight makes it difficult for him/her to learn to walk, run, skip, or jump.. 27. My child's vision gets in the way of his/her learning. 28. My child enjoys watching television, videos, or playing video games. 29. My child likes to travel on family vacations. 30. My child enjoys playing with others (sisters and brothers or friends). 31. My child enjoys drawing, painting or other art activities.

Table 3.6 Items (n=45) allocated to six domains in the adapted CVFQ (continued)

Family impact	32. How much of the time do you worry about yourchild's eyesight? 33.How much time do you need to spend on treatment for your child's vision problem (eye doctor appointment, patching, eye drops and therapy)? 34.Does the time you spend on your child's vision problem (eye doctor appointments, patching, eye drops, therapy) take away from time you would like to spend with your other children or husband/wife? 35.Do you and other family members (your spouse or parents) argue about the medical care your child is getting or about treatment that the doctor has prescribed? 36.I am bothered by other people's comments about my child's vision or eyes when I take him/her to a store or mall. 37. My child feels different from other children. 38. I notice other children looking at my child. 39. My child is teased because of his/her vision problem. 40. I worry that my child may not be able to read, watch TV, or drive a car.
Treatment	41.I have trouble applying eye/vision treatment (for example, putting on an eye patch or glasses, giving eye drops or other medication). 42.My child is uncomfortable when treated (for example, while wearing a patch or glasses or when you put in eye drops). 43.My child is less active when having treatment (for example, when wearing a patch or glasses, or when taking eye drops or medication). 44.I worry when my child refuses treatment (for example, pulls off the patch or glasses, or squeezes eye shut when trying to put in eye drops). 45.I forget to give my child treatment.

3.1.4.2 Factor analysis and domain identification

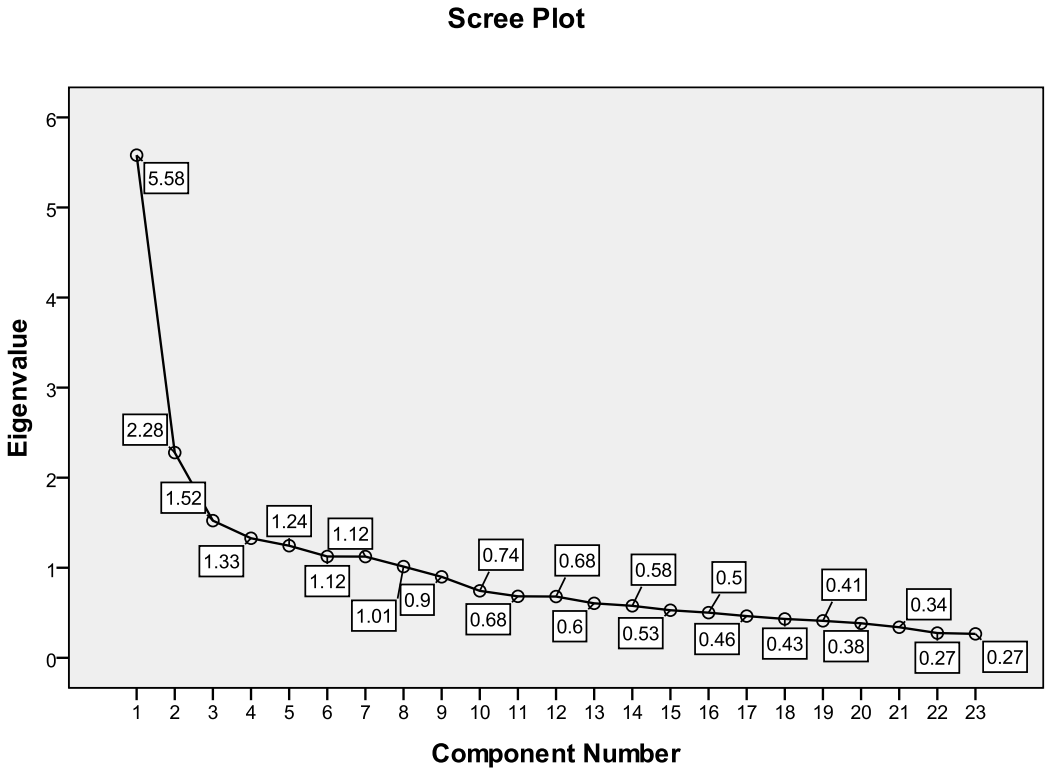
A questionnaire set derived from the preliminary test was sent out for the pilot study in children with ID in China. Of the 200 participants who participated, 173 returned the questionnaires; 168, 166 and 149 participants completed the AUQUEI, LVP-FVQ and CVFQ, respectively.

3.1.4.2.1 AUQUEI

The 23 items of the AUQUEI were subjected to principal component analysis (PCA). Prior to performing PCA, the suitability of the data for factor analysis was assessed. Inspection of the correlation matrix revealed the presence of many coefficients of 0.3 or greater. The Kaiser-Meyer-Oklin (KMO) value was 0.8, exceeding the recommended value of 0.6. The Barlett's Test of Sphericity reached statistical significance ($P<0.01$), supporting appropriateness of factor analysis**.**

PCA revealed the presence of eight components with Eigenvalues exceeding 1 (Figure 3.7), which explained cumulatively 66.2% of the variance (Table 3.9). Using Catell's scree test, an inspection of the scree plot revealed three components as confirmed factors (Figure 3.7). Thus, these components were retained for further investigation. This finding was further supported by the results of the parallel analysis (Table 3.7), which showed only three components with Eigenvalues exceeding the corresponding criterion values for a randomly generated data matrix of the same size (23 variables × 168 participants).

Figure 3.7 Scree plots showing the factor structures for the adapted AUQUEI



The number at each point is the Eigenvalue of each item (component number). The scree plots yielded 8 components with an Eigenvalue >1. However, the cut-off point is the fourth, where the plots plateau, after this point the increase in variance is relatively small.

Table 3.7 Comparison of the Eigenvalues derived from the principal component analysis (PCA) and the corresponding criterion values obtained from the parallel analysis in AUQUEI

Component number	Actual Eigenvalue from PCA	Criterion value from parallel analysis	Decision
1	5.58	1.73	accept
2	2.28	1.61	accept
3	1.52	1.52	accept
4	1.33	1.43	reject

To aid in the interpretation of these three components, Varimax rotation was performed. The rotated solution revealed the presence of a simple structure, with the three components showing a number of strong loadings and all variables loading substantially on one component (Table 3.8). The three components solution explained a total of 40.8% of the variance (Table 3.9).

Table 3.8 Component matrix showing loadings on items after each factor was extracted, using principal component analysis [a]

	Component		
	1	***2***	***3***
When I move (walk, run, jump)	.67	-.02	-.31
When I make a drawing	.65	.08	-.08
When I am playing outside	.62	-.30	.22
When my work is marked at school	.59	.04	-.30
At school	.59	-.09	.03
When I do my homework	.57	.20	-.24
When I read a book	.57	.24	-.36
My brothers and sisters make me	.53	-.14	.04
When I play a sport	.53	.01	-.40
When I am having dinner with my family	.50	-.11	.35
When people tell me what to do	.49	.19	-.38
When it is holiday	.48	-.43	.13
When I am eating	.47	-.22	.30
When I am with my grandparents	.46	-.28	-.03
When I play alone	.44	.15	.00
When I am not sick	.43	-.24	.00
When I watch television	.38	-.33	.26
When I go to doctor's	.37	.60	.27
When I stay in hospital	.40	.58	.35
When I am sick	.22	.57	.17
When I take medicine	.40	.49	.30
On my birthday	.43	-.49	.16
When I go to bed at night	.18	-.00	.40

a. 3 components extracted.

Table 3.9 Results of the principal component analysis showing initial Eigenvalues of the components (factors) extracted and cumulative percentage variance of the three components derived (indicated with asterisk)

Component	Initial Eigenvalues		
	Total	**% of Variance**	**Cumulative %**
1	5.58	24.27	24.27
2	2.28	9.91	34.17
3	1.52	6.62	40.79*
4	1.33	5.77	46.56
5	1.24	5.41	51.97
6	1.13	4.89	56.86
7	1.12	4.89	61.75
8	1.01	4.40	66.15
9	.90	3.91	70.06
10	.74	3.23	73.29
11	.68	2.97	76.26
12	.68	2.96	79.22
13	.61	2.63	81.85
14	.58	2.51	84.36
15	.53	2.30	86.65
16	.50	2.18	88.83
17	.46	2.01	90.85
18	.43	1.88	92.72
19	.41	1.78	94.51
20	.38	1.67	96.18
21	.34	1.48	97.65
22	.28	1.20	98.85
23	.27	1.16	100.00

Table 3.10 Domains and items confirmed from the adapted AUQUEI

School Life	4. At school, I feel 11. When I make a drawing, I feel 14. When I move (walk, run, jump), I feel 18. When people tell me what to do, I feel 19. When I am playing outside, I feel 20. When I play a sport, I feel 21. When I do my homework, I feel 22. When I read a book, I feel 23. When my work is marked at school, I feel 8. When I play alone*
Health care	5. When I go to the doctor's, I feel 7. When I stay in hospital, I feel 9. When I take medicine, I feel 16. When I am sick, I feel
Family life	1. When I am having dinner with my family, I feel 3. My brothers and sisters make me feel 6. On my birthday, I feel 10. When it is holiday, I feel 12. When I am with my grandparents, I feel 13. When I watch television, I feel 15. When I am eating, I feel 17. When I am not sick, I feel 2. When I go to bed at night, I feel?*

In the AUQUEI, two items (with asterisks in Table 3.10) were identified for further investigation, due to low loadings on all factors and/or because the Cronbach's α coefficient increased significantly if the item was deleted. Further analysis of item reduction will be discussed in Section 3.2.4.2.3.

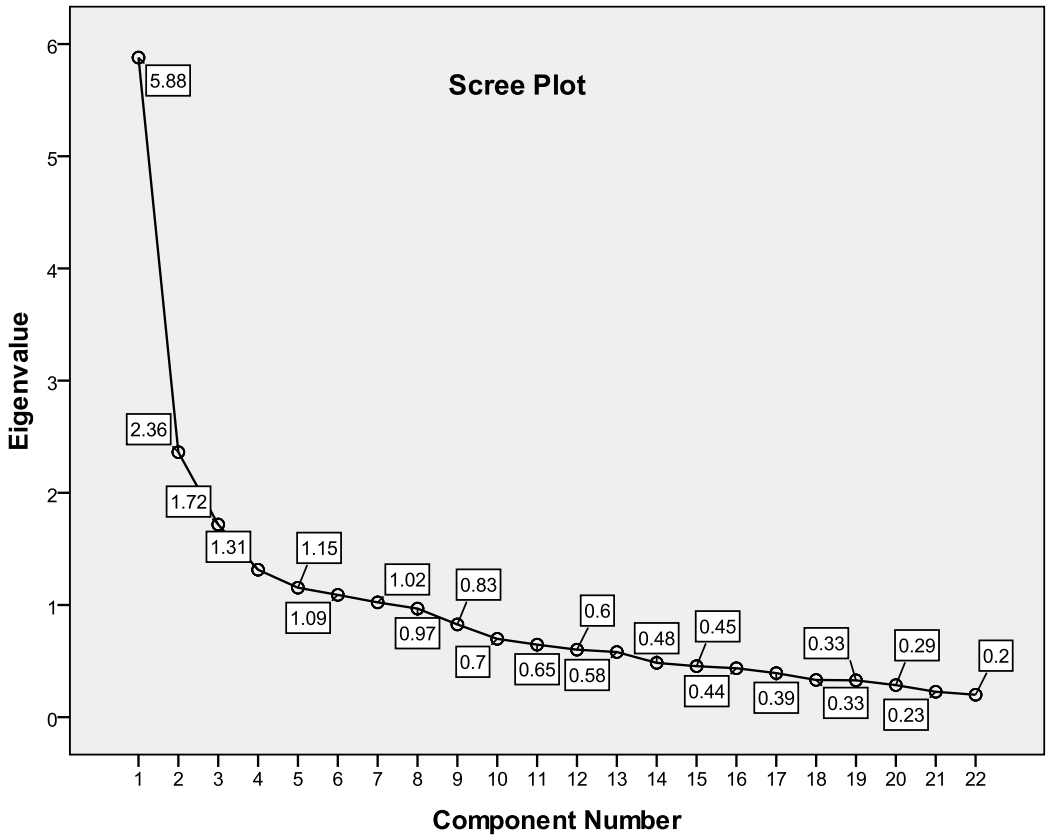
3.1.4.2.2 LVP-FVQ

Similarly, 22 items from the LVP-FVQ were subjected to PCA. The KMO measure was 0.8 and the Barlett's Test of Sphericity reached statistical significance ($P<0.01$), supporting the appropriateness of factor analysis.

The rotated solution revealed the presence of a simple structure, with the three components showing a number of strong loadings and all variables loading substantially on one component (Table 3.11). An inspection of the scree plot revealed an inflection after the fourth component (Figure 3.8). Using Catell's scree test, it was decided to retain the first three components for further investigation (Table 3.11). The results from the Parallel Analysis also showed that there were only three factors with Eigenvalues

that exceeded the corresponding criterion values for a randomly generated data matrix of the same size (22 variables × 168 participants, Table 3.12). PCA revealed the presence of seven components (factors) with Eigenvalues exceeding 1, which explained cumulatively 66.1% of the variance (Table 3.13).

Figure 3.8 Scree plots showing factor structures for the adapted LVP-FVQ



The number at each point is the Eigenvalue of each item (component number). The scree plots yielded 7 factors with an Eigenvalue >1. However, the cut-off point was the fourth item (component 4), where the plots plateau; after this point, the variance is relatively small.

Table 3.11 Component matrix showing loadings on items after each factor was subtracted using principal component analysis

	Component		
	1	***2***	***3***
Can you copy from the board in class	.74	-.27	.05
Can you write your name	.69	-.27	.14
Can you do maths	.63	-.37	-.03
Can you write the number from 1 to 10	.63	-.13	-.20
Can you read a book by yourself	.63	-.20	-.14
Can you tie up your shoelaces by yourself	.58	-.21	.29
Can you remember words easily	.55	-.39	-.01
Is your handwriting neat	.54	-.20	-.30
Can you see bus numbers clearly	.53	.05	.23
Can you put toothpaste on your toothbrush by yourself	.52	.02	.17
Can you finish your homework on time	.49	-.14	-.27
Can you see the picture in your books clearly	.49	.47	-.23
Can you draw a straight line on paper without a ruler	.46	-.31	-.05
Can you see the TV clearly	.44	.39	.39
Can you hit a ball when you play	.42	.18	.31
Can you see a person's face across the road	.38	.25	.35
Can you kick a ball when you play	.35	.07	-.26
Can you climb up and down stairs	.43	.69	.05
Can you find your favourite toy at play time	.38	.54	-.48
Can you find food on your plate when eating	.37	.53	.23
Can you pick up a red pencil from a box of pencils	.37	.35	-.53
Do you bump into things	.16	.09	.48

Table 3.12 Comparison of the Eigenvalues from the principal component analysis and the corresponding criterion values obtained from the parallel analysis in LVP-FVQ

Component number	**Actual Eigenvalue from PCA**	**Criterion value from parallel analysis**	**Decision**
1	5.88	1.72	accept
2	2.36	1.59	accept
3	1.72	1.49	accept
4	1.31	1.41	reject

Varimax rotation was performed on the three factors, which explained a total of 45.3% of the variance (Table 3.13). In the LVP-FVQ, two items (with asterisks in Table 3.14) were identified for further investigation based on the criterion used in AUQUEI.

Table 3.13 Results of the principal component analysis showing initial Eigenvalues of the component concluded (factors) and cumulative percentage on variance of three components derived (indicated with asterisk)

Component	Initial Eigenvalues		
	Total	% of Variance	Cumulative %
1	5.88	26.73	26.73
2	2.36	10.74	37.46
3	1.72	7.81	45.27*
4	1.31	5.97	51.24
5	1.15	5.25	56.49
6	1.09	4.95	61.44
7	1.02	4.65	66.09
8	.97	4.40	70.49
9	.83	3.76	74.25
10	.70	3.17	77.42
11	.65	2.94	80.36
12	.60	2.74	83.09
13	.58	2.64	85.73
14	.48	2.20	87.93
15	.45	2.07	90.00
16	.44	1.98	91.98
17	.39	1.79	93.77
18	.33	1.51	95.27
19	.33	1.49	96.77
20	.29	1.30	98.06
21	.23	1.03	99.09
22	.20	.91	100.00

Table 3.14 Domains confirmed from the LVP-FVQ with principal component analysis and internal consistency by Cronbach's α coefficient

School activities	After-school activities	Item identification
Can you copy from the board in class?	Can you tie up your shoelaces by yourself?	Can you find your favourite toy at play time?
Can you do maths?	Can you find food on your plate when eating?	
Can you write your name?	Can you put toothpaste on your toothbrush by yourself?	Can you pick up a red pencil from a box of pencils?
Can you remember words easily?	Can you see the TV clearly?	
Can you read a book by yourself?	Can you climb up and down stairs?	Can you see the picture in your books clearly?
Can you write the number from 1 to 10?	Can you see a person's face across the road?	
Is your handwriting neat?	Can you hit a ball when you play?	Can you kick a ball when you play?*
Can you draw a straight line on paper without a ruler?	Can you see bus numbers clearly?	
Can you finish your homework on time?	Do you bump into things?*	

3.1.4.2.3 Domains in AUQUEI, LVP-FVQ and CVFQ

Thus, the following three domains were defined for the AUQUEI: School life, Family life and Health care. Using the same procedure, the following three domains were determined from the LVP-FVQ: School activities, After-school activities and Item identification. The previously defined domains of CVFQ were: General health, General vision, Competence, Personality, Family impact, and Treatment. For items in the domain of "Treatment", the percentage of missing data accounted for more than 50%, thus this domain was excluded. The internal consistency reliability (Cronbach's α) of the domains for each of the questionnaires is presented in Tables 3.15 to 3.17. Most Cronbach's α values ranged from 0.70 to 0.85, which indicate good internal consistency reliability.

Table 3.15 Domains and internal consistency reliability of domains derived from the AUQUEI

Domain	Items	No. of items	Cronbach's α
School life	4,11,14,18,20-23	8	0.81
Family life	1,3,6,10,12,13,15,17,19	9	0.76
Health care	5,7,9,16	4	0.73

Table 3.16 Domains and internal consistency reliability of domains derived from the LVP-FVQ

Domain	Items	No. of items	Cronbach's α
School activities	14-22	9	0.84
After-school activities	1,4,5,8-12	8	0.69
Item identification	6,7,13	3	0.69

Table 3.17 Domains and internal consistency reliability of domains derived from the CVFQ

Domain	Items	No. of items	Cronbach's α
General Health	1	1	N/A
General Vision	2, 3	2	0.70
Competence	4-19	16	0.82
Personality	20-31	12	0.74
Family Impact	32-40	9	0.73

Measures of associations between named domains indicated that, first, some domains were highly or moderately correlated within the questionnaire. These included: the adapted AUQUEI domains "family life" and "school life" ($r=0.54$, $p<0.01$); the adapted LVP-FVQ domains vision-related "school activities" and vision-related "After-school activities" ($r=0.49$, $p<0.01$); and the adapted CVFQ, domains "general health" and "general vision" ($r=0.32$, $P<0.01$), "general health" and "family impact" ($r=0.34$, $P<0.01$), and "general vision" and "family impact" ($r=0.45$, $P<0.01$). Second, the domains of vision-related "school activities" in the LVP-FVQ and "school life" in the AUQUEI were found to be moderately correlated ($r=0.27$, $p<0.01$).

3.1.4.3 Vision screening

Of the 107 participants who underwent vision screening in the pilot study, 35 (33%) had some form of visual abnormality, as defined in Table 2.3. Of these, 11 achieved acuity

of at least 20/30 with refractive correction, indicating that the visual abnormality was uncorrected refractive error in these cases. The remaining 24 had corrected acuity poorer than 20/30 in at least one eye, with the following abnormalities: 6 with constant strabismus (4 unilateral; 2 alternating), 9 with anisometropia, 3 with both strabismus and anisometropia, and 6 with suspected pathology. All of the 35 participants with visual abnormality were referred for further assessment.

3.1.5 Discussion

Based on feedback from the advisory panels and preliminary testing in a small group of children with ID, three questionnaires, previously validated for use in non-disabled children, were modified for use in children with ID. The revised questionnaires were administered to a Chinese population (See Appendix IV: Sample AUQUEI, LVP-FVQ and CVFQ in Chinese), where questionnaires were completed within small groups of children in a familiar environment, under the guidance of familiar personnel. Under such circumstances, the questionnaires were well-comprehended and appeared to be appropriate and useful. Moreover, the responses to the domains of the questionnaires were reliable as evidenced by good internal consistency.

The results of the pilot study suggest that children with ID have a sense of both their vision-related performance (VQoL) and HQoL. Highly correlated domains within questionnaires reflected good content validity. Note that high correlation between domains may suggest an overall measure of QoL rather than subscale measures. However, factor analysis clearly identified separate subscales relating to different facets of QoL. The associations between domains in the VQoL instrument (LVP-FVQ) and the HQoL instrument (AUQUEI) indicate a link between visual function and HQoL in children with ID. This suggests that the instrument developed here may be used to explore the extent to which children with ID suffer reduction in their HQoL as a result of visual abnormalities. However, it is possible that the LVP-FVQ, having been developed for a low vision population, may not include items that are suitable for a population with less severe visual abnormalities. This point is further discussed in section 3.2.5.

It is worth noting that some items included in the original and adapted LVP-FVQ which refer to ability to see and to navigate may be influenced by physical and intellectual capability. For example, a response to the question "Can you copy from the board in class?" will depend upon visual functions such as acuity (to see detail on the board) but also on other factors such as physical coordination and cognition. However, the items are designed to test vision-*related* factors, and while vision is at the core of these, these are influenced by a range of functions and abilities.

The number of domains identified in the present study differs from those derived from the original questionnaires, since some items and the questionnaires were originally validated for use in children with ND. While the number of domains differed, there was similarity between the original and modified questionnaires in terms of the nature of the domains (See Table 3.1, Table 3.4 and Table 3.5). The modified LVP-FVQ and AUQUEI questionnaires included one (School activities) and two domains (School life and Family life), respectively, that were identical to those established previously in children with ND. This consistency between domains across different populations suggests that some aspects in HQoL and VQoL are similar for children with and without ID.

There was good internal consistency for each domain in the CVFQ, after one domain (domain of treatment) was excluded due to a low response rate. This low response rate indicated the inapplicability of this domain for children with ID. Although the domains derived from the application of the CVFQ in children with ND (Felius et al, 2004) were the same as those derived from children with ID in this study, the items within each domain differed across children with and without ID. This suggests diverse perspectives of the parents and carers across these groups of children. This finding is explored further in Chapter 4, taking into account demographic and vision status information. In addition, the associations that were found between the domains of "general health", "general vision" and "family impact" imply that health and vision-related items impact not only children, but also significantly affect the children's families.

The results of vision screening are in agreement with previous reports of a high incidence of visual abnormality in children with ID. As noted in Chapter 1, visual abnormality is a factor that can often be corrected, at least during childhood. These findings reinforce the importance of regular eye examinations and treatment as appropriate for children with ID.

3.2 Further validation and refinement of instrument using Rasch analysis

In Section 3.1, the testing of domains in each instrument for use in children with ID was described and the internal consistency was found to be robust after conventional validation approaches, based on classical test theory. However, the validity of instruments that use the Likert scale, where the score is calculated from the sum of each item and uniform changes across categories are assumed, has been questioned (Wright & Masters, 1982). For example, in a Likert-scaled HQoL questionnaire, such as the AUQUEI as applied in this study, "very happy" coded as "4" in data analysis, is used to represent twice the level of happiness as "happy" coded as 2 for each of the items. Thus, a scale of this kind may not correctly reflect category steps. As addressed in Chapter 1, Rasch analysis offers a means by which this potential confound is minimised by converting ordinal values to interval measures allowing item difficulty to be appraised accordingly(Garamendi, Pesudovs, Stevens, & Elliott, 2006; Massof, 2002). In addition, the targeting of the instrument to the population of interest (i.e., whether the items included in the instruments match the capacity of participants) is depicted on a common scale shared by item and participants.

3.2.1 Aim

To improve the psychometric properties (validity and reliability) of the adapted LVP-FVQ, AUQUEI and CVFQ by Rasch analysis.

3.2.2 Hypothesis

The psychometric properties of the adapted instruments will be improved after Rasch analysis.

3.2.3 Methods

3.2.3.1 Questionnaire selection and adaptation

The instruments used for validation are described in Section 3.1.

3.2.3.2 Participants

The participants were the same as described in Section 3.1.

3.2.3.3 Data analysis

General methodology relating to Rasch analysis is described in Chapter 2 (see Section 2.4.4). Analysis methods specific to this pilot study are outlined below.

Appropriateness was assessed by determining the degree of missing data and the ceiling/floor effect. Missing data were defined as any item to which more than 10% of respondents did not respond. The ceiling/floor effect was defined as more than 90% (on the dichotomous LVP-FVQ scale) or 50% (on the four-option AUQUEI scale and five-option CVFQ scale) of respondents selecting one response category at the high or low end of the scale (Langelaan et al., 2007). After examination of skew, kurtosis and normality, items not compliant with normality were assessed in terms of the item context and its match or mis-match with adjacent items on the scale.

Rasch analysis was performed using Winsteps 3.35 (Linacre, 2008) with the joint maximum likelihood estimation using the Partial Credit Model (Linacre, 2002; Pesudovs, Garamendi, & Elliott, 2006; Pesudovs, et al., 2003). Ordinal data were transformed to an interval scale, using a unit known as the logit (see section 2.2.4). Persons with poor fit statistics (e.g., providing contradictory responses) were identified, response patterns were examined, and the data relating to that person were excluded from analysis if appropriate (e.g., the person clearly misunderstood a question). Poorly fitting items were also identified as overfit (mean square value of item < 1.0 Rasch index), indicating low variation in response pattern, probably due to redundancy; or as underfit (mean square value of item >1.3) suggesting the response pattern is unlikely to conform with the Rasch model. Moreover, the unidimensionality of each instrument was tested using fit statistics and Rasch-residual-based Principal Component Analysis (PCAR) to determine whether items were consistent with a single underlying theme.

Since multiple scale types were used in the original CVFQ, in the modified questionnaire, a system of uniform coding was applied post-hoc where appropriate, such that a lower score represented lower endorsement levels based on proxy HQoL or VQoL in all the items. For example, in the quality scale, “excellent” was recoded from 1 to 5

while “poor” was recoded from 5 to 1. In other subscales of frequency, difficulty and agreement, the rescoring applied to the individual item. Therefore, the new scores of the CVFQ are in accordance with the principle of the rating scale model of Rasch analysis, where a lower score represents lower manifestation of the underlying trait.

3.2.4 Results

3.2.4.1 Descriptive statistics

After initial analysis for appropriateness, four items from the AUQUEI and six items from the LVP-FVQ, which did not provide normally distributed data, were identified (items with asterisks in Skew and Kurtosis sections of Table 3.18 and Table 3.19). Thirteen items from the CVFQ that did not provide normal distribution or have a high percentage of missing data were identified (items with asterisks in Table 3.20).

Table 3.18 Assessment of AUQUEI item quality in children with ID

Item	Description	Skew	Kurtosis	Missing Data (%)	Ceiling Effect (%)	Infit Mean Square	Outfit Mean Square	Item Calibration (SE)
1	Having dinner with my family	-1.2	2.0	1.2	53.6 *	0.9	0.8	-0.7 (0.1)
2	Go to bed at night	-0.6	0.1	1.8	31.0	1.2	2.4	0.1 (0.1)
3	My brothers and sisters make me	-1.0	0.6	1.2	49.4	1.0	0.9	-0.5 (0.1)
4	At school	-1.1	1.5	1.8	50.6 *	0.8	0.8	-0.6 (0.1)
5	Go to doctor's	-0.7	-0.2	0.6	13.1	1.3	1.3	1.9 (0.1)
6	On my birthday	-2.3 *	5.9 *	1.2	77.4 *	1.0	1.0	-1.7 (0.1)
7	Stay in hospital	0.8	0.3	0.6	8.9	1.1	1.0	2.2 (0.1)
8	Play alone	0.0	-1.1	0.6	26.8	1.2	1.2	0.8 (0.1)
9	Take medicine	0.6	-0.3	1.2	10.7	1.2	1.2	2.0 (0.1)
10	When it is holiday	-1.5	1.3	1.8	70.2	1.0	1.2	-1.4 (0.1)
11	Make a drawing	-0.6	-0.3	1.2	41.1	0.6	0.6	-0.3(0.1)
12	With my grandparents	-1.7	4.0 *	1.2	66.7 *	1.0	0.9	-1.3 (0.1)
13	Watch television	-1.2	1.8	1.2	56.5 *	1.0	1.3	-0.8 (0.1)
14	Move (walk, run, jump)	-0.8	0.1	1.2	44.0	0.9	0.8	-0.2 (0.1)
15	Eating	-1.4	2.8 *	1.8	56.0 *	0.9	0.8	-0.9 (0.1)
16	Sick	1.1	1.7	2.4	4.8	1.3	1.3	2.8 (0.1)
17	Not sick	-1.4	2.5 *	1.8	52.4 *	1.0	1.2	-0.7 (0.1)
18	People tell me what to do	-0.7	- 0.2	2.4	39.3	1.0	1.0	0.0 (0.1)
19	Playing outside	-0.9	0.8	1.2	54.2 *	0.7	0.6	-0.8 (0.1)
20	Play a sport	-0.9	0.6	1.8	43.5	1.0	1.1	-0.2 (0.1)
21	Do my homework	-0.6	-0.4	1.2	35.7	1.1	1.0	0.2 (0.1)
22	Read a book	-0.4	-0.7	1.8	29.8	1.0	1.0	0.4(0.1)
23	My work is marked at school	-0.7	0.0	1.2	39.3	0.8	0.8	-0.1 (0.1)

Descriptive statistics of skew, kurtosis, percentage ceiling effect (percentage answers of "very happy") and Rasch analysis are shown. Fit statistics (infit and outfit) are indicated by mean square values and item calibration indicates the location of each item in the item-person map (the difficulty of the item). Asterisks indicate items not meeting criteria for skew, kurtosis, ceiling effect or Rasch fit and need further consideration. Each item was asked in the form "When …, I feel:"or "…, I feel:" as appropriate.

Table 3.19 Assessment of LVP-FVQ item quality in children with ID

Item	Description	Skew	Kurtosis	Missing Data (%)	Ceiling Effect (%)	Infit Mean Square	Outfit Mean Square	Item Calibration (SE)
1	Climb up and down stairs	-7.3 *	52.3 *	0.6	98.2 *	0.9	0.8	-3.1 (0.7)
2	Bump into things	-0.6	-1.7	0.6	64.1	1.5 *	1.9 *	1.5 (0.2)
3	Kick a ball when you play	-0.9	-1.1	1.2	71.1	1.3	1.3	1.1 (0.2)
4	Hit a ball when you play	-2.7	5.1	1.2	89.8	1.0	1.1	-0.7 (0.3)
5	Find food on your plate when eating	-4.6 *	19.2 *	1.8	95.8 *	1.0	1.4	-1.9 (0.4)
6	Find your favourite toy at play time	-4.6 *	18.9 *	3.0	95.7 *	1.0	0.9	-1.9 (0.4)
7	Pick up a red pencil from a box of pencils	-2.3	3.3	4.2	87.6	1.1	1.5	-0.4 (0.3)
8	Put toothpaste on your toothbrush by yourself	-2.2	3.1	2.4	87.2	1.0	0.8	-0.3 (0.3)
9	Tie up your shoelaces by yourself	-0.4	-1.8	0.6	60.5	0.9	0.9	1.8 (0.2)
10	See the TV clearly	-4.0 *	14.0 *	1.2	94.6 *	1.0	0.6	-1.6 (0.4)
11	See a person's face across the road	-1.8	1.2	0.6	83.2	1.2	1.2	0.1 (0.2)
12	See bus numbers clearly	-1.8	1.3	3.0	83.4	0.9	0.9	0.0 (0.2)
13	See the pictures in your books clearly	-3.7 *	11.7 *	3.6	93.8 *	1.0	0.7	-1.4 (0.4)
14	Read a book by yourself	-1.4	0.1	3.0	79.1	0.9	0.7	0.4 (0.2)
15	Draw a straight line on paper without a ruler	0.2	- 2.0	1.2	45.8	1.1	1.1	2.7 (0.2)
16	Copy from the board in class	-1.3	-0.3	1.2	77.1	0.7	0.5	0.6 (0.2)
17	Write the numbers from 1 to 10	-2.9 *	6.3 *	2.4	90.9 *	0.8	0.4	-0.9 (0.3)
18	Write your name	-1.8	1.2	1.8	83.0	0.7	0.7	0.1 (0.2)
19	Remember words easily	-0.4	-1.9	1.8	59.4	0.9	1.1	1.8 (0.2)
20	Finish your homework on time	-2.1	2.4	1.8	86.1	1.0	0.8	- 0.2 (0.3)
21	Do maths	-1.0	-1.0	3.0	72.4	0.8	0.9	1.0 (0.2)
22	Is your handwriting neat	-0.8	-1.5	3.0	67.5	1.0	0.9	1.3 (0.2)

Descriptive statistics of skew, kurtosis, percentage ceiling effect (percentage answers of "yes") and Rasch analysis are shown. Fit statistics (infit and outfit) are indicated with mean square values and item calibration indicates the location of each item in the item-person map (the difficulty of the item). Asterisks indicate items not meeting criteria for skew, kurtosis, ceiling effect or Rasch fit and need further consideration. Each item was asked in the form "Can you…?" (except for item 2 "Do you bump into thing?" and item 22 "Is your handwriting neat?").

