

**Quality of Life and Self-Report in Children and Adolescents with  
Cognitive Disability**

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**ATTESTATION OF AUTHORSHIP**

I hereby declare that this submission is my own work and that, to the best of my knowledge and belief, it contains no material previously published or written by another person nor material which to a substantial extent has been accepted for the award of any other degree or diploma of a university or other institution of higher learning, except where due acknowledgement is made.

Name Erika Ikeda

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As this research involved human participants, I would like to acknowledge that this thesis has received approval from the following ethics committee: Auckland University of Technology Ethics Committee (date of approval: 19 September 2011, reference number: 11/206) (see Appendix A for complete proofs).

## **ABSTRACT**

The purpose of this research was to initiate the development of a tool that could be used by children and adolescents with autism spectrum disorders (ASDs) or mild intellectual disability (MID) to communicate perspectives of their quality of life (QOL). This research gathered direct verbal responses and opinions from parents and children and adolescents with ASDs or MID to assess the content validity of QOL instrument, Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) Child Self-Report. The PedsQL questionnaire is designed to measure QOL in children and adolescents based on multidimensional domains of physical, emotional, social and school functioning.

This research was qualitative descriptive and consisted of Phase one and two. By using sample of convenience and purposive sampling techniques, participants were recruited from a special school, a school with a special unit in Auckland region and via two private service providers, the Centre for Autism and Related Disorders and the Wilson Home Trust in each phase. Focus groups and individual interviews in Phase one and the interviews in Phase two were employed to obtain the participants' perspectives and feedback on the appropriateness of the PedsQL questionnaire. The questionnaire was presented in both phases, and completed by the children and adolescents in Phase two. Thematic and content analyses were conducted in both phases to discover key themes and categories.

The findings showed that the PedsQL questionnaire was inappropriate for children and adolescents with ASDs or MID in terms of: (1) a number of missing and irrelevant questions on social domain, (2) use of abstract concepts and words, and (3) monotonous formatting, response scales and presentation style. Cognitive and language ability and disease-specific symptoms such as impaired social relationships strongly influenced the comprehension and appraisal of the questionnaire. The modification of the PedsQL questionnaire was undertaken after Phase one including use of concrete wording, visuals, facial expressions and one-by-one presentation style, which assisted the children's and adolescents' understanding of and engagement with the questionnaire in Phase two. Thus, the development of the PedsQL questionnaire for children and adolescents with ASDs or MID may enhance the accuracy of their self-rating and obtain their unique perception of QOL. Based on this research, future research will explore

further modification of the PedsQL questionnaire for use in children and adolescents with ASDs or MID as well as the use of different modes of administration such as qualitative interviews and electronic devices.

## **CHAPTER 1: INTRODUCTION**

### **BACKGROUND**

In the last few decades, autism spectrum disorders (ASDs) including autistic disorder, Asperger disorder and atypical autism have been widely recognised as an “epidemic” of mental health conditions, which necessitated the rapid growth in research regarding these conditions (Elsabbagh et al., 2012). The prevalence of ASDs has increased worldwide over time, and the latest global prevalence rate of ASDs was estimated at one child out of 160 (Elsabbagh et al., 2012). In New Zealand, the prevalence rate of ASDs is one child per 100, and estimated number of individuals with ASDs is over 40,000 (Ministry of Education, 2013). From clinical and healthcare perspectives, ASDs are a cause of high service utilisation due to their early onset, lifelong persistence, high degree of related impairment, and absence of effective treatment and intervention for the main problems such as impaired social relationships and interactions (Cremeens, Eiser, & Blades, 2006; Simonoff et al., 2008; Varni, Burwinkle, & Lane, 2005).

The core defining attributes of ASDs are impaired social interactions, impaired communication, repetitive behaviours and restricted interest (American Psychiatric Association [APA], 2000; Plimley, 2007). The ASDs are associated with impaired cognitive abilities, epilepsy and high rates and various types of child psychiatric comorbidity such as anxiety disorder, attention deficit hyperactivity disorder (ADHD), and oppositional defiant disorder (Elsabbagh et al., 2012; Farley et al., 2009; Kamp-Becker et al., 2011; Sheldrick, Neger, Shipman, & Perrin, 2011; Simonoff et al., 2008). However, differentiation between ASDs and intellectual disability (ID) is still blurred because of the attribute of diagnostic overlap between psychiatric symptoms and intellectual impairment (Simonoff et al., 2008). In New Zealand, for instance, around two percent of children and adolescents were diagnosed with ID (Ministry of Health, 2005, 2011) and approximately 28% of children and adolescents with ID had treatment or care for mental health problems including ADHD, anxiety disorder, autism spectrum and mood disorder (Ministry of Health, 2011). Furthermore, both ASDs and ID significantly impact on personal development and social, psychological, physical and material well-being in children and adolescents such as school activities, education, peer

relationships and healthcare costs (McIntyre, Kraemer, Blacher, & Simmerman, 2004; Ministry of Health, 2005, 2011; Schalock, 2004; Simonoff et al., 2008).

The World Health Organisation Quality of Life Group (WHOQOL Group, 1995) defined quality of life (QOL) as “an individual’s perception of their position in life, in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (p. 1405). QOL is generally conceptualised as a multidimensional construct involving broad domains such as physical, mental and social well-being (Coghill, Danckaerts, Sonuga-Barke, Sergeant, & the ADHD European Guidelines Group, 2009; Davis et al., 2006; Eiser & Morse, 2001). QOL also encompasses subjective and objective perspectives which are determined by personal appraisal and functional assessment, respectively (Eiser & Morse, 2001; Schalock, 2004). Health-related quality of life (HRQOL) is more specific than QOL, and defined as the impact of health, illness and disorder on QOL (Drotar, 2004; Eiser & Morse, 2001). Recently, QOL and HRQOL for children and adolescents have received greater attention in relation to the evaluation of treatment efficacy and well-being among paediatric populations with chronic and mental health conditions (Coghill et al., 2009; Upton, Lawford, & Eiser, 2008). In paediatric trials and practice settings, QOL and HRQOL are viewed as “gold standards” against which other health outcomes should be assessed (Cummins, 2005; Eiser & Morse, 2001; Schalock, 2004). Outcomes of QOL and HRQOL are used for the evaluation of clinical decision-making, cost-effectiveness analyses of treatment and intervention, and strategies for improving treatment (Coghill et al., 2009; Matza, Swensen, Flood, Secnik, & Leidy, 2004; Simonoff et al., 2008; Varni et al., 2005). In public and healthcare service settings, QOL and HRQOL are utilised as a consumer’s voice and the evaluation of healthcare system, service planning and service management (Coghill et al., 2009; Matza et al., 2004; Simonoff et al., 2008; Varni et al., 2005). QOL and HRQOL also impacts on public policy, service delivery principles and disability reform (Schalock, 2004).

To date, a number of generic QOL and HRQOL measures for children and adolescents with chronic diseases and mental health problems have emerged and developed, such as Child Health and Illness Profile (CHIP), Child Health Questionnaire (CHQ) and Pediatric Quality of Life Inventory (PedsQL) (Coghill et al., 2009; Dey, Landolt, & Mohler-Kuo, 2012; Eiser & Morse, 2001). Paediatric QOL and HRQOL measures

involve at least one of two versions: self-report and proxy-report. For children and adolescents who are too young, ill, or unable to respond themselves due to their impaired cognitive and linguistic ability, proxy ratings usually by parents, clinicians or teachers are provided (Eiser & Morse, 2001). To the extent that QOL and HRQOL are subjective, it is necessary to be assessed from the child's perspective based on self-rating wherever possible (Coghill et al., 2009; Eiser & Morse, 2001; Matza et al., 2004). Although proxy-reports may be valuable and accurate, proxy ratings are based on parents' point of views which are affected by their own well-being and experience as well as expectations of their children (Eiser & Morse, 2001).

It is important to determine relationships between self- and proxy-ratings depending on age, developmental stage and health status (Bouman, Koot, Van Gils, & Verhulst, 1999; Eiser & Morse, 2001; Matza et al., 2004). Due to the increasing recognition of the value of self-reports, there is a need to develop appropriate QOL and HRQOL measures for children and adolescents (Cremeens et al., 2006; Matza et al., 2004). The appropriateness of the instruments including the wording, sentence, structure, content, response scales and presentation styles are determined by the chronological and developmental age and types of disorder (Cremeens et al., 2006; Matza et al., 2004). Several studies examined different response scales (i.e. Likert scales, facial expression and visual analogue scales) and presentation styles (i.e. written, pictorial, verbal, and computerised) (Cremeens et al., 2006). It is evident that the child-friendly formatting and presentation of the measures is likely to improve the completion rate and reliability of self-reports (Coghill et al., 2009).

Research on paediatric QOL and HRQOL has primarily highlighted chronic and acute diseases but little attention has been given to psychiatric disorders (Matza et al., 2004). To date, research that investigated QOL and HRQOL in children and adolescents with mental health problems, particularly ADHD (Coghill et al., 2009; Danckaerts et al., 2010; Dey et al., 2012; Pongwilairat, Louthrenoo, Charmsil, & Witoonchart, 2005; Varni & Burwinkle, 2006). Bastiaansen, Koot, Bongers, Varni and Verhulst (2004) reported that children with psychiatric disorders had impaired QOL compared to healthy children. Another study by Bastiaansen, Koot, Ferdinand and Verhulst (2004) compared QOL scores between child psychiatric disorders such as attention-deficit and disruptive behaviour disorders, anxiety disorders, pervasive developmental disorders (PDDs) and

mood disorders. The results showed that QOL ratings by clinicians were lower in children with PDDs, so-called ASDs, including autistic disorder and Asperger disorder than other psychiatric disorders (Bastiaansen, Koot, Ferdinand, et al., 2004).

The higher prevalence of children and adolescents with ASDs and the increasing necessity of healthcare services for this population are widely recognised (Elsabbagh et al., 2012; Plimley, 2007). Nevertheless, there are a limited number of studies on QOL and HRQOL conducted in children and adolescents with ASDs. Plimley (2007) identified that specific considerations of QOL for individuals with ASDs were necessary across a number of domains, such as physical well-being, emotional well-being, social inclusion, interpersonal relations, personal development, self-determination, rights and material well-being. Taking account of the specific characteristics and issues for children and adolescents with ASDs, the general concept of QOL and HRQOL may not be relevant to this population. Consequently, measurement of QOL and HRQOL for use in children and adolescents with ASDs may need to be examined and designed specifically for this population.

The U.S. Department of Health and Human Services Food and Drug Administration (FDA) (2009) made several recommendations on research design for the modification of patient-reported outcome instruments:

- 1) A small nonrandomised study: To compare the distribution of responses between different versions.
- 2) A qualitative study: To confirm content validity in a new population.
- 3) A small randomised study: To ascertain the measurement properties in the new population. (pp. 20-21)

It is also important that the research identifies the intended population and condition to use for the instrument (FDA, 2009). In keeping with this guideline, the present research on the development of QOL measurement for use in children and adolescents with ASDs or mild intellectual disability (MID) were undertaken.

The research question, “What is an appropriate QOL measurement for children and adolescents with ASDs or MID?” was developed to address the gap in research and



practice. The aim of this research was to initiate the development of a tool that could be used by children and adolescents with ASDs or MID to communicate perspectives of their QOL. In order to achieve this aim, two objectives were established; (1) to examine the content validity of a child self-report QOL inventory, Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) 4.0 Generic Core Scales, among children and adolescents with ASDs or MID; (2) to develop a child self-report QOL inventory, based on the PedsQL items, appropriate for children and adolescents with ASDs or MID.

### **THESIS RATIONALE**

This research is part of a larger project, SNAP! for Health (Special children, Nutrition, Activity, and Participation) to be implemented in partnership with special needs schools, teachers and families. The programme is a school-based intervention that aims to improve health and well-being in children with cognitive disability. The development of a valid and reliable child self-report of QOL is needed to evaluate the effectiveness of the intervention with relation to the improvement of overall health and well-being among this population. However, we identified a gap in the literature regarding the use of appropriate child self-reports of QOL in this population. In particular, there is little research on the validation of QOL measures using a qualitative research approach. The examination of this issue will be beneficial specifically to vulnerable populations, children and adolescents with cognitive disability (Sullivan-Bolyai, Bova, & Harper, 2005). Thus, this research was aimed to initiate the development of a tool that could be used by children and adolescents with cognitive disability, specifically autism spectrum disorders or mild intellectual disability, to communicate perspectives of their QOL.

### **Choice of Research Design**

A qualitative descriptive study was employed to answer the research question: “What is an appropriate QOL measurement for children and adolescents with autism spectrum disorders (ASDs) or mild intellectual disability (MID)?” A qualitative descriptive study is suitable for this research because:

- 1) It aims to address the who, what and where of experiences and perceptions, which allows the researchers to obtain preliminary insight into and knowledge of

the development of a QOL instrument for children and adolescents with ASDs or MID.

- 2) The subjective perspective of a qualitative descriptive study is in concordance with the concept of QOL, which respect the informants' points of views and reflect their perspectives.
- 3) It can provide information on the important conception of QOL in children and adolescents with ASDs and MID beyond the existing confines which cannot be identified through a deductive quantitative approach.

A qualitative descriptive study refers to an inductive and data-driven pragmatic study approach. It also embraces the concept of a naturalistic inquiry in which there are no “pre-selection of variables to study, manipulation of variables and a priori commitment to anyone theoretical view of a target phenomenon” (Sandelowski, 2000, p. 337). A qualitative descriptive study is recognised as a categorical alternative to phenomenology, grounded theory, ethnography or a narrative study (Neergaard, Olesen, Andersen, & Sondergaard, 2009; Sandelowski, 2000). In clinical and healthcare practice, a qualitative descriptive study is useful for the development of questionnaire and needs assessments due to the nature of rich and subjective information based on patients' points of view regarding health-related concerns and issues (Neergaard et al., 2009; Sullivan-Bolyai et al., 2005). In this research, a qualitative descriptive study was employed to effectively reflect participants' views on the development of the PedsQL child self-report.

### **Choice of Subjects**

This research identified that children and adolescents with ASDs and ID were vulnerable in terms of physical, psychological and social impairments. Children and adolescents with ASDs have deficits in reciprocal social and emotional interactions, communication and flexibility of thought and behaviour, which considerably impact on their well-being (APA, 2000; Plimley, 2007). According to Ministry of Health (2011), over a quarter of children and adolescents with ID had chronic diseases including coronary heart disease, chronic respiratory disease, diabetes, chronic kidney disease, cancer and morbid obesity. They also experienced difficulty participating in play, sports

or games, and making friends (Ministry of Health, 2005). Furthermore, the growing prevalence rate and service use of this population indicates an urgent need for a reliable self-appraisal of their QOL in the context of family, school and community (Elsabbagh et al., 2012; Ministry of Health, 2011; Simonoff et al., 2008).

In this research, participants were selected using sample of convenience and a purposive sampling technique. Sample of convenience is a non-probability sampling technique in which participants are selected due to their accessibility to the researcher (Marshall, 1996). Sample of convenience is useful for this research due to the limited number of available target population and time constraint. This technique was used in Phase one, and both sample of convenience and a purposive sampling was applied in Phase two. According to Sandelowski (2000), a purposive sampling refers to a strategy to obtain individuals who are considered as “information-rich” for the aim of research (p. 338). In this study, three participants from Phase one were selected for Phase two study due to the parent’s sufficient knowledge and experience in the area of ASDs or ID.

### **Choice of Data Collection Methods**

Focus groups and interviews were employed to collect a variety of in-depth data regarding the appropriateness of the PedsQL self-reports for use in children and adolescents with ASDs or MID. These methods are recommended in a qualitative descriptive approach in order to identify key aspects of the *who*, *what* and *where* of experiences and perceptions of participants (Neergaard et al., 2009; Sandelowski, 2000). Taking into account of the unique characteristics of ASDs and MID such as impaired psychosocial functioning (Plimley, 2007; Schalock, 2004), focus groups and interviews are suitable to discover underlying problems pertinent to their QOL. The combination of focus groups and interviews are advantageous due to the scope to obtain a broad range of information through focus groups and attain more specific and detailed opinions, experiences and feedback through interviews (Morgan, 1996).

### **Choice of Measure**

On the basis of available literature and student and supervisors discussions it was agreed that the Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) 4.0 Generic Core Scales Child

Self-Report was the most suitable QOL instrument for use in children and adolescents with cognitive disability. The PedsQL 4.0 Generic Core Scales was initially designed for healthy children and adolescents and those with acute and chronic health conditions (Varni, n.d.). Recently the PedsQL child self-report has been used in children and adolescents with psychiatric disorders and established its psychometric properties such as attention deficit hyperactivity disorder (ADHD) (Bastiaansen, Koot, Bongers, et al., 2004; Bastiaansen, Koot, Ferdinand, et al., 2004; Pongwilairat et al., 2005; Sawyer et al., 2002; Varni & Burwinkle, 2006) and autism spectrum disorders (ASDs) or pervasive developmental disorders (PDD) (Bastiaansen, Koot, Ferdinand, et al., 2004; Sheldrick et al., 2011; Shipman, Sheldrick, & Perrin, 2011). However, lack of information on psychometric properties and mixture of other psychometric disorders were always problematic for identifying the findings in children and adolescents with ASDs and MID. In addition, there is little evidence that the content appropriateness of the PedsQL child self-report was qualitatively analysed and verified for use in children and adolescents with ASDs or MID.

### **Choice of Analysis**

This research conducted content and thematic analyses to identify key themes and categories from data in Phase one and two. Content analysis is a way to analyse the manifest as well as latent content of data reflectively and interactively, and it yields a straight descriptive summary of the patterns of data (Neergaard et al., 2009; Sandelowski, 2000). However, it also implies lack of interpretation of the meaning of data (Neergaard et al., 2009; Sandelowski, 2000). In order to move beyond description and produce an in-depth conceptual description and understanding the meaning of data, thematic analysis was employed (Neergaard et al., 2009; Sandelowski, 2000).

In relation to this research, content analysis provides knowledge and a practical strategy for developing the PedsQL self-report for use in children with ASDs or MID. In contrast, thematic analysis provides a rich and detailed account of data from focus groups and interviews regarding the appropriateness of the PedsQL self-report for use in children with ASDs or MID. Therefore, content analysis was conducted to identify key factors which impact on the appropriateness of the PedsQL questionnaire by abstracting meaning units within the data set (Elo & Kyngäs, 2008; Graneheim & Lundman, 2004).

Thematic analysis was used to interpret insight and perception of children with ASDs or MID and their parents towards the PedsQL questionnaire by identifying themes, patterns or implications within the data set (Braun & Clarke, 2006).

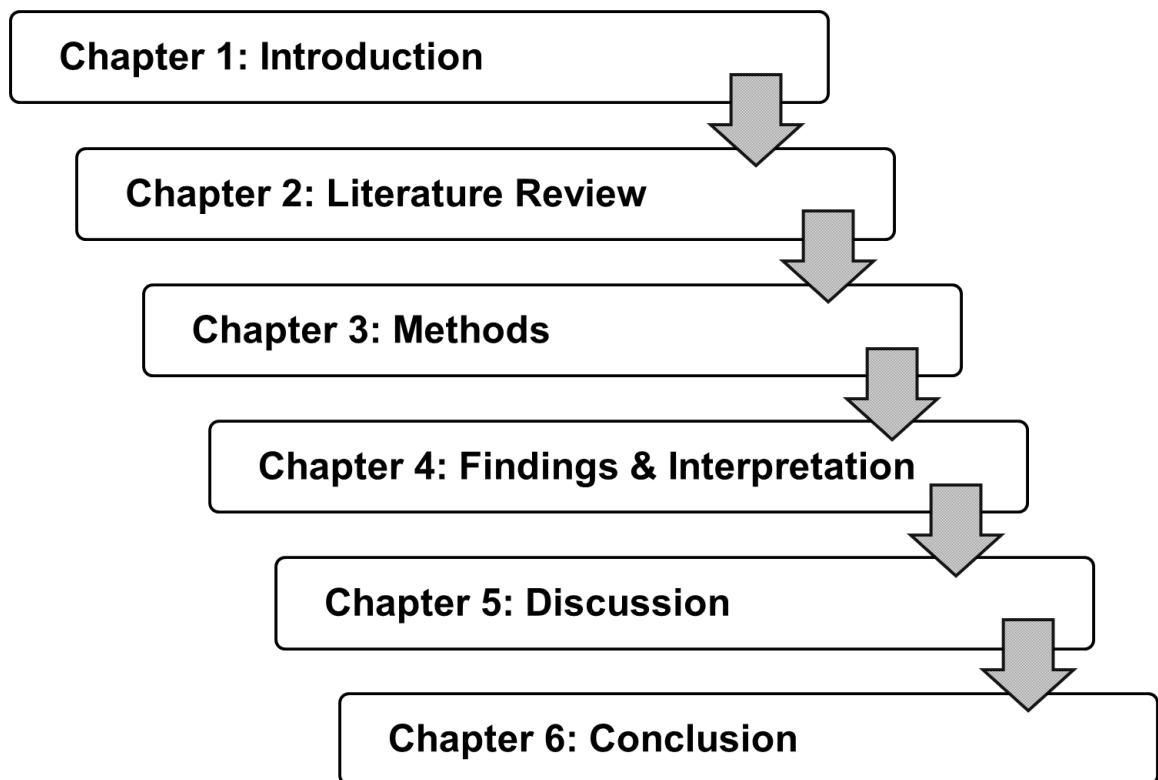
## **ORIGINALITY OF THE THESIS**

Over the last decade the incorporation of quality of life (QOL) and health-related quality of life (HRQOL) assessment in clinical practice and healthcare services has been highlighted to enhance patient health and healthcare utilisation as well as the efficacy healthcare programmes and interventions (Coghill et al., 2009; Varni et al., 2005). In paediatric clinical practice, the value of QOL and HRQOL measurement as broader psychosocial outcomes (Matza et al., 2004) and patient-reported outcomes (PROs) (Varni et al., 2005) has been acknowledged in terms of children's right to express their own views and participate in medical treatment and healthcare services (Cremins et al., 2006; United Nations Children's Fund, 2005), and cost-effectiveness of treatments (Coghill et al., 2009; Tilford et al., 2012). Consequently, QOL and HRQOL have been considered as "gold standard" of patients' outcomes and there has been an emerging body of literature on QOL and HRQOL instruments to accurately elicit patients' perspectives.

This research is unique in terms of the use of qualitative research approach, namely qualitative description, instead of quantitative research method to assess the content validity of QOL and HRQOL measures. A qualitative research approach is suitable for vulnerable populations such as children and adolescents with cognitive disability to obtain rich and valuable data and describe their perception and experience of their world. This approach is particularly important to identify the needs of children and adolescents with cognitive disability, which is different from other populations. The development of QOL and HRQOL measures using qualitative descriptive study design is a holistic approach to take account of not only individual characteristics (e.g. developmental change) but also environmental attributes (e.g. cultural issues).

## THESIS ORGANISATION

This thesis consists of six chapters (see Figure 1.1.). Chapter 2 is the critical review of the literature for the thesis on measurement of QOL in children and adolescents with autism spectrum disorders. The review aims to examine the appropriate QOL and HRQOL measurement for children and adolescents with ASDs and identify strengths and limitations of QOL and HRQOL instruments for use in this population. Chapter three presents the research methods that were utilised for the study. This chapter outlines the research design, sampling, data collection methods, procedure and data analysis that were employed. The development of the Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) 4.0 Generic Core Scales Child Self-Report will also be explicated in this chapter. Chapter four synthesises and interprets the key findings from the focus group discussions and interviews. Chapter five discusses and reflects on these key findings in relation to pertinent literature. Chapter six summarises these key findings and evaluate their significance in regards to its limitations and implications. Recommendations for further research are also provided.



*Figure 1.1.* Thesis structure.

## **CHAPTER 2: LITERATURE REVIEW**

### **Measurement of Quality of Life in Children and Adolescents with Autism Spectrum Disorders: A Critical Review**

#### **SUMMARY**

Measurement of quality of life (QOL) and health-related quality of life (HRQOL) is important in monitoring well-being in children and adolescents with autism spectrum disorders (ASDs). In this review, the aim is to investigate the characteristics of QOL and the appropriateness of QOL measures in children and adolescents with ASDs. MEDLINE, CINAHL Plus with Full Text and SPORTDiscus with Full Text via EBSCO Health Database, PsycINFO and ProQuest Health and Medicine (up to February 2013) databases were searched and articles were identified. The following inclusion criteria were used; articles which reported original research and measured QOL in children and adolescents with ASDs, and participants aged 5 to 18 years. Searches were limited to articles from peer-reviewed journals, in English, and those available in full-text. Additional limitations by document type (i.e. article) and subject (i.e. humans, child, adolescent, children and youth, autism, autistic disorder and Asperger syndrome) to the ProQuest Health and Medicine. The search identified 10166 titles. Thirty-two articles were selected for full review and 10 met the inclusion criteria. The review identified a number of QOL measures in children and adolescents with ASDs. The most commonly used instrument was the Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) 4.0 Generic Core Scales. Self-reports were uncommon and the reliability and validity of QOL measures were not sufficiently reported for this population. Large discrepancies were found between self-reports and proxy-reports. Despite the differences in study design and methodological quality, there was consistency in the results among studies, in that children and adolescents with ASDs provided lower QOL scores, particularly for social domains, compared to their healthy counterparts. From this review, future research

should focus on examining the appropriateness, reliability and validity of QOL instruments for use in children and adolescents with ASDs.

## INTRODUCTION

Autism spectrum disorders (ASDs) are high prevalent mental disorders of childhood and adolescence at the onset of an early age and impacts on the acquisition of developmental milestones (Filipek et al., 1999; Ozonoff, Goodlin-Jones, & Solomon, 2005). The ASDs refer to a wide variety of cognitive and neurobehavioral related disorders defined by three core symptoms: (1) impairment in social interactions, (2) impairment in verbal and nonverbal communication, and (3) restricted, repetitive and stereotyped patterns of behaviours, interests and activities (APA, 2000). According to the Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV) (APA, 2000), ASDs involves five possible diagnoses: Autistic disorder, Asperger disorder, childhood disintegrative disorder, Rett disorder, Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) and atypical autism, which are consistent with the International Classification of Disease, 10th edition (ICD-10) (World Health Organization [WHO], 2010). Individuals with ASDs often have psychiatric comorbidities such as attention deficit hyperactivity disorder (ADHD), anxiety disorder, mood disorders, and oppositional defiant and conduct disorders (Brereton, Tonge, & Einfeld, 2006; Kamp-Becker et al., 2011; Simonoff et al., 2008). Although the nature of ASDs symptoms alters with age, ASDs have lifelong detrimental impacts on physical, mental, social and academic development, which, in turn, affect well-being of individuals with ASDs (Kamp-Becker et al., 2011; Ozonoff et al., 2005; Rao, Beidel, & Murray, 2008). In children and adolescents with ASDs, psychosocial impairments such as social skills deficits (e.g. eye contact), impaired social competence (e.g. peer relationships) and limited social emotional reciprocity permeate all domains of social, emotional and academic functioning in the course of everyday life (Knott, Dunlop, & Mackay, 2006; Ozonoff et al., 2005; Rao et al., 2008).

Quality of life (QOL) is defined as, “an individual’s perception of their position in life, in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” (WHOQOL Group, 1995, p. 1405). The



concept of QOL is multidimensional framework in which physical, psychological and social components are influenced by personal characteristics and environmental variables (Cummins, 2005; Schalock, 2004). This concept is related to the definition of 'health' which refers to "a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity" (WHO, 1946, p. 1315). Health-related quality of life (HRQOL) is more specific to the impact and characteristics of a particular illness, disorder and medical treatment on the daily functioning and well-being of a patient (Dey et al., 2012; Drotar, 2004; Limbers, Heffer, & Varni, 2009). For children and adolescents, child development stages are also important factors of their HRQOL (Dey et al., 2012). QOL and HRQOL are measured by subjective personal appraisal as well as objective functional assessment, and regarded as "gold standard" instruments against which any other health outcomes should be assessed (Cummins, 2005; Eiser & Morse, 2001; Schalock, 2004). Subjective and objective measurement strategies encompass surveys, questionnaires, interviews and observations (Schalock, 2004). In a paediatric population, QOL and HRQOL are predominantly measured by individual self-appraisal using questionnaires although interviews and observations are occasionally integrated (Coghill et al., 2009; Davis et al., 2006; Eiser & Morse, 2001; Matza et al., 2004).

Several QOL and HRQOL generic instruments have been utilised to measure quality of life in children and adolescents with ASDs, such as the Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) (Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011), the World Health Organization Quality of Life Instrument, Abbreviated Version (WHOQOL-BREF) (Jennes-Coussens, Magill-Evans, & Koning, 2006; Kamp-Becker, Schroder, Remschmidt, & Bachmann, 2010), the Inventar zur Erfassung der Lebensqualität bei Kindern und Jugendlichen (ILK; Inventory for the Assessment of Quality of Life in Children and Adolescents) (Kamp-Becker et al., 2011), the Quality of Communication Life Scale (QCL) (Burgess & Turkstra, 2010), the Health Utilities Index 3 (HUI3) (Tilford et al., 2012) and the Quality of Well-Being Self-Administered Scale (QWB-SA) (Tilford et al., 2012).

Based on their conceptual frameworks, three measures (PedsQL, WHOQOL-BREF and ILK) were designed for measuring health outcomes based on a multidimensional construct (Jennes-Coussens et al., 2006; Kamp-Becker et al., 2011; Kamp-Becker et al.,

2010; Kuhlthau et al., 2010; Lee, Harrington, Louie, & Newschaffer, 2008; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011). Two measures (HUI3 and QWB-SA) were formulated to evaluate health economics such as cost-effectiveness (Tilford et al., 2012). One measure (QCL) was developed to assess the effects of communication disorders on communication QOL (Burgess & Turkstra, 2010). Differences in the purpose of QOL measurement designs imply that QOL outcomes obtained by one measure may be less correlated with those attained by another measure. There is a need for a standard QOL and HRQOL instrument for use with children and adolescents with ASDs.