Table 3.20 Assessment of CVFQ item quality for proxy responses

Item	Description	Skew	Kurtosis	Missing Data (%)	Ceiling Effect (%)	Infit Mean Square	Outfit Mean Square	Item Calibration (SE)
1	Overall health	0.3	1.0	1.4	11.7	0.5*	0.5*	0.3 (0.1)
2	Eyesight	0.3	0.1	0.7	6.9	0.8	0.8	0.9 (0.1)
3	Eyesight with the affected eye	-0.8	0.9	28.3	4.1	0.9	0.9	1.7 (0.1)
4	Feed himself/herself	3.9*	18.9*	0.7	89.0*	1.2	0.9	-2.4 (0.2)
5	Recognise faces across room	6.2*	41.8*	0.7	95.9*	1.3	1.3	-3.6 (0.4)*
6	Dress himself	2.9*	10.2*	0.0	80.0*	1.2	0.9	-1.7 (0.2)
7	Brush teeth	3.7*	16.0*	0.7	85.5*	1.4*	1.1	-2.0 (0.2)
8	Wash face	2.5*	5.6*	0.0	84.1*	1.0	0.7	-2.1 (0.2)
9	Ride a bike	0.7	-0.9	7.6	39.3	1.4*	1.4*	0.3 (0.1)
10	Play a sport	1.4	1.1	0.7	51.7*	1.4*	1.3	-0.3 (0.1)
11	Locate a small piece of food	2.9*	8.1*	2.1	85.5*	1.2	1.0	-2.3 (0.2)
12	Pour liquid into a cup	3.6*	15.6*	0.7	87.6*	1.3	1.0	-2.3 (0.2)
13	Dial a telephone	2.0	3.7*	2.1	71.0*	1.3	1.1	-1.2 (0.1)
14	Help with chores	1.4	0.9	3.4	62.8*	1.0	0.9	-0.9 (0.1)
15	Tell what time it is	1.4	1.0	2.1	46.9	1.4*	1.3	-0.2 (0.1)
16	Identify coins	1.9	2.8*	2.1	68.3*	1.8*	1.6*	-0.8 (0.1)
17	Eyesight make it difficult to find something	1.1	0.7	25.5	13.1	1.3	1.5*	0.3 (0.1)
18	Bumps into people	1.2	1.0	2.1	57.2*	1.1	1.1	-0.9 (0.1)
19	Trips over curbs or steps	0.7	-0.5	3.4	50.3*	0.8	0.8	-0.8 (0.1)
20	Happy most of time	-1.1	3.0	0.0	2.1	0.6*	0.8	0.0 (0.1)

Table 3.20 Assessment of CVFQ item quality for proxy responses (continued)

Item	Description	Skew	Kurtosis	Missing Data (%)	Ceiling Effect (%)	Infit Mean Square	Outfit Mean Square	Item Calibration (SE)
21	Visit with relatives	-0.7	0.7	4.8	1.4	1.0	1.1	0.3 (0.1)
22	Make friends easily	-0.2	-0.6	2.1	3.4	0.8	0.8	1.1 (0.1)
23	Affectionate	0.0	-0.7	1.4	2.1	1.3	1.3	1.1 (0.1)
24	Get along well with others	-0.3	-0.4	0.7	2.1	0.7	0.7	0.8 (0.1)
25	Enjoy looking at books	-0.4	-0.5	5.5	4.1	1.1	1.1	0.9 (0.1)
26	Eyesight make difficult to move, run and jump	1.3	1.8	30.1	26.2	1.6*	1.7*	2.8 (0.2)
27	Vision gets in the way of learning	0.9	1.7	29.7	14.5	1.5*	1.8*	2.6 (0.2)
28	Enjoy watching TV, play video game	-0.7	0.5	0.7	2.1	0.9	1.0	0.3 (0.1)
29	Like to travel on vacation	-0.6	0.2	1.4	2.8	0.9	0.9	0.4 (0.1)
30	Enjoy playing with others	-0.8	0.6	0.7	3.4	0.9	0.9	0.4 (0.1)
31	Enjoy drawing	-0.2	-0.3	4.1	2.8	0.9	0.9	0.7 (0.1)
32	Worry about eyesight	-0.2	-0.9	1.4	14.5	1.4*	1.5*	1.2 (0.1)
33	Spend on vision treatment	0.0	-1.1	11.0	22.1	1.3	1.3	0.6 (0.1)
34	Take away time for therapy	0.5	-0.8	18.6	35.2	1.4*	1.4*	0.1 (0.1)
35	Argue about medical care	0.7	-0.6	11.0	41.4	1.5*	1.5*	0.0 (0.1)
36	Bothered by other people's comments	0.7	0.6	26.2	11.7	0.7	0.7	0.3 (0.1)
37	Feel different from others	0.0	-0.8	2.8	9.0	0.9	0.9	1.0 (0.1)
38	Other children looking at mine	-0.2	-0.6	2.8	4.8	0.9	0.9	1.3 (0.1)
39	Teased for vision problem	1.1	1.8	23.4	11.0	0.7	0.7	0.3 (0.1)
40	Worry about can't read, watch, drive	0.5	-0.3	2.8	17.9	0.9	1.0	0.4 (0.1)

Descriptive statistics of skew, kurtosis, percentage ceiling effect and Rasch analysis are shown. Fit statistics (infit and outfit) are indicated by mean square values and item calibration indicates the location of each item in the item-person map (the difficulty of the item). Asterisks indicate items not meeting criteria for skew, kurtosis, ceiling effect or Rasch fit and need further consideration. Each item was asked in the forms of frequency, difficulty, agreement, as appropriate.

3.2.4.2 Rasch analysis

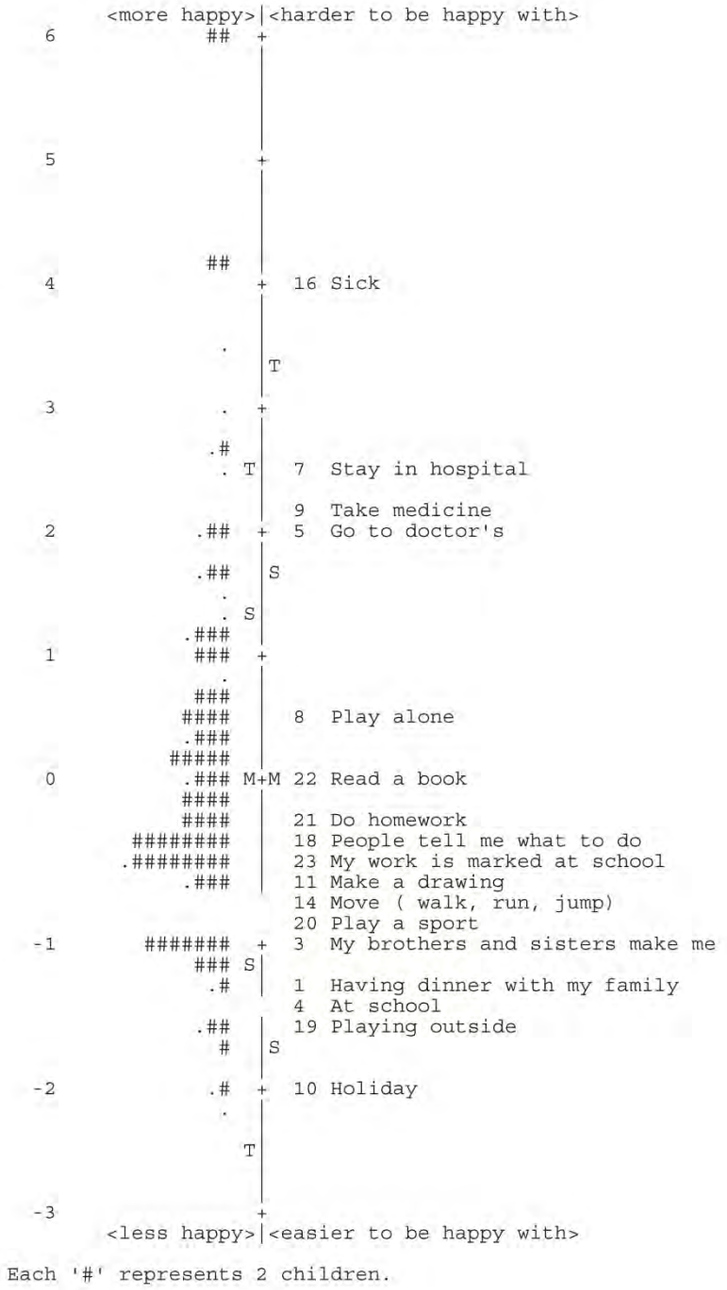
3.2.4.2.1 Person-item map

Figure 3.9 shows children's VQoL (degree of activity difficulty) for the adapted 22-item LVP-FVQ. The right side of the logit scale indicates the number of participants with a given logit value on VQoL. Person and item appear in descending order of ability and difficulty on a common scale. Item-person misalignment was found in the adapted LVP-FVQ, shown by a lack of appropriate items for participants at the top of the scale. Thus items in this questionnaire described tasks that were well within the ability of the respondents. A more even spread of items was obtained for the 17-item AUQUEI scale (degree of activity enjoyment) after item reduction (Figure 3.10), indicating that AUQUEI items align better with respondents than LVP-FVQ items. Similarly, lack of alignment with person ability and item difficulty was found in the adapted CVFQ scale (Figure 3.11). The overall fit results may be summarised as follows (see Table 3.18, 3.19 and 3.20): infit range, 0.7-1.5; outfit range, 0.4-1.9.

Figure 3.9 Person-item map of the LVP-FVQ, showing the distribution of the Rasch calibrated respondents scores (right) and item locations (left)

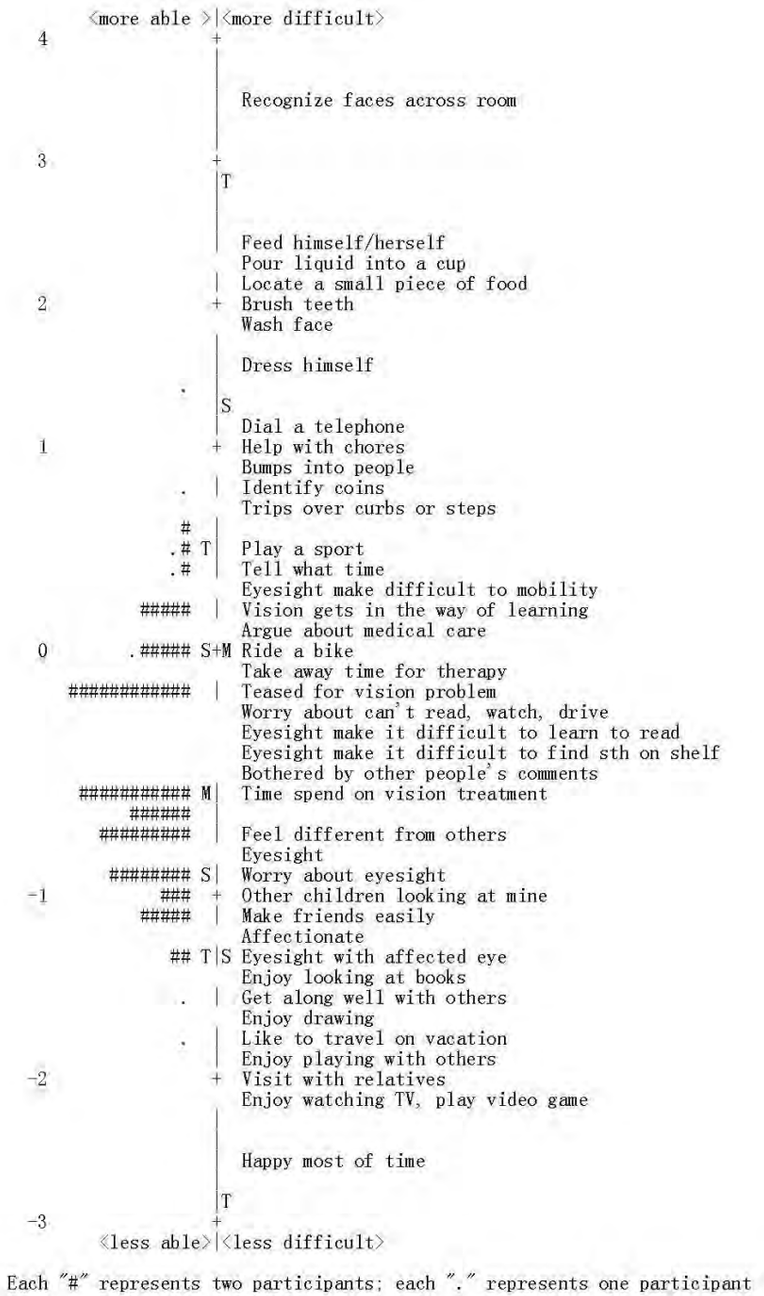
Each "#" represents three children. More able children and more difficult items are near the top. The scale is in log units. M indicates mean; S indicates 1 SD from the mean; T indicates 2 SD from the mean.

Figure 3.10 Person-item map of the Rasch-scaled AUQUEI after item reduction and category collapse, showing the distribution of the Rasch calibrated respondents' scores (right) and item locations (left)



Each "#" represents two children. More life-satisfied children and less favourable activities are near the top. The scale is in log units. M, mean; S, 1 SD from the mean; T, 2 SD from the mean.

Figure 3.11 Person-item map of the Rasch-scaled CVFQ, showing the distribution of the Rasch calibrated respondents' scores (right) and item locations (left)



Lack of corresponding person (participants) match with the top and bottom sections of the items. Each "#" represents two children. More able children and more difficult activities are near the top. The scale is in log units. M, mean; S, 1 SD from the mean; T, 2 SD from the mean.

3.2.4.2.2 Category reduction

Eight out of 23 items in the original AUQUEI questionnaire were found to have disordered thresholds. This indicated that the rating scale of this questionnaire needed to be redesigned for use in this population. In its original form, the AUQUEI rating scale consists of four categories: “not happy at all”, “not happy”, “happy” and “very happy”. The least frequently selected category in this questionnaire was “not happy”, which was not used by 132 out of the 168 participants. Subsequent to initial Rasch analysis, this underutilisation was overcome by combining response 1 “not happy at all” and response 2 “not happy”. Consequently, the scores for all items were recorded by collapsing 4 categories to 3 categories. The category collapse improved targeting of items to participants, as indicated by a reduction in the difference between the mean value for the children and the mean value of the item from 1.30 to 0.5. In addition, category reduction also succeeded in improving person separation from 2.08 to 2.41. This improvement is also reflected in item and person reliability, as illustrated in Table 3.21. With further item modifications, all items showed ordered thresholds. The CVFQ polytomous scales and LVP-FVQ dichotomous rating scales yielded no disordered thresholds.

Table 3.21 Person separation, targeting of items to participants, person and item reliability and residual variance explained by measures for original and revised versions of the AUQUEI

	23 Item AUQUEI ***(4 Categories)***	**23 Item AUQUEI** ***(3 Categories)***	**19 Item AUQUEI** ***(3 Categories)***	**17 Item AUQUEI** ***(3 Categories)***
Person separation	2.08	2.41	2.27	2.18
Difference between item and subject means (subject SEM)	1.30 (0.08)	0.50 (0.10)	0.21 (0.10)	0.03 (0.10)
Cronbach's α (KR-20) person reliability	0.88	0.91	0.89	0.89
Item reliability	0.99	0.99	0.99	0.99
Residual Variance explained by measures (Unexplained variance in 1st contrast) (%)	70.7 (3.4)	84.0 (1.7)	83.2 (1.9)	85.1 (1.9)

3.2.4.2.3 Item reduction

Consistent with previous work on questionnaire development (Pesudovs, et al., 2003), Rasch indices, such as the overall model fit, item and item/person separation reliability, and difference between the item difficulty and person ability were examined. Given the iterative process of item reduction, a number of criteria (see Table 3.13) were followed during the procedure. In the AUQUEI, four items with high skew, kurtosis and ceiling effect were identified based on data distribution. Removal of these items from the present instrument resulted in improved targeting of items to the participants (difference of mean value between items and participants decreased from 0.50 to 0.21). Item 2 ("When I go to bed at night I feel…") and item 13 ("When I watch television I feel…") with poor fit statistics (infit>1.5) and disordered categories were also eliminated, thus, a total of 6 items were excluded from the AUQUEI. Item removal could be continued to further improve item fit, but this would negatively affect person separation. For this reason, the modifications to this questionnaire were a compromise between item fit and person separation, with some items retained in the interests of maintaining a reasonable level of person separation. The original validation of the AUQUEI found internal consistency as indicated by a Cronbach α of 0.88, which remained consistent with the shortened version (0.89); item reliability was also unchanged (0.99). In general, the levels of variance explained by Rasch measures >60% and variance not explained by Rasch measures, but explained by first contrast <5%, are accepted as an indication of the unidimensionality of items without redundancy in a construct. The variance of the data that can be explained by Rasch measures determined by PCAR was found to be 70.7% originally, increasing to 85.1% in the shortened version. The variance of data not explained by Rasch measures, but explained by first contrast, changed from 3.4% to 1.9%, respectively. These findings indicated that the Rasch model fit is improved after category and item reductions. In the adapted AUQUEI, 17 of the 23 original items remained (Table 3.21). Following a similar process, six items were removed from the LVP-FVQ. However, given the low person separation reliability of 1.35 (See Appendix 3), the instrument is unlikely to distinguish between at least two groups of children with normal vision or mild visual abnormalities, as suggested by the Rasch analysis. Thus, further Rasch-scale conversion and analysis was not applicable to the adapted

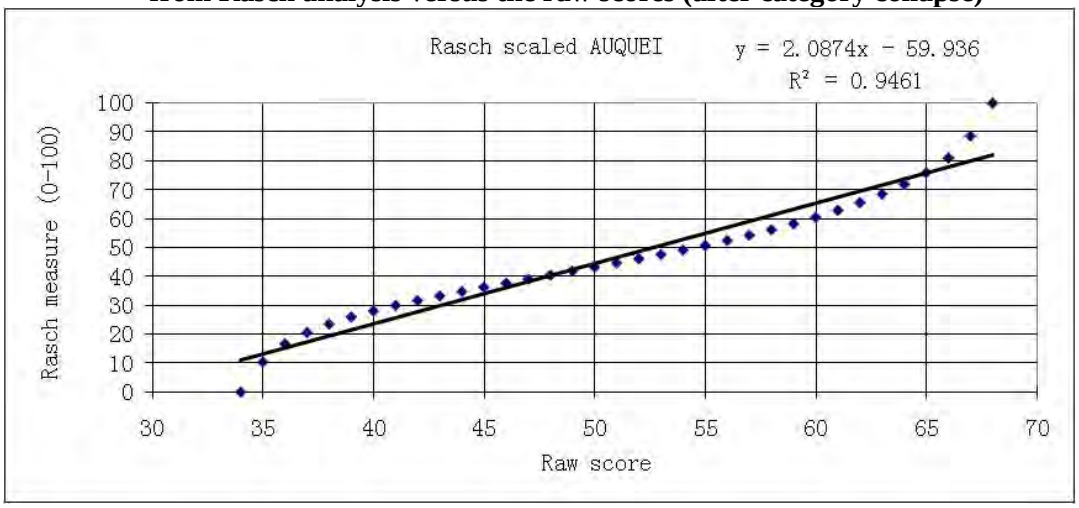
LVP-FVQ. For the same reason in CVFQ, with item exclusion and category adjustment, person separation reliability was still low (pre: 1.78, post: 1.95), and did not reach the criterion of 2.00. Therefore, CVFQ was not subjected to further analysis and score conversion.

The hierarchy of items in the adapted AUQUEI as listed in Figure 3.10 shows that the least enjoyable HQoL activities for children with ID were indicated by the items "When I am sick I feel…", followed by "When I stay in hospital I feel…", "When I take medicine I feel…" and "When I go to the doctor I feel…". In contrast, the most enjoyable HQoL activities were "When I am on holiday I feel…", "When I am playing outside I feel…", "When I am at school I feel…" and "When I am having dinner with family I feel…". Note that least enjoyable activities relate to health care needs while the most enjoyable are related to socialisation and school. This pattern is consistent with the expected degree of enjoyment of a diverse range of daily activities. No such pattern was derived from the LVP-FVQ and CVFQ, given a lack of person separation reliability indicated by Rasch analysis.

3.2.4.2.4 Rasch scale conversion

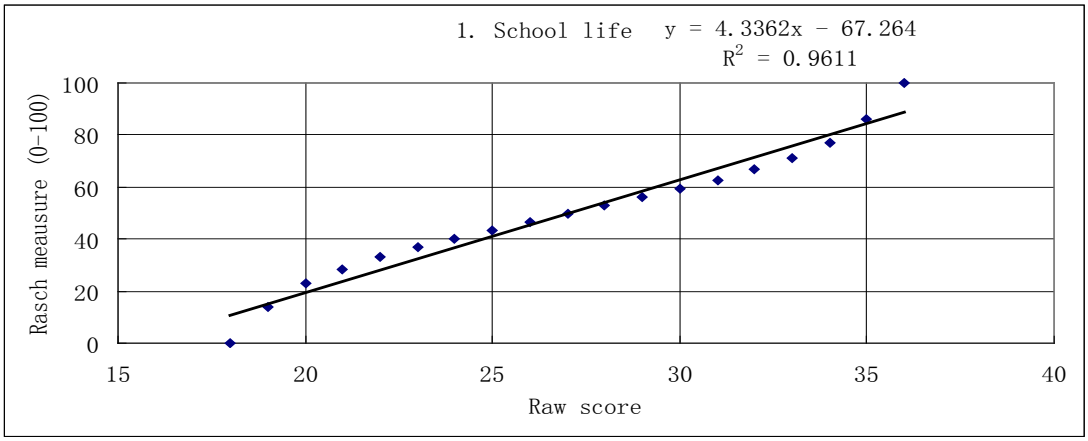
The AUQUEI questionnaire raw score (ordinal score from the Likert scale) was converted to a Rasch person measure. The transformation resulted in a linear interval scale, which allowed further parametric statistical analysis. In order to predict the measure from the raw score, the total scale measure was converted using the following formula: Rasch measure (0-100 scale) = 2.084×Raw score - 59.936(Figure 3.12); for subscale (domain) of "School life", Rasch measure (0-100 scale) = 4.3362 ×Raw score – 67.264; for subscale of "Family life", Rasch measure (0-100 scale) = 11.705×Raw score – 90.364; for subscale of "Health care" Rasch measure (0-100 scale) = 11.178×Raw score – 85.114 (constants in these formulae are taken from line fitting values in Figures 3.12 and 3.13).

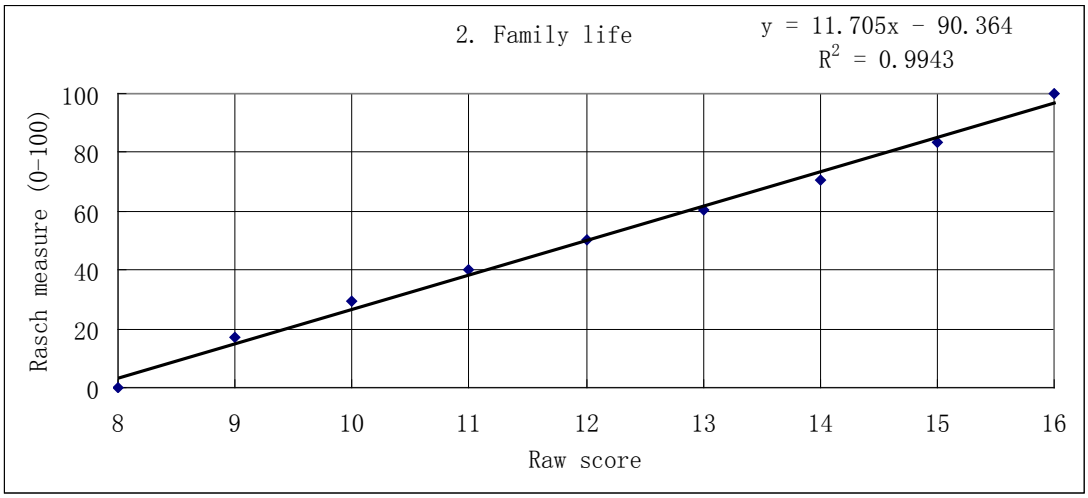
Figure 3.12 Scatter plot of Rasch scaled AUQUEI indicates the person measure estimated from Rasch analysis versus the raw scores (after category collapse)

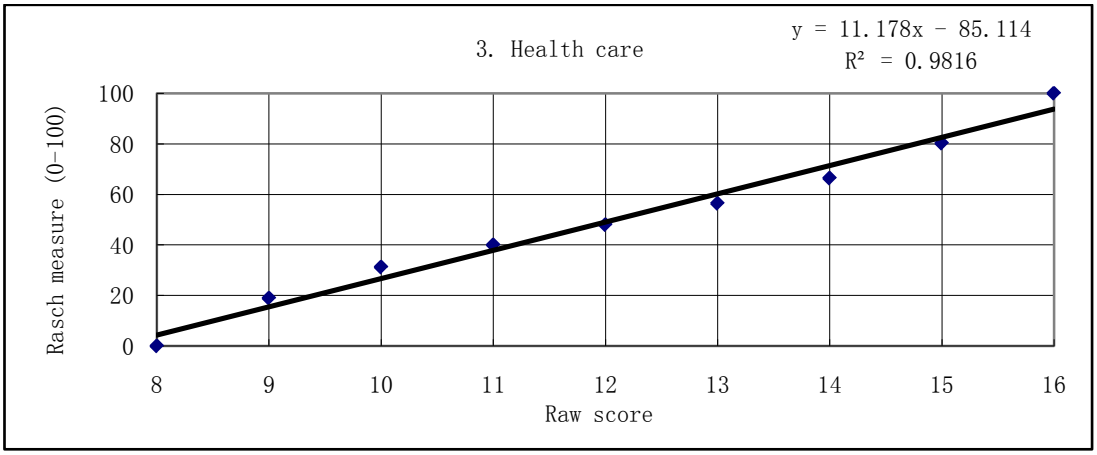


The trend line is generated to simplify the relationship between raw score and Rasch scaled score for future use.

Figure 3.13 Rasch scores for each of the subscales of School life, Family life and Health care assessed using the AUQUEI as a function of raw scores in each subscale after category collapse.







The trend line is generated to simplify the relation between raw score and Rasch scaled measure in following domains: 1) School life, 2) Family life, 3) Health care.

3.2.4.3 Instrument developed for use in main study

In this chapter, the adapted AUQUEI was developed with both conventional methods and Rasch analysis, which helped to further develop an interval-scaled instrument for use in children with ID. On the other hand, the adapted LVP-FVQ and CVFQ did not reach acceptable person separation reliability and were not considered to be suitable for development of an interval-scaled instrument. However, since the results produced from the conventional validation process indicated that these two scales were valid to use with a dichotomous scale and Likert scale respectively, the adapted LVP-FVQ and CVFQ were selected for use in the main study with valid items derived from conventional analysis.

Thus the new instrument kit for use in the assessment of HQoL and VQoL in children with ID in the main study was titled “Health- and Vision-related Quality of life Scales (HVQoLS)”, and included the following subscales and items from the validated and adapted AUQUEI, LVP-FVQ and CVFQ (Table 3.22-Table 3.24).

Table 3.22 Subscales and items of Rasch scored HQoL (AUQUEI)

School life
1. At school, I feel 2. When I play alone, I feel 3. When I make a drawing, I feel 4. When I move (walk, run, jump), I feel 5. When people tell me what to do, I feel 6. When I play a sport, I feel 7. When I do my homework, I feel 8. When I read a book, I feel 9.When my work is marked at school, I feel
Family life
10. When I am having dinner with my family, I feel 11. My brothers and sisters make me feel 12. When it is holiday, I feel 13. When I am playing outside, I feel
Health care
14. When I go to the doctor's, I feel 15. When I stay in hospital, I feel 16. When I take medicine, I feel 17. When I am sick, I feel

Table 3.23 Subscales and items of VQoL (LVP-FVQ)

School activities
1. Can you copy from the board in class? 2. Can you do maths? 3. Can you write your name? 4. Can you remember words easily? 5. Can you read a book by yourself? 6. Can you write the numbers from 1 to 10? 7. Is your handwriting neat? 8. Can you draw a straight line on paper without a ruler? 9. Can you finish your homework on time?
After school activities
10. Can you tie up your shoelaces by yourself? 11. Can you find food on your plate when eating? 12. Can you put toothpaste on your toothbrush by yourself? 13. Can you see the TV clearly? 14. Can you climb up and down stairs? 15. Can you see a person's face across the road? 16. Can you hit a ball when you play? 17. Can you see bus numbers clearly?
Item identification
18. Can you find your favourite toy at play time? 19. Can you pick up a red pencil from a box of pencils? 20. Can you see the picture in your books clearly?

Table 3.24 Subscales and items of HQoL and VQoL (CVFQ)

General health
1. In general, would you say that your child's overall health is:(Q)
General vision
2. At the present time, would you say that your child's eyesight (using both eyes) is: (Q) 3. If your child has an eyesight problem for only one eye, would you say that your child's eyesight in the affected eye is: (Q)
Competence
4. My child can feed himself/herself.(D) 5. My child can recognise faces (friends, relatives) across a room.(D) 6. My child can dress himself/herself.(D) 7. My child can brush his/her teeth.(D) 8. My child can wash his/her face.(D) 9. My child can ride a bicycle.(D) 10. My child can play a sport or an active game (for example, football).(D) 11. My child can locate a small piece of food (a raisin or Cheerio) and grasp it.(D) 12. My child can pour liquid into a cup or glass.(D) 13. My child can dial a telephone.(D) 14. My child helps with chores.(D) 15. My child can tell what time it is.(D) 16. My child can identify coins.(D) 17. My child's eyesight makes it difficult for him/her to find something on a crowded shelf or in a closet.(A) 18. My child bumps into people, walls, or furniture.(F) 19. My child trips over curbs or steps.(F)
Personality
20. My child is happy most of the time.(A) 21. My child likes to visit with relatives.(A) 22. My child makes friends easily.(A) 23. My child is affectionate.(A) 24. My child gets along well with our other children and friends.(A) 25. My child enjoys looking at books.(A) 26. My child's eyesight makes it difficult for him/her to learn to walk, run, skip, or jump.(A) 27. My child's vision gets in the way of his/her learning.(A) 28. My child enjoys watching television, videos, or playing video games.(A) 29. My child likes to travel on family vacations.(A) 30. My child enjoys playing with others (sisters and brothers or friends).(A) 31. My child enjoys drawing, painting or other art activities.(A)

Table 3.24 Subscales and items of HQoL and VQoL (CVFQ) (Continued)

Family impact
32. How much of the time do you worry about your child's eyesight?(F)
33.How much time do you need to spend on treatment for your child's vision problem (eye doctor appointment, patching, eye drops, therapy)?(F)
34.Does the time you spend on your child's vision problem (eye doctor appointments, patching, eye drops, therapy) take away from time you would like to spend with your other children or husband/wife?(F)
35.Do you and other family members (your spouse or parents) argue about the medical care your child is getting or about treatment that the doctor has prescribed?(F)
36. I am bothered by other people's comments about my child's vision or eyes when I take him/her to a store or mall.(A)
37. My child feels different from other children.(A)
38. I notice other children looking at my child.(A)
39. My child is teased because of his/her vision problem.(A)
40. I worry that my child may not be able to read. (A)

Indicated in brackets behind each item is the type of ordinal scale used: (Q) Quality scale, (F) Frequency scale, (A) Agreement scale, (D) Difficulty scale.

Four categories of "Not happy at all"(1), "Not happy"(1), "Happy"(2) and "Very happy"(3) were used in the section of "child's perception". After category collapse, the response options "Not happy at all" and "Not happy" were combined as one option, i.e. "Not happy", giving the children three choices for each item in the AUQUEI. Further transforming scores to interval scales followed the formulae derived from the plots shown in Figure 3.12 and Figure 3.13. A dichotomous option of "Yes" (1) or "No" (0) was used in the LVP-FVQ. For the HQoL and VQoL domains in the modified CVFQ section of the new instrument, which represent proxy perception, items were allocated to five types of scales, Quality (Q): Excellent (4), Very good (3), Good (2), Not bad (1), Poor (1); Frequency (F): Never (4), Seldom (3), Sometimes (2), Often (1), Usually (1); Agreement (A): Strongly disagree (4), Disagree (3), Slightly agree (2), Agree (1), Strongly agree (1); Difficulty (D): Easy (4), With a little difficulty (3), With difficulty (2), With extreme difficulty (1), Can't complete (1). Coding of items 20-25, 28-31 were reversed, in accordance to the criterion that lower scores represent lower underlying traits in the context of HQoL and VQoL.

3.2.5 Discussion

The three instruments selected here were initially developed for use in children with ND but the present study included children with ID. Thus, the process of development and refinement of the questionnaires was conducted to maximise the likelihood that items

would be comprehensible and meaningful to children in this group. The items not applicable to the lives of children with ID, or those beyond their comprehension were deleted or rewritten. Rasch analysis identified misalignments between some items and participants' lifestyles and capabilities, suggesting that these items were not appropriate for this population.