QOL and HRQOL instruments for children and adolescents are mainly categorised into two versions: child self-report and parent proxy-report. Considering that the QOL and HRQOL construct is intrinsically subjective in nature and refers to an individual's perceptions and appraisal of their life, it is assumed that QOL and HRQOL should preferably be assessed from a child's perspective (Coghill et al., 2009; Ravens-Sieberer et al., 2006). However, there are general issues regarding the use of self-reports and specifically in relation to children with mental health problems. Age is one of the factors that influence a child's ability to provide an empirically reliable self-report on QOL and HRQOL (Bell, 2007; Coghill et al., 2009; Matza et al., 2004). The age of a child with mental health conditions is strongly related to child developmental stages, specifically cognitive and language development (Coghill et al., 2009; Varni et al., 2005). Matza et al. (2004) reported that the level of language comprehension improved with a child's age. For example, children at the age of five are able to understand concrete concepts such as pain but may not be able to interpret subjective or abstract health-related notions such as emotional impact of illness (Coghill et al., 2009; Matza et al., 2004). Children and adolescents with psychiatric disorders have lower developmental age than their chronological age, and more difficulty to complete the age-matched self-reports (Coghill et al., 2009). This is not only because of their language impairments but also difficulties with memory and recall (Coghill et al., 2009). For children and adolescents with mental health problems, disorder-specific symptoms and impairments also affect QOL and HRQOL assessment. For instance, children with autism may have problems in reporting on peer relationships due to their impaired social competence (Danckaerts et al., 2010). In this regard, parent proxy-reports provide

supplemental information on a child's QOL and HRQOL in a comprehensive and integrative manner (Danckaerts et al., 2010; Matza et al., 2004). Still, it needs to be considered that several studies have reported that there are medium to large effect sizes between child self-reports and parent proxy-reports in children and adolescents with psychiatric disorders (Danckaerts et al., 2010; Kamp-Becker et al., 2011; Sheldrick et al., 2011; Varni & Burwinkle, 2006).

Empirical evidence on QOL and HRQOL in children and adolescents with ASDs may contribute to their well-being within diverse contexts of the family, school and community. It may also support and assist in developing strategies for health service planning. Ozonoff et al. (2005) claimed that there were no QOL and HRQOL measures that were designed for children and adolescents with ASDs. Moreover, no reviews were found comparing QOL and HRQOL measures and outcomes from the instruments in children and adolescents with ASDs across studies. Thus, the aims of this review were (1) to critically review studies that measured QOL in children and adolescents with ASDs; (2) to examine the appropriate measurement of QOL and HRQOL for children and adolescents with ASDs; and (3) to identify strengths and limitations of QOL and HRQOL measures in the existing literature for use in children and adolescents with ASDs.

## **METHODS**

### **Search**

A literature searches was conducted using MEDLINE, CINAHL Plus with Full Text and SPORTDiscus with Full Text via EBSCO Health Database, PsycINFO and ProQuest Health and Medicine (from 1990 up to February 2013). Any studies that measured QOL or HRQOL in children and youth with autism spectrum disorders were searched through the databases. Searches were conducted using the following key terms: (child\* OR 'youth' OR adolescen\* OR kid\* OR teenager\*) AND (autis\* OR Kanner\* OR Asperger\*) AND ('quality of life' OR well\*). Searches were limited to articles written in 'English', 'Full-text' or 'PDF Full-text', 'peer reviewed journal', studies with children and adolescents between 5 and 18 years of age. Additional limitations were applied in the ProQuest Health and Medicine database: document type

of ‘article’, subject of ‘humans’, ‘child’, ‘adolescent’, ‘children and youth’, ‘autism’, ‘autistic disorder’ and ‘Asperger syndrome’. Reference lists of relevant articles that were not captured by the search were consulted for additional materials.

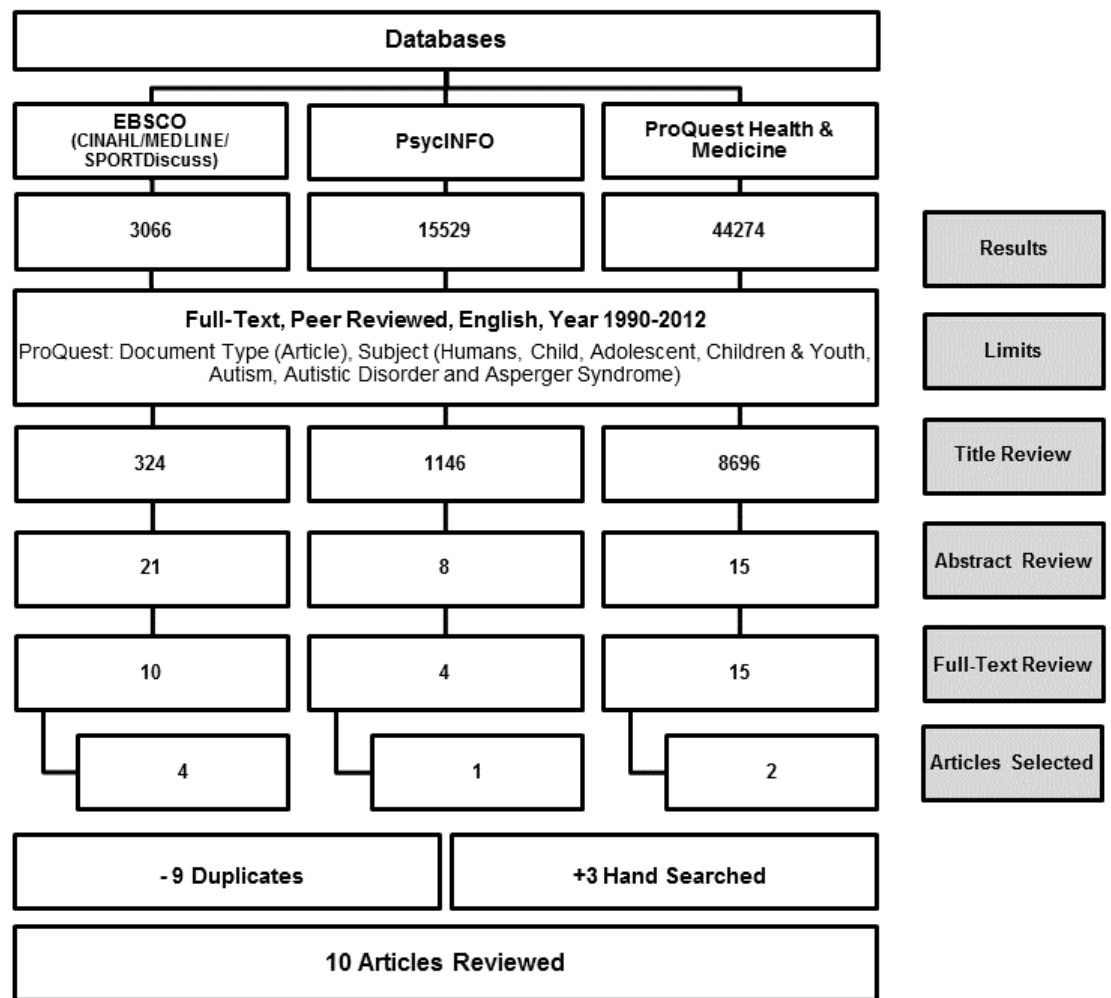
### **Study Selection**

All titles and abstracts were screened and evaluated for eligibility by the student (E.I.). The student and supervisor (E.H. and E.I.) fully reviewed articles and independently assessed full-texts of eligible studies. Disagreements were reconciled by discussion. Studies were included for review if they met the following criteria: QOL or HRQOL in children or adolescents with ASDs were measured and reported. Articles were excluded if at least one of the following exclusion criteria was met: (1) QOL or HRQOL of parents of children with ASDs were reported; (2) majority of participants were at an age other than 5 to 18 years; (3) studies focused on children or adolescents with attention deficit hyperactivity disorder (ADHD) or other mental disorders; (4) only functional assessments of social competence, social skills, relationships, friendship or emotional recognition and understanding were reported; (5) symptoms of ASDs, sleep problems, anxiety or depression were only measured as QOL or HRQOL outcomes.

## **RESULTS**

### **Study Selection**

The search process of study selection is outlined in Figure 2.1. The first search step identified 62,869 articles. After limiting to full-text, peer reviewed journals, language of English and published year between 1990 and 2012, 18,379 titles remained. Additional limitations were applied to the ProQuest Health and Medicine by document type (i.e. article) and subject (i.e. humans, child, adolescent, children and youth, autism, autistic disorder and Asperger syndrome), and 10,166 titles left. Based on the titles and abstracts of these articles, 10,137 articles were discarded. The second search step of reference lists resulted in an additional three articles. These three articles and those 29 articles identified in the databases were retained for full review, and 10 articles met the inclusion criteria.



**Figure 2.1.** Flow diagram of the article selection process. – removed; + added.

## Data Extraction

Characteristics of included studies were extracted in the following categories: source, methods (design), participants (age, gender and disability), measure (QOL or HRQOL and diagnosis), outcomes and study conclusions. Main attributes are summarised and presented in Table 2.1. The table was arranged alphabetically by first author and when more than one study was conducted by the same author, chronologically.

**Table 2.1**

*Summary of Included Studies Assessing Quality of Life and Health-Related Quality of Life in Children and Adolescents with ASDs*

Source	Participants; mean age $\pm$ SD or range (years)	QOL measure	Rater	Diagnostic measure	Main outcomes
Burgess and Turkstra (2010)	14 adolescents with AS (14 boys); 16.87 $\pm$ 1.93. 15 typically developing adolescents (15 boys); 15.88 $\pm$ 2.06.	QCL	Child, parent		The total QCL ratings in the AS group was significantly lower than those in the typically developing (TD) group ( $p = .01$ ). The effect size between the AS and the TD groups was large and considered as clinically meaningful. The QCL total scores were higher in the AS self-report than in AS proxy-report ( $p = .07$ ).
Jennes-Coussens, Magill-Evans, and Koning (2006)	12 adolescents with AS (12 boys); 20.3 $\pm$ 1.3. 13 healthy adolescents (13 boys); 20.5 $\pm$ 1.3.	WHOQOL-BREF	Child		The overall QOL and the Psychological, Social and Environment domain scores in adolescents with AS were lower than those in healthy adolescents ( $ES = .45$ ). The scores on Social ( $p = .002$ ) and Physical Health ( $p = .006$ ) in adolescents with AS were significantly lower than those in healthy adolescents.
Kamp-Becker et al. (2011)	42 children with AS, HFA or AA (39 boys, 3 girls); 12.7 $\pm$ 2.6.	ILK	Child, parent	ADOS-G, VABS	Compared the mean LQ 0-28 <sup>b</sup> scores of the self-report and proxy-report to a standard sample, the self-report score was at 47th (SD $\pm$ 31.4) percentile and the proxy-report score was at 33rd (SD $\pm$ 27.6) percentile. Compared with a psychiatric sample, the self-report score was at 69th (SD $\pm$ 28.0) percentile and the proxy-report score was at 67th (SD $\pm$ 29.9) percentile. There were no significant associations between the HRQOL and the ADOS summary score, and between the HRQOL and the VABS scores.

Kamp-Becker, Schroder, Renschmidt, and Bachmann (2010)	26 adolescents and young adults with AS, HFA or AA (26 boys); $21.6 \pm 3.4$ .	WHOQOL-BREF	Child	ADOS-G, VABS	The Social domain was the most impaired and had the lowest QOL score. Compared the mean QOL scores of patients with ASDs to a reference healthy sample, the scores were significantly lower in the Physical, Psychological and Social domains and the overall score ( $p < .001$ ). Compared with a reference sample with SSD, the scores were significantly higher in the Physical, Psychological and Environment domains. A significant correlation were found between the VABS domain of "daily living skills" and the QOL Physical ( $p = .039$ ), Psychological ( $p = .030$ ) and the summary ( $p = .012$ ) scores. The VABS domain of "daily living skills" ( $p = .002$ ) and the ADOS-G summary score ( $p = .028$ ) were optimum predictors of the total QOL score.
Kuhlthau et al. (2010)	286 children with AD, AS or PDD-NOS (236 boys, 50 girls); 2-18.	PedsQL 4.0 Generic Core Scales	Parent	Vineland-II, SRS, RBS-R, CBCL	The total score for children with ASDs was significantly lower than the national norm for a healthy sample and for a chronic health condition sample ( $p < .001$ ). Differences of the scores on Social Functioning between children with ASDs and with chronic condition, and between ASDs children and healthy children were significant. A significant association between the Social Functioning scores and the Vineland-II Adaptive Behaviour Composite scores was found. There were consistent negative correlations between the PedsQL scores and the SRS scores, and between all PedsQL except School Functioning scores and the RBS-R scores. The CBCL scores were negatively correlated with the PedsQL scores.

Lee, Harrington, Louie, and Newschaffer (2008)	483 children with autism (372 boys, 111 girls); 3-17. 6319 children with ADD or ADHD (4549 boys, 1769 girls) <sup>a</sup> ; 3-17. 58953 children without disabilities or mental conditions (28264 boys, 30632 girls) <sup>a</sup> 3-17.	<i>Quality of Life</i>	Parent		Children aged 3-5 with autism were less likely to attend a religious service than the unaffected control group ( $p < .05$ ). Compared to the two comparison groups, the autism group aged 6-11 were more likely to repeat a grade and miss school for more than a week ( $p < .05$ ), and less likely to attend a religious service and participate in activities/events ( $p < .05$ ). Compared with the unaffected control group, the autism group aged 12-17 were more likely to repeat a grade and miss more than a week of school ( $p < .05$ ), and less likely to attend a religious service, take participate in activities/events and community service ( $p < .05$ ).
Limbers, Heffer, and Varni (2009)	22 children with AS (20 boys, 2 girls); $9.25 \pm 2.15$ .	PedsQL 4.0 Generic Core Scales	Parent	PedsQL™ Cognitive Functioning Scale, ASDS	The scores of all HRQOL scales, except the Missed School <sup>c</sup> , in children with AS were significantly lower than those in healthy children ( $p < .001$ ). Significant differences between healthy children and children with AS were found on the Social Functioning. The Cognitive Functioning was significantly correlated with the Psychosocial Health Summary Score <sup>c</sup> (ES = .77) and the School- Related Cognitive Functioning <sup>c</sup> (ES = .76). The Psychosocial Health Summary Score <sup>c</sup> (ES = .59) and the School-Related Cognitive Functioning (ES = .50) were significantly correlated with the ASDS Maladaptive Scale. The effect size between the Social Functioning and the ASDS Social Scale was medium (ES = .30).