The LVP-FVQ was initially developed and validated in a population of children with low vision and ND. When applied to a sample of children with ID in this study, the psychometric properties of this instrument, such as person separation were not found to reach acceptable levels. Low item precision and misalignment might be attributed to the fact that the original items were used for children with low vision. Thus, many items may have been relatively easy for the population tested here. This is not surprising since just over one-third of the children had a form of visual abnormality, but most did not have low vision. Further study in children with ID who have a wide range of visual abilities is required to gain a better understanding of the contribution of vision to HQoL. Similarly, the CVFQ was developed initially and validated in a population of children from an eye hospital, and thus, some items were focussed on the children's vision condition and treatment. Given the relatively higher vision capacity in current study groups, low item precision and misalignment was revealed. Therefore, further Rasch analysis was not applicable for this subscale.

Rasch analysis indicated disordered categories in the AUQUEI, and modifications allowed correct ordering. For this reason, the instrument was modified with the intention of ensuring that thresholds were correctly ordered. The disordered threshold on the four-category scale suggested that simplification of the rating scale was required when children were asked to respond to questions relating to their HQoL. Good person separation was demonstrated by Rasch analysis (Figure 3.10) inferring that different people within the population expressed different levels of enjoyment of daily activities, such as those related to health care, school and leisure time, and peer/family interaction. Those items listed in the AUQUEI matched appropriately with children's perception of HQoL, as implied by the person-item targeting (Figure 3.10). With appropriate design

and questionnaire administration, children with ID are able to indicate the way in which activities affect their HQoL. The modified AUQUEI appears to be a useful tool for assessment of HQoL in children with ID.

The relationship between self-reported VQoL and HQoL could not be explored, given the limited person separation in the LVP-FVQ. In addition, criterion validity of the instruments in terms of the extent to which the instruments were related to other measures such as visual function (Gothwal, et al., 2003), is an unknown factor. Due to limited subjects with visual impairment recruited in this pilot study, the relationship between HQoL, VQoL and measures of visual function such as visual acuity and stereoacuity in children with ID are explored in the main study (Chapter 4).

3.2.6 Conclusion

Rasch analysis allowed modifications of instruments that had previously been validated for a different population, and to test their applicability to the population of children with ID here. The AUQUEI is useful in providing information on HQoL in children with ID, while the LVP-FVQ and CVFQ were not found to be appropriate for Rasch analysis, as used in children with ID and with visual impairment. Therefore, for the latter questionnaires raw scores derived from the dichotomous and Likert scales were used in the main study.

CHAPTER 4 MAIN STUDY: APPLICATION OF HEALTH- AND VISION-RELATED QUALITY OF LIFE SCALES IN CHILDREN WITH ID

Chapter 3 described how conventional questionnaire development, i.e. classical theory test, and modern theory of questionnaire development, i.e. Rasch analysis, were used to modify instruments that had previously been validated for children with ND, and to test their applicability in children with ID. The Health- and Vision-related Quality of life Scale (HVQoLS) that was developed for use in this study included the adapted items from the AUQUEI, LVP-FVQ, and CVFQ. Item reduction and category collapsing were conducted to enhance the applicability of the instruments to the population of interest. The instruments that were adapted for use in this study have not been used previously in a large sample population with heterogeneous characteristics, and have not been used to compare HQoL or VQoL in children with and without ID.

The influence of reduced visual function, such as low visual acuity, on HQoL and VQoL in children with ND has been widely addressed as discussed in Chapter 1 (Birch, et al., 2007; Chak & Rahi, 2007; Felius, et al., 2004; Gothwal, et al., 2003; Nirmalan, et al., 2004; Quinn, et al., 2004). However, the impact of reduced visual function has not been explored in children with ID. Therefore, the link between objective visual function (determined by clinical assessment) and subjective perceptions of HQoL and VQoL (determined by questionnaire results) in this population is not clear. Given the evidence that visual abnormality and impairment have a negative impact on the quality of life of children with ND, this study aimed to explore the extent of the impact of visual abnormality and impairment on HQoL and VQoL in children with and without ID using valid instruments.

4.1 Characterising visual status in children with and without ID

4.1.1 Aim

To characterise visual status in developmental age matched children with and without ID

4.1.2 Methods

4.1.2.1 Participants

299 children with ID were recruited from the Developmental Assessment Department, Shanghai Children's Medical Centre (SCMC, n=53), and the Shanghai School of Special Education (SSSE, n=246). In addition, 143 children with ND were recruited from SCMC (n=32), the Shanghai Fushan Primary School (SFPS, n=95), and the Shanghai Low Vision School (SLVS, n=16). The children with ID were aged from eight to eighteen years, while the children with ND were aged from four to nine years, giving an approximate equivalence in cognitive capacity (See Section 2.2.7). Given that all schools and hospitals are located in the same district in Shanghai, the participants in this study hold similar socioeconomic characteristics.

4.1.2.2 Selected characteristics

The participants were classified in terms of developmental age, gender, IQ scores and visual functions. The latter was defined according to best corrected binocular visual acuity (BCBV) and stereoacuity. Vision screening was designed to identify visual abnormality, which is defined in Table 2.4. Participants with decreased visual acuity (poorer than 0.2 in LogMAR record), but no ocular pathological cause detected by vision screening were defined as having amblyopia (Simons, 2005). The method of vision screening is described in Section 2.3. Participants found to have significant visual abnormality were referred for further assessment as appropriate.

4.1.2.3 Participants grouping

The participants were categorised on the basis of the following two measures of visual function.

1. BCBV: Participants were divided into three subgroups based on BCBV: normal vision, visual abnormality, and visual impairment (as defined previously in Table 2.3).

2. Stereopsis: Children with stereoacuity better than 100 seconds of arc were allocated to the normal stereopsis group and those with stereoacuity equal to or worse than 100 seconds of arc were allocated to the abnormal stereopsis group. In order to compare interocular differences in visual acuity in participants with abnormal stereopsis, children in this group were divided into the following subgroups: stereopsis deficit (100”- 400”) and stereopsis impairment (800” or worse).

4.1.2.4 Data analysis

The associations between visual functions (BCBV, stereoacuity) and IQ scores were examined using Pearson’s correlation analyses. An independent sample t-test was used to compare visual functions between children with and without ID, and across gender within each of those groups. Interocular differences in visual acuity were compared across three subgroups of children with ID (normal stereopsis, stereopsis deficit and stereopsis impairment) using an ANOVA test with a Bonferroni-adjusted post hoc test.

4.1.3 Results

4.1.3.1 Participants

Reliable records of chronological age and IQ score were available for 251 (83.9%) children with ID and 138 (96.5%) children with ND. Data on participants’ age, gender and IQ are shown in Table 4.1. The developmental age (as defined in section 2.2.7) of children with ID was similar to that of children with ND, while gender was slightly biased toward males in children with ID.

Table 4.1 Demographic characteristics of the 389 participants with and without ID

Subgroup	**Age in years (mean ± SD)**	**Gender (% male)**	**IQ (mean ± SD)**
Children with ID (n=251)	Chronological:13.1±3.3 Developmental:6.3±1.9	64.9	48.5±9.2
Children with ND (n=138)	Chronological: 6.6±1.8	52.9	WNR[a]

[a] *WNR: Within normal range*

4.1.3.2 Vision screening

241 children with ID completed vision screening, excluding 58 children who failed to complete the screening because their cooperation could not be maintained. Of the children who completed vision screening, 93 (38.6%) were found to have at least one form of visual abnormality (see Table 4.2 for details of abnormalities). All 93 children were referred for further assessment. Of the 41 children with refractive error, which consisted of hyperopia, myopia, astigmatism or anisometropia, 21 achieved acuity of at least 20/30 with refractive correction, indicating that the dominant (22.6% of 93; other percentages below are out of 241) visual abnormality was uncorrected refractive error in these cases. 72 (29.9%) children who could not improve their vision with refractive correction, had at least one of the following abnormalities: 31 with constant strabismus (19 unilateral, 12 alternating); 7 with anisometropia and strabismus; and 25 with suspected pathology of the retina or ocular media. The remaining 47 (19.6%) children were considered to be amblyopic, and of these, 21 had unknown ocular aetiology.

Overall, the prevalence of visual abnormalities in children with ID as defined here was: refractive error 41 (17.1%), strabismus 31 (12.9%), anterior ocular abnormality 6 (2.5%), posterior ocular abnormality 19 (7.9%), and unknown aetiology 21 (8.7%). In addition to visual abnormality, five children were found to have colour vision abnormality (2.1%).

Table 4.2 Visual abnormalities identified in 93 intellectually disabled children with visual abnormality during vision screening

Visual Abnormality	Definition	Number of Subjects
Reduced visual acuity	More than 2-line interocular difference or better eye VA poorer than 20/30.	88
Anisometropia	More than 1.00DS or DC.	14
Manifest strabismus	Any, apparent on cover test.	31
Hyperopia	More than +3.50D in any meridian.	5
Myopia	More than -3.00D in any meridian.	20
Media opacity	Any, apparent on direct ophthalmoscopy.	8
Astigmatism	More than 1.50DC at 90 or 180 degrees, or more than 1.00DC at an oblique meridian.	15
Optic neuropathy	Any, apparent on direct ophthalmoscopy.	17

Vision screening was completed in 111 children with ND. Excluding the16 children

with low vision, the prevalence of visual abnormalities of the remaining children was: refractive error 4 (4.2%), strabismus 3 (3.2%), anterior ocular abnormality 1 (1%), suspected posterior ocular abnormality 2 (2.1%), amblyopia or unknown pathology in visual pathway 4 (4.2%), and colour vision abnormality 1 (1%). In the 16 children with low vision, 7 had congenital cataract, 5 had albinism, 3 had retinopathy of prematurity, 2 had cerebral visual impairment, and 1 had congenital glaucoma.

Table 4.3 and figures 4.1 to 4.4 show percentages of participants with ND and ID, with normal, abnormal or impaired vision, and classified according to level of BCBV and stereoacuity. The 263 children with ID who provided visual acuity data were divided into three subgroups according to BCBV. Of these, 153 (58.2%) were allocated to the normal vision subgroup, 69 (26.2%) to the visual abnormality subgroup, and 41 (16.6%) were allocated to the visual impairment subgroup (see table 4.3, Figure 4.1 and Figure 4.3). Of the 224 children with ID who had a valid record for stereoacuity, 64 (28.6%) had normal stereopsis (stereoacuity< 100”), and the other 160 (71.4%) had stereopsis deficit (stereoacuity ≥ 100”) (see table 4.3, Figure 4.2 and Figure 4.4). In the 108 (48%) children with stereopsis impairment (stereoacuity≥ 800”), 43 had normal BCBV, 29 had amblyopia, 20 had refractive error (without amblyopia), 5 had strabismus (without amblyopia), and 11 had ocular pathology (data not shown in figures). In the 118 children with ND, 93 (78.8%) had normal stereopsis and 25 (21.2%) had stereopsis deficit, including 15 (12.7%) with stereopsis impairment (see Figure 4.4).

Figure 4.1 Comparison of best corrected binocular vision in children with and without intellectual disabilities

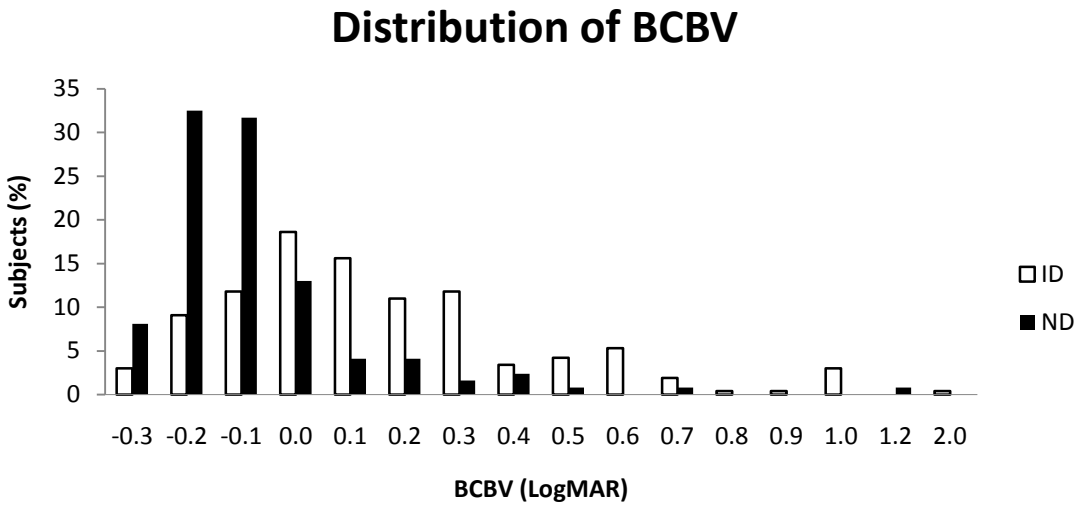


Figure 4.2 Comparison of stereoacuity in children with and without intellectual disabilities

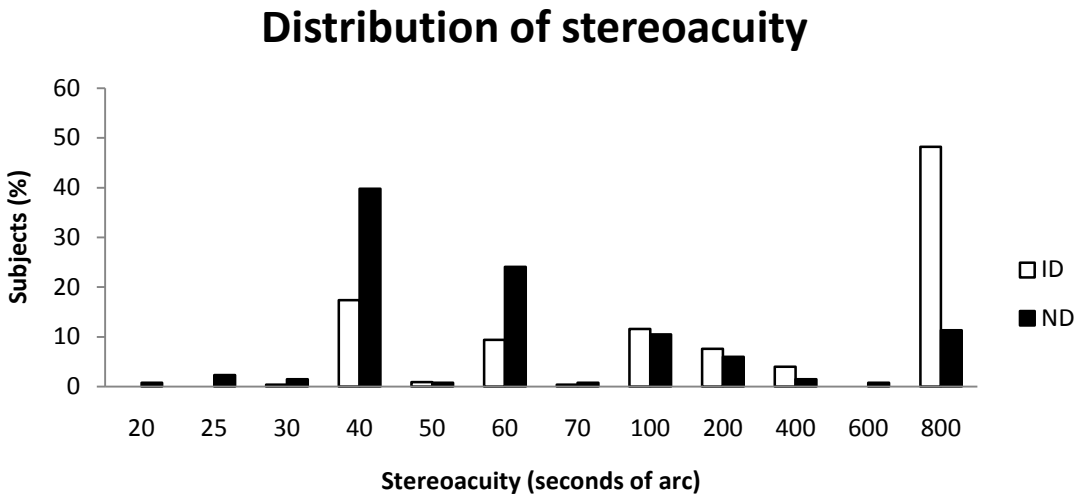


Figure 4.3 Percentage of children with normal or abnormal vision (as defined in text; see also Table 2.3) and with normal development (ND) and intellectual disability (ID)

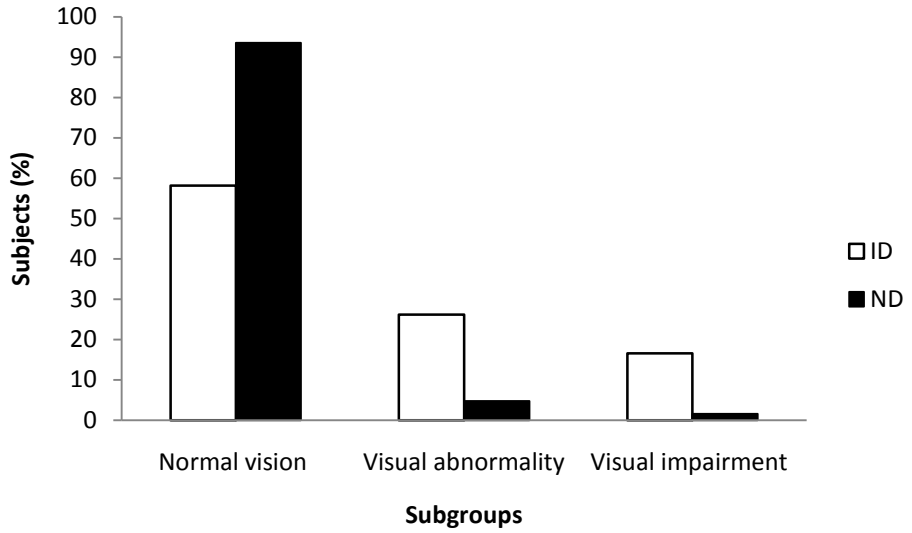
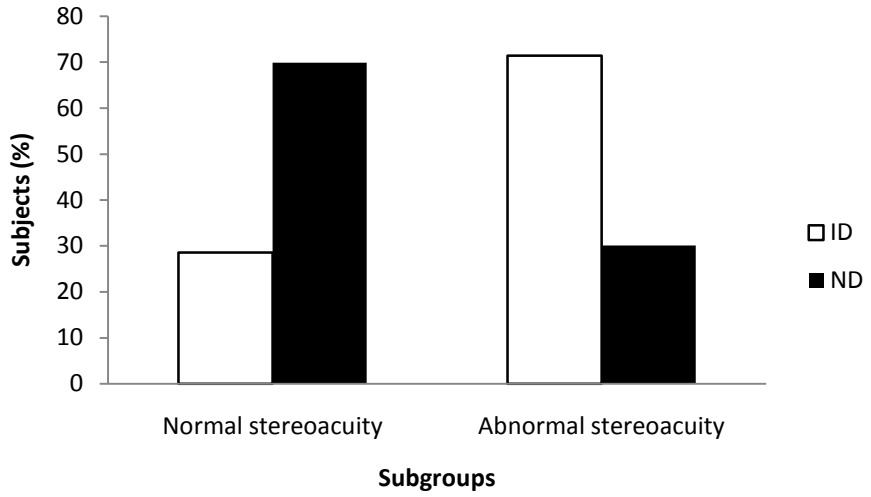


Figure 4.4 Percentage of children with normal or abnormal stereoacuity, and with normal development (ND) or intellectual disability (ID)



In children with ID, a significant association (r=0.38, P<0.01) was found between BCBV and stereopsis. Male participants (mean=0.05, SD=0.2) had significantly better logMAR acuity than female participants (mean=0.10, SD=0.3, P<0.01). No gender differences were found for stereopsis. In children with ND, a significant association (r=0.25, P<0.01) was found between BCBV and stereopsis. No gender differences were found in BCBV or stereopsis in this group.

An association was found between IQ score and BCBV (r=0.21, P<0.01), and between IQ score and stereoacuity (r=0.30, P<0.01). Consistent with this, children with ND had better BCBV and stereopsis than children with ID (P<0.01). In children with ID, those with mild ID had better BCBV and stereopsis than those with moderate ID (P<0.01, Table 4.3).

Of the 183 children with ID and a valid IQ score, 127 had moderate ID and 56 had mild ID. Visual impairment was found in 19 (14.8%) of the children with moderate ID and 3 (5.5%) of the children with mild ID. Normal vision was found in 78 (60.9%) of the children with moderate ID and 39 (70.9%) of the children with mild ID. The remainder were classified as having visual abnormality (see definition in Table 2.3, Section 2.2.7).

Table 4.3 Comparisons of visual function in children with ID and children with ND

Subgroup	**Mean score (SD) of best corrected binocular vision (LogMAR)**	**Mean score (SD) of stereopsis (stereoacuity in arc seconds)**
ID	0.16 (0.3), n=263	442.1(353.6), n=224
ND	-0.08 (0.2), n=123	77.3 (98.2), n=118
Level of significance for the difference between mean scores	**P<0.01**	**P<0.01**
Moderate ID	0.15 (0.3), n=127	424.6 (354.5), n=109
Mild ID	0.03 (0.2), n=56	265.7 (314.8), n=55
Level of significance for the difference between mean scores	**P<0.01**	**P<0.01**

A post hoc comparison using the Bonferroni test was applied to the interocular (between right and left eye) difference in distance VA and near VA in subgroups of children with ID and with diverse stereopsis status. Statistically significant differences were found between children with normal stereopsis and stereopsis impairment (Table 4.4). As expected, the results indicated that the mean interocular differences in distance and near VA for children with stereopsis impairment were significantly higher than those of children with stereopsis deficit or normal stereopsis. There were no significant differences between children with stereopsis deficit or normal stereopsis.

Table 4.4 Comparison of interocular difference in children with ID and with diverse stereopsis status

Interocular difference	Normal stereopsis vs. stereopsis deficit	Normal stereopsis vs. stereopsis impairment	Stereopsis deficit vs. stereopsis impairment
Difference in distance VA (LogMAR)	Mean= 0.06, SD=0.06 P>0.05	Mean=0.16,SD=0.05 **P<0.01**	Mean=0.10, SD=0.06 P>0.05
Difference in near VA (LogMAR)	Mean= 0.04, SD=0.04 P>0.05	Mean=0.08, SD=0.03 **P<0.05**	Mean=0.04, SD=0.03 P>0.05

4.1.4 Discussion

The significantly higher prevalence of visual abnormalities in children with ID than those with ND is consistent with previous findings (Kwok, et al., 1996; Woodhouse, Adler, & Duignan, 2003; Woodhouse, Griffiths, & Gedling, 2000; Woodhouse, et al., 1997; Woodruff, 1977; Woodruff, Cleary, & Bader, 1980). Among these visual abnormalities, refractive error was the most common and correctable. However, comparison of the prevalence of visual abnormalities across studies is limited by the different definitions of refractive error that are used among studies in the published literature. In the present study, refractive error occurred in 17% of children in the ID group, which is approximately four times higher than the prevalence of refractive error in the control group (4.2%). In children with ID, about 50% of those with refractive error achieved at least 6/9.5 (LogMAR: 0.2) visual acuity with optical correction, while only six children (15%) presented wearing prescription glasses. These findings indicate that there is a significant demand for refractive correction in children with ID.

The prevalence of amblyopia was about five times higher in children with ID (19.6%, 47/241) compared to children with ND (4.2%, 4/95). This figure does not include those who were unable to undergo vision screening. Apart from those with unexplained decreased BCBV (8.7%, 21/241), which could not be firmly diagnosed by vision screening, at least 72 (30.0%, 72/241) children could have had better corrected vision if they had undergone vision screening earlier.

Interestingly, a proportion of children (3.0% in children with ID, 8.1% in children with

ND) achieved BCBV at 6/3 (LogMAR; -0.3), higher than might be expected based on previous findings in this age range (Vision in Preschoolers Study Group, 2004). This finding may reflect the fact that visual acuity measured in the present study was in binocular viewing conditions, while previous studies have recorded monocular acuities (Cagenello et al, 1993). In addition, Lea symbol acuity is reportedly higher than other commonly used acuity tests such as Bailey-Lovie in school-aged children and HOTV and Landot C in young children (Dobson, Maguire, Orel-Bixler, Quinn, & Ying, 2003; Vision in Preschoolers Study Group, 2004). Dobson et al (2003) attributed this difference to a relatively low number of optotypes on the Lea symbols test, with a correspondingly higher chance of guessing correctly (Dobson et al, 2003).

A substantial proportion (29%, 21/72) of this group had no ocular aetiology such as strabismus or refractive error. This finding implies that the reason for the decreased VA in these children may be a result of disorders located in the visual pathway or at higher levels in the brain. Given the high prevalence of cerebral disorders in children with ID, the cause of decreased vision in children with ID requires further investigation. In such cases, categorisation of amblyopia might not be appropriate to apply in this subgroup.

Although a high rate of stereopsis impairment (stereoacuity $\geq$ 800") was found in children with ID, only 60.2% of these children were found to have visual abnormality by visual screening. The reasons for the mismatch between the stereopsis test and other screening results might be due to two factors. First, findings from previous studies (L. S. Nielsen, L. Skov, & H. Jensen, 2007a) suggest that stereopsis measures are not always in accordance with a diagnosis of amblyopia orstrabismus in children with ID. This is possibly because these children may not understand the test or may be reluctant to respond. Second, the poor stereopsis in some children with ID is unsurprising in view of high interocular difference in VA. As indicated in Table 4.4, the interocular difference in VA in children with stereopsis impairment is larger than those with normal stereopsis, which indicates that significant interocular differences in VA are likely to contribute to the high rate of abnormal stereopsis in children with ID.

Most reports of visual outcomes use best corrected visual acuity as a main indicator for

visual function. In this study, the strong association between stereoacuity and BCBV, and the strong distinction in stereoacuity between children with and without ID, suggests that stereoacuity may be an additional indicator for visual function in future research, if its reliability is confirmed. However, measures of effectiveness (i.e., sensitivity, specificity, positive predictive value and negative predictive value) of stereopsis tests in children with ID are needed before stereopsis can be used as an indicator of visual function in this population.

As described in Chapter 1 (see Section 1.2), a high prevalence of colour vision defects has been reported in children with ID (Perez-Carpinell, de Fez, & Climent, 1994). However, in the present study, the rate of colour vision abnormality in children with ID is low (2.1%, Section 4.1.3.2), and similar to the prevalence found in a population-based study of children with ND (Cosstick, Robaei, Rose, Rochtchina, & Mitchell, 2005) and found in male athletes participating in the Special Olympic Games (Woodhouse, et al., 2003). Taken together, these findings suggest that the prevalence of colour vision defects is similar in populations with and without ID.

Given the poor communication capacity of children with moderate ID, their needs may be neglected by parents and clinical practitioners. In the present study, those with moderate ID were more likely to have poorer BCBV and stereoacuity than those with mild ID. This fact, together with the low rate of optical correction for refractive errors, indicates a need for primary eye care services for the population of children with ID, especially those with moderate to severe ID.

4.2 Children's perceptions of health- and vision- related quality of life

4.2.1 Aims

1.To evaluate the extent of the impact of visual abnormality and impairment on HQoL and VQoL in children with and without ID

2. To compare perceptions of HQoL and VQoL between children with and without ID

4.2.2 Hypotheses

1. Visual abnormality and impairment have a negative impact on HQoL and VQoL in children with ID.

2. Perception of HQoL and VQoLis lower in children with ID than in children with ND.

4.2.3 Methods

4.2.3.1 Participants grouping

Participants and subgroups were described in Sections 2.2.7 and 4.1.2.1.

4.2.3.2 Questionnaire instrument

The HVQoLS (see Section 3.2.4.3) developed in the pilot study was given to each participant at SCMC by the investigator. Recruitment was by invitation. At the SCMC, participants were instructed to complete the questionnaire after they underwent a routine developmental assessment consultation with a paediatrician. Demographic information was recorded, based on respondents' answers in the background information section of the questionnaire or from their clinical records. For the participants from each of the schools (SSSE, SFPS, SLVS), the questionnaires were completed in class and returned to the investigator (as described in Section 2.2.9). The demographic information on these participants was obtained from the schools' medical records.

4.2.3.3 Subscales

The subscales of HVQoLS investigated in this section are derived from children's perception, including "School life", "Family life" and "Health care" of HQoL; "School

activities", "Afterschool activities" and "Item identification" of VQoL. The higher scores in each domain indicate higher HQoL and VQoL, respectively. The instruments and manner of questionnaire administration to children with and without ID were identical (see Section 2.2.9).

4.2.3.4 Data analysis

As described in Table 2.6 (see Section 2.5.1) the associations between visual function (BCBV, stereoacuity), developmental age, IQ scores, and composite scores of the HVQoLS subscales in children with ID were examined using Pearson's correlation analyses. HQoL and VQoL were explored in subgroups of children with ID with normal vision, visual abnormality and visual impairment using an ANOVA test with a Bonferroni-adjusted post hoc test. An independent sample t-test was used to compare HQoL and VQoL in children with and without ID. The influences of developmental age and gender on children's perception of HQoL and VQoL were explored using a two-way between groups ANOVA based on developmental age and gender subgroups.

4.2.4 Results

Children with ND were found to have higher scores than children with ID in HQoL subscales of "Overall HQoL", "School life" and "Family life" ($P<0.01$; Table 4.5). In contrast, there were no significant differences revealed in the VQoL subscales between children with and without ID (Table 4.5).

The association between measures of visual function (BCBV and stereopsis) and HQoL and VQoL was explored. A correlation matrix is shown in Table 4.6. BCBV correlated moderately with children's perception of VQoL in those with ND, but not in children with ID. There was no significant correlation between visual function and subscales of HQoL in children with or without ID. Developmental age was associated with VQoL, especially in the subscales relevant to school activities in children with and without ID. IQ score had a mild association with BCBV and stereoacuity in children with ID. This result is in line with the comparison of visual function in children with mild and moderate ID in the previous section (Table 4.3).

The impact of visual abnormality on children's HQoL and VQoL was further investigated by comparing findings between children with normal and abnormal BCBV and stereopsis. In children with ID, there were no significant differences in any of the subscales of HQoL and VQoL between children with normal and abnormal BCBV (Table 4.7a). In children with ND, significant differences in "Overall VQoL" and in "After-school activities" and "Item identification" were found between children with normal and abnormal BCBV (Table 4.7a). When comparing the impact of abnormal stereopsis in children with ID, a significant difference was found in the HQoL subscale of "Family life". In children with ND, there were no differences between children with normal and abnormal stereopsis in "Overall HQoL" or the HQoL subscales, but significant differences were found for "Overall VQoL" and "School-activities" and "After-school activities" between subgroups with normal function and abnormal stereopsis (Table 4.7b). A post hoc comparison using the Bonferroni test was applied to the subscales of "Overall VQoL", "After-school activities" and "Item identification" in subgroups of children with ND. Statistically significant differences were found between children with normal BCBV, visual abnormality and visual impairment (Table 4.8). The results indicated that the mean score for children with visual impairment was significantly lower compared with children with normal vision for "Overall VQoL", "After-school activities" and "Item identification"; and was significantly lower compared with children with visual abnormality for "After-school activities" (Table 4.8). There were no significant differences between children with visual abnormalities and normal vision.

Table 4.5 Comparison of HQoL and VQoL between children with and without ID

	Subscales	**ND**	**ID**
HQoL	Overall HQoL	Mean=45.09, SD=10.9	Mean=40.38, SD=10.9 **P<0.01**
	School life	Mean=55.27, SD=14.3	Mean=49.17, SD=15.8 **P<0.01**
	Family life	Mean=74.77, SD=15.4	Mean=67.59, SD=18.6 **P<0.01**
	Health care	Mean=2.18, SD=25.3	Mean=2.83, SD=24.0 P=0.80
VQoL	Overall VQoL	Mean=17.07, SD=1.7	Mean=17.00, SD=2.4 P=0.84
	School activities	Mean=7.05, SD=2.4	Mean=7.19, SD=1.7 P=0.52
	After-school activities	Mean=7.01, SD=1.0	Mean=7.07, SD=1.2 P=0.62
	Item-identification	Mean=2.87, SD=0.4	Mean=2.86, SD=0.4 P=0.75

Table 4.6 Pearson's correlation matrix between Age, IQ, Visual function, and HVQoLS subscales scores[a].

Subgroups	Characteristics	BCBV (LogMAR)	Stereoacuity	School Activities [b]	After-school Activities [c]
ID (N=182)	Developmental age			0.23	0.28
	IQ score	-0.19	-0.25		
	BCBV[d]		0.38		
ND (N=118)	Chronological age			0.33	0.36
	BCBV [d]		0.25	-0.41	-0.36

[a]*Only statistically significant correlation coefficients(r) (P <0.01) shown in the table*
[b]*VQoL "School Activities" subscale score (HVQoLS)*
[c]*VQoL "After-school Activities" subscale score (HVQoLS)*
[d]*BCBV recorded with LogMAR, where the lower score indicates higher visual ability*

Table 4.7a Comparison of the impact of abnormal BCBV on HQoL and VQoL in children with and without ID

Subgroup	Results from ANOVA on HQoL subscales				Results from ANOVA on VQoL subscales			
	Overall HQoL[a]	School Life	Family life	Health care	Overall VQoL[b]	School activities	After-school activities	Item identification
ID (N=248)	F(2,232)=0.19 P=0.83	F(2,242)=0.24 P=0.79	F(2,238)=0.34 P=0.72	F(2,245)=0.99 P=0.38	F(2,239)=1.53 P=0.22	F(2,245)=0.85 P=0.43	F(2,243)=1.17 P=0.31	F(2,249)=0.09 P=0.91
ND (N=133)	F(2,120)=0.54 P=0.58	F(2,124)=0.1.56 P=0.21	F(2,126)=0.05 P=0.95	F(2,127)=0.54 P=0.59	F(2,27)=6.16 **P<0.01**	F(2,27)=1.39 P=0.27	F(2,27)=12.37, **P<0.01**	F(2,25)=3.76 **P=0.04**

[a] *Total score of HQoL items with Rasch scale,* [b] *Total score of VQoL items with Likert scale*
P values indicate significance level of the differences in HQoL or VQoL in subgroups of diverse BCBV, i.e. Normal vision, Visual impairment and Visual abnormality.