Sheldrick, Neger, Shipman, and Perrin (2011)	39 adolescents with ASDs (AD, PDD or AS) (73% boys, 27% girls); $14.8 \pm 1.9$ .	PedsQL 4.0 Generic Core Scales	Child, parent		The self-report scores were significantly correlated with the parent report scores on Physical Functioning ( $p < .01$ ), School Functioning ( $p < .05$ ) and the total score ( $p < .05$ ); and with the proxy-report scores on all domains ( $p < .01$ ; Emotional Functioning, $p < .05$ ) and the total score ( $p < .01$ ). Correlations between the self-report and the proxy-report were significant on the total score ( $p < .01$ ) and Social ( $p < .01$ ) and School ( $p < .05$ ) Functioning. Differences between the self-report scores and the parent report scores were significant for the total score ( $p < .01$ ) and Emotional ( $p < .01$ ), Social ( $p < .01$ ) and School ( $p < .05$ ) Functioning; and between the self-report scores and the proxy-report scores were significant for the total score ( $p < .05$ ) and Emotional ( $p < .05$ ) and Social ( $p < .01$ ) Functioning.
Shipman, Sheldrick, and Perrin (2011)	39 adolescents with ASDs (AD, PDD or AS) (73% boys, 27% girls); $14.8 \pm 1.9$ .	PedsQL 4.0 Generic Core Scales	Child, parent	RSES, SMFQ, SCARED	All scores of the self-report were below the normative mean ( $p < .05$ ). The scores of the self-report were higher than those of the proxy-report. Moderate to large positive correlations with the RSES and moderate to large negative correlations with the SCARED and the SMFQ were reported.

Tilford et al. (2012)	150 children with AD, PDD-NOS or AS (128 boys, 22 girls); $8.6 \pm 3.3$ .	HUI3, QWB- SA	Parent	ADOS, Vineland- II	Speech problems were the highest percentage of problem responses in both HUI3 and QWB-SA. Problems in Cognition, Emotion and Pain domains were found in the HUI3. The percentage of children who had "confusion and memory loss" on the QWB-SA was high. The HUI3 summary score was significantly correlated with all Vineland-II domain scores and the composite score ( $\rho = .373 - .552$ ; $p < .001$ ). The QWB-SA overall score was significantly correlated with the Vineland-II composite and all domains, except Motor Skill, scores ( $\rho = .212 - .248$ ; $p < .001$ ).
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*Note.* AS = Asperger syndrome. HFA = High-functioning autism. AA = Atypical autism. AD = Autistic disorder. PDD-NOS = Pervasive developmental disorder-not otherwise specified. ADD = Attention deficit disorder. ADHD = Attention deficit hyperactivity disorder. PDD = Pervasive developmental disorder. ASDs = Autism spectrum disorders. SSD = Schizophrenia-spectrum disorders. QCL = Quality of Communication Life Scale. WHOQOL-BREF = World Health Organization Quality of Life Instrument, Abbreviated Version. ILK = Inventar zur Erfassung der Lebensqualität bei Kindern und Jugendlichen (Inventory for the Assessment of Quality of Life in Children and Adolescents). PedsQL = Pediatric Quality of Life Inventory<sup>TM</sup>. HUI3 = Health Utilities Index 3. QWB-SA = Quality of Well-Being Self-Administered Scale. ADOS-G = Autism Diagnostic Observatory Schedule-Generic. VABS = Vineland Adaptive Behaviour Scales. Vineland-II = Vineland Adaptive Behaviour Scales, Second Edition, Survey Interview Form. SRS = Social Responsiveness Scale. RBS-R = Repetitive Behaviour Scale-Revised. CBCL = Achenbach Child Behaviour Checklist. ASDS = Asperger Syndrome Diagnostic Scale. RSES = Rosenberg Self-Esteem Scale. SMFQ = Short Mood and Feelings Questionnaire. SCARED = Screen for Child Anxiety Related Emotional Disorders. ADOS = Autism Diagnostic Observation Schedule. SD = standard deviation. ES = effect size (Cohen's *d*). <sup>a</sup>Numbers might not add up to the total number due to missing values. <sup>b</sup>LQ 0-28 score shows the patients' HRQOL level. <sup>c</sup>Psychosocial Health Summary Score includes Emotional Functioning and Social Functioning scales, and School Functioning Summary Score involves School-Related Cognitive Functioning and Missed School scales.

## Coding of Results

The age of participants in each study was determined from the reported mean or range. Results of studies for children and adolescents were synthesised due to the small number of studies. In two studies (Kuhlthau et al., 2010; Lee et al., 2008), a small percentage of children were of ages between two and four years, and the other two studies (Jennes-Coussens et al., 2006; Kamp-Becker et al., 2010) involved participants aged between 19 and 28 years. These studies were included in this review. The review also included two consecutive studies in which two different designs of reliability and validity studies of self-completed QOL were conducted (Sheldrick et al., 2011; Shipman et al., 2011). Demographic data from participants and the mean HRQOL scores from self- and proxy-reports were duplicated between these studies. In the included studies, approaches to assess QOL or HRQOL in children and adolescents with ASDs incorporated child self-reports, parent proxy-reports and parent reports.

## Synthesis of Results

### *Type of instruments for QOL and HRQOL in children and adolescents with ASDs*

A summary of QOL and HRQOL measurements that were used in the included studies is presented in Table 2.2. All 10 studies utilised questionnaires as a measure of QOL or HRQOL in children and adolescents with ASDs. Four studies (Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011) used the Pediatric Quality of Life Inventory<sup>TM</sup> (PedsQL) 4.0 Generic Core Scales, two studies (Jennes-Coussens et al., 2006; Kamp-Becker et al., 2010) used the World Health Organization Quality of Life Instrument, Abbreviated Version (WHOQOL-BREF) and the other four studies used the Inventar zur Erfassung der Lebensqualität bei Kindern und Jugendlichen (ILK; Inventory for the Assessment of Quality of Life in Children and Adolescents) (Kamp-Becker et al., 2011), the Quality of Communication Life Scale (QCL) (Burgess & Turkstra, 2010), the *Quality of Life* (Lee et al., 2008), and the Health Utilities Index 3 (HUI3) and the Quality of Well-Being Self-Administered Scale (QWB-SA) (Tilford et al., 2012). The *Quality of Life* is a selection of items from the National Survey of Children's Health in the US which are considered relevant to 'QOL' by Lee et al. (2008). Five out of seven QOL or HRQOL measures (PedsQL, WHOQOL-BREF,

ILK, *Quality of Life* and HUI3) specified the age for use, and three of them (PedsQL, ILK and *Quality of Life*) were designed for children and adolescents, namely child age between 5 to 18 years. The number of items varied from 5 to 71. Some of the items are integrated into multidimensional domains. The domains ranged from four to eight including physical (PedsQL, WHOQOL-BREF, ILK, HUI3 and QWB-SA), emotional / mental / psychological (PedsQL, WHOQOL-BREF, ILK and HUI3), social (PedsQL, WHOQOL-BREF, ILK and QWB-SA), school (PedsQL and ILK), and communication / speech (QCL and HUI3). In terms of psychometric properties, the PedsQL was the only one in which both reliability and validity were established in children and adolescents with ASDs. Five measures (WHOQOL-BREF, ILK, QCL, HUI3 and QWB-SA) determined only validity in which three of them reported reliability from the other studies. Neither reliability nor validity information was provided in the *Quality of Life* (Lee et al., 2008). The types of validity involved construct-related validity including discriminant and convergent validity, and criterion-related validity such as concurrent validity. Reliability was classified into internal consistency and reproducibility including test-retest reliability and inter-rater agreement.

**Table 2.2**

*Generic Quality of Life and Health-Related Quality of Life Instruments Included in the Review*

Measure	References	Report	Child age (year)	Item (no.)	Domain (no.)	Domain (name)	Reliability	Validity <sup>k</sup>	
Pediatric Quality of Life Inventory (PedsQL) 4.0 Generic Core Scale	Kuhlthau et al. (2010), Limbers et al. (2009), Sheldrick et al. (2011), and Shipman et al. (2011)	Self	5-18 <sup>c</sup>	23	4	Physical Functioning <sup>g</sup>	Internal consistency	Discriminant	
		Proxy <sup>b</sup>	2-18 <sup>d</sup>			Emotional Functioning <sup>g</sup>	Inter-rater <sup>j</sup>	Convergent	
		Parent <sup>b</sup>				Social Functioning <sup>g</sup>		Concurrent	
						School Functioning <sup>g</sup>			
World Health Organization Quality of Life Instrument, Abbreviated Version (WHOQOL-BREF)	Jennes-Coussens et al. (2006) and Kamp-Becker et al. (2010)	Self	≥ 18 <sup>e</sup>	24	4	Physical Health	Internal consistency <sup>j</sup>	Discriminant	
						Psychological Health	Test-retest <sup>j</sup>	Convergent	
						Social Relationships		Concurrent	
					Environment				
Inventory for the Assessment of Quality of Life in Children and Adolescents (ILK) <sup>a</sup>	Kamp-Becker et al. (2011)	Self	6-18	9	6	School	Internal consistency <sup>j</sup>	Discriminant	
		Proxy	6-18	11		Family	Test-retest <sup>j</sup>		
							Social Relations with Peers		
							Hobbies and Recreational Activities		
							Physical Health		
				Mental Health					

Quality of Communication Life Scale (QCL)	Burgess and Turkstra (2010)	Self Proxy		18		Total Score <sup>h</sup> Summary Item <sup>h</sup>	Inter-rater <sup>j</sup>	Discriminant
<i>Quality of Life</i>	Lee et al. (2008)	Parent	3-5 6-11 12-17	5 7 7				
Health Utilities Index 3 (HUI3)	Tilford et al. (2012)	Proxy	≥ 5	45	8	Vision Hearing Speech Ambulation Dexterity Cognition Emotion Pain		Discriminant Convergent
Quality of Well-Being Self-Administered Scale (QWB-SA)	Tilford et al. (2012)	Proxy		71 <sup>f</sup>	4	Mobility <sup>i</sup> Physical Activity <sup>i</sup> Social Activity <sup>i</sup> Symptoms (CPX)		Discriminant Convergent

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*Note.* <sup>a</sup>German version. <sup>b</sup>Proxy and parent versions use the same form. <sup>c</sup>Ages 5-7 (young child), 8-12 (child) and 13-18 (teen). <sup>d</sup>Ages 2-4 (toddlers), 5-7 (young children), 8-12 (children) and 13-18 (teens). <sup>e</sup>Age-specific values from a reference sample are available for an age range between 18 and 85 years for the German version of the WHOQOL-BREF. <sup>f</sup>Two conditions (sexuality and hangovers) in the CPX scale were excluded in the study. <sup>g</sup>Summary scores include Total Scale Score (23 items), Physical Health Summary Score (Physical Functioning; 8 items), Psychosocial Health Summary Score (Emotional, Social, School Functioning; 15 items). <sup>h</sup>Total Score involves 17 items and Summary Item is a rating of overall QOL, "In general, my quality of life is good". <sup>i</sup>Functioning Scale. <sup>j</sup>The reliability of a measure was not determined in children and adolescents with ASDs but in other population. <sup>k</sup>Discriminant validity and convergent validity are categories of construct-related validity; and concurrent validity is a type of criterion-related validity.

*Agreement between Child self-reports and Parent Ratings on QOL and HRQOL*

Six studies (Burgess & Turkstra, 2010; Jennes-Coussens et al., 2006; Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Sheldrick et al., 2011; Shipman et al., 2011) used self-reports, seven studies (Burgess & Turkstra, 2010; Kamp-Becker et al., 2011; Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011; Tilford et al., 2012) applied proxy-reports, and two studies (Lee et al., 2008; Sheldrick et al., 2011) employed parent reports. One study (Sheldrick et al., 2011) investigated correlations between adolescent self-reports and parent proxy-reports as well as standard parent reports. In this study, the same form was applied to proxy and parent versions. Parents were asked “how much of a problem has your teen had with...” for the standard parent report, and “pretending that you are your son or daughter, answer the following questions as you think they are answering them” for the parent proxy-report (Sheldrick et al., 2011). For the total PedsQL score, correlations between self-reports and standard parent reports ( $r = .40, p < .05$ ) were lower than those between self-reports and parent proxy-reports ( $r = .58, p < .01$ ) (Sheldrick et al., 2011). Differences between child self-ratings and standard parent and parent proxy ratings were large in the domains of Emotional and Social Functioning (Sheldrick et al., 2011). Three studies (Burgess & Turkstra, 2010; Kamp-Becker et al., 2011; Shipman et al., 2011) compared child self-reports and parent proxy-reports. In the ILK, the correlation between self- and proxy-reported HRQOL was medium ( $r = .45$ ), and the total LQ 0-28 scores between self- and proxy-reports were significantly different ( $p \leq .000$ ) (Kamp-Becker et al., 2011). The total scores of quality of communication life and the summary item score of QOL in adolescents with Asperger syndrome were higher than those obtained from mother proxy-reports (Burgess & Turkstra, 2010). Nevertheless, the patterns of ratings from self- and proxy-reports were comparable (Burgess & Turkstra, 2010). Similarly, the scores of the PedsQL Emotional, Social and School Functioning scales in adolescents with ASDs were higher than the parent proxy scores although the Physical Functioning scores were lower in the adolescents (Shipman et al., 2011). These consistent findings indicated that discrepancies between self-reports and parent proxy-reports were large with regard to QOL and HRQOL ratings.



*Comparison of QOL and HRQOL between ASDs and Healthy or Clinical Samples*

In nine studies (Burgess & Turkstra, 2010; Jennes-Coussens et al., 2006; Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Lee et al., 2008; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011), QOL and HRQOL in children adolescents with ASDs were compared with those in a sample classified as healthy. Four of these studies (Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Lee et al., 2008) also examined differences in QOL and HRQOL between children and adolescents with ASDs and clinical samples including psychiatric disorders (Kamp-Becker et al., 2011), schizophrenia-spectrum disorder (Kamp-Becker et al., 2010), chronic health conditions (Kuhlthau et al., 2010), and attention deficit disorder (ADD) and attention deficit hyperactivity disorder (ADHD) (Lee et al., 2008). Four studies consistently found that total scores and all domains scores of the PedsQL in children and adolescents with ASDs were lower than those in an age-matched healthy sample, in which two studies used both self- and proxy-reports (Sheldrick et al., 2011; Shipman et al., 2011) and the others used only proxy-reports (Kuhlthau et al., 2010; Limbers et al., 2009). In particular, the domain with the greatest difference between ASDs and healthy samples in proxy-reports was Social Functioning ( $p < .01$ ) (Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011). Likewise, two studies with WHOQOL-BREF self-reports revealed that adolescents with Asperger syndrome had lower scores on the overall and all domains compared to healthy individuals, and substantial differences were found in the domain of Social Relationships (Jennes-Coussens et al., 2006; Kamp-Becker et al., 2010). Other three studies showed that QOL and HRQOL in children and adolescents with ASDs were worse compared to a healthy sample (Burgess & Turkstra, 2010; Kamp-Becker et al., 2011; Lee et al., 2008).

In comparison with chronic health condition normative populations, the proxy PedsQL scores were significantly lower on the total and Emotional and Social Functioning scales ( $p < .001$ ), but similar on Physical and School Functioning scales (Kuhlthau et al., 2010). Parents reported that children with ASDs, specifically at the age between 6 and 11 years, had more problems with school life and activity participation compared to children with ADD or ADHD (Lee et al., 2008). Compared with a psychiatric sample, however, self-reported HRQOL was better in children and adolescents with ASDs

(Kamp-Becker et al., 2011). In the WHOQOL-BREF self-reports, adolescents with ASDs had higher scores on Physical Health, Psychological Health and Environment domains, and similar scores on Social Relationships compared to schizophrenia-spectrum disorder patients (Kamp-Becker et al., 2010).