Table 4.7b Comparison of impact of abnormal stereopsis on HQoL and VQoL in children with and without ID

Subgroup	Results from independent t-test on HQoL subscales				Results from independent t-test on VQoL subscales			
	Overall HQoL[a]	**School Life**	**Family life**	**Health care**	**Overall VQoL[b]**	**School activities**	**After-school activities**	**Items identification**
ID (N=211)	t(199)=1.36 P=0.18	t(206)=1.03 P=0.31	t(202)=1.94 **P=0.04**	t(208)=0 P=1.00	t(204)=0.05 P=0.95	t(209)=0.21 P=0.83	t(206)=-0.56 P=0.58	t(211)=0.79 P=0.43
ND (N=129)	t(118)=1.41 P=0.16	t(122)=0.70 P=0.49	t(124)=1.31 P=0.18	t(125)=0.21 P=0.98	t(124)=-3.30 **P<0.01**	t(124)=-2.14 **P=0.03**	t(53)=-3.45 **P<0.01**	t(43)=-1.28 P=0.21

Table 4.8 Results from ANOVA with Bonferroni post hoc test on VQoL subscales in children with ND

Subscale of VQoL	Visual impairment	Visual abnormality	Normal vision
Overall VQoL	Mean=14.61, SD=3.9	Mean=16.89, SD=2.7 P=0.08	Mean=17.49, SD=2.1 **P<0.01**
After-school activities	Mean=5.67, SD=1.5	Mean=6.89, SD=1.1 **P<0.01**	Mean=7.24, SD=0.7 **P<0.01**
Item identification	Mean=2.50, SD=0.9	Mean=2.78, SD=0.4 P=0.25	Mean=2.94, SD=0.2 **P<0.01**

P values indicate significance level of the differences in subscales of VQoL between subgroups of Visual abnormality, Visual impairment and Normal Vision.

The preliminary associations shown in Table 4.6 between developmental age and the VQoL subscales, as measured by the HVQoLS, in children with ID were analysed further. A two-way between-group analysis of variance was conducted to explore the impact of developmental age and gender in children with ID on the "School activities" and "After-school activities" VQoL subscale. Participants were divided into two groups according to their developmental age (Young group: 3-6 years, n=85; Older group: 7-10 years, n=62). For "School activities", the interaction effect between gender and developmental age groups was not statistically significant ($F(1,141)=1.21$, $P=0.27$). There was a statistically significant main effect for developmental age ($F(1,141)=4.60$,$P<0.05$). Therefore, gender was not considered to influence the variance in scores. Similar results were also found for "After-school activities" with main effect of developmental age ($F(1,143)=12.02$, $P<0.01$).

The association between VQoL and HQoL subscale scores was explored using Pearson's correlation (Table 4.9). In children with ID, VQoL was associated with the subscale "School life" of HQoL ($r=0.21$, $P<0.01$). HQoL was associated with subscales "School activities" ($r=0.20$, $P<0.01$) and "Item identification" of VQoL ($r=0.21$, $P<0.01$). In children with ND, Overall HQoL was not associated with any of the VQoL subscales whereas Overall VQoL was associated with the HQoL subscale "School life" ($r=0.32$, $P<0.01$). All of the subscales within the HQoL were correlated with each other, as well as the subscales within the VQoL, indicating robust internal consistency of domains of HQoL and VQoL respectively.

Table 4.9 Pearson's correlation matrix[a] showing correlation coefficients between overall and subscale scores of HQoL and VQoL

		HQoL subscales			VQoL subscales		
		School life	**Family life**	**Health care**	**School activities**	**After-school activities**	**Item identification**
ID (N=259)	Overall HQoL score	0.90	0.66	0.57	0.20		0.21
	Overall VQoL score	0.21			0.95	0.82	0.49
ND (N=134)	Overall HQoL score	0.87	0.64	0.70			
	Overall VQoL score	0.32			0.93	0.83	0.43

[a]*Only correlation coefficients statistically significant (P <0.01) shown in the table*

4.2.5 Discussion

The study described in this section explored the relationship between objective measures of visual function and subjective measures of HQoL and VQoL from children's own perceptions. The impact of visual abnormality on most HQoL and VQoL subscales in children with ID was not statistically significant and does not support the hypothesis that "Visual abnormality and impairment have an adverse impact on HQoL and VQoL in children with ID". There are at least two possible explanations for this unexpected finding. First, the impact of visual abnormality in children with ID may be lower than that of their coexisting general physical or psychological impairments, thus the impact of visual abnormality is relatively insignificant when identified by the questionnaire method used here. Second, as discussed in Chapter 3, this finding may in part reflect the low sensitivity of the VQoL instrument, as indicated by the low person separation in the Rasch results (See Section 3.2.4.2.3). Thus, the findings presented here do not point toward a lack of impact of visual abnormality on VQoL in children with ID, and suggest that further refinement of the instrument is needed before this question can be addressed. In contrast, visual abnormality, categorised according to BCBV and stereopsis, was found to impact VQoL in children with ND. This may reflect the fact that the ND group included children from a low vision school, and is concordant with previous findings (Felius, et al., 2004; Gothwal, et al., 2003; Kuang, Hsu, Chou, Tsai, & Chou, 2005).

In the present study, visual functions were found to be associated with IQ score. Similarly, the study by Nielsen et. al. (2007b) concluded that visual impairment was more significant in children with lower IQ. Several studies concluded that visual abnormality, such as myopia, is strongly associated with systemic conditions (Logan, Gilmartin, Marr, Stevenson, & Ainsworth, 2004; Marr, Halliwell-Ewen, Fisher, Soler, & Ainsworth, 2001). However, none of the studies, including the present one, revealed a causal relationship between visual impairment and low IQ. On one hand, visual impairment has been found to be a barrier to children's motor skill performance (Houwen, Hartman, & Visscher, 2009; Houwen, Visscher, Hartman, & Lemmink, 2007; Houwen, Visscher, Lemmink, & Hartman, 2008), which would prevent children from

achieving normal scores in an IQ test. On the other hand, lower IQ indicates a higher risk of visual impairment due to brain damage, cerebral disorders and prematurity (L. S. Nielsen, L. Skov, et al., 2007b).Given the higher risk of visual abnormality in children with severe ID (L. S. Nielsen, L. Skov, et al., 2007a; Woodhouse, Adler, & Duignan, 2004; Woodhouse, et al., 2003), visual examinations are recommended as early as possible. For children with visual impairment, a thorough cerebral function examination is highly recommended (Gronqvist, Flodmark, Tornqvist, Edlund, & Hellstrom, 2001). Further studies on whether reduced visual function has an impact on intellectual development in children with ID are needed, as visual impairment could impede children in learning and adapting to the environment and community.

There were significant differences in the VQoL subscales among children with ND with diverse visual functions (BCBV and stereoacuity). The relationship between visual function (BCBV) and VQoL was significant in children with ND and was identified in the subscales of “School activities” and “After school activities”, which suggests that the impact of visual abnormality might be evident when children approach school-age. Apart from visual function, developmental/chronological age was also found to influence VQoL, as indicated by significant correlations with the VQoL subscales (Table 4.6). These findings suggest that it is possible that children become aware of the importance of vision as they experience the increased visual demands during this period such as reading, drawing, and other academic tasks involving higher visual skills.

In children with ND, the impact of visual abnormality on “School activities” was much lower compared with the impact on the other VQoL subscales. This could be because of the children’s familiarity with their environment. In the low vision schools in Shanghai where the questionnaire was conducted, school facilities and reading material were adapted to suit the needs of children with visual impairment. However, in after-school activities, it is more likely that the facilities are designed for children in general, and are not designed specifically for children with visual impairment. This could explain the difference in the influence of visual abnormality in children for school-related and non-school-related activities.

In children with ID, some weak associations were found between subscales of children's self-reported HQoL and VQoL. Given the satisfactory reliability of the subscales, indicated by the internal consistency, school-related activities were highlighted as an important element for children of school-age. This is not surprising, considering the higher visual demand that is required in diverse study activities. This finding also highlights the importance of timely eye care services for children prior to school age. Given the higher risk of visual abnormality in children with ID compared with children with ND, these children may become aware of the impact of their visual abnormality as they encounter challenges during school-related activities, which they might not notice in other domains.

4.3 Proxy perception of impact of visual abnormality in children with ID

Parents' opinions, as a proxy view of children's HQoL and VQoL, collected by questionnaires, are widely applied in health care outcomes research, especially for young children and for those with ID. However, previous research has indicated that there may be discrepancies between self and proxy opinions of quality of life. A poor agreement between children's and proxy ratings should not automatically be a criterion for rejection when assessing a HQoL instrument, as this difference might stem from a genuine difference in opinions between the two parties (Eiser & Morse, 2001a). Despite the obvious limitations and potential biases involved in proxy assessment, such reports provide additional information of a child's HQoL and VQoL from the parents' point of view.

4.3.1 Aims

1. To investigate the impact of visual abnormality on proxy HQoL and VQoL subscales.
2. To investigate the association, if any, between proxy and children's perceptions of HQoL and VQoL.
3. To compare proxy views of HQoL and VQoL between parents of children with and without ID.

4.3.2 Hypotheses

1. Children with visual abnormality have lower HQoL and VQoL as viewed by parents.
2. Difference exists between proxy and children's perceptions of HQoL and VQoL.
3. Proxy views of HQoL and VQoL are different between parents of children with and without ID.

4.3.3 Methods

4.3.3.1 Participants and grouping

Parents were divided into subgroups of normal vision, visual abnormality and visual impairment, based on the BCBV of their children. The recruitment and grouping of children have been described previously (see Section 2.2.7).

4.3.3.2 Questionnaire

The proxy section of the HVQoLS was given to parents of participating children at the SCMC by the investigator. Recruitment was by invitation. At the SCMC, parents completed the questionnaire while in the clinic. For the parents of the children recruited from each of the schools (SSSE, SFPS, SLVS), the questionnaires were taken home and returned to the investigator the next day. All parents were provided with an instruction letter on how to complete the questionnaire.

4.3.3.3 Subscales

The subscales of HVQoLS investigated in this section are derived from parents' perception, including "General health", "General vision", "Competence", "Personality" and "Family impact".

4.3.3.4 Data analysis

Proxy views of HQoL and VQoL were compared among subgroups of participants according to their visual function (BCBV), using at-test, an ANOVA test with a Bonferroni-adjusted post hoc test, and, where appropriate, associations of perspectives of HQoL and VQoL between children's and the proxy view were explored via a Pearson's correlation test.

4.3.4 Results

A total of 270 parents of children with ID and 127 parents of children with ND agreed to complete the proxy section of the HVQoLS. Of the children with ID, 239 completed the vision screening, thus, their parents were divided into groups including children with normal vision (n=141), visual abnormality (n=61) and visual impairment (n=37). Of the children with ND, 124 completed the vision screening and their parents were divided into groups including children with normal vision (102), visual abnormality (n=5), and visual impairment (n=17).

The proxy view of children's HQoL and VQoL was lower for parents of children with ID compared with parents of children with ND for all subscales and this difference was statistically significant for "General Health" and "Personality" (Table 4.10).

Additionally, the proxy scores on the HVQoLS subscales were compared between subgroups with and without visual impairment in children with and without ID. In children with ID, "General vision" and "Family impact" scored significantly higher in the normal visual function subgroups than those with visual abnormality and visual impairment. In children with ND, scores for "General vision" and "Competence" (Table 4.11) were higher than in children with ID. No other significant differences were found. In children with ND, "General vision" was significantly higher in the normal vision subgroup compared with the visual abnormality subgroup, and "Competence" was significantly higher in the normal vision subgroup compared with the visual impairment subgroup. No statistical significance was found in the other subgroup comparisons (Table 4.12).

Associations between the proxy HVQoLS subscales were explored (Table 4.13). Several relationships were found for parents' views of whether general health and general vision status affected both their children's life and family life. In children with ID, "General vision" was associated with children's "Personality" (r=0.20), and children's "Competence" (r=0.35) and "Personality" (0.46) were associated with "Family impact". In children with ND, children's "General health" was associated with "Personality"

(r=0.39), “General vision” was associated with “Competence” (r=0.32),and children’s “Personality” correlated highly with “Family impact” (r=0.64). In addition, “General health” and “General vision” were highly correlated in both children with and without ID (r=0.41 and 0.46).

Table 4.10 Comparison of HQoL and VQoL scores between children with and without ID from proxy view

Subscales	ND	ID
General health	Mean=3.59, SD=1.0	Mean=3.18, SD=1.0 **P<0.01**
General vision	Mean=6.62, SD=1.7	Mean=6.36, SD=1.6 P=0.27
Competence	Mean=76.69, SD=5.7	Mean=75.07, SD=6.7 P=0.16
Personality	Mean=39.99 SD=8.9	Mean=36.65, SD=7.9 **P<0.01**
Family impact	Mean=35.45, SD=4.0	Mean=35.19, SD=4.1 P=0.73

Table 4.11 Impact of visual abnormality on HQoL and VQoL from proxy views

Subgroup	Results from ANOVA on HQoL and VQoL subscales				
	General health	General vision	Competence	Personality	Family impact
ID (N=239)	F(2,236)=0.92 P=0.40	F(2,153)=6.24 **P<0.01**	F(2,70)=0.45 P=0.64	F(2,132)=2.54 P=0.08	F(2,122)=3.55 **P<0.05**
ND (N=124)	F(2,121)=0.69 P=0.50	F(2,60)=6.74 **P<0.01**	F(2,46)=18.38 **P<0.01**	F(2,63)=0.71 P=0.49	F(2,35)=2.28 P=0.12

P values indicate significance level of the differences in proxy HQoL or VQoL in subgroups of diverse BCBV, i.e. Normal vision, Visual impairment and Visual abnormality.

Table 4.12 Results from ANOVA with Bonferroni post hoc test on scores of HQoL and VQoL subscales

Subgroup	Subscale of VQoL	Normal vision	Visual abnormality	Visual impairment
ID	General vision	Mean=6.70, SD=1.4	Mean=5.92,SD=1.4 **P<0.01**	Mean=5.83, SD=2,2 **P<0.05**
	Family impact	Mean=36.24, SD=4.3	Mean=34.05, SD=4.2 **P<0.05**	Mean=34.96, SD=3.1 P=0.53
ND	General vision	Mean=6.96, SD=1.3	Mean=4.25, SD=0.5 **P<0.01**	Mean=5.83, SD=2.6 P=0.11
	Competence	Mean=78.74, SD=1.3	Mean=78.67, SD=1.5 P=1.0	Mean=71.82, SD=6.7 **P<0.01**

P values indicate significance level of the differences in subscales of proxy VQoL between subgroups of Visual abnormality, Visual impairment and Normal vision.

Table 4.13 Pearson's correlation matrix showing correlation coefficients between subscales of proxy HQoL and VQoL[a]

Subgroups	Subscales	Competence	Personality
ID (N=270)	General vision [b]		0.20
	Family impact	0.35	0.46
ND (N=127)	General health		0.39
	General vision	0.32	
	Family impact		0.64

[a]*Only correlation coefficients that are statistically significant (P <0.01) are shown in the table*
[b] *"General health" and "General vision" were highly correlated with r=0.41 and 0.46.in ID and ND subgroups*

Table 4.14 Pearson's correlation matrix showing correlation coefficients on subscales of HVQoLS between proxy and children's views

Subgroups	Subscales	VQoL[b]	School activities	After-school activities	Item identification	HQoL[c]	School life	Family life	Health care
ID (N=270)	General health	0.15*	0.14*	0.13*		0.16*			0.20**
	General vision					0.20*	0.17*		0.30**
	Competency			0.27*					0.28*
	Personality	0.24**	0.28**						
	Family impact							0.22*	0.23**
ND (N=131)	General vision								0.26*
	Competency	0.33*		0.30*	0.33*		0.45**		

[a]***. Correlation is significant at the 0.01 level (2-tailed), *. Correlation is significant at the 0.05 level (2-tailed).*
[b]*VQoL is sum scores of subscales of VQoL;* [c] *HQoL is sum scores of subscales of HQoL*

The associations between the proxy and children's perceptions of HQoL and VQoL were explored (Table 4.14). Several subscales of children's perception were mildly or moderately correlated with the subscales of proxy perception. In children with ID, meaningful associations were found between: "General health" and "Health care" (r=0.20); "General health" and "HQoL" (r=0.16); "General vision" and "Health care" (r=0.30); "Competency" and "After-school activities" (r=0.27); "Personality" and "VQoL" (r=0.24), "Personality" and "School activities" (r=0.28); "Family impact" and "Family life" (r=0.22); and "Family impact" and "Health care" (r=0.23). In children with ND, "General vision" only correlated with "Health care" (r=0.26), while "Competency" was associated with several subscales, including "VQoL" (r=0.33), "After-school activities" (r=0.30), "Item identification" (r=0.33) and "School life" (r=0.45).

4.3.5 Discussion

Parents of children with and without ID had a clear awareness of their children's visual status, as reflected in the subscale of "General vision" showing significantly higher scores in children with normal vision than those with visual abnormality and visual impairment. Also, parents were aware that visual abnormality and impairment could influence some aspects of their children's lives. For parents of children with ID, visual abnormality was considered to have a significant influence on family life. For parents of children with ND, visual impairment affected competence. This implied that for children with ND and visual impairment, parents realized that visual abnormality or impairment was a significant barrier to their children's abilities (competency) in daily activities. For children with ID, the extent to which vision affects these abilities, in the parents' view, may be concealed by other skills deficits due to general developmental delay of children with ID. The addition of a visual abnormality might not significantly influence the performance of children with ID, whilst its impact in children with ND is more apparent.

Although the mean HQoL and VQoL scores of the normal vision subgroups were higher than the scores for the visual abnormality or visual impairment subgroups, the scores did not always differentiate between “Visual abnormality” and “Visual impairment”, in terms of their impact on the subscales of children’s HQoL and VQoL. This indicates that the sensitivity of the scale was not sufficient to differentiate between subgroups of children with visual problems, or that the difference between the subgroups in their perceptions of HQoL and VQoL was insignificant. This finding questions the suitability of this scale in clinical applications, where many cases will involve a certain level of visual abnormalities. A lack of sensitivity of the VQoL scale in this population was also highlighted during the validation phase with Rasch analysis, which showed that the scale lacked person separation. Therefore, it would be desirable to include more items that focus on the range of visual abilities of the target population.

The significant correlation between proxy HQoL and VQoL scores indicated that in the parents’ view, their children’s visual function was associated with their development and maturity, as reflected in the subscales of “Competence” and “Personality”. Parents, regardless of their children’s IQ, tended to associate these aspects of their children with family life. A child with higher scores on “Competence” and “Personality” would be more likely to have a positive impact on their family. This impact might be indirectly attributed to good vision, since vision was associated with the “Competence” and “Personality” of children. However, more direct evidence of the relationship between these factors was not examined in this study. Further investigation into how visual abnormality influences children’s development and their family life, especially in those with ID is warranted.

In addition, the results in this study showed some agreement between children’s subjective self-assessment and their parents’ proxy opinions of HQoL and VQoL. In children with ID, even though different items were included in the proxy and

self-assessment of HQoL and VQoL, children's perceptions were associated with the proxy opinion of health care and family impact. In children with ND, parents' views of their children's competence were associated with children's self-perceptions of their school life and other vision-related, after-school activities. These associations between children's perception of HQoL and VQoL and proxy views support the use of proxy views as additional, but not replacement, sources of information of children's HQoL and VQoL.

4.4 Conclusion

These findings demonstrate a link between perceptions of HQoL and VQoL by children and their parents, and objective findings by clinical examination. Such a questionnaire can be applied in future in combination with other vision screening programs. The results indicated that different perceptions of HQoL and VQoL existed between children with and without ID, and their parents. While HQoL is relatively low in children with ID, visual abnormalities that commonly occur in children with ID do not impact their HQoL and VQoL in the children's view, but do have an impact in the parents' view. This finding may reflect different perceptions between children and their parents of the extent of impact of visual abnormality on HQoL and VQoL. Together, these findings suggest that a more suitable VQoL scale is still needed for children with ID, especially for those with mild to moderate visual abnormality.

CHAPTER 5 OVERVIEW AND FUTURE WORK

5.1 Overview

The work described in this thesis is the first to investigate HQoL and VQoL and associations between these subjective perceptions and objective measures of visual function in children with ID. Given the lack of available tools to assess HQoL and VQoL in children with ID, this thesis describes the development of suitable instruments for use in this population.

This chapter revisits the research questions (RQs) introduced in Chapter 1 and shows how each of these was addressed. In particular, the development of the instrument for assessment of HQoL and VQoL and its generalizability are discussed, as are the results of application of the instrument presented in Chapter 4. The theoretical, methodological, clinical and societal contributions of the research are outlined, and limitations of this present research explained. Finally, suggestions for future research directions are proposed.

5.2 Conclusions related to the research questions

5.2.1 Measurement of HQoL and VQoL in children with ID (Research Question 1, RQ1)

The process of instrument development described in this study could provide a model for future instrument development by researchers and eyecare practitioners who are involved in service delivery to children with ID. The pilot study demonstrated the validity and reliability of the modified instruments, through conventional methods of questionnaire development. Further refinements using modern questionnaire development methods reinforced the robust psychometric properties of the HQoL instruments and revealed further modifications that could be made to improve VQoL instruments for use in children with ID. Following a validation process, the modified HVQoLS was found to have acceptable levels of construct and content validity. This

instrument was well comprehended by children with mild and moderate ID and can be used as an outcomes assessment tool for health and eyecare practitioners working with children with ID at these levels of severity. However, given limited communication and reading comprehension capacity, the questionnaire developed here is not applicable in children with severe ID or autistic spectrum disorders (See Section 2.2.5.2). For these subgroups, perhaps clinical and daily life observation would be more beneficial to better understand their QoL, and this question should be the subject of further research.

5.2.2 Difference in self-perceived HQoL and VQoL between children with and without ID (RQ2)

Using the HVQoLS, this study showed that children with ID had a measureable awareness of their HQoL and VQoL that differed substantially from the awareness of children without ID. This finding was particularly relevant, given that identical items were included in the instruments used in both groups of children. In addition, this study explored the differences in self-perceived and proxy measures of HQoL and VQoL in children with and without ID.

5.2.3 Difference in proxy-perceived HQoL and VQoL between children with and without ID (RQ3)

Visual abnormality was identified by proxy views from parents of children with ID as a contributor to HQoL and VQoL, but this was not the case in the views from the children with ID themselves. This contradiction confirmed the different views between children and parents towards children's QoL and indicates that both self-perceived and proxy views of QoL should be taken into consideration when assessing QoL in children with ID.

5.2.4 Impact of visual abnormality on HQoL and VQoL in children with and without ID (RQ4)

Moreover, the significantly higher risk of visual abnormality in children with ID compared with those without ID has demonstrated the importance of vision screening and optimal visual correction for children with ID. The findings presented here suggest that in spite of other potential consequences of ID, prompt correction of visual abnormalities, most commonly refractive error, should be a focus for primary eye care practitioners, especially for those involved in the health care of children with ID.

5.3 Contributions of the study

Overall, the process of instrument development in this study provides a suitable procedure for instrument selection, modification and administration in children with ID. In particular, this study highlights the need for extra awareness in order to acquire reliable responses from children with ID, and the importance of a feasible instrument format and method of questionnaire administration that is designed specifically for children with special needs.

5.3.1 Theoretical contribution

Research into HQoL and VQoL in children with ID is still at an early stage and its application in clinical assessment has not been addressed in the literature. Existing instruments for measuring HQoL either do not focus on vision-related problems, or are not specific for children with ID, and thus, are inappropriate for this application. This research has attempted to add to the base of knowledge in several ways by clarifying and expanding existing knowledge. The study firstly developed an instrument to be used in the children with ID and provided evidence (such as rational ranking of QoL related activities, clear patterns (domains) of the HQoL and VQoL perceptions; Sections 3.1.4.2.3 and 3.2.4.2.3) that children with ID have a sense of both VQoL and HQoL which is accessible using appropriate tools.

5.3.2 Methodological contribution

A major strength of this study was that instrument development was conducted in stages. First, the prototype questions were informed by an advisory group including parents, teachers and health care providers of children with ID. The instrument format was refined by pilot work in children with and without ID and their parents. Second, development of the instrument in this study included conventional and more recent validation techniques, which strengthened the psychometric properties of the instrument. Rasch analysis was used as part of the questionnaire validation procedure and the psychometric properties of the instruments were described by parameters from the model index. This approach identified limitations in the psychometric properties of the instrument and indicated where further modifications were required. Any redundant items were identified and excluded, and categories were collapsed where appropriate. This approach has some advantages over conventional questionnaire validation methods of correlations and factor analysis (see section 1.5.3). Following this process, the targeting of the instrument towards the study population was enhanced, so that the items included in the instrument were matched better with the capability of the participants. In addition, as one of the advantages of Rasch analysis, the raw data from the questionnaire were converted into an interval scale, which strengthened the statistical application of these data in multivariate analyses. Through these adjustments, the results provided a meaningful interpretation of HQoL and VQoL in children with ID. Consequently, the construct validity and internal consistency reliability of the instrument were considerably improved and the instrument was considered ready for use in the population of children with ID.

5.3.3 Clinical contribution

Using the questionnaire developed here, the link between clinical measures of visual function and subjective perception of HQoL and VQoL was explored. It is important to probe this issue in children with ID to establish the impact of visual abnormality on perceptions of HQoL and VQoL from children and their parents, particularly in view

of the very high incidence of ocular and visual abnormalities in this population. Given the limited comprehension and communication capacity of these children, their vision care needs are sometimes neglected or concealed by other physical conditions. The perspectives of HQoL and VQoL held by children with ID and their parents are described in detail in this thesis. These perspectives and the questionnaire itself may prove informative and useful for eye and health care providers working with children with ID.

5.3.4 Societal contribution

It has been noted that in recent years there has been a paradigm shift in attitudes and policies relating to people with ID. This shift incorporates a move from the statutory provision of care to a commitment by the community to provide support for opportunities to work, which requires a closer relationship between people with ID and others in the community (Section 1.1.3). Obviously, people with ID are confronted with obstacles when they contribute to society. Skills set, ability to socialise and education levels are factors that may impose limits on the scope for community engagement in any population. In particular, the high prevalence of visual impairment in the population of people with ID may impose additional disability to those already with impaired daily functioning. Therefore, it is imperative to explore the health status, quality of life and social involvement of individuals with ID, which links to advancement of the wellbeing of this population. The findings of the present study provide some insight into the perspectives of children with ID, the extent to which quality of life can be measured based on such perspectives, and the extent to which visual abnormality may impact on quality of life in those individuals. The findings also illustrate the high incidence of visual abnormality in this population, and the need for correction of treatable abnormalities as a part of enhancing life skills for people with ID (Section 4.1.4.2).

5.4 Limitations of the study

The limitations to the study methodology include the generation of items and the use of a convenience sample. Prototype items were not originally generated from advisory panels, but were adapted from existing instruments (AUQUEI, LVP-FVQ and CVFQ). Given that two of these original instruments were designed for children with visual impairment or severe ocular pathology, it is unsurprising that many items were relatively easy for the participants in the present study and were excluded in the validation process. Exclusion of these items from the VQoL section of the finalised HVQoLS instrument resulted in a lower person separation compared with the original instruments. For the same reason, the VQoL of most children with higher visual capability could not be thoroughly investigated due to a lack of appropriate questions for children with mild visual abnormalities. Thus in its present form, the instrument would not be applicable in assessments of the impact of changes in visual function on quality of life in children with ID, limiting its future application.

5.5 Suggestions for future research directions

The instrument developed in this study may provide a useful measure of subjective visual function alongside clinical measures of visual function with additional items describing VQoL in children with near normal vision.

For future research, it will be important to explore whether the outcomes of interventions to correct visual abnormalities can be measured in both objective visual function and in subjective measures of HQoL and VQoL in children. This is particularly important in view of the high prevalence of ocular and visual abnormalities in children with ID, and the need to understand the extent to which treatment of those abnormalities benefits the child. However, as noted above, the questionnaire developed here required further refinement to allow this type of assessment.

The present research was conducted in China, and it may be that the impact of visual abnormality on quality of life in children with ID depends, to some extent, on cultural factors. The perception of HQoL and VQoL could possibly vary across countries. Further study with adequate sample sizes from populations of interest will help to understand the effects of ethnicity, economic development and culture on subjective perceptions and the impact of other health interventions. Based upon these preliminary results, the construct and item content should be studied more thoroughly prior to further application in alternative populations.

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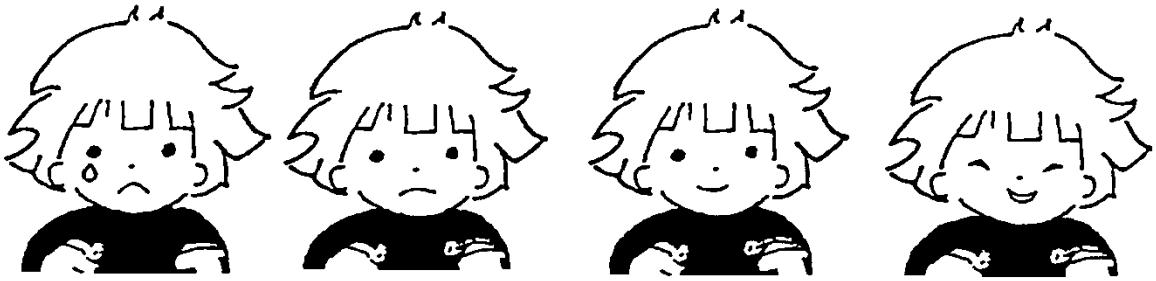
Appendix 1 Original instruments

Child self questionnaire (AUQUEI)

Items 1 and 2 differ in format slightly to the rest. In Item 1, children are asked to answer 'Why?' to each option. In Item 2, they are given a frequency rating scale of feeling this way, from 1 (never) to 4 (very often). Items 4 - 29 all follow the same style as Item 3. The four faces represent feeling towards the prompt on the top left. Children are required to respond by filling in the circle below the representative picture.

CHILD SELF REPORT FORM

Sometimes you are…

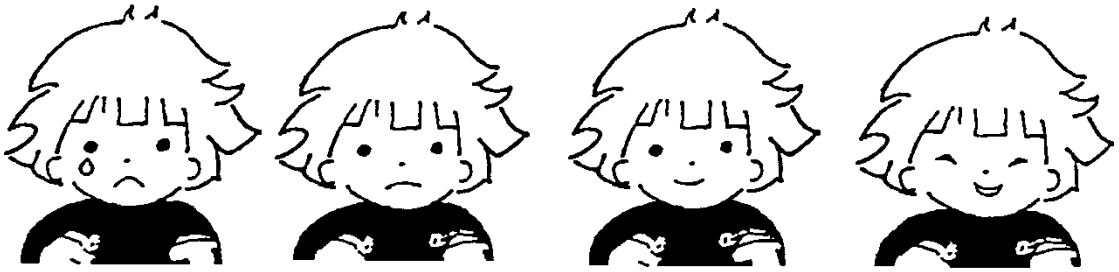


Not happy at all Say why...

Unhappy Say why...

Happy Say why...

Very happy Say why...



How often are you…

Not happy at all

Never ☐
Occasionally ☐☐
Often ☐☐☐
Very often ☐☐☐☐

Unhappy

Never ☐
Occasionally ☐☐
Often ☐☐☐
Very often ☐☐☐☐

Happy

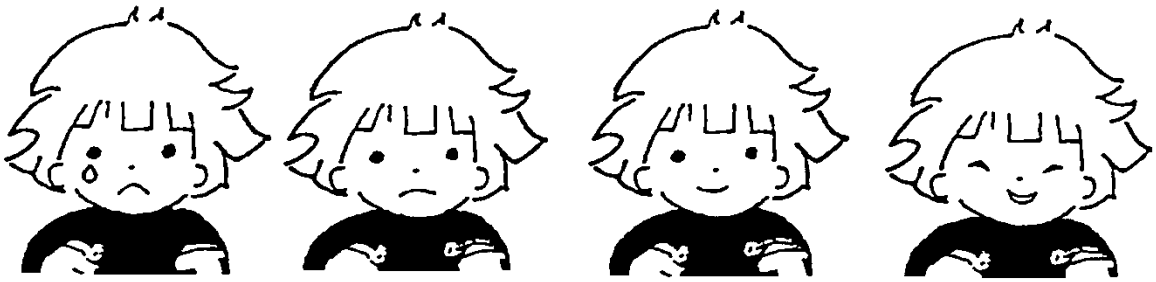
Never ☐
Occasionally ☐☐
Often ☐☐☐
Very often ☐☐☐☐

Very happy

Never ☐
Occasionally ☐☐
Often ☐☐☐
Very often ☐☐☐☐

*Colour in the boxes that match your reply

3. How do you feel when you're having dinner with your family ?



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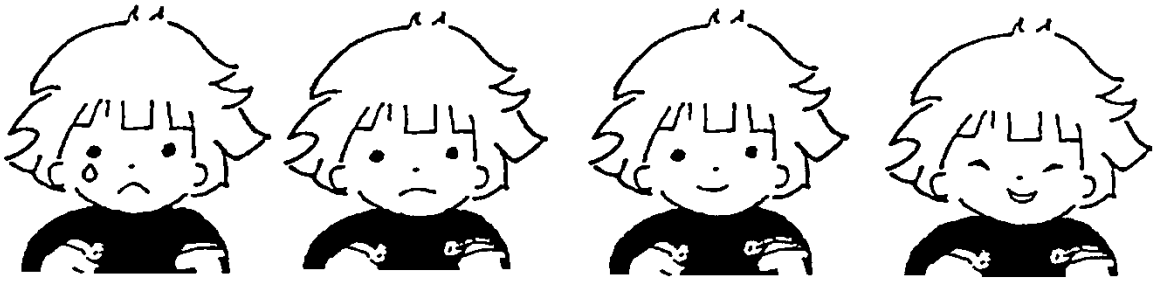
4. How do you feel when you go to bed at night ?



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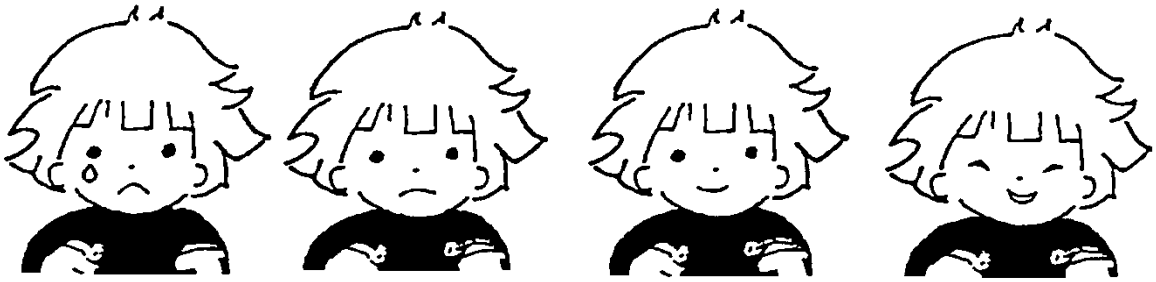
5. If you have brothers and sisters, how do you feel when you play with them ?



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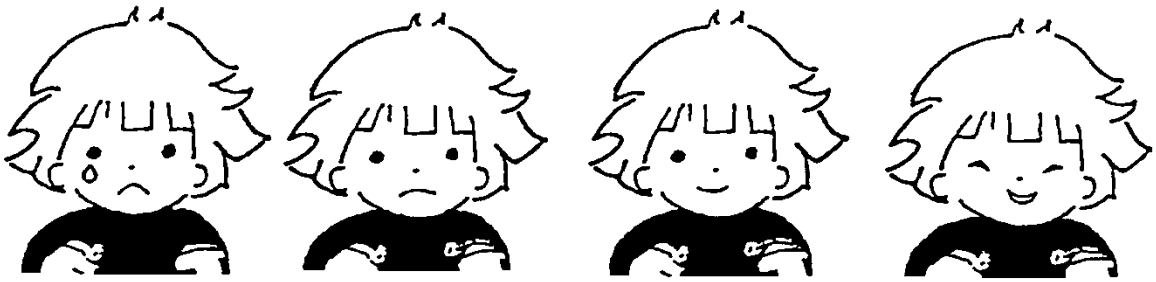
6. How do you feel when you're asleep at night ?



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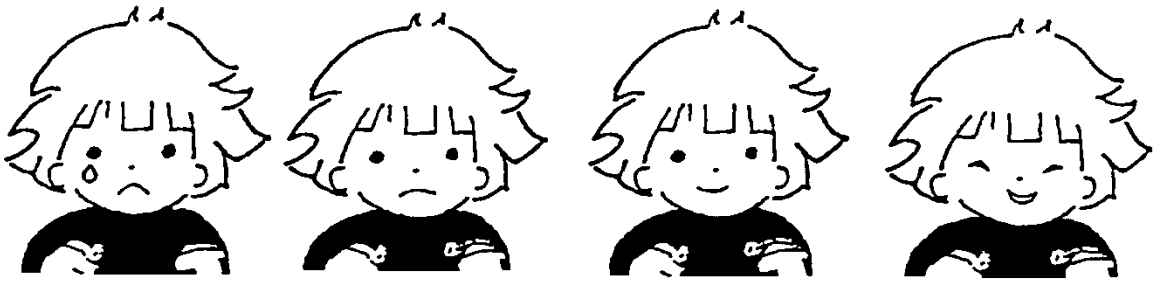
7. How do you feel when you are at school ?



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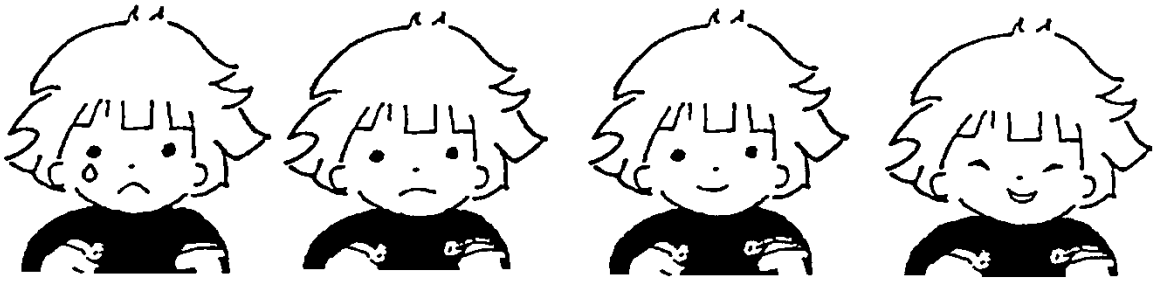
8. How do you feel when you watch a picture of yourself ?



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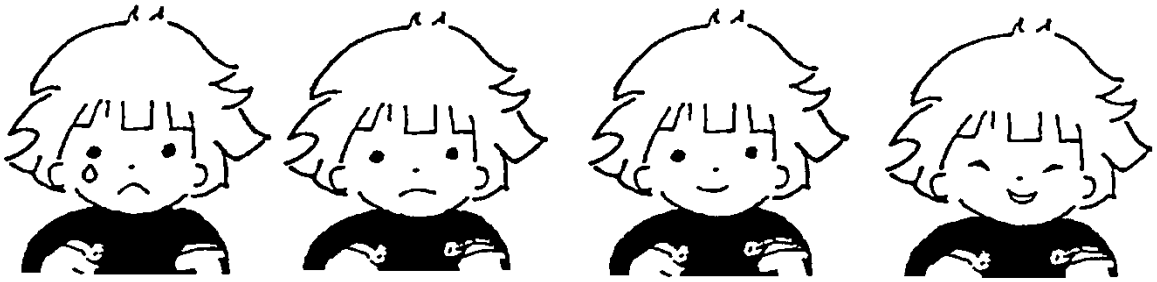
9. How do you feel when you go to the doctor's ?



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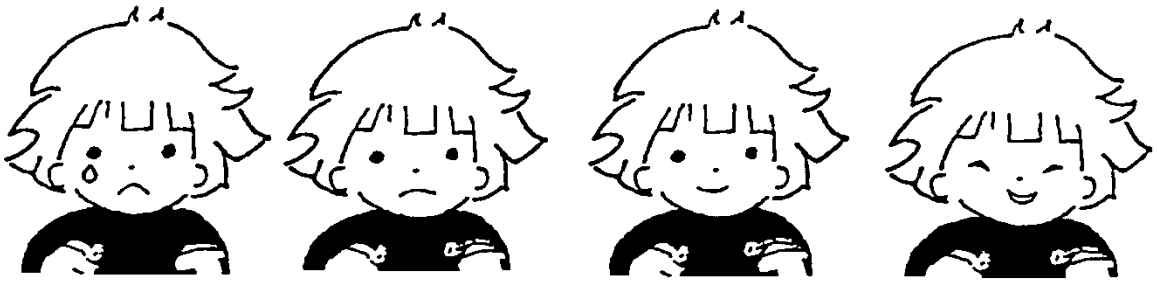
10. How do you feel when you think about your father ?



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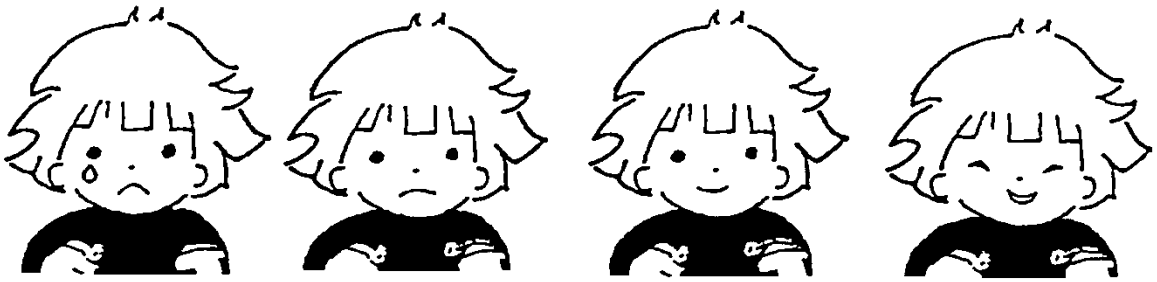
11. How do you feel on your birthday ?



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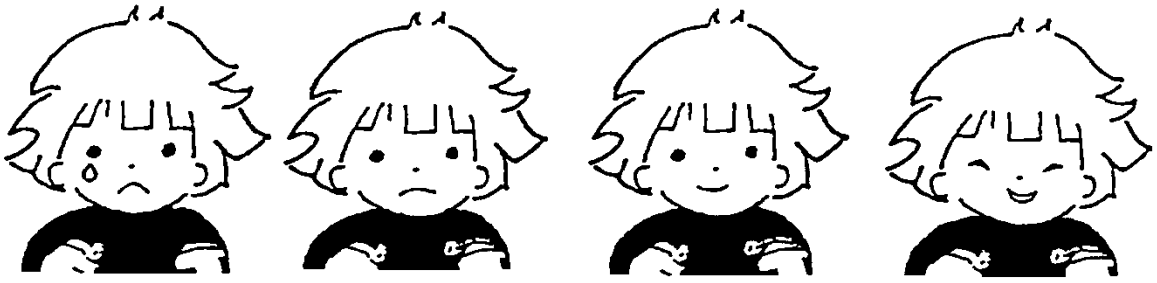
12. How do you feel when you think about your mother ?



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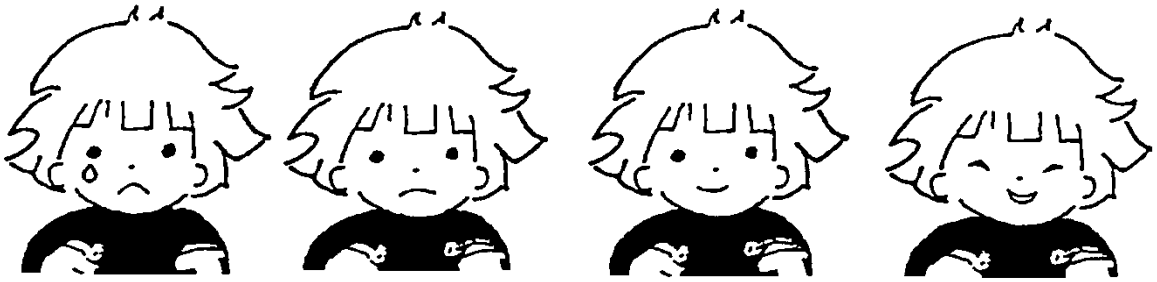
13. How do you feel when you stay in hospital ?



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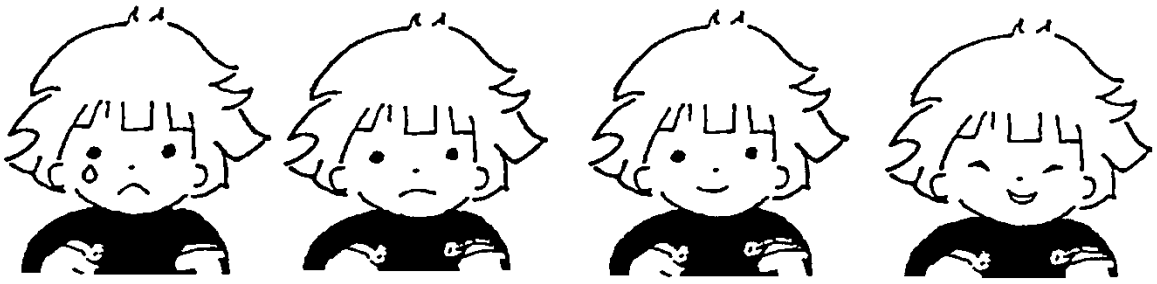
14. How do you feel when you play alone ?



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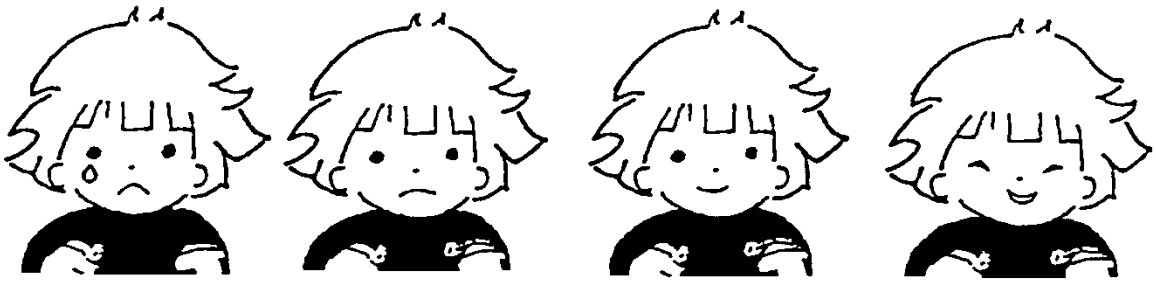
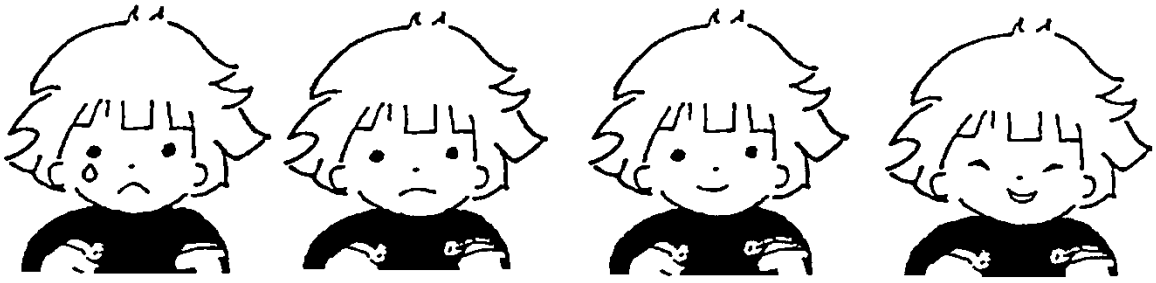
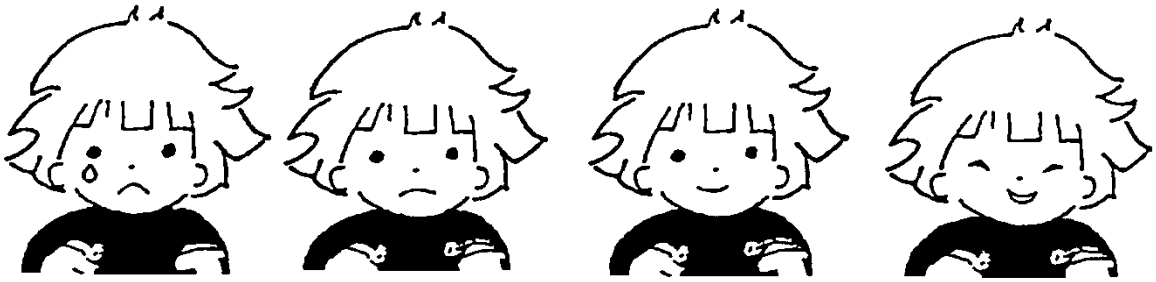
15. How do you feel when your parents are talking about you ?



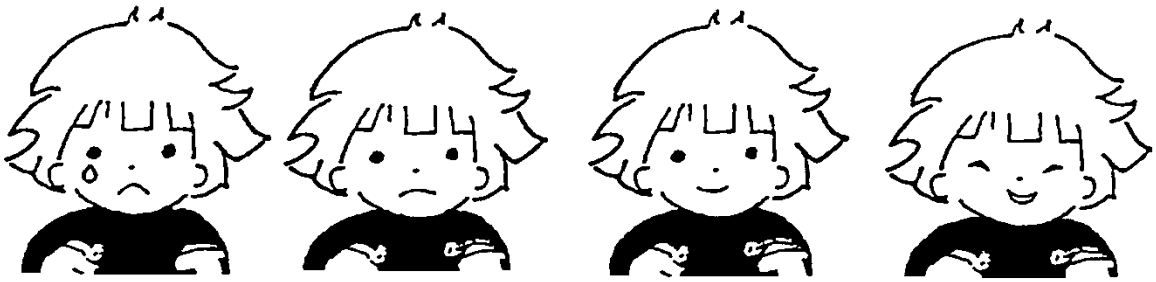
16. How do you feel when you spend the night away from home ?



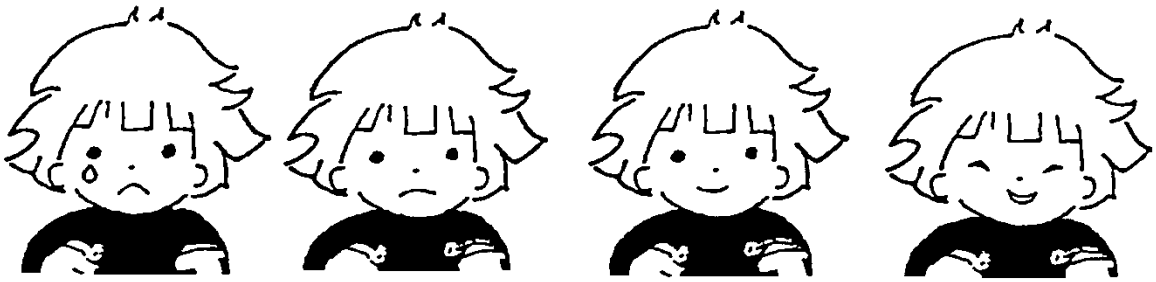
17. How do you feel when people ask you how to show what you're able to do ?



18. How do you feel when your friends are talking about you ?



19. How do you feel when you take a medicine ?



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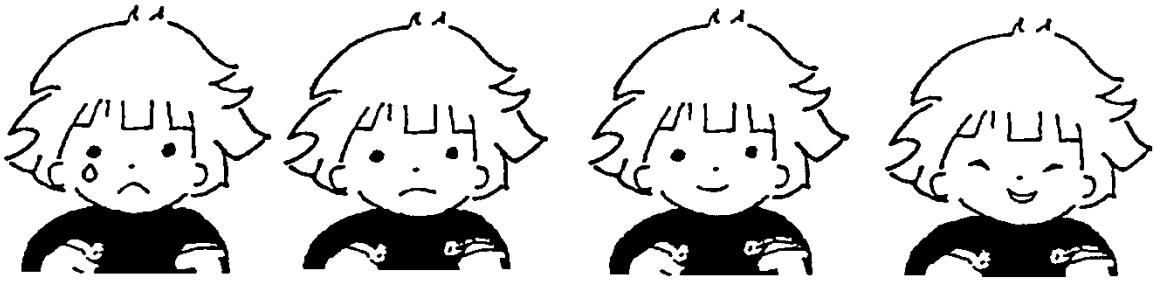
20. How do you feel during the holidays ?



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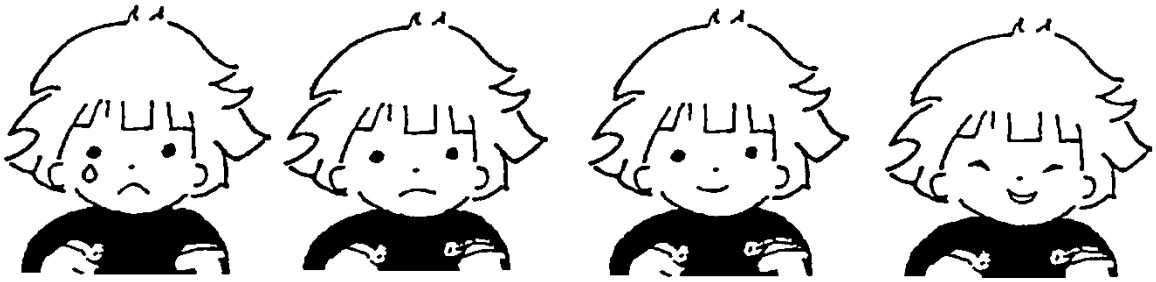
21. How do you feel when you make a drawing ?



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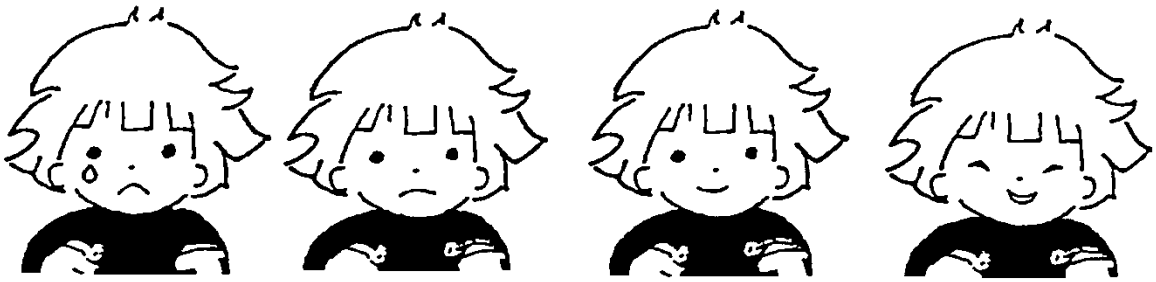
22. How do you feel when you see yourself as a grown-up ?



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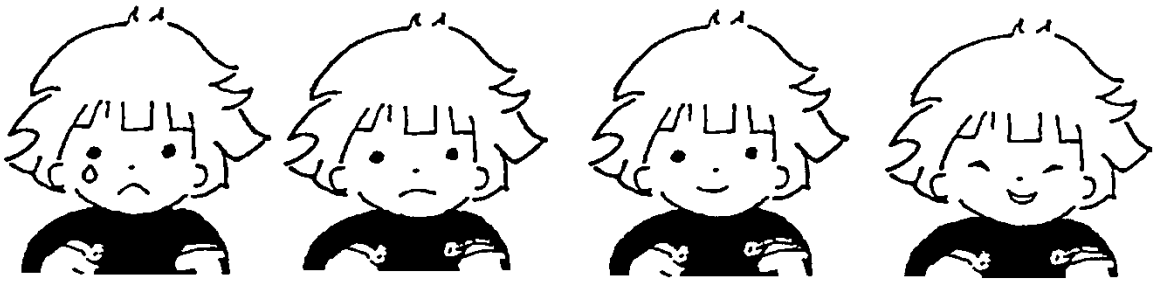
23. How do you feel when you are with your grand-parents ?



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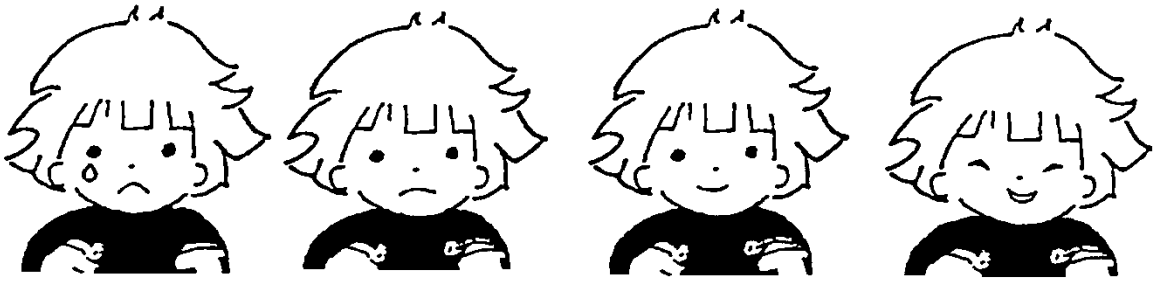
24. How do you feel when you watch television ?



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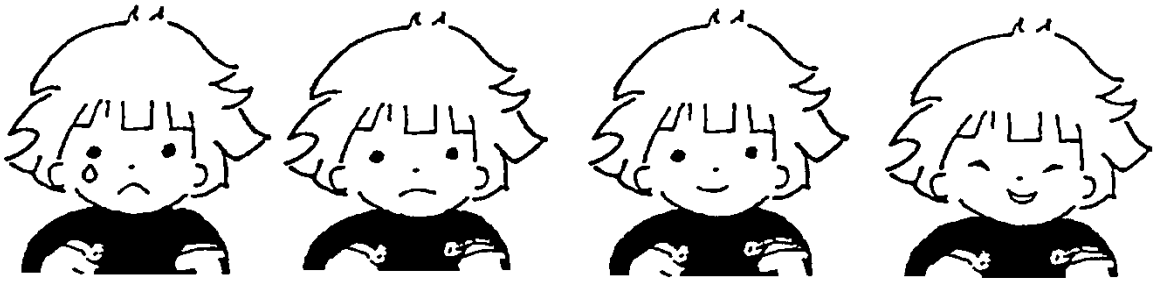
25. How do you feel when you move (walk, run, jump)?



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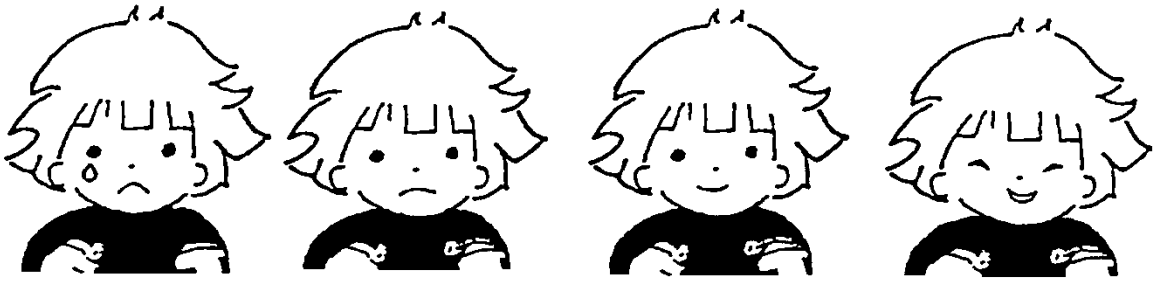
26. How do you feel when you are eating?



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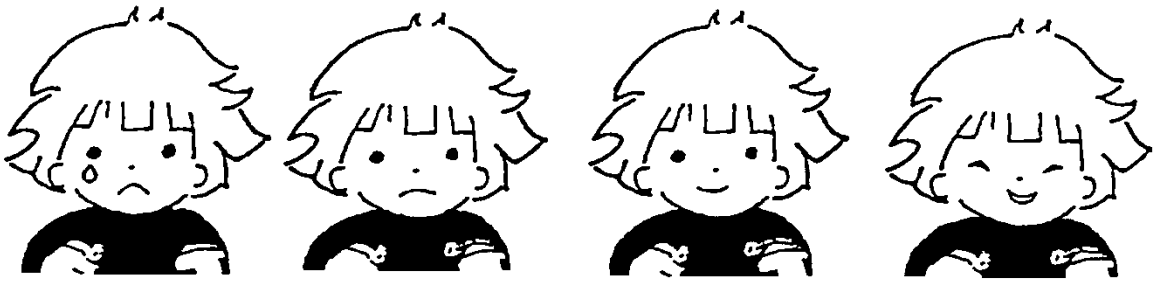
27. Some days you are well, some days you are sick, how do you feel when you think about your health?



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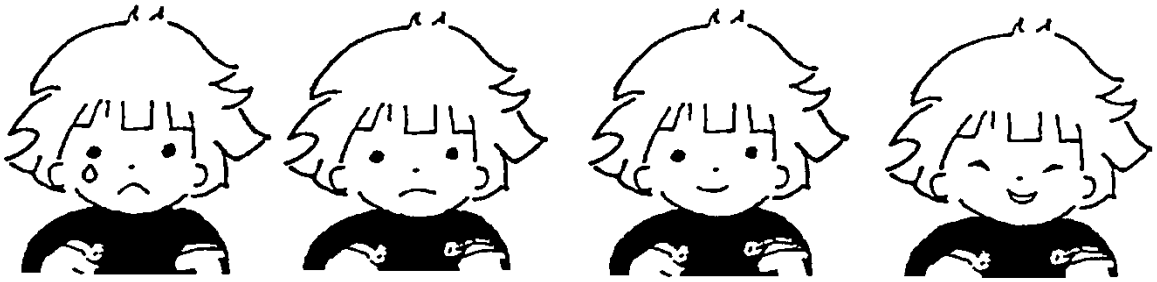
28. How do you feel when people tell you what to do?



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THANK YOU FOR COMPLETING THIS QUESTIONNAIRE!!!

LV Prasad-Functional Vision Questionnaire (LVP-FVQ)

1. Do you have any difficulty in making out whether the person you are seeing across the road is a boy or a girl, during the day?

____Yes ____No ____Not applicable

If yes, how much difficulty do you have?

● Little
● A moderate amount
● A great deal
● Unable to do the activity
(Note: the same response options were used for questions 1–19.)

2. Do you have any difficulty in seeing whether somebody is calling you by waving his or her hand from across the road?
3. Do you have difficulty in walking alone in the corridor at school without bumping into objects or people?
4. Do you have any difficulty in walking home at night (from tuition or a friend's house) without assistance when there are streetlights?
5. Do you have any difficulty in copying from the blackboard while sitting on the first bench in your class?
6. Do you have difficulty in reading the bus numbers?
7. Do you have any difficulty in reading the other details on the bus (such as its destination?)
8. Do you have any difficulty in reading your textbooks at an arm's length?
9. Do you have any difficulty in writing along a straight line?
10. Do you have any difficulty in finding the next line while reading when you take a break and then resume reading?
11. Do you have any difficulty in locating dropped objects (pen, pencil, eraser) within the classroom?
12. Do you have any difficulty in threading a needle?

13. How much difficulty do you have in distinguishing between 1 rupee and 2 rupee coins (without touching)?
14. Do you have difficulty in climbing up or down stairs?
15. Do you have difficulty in lacing your shoes?
16. Do have difficulty in locating a ball while playing in the daylight?
17. Do you have difficulty in applying paste on your toothbrush?
18. Do you have difficulty in locating food on your plate while eating?
19. Do you difficulty in identifying colours (e.g., while colouring)?
20. How do you think your vision is compared with that of your normal-sighted friend? Do you think your vision is:

- As good as your friend's
- A little bit worse than your friend's
- Much worse than your friend's

Children's Visual Function Questionnaire

•The CVFQ is a set of 2 instruments (one targeted to children under 3 years of age, one to children aged 3 to 7 years) developed to measure the impact of visual impairment on the quality of life of young children and their families. Its intended use is primarily for research purposes and may include the assessment of efficacy of different therapeutic approaches for eye disorders in infancy and early childhood and of methods of early visual stimulation and rehabilitation.

•The CVFQ is a public document available without charge to all researchers provided that they identify the measure as such in all publications. Users should also cite the following article:

Felius J, Stager DR Sr, Berry PM, Fawcett SL, Stager DR Jr, Salomão SR, Berezovsky A, Birch EE. Development of an instrument to assess vision-related quality of life in young children. American Journal of Ophthalmology, 2004; 138(3): 362-372.

•A note of caution: Although the 2 instruments (for the 2 age groups) are different, they show considerable overlap, which may lead to confusion. The footer on each page identifies the version to which it belongs.

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PROTOTYPE INSTRUMENT:

The following is the full list of items belonging to the prototype instrument used in phase 1. Indicated in brackets behind each item is the type of ordinal scale used for the response options: [Q] Quality scale, [F] Frequency scale, [A] Agreement scale, [D] Difficulty scale. For some items an extra response option was included: [NA] "Does not apply to my child," [TY] "My child is too young to attempt this." The questionnaire items were preceded by a cover page with instructions and a statement of the purpose of this research, and followed by a series of demographic questions (available as supplemented content on the Internet at AJO.com).