*Relationships between Subjective QOL and HRQOL Measures and Objective ASDs-Specific Measures*

Six studies investigated correlations between QOL or HRQOL and ASDs-specific symptoms and functional abilities (Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Limbers et al., 2009; Shipman et al., 2011; Tilford et al., 2012). Measures of ASDs-specific symptoms were classified into diagnostic scales (Autism Diagnostic Observation Schedule [ADOS], Autism Diagnostic Observatory Schedule-Generic [ADOS-G] and Asperger Syndrome Diagnostic Scale [ASDS]) and behaviour scales (Vineland Adaptive Behaviour Scales [VABS], Vineland Adaptive Behavior Scales, Second Edition, Survey Interview Form [Vineland-II], Repetitive Behavior Scale-Revised [RBS-R] and Achenbach Child Behavior Checklist [CBCL]). There were no significant correlations between autism diagnostic scales (ADOS and ADOS-G) and QOL or HRQOL measures (HUI3, ILK, QWB-SA, and WHOQOL-BREF) (Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Tilford et al., 2012). The PedsQL Physical Health Summary Score showed a medium correlation with the ASDS Sensorimotor Scale (effect size [ES] = .35), and the PedsQL Psychosocial Health Summary Score (ES = -.59) and School-Related Cognitive Functioning (ES = -.50) were significantly associated with the ASDS Maladaptive Scale (Limbers et al., 2009). Some significant correlations between QOL or HRQOL measures (HUI3, ILK, PedsQL, QWB-SA and WHOQOL-BREF) and behaviour scales (VABS, Vineland-II, CBCL and RBS-R) were found. The Vineland-II Composite Score was significantly correlated with the PedsQL Social Functioning ( $\beta = .34, p < .001$ ), HUI3 ( $p = .52, p < .001$ ) and QWB-SA ( $p = .25, p < .001$ ) (Kuhlthau et al., 2010; Tilford et al., 2012). The VABS Daily Living Skills was significantly associated with the WHOQOL-BREF Physical Health ( $r = .43, p = .04$ ), Psychological Health ( $r = .45, p = .03$ ) and overall score ( $r = .52, p = .01$ ) (Kamp-Becker et al., 2010). However, there were no significant correlations between VABS and ILK (Kamp-Becker et al., 2011). The PedsQL scales had

significantly negative correlations with the CBCL ( $p < .01$ ) and RBS-R ( $p < .01$ ) scales (Kuhlthau et al., 2010).

Instruments for assessing functional impairments related to ASDs involved cognitive scale (PedsQL Cognitive Functioning Scale), emotional scales (Rosenberg Self-Esteem Scale [RSES], Short Mood and Feelings Questionnaire [SMFQ] and Screen for Child Anxiety Related Emotional Disorders [SCARED]), and social scale (Social Responsiveness Scale [SRS]). The PedsQL Cognitive Functioning Scale was significantly correlated with the self-rated PedsQL total score ( $ES = .61, p < .05$ ), Psychosocial Health ( $ES = .77, p < .001$ ), Social Functioning ( $ES = .53, p < .05$ ), School Functioning ( $ES = .75, p < .01$ ) and School-Related Cognitive Functioning ( $ES = .76, p < .01$ ) (Limbers et al., 2009). The self-reported PedsQL on all domains were also significantly correlated with the RSES, SMFQ and SCARED (Shipman et al., 2011). A positively moderate to large association with RSES ( $r = .36$  to  $.74$ ) and negatively moderate to large correlations with SCARED ( $r = -.35$  to  $-.69$ ) and SMFQ ( $r = -.45$  to  $-.72$ ) were found (Shipman et al., 2011). All domains of the proxy-reported PedsQL were negatively correlated with the SRS, particularly with the largest coefficients for the total, Psychosocial and Social scales (Kuhlthau et al., 2010).

## DISCUSSION

The review found that QOL and HRQOL in children and adolescents with ASDs was significantly poorer than the general paediatric population (Burgess & Turkstra, 2010; Jennes-Coussens et al., 2006; Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Lee et al., 2008; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011). Particularly, children and adolescents with ASDs and their parents perceived that social domains (e.g. social support, personal relationships and peer relationships) were the most problematic (Jennes-Coussens et al., 2006; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011). Although six out of 10 studies (Burgess & Turkstra, 2010; Jennes-Coussens et al., 2006; Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Sheldrick et al., 2011; Shipman et al., 2011) used child self-reports, eight out of 10 studies utilised parent ratings to measure QOL and HRQOL in children and adolescents

with ASDs (Burgess & Turkstra, 2010; Kamp-Becker et al., 2011; Kuhlthau et al., 2010; Lee et al., 2008; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011; Tilford et al., 2012). However, there was consistent evidence across studies that self-reporting QOL and HRQOL in children and adolescents with ASDs were significantly different from parent proxy-reporting QOL and HRQOL (Burgess & Turkstra, 2010; Kamp-Becker et al., 2011; Sheldrick et al., 2011; Shipman et al., 2011). The PedsQL which involved both self- and proxy-reports and established psychometric properties was the most commonly used (Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011). In this section, the appropriateness and limitations of the QOL and HRQOL instruments in children and adolescents with ASDs are discussed. Implications and recommendations for future QOL research are also provided.

#### *Appropriate Measurement of QOL and HRQOL in Children and Adolescents with ASDs*

Considering the definition of QOL, three instruments (PedsQL, WHOQOL-BREF and ILK) embrace the most common domains of QOL and HRQOL which refer to physical, mental / emotional / psychological and social functioning (Cummins, 2005; Schalock, 2004; WHOQOL Group, 1995). The other measures contained at least one of the domains, which implied that there was inconsistency across the instruments with regard to the domains of QOL and HRQOL. Children and adolescents with ASDs scored lower than those without ASDs on the social domain measured by the PedsQL and WHOQOL-BREF (Jennes-Coussens et al., 2006; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Limbers et al., 2009; Sheldrick et al., 2011; Shipman et al., 2011). Considering that every social context such as the family, peer groups, school and community are likely to contribute to children's life (Matza et al., 2004), it is also critical to assess QOL and HRQOL of children and adolescents with ASDs within their relevant contexts. Due to the fact that QOL and HRQOL measures consist of a multidimensional construct and different scales and items, it is difficult to directly compare QOL and HRQOL across the instruments (Coghill et al., 2009).

The value of self-completed reports of QOL and HRQOL in children and adolescents with mental health conditions was highlighted in several studies (Coghill et al., 2009; Danckaerts et al., 2010; Dey et al., 2012; Matza et al., 2004). This trend originated from two main aspects: the importance of children's rights to reflect their own perspectives

on medical treatment (Coghill et al., 2009; Cromeens et al., 2006; United Nations Children's Fund [UNICEF], 2005) and considerable disagreement between child self-reports and parent proxy-reports on QOL and HRQOL (Coghill et al., 2009; Danckaerts et al., 2010; Matza et al., 2004). The findings from this review showed that children and adolescents with ASDs were likely to have different perspectives of QOL and HRQOL from their parents, which can be captured by only self-reports. (Burgess & Turkstra, 2010; Kamp-Becker et al., 2011; Sheldrick et al., 2011; Shipman et al., 2011). In the included studies, four measures (PedsQL, WHOQOL-BREF, ILK and QCL) incorporated child self-reports, and two of them (PedsQL and ILK) were specifically designed for children and adolescents for ages between 5 and 18 years. In terms of a child-appropriate QOL and HRQOL measure, it is critical that the questionnaire is formulated and validated for children and adolescents depending on their age, disorder and developmental stages (Coghill et al., 2009; Matza et al., 2004). However, all studies utilised generic QOL and HRQOL measures for children and adolescents with ASDs, and there were no QOL and HRQOL measures that were specifically developed for this population.

The review identified that almost all QOL and HRQOL measures failed to establish psychometric properties for children and adolescents with ASDs (see Table 2.2). According to Lohr (2002), QOL and HRQOL instruments are required to establish reliability (i.e. internal consistency and reproducibility including test-retest reliability and inter-rater agreement) and validity (i.e. content-related, construct-related and criterion-related validity). Within the seven measures, only the PedsQL met both criteria of reliability and validity for children and adolescents with ASDs although reproducibility and content-related validity have not been established for this population. The WHOQOL-BREF met criteria of validity but no evidence was found regarding reliability in the ASDs population. The ILK, QCL, HUI3 and QWB-SA demonstrated only construct-related validity such as discriminant and convergent validity. No information of psychometric properties was provided regarding the *Quality of Life* (Lee et al., 2008). Some of the studies reported the reliability and validity of the instruments based on the previous research which did not include children and adolescents with ASDs. For example, Bastiaansen, Koot, Bongers et al. (2004) reported all criteria of the reliability, with the exception of reproducibility, and validity of the PedsQL in children

with psychiatric disorders. As the study did not identify what kind of psychiatric disorders were included, it is difficult to determine the psychometric properties of the PedsQL specifically for children and adolescents with ASDs. Consequently, there was little evidence that the QOL and HRQOL measures were reliable for use in children and adolescents with ASDs.

#### *Differentiation between QOL, Health Status and Functional Status*

Six studies examined correlations between QOL or HRQOL measures and clinical measures (Kamp-Becker et al., 2011; Kamp-Becker et al., 2010; Kuhlthau et al., 2010; Limbers et al., 2009; Shipman et al., 2011; Tilford et al., 2012). Significant correlations and at least medium effect sizes with clinical measures demonstrate the convergent validity of QOL and HRQOL questionnaires (Bastiaansen, Koot, Bongers, et al., 2004; Eiser & Morse, 2001). The PedsQL was significantly correlated with ASDs-related measures of symptoms (i.e. ASDs, Vineland-II, RBS-R and CBCL) and functional impairments (i.e. PedsQL Cognitive Functioning, RSES, SMFQ, SCARED and SRS) (Kuhlthau et al., 2010; Limbers et al., 2009; Shipman et al., 2011). These findings showed the convergent validity of the PedsQL but may also be interpreted that the PedsQL has items that overlap with symptoms and functional impairments of ASDs. However, *health status* which refers to the presence or absence of symptoms, and *functional impairment*, which refers to an objective measurement of impact on functioning, need to be differentiated from QOL, which is defined as self-perception of well-being (Coghill et al., 2009; Drotar, 2004). Although these correlations between QOL, symptoms and impairments do not necessarily imply that both items are identical constructs or factors, there may be the risk that QOL outcomes are used interchangeably for health status and functional impairments (Coghill et al., 2009; Drotar, 2004). However, subjective QOL and HRQOL measures need to be disentangled from objective assessment of symptoms and functional impairment as the concepts of QOL and HRQOL are different from those of health and functional status (Coghill et al., 2009; Davis et al., 2006; Drotar, 2004; Smith, Avis, & Assmann, 1999). In children and adolescents with ASDs, this issue may be caused by a respondent's confusion between their perceptions of *think* and *feel* and their conditions of *is* and *can* due to deficiency in recognition of their self-insight (Coghill et al., 2009; Mitchell & O'Keefe, 2008).

Another possibility may result from lacking the precise wording of the questions which refer to distinctions between psychopathology (e.g. “Are you feeling happy?”) and QOL (e.g. “Are you feeling as happy as you think you should?”) (Coghill et al., 2009). It is not clear from the findings that the wording of each questionnaire is comprehensible to children and adolescents with ASDs to report their perceptions independently of their symptoms and impairments. Considering that children and adolescents with ASDs have impaired cognitive and language abilities, their interpretation and understanding of the questions may be different from other populations.

### *Implications and Recommendations for Future Studies*

This review showed that the PedsQL was the most commonly used in children and adolescents with ASDs and established reliability and validity in the ASDs population. In this regard, the PedsQL exhibited substantial contribution toward measuring QOL and HRQOL in children and adolescents with ASDs. The PedsQL would be advantageous to this population in terms of a simple structure (i.e. four scales and three summary scores), the length of the questionnaire (i.e. 23 items), age-appropriate versions (i.e. young child, child and teen) and established psychometric properties (i.e. reliability and validity). However, it is not clear that the same wording, formatting and design of the PedsQL questionnaire are relevant to children and adolescents with ASDs considering their unique mental and cognitive functioning. Furthermore, the reliability and validity of the PedsQL may need to be established for children and adolescents with ASDs across the wide range of age and different levels of disability. Future research should investigate the content appropriateness, accuracy and trustworthiness of the self-reported PedsQL for use in children and adolescents with ASDs.

### **Limitations**

Due to scant research regarding QOL and HRQOL in children and adolescents with ASDs within which different methodologies (i.e. data analyses) and instruments (i.e. QOL, HRQOL, clinical and cognitive measures) had been applied, it was not possible to compare QOL and HRQOL measures and outcomes among studies. In some studies, it was not clearly described which versions of the QOL and HRQOL instrument (e.g. age-specific reports) were used in the studies. In addition, there was a deficiency of

information about the reliability and validity of the QOL and HRQOL measures in children and adolescents with ASDs. With the exception of three studies (ASDs samples:  $n = 150$ ,  $286$  and  $483$ ) (Kuhlthau et al., 2010; Lee et al., 2008; Tilford et al., 2012), sample sizes of the individual study were small (ASDs samples:  $n = 12-42$ ). Participants in the studies were predominantly boys and without intellectual disability for example high-functioning autism and Asperger syndrome ( $IQ > 70$ ).

## CONCLUSIONS

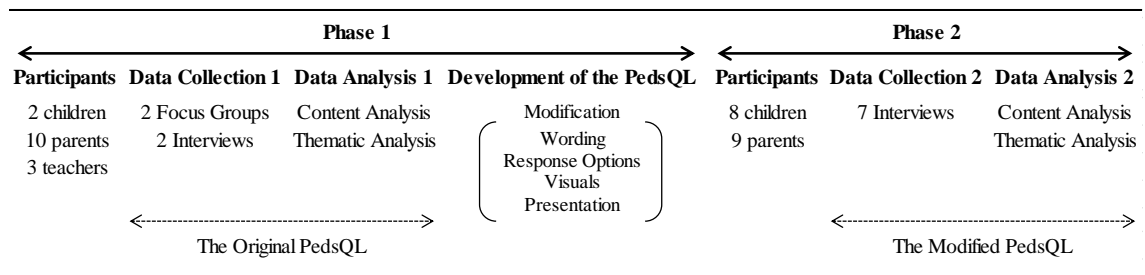
This review demonstrated that children and adolescents with ASDs experienced lower levels of QOL and HRQOL across a variety of domains. In particular, children and adolescents with ASDs showed lower scores in the domain of social functioning which is one of the characteristics of ASDs. Considerable differences between self- and proxy-reported QOL and HRQOL emphasised the importance and development of self-report in children and adolescents with ASDs. From this review, the PedsQL seemed to be an appropriate QOL and HRQOL measure for use in children and adolescents with ASDs. It is vital for future research to investigate further psychometric properties and the content appropriateness of the PedsQL self-report for use in children and adolescents with ASDs.