1. In general, would you say that your child's overall health is: [Q]
2. At the present time, would you say that your child's eyesight using both eyes is: [Q]
3. If your child has an eyesight problem for only one eye, would you say that your child's eyesight in the affected eye is: [Q] [NA]
4. How much of the time do you worry about your child's eyesight? [F]
5. How much time do you need to spend on treatment for your child's vision problem (eye doctor appointments, patching, eye drops, therapy)? [F]
6. Does the time you spend on your child's vision problem (eye doctor appointments, patching, eye drops, therapy) take away from time you would like to spend with your other children or husband/wife? [F]
7. Do you and other family members (your spouse or parents) argue about the medical care your child is getting or about treatment that the doctor has prescribed? [F]
8. I am afraid that my child will never have good vision. [A] [NA]
9. I am bothered by other people's comments about my child's vision or eyes when I take him/her to a store or mall. [A] [NA]
10. My child likes to try new things. [A] [NA]
11. Taking my child to the eye doctor is stressful. [A] [NA]
12. I think that my child's vision will improve. [A] [NA]
13. My child feels different from other children. [A] [NA]
14. My child is happy most of the time. [A] [NA]
15. I notice other children looking at my child. [A] [NA]
16. My child likes to visit with relatives. [A] [NA]
17. My child is teased because of his/her vision problems. [A] [NA]
18. My child cries a lot. [A] [NA]

19. I worry that my child may not be able to read, watch TV, or drive a car. [A] [NA]

20. My child makes new friends easily. [A] [NA]

21. My child is affectionate. [A] [NA]

22. My child gets along well with our other children and friends. [A] [NA]

23. My child gets angry or frustrated because of his vision problem. [A] [NA]

24. We stay at home a lot because of my child's vision problem. [A] [NA]

25. My child can feed himself/herself. [D] [TY]

26. My child plays with toys. [D] [TY]

27. My child can recognise faces (friends, relatives) across a room. [D] [TY]

28. My child can imitate others (make a face, stick tongue out, play peek-a-boo). [D] [TY]

29. My child can dress himself/herself. [D] [TY]

30. My child can brush his/her teeth. [D] [TY]

31. My child can wash his/her face. [D] [TY]

32. My child adjusts to changes in lighting (going out into bright sunlight or entering a dark room or theater.) [D] [TY]

33. My child can ride a bicycle. [D] [TY]

34. My child can play a sport or active game (for example, tag). [D] [TY]

35. My child will track a mobile or a moving toy. [D] [TY]

36. My child can locate a small piece of food (a raisin or Cheerio) and grasp it. [D] [TY]

37. My child can pour liquid into a cup or glass. [D] [TY]

38. My child can dial a telephone. [D] [TY]

39. My child helps with chores. [D] [TY]

40. My child can tell what time it is. [D] [TY]

41. My child can identify coins. [D] [TY]

42. My child enjoys looking at books. [A] [TY]

43. My child is interested in playing with our pet(s). [A] [TY]

44. My child has a regular sleep routine. [A] [TY]

45. My child's eyesight makes it difficult for him/her to learn to walk, run, skip, or jump. [A] [TY]

46. My child's vision gets in the way of his/her learning. [A] [TY]

47. My child's eyesight has made it difficult for him/her to learn to read. [A] [TY]

48. My child enjoys watching television, videos, or playing video games. [A] [TY]

49. My child likes to travel on family vacations. [A] [TY]

50. My child enjoys playing with others (sisters and brothers or friends). [A] [TY]

51. My child enjoys drawing, painting or other art activities. [A] [TY]

52. My child's eyesight makes it difficult for him/her to find something on a crowded shelf or in a closet. [A] [TY]

53. My child makes eye contact with me and smiles. [F] [TY]

54. My child bumps into people, walls, or furniture. [F] [TY]

55. My child trips over curbs or steps. [F] [TY]

56. My child bumps into other people. [F] [TY]

57. I have trouble applying treatment (for example, putting on an eye patch or glasses, giving eye drops or other medication). [F]

58. My child is uncomfortable when treated (for example, while wearing a patch or glasses or when you put in eye drops). [F]

59. My child is less active when treated (for example, when wearing a patch or glasses, or when taking eye drops or medication). [F]

60. I worry when my child refuses treatment (for example, pulls off the patch or glasses, or squeezes eye shut when trying to put in eye drops). [F]

61. I sometimes forget to treat my child. [F]

Appendix 2 Adapted instruments in pilot study

Instructions for Completion of the Enclosed Questionnaires

There are four different questionnaires and one child's information sheet enclosed in this envelope. Two of the questionnaires are designed for testing children's quality of life, to be completed with the help of parents or caregivers whenever necessary, and the others to be completed by the child's parent or main caregiver. The child information sheet is designed for the statistical analysis for the study. A set of instructions for completion of each questionnaire is given below. The child may need the help of a parent or caregiver to complete these questionnaires. The questionnaires may be completed at the child's own pace. **The parent/caregiver should explain the meaning of each question if necessary, but should avoid guiding the child's response.** It is possible that some items of this questionnaire are beyond the knowledge or experience of the child. Please omit questions that cannot be answered for this reason, and leave a cross mark beside the question.

Instruction for Children's Quality of Life Questionnaire

This questionnaire aims to access children's quality of life. The four faces with different expressions are intended to illustrate a range of feelings. The child is required to respond by pointing to the representative picture that matches his/her feeling, following the interpretation of each question by the parent/caregiver if necessary. The adult tester circles the answer (one response only) respectively.

Instruction for Vision Impact Questionnaire

This questionnaire is also for the child, by pointing to a "yes" or "no" cartoon, in response to each question (illustrated by a smiley face or X cross, respectively). The adult tester circles the answer (one response only) respectively.

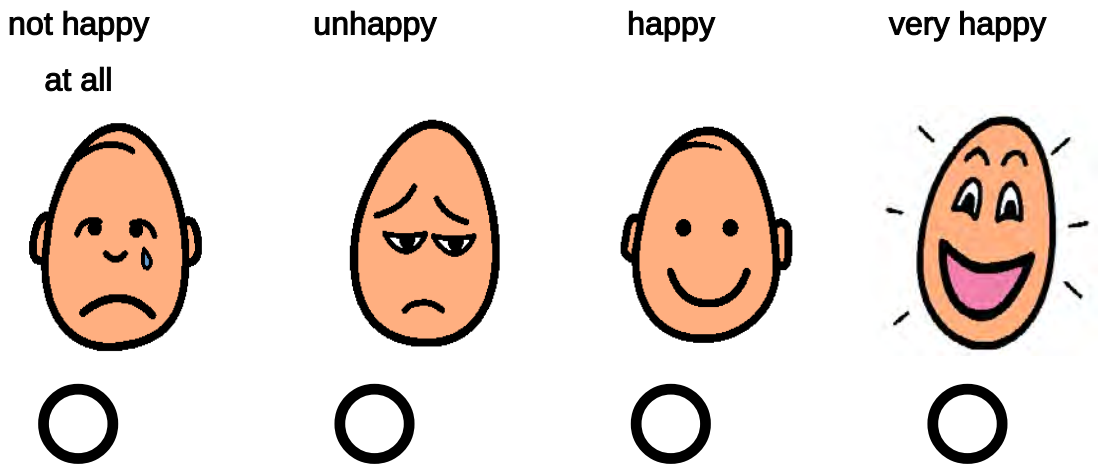
Instruction for Children's Visual Function Questionnaire for Parent/Caregiver

This questionnaire is for completion by the parent/caregiver. The questions are about the respondent's opinions on the child's vision. In response to each question, please tick one option.

Children's Quality of Life Questionnaire

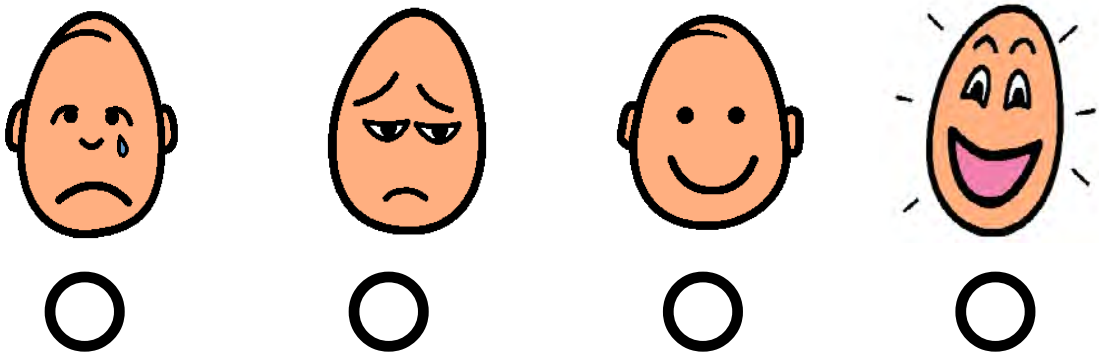
The four faces show feelings. Children are asked to point to the picture showing how they feel, on each question. An adult should read out the questions to the child and fill in the circle to indicate the child's response. Before reading any questions, the adult should show the face pictures (below) and explain the procedure to the child.

*Pointing to the face that matches your reply



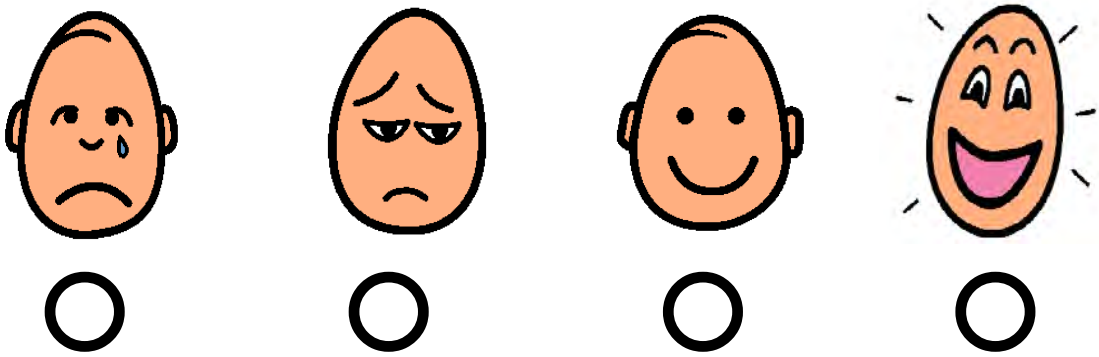
1. When I am having dinner with my family, I feel

not happy at all | unhappy | happy | very happy



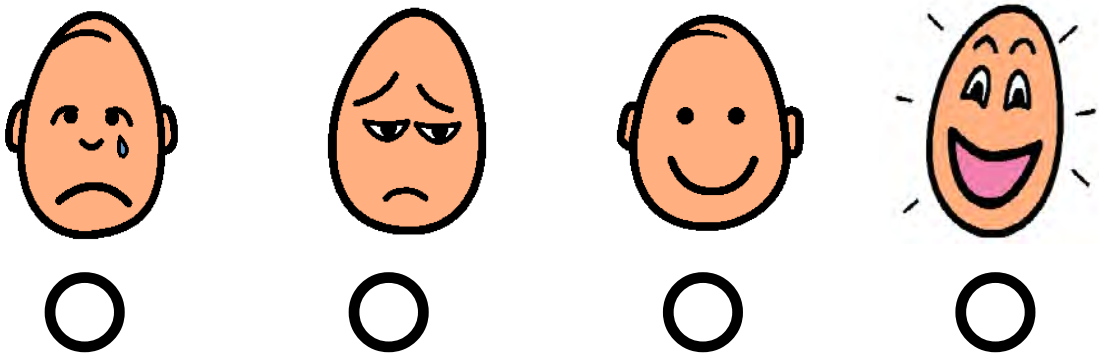
2. When I go to bed at night, I feel

not happy at all | unhappy | happy | very happy



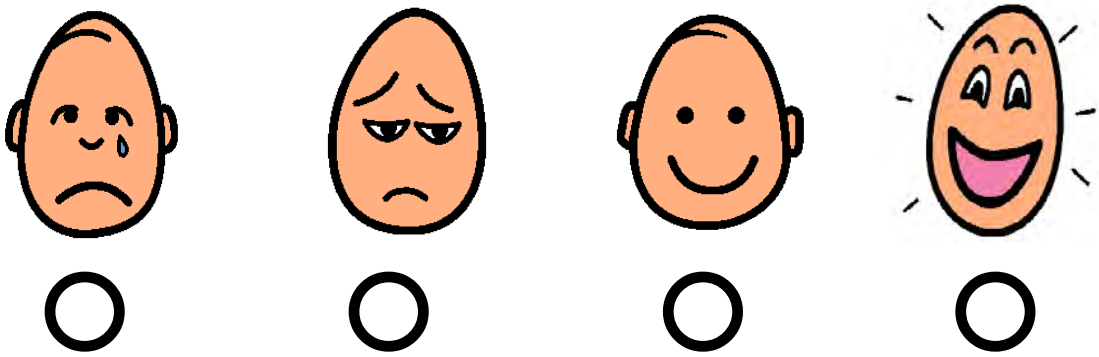
3. My brothers and sisters make me feel

not happy at all | unhappy | happy | very happy



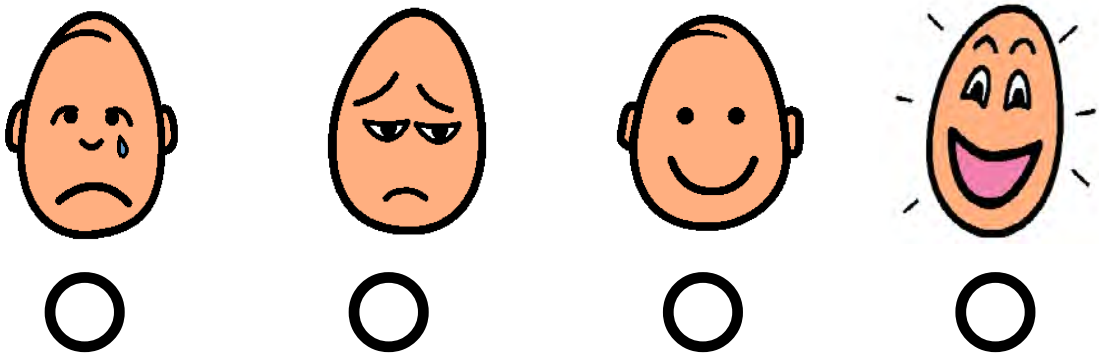
4. At school, I feel

not happy at all | unhappy | happy | very happy



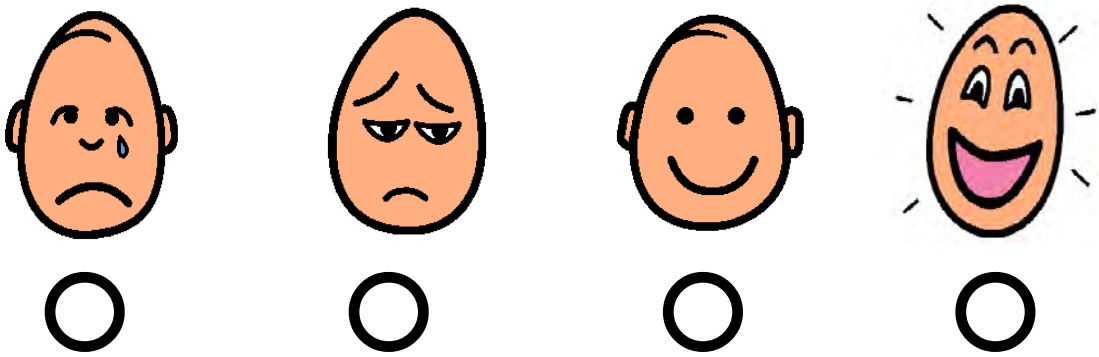
5. When I go to the doctor's, I feel

not happy at all | unhappy | happy | very happy



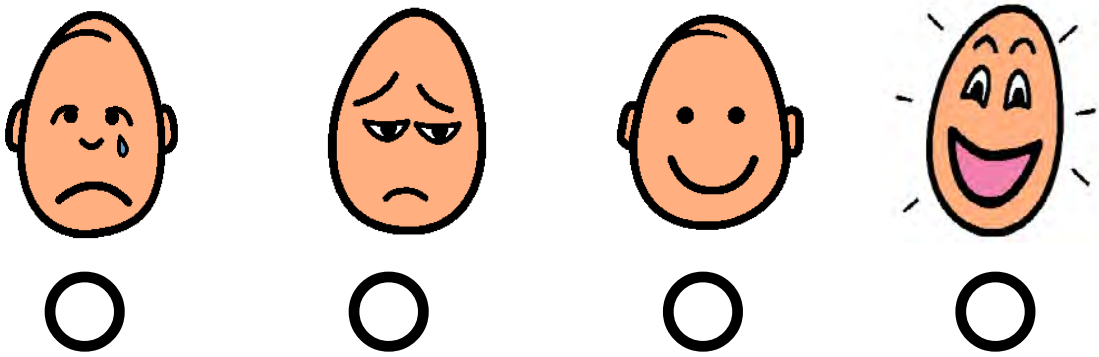
6. On my birthday, I feel

not happy at all | unhappy | happy | very happy



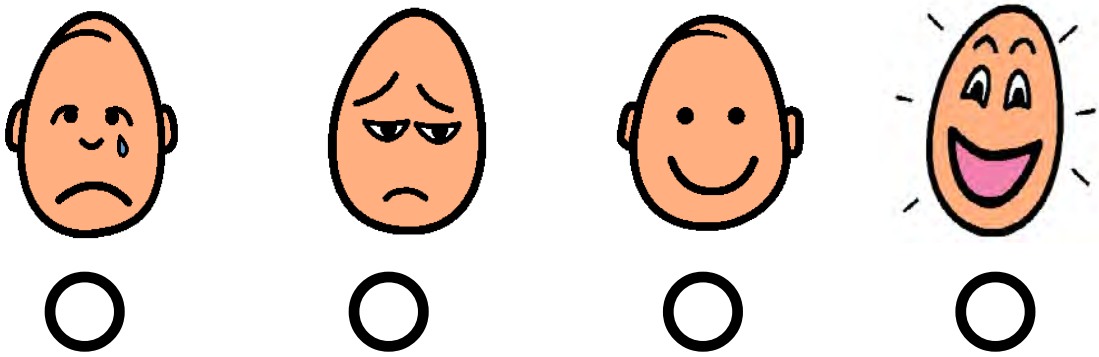
7. When I stay in hospital, I feel

not happy at all | unhappy | happy | very happy



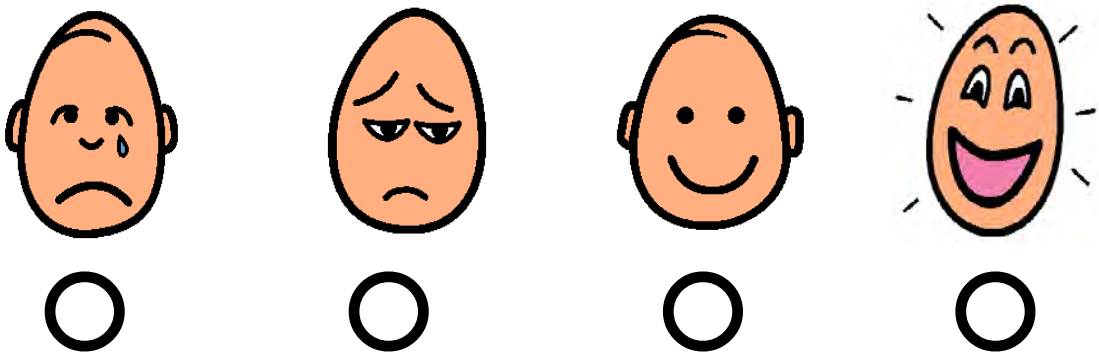
8. When I play alone, I feel

not happy at all | unhappy | happy | very happy



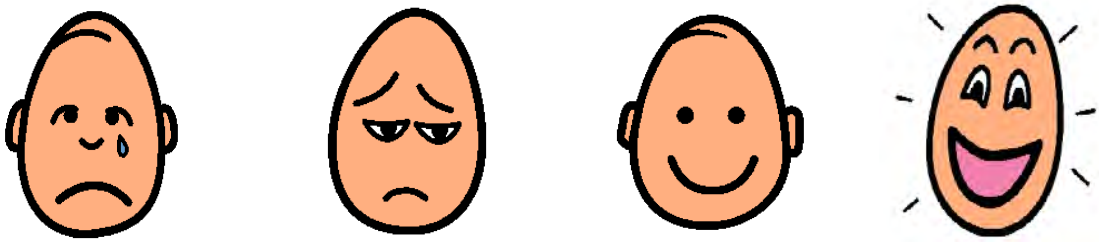
9. When I take medicine, I feel

not happy at all | unhappy | happy | very happy



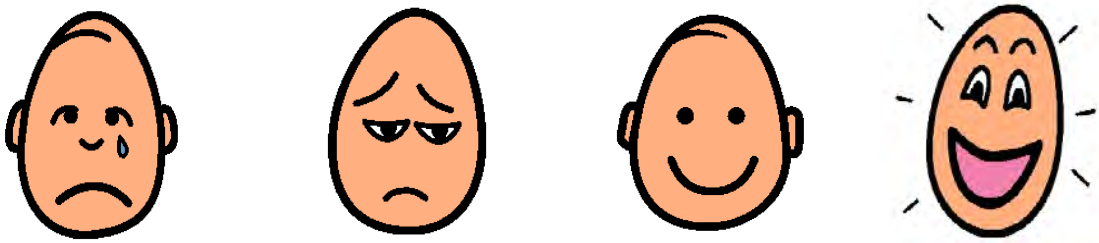
10.When I am on holiday, I feel

not happy at all | unhappy | happy | very happy



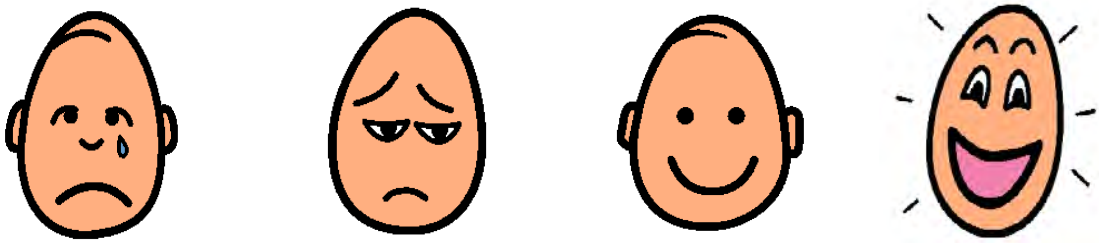
11. When I make a drawing, I feel

not happy at all | unhappy | happy | very happy



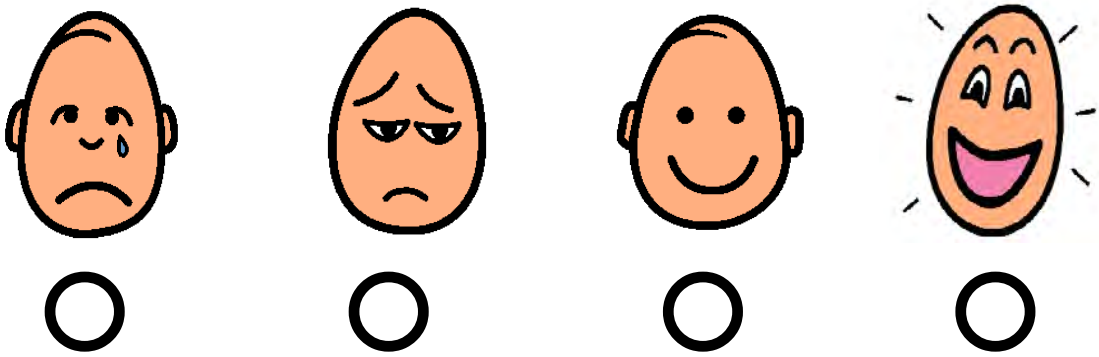
12. When I am with my grandparents, I feel

not happy at all | unhappy | happy | very happy



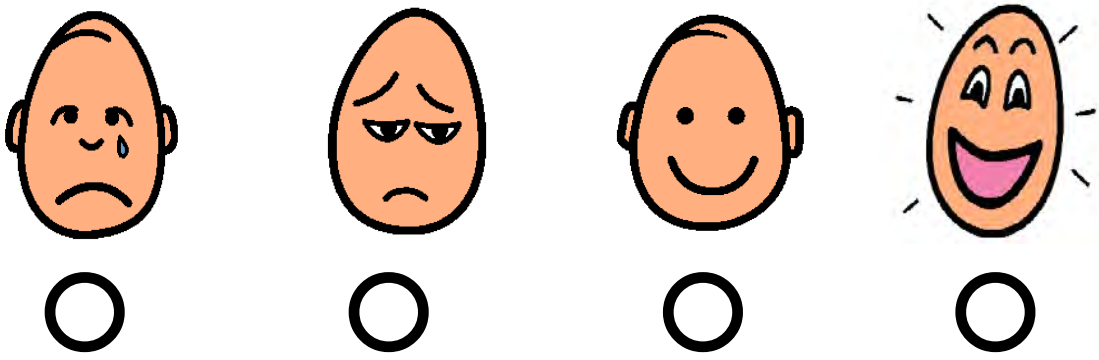
13. When I watch television, I feel

not happy at all | unhappy | happy | very happy



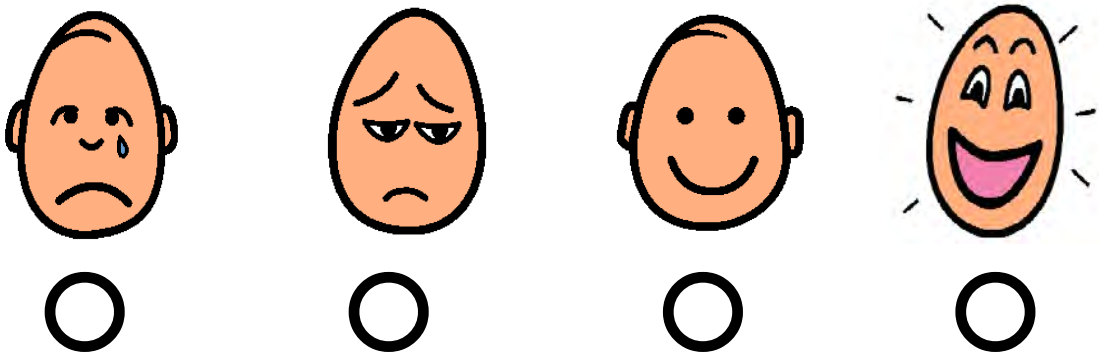
14. When I move (walk, run, jump), I feel

not happy at all | unhappy | happy | very happy



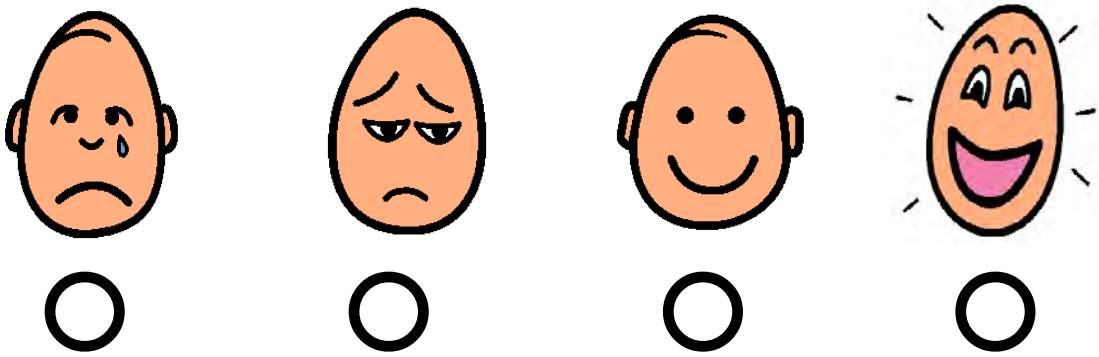
15. When I am eating, I feel

not happy at all | unhappy | happy | very happy



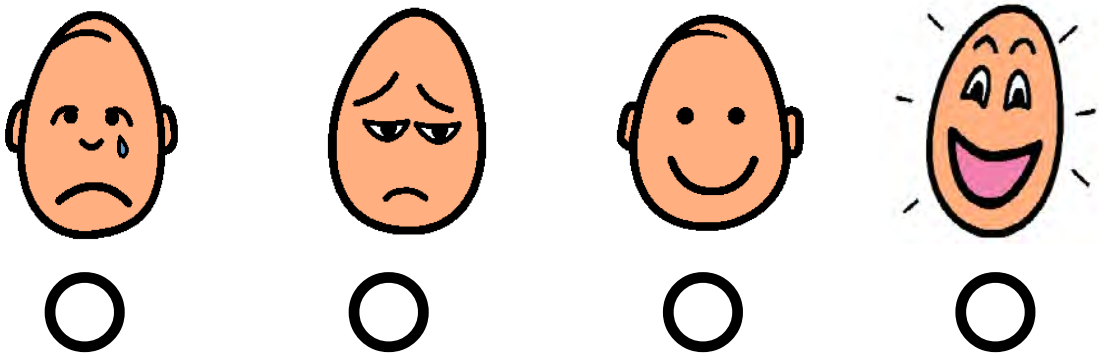
16. When I am sick, I feel

not happy at all | unhappy | happy | very happy



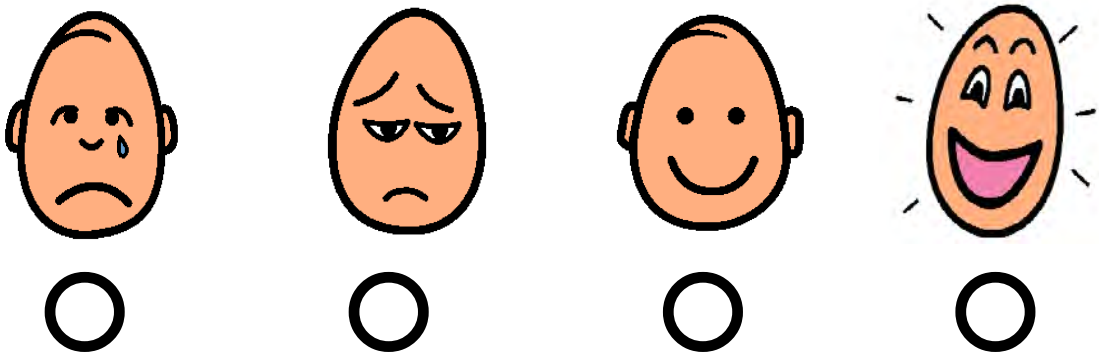
17. When I am not sick, I feel

not happy at all | unhappy | happy | very happy



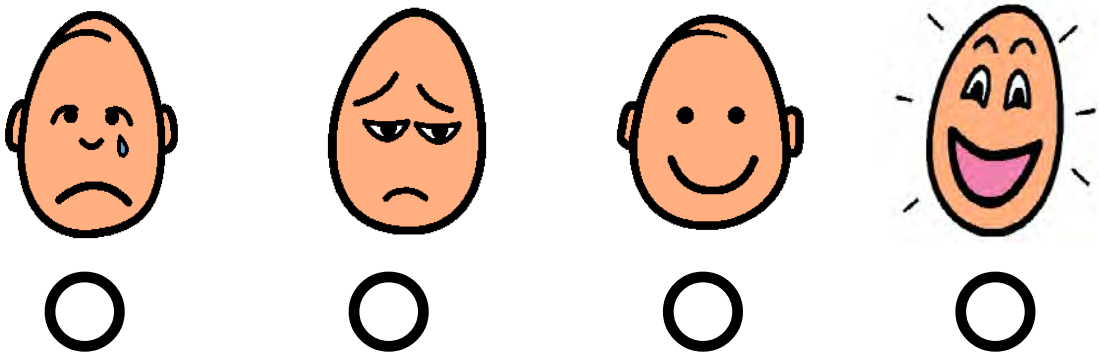
18. When people tell me what to do, I feel

not happy at all | unhappy | happy | very happy



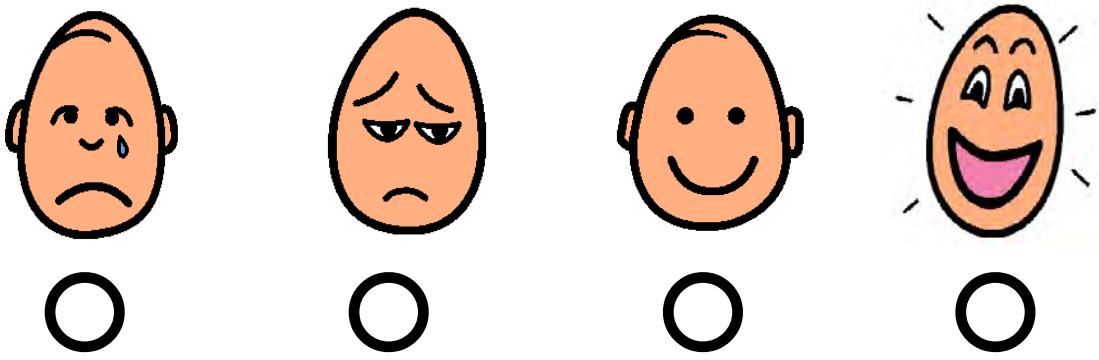
19. When I am playing outside, I feel

not happy at all | unhappy | happy | very happy



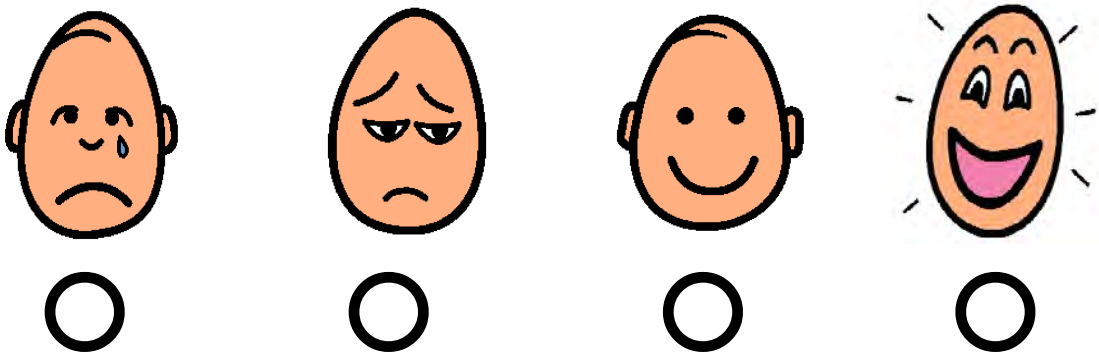
20. When I play a sport, I feel

not happy at all | unhappy | happy | very happy

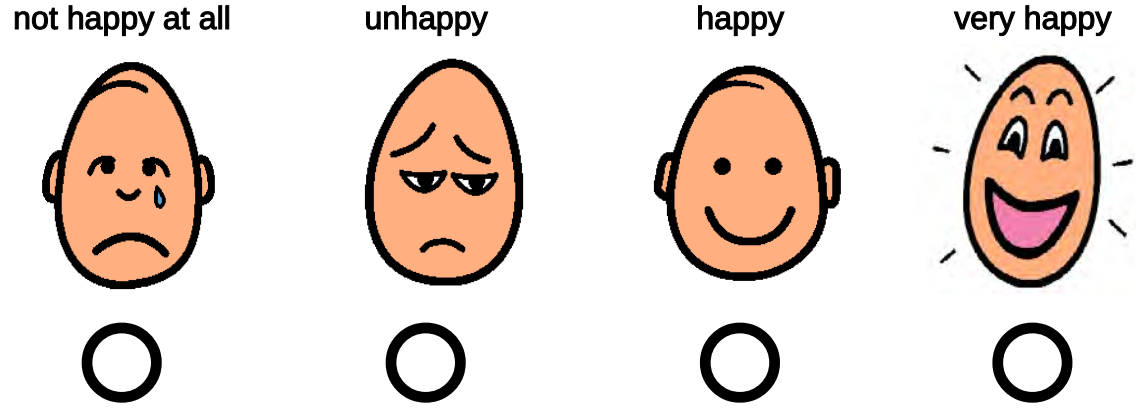


21. When I do my homework, I feel

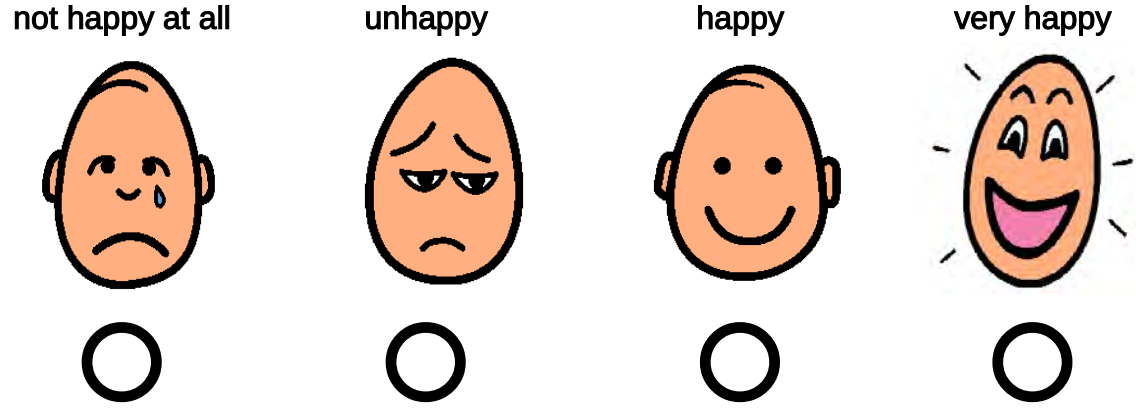
not happy at all | unhappy | happy | very happy



22. When I read a book, I feel



23. When my work is marked at school, I feel



THANK YOU FOR COMPLETING THIS QUESTIONNAIRE!!!

Vision related Quality of life Questionnaire

An adult should read the following questions to the child. The child should be asked to respond by pointing to the representative picture. The happy face on yellow indicates 'Yes' and the sad face on red indicates 'No'. The adult should then circle either 'Yes' or 'No' as appropriate.

1. Can you climb up and down stairs ?

YES  NO 

2. Do you bump into things ?

YES  NO 

3. Can you kick a ball when you play ?

YES  NO 

4. Can you hit the ball when you play ?

YES  NO 

5. Can you find food on your plate when eating ?

YES  NO 

6. Can you find your favourite toy at play time ?

YES  NO 

7. Can you pick up a red pencil from a box of pencils?

YES  NO 

8. Can you put toothpaste on your toothbrush by yourself?

YES  NO 

9. Can you do (tie) up your shoelaces by yourself?

YES  NO 

10. Can you see the TV clearly?

YES  NO 

11. Can you see a person's face across the road ?

YES  NO 

12. Can you see bus numbers clearly ?

YES  NO 

13. Can you see the pictures in your books clearly?

YES  NO 

14. Can you read a book by yourself?

YES  NO 

15. Can you draw a straight line on paper without a ruler ?

YES  NO 

16. Can you copy from the board in class?

YES  NO 

17. Can you write the numbers from 1 to 10?

YES  NO 

18. Can you write your name ?

YES  NO 

19. Can you remember words easily?

YES  NO 

20. Can you finish your homework on time ?

YES  NO 

21. Can you do maths ?

YES  NO 

22. Is your handwriting neat?

YES  NO 

THANK YOU FOR DOING THIS !!!

Children's visual function questionnaire

Instruction to the respondents

Take the time to read each question carefully.

- It is important that you answer ALL questions.
- Try to give only ONE answer to each question.
- If your child has GLASSES or has to wear a PATCH, try to think of a typical situation or an average day for your child when you answer the questions

This questionnaire is adapted from CVFQ v.3 2004 @ J. Felius & E. Birch, Retina Foundation of the Southwest

Please answer these questions about your child's health and vision:

1. In general, is your child's overall health:

❒ Excellent ❒ Very Good ❒Good ❒Fair ❒Poor

2. At the present time, is your child's eyesight when using both eyes:

❒Excellent ❒Good ❒Fair ❒Poor ❒Very Poor ❒Blind

3. If your child has an eyesight problem for only one eye, is your child's eyesight in the affected eye:

❒Excellent ❒Good ❒Fair ❒Poor ❒Very Poor ❒Blind
❒Does not apply to my child

4. Do you worry about your child's eyesight?

❒Never ❒Once in a while ❒Sometimes ❒Often ❒Always

5. How much time do you spend on caring for your child's vision (such as: eye doctor appointments, patching, eye drops, therapy)?

❒Once a month or less (or never) ❒Once a week ❒Once a day
❒ A few hours each day ❒Most of the day

6. Does the time you spend on your child's vision (eye doctor appointments, patching, eye drops, therapy) take away from time you would like to spend with your other children or husband/wife?

❒Never ❒Once in a while ❒Sometimes ❒Often ❒Always

7. Do you and other family members (your spouse or parents) argue about the medical care your child is getting or about treatment that the doctor has prescribed?

❒Never ❒❒Once in a while ❒❒Sometimes ❒❒Often ❒❒Always

We would like to know how you feel about your child's vision.

Please indicate how much you agree with each of the following statements.

	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree	Does not apply to my child
8. It bothers me when other people comment about my child's vision or eyes when I take him/her to a store or mall.						
9. My child feels different from other children.						
10. My child is happy most of the time.						
11. I notice other children looking at my child.						
12. My child is teased because of his/her vision problems.						
13. My child makes new friends easily.						
14. My child is affectionate.						
15. My child gets along well with our other children and friends.						
16. My child likes to visit with relatives.						
17. I worry that my child may not be able to read, watch TV.						

How does your child's eyesight affect his/her activities?

Please indicate how much difficulty your child has with the following activities because of his/her vision condition.

	No difficulty because of eyesight	A little difficult because of eyesight	Moderate difficulty because of eyesight	Extreme difficulty because of eyesight	Cannot do this at all because of eyesight	My child is unable to attempt this
18. My child can recognise faces (friends, relatives) across a room.						
19. My child can dress himself/herself.						
20. My child can brush his/her teeth.						
21. My child can wash his/her face.						
22. My child can ride a bicycle.						
23. My child can play a sport or active game (for example, tag).						
24. My child can pour liquid into a cup or glass.						
25. My child can dial a telephone.						
26. My child helps with chores.						
27. My child can tell what time it is.						
28. My child can identify coins.						
29. My child can locate a small piece of food (a raisin) and grasp it.						
30. My child can feed himself/herself.						

How does your child's eyesight affect his/her activities? (Continued.)

Please indicate how much you agree with each of the following statements.

	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree	Does not apply to my child
31. My child enjoys looking at books.						
32. My child's eyesight makes it difficult for him /her to learn to walk, run, skip, or jump.						
33. My child's vision gets in the way of his/her learning.						
34. My child enjoys watching television, videos, or playing video games.						
35. My child likes to travel on family vacations.						
36. My child enjoys playing with others (sisters and brothers or friends).						
37. My child enjoys drawing, painting or other art activities.						
38. My child's eyesight makes it difficult for him /her to find something on a crowded shelf or in a closet.						
39. My child bumps into people, walls, or furniture.						

Please indicate how often this happens:

	Never	Once in a While	Some-times	Often	Always	My child is too young to attempt this
40. My child trips over curbs or steps.						

Questions about the treatment of your child's eye condition.

41. Is your child currently being treated for his/her eye condition? (Treatment includes eyeglasses, contact lenses, intraocular lenses, patching, eye drops, or other treatment). Please circle one:

YES / NO

If your answer to question 35 was **NO**, it is the end of this questionnaire.
If your answer to question 35 was **YES**, please answer the **following questions**:

	Never	Once in a While	Sometimes	Often	Always
42. I have trouble applying treatment (for example, putting on an eye patch or glasses, giving eye drops or other medication).					
43. My child is uncomfortable when treated (for example, while wearing a patch or glasses or when you put in eye drops).					
44. My child is less active when treated (for example, when wearing a patch or glasses, or when taking eye drops or medication).					
45. I worry when my child refuses treatment (for example, pulls off the patch or glasses, or squeezes eye shut when trying to put in eye drops).					
46. I sometimes forget to treat my child.					

Sample questionnaires in Chinese

儿童视觉相关生活质量调查问卷

(改编自 LVP-FVQ)

被调查儿童要求在回答每个问题时，用手指出哪个脸谱所代表的感觉最符合他们的答案。黄色的快乐脸谱表示“能”，红色的沮丧脸谱表示“不能”。成人调查者读出每个问题，并圈出被调查儿童给出的答案。

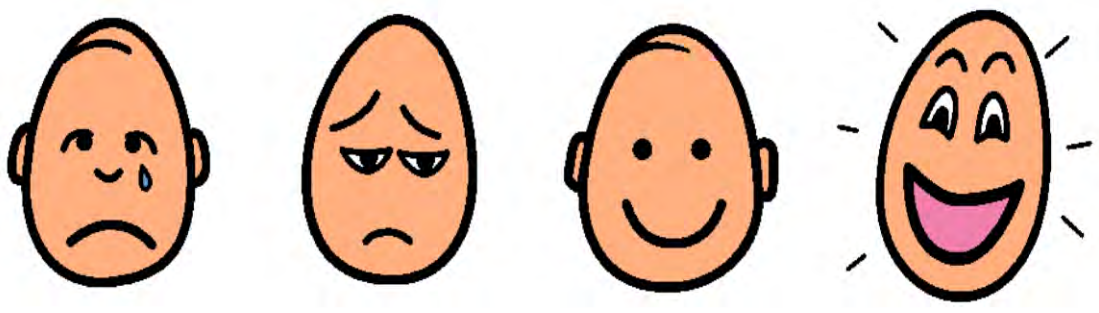
1. 你能自己上下楼梯吗？

能  不能 

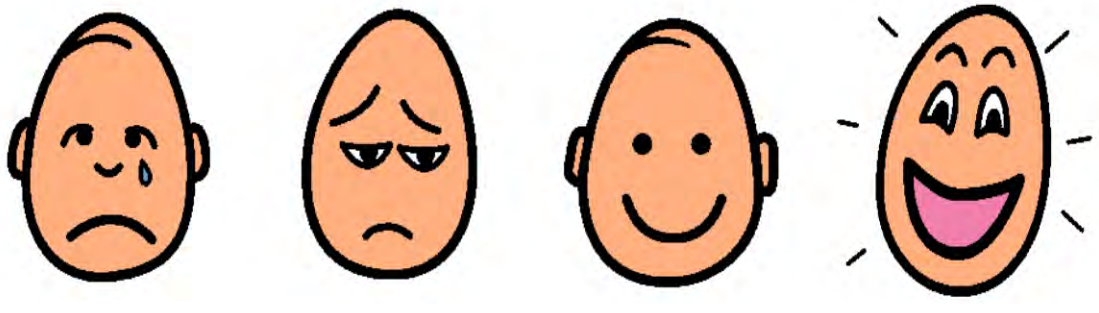
2. 你走路时会撞到东西吗？

会  不会 

1. 当我在家吃饭时，我觉得：



2. 当我晚上睡觉时，我觉得：



Appendix 3 Supplemental Rasch analysis results on validation of the AUQUEI, LVP-FVQ and CVFQ

1) AUQUEI (before item and scale reduction)

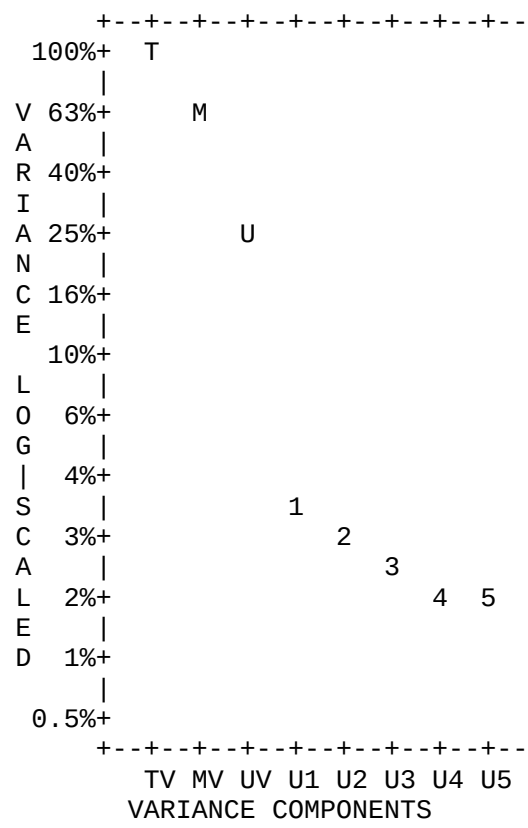
```
   SUMMARY OF 167 MEASURED (NON-EXTREME) PERSONS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-------------------------------------------------------------------------------|
| MEAN      70.4      22.7        1.30     .34      1.10    -.1   1.05    -.2 |
| S.D.       8.7       1.2         .99     .10       .66    2.0    .74    1.8 |
| MAX.      91.0      23.0        5.38    1.04      3.44    5.3   5.44    5.2 |
| MIN.      27.0       9.0        -.57     .29       .20   -4.5    .25   -3.9 |
|-------------------------------------------------------------------------------|
| REAL RMSE    .43  ADJ.SD     .89  SEPARATION  2.08  PERSON RELIABILITY  .81 |
|MODEL RMSE    .36  ADJ.SD     .92  SEPARATION  2.58  PERSON RELIABILITY  .87 |
| S.E. OF PERSON MEAN = .08                                                     |
+-------------------------------------------------------------------------------+
  MAXIMUM EXTREME SCORE:      1 PERSONS
        VALID RESPONSES:  98.6%

     SUMMARY OF 168 MEASURED (EXTREME AND NON-EXTREME) PERSONS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-------------------------------------------------------------------------------|
| MEAN      70.5      22.7        1.33     .35                                 |
| S.D.       8.9       1.2        1.07     .15                                 |
| MAX.      92.0      23.0        6.64    1.84                                 |
| MIN.      27.0       9.0        -.57     .29                                 |
|-------------------------------------------------------------------------------|
| REAL RMSE    .45  ADJ.SD     .97  SEPARATION  2.15  PERSON RELIABILITY  .82 |
|MODEL RMSE    .38  ADJ.SD    1.00  SEPARATION  2.60  PERSON RELIABILITY  .87 |
| S.E. OF PERSON MEAN = .08                                                     |
+-------------------------------------------------------------------------------+
PERSON RAW SCORE-TO-MEASURE CORRELATION = .85 (approximate due to missing data)
CRONBACH ALPHA (KR-20) PERSON RAW SCORE RELIABILITY = .88 (approximate due to missing data)

     SUMMARY OF 23 MEASURED (NON-EXTREME) ITEMS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-------------------------------------------------------------------------------|
| MEAN     511.3     164.7         .00     .12      1.00     .0   1.05     .3 |
| S.D.      91.3        .8        1.16     .02       .17    1.6    .34    2.2 |
| MAX.     619.0     166.0        2.77     .17      1.28    2.4   2.36    8.0 |
| MIN.     283.0     163.0       -1.72     .11       .64   -3.8    .61   -3.0 |
|-------------------------------------------------------------------------------|
| REAL RMSE    .13  ADJ.SD    1.15  SEPARATION  9.02  ITEM   RELIABILITY  .99 |
|MODEL RMSE    .12  ADJ.SD    1.15  SEPARATION  9.26  ITEM   RELIABILITY  .99 |
| S.E. OF ITEM MEAN = .25                                                       |
+-------------------------------------------------------------------------------+
UMEAN=.000 USCALE=1.000
ITEM RAW SCORE-TO-MEASURE CORRELATION = -.99 (approximate due to missing data)
3787 DATA POINTS. APPROXIMATE LOG-LIKELIHOOD CHI-SQUARE: 6771.06
```

STANDARDIZED RESIDUAL VARIANCE SCREE PLOT

Table of STANDARDIZED RESIDUAL variance (in Eigenvalue units)

		Empirical		Modeled
Total variance in observations	=	78.6 100.0%		100.0%
Variance explained by measures	=	55.6 70.7%		71.8%
Unexplained variance (total)	=	23.0 29.3%	100.0%	28.2%
Unexplned variance in 1st contrast	=	2.7 3.4%	11.6%	
Unexplned variance in 2nd contrast	=	2.1 2.7%	9.2%	
Unexplned variance in 3rd contrast	=	1.6 2.1%	7.1%	
Unexplned variance in 4th contrast	=	1.5 1.9%	6.6%	
Unexplned variance in 5th contrast	=	1.5 1.9%	6.4%	

VARIANCE COMPONENT SCREE PLOT



ITEM STATISTICS: MISFIT ORDER

ENTRY NUMBER	RAW SCORE	COUNT	MEASURE	MODEL S.E.	INFIT MNSQ	INFIT ZSTD	OUTFIT MNSQ	OUTFIT ZSTD	PTMEA CORR.	EXACT MATCH OBS%	EXACT MATCH EXP%	ITEM
2	508	164	.13	.11	1.23	2.1	2.36	8.0	A .21	55.5	51.2	ITEM2 when i go to bed at night
16	283	163	2.77	.12	1.28	2.4	1.27	2.2	B .40	46.0	53.7	ITEM16 when i am sick
13	579	165	-.84	.13	.96	-.3	1.28	1.5	C .35	64.2	59.1	ITEM13 when i watch television
5	357	166	1.89	.11	1.26	2.4	1.25	2.3	D .49	54.2	49.1	ITEM5 when i go to doctor's
8	458	166	.77	.11	1.22	2.1	1.20	1.8	E .46	47.0	48.9	ITEM8 when i play alone
17	567	164	-.69	.13	1.04	.4	1.21	1.3	F .35	70.7	58.0	ITEM17 when i am not sick
9	349	165	1.96	.11	1.20	1.9	1.15	1.4	G .53	50.3	49.0	ITEM9 when i take medicine
10	601	164	-1.35	.15	.98	-.1	1.17	.8	H .35	75.6	69.3	ITEM10 when it is holiday
20	537	164	-.24	.12	1.04	.4	1.11	.8	I .42	57.9	54.2	ITEM20 when i play a sport
7	335	166	2.15	.11	1.07	.7	1.00	.0	J .55	56.6	48.8	ITEM7 when i stay in hospital
21	507	165	.17	.11	1.05	.5	1.01	.1	K .51	54.5	51.2	ITEM21 when i do my homework
6	619	165	-1.72	.17	1.04	.3	1.02	.2	L .32	78.2	77.0	ITEM6 on my birthday
18	518	163	-.04	.11	1.01	.2	.95	-.3	k .48	58.3	52.5	ITEM18 when people tell me what to do
22	484	164	.40	.11	1.01	.2	.99	.0	j .51	48.2	49.7	ITEM22 when i read a book
12	600	165	-1.25	.15	.98	-.1	.88	-.5	i .35	66.7	67.1	ITEM12 when i am with my grandparents
3	556	165	-.47	.12	.96	-.4	.93	-.4	h .44	59.4	56.3	ITEM3 my brothers and sisters make me
1	572	165	-.72	.13	.91	-.8	.83	-1.0	g .43	69.1	58.3	ITEM1 when i am having dinner with my family
14	538	165	-.21	.12	.88	-1.1	.84	-1.2	f .53	58.8	54.1	ITEM14 when i move (walk, run, jump)
15	576	164	-.85	.13	.88	-1.0	.84	-.9	e .42	71.3	59.1	ITEM15 when i am eating
23	532	165	-.13	.12	.84	-1.6	.78	-1.7	d .51	62.4	53.6	ITEM23 when my work is marked at school
4	562	164	-.62	.13	.83	-1.6	.80	-1.3	c .47	68.9	57.4	ITEM4 at school
19	578	165	-.83	.13	.66	-3.2	.61	-2.6	b .49	69.1	58.9	ITEM19 when i am playing outside
11	543	165	-.29	.12	.64	-3.8	.63	-3.0	a .57	75.8	55.1	ITEM11 when i make a drawing
MEAN	511.3	164.7	.00	.12	1.00	.0	1.05	.3		61.7	56.2	
S.D.	91.3	.8	1.16	.02	.17	1.6	.34	2.2		9.4	6.9	

SUMMARY OF CATEGORY STRUCTURE. Model="R"

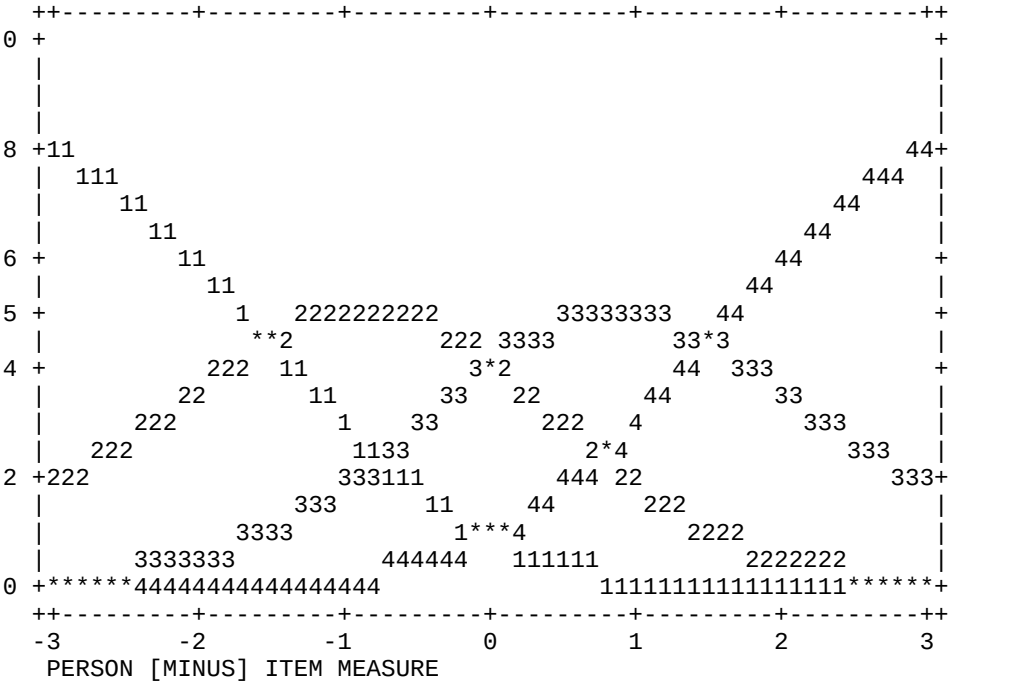
CATEGORY LABEL	SCORE	OBSERVED COUNT	%	OBSVD AVRGE	SAMPLE EXPECT	INFIT MNSQ	OUTFIT MNSQ	STRUCTURE CALIBRATN	CATEGORY MEASURE	
1	1	258	7	-.60	-1.15	1.76	2.02	NONE	(-2.76)	1
2	2	667	17	-.20	.06	.81	1.00	-1.52	-.84	2
3	3	1281	33	1.16	1.21	.81	.73	.02	.85	3
4	4	1581	41	2.36	2.29	.93	.95	1.50	(2.75)	4
MISSING		54	1	1.27						

OBSERVED AVERAGE is mean of measures in category. It is not a parameter estimate.

CATEGORY LABEL	STRUCTURE MEASURE	S.E.	SCORE-TO-MEASURE AT CAT.	----ZONE----		50% CUM. PROBABLTY	COHERENCE M->C	C->M	ESTIM DISCR	
1	NONE		(-2.76)	-INF	-1.90		48%	15%		1
2	-1.52	.08	-.84	-1.90	.01	-1.70	55%	49%	.41	2
3	.02	.05	.85	.01	1.90	.01	51%	75%	1.07	3
4	1.50	.04	(2.75)	1.90	+INF	1.68	80%	63%	1.23	4

M->C = Does Measure imply Category?
C->M = Does Category imply Measure?

CATEGORY PROBABILITIES: MODES - Structure measures at intersections



```
TABLE 12.2 C:\DOCUMENTS AND SETTINGS\DCUI\DESKTOP ZOU146WS.TXT Aug  9 20:04 2010
INPUT: 168 PERSONS  23 ITEMS  MEASURED: 168 PERSONS  23 ITEMS  4 CATS       3.63.0
-------------------------------------------------------------------------------

       PERSONS MAP OF ITEMS
               <more>|<rare>
    6             .  +
                     |
                     |
                     |
                  .  |
                     |
    5                +
                     |
                .#   |
                     |
                     |
                .#   |
    4                +
                     |
                     |
                 #   |
                   T |
                 .   |
    3            #   +
                 .   |  ITEM16 when i am sick
                 #   |
                     |
               .## S|T
                ##   |  ITEM7 when i stay in hospital
    2           .##  +  ITEM9 when i take medicine
           .######   |  ITEM5 when i go to doctor's
            .#####   |
              ####   |
              #### M |
             ######  |S
    1    .##########  +
        .###########  |  ITEM8 when i play alone
             .#####   |
           .#######   |
              ##### S|  ITEM22 when i read a book
                 ##  |  ITEM2 when i go to bed at night
                     |  ITEM21 when i do my homework
    0             #  +M ITEM18 when people tell me what to do
                  .  |  ITEM14 when i move ( walk, run, jump)
                     |  ITEM20 when i play a sport
                     |  ITEM23 when my work is marked at school
                     |  ITEM11 when i make a drawing
                .#   |  ITEM3 my brothers and sisters make me
                   T |  ITEM1 when i am having dinner with my family
                     |  ITEM17 when i am not sick
                     |  ITEM4 at school
                     |  ITEM13 when i watch television
                     |  ITEM15 when i am eating
                     |  ITEM19 when i am playing outside
   -1                +
                     |S
                     |  ITEM10 when it is holiday
                     |  ITEM12 when i am with my grandparents
                     |
                     |  ITEM6 on my birthday
                     |
   -2                +
               <less>|<frequ>
 EACH '#' IS 2.
```

2) AUQUEI (after item and category reduction)

```
     SUMMARY OF 164 MEASURED (NON-EXTREME) PERSONS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN      50.9      16.8         .03     .46      1.04    -.2   1.09     .1  |
| S.D.       6.5        .9        1.26     .08       .59    1.7   1.03    1.3  |
| MAX.      66.0      17.0        4.16     .84      4.12    4.1   9.90    5.0  |
| MIN.      20.0       7.0       -2.21     .41       .14   -4.6    .14   -2.0  |
|-------------------------------------------------------------------------------|
| REAL RMSE    .53  ADJ.SD    1.15  SEPARATION  2.18  PERSON RELIABILITY  .83  |
|MODEL RMSE    .47  ADJ.SD    1.18  SEPARATION  2.53  PERSON RELIABILITY  .86  |
| S.E. OF PERSON MEAN = .10                                                     |
+-------------------------------------------------------------------------------+
  MAXIMUM EXTREME SCORE:      4 PERSONS
        VALID RESPONSES:  98.6%

     SUMMARY OF 168 MEASURED (EXTREME AND NON-EXTREME) PERSONS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN      51.3      16.8         .18     .49                                   |
| S.D.       6.9        .9        1.59     .23                                   |
| MAX.      68.0      17.0        6.44    1.89                                   |
| MIN.      20.0       7.0       -2.21     .41                                   |
|-------------------------------------------------------------------------------|
| REAL RMSE    .60  ADJ.SD    1.47  SEPARATION  2.46  PERSON RELIABILITY  .86  |
|MODEL RMSE    .54  ADJ.SD    1.49  SEPARATION  2.74  PERSON RELIABILITY  .88  |
| S.E. OF PERSON MEAN = .12                                                     |
+-------------------------------------------------------------------------------+
PERSON RAW SCORE-TO-MEASURE CORRELATION = .88 (approximate due to missing data)
CRONBACH ALPHA (KR-20) PERSON RAW SCORE RELIABILITY = .89 (approximate due to missing data)

     SUMMARY OF 17 MEASURED (NON-EXTREME) ITEMS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN     491.0     161.7         .00     .15      1.06    -.1   1.11     .2  |
| S.D.      76.4        .9        1.63     .04       .32    2.0    .35    1.4  |
| MAX.     589.0     163.0        3.98     .30      1.94    3.6   2.03    3.1  |
| MIN.     335.0     160.0       -2.02     .13       .70   -3.5    .71   -2.0  |
|-------------------------------------------------------------------------------|
| REAL RMSE    .18  ADJ.SD    1.62  SEPARATION  8.98  ITEM   RELIABILITY  .99  |
|MODEL RMSE    .16  ADJ.SD    1.62  SEPARATION 10.27  ITEM   RELIABILITY  .99  |
| S.E. OF ITEM MEAN = .41                                                       |
+-------------------------------------------------------------------------------+
               DELETED:      6 ITEMS
UMEAN=.000 USCALE=1.000
ITEM RAW SCORE-TO-MEASURE CORRELATION = -.99 (approximate due to missing data)
2749 DATA POINTS. APPROXIMATE LOG-LIKELIHOOD CHI-SQUARE: 3981.89
```

SUMMARY OF CATEGORY STRUCTURE. Model="R"

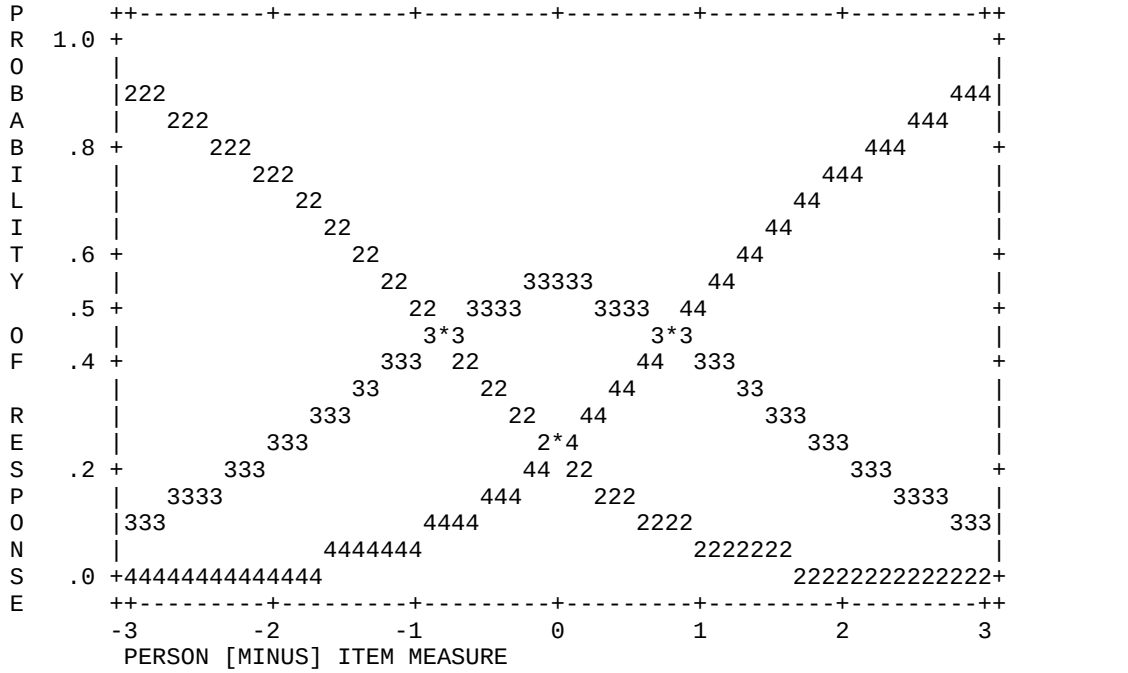
CATEGORY LABEL	OBSERVED SCORE	OBSERVED COUNT	OBSERVED %	OBSVD AVRGE	SAMPLE EXPECT	INFIT MNSQ	OUTFIT MNSQ	STRUCTURE CALIBRATN	CATEGORY MEASURE	
2	2	865	31	-1.92	-1.95	1.05	1.30	NONE	(-2.04)	1
3	3	919	33	.11	.17	.88	.93	-.82	.00	3
4	4	965	35	1.70	1.67	.99	1.13	.82	(2.04)	4
MISSING		39	1	.11						

OBSERVED AVERAGE is mean of measures in category. It is not a parameter estimate.

CATEGORY LABEL	STRUCTURE MEASURE	STRUCTURE S.E.	SCORE-TO-MEASURE AT CAT.	SCORE-TO-MEASURE ----ZONE----	50% CUM. PROBABLTY	COHERENCE M->C	COHERENCE C->M	ESTIM DISCR	
2	NONE		(-2.04)	-INF -1.16		82%	62%		1
3	-.82	.06	.00	-1.16 1.16	-.97	52%	76%	.94	3
4	.82	.05	(2.04)	1.16 +INF	.97	80%	63%	1.08	4

M->C = Does Measure imply Category?
C->M = Does Category imply Measure?