## **CHAPTER 3: METHODS**

### **RESEARCH DESIGN**

This research was a qualitative descriptive study (Neergaard et al., 2009; Sandelowski, 2000) and consisted of two phases: Phase one and Phase two (see Figure 3.1.). Participants were recruited through a special school and a school with a special unit in Auckland region and via two service providers; the New Zealand branch of Centre for Autism and Related Disorders and the Wilson Home Trust, also located in Auckland. Two children with autism spectrum disorders (ASDs) or mild intellectual disability (MID), ten parents and three teachers participated in Phase one. Two focus group discussions and two individual interviews were conducted at the special school and at the university. The participants were provided with the Pediatric Quality of Life Inventory<sup>TM</sup> Child Self-Report (i.e. the original PedsQL questionnaire) and asked about their experiences, knowledge and perception in relation to the original PedsQL questionnaire. Data from the focus groups and interviews were analysed using content and thematic analyses. On the basis of the findings from Phase one, the original PedsQL questionnaire was modified for use in children and adolescents with ASDs or MID (i.e. the modified PedsQL questionnaire). The amendment of the original PedsQL questionnaire encompassed wording, response scales, format and presentation. In Phase two, eight children with ASDs and their parents ( $n = 9$ ) participated in individual interviews. After the administration of the modified PedsQL questionnaire with the children and adolescents, they and their parents were interviewed regarding the modified PedsQL questionnaire. Content and thematic analyses were conducted for identifying key themes and categories from the interviews. Mean and standard deviation of time for completion and completion rates of the modified PedsQL were reported.



**Figure 3.1.** Qualitative descriptive research design.

### PHASE ONE

Fifteen participants were recruited using convenience sampling, and focus groups and semi-structured interviews were employed for data collection (see Figure 3.1.). The original PedsQL questionnaire was provided to the participants as the main topic of the discussions or interviews. A facilitator and an interviewer led the focus groups and interviews with a couple of open-ended questions about the appropriateness of the original PedsQL questionnaire. The data were analysed using content and thematic analysis techniques to identify key themes and categories. After the data analysis, the modification of the original PedsQL questionnaire was conducted regarding wording, response options, formatting and presentation style.

### PHASE TWO

After the modification of the original PedsQL questionnaire, 17 participants including children and adolescents and their parents were recruited for individual interviews using convenience and purposive sampling (see Figure 3.1.). The modified PedsQL was first trialled by the children and adolescents, and then an interviewer asked the parents and children about the appropriateness and effectiveness of the modified PedsQL questionnaire. Content and thematic analyses were undertaken for data analysis.

## SAMPLING

This research employed sample of convenience and purposive sampling techniques. Convenience sampling involves recruiting the most accessible individuals, while a purposive sampling selects individuals who are “information-rich” for the aim and purpose of the study (Marshall, 1996; Sandelowski, 2000, p. 338). In Phase one, convenience sampling was utilised due to time constraints and the small number of possible key informants who involved children, parents and teachers. Children, parents and teachers were considered as important informants able to provide holistic knowledge regarding the children’s quality of life at home, school and other environments. In Phase two, in addition to convenience sampling, purposive sampling was also used to obtain effective feedback on the modification of the original PedsQL questionnaire. Children and their parents were identified as key informants who were able to provide knowledge regarding themselves (children) and their children. Three participants whom the researchers identified as key informants based on the child’s age and types (i.e. autistic disorder or Asperger syndrome) and levels of disability (i.e. mild to moderate) were asked to participate in interviews in Phase two.

With regard to sample size, Marshall (1996) described that “an appropriate sample size for a qualitative study is one that adequately answers the research question” (p. 523). Lewis, Kellett, Robinson, Fraser and Ding (2004) also suggested that a small sample size is preferable in a qualitative descriptive research. Considering that few new themes and categories emerged between Phase one and two, the data saturation were likely to be reached and depth of data were obtained from both phases. Therefore, the total of 15 and 17 participants from each phase was appropriate.

Participants were included if (1) children or an adolescents were at the age between five and 18; (2) children or adolescents had clinical diagnosis of ASDs; and (3) children and adolescents were not diagnosed as ASDs but had MID (approximate  $IQ \geq 50$ ) (APA, 2000; WHO, 2010). However, children and adolescents or their parents were excluded if (1) children and adolescents had not been diagnosed as ASDs but had moderate, severe and profound intellectual disability (approximate  $IQ < 50$ ) (APA, 2000; WHO, 2010); and (2) children and adolescents had physical impairments. The diagnosis and severity of ASDs or ID which were assessed by specialists such as paediatricians,

psychiatrists or clinical psychologists were reported by parents/guardians or teachers of special need school (following consent from parents). Additionally, specialists' and teachers' advice and judgement on the eligibility of children and adolescents for this research were considered to determine their participation. According to the Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV) (APA, 2000), ASDs involves six diagnoses of autistic disorder, Asperger disorder, childhood disintegrative disorder, Rett disorder, Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), and atypical autism, which are consistent with the International Classification of Disease, 10th edition (ICD-10) (WHO, 2010). In this research, autistic disorder (i.e. autism, autism spectrum disorder and high-functioning autism) and Asperger disorder (i.e. Asperger syndrome) were identified under the diagnosis of ASDs.

Characteristics of participants are shown in Tables 3.1 (Phase one) and 3.2 (Phase two). Children and adolescents who participated in Phase one and two varied in terms of age and disability levels, but resembled each other in terms of gender and types of disability.

**Table 3.1**

*Descriptive Characteristics of Participants in Phase One*

Form	Participant (N)	Child Disability
Focus Group		
1	Children (2), parents (4), teachers (3)	GDD (P), Epilepsy (C), ADHD (P)
2	Parents (4)	ASD (P), Down Syndrome (P)
Interview		
1	Parent (1)	ASD (P)
2	Parent (1)	ASD/HFA (P), ADHD (C), AS (C)

*Note.* N = Number. GDD = Global Developmental Delay. ADHD = Attention Deficit Hyperactivity Disorder. ASD = Autism Spectrum Disorder. AS = Asperger syndrome. HFA = High-functioning Autism. P = Primary disorder. C = Comorbidity.

The children and adolescents were all boys from 6 to 16 years of age and predominantly diagnosed as autistic disorder (autism) with low to high functioning. Other primary

diagnoses included global developmental delay, attention deficit hyperactivity disorder (ADHD), Down syndrome. Most of the children and adolescents also had moderate to mild intellectual disability (MID). Some children and adolescents who were primarily diagnosed as autism had comorbidities such as ADHD, Asperger syndrome, global developmental delay and epilepsy. All children and adolescents with ASDs who participated in Phase two were boys. This is not unusual as it is evident that the majority of individuals with ASDs are male (Elsabbagh et al., 2012).

**Table 3.2**

*Descriptive Characteristics of Participants in Phase Two*

Child	Gender	Age	Disability
1	M	6	AD (P)
2	M	7	AD (P), GDD (C), ADD (C), Epilepsy (C), Syringomyelia (C)
3	M	16	AD (P)
4	M	10	AD (P), ADHD (C)
5	M	10	AD/HFA (P), AS (C), ADHD (C)
6	M	14	AD (P), Chromosomal Disorder (C)
7	M	6	AD (P), ADHD (C)
8	M	15	AD (P), AS (C), ADHD (C)

*Note.* M = Male. AD = Autistic Disorder. GDD = Global Developmental Delay. ADD = Attention Deficit Disorder. ADHD = Attention Deficit Hyperactivity Disorder. AS = Asperger syndrome. HFA = High-Functioning Autism. P = Primary disorder. C = Comorbidity.

## RECRUITMENT

Participants were recruited through a special school, which is one catering for students who have special education needs, and a mainstream school with a special unit in Auckland region, and via two service providers, the New Zealand branch of Centre for

Autism and Related Disorders and the Wilson Home Trust in Auckland. The invitation to the research was given to potential participants through advertisements and letters. In Phase one, two focus groups and two individual interviews were conducted. The number of participants in each focus group was nine and four. The first focus group consisted of children or adolescents with ASDs or MID ( $n = 2$ ), parents ( $n = 4$ ), and special school teachers ( $n = 3$ ) whereas the second one included only parents ( $n = 4$ ). The individual interviews were undertaken with parents ( $n = 2$ ). In Phase two, seven individual interviews with children or adolescents with ASDs ( $n = 8$ ) parents ( $n = 9$ ) were conducted.

### **INFORMED CONSENT**

Ethics for this research was approved by AUTECH (reference number 11/206). Information sheets were provided to children (see Appendix B for complete proof) and their parents and teachers (see Appendix C for complete proof) and a principal (see Appendix D for complete proof). Access to the school (see Appendix E for complete proof) for a focus group was obtained from the principal. Assent from children (see Appendix B for complete proof), consent from parents and teachers (see Appendix F for complete proof) and focus group consent (see Appendix G for complete proof) were received prior to study commencement. As children and adolescents with autism spectrum disorders and mild intellectual disability are a vulnerable population in terms of their age and severity of disability, consultation with parents and teachers was undertaken prior to the data collection. Where this was the case, researchers gave special consideration to children' and adolescents' needs, particularly with respect informed consent. Few attempts were made by researchers to give voice to the children and adolescents and to understand their experiences, likes and dislikes, perceptions and actions in their own terms (Morrow & Richards 1996).

## QUALITY OF LIFE MEASURE

### **Pediatric Quality of Life Inventory™ Generic Core Scales Child Self-Report: The Original PedsQL Questionnaire**

In Phase one, the Paediatric Quality of Life Inventory™ 4.0 Generic Core Scales Child Self-Report for *young child* ages five to seven years (refer to “the original PedsQL” in this research) were utilised. The PedsQL questionnaire also involves the other versions of child self-report including for *child* (ages 8 to 12 years) and for *teen* (ages 13 to 18 years). We (supervisors and myself) identified the PedsQL questionnaire as the most appropriate for children and adolescents with ASDs or MID aged from 5 to 18 years because the PedsQL questionnaire might be comprehensible for children and adolescents regardless of their cognitive levels (Cremeens et al., 2006; Riley, 2004). The PedsQL questionnaire consists of total 23 items in four distinct dimensions: physical functioning (eight items), emotional functioning (five items), social functioning (five items) and school functioning (five items) (Varni, Seid, & Kurtin, 2001) (see Appendix H for complete proofs). The PedsQL questionnaire contains a three-point rating scale: “not at all”, “sometimes” and “a lot” (Varni et al., 2001). The reliability and validity of the PedsQL self-reports have been established in children and adolescents with and without chronic and mental health conditions (Coghill et al., 2009; Danckaerts et al., 2010; Dey et al., 2012; Varni, Burwinkle, Seid, & Skarr, 2003). Among adolescents with high-functioning ASDs (IQ > 70), high internal consistency reliability, low inter-rater agreement, moderate to high construct validity and moderate concurrent validity were established in the PedsQL self-reports (Sheldrick et al., 2011; Shipman et al., 2011).

## DATA COLLECTION

The purpose of data collection is to discover the *what*, *who*, *where* and *why* of experiences and perceptions of participants relevant to a specific topic (Sandelowski, 2000). In reflecting on this purpose, the research question: “What is an appropriate QOL measurement for children and adolescents with ASDs or MID?” was explored through focus group discussions and semi-structured interviews.

## **Focus Groups**

Focus groups lasted up to 60 minutes. Researchers (the student and two supervisors) actively involved in creating the discussions and were consistently assigned to a facilitator (one supervisor) and note takers (the student and another supervisor) (Morgan, 1996). The facilitator led the discussion, and the note takers recorded the focus group feedback during the conversation. Special considerations were given to children with regard to encouraging them to speak their own views at ease, and to all participants to welcoming different and similar perspectives in the discussion. These considerations were given to generate interactive discussions in which interactions produce valuable data on the degree of consensus and disagreement among participants (Morgan, 1996).

Several procedures were followed to make sure that the focus groups were conducted in the most effective and efficient way possible. Firstly, venues were carefully selected for each group to be easily accessible and feel comfortable to speak about their experiences and perception. A classroom at special school and a meeting room at the university were organised. Secondly, each session began with informal conversation and “ice-breaking” interaction which can create relaxing environment (Powell & Single, 1996, p. 502). Then, a brief introduction of the research topic was described to orientate the participants to the main focus of the discussion. Lastly the focus groups were audio-taped and the student transcribed them immediately after the discussions while the information remained fresh in their minds. Each transcription and note was doubly checked by the two supervisors who participated in the discussion.

## **Interviews**

Interviews were semi-structured and between 30 and 60 minutes in duration. Special consideration was given particularly to children and adolescents with ASDs regarding an environment in which the children and adolescents feel comfortable. According to Plimley (2007), individuals with ASDs are more likely to feel stressed under the following situations: Change, ritual events, emotive events, sensory contact, social interactions and unpredictability. Therefore, researchers (the student and one supervisor) consulted parents in advance to ensure an environment where their children felt comfortable to stay and speak about their experiences. Before each interview started,



the researchers allowed time for children to familiarise themselves with the university environment, if the interview was conducted at the university, and the researchers.

The interviews began with a brief introduction to explain the research topic and its purpose to the children and their parents. The researchers were consistently assigned to an interviewer (one supervisor) and a note taker (the student). The interviewer led the interview with parents and the children. In Phase two, the interviewer also assisted the children and adolescents, if necessary, while they were answering the modified PedsQL questionnaire. Although the children and adolescents were supported to complete the whole questionnaire, they were encouraged to have a rest between questions. If the children were not able to understand or were unwilling to answer a question, they skipped the question. The note taker documented the conversation feedback as well as the children's behaviours during the interview. The interviews were audio-taped and transcribed by the student and each transcription and note was doubly checked by the supervisors who conducted the interviews.

#### *Open-ended Questions in Focus Groups and Interviews*

Specific open-ended questions related to the appropriateness of the original and modified PedsQL questionnaires were asked to guide the discussion and interview: *"What did you think about this questionnaire?"*; *"What aspects of quality of life for the children were missing or not relevant?"*; *"Which questions were unclear or confusing to the children? Why?"*; *"What about wording / response scales / format that was / were used in the questionnaire?"* For the interviews, however, the emphasis was more on personal aspects of "your child/children" for parents and "you" for children and adolescents to attain detailed behavioural, attitudinal and experiential information (Powell & Single, 1996). In Phase two, questions regarding parental interpretation of the children's behavioural responses to the modified PedsQL questionnaire were also sought for the evaluation of the children's experiences.

These open-ended questions encourage participants to evaluate the issues of importance to the participants using their own ideas and feelings (Kitzinger, 1995; Krueger & Casey, 2000). The open-ended questions also prompt the participants to generate their own questions and develop the exploration of relevant experiences (Kitzinger, 1995).

The flexibility of the open-ended questions could provide a wide range of rich data in relation to the participants' experiences and perceptions of the original and modified PedsQL questionnaires.

## **DATA ANALYSIS**

Two strategies of content analysis and thematic analysis were used to analyse the data from transcriptions and notes. The definition and procedure of each technique are described below.

### **Content Analysis**

Content analysis is a common analysis technique for qualitative description (Neergaard et al., 2009; Sandelowski, 2000). In this research, the purpose of content analysis was (1) to assess the appropriateness of the format, wording and presentation of the PedsQL questionnaire in children and adolescents with ASDs or MID; and (2) to identify relevant or irrelevant dimensions of the PedsQL questionnaire to children and adolescents with ASDs or MID. The data were analysed according to an inductive content analysis process (Elo & Kyngäs, 2008). The process involved coding, categorisation and summarizing (Elo & Kyngäs, 2008; Neergaard et al., 2009; Sandelowski, 2000). The student read the data several times and simultaneously wrote down key words and phrases to understand and become familiar with the content. The key words and phrases were labelled into each meaning unit which has been referred to as coding. The list of extracted codes then were grouped together to create subcategories and categories depending on similarities in the extracts. The main categories were defined and described through interpretation of each category. All processes were conducted by the student and formulated categories and descriptions of each category were double-checked by one of the supervisors (T.W.) to strengthen integrity and trustworthiness of the findings (Neergaard et al., 2009).

### **Thematic Analysis**

In order to capture further insight into experiences and perception of the participants, thematic analysis was also conducted. The purpose of thematic analysis in this research

was (1) to identify the perception of QOL measurement in children and adolescents with ASDs or MID; and (2) to analyse the interpretation of and attitudes towards the PedsQL questionnaires in children and adolescents with ASDs or MID. Thematic analysis was conducted based on the selective or highlighting approach (van Manen, 1990). The student first listened to recordings or read documents several times. The statement(s) or phrase(s) that appeared specifically important and to disclose about experience or perception of children and adolescents with ASDs were highlighted or underlined. The similar statement(s) and phrase(s) were grouped together. Then, themes and subthemes were discovered and defined through interpreting and seeing the meaning. Elements were identified within the themes and subthemes. The last process was collaboratively conducted with the student and the supervisor (T.W.), which included examination, articulation, re-interpretation, omission, addition or reformulation of the themes (van Manen, 1990, p. 100). The collaborative analysis improved research rigour and determined the credibility and integrity of the findings (Neergaard et al., 2009; van Manen, 1990).

## **CHAPTER 4: FINDINGS AND INTERPRETATION**

This chapter will present findings of the focus group discussions and individual interviews with children and adolescents, parents and teachers in Phase one and two. This chapter consists of five main parts. Firstly, the process of developing the PedsQL questionnaire will be described. Secondly, three key themes which were discovered through thematic analysis in Phase one and two will be presented together. According to Van Manen (1990), “theme is the process of insightful invention, discovery or disclosure” (p. 88) of the data through interpretation, and is not simply conceptual formulations or categorical accounts. Therefore, the interpretation of the data to uncover thematic aspects of the experiences and situations will be also introduced with the findings. Thirdly, key categories which were identified through content analysis in Phase one and two will be presented separately. Since themes embodied the meaning and interpretation of the data, the findings through thematic analysis were broader and deeper than those through content analysis. Consequently, the themes were presented before the categories to capture the significance of the findings. Some findings identified through content analysis were associated with the themes. Lastly, the completion rate and time for completion of the PedsQL questionnaire in Phase two will be reported.

### **THE DEVELOPMENT OF THE ORIGINAL PEDSQL QUESTIONNAIRE: THE MODIFIED PEDSQL QUESTIONNAIRE**

The findings from Phase one identified that the Paediatric Quality of Life Inventory™ 4.0 Child Self-Report ages 5 to 7 years (i.e. the original PedsQL) needed to be modified in order to be used for children and adolescents for ASDs or MID. Through discussions among the four researchers (the student and supervisors), several modifications to the original PedsQL questionnaire were made (i.e. the modified PedsQL). The modification of the original PedsQL questionnaire included rewording, changes in response scales and presentation style, and use of visuals (see Appendices H and I for complete proofs). The modified PedsQL questionnaire was completed by children and adolescents with ASDs in Phase two. The number of questions which were skipped (i.e. completion rate) and the time for completion of the modified PedsQL questionnaire were recorded.

### **THEMATIC ANALYSIS OF FOCUS GROUPS AND INTERVIEWS: PHASE ONE AND TWO**

Three themes of *concreteness*, *relationships* and *perception* were identified in relation to the perception of quality of life (QOL) measurement in children and adolescents with ASDs or MID, and their attitudes toward the PedsQL questionnaire (see Table 4.1 and 4.2). The same themes were identified through the analysis of data from Phase one and two although more subthemes were recognised in the data from Phase two. Therefore, all data from Phase one and two are presented and described together based on the themes and subthemes from Phase two.

**Table 4.1**

*Themes from Thematic Analysis of Focus Groups and Interviews in Phase One*

Theme	Definition	Subtheme	Definition	Examples of quotations
Concreteness	Something that is tangible, based on reality of specific events or experience and is often perceptible through the senses	Who / Whom	A description of a person or people in the questionnaire	"If possible, the use of 'name of the child' instead of 'you' when asking the child about their health and activities."; "Maybe it is better to ask questions about a specific friend rather than in general about friends."
		When	The time or period that something is done or happens in a particular situation	"Specifying a certain time or a certain day such as 'Monday' or 'today'."; "Asking the children to think about the questions in terms of 'the past month' would be a challenge. 'Lately' or 'past week' would be better."
		Where	A particular place or part that is likely to be known by the child, or a variety of interpretations of context	"It's better to use 'wide contexts' rather than time, such as 'at school' or 'at home'."; The question of 'I worry about what will happen to you', where and when?"

		What	Particular information or details about the thing or things	"It may be useful to separate 'classwork' from 'homework' in terms of keeping up with 'schoolwork'."; "Without particular reference to a specific sporting activity or exercise that the children like or try to participate in, it may not make sense."
Relationships	A particular type of connection between individuals in the context of social, cultural and other influences, which varies from family or kinship relations, friendship or peer relationships to relations with neighbourhoods	Social Interactions	The process of communication between two or more individuals and one or more types of behaviours in the context of an individual's family, friends or peers	"The children want to make friends, but at the same time they feel overwhelmed."; "The children cope better with adults."
		Self	An individual person as the object of his or her own reflective consciousness and the state of not interacting with others	"Questions on social functioning such as 'getting along with other children' are not relevant for low-functioning children with autism because they don't want to play."; "The children actually prefer to play alone and don't necessarily want or desire the participation and interaction with other children."
Perception	The way we interpret or make sense of sensory input which is attained via five senses, namely sight, hearing, smell, taste and touch.	Visuals	Pictures that are illustrated in the questionnaire to make the meaning of each question clearer	"Possibly the use of visual images may also be helpful."; "Using pictures from their world ideally to supplement questions."; "The use of visuals would likely contribute to a better understanding of the questions by the children."

It directly influences or  
controls thoughts,  
feelings, behaviours or  
a combination of these

Feelings

A child's emotions that are aware  
through the mind and the senses, or  
emotional expressions that are  
demonstrated as pictures

"The use of 'happy face' or 'sad face' symbols for the  
questions around feelings."; "Use visuals together  
with emotions such as smiley faces would be more  
easily understood."; "Use of emotive language may  
be difficult for some children to understand."

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### **Theme One: *Concreteness***

*Concreteness* is defined as something that is tangible, based on reality of specific events or experience and is often perceptible through the senses (Hobson, 2012; Rosa, Catricalà, Vigliocco, & Cappa, 2010). This is in contrast to abstract which refers to something that cannot be perceived directly in the senses (Rosa et al., 2010). Being concrete implies that concepts become more perceivable, more imaginable, or easier to contextualise (Rosa et al., 2010), and it avoids issues of vagueness, ambiguity or generalisation. Moving from concrete to abstract thinking is one of the milestones of normal childhood development.

According to Piaget's cognitive developmental theory, children about age 7 to 11 are at the concrete-operational stage (Shaffer & Kipp, 2007a). In this stage, children rapidly acquire cognitive operations and apply these important new skills when thinking about objects and events that they have experienced (Shaffer & Kipp, 2007a). A cognitive operation is an internal mental activity that enables children to modify and reorganize their images and symbols to reach a logical conclusion (Shaffer & Kipp, 2007a). When children turn 12 to 13 years of age, they start to think abstractly that represents the attainment of formal operational thinking (White, Hayes, & Livesey, 2010d). This increased ability of abstract thinking allows the children to consider alternative ways of organizing the world and to think more deeply about abstract concepts such as truth, justice, freedom and morality (White et al., 2010d). This cognitive transition from concrete to formal operational thinking, however, is not likely to occur properly in children with ASDs; consequently, they have difficulty dealing with abstract concepts.

Children with ASDs often demonstrate impairment in verbal and non-verbal communication which are associated with cognitive and language malfunctioning (Filipek et al., 1999; Hobson, 2012). Atypical patterns of cognitive processing such as concrete and abstract thinking in children with autism are probably due to neurological abnormalities (Grandin, 2000; Quill, 1995). Individuals with ASDs are limited in their ability to think abstractly in which the process of interpretation is required (Church, Alisanski, & Amanullah, 2000; Hobson, 2012; Ropar & Peebles, 2007). Instead, they pay more attention to concrete characteristics of stimulus (Hobson, 2012; Ropar & Peebles, 2007). Children with high-functioning autism and Asperger syndrome, for

example, find concrete words or factual knowledge easier to read than abstract words (Church et al., 2000; Ropar & Peebles, 2007).

Differences in the way children and adolescents with ASDs think concretely can become problematic when they deal with questions or questionnaires that involve abstract concepts. Hobson (2012) describes how individuals with autism have difficulty with forming a fixed and categorical frame of reference that transcends the immediate context of their experience. For typically developing children around the age of 12 years, abstract thinking is a developmental landmark (White et al., 2010d). However, the present study revealed that not only the participants aged younger than 12 years but also the older participants were confused by and could not comprehend general and conceptual ideas. In this respect, it is critical to identify abstract concepts in the original PedsQL questionnaire and determine what is concrete for children and adolescents with ASDs in the different contexts of each question.

Questions about social functioning in both PedsQL questionnaires that focused on social interaction or relationships used abstract terminology of *who/whom* such as “you” and “other kids”. The definition of *who/whom* refers to a description of a person or people in the questionnaire. The abstract concept of *who/whom* was difficult for the children and adolescents with ASDs to comprehend as it introduced people in a general rather than specific way. In response to this, parents from Phase one and two of the focus groups and interviews talked about using the names of the child with ASDs instead of “you” and “other kids”. An example of this was “(Jack) likes to play with (William and Liam).”

Another difficulty for children and adolescents with ASDs was orientating to the general idea of *when*. *When* is used to describe the time or period that something is done or happens in a particular situation. However *when* is asked as a question, it is an inquiry into a more general point in time or a period of time, such as “the past one month” or “the last few weeks.” It was hard for the children and adolescents with ASDs to generalise about their answers over the period of time. Bell (2007) also indicated that short recall periods are more suitable for children due to their limited memory capacity. Parents from Phase one of the interviews suggested that it might be possible for children with autism to respond a question if the period is more specified at a point of time, such

as “yesterday.” In addition, the concept of “yesterday” needs to be connected to a specific day because the children found it difficult to grasp the idea of “yesterday.” For example, the concept of “yesterday” was explained as “Yesterday was Tuesday,” before the children answered the questionnaire. Regarding questions about school functioning, however, the idea of “yesterday” was modified if a child and an adolescent did not have school “yesterday.” In this case, a certain day in the past will be provided to remind the child and adolescent of his school days, for instance “on Friday.” Children and adolescents with autism also tend to consider specificity of time in a day. On the second question in school functioning of the modified PedsQL questionnaire, an adolescent with autism and his mother from Phase two of the interview had conversation of whether he was sick in bed or not. The adolescent maintained that he was sick in bed yesterday. The mother asked him,

*“When were you sick, at night time or during the day?”*

He answered,

*“I was sick at night time.”*

This result highlighted the importance of specification of *when*. The question needs to be described with a particular time frame such as “daytime” or “night time” in order to obtain an accurate response from the child.

The information of *where* also helps children and adolescents with ASDs to visualise a situation that they have experienced. *Where* refers to a particular place or part that is likely to be known by the child or adolescent. *Where* also encompasses a variety of interpretations of context, for example “where it is”. Identifying *where* in each question allows children and adolescents with ASDs to understand the meaning of the questions and smoothly link each question to their memories. In response to this comment from Phase one of the focus group, all questions about school functioning were modified with the information of *where* which is either “at school” or “at home.” A parent commented on Question 7 of the physical functioning domain of the modified PedsQL questionnaire that it might be appropriate to ask a child a further question such as “where he/she hurt,” if the child answered “a lot” or “sometimes.” Use of the general term of “body” in this question may result in guessing an answer instead of judging it from their experience.

As regards the second question in school functioning of the modified PedsQL questionnaire, a parent asked an adolescent to ensure that he actually meant that he had felt sick,

*“Where are you sick or sore?”*

The parent explained later that he sometimes had a headache, which is not necessarily categorized as traditional sickness but he might answer “he was sick.” Again, the word of “sick” may to be misinterpreted by the children and adolescents with ASDs or MID. Clearly highlighting *where* or *which part* in a question and even its answer may allow children and adolescents with ASDs to interpret the question correctly and to obtain the answer which are inferred from circumstances of their life.

General terms of physical activities such as “sports” and “exercise” used in the third question in physical functioning of the original PedsQL questionnaire were hardly understood by children and adolescents with ASDs. It is necessary when working with children and adolescents with ASDs to explicate *what* kind of activities each question indicates. The notion of *what* implies particular information or details about the thing or things. In Phase one of the interviews, one parent suggested that it may not make sense to the children if there is no particular reference to a specific sporting activity or exercise that children with ASDs like or try to participate in. In Question 6 in physical functioning domain of the modified PedsQL questionnaire, another ambiguous phrase, “to help in the house” was often highlighted in Phase two of the interviews. One child was not able to understand the meaning of this question. Three parents from Phase two of the interviews provided an example of their children’s chores at home such as “mopping”, “vacuuming” and “tidying up a room” when the question was read out. These results show that specific activities for each child need to be identified through their parents or caregivers before the questionnaire is conducted.

Considering these consistent findings, the description of concrete contents and contexts in a questionnaire may be critical for children and adolescents with ASDs. The concept of concreteness indicates that one circumstance cannot be converted into others. When context of each question is precisely set by specific information of person, time and place, the questionnaire may provide only a snapshot of a child’s or an adolescent’s

QOL instead of the present QOL in general. It is possible that a child or an adolescent may answer the same questionnaire differently depending on the situation where he or she is at this moment. It was often reported that children and adolescents with ASDs had a weekly routine. It means asking questions about a point of time such as “yesterday” may cause a wide diversity of answers over a period of time. In addition, a questionnaire which is tailored for a child or an adolescent with ASDs may not be appropriate for the other children or adolescents. It means that it will be difficult to create a general questionnaire among children and adolescents with ASDs, and variability will exist within the questionnaire.

**Table 4.2**

*Themes from Thematic Analysis of Interviews in Phase Two*

Theme	Definition	Subtheme	Definition	Element	Examples of quotations
Concreteness	Something that is tangible, based on reality of specific events or experience and is often perceptible through the senses	Who / Whom	A description of a person or people in the questionnaire		"Do you like to play with [the child's friends name listed]?"
		When	The time or period that something is done or happens in a particular situation		"Yesterday was Tuesday."; "At night time or during the day?"
		Where	A particular place or part that is likely to be known by the child, or a variety of interpretations of context		"Where were you sick? At home?"; "Where were you sick in your tummy or head or arm, where were you sick?"
		What	Particular information or details about the thing or things		"Is it easy for you to tidy up or is it easy for you to make your own bed, is it easy?"; "Like tidy up"; "I (the parent)'d make them (the children) vacuum and mop."