CATEGORY PROBABILITIES: MODES - Structure measures at intersections



```
TABLE 12.2 C:\DOCUMENTS AND SETTINGS\DCUI\DESKTOP ZOU696WS.TXT Aug  9 20:16 2010
INPUT: 168 PERSONS  23 ITEMS  MEASURED: 168 PERSONS  17 ITEMS  3 CATS       3.63.0
-------------------------------------------------------------------------------

       PERSONS MAP OF ITEMS
               <more>|<rare>
    6            ##  +
                     |
                     |
                     |
                     |
                     |
    5                +
                     |
                     |
                     |
                     |
                ##   |
    4                +  ITEM16 when i am sick
                     |
                     |
                 .   |
                     |T
                     |
    3              .  +
                     |
                .#   |
                 . T|  ITEM7 when i stay in hospital
                     |
                     |  ITEM9 when i take medicine
    2           .##  +  ITEM5 when i go to doctor's
                     |
                .##  |S
                  .  |
                  . S|
               .###  |
    1           ###  +
                  .  |
                ###  |
               ####  |  ITEM8 when i play alone
               .###  |
              #####  |
    0          .### M+M ITEM22 when i read a book
               ####  |
               ####  |  ITEM21 when i do my homework
           ########  |  ITEM18 when people tell me what to do
          .########  |  ITEM23 when my work is marked at school
               .###  |  ITEM11 when i make a drawing
                        ITEM14 when i move ( walk, run, jump)
                        ITEM20 when i play a sport
   -1       #######  +  ITEM3 my brothers and sisters make me
                ### S|
                 .#  |  ITEM1 when i am having dinner with my family
                        ITEM4 at school
                .##  |  ITEM19 when i am playing outside
                  #  |S
                     |
   -2             .#  +  ITEM10 when it is holiday
                   .  |
                      |
                    T|
                     |
                     |
   -3                +
               <less>|<frequ>
 EACH '#' IS 2.
```

3) LVP-FVQ (before item reduction)

```
     SUMMARY OF 149 MEASURED (NON-EXTREME) SUBJECTS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN      17.0      21.7        2.07     .72      1.00     .1    .97     .1  |
| S.D.       3.7       1.0        1.33     .21       .24     .8    .99     .8  |
| MAX.      21.0      22.0        3.79    1.08      2.00    2.3   7.88    4.1  |
| MIN.       4.0      15.0       -2.02     .50       .54   -2.4    .18   -1.6  |
|-------------------------------------------------------------------------------|
| REAL RMSE    .78  ADJ.SD    1.08  SEPARATION  1.39  SUBJEC RELIABILITY  .66  |
|MODEL RMSE    .75  ADJ.SD    1.10  SEPARATION  1.48  SUBJEC RELIABILITY  .69  |
| S.E. OF SUBJECT MEAN = .11                                                    |
+-------------------------------------------------------------------------------+
  MAXIMUM EXTREME SCORE:     17 SUBJECTS
  MINIMUM EXTREME SCORE:      1 SUBJECTS
      LACKING RESPONSES:      1 SUBJECTS
        VALID RESPONSES:  98.6%

     SUMMARY OF 167 MEASURED (EXTREME AND NON-EXTREME) SUBJECTS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN      17.4      21.7        2.33     .84                                 |
| S.D.       4.0       1.0        1.66     .41                                 |
| MAX.      22.0      22.0        5.09    1.88                                 |
| MIN.        .0      15.0       -5.22     .50                                 |
|-------------------------------------------------------------------------------|
| REAL RMSE    .96  ADJ.SD    1.35  SEPARATION  1.42  SUBJEC RELIABILITY  .67  |
|MODEL RMSE    .93  ADJ.SD    1.37  SEPARATION  1.47  SUBJEC RELIABILITY  .68  |
| S.E. OF SUBJECT MEAN = .13                                                    |
+-------------------------------------------------------------------------------+
SUBJECT RAW SCORE-TO-MEASURE CORRELATION = .94 (approximate due to missing data)
CRONBACH ALPHA (KR-20) SUBJECT RAW SCORE RELIABILITY = .85 (approximate due to missing data)

     SUMMARY OF 22 MEASURED (NON-EXTREME) ACTIVITYS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN     115.4     146.9         .00     .29       .98     .0    .96     .1  |
| S.D.      22.4       1.9        1.40     .12       .18    1.6    .34    1.4  |
| MAX.     147.0     149.0        2.65     .73      1.54    5.0   1.87    5.0  |
| MIN.      60.0     143.0       -3.12     .19       .68   -3.1    .39   -2.8  |
|-------------------------------------------------------------------------------|
| REAL RMSE    .32  ADJ.SD    1.36  SEPARATION  4.29  ACTIVI RELIABILITY  .95  |
|MODEL RMSE    .31  ADJ.SD    1.36  SEPARATION  4.35  ACTIVI RELIABILITY  .95  |
| S.E. OF ACTIVITY MEAN = .30                                                   |
+-------------------------------------------------------------------------------+
UMEAN=.000 USCALE=1.000
ACTIVITY RAW SCORE-TO-MEASURE CORRELATION = -.96 (approximate due to missing data)
3232 DATA POINTS. APPROXIMATE LOG-LIKELIHOOD CHI-SQUARE: 2272.13
```

```
TABLE 12.2 Vision Data                              ZOU574WS.TXT Aug  9 20:55 2010
INPUT: 168 SUBJECTS  22 ACTIVITYS  MEASURED: 167 SUBJECTS  22 ACTIVITYS  2 CATS
--------------------------------------------------------------------------------

       SUBJECTS MAP OF ACTIVITYS
               <more>|<rare>
    4        .#####  +
        .##########  |
                  .  |
                  .  |
                    S|
                     |
                     |
    3         #####  +
                     |T
                     |  15 can you draw a straight line on paper without a ruler
                     |
          .#######   |
                 #   |
                 .   |
    2          #### M+
                 .   |  19 can you remember words easily
                ###  |  9 can you tie up your shoelaces by yourself
                 .   |  2 do you bump into things
                 .   |S
                .#   |  22 is your handwriting neat
                     |
    1           .##  +  21 can you do maths
                        3 can you kick a ball when you play
                .###  |
                     S|
                  .#  |  16 can you copy from the board in class
                      |  14 can you read a book by yourself
                 .##  |
                      |  18 can you write your name
    0             ##  +M 11 can you see a person's face across the road
                        12 can you see bus numbers clearly
                      |  20 can you finish your homework on time
                   #  |  8 can you put toothpaste on your toothbrush by yourself
                   .  |  7 can you pick up a red pencil from a box of pencils
                   . T|
                   .  |  4 can you hit a ball when you play
                      |  17 can you write the number from 1 to 10
   -1                 +
                      |
                      |
                      |S 13 can you see the picture in your books clearly
                      |  10 can you see the TV clearly
                      |
                      |  5 can you find food on your plate when eating
                        6 can you find your favorite toy at play time
   -2              .  +
                      |
                      |
                      |
                      |
                      |
                      |T
   -3                 +
                      |  1 can you climb up and down stairs
                      |
                      |
                      |
                      |
                      |
   -4              .  +
                <less>|<frequ>
 EACH '#' IS 3.
```

4) LVP-FVQ (after item reduction)

```
TABLE 3.1 Vision Data                              ZOU778WS.TXT Aug  9 21:02 2010
INPUT: 168 SUBJECTS  22 ACTIVITYS  MEASURED: 167 SUBJECTS  16 ACTIVITYS  2 CATS
-------------------------------------------------------------------------------

     SUMMARY OF 140 MEASURED (NON-EXTREME) SUBJECTS
+-----------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT    |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-----------------------------------------------------------------------------|
| MEAN      11.5      15.8        1.46     .76      1.00     .1   1.03     .1 |
| S.D.       3.3        .8        1.38     .19       .25     .7    .79     .9 |
| MAX.      15.0      16.0        3.19    1.08      1.68    2.7   5.47    3.2 |
| MIN.       3.0      11.0       -1.76     .56       .55   -2.2    .20   -2.0 |
|-----------------------------------------------------------------------------|
| REAL RMSE    .82  ADJ.SD    1.11  SEPARATION  1.35  SUBJEC RELIABILITY  .64 |
|MODEL RMSE    .78  ADJ.SD    1.14  SEPARATION  1.45  SUBJEC RELIABILITY  .68 |
| S.E. OF SUBJECT MEAN = .12                                                  |
+-----------------------------------------------------------------------------+
  MAXIMUM EXTREME SCORE:     26 SUBJECTS
  MINIMUM EXTREME SCORE:      1 SUBJECTS
      LACKING RESPONSES:      1 SUBJECTS
        VALID RESPONSES:  98.7%

     SUMMARY OF 167 MEASURED (EXTREME AND NON-EXTREME) SUBJECTS
+-----------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT    |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-----------------------------------------------------------------------------|
| MEAN      12.1      15.8        1.90     .94                                 |
| S.D.       3.5        .8        1.74     .45                                 |
| MAX.      16.0      16.0        4.51    1.88                                 |
| MIN.        .0      11.0       -4.36     .56                                 |
|-----------------------------------------------------------------------------|
| REAL RMSE   1.06  ADJ.SD    1.38  SEPARATION  1.30  SUBJEC RELIABILITY  .63 |
|MODEL RMSE   1.04  ADJ.SD    1.40  SEPARATION  1.35  SUBJEC RELIABILITY  .64 |
| S.E. OF SUBJECT MEAN = .14                                                  |
+-----------------------------------------------------------------------------+
SUBJECT RAW SCORE-TO-MEASURE CORRELATION = .95 (approximate due to missing data)
CRONBACH ALPHA (KR-20) SUBJECT RAW SCORE RELIABILITY = .84 (approximate due to missing data)

     SUMMARY OF 16 MEASURED (NON-EXTREME) ACTIVITYS
+-----------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT    |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-----------------------------------------------------------------------------|
| MEAN     100.4     138.2         .00     .24       .99    -.1   1.03     .0 |
| S.D.      20.2       1.6        1.03     .03       .19    1.5    .40    1.3 |
| MAX.     124.0     140.0        2.29     .31      1.35    3.0   1.88    2.6 |
| MIN.      51.0     135.0       -1.45     .20       .68   -3.0    .34   -2.7 |
|-----------------------------------------------------------------------------|
| REAL RMSE    .25  ADJ.SD    1.00  SEPARATION  3.94  ACTIVI RELIABILITY  .94 |
|MODEL RMSE    .24  ADJ.SD    1.00  SEPARATION  4.08  ACTIVI RELIABILITY  .94 |
| S.E. OF ACTIVITY MEAN = .27                                                 |
+-----------------------------------------------------------------------------+
      LACKING RESPONSES:      6 ACTIVITYS
UMEAN=.000 USCALE=1.000
ACTIVITY RAW SCORE-TO-MEASURE CORRELATION = -.99 (approximate due to missing data)
2211 DATA POINTS. APPROXIMATE LOG-LIKELIHOOD CHI-SQUARE: 1798.47
```

```
TABLE 12.2 Vision Data                              ZOU778WS.TXT Aug  9 21:02 2010
INPUT: 168 SUBJECTS  22 ACTIVITYS  MEASURED: 167 SUBJECTS  16 ACTIVITYS  2 CATS
--------------------------------------------------------------------------------

       SUBJECTS MAP OF ACTIVITYS
              <more>|<rare>
    4     .########  +
                     |
                     |
                     |
                     |
                     |
                     |
                     |
         .##########  |
                     |
    3                +
                     |
                    S|
                     |
                     |
                     |
                     |
           #######   |  can you draw a straight line on paper without a ruler
                     |
                  .  |T
    2             .  +
                     |
           .######  |
                  #  |
                     |
                    M|
                     |  can you remember words easily
                ###  |  can you tie up your shoelaces by yourself
                     |
                     |
    1                +S
                .##  |
                     |  is your handwriting neat
                     |
                 .#  |  can you kick a ball when you play
                  .  |  can you do maths
                     |
               .###  |
                     |
                    S|  can you copy from the board in class
    0          .###  +M
                     |  can you read a book by yourself
                     |
                   #  |
                     |
                     |  can you see a person's face across the road
                        can you see bus numbers clearly
                        can you write your name
                     |
                .##  |
                     |  can you finish your homework on time
                     |  can you put toothpaste on your toothbrush by yourself
   -1             .  +S can you pick up a red pencil from a box of pencils
                     |
                     |
                  . T|  can you hit a ball when you play
                  .  |  can you write the number from 1 to 10
                     |
                     |
                     |
                   #  |
                     |
   -2                .  +
                <less>|<frequ>
 EACH '#' IS 3.
```

5) CVFQ (before item and category reduction)

```
  SUMMARY OF 145 MEASURED PERSONS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-------------------------------------------------------------------------------|
| MEAN     144.1      37.4        1.14     .20      1.07     .0   1.06     .0 |
| S.D.      15.8       3.3         .46     .03       .56    1.9    .69    1.7 |
| MAX.     180.0      40.0        2.91     .33      4.34    6.3   4.81    6.5 |
| MIN.      96.0      23.0         .06     .17       .32   -3.8    .33   -3.0 |
|-------------------------------------------------------------------------------|
| REAL RMSE    .23  ADJ.SD     .41  SEPARATION  1.78  PERSON RELIABILITY  .76 |
|MODEL RMSE    .20  ADJ.SD     .42  SEPARATION  2.07  PERSON RELIABILITY  .81 |
| S.E. OF PERSON MEAN = .04                                                     |
+-------------------------------------------------------------------------------+
        VALID RESPONSES:  93.4%
PERSON RAW SCORE-TO-MEASURE CORRELATION = .70 (approximate due to missing data)
CRONBACH ALPHA (KR-20) PERSON RAW SCORE RELIABILITY = .89 (approximate due to missing data)

     SUMMARY OF 40 MEASURED ITEMS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL         INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD |
|-------------------------------------------------------------------------------|
| MEAN     522.3     135.4         .00     .12      1.10     .3   1.06     .2 |
| S.D.     127.2      13.7        1.22     .06       .35    2.7    .33    2.5 |
| MAX.     714.0     145.0        2.36     .41      2.02    5.8   1.92    5.8 |
| MIN.     190.0     101.0       -3.43     .08       .46   -6.1    .48   -5.8 |
|-------------------------------------------------------------------------------|
| REAL RMSE    .15  ADJ.SD    1.21  SEPARATION  7.93  ITEM   RELIABILITY  .98 |
|MODEL RMSE    .14  ADJ.SD    1.21  SEPARATION  8.83  ITEM   RELIABILITY  .99 |
| S.E. OF ITEM MEAN = .20                                                       |
+-------------------------------------------------------------------------------+
               DELETED:     15 ITEMS
UMEAN=.000 USCALE=1.000
ITEM RAW SCORE-TO-MEASURE CORRELATION = -.91 (approximate due to missing data)
5417 DATA POINTS. APPROXIMATE LOG-LIKELIHOOD CHI-SQUARE: 11820.12
```

```
INPUT: 145 PERSONS  55 ITEMS  MEASURED: 145 PERSONS  40 ITEMS  5 CATS       3.63.0
--------------------------------------------------------------------------------

       PERSONS MAP OF ITEMS
               <more>|<rare>
    3             .  +
                     |
                  .  |
                     |T ITEM45 eyesight make difficult to mobility
                  .  |
    2             .# T+  ITEM46 vision gets in the way of learning
                 .###  |
                  ### S|
              .#######  |  ITEM3 eyesight with affected eye
             ######### M|S ITEM15 other children looking at mine
                        ITEM4 worry about eyesight
    1         ########  +  ITEM13 feel different from others
                        ITEM20 make friends easily
                        ITEM21 affectionate
              .#######  |  ITEM2 eyesight
                        ITEM22 get along well with others
                        ITEM42 enjoy looking at books
                 .#### S|  ITEM5 spend on vision treatment
                        ITEM51 enjoy drawing
                    .#  |  ITEM1 overall health
                        ITEM16 visit with relatives
                        ITEM17 teased for vision problem
                        ITEM19 worry about can't read, watch, drive
                        ITEM33 ride a bike
                        ITEM48 enjoy watching TV, play video game
                        ITEM49 like to travel on vacation
                        ITEM50 enjoy playing with others
                        ITEM52 eyesight make it difficult to find sth on shelf
                        ITEM9 bothered by other people's comments
                   . T|  ITEM6 take away time for therapy
                        ITEM7 argue about medical care
    0              .  +M ITEM14 happy most of time
                        ITEM34 play a sport
                        ITEM40 tell what time
                      |
                      |
                      |  ITEM41 identify coins
                      |  ITEM39 help with chores
                        ITEM54 bumps into people
                        ITEM55 trips over curbs or steps
   -1                 +  ITEM38 dial a telephone
                      |S
                      |  ITEM29 dress himself
                      |
                      |  ITEM30 brush teeth
                        ITEM31 wash face
   -2                 +  ITEM36 locate a small piece of food
                      |  ITEM25 feed himself
                        ITEM37 pour liquid into a cup
                      |T
                      |
                      |
   -3                 +
                      |
                      |  ITEM27 recognize faces across room
                      |
                      |
   -4                 +
               <less>|<frequ>
 EACH '#' IS 3.
```

6) CVFQ (after item and category reduction)

```
   SUMMARY OF 145 MEASURED PERSONS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL          INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN     108.1      37.4         .66     .22      1.06     .0   1.06     .0  |
| S.D.      13.5       3.3         .54     .02       .51    1.8    .69    1.7  |
| MAX.     143.0      40.0        2.67     .36      3.33    5.9   5.20    6.8  |
| MIN.      68.0      23.0        -.60     .20       .34   -3.9    .35   -2.8  |
|-------------------------------------------------------------------------------|
| REAL RMSE    .25  ADJ.SD     .48  SEPARATION  1.95  PERSON RELIABILITY  .79  |
|MODEL RMSE    .22  ADJ.SD     .49  SEPARATION  2.23  PERSON RELIABILITY  .83  |
| S.E. OF PERSON MEAN = .04                                                     |
+-------------------------------------------------------------------------------+
        VALID RESPONSES:  93.4%
PERSON RAW SCORE-TO-MEASURE CORRELATION = .80 (approximate due to missing data)
CRONBACH ALPHA (KR-20) PERSON RAW SCORE RELIABILITY = .87 (approximate due to missing data)

     SUMMARY OF 40 MEASURED ITEMS
+-------------------------------------------------------------------------------+
|           RAW                          MODEL          INFIT        OUTFIT     |
|          SCORE     COUNT     MEASURE   ERROR      MNSQ   ZSTD   MNSQ   ZSTD  |
|-------------------------------------------------------------------------------|
| MEAN     392.0     135.4         .00     .13      1.08     .2   1.06     .2  |
| S.D.     112.8      13.7        1.33     .06       .28    2.3    .31    2.2  |
| MAX.     570.0     145.0        2.82     .41      1.67    4.2   1.85    4.7  |
| MIN.     127.0     101.0       -3.53     .09       .49   -6.4    .51   -5.9  |
|-------------------------------------------------------------------------------|
| REAL RMSE    .16  ADJ.SD    1.32  SEPARATION  8.17  ITEM   RELIABILITY  .99  |
|MODEL RMSE    .15  ADJ.SD    1.32  SEPARATION  9.04  ITEM   RELIABILITY  .99  |
| S.E. OF ITEM MEAN = .21                                                       |
+-------------------------------------------------------------------------------+
                DELETED:     15 ITEMS
UMEAN=.000 USCALE=1.000
ITEM RAW SCORE-TO-MEASURE CORRELATION = -.95 (approximate due to missing data)
5417 DATA POINTS. APPROXIMATE LOG-LIKELIHOOD CHI-SQUARE: 10757.53
```

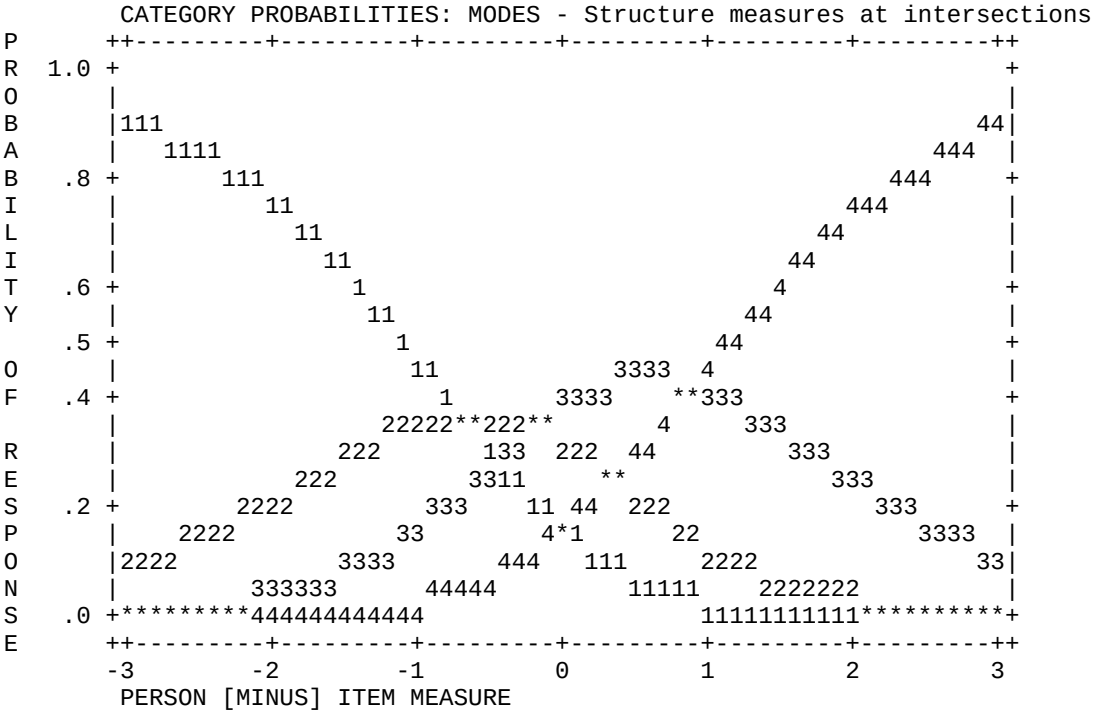
SUMMARY OF CATEGORY STRUCTURE. Model="R"

CATEGORY LABEL	SCORE	OBSERVED COUNT	%	OBSVD AVRGE	SAMPLE EXPECT	INFIT MNSQ	OUTFIT MNSQ	STRUCTURE CALIBRATN	CATEGORY MEASURE	
1	1	780	13	-.64	-.78	1.22	1.36	NONE	(-2.12)	1
2	2	1040	18	-.11	-.10	1.02	1.19	-.71	-.63	3
3	3	1569	27	.41	.59	1.00	.76	-.19	.58	4
4	4	2028	35	1.94	1.84	.87	.95	.90	(2.20)	5
MISSING		383	7	-.27						

OBSERVED AVERAGE is mean of measures in category. It is not a parameter estimate.

CATEGORY LABEL	STRUCTURE MEASURE	S.E.	SCORE-TO-MEASURE AT CAT.	----ZONE----	50% CUM. PROBABLTY	COHERENCE M->C	C->M	ESTIM DISCR	
1	NONE		(-2.12)	-INF -1.42		72%	21%		1
2	-.71	.05	-.63	-1.42 -.04	-1.09	36%	50%	.63	3
3	-.19	.04	.58	-.04 1.43	-.08	42%	60%	.78	4
4	.90	.04	(2.20)	1.43 +INF	1.16	85%	62%	1.27	5

M->C = Does Measure imply Category?
C->M = Does Category imply Measure?



```
TABLE 12.2 \\TSCLIENT\I\CARER WITH RECODE AND MIS ZOU498WS.TXT Aug  9 21:41 2010
INPUT: 145 PERSONS  55 ITEMS  MEASURED: 145 PERSONS  40 ITEMS  4 CATS       3.63.0
-------------------------------------------------------------------------------

       PERSONS MAP OF ITEMS
               <more>|<rare>
    3                +
                     |  ITEM45 eyesight make difficult to mobility
                   .  |T ITEM46 vision gets in the way of learning
                   .  |
                      |
                      |
    2                 +
                   #  |
                  .# T|  ITEM3 eyesight with affected eye
                 .##  |
                .####  |S ITEM15 other children looking at mine
                  ### S|  ITEM4 worry about eyesight
    1         #######  +  ITEM13 feel different from others
                         ITEM20 make friends easily
                         ITEM21 affectionate
              #######  |  ITEM2 eyesight
                         ITEM22 get along well with others
                         ITEM42 enjoy looking at books
           .########## M|  ITEM5 spend on vision treatment
                         ITEM51 enjoy drawing
           ###########  |  ITEM19 worry about can't read, watch, drive
                         ITEM49 like to travel on vacation
            #########  |  ITEM1 overall health
                         ITEM16 visit with relatives
                         ITEM17 teased for vision problem
                         ITEM33 ride a bike
                         ITEM48 enjoy watching TV, play video game
                         ITEM50 enjoy playing with others
                         ITEM52 eyesight make it difficult to find sth on shelf
                         ITEM9 bothered by other people's comments
              ###### S|  ITEM6 take away time for therapy
    0           .###  +M ITEM14 happy most of time
                         ITEM7 argue about medical care
                .##  |  ITEM40 tell what time
                  . T|  ITEM34 play a sport
                 .#  |
                  .  |
                     |  ITEM39 help with chores
                         ITEM41 identify coins
                         ITEM54 bumps into people
                         ITEM55 trips over curbs or steps
   -1                +
                     |  ITEM38 dial a telephone
                     |S
                     |  ITEM29 dress himself
                     |
                     |  ITEM30 brush teeth
   -2                +  ITEM31 wash face
                     |  ITEM36 locate a small piece of food
                        ITEM37 pour liquid into a cup
                     |  ITEM25 feed himself
                     |
                     |T
                     |
   -3                +
                     |
                     |
                     |  ITEM27 recognize faces across room
                     |
                     |
   -4                +
               <less>|<frequ>
 EACH '#' IS 2.
```

Appendix 4 Ethical application and cover letter of quesionnaire

5 July 2005

Human Research Ethics Committee

The University of NSW

NSW 2052

THE UNIVERSITY OF
NEW SOUTH WALES

Catherine M. Suttle
School of Optometry and Vision Science

Dear Sir or Madam:

Re: HREC 04110 Impact of visual abnormality on quality of life in children with intellectual disability

I would like to investigate the visual-related quality of life in the schools as following:

Damin School for Special Education, Ningbo, China

Lujiazhui School for Special Education, Shanghai, China

The investigation will include conducting vision tests and distributing a questionnaire package, which is to be completed by parents/caregivers and children. All the research activities conducted in above-mentioned schools will be under supervision of the investigators of HREC 04110.

I am hereby applying to the committee for the approval of the investigation.

Sincerely yours,

Catherine M. Suttle

Lecturer

School of Optometry and Vision Science
University of New South Wales
Sydney NSW 2052, Australia
Email C.Suttle @unsw.edu.au
Phone (61)2 9385 4620
Fax (61)2 9313 6243

30 Nov. 2005

THE UNIVERSITY OF
NEW SOUTH WALES

Catherine M. Suttle
School of Optometry and
Vision Science

Human Research Ethics Committee

UNSW

Dear Sir or Madam:

Re: HREC 04110 Impact of visual abnormality on quality of life in children with intellectual disability

I would like to request two modifications the above study. Firstly, we wish to offer our quality of life questionnaires to children in the school in China. The questionnaires would be provided to children by their school-teacher, for each child to take home, and if the child and his/her parents choose to complete the questionnaire, it would be returned to the school for collection by us. No separate Subject Information and Consent form would be provided for completion of the questionnaire, since completion itself implies consent.

Secondly, we wish to conduct our vision tests at the school. In this case, our Information and Consent form would be offered to parents, and children whose parents agree to their participation would undergo our visual function tests at their school. The collaborator in China has offered access to large number of children as subjects in our research project. The PhD student Yu.Cui, who had worked in China as ophthalmologist, will conduct the quesionnaire.

Copies of letters of support from the head teacher of the school is provided herewith. I hope that the Committee will consider the above amendments at their next meeting.

Yours truly,

Catherine M. Suttle

Lecturer

School of Optometry and Vision Science

University of New South Wales

Sydney NSW 2052, Australia

Email C.Suttle@unsw.edu.au

Phone (61)2 9385 4620

Fax (61)2 9313 6243

THE UNIVERSITY OF
NEW SOUTH WALES

Optometry and Vision Science
THE UNIVERSITY OF NEW SOUTH WALES

Catherine M. Suttle PhD MCOptom
School of Optometry and Vision Science

Dear Parent/carer

Here at the School of Optometry and Vision Science, University of New South Wales, we are conducting a study of vision care and its impact on quality of life in children with developmental delay. As part of the study, we are conducting a survey. We enclose a set of questionnaires, which we hope you and your child will take the time to complete. Two questionnaires are to be completed by the caregiver, and three questionnaires are to be completed by the child (with help if needed). The enclosed instructions give further details to guide you with completion of the questionnaires. The questionnaires are, and will remain, anonymous, will provide us with very valuable information on your child's eye care. A pre-paid envelope is enclosed, for return of the questionnaires.

If you are happy to complete the questionnaires, please do so, and return it to us in the pre-paid envelope.

If you do not wish to complete the questionnaires, please disregard this letter.

Thank you.

Yours sincerely,

Catherine M. Suttle PhD, MCOptom

Fiona Stapleton PhD, MSc, MCOptom

THE UNIVERSITY OF
NEW SOUTH WALES

Catherine M. Suttle
School of Optometry and Vision Science

Approval No (04110)

THE UNIVERSITY OF NEW SOUTH WALES

SOUTH EAST AREA HEALTH SERVICE

PARTICIPANT INFORMATION STATEMENT AND CONSENT FORM

Impact of Visual Abnormality on Quality of Life in Children with Intellectual Disability

We invite you and your child to participate in a study investigating the impact of visual abnormality on quality of life in children with and without intellectual disability. Visual abnormalities are very common in children with intellectual disability. Visual abnormalities have a negative impact on quality of life in children and adults in the general population, but it is not known whether such abnormalities have a similar impact in the population of children with intellectual disability. We hope to learn whether children with intellectual disability suffer a reduction in their quality of life as a result of visual abnormality. Your child was selected as a possible participant in this study because he/she is within the age range we intend to study.

If you decide to participate, a registered optometrist will examine your child's vision using standard clinical procedures. In addition, your child's quality of life will be assessed by means of one or more short questionnaires, to be completed by the child or

by a parent or guardian. You may be asked to return for the same tests and questionnaires on a second occasion. The vision tests are of approximately 30 minutes' duration, and the questionnaires will take approximately 30 minutes to complete, on each occasion (a total of approximately one hour, on two occasions).

Any information that is obtained in connection with this study and that can be identified with you or your child will remain confidential and will be disclosed only with your permission or except as required by law. If you give us your permission by signing this document, we plan to discuss and publish the results, at scientific conferences and in scientific journals. In any publication, information will be provided in such a way that you cannot be identified. Complaints may be directed to the Ethics Secretariat, The University of New South Wales, Sydney 2052 Australia (phone 9385 4234, fax 9385 6648, email ethics.sec@unsw.edu.au). Alternatively, you can contact Ms. Liu Jia Feng (phone 0574-56443233, fax 0574-56443233), the head of Damin School, Ningbo,who will forward your complaints to the Ethics Secretariat, The University of New South Wales.

Your decision whether or not to participate will not prejudice your or your child's future relations with The University of New South Wales. If you decide to participate, you are free to withdraw your consent and to discontinue participation at any time without prejudice.

If you have any questions, please feel free to ask us. If you have any additional questions later, Mr.Yu Cui (021-65201023, email danielcui@vip.sina.com) will be happy to answer them. You will be given a copy of this form to keep.

You are making a decision whether or not to participate. Your signature indicates that, having read the Participant Information Statement, you have decided to take part in the study.

…… ……

Signature of Research Participant Signature of Witness

…… …..…….

(Please PRINT name) (Please PRINT name)

…… ……..…….

Date Nature of Witness

Appendix 5 Publications and presentations

Original publications related to the thesis

Cui Y, Stapleton F, Suttle C. Developing an instrument to assess vision-related and subjective quality of life in children with intellectual disability: data collection and preliminary analysis in a Chinese population, Ophthalmic Physiol Opt. 2008 May;28(3):238-46

Cui Y, Stapleton F, Suttle C, Bundy A. Health- and Vision-Related Quality of Life in Intellectually Disabled Children. Optometry and Vision Science, 2010 Jan;87(1):37-44.

Abstracts related to the thesis

Cui Y, Stapleton F, Suttle C. Visual Impact on Quality of Life in Children with Intellectual Disability, proceedings in 11th Scientific Meeting in Optometry, Australia, published as abstract in Clin Exp Optom 2006; 89: 2: 104–117.

Cui Y, Stapleton F, Suttle C. Development of Questionnaires to Assess Vision and Health-Related Quality of Life in School Children with Intellectual Disability, poster presentation at Annual Meeting of American Academy Optometry, 2007.

Cui Y, Stapleton F, Suttle C. Vision in Children with Intellectual Disability, poster presentation at Australian Ophthalmic and Vision Science Meeting, 2008.

Cui Y, Stapleton F, Suttle C. An Assessment of Health and Vision-related Quality of Life in Children with Intellectual Disabilities, poster presentation at UNSW Research Showcases, 2009.

Cui Y, Stapleton F, Suttle C. Health- and vision-related quality of life in children with intellectual disability, poster presentation at 13th Biennial Scientific Meeting, 7th Educators' Meeting in Optometry,2010.