Relationships	A particular type of connection between individuals in the context of social, cultural and other influences, which varies from family or kinship relations, friendship or peer relationships to relations with neighbourhoods	Social Interactions	The process of communication between two or more individuals and one or more types of behaviours in the context of an individual's family, friends or peers	Friends	"I (the parent) just think it's difficult all of that area so trying to get kids to play with friends and the whole thing."; "He (the child) doesn't need a friend around."
				Family	"Socially their (the children's) brothers are totally different from other kids."; "The most important thing in his (the child's) life is his family, you know, we spend a lot of time with extended family."; "He (the older brother) found me (the child) as a big embarrassment to him."
				Adults	"Their (the children's) ability to have relationships with adults and teachers and... I (the parent) think it's really really important."; "They (the children) are much more comfortable talking to adults than they are with, you know, their peers."
				Community	"I (the parent) am actually looking into building community. That's for the child."; "I (the parent) think it is quite important because if I want them (the children) to be eventually something that happens in the community."

		Self	An individual person as the object of his or her own reflective consciousness and the state of not interacting with others		"I (the child) just don't usually hang out with people. I just hang by myself."; "I (the parent) guess it could be tailored to the individual activities that the child likes doing."
Perception	The way we interpret or make sense of sensory input which is attained via five senses, namely sight, hearing, smell, taste and touch. It directly influences or controls thoughts, feelings, behaviours or a combination of	Visuals	Pictures that are illustrated in the questionnaire to make the meaning of each question clearer		"Picture expression is very good. He (the child) gets it quite quickly."; "Obviously kids with autism are very visual so yeah...pictures are good too because they are so simple."; "Pictures are definitely."
		Feelings	A child's emotions that are aware through the mind and the senses, or emotional expressions that are demonstrated as pictures	Facial Expression	"He (the child) quite likes TV but he only watches for a very short period of time. So I don't think he'd feel sad if we go for walk instead watching TV."
				Mental States	"Some people with what I (the child) have could be more the unexpected. Some might have bad temper, some might be a little distractive. And some can just have real bad meltdowns."; "I (the parent) am not sure that's necessarily how he (the child) is because one of them he said he was angry and sad and next one he was very happy so I don't know that is that."



these	Behaviour	A particular way that the child answers the questionnaire	Repetition	"He (the child) is still gonna say 'happy' because what he has been doing, it's about a routine and a pattern that is responded to."; "What I (the parent) am just saying was that to avoid that, you know, repetition and just answering the same thing."
			Preference	"He (the child) chose 'a lot' but he would probably choosing a smiley face."; "If you got 'yes' or 'no', he (the child) would always probably do 'yes' I (the parent) think."; "Most of the time he (the child) answers 'yes' for everything anyway."

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## **Theme Two: *Relationships***

A *relationship* refers to a particular type of connection between individuals in the context of social, cultural and other influences, which varies from family or kinship relations, friendship or peer relationships to relations with neighbourhoods. *Relationships* encompass aggregations of interactions between two individuals (Hinde, 1976, 1995; Rubin, Bukowski, & Lausen, 2011). Characteristics of a *relationship* are determined by the content, quality and pattern of interactions; the extent to which the interactions are reciprocal or complementary; and the level of cognitive and moral abilities (Hinde, 1976, 1995).

The level of cognitive, emotional, moral and personality development is strongly associated with that of social development (White, Hayes, & Livesey, 2010c). The degree of social achievement in early childhood including attachment, play, self-esteem and social relationships influences the development of socialisation and social competence in later life (White et al., 2010c). Compared to typically developing children, children with ASDs have certain impairments in perceiving, producing and understanding emotions, and verbal and non-verbal communication (Filipek et al., 1999; Travis, 1998). These impairments create obstacles to successfully engaging in social interactions and maintaining relationships (Travis, 1998). In the study by Church and Coplan (1995), parents of preteens with high-functioning autism reported that their children often had awkward peer relationships or friendships with younger children. One child described that he was able to sustain relationships with his peers if interactions were anticipated; however, if something was changed, the child got confused and would go into his long monologues (Church & Coplan, 1995). Adolescents with Asperger syndrome are often unable to make reciprocal social interactions and tend to be seen as self-centred with a deficit of understanding of others (Carrington, Templeton, & Papinczak, 2003). Individuals with Asperger syndrome find it hard to interpret implicit social cues such as non-verbal body language (Carrington et al., 2003). It is evident that the ability of initiating, sustaining or maintaining relationships among children and adolescents with ASDs is highly variable but still problematic. These findings were consistent with the results from the present study.

Under the theme of *relationships*, two subthemes were identified: *Social interactions* and *self*. *Social interactions* refer to the process of communication between two or more individuals and one or more types of behaviours in the context of an individual's family, friends or peers. All five questions about social functioning in both PedsQL questionnaires involved social interactions with "friends". However, other three essential interactions for children and adolescents with ASDs were also identified in this study: "Family", "adults" and "community". In response to the questions about social functioning especially regarding social interactions with friends, one parent of an adolescent with autism from Phase two explained one of the reasons for not interacting with friends.

*"Teenagers are ... (pause)... there are so many rules about being teenagers. That is impossible (to interact with them)."*

Two parents from Phase one and two of the interviews reported that the child and adolescent did not need friends around him. Another parent from Phase one of the interview explained that the child interacted with his classmates, which was reported by the child as "a lot" but by the parent as "sometimes". The parent explained that he did not realise that he did not socialise well. It is apparent that interactions with friends or classmates are difficult for children and adolescents with ASDs. Interactions with family including parents, siblings and extended family are important elements for children and adolescents with ASDs. In Phase two of the interview, one parent reported on interactions with a brother of the child with autism;

*"The older brother makes a point of engaging them and he comes up with play...and they readily interact with him. I think the harder part for the kids with spectrum is other people not their own family."*

Another parent from Phase two of the interview described,

*"The most important thing in his life is his family, you know, we spend a lot of time with our extended family. So I did think that there's actually nothing in the questionnaire about family."*

Children and adolescents with ASDs feel more comfortable to communicate and more likely to establish relationships with adults such as teachers and teacher aides than with peers. Parents from Phase one and two of the interviews pointed out the importance of having relationships with teachers or teachers' aides since adolescents with ASDs have difficulty and a lack in getting along with their peers. Complex changes in the structure of peer relationships from childhood to adolescence may hinder adolescents with ASDs from interacting with peers. Community interactions are a more future target for children and adolescents with ASDs in later life. In Phase two of the interview, one parent of a primary school child with autism described that they were looking into building community for the child and identifying the neighbours who could support him. Another parent from Phase two of the interview also described identifying the persons with whom the children play sports or do any physical activities is important as the children would eventually join something that happens in the community. The findings showed that the key social interactions in QOL of children and adolescents with ASDs were taking place rarely with friends or peers, more often with family and adults, and later in the community.

Although questions about social functioning in both PedsQL questionnaires mainly focus on relationships with friends, children and adolescents with ASDs have difficulty with interacting with peers and are more often by themselves. *Self* refers to an individual person as the object of his or her own reflective consciousness and the state of not interacting with others. Children and adolescents with ASDs preferred to be alone and do their own individual activities. One adolescent with Asperger syndrome commented,

*"I just don't usually hang out with people. Just by myself go for walk, do my own thing. I'm not so interested in hanging out with them."*

When he talked about being by himself in his spare time, he also described it as

*"I just say I have my moment. Usually at home I have my special moment."*

One parent from Phase two of the interview suggested that individual activities that the child likes doing are crucial for his QOL. Another parent from Phase two of the interview reported that the adolescent was usually watching DVDs or playing sports

otherwise there is nothing to do. Though playing sports in general is an opportunity to socialise with others, children and adolescents with ASDs are unlikely to socialise through sports. Nevertheless, they are still able to play individual and team sports without having socialised well. For children and adolescents with ASDs, ‘doing or participating in sports’ and ‘socialising through sports’ are not synonymous. These results revealed that self-activities or activities in their own time are important aspects of their QOL; however, there are no questions regarding the individual activities in either PedsQL questionnaire.

It is evident that for children and adolescents with ASDs building relationships and having social interactions are challenging parts of their life. They prefer to be by themselves and do their own things. However, socialisation is still a goal for their QOL. One adolescent with Asperger syndrome commented,

*“I’m trying to start social life but so far... (pause)... I think it’s not working.”*

Parents also try to make opportunities for their children to socialise with others such as through participating in sports. There may be a gap between the parental goal of fostering children’s and adolescents’ relationships and how the children and adolescents have actually perceived or experienced social interactions. Considering the concept of social relationships in children and adolescents with ASDs is different from that in typically developing children and adolescents, the PedsQL questionnaire need to embrace aspects of social interactions with not only friends or peers but family and adults including teachers and teacher aides. It is also necessary to recognise the importance of children’s and adolescents’ own time and activities.

### **Theme Three: *Perception***

*Perception* including “recognition” and “identification” refers to the way we interpret or make sense of sensory input which is attained via five senses, namely sight, hearing, smell, taste and touch (Shaffer & Kipp, 2007b; White, Hayes, & Livesey, 2010b). It also directly influences or controls thoughts, feelings, behaviours or a combination of these. *Perception* is obtained and progressed through interaction between inborn perceptual abilities and a child’s experiences in the environment (White et al., 2010b). By the age of six years, typically developing children have begun to have depth

perception, and have attained an ability to recognise colours and different facial expressions (White et al., 2010b). However, it is evident that sensory peculiarities are considered as integral features of autism and consequently affect the development of the perceptual system in individuals with autism (Iarocci & McDonald, 2006). Individuals with autism have reported difficulties in their subjective sensory-perceptual experience particularly in the reception (input) and processing (interpret) of sensory information (Iarocci & McDonald, 2006). Children with Asperger syndrome also have a noteworthy impairment in ability to integrate sensory information and demonstrate atypical and unique behaviours (Church et al., 2000). It is evident that perceptual peculiarities and consequent emotional and behavioural abnormalities are core symptoms in individuals with ASDs.

The development of perception progresses through fundamental abilities to selectively attend to, and spatially and temporally integrate multiple sensory information from different modalities (Iarocci & McDonald, 2006). Atypical perception in individuals with autism has effects on the integration of multiple sensory inputs (Iarocci & McDonald, 2006). The perceptual experience of individuals with autism demonstrated that visual perception is likely to predominate in the integrated audio-visual sensory contexts (Iarocci & McDonald, 2006). Grandin (2000) who was diagnosed with autism reported that her perceptual and thinking process carries on in pictures rather than in language and words. In the study of cerebral processing on an embedded figures task, individuals with autism used more visual parts of the brain rather than memory systems, which typically developing individuals mainly used (Ring et al., 1999). Individuals with ASDs also have impairments in the cognitive processing of emotions including problems in identifying and describing feelings, and difficulties in recognising feelings from the physical sensations of emotional arousal (Hill, Berthoz, & Frith, 2004). Children with autism at all ages and ability levels have abnormalities in characteristics of emotional expression and emotional recognition (Travis, 1998). Compared to adolescents with ASDs, however, younger children with ASDs struggled for recognising basic facial expressions and understanding complex emotions (Stichter, O'Connor, Herzog, Lierheimer, & McGhee, 2012). Rutherford and McIntosh (2007) reported that individuals with autism have a different process of recognising facial expressions, which tend to focus on discrete facial features rather than the configuration

of a combination of features. Behaviours of individuals with ASDs tend to be inflexible and follow a certain pattern or routines with specific details (Church et al., 2000; Ozonoff, Pennington, & Rogers, 1991). Many children with ASDs are more likely to have a particular interest or activity, and repetitively engage in a fixed type of behaviour (Church et al., 2000; Ozonoff et al., 1991).

In Phase one of focus groups and interviews, all parents consistently reported that the use of *visuals* is helpful for children and adolescents with ASDs to understand the meaning of each question. *Visuals* refer to pictures that are illustrated in the questionnaire to make the meaning of each question clearer. One parent from Phase two of the interview reported the use of visual images might be helpful for children with ASDs in any level of reading ability. Another parent interviewed in from Phase two described that the use of visuals would possibly contribute to better understanding of the questions by the children. In response to this feedback, the original PedsQL questionnaire was modified and illustrated with pictures in each question (the modified PedsQL questionnaire) (see Appendices H and I for complete proofs). In Phase two of interviews using the modified PedsQL questionnaire, five out of six parents and one adolescent provided positive comments on the visuals in the questionnaire. One parents described,

*“Picture expression is very good. He gets it quite quickly. I think the pictures help him to answer the question faster, but the fact is that he can read.”*

Another parent mentioned,

*“Pictures are definitely. Pictures are really help. The idea of visuals is of course to have less word and let the pictures do the talking.”*

Other comments on the visuals such as “simple”, “not distractive” and “get the child’s attention” were reported. These remarks demonstrate that the use of visuals in the questionnaire is beneficial for children and adolescents with ASDs in term of comprehension of the meaning of each question.

The PedsQL self-reports for children and adolescents are based on personal appraisal of their well-being on the domain of physical, emotional, social and school functioning.

For children and adolescents with ASDs, however, it is hard to recognise or reflect on their own feelings in relevant to each question. *Feelings* refer to a child's or an adolescent's emotions that are aware through the mind ("mental state") and the senses, or emotional expressions such as "facial expressions" that are demonstrated as pictures. Both of the original and modified PedsQL questionnaires include three-point response scale with facial expressions. In the original PedsQL questionnaire, the construction of each item is based on assessment of difficulties such as "how much of a problem", and the responses of "not at all", "sometimes" and "a lot" corresponded to "smiley", "neutral" and "sad" faces, respectively. The contrary between negative statements and positive responses might confuse children and adolescents with ASDs to answer questions. It is recommended that negative formulations should be avoided in a questionnaire for children (Bell, 2007). Therefore, in the modified PedsQL questionnaire, some questions were reworded from negative to positive wording such as "easy" instead of "hard" to highlight positive aspects of life and happiness. The responses of each question were also switched to match them to relevant facial expressions; for example, "not at all" was represented with "smiley" face. One parent from Phase one of the interview reported that use of smiley faces would be more easily understood for children with ASDs. On the contrary, one parent from Phase two of the interviews pointed out that facial expressions were quite confusing because they sometimes did not correspond to the child's own feelings. In the warm-up question of "how much TV did [name of the child] watch yesterday", the child would not feel "sad" if he could go for a walk instead of watching TV. It is possible that the use of facial expressions in the response options may cause confusion in terms of disagreement between a child's own feelings and corresponding facial expressions.

The nature of mental states in children and adolescents with ASDs is different from that in typically developing children and adolescents. In the study by Church et al. (2000), parents of middle school children with Asperger syndrome reported that their children were emotionally more unstable than typically developing peers. In the present study, the modified PedsQL questionnaire included four questions on mental states, namely feelings of fear, sadness, anger and happiness. It is evident that the mood of children and adolescents with ASDs fluctuates considerably in a day. One parent from Phase two of the interview doubted why the child answered that he felt angry, sad, scared and



happy in a day. This enquiry can be resolved by another adolescent's description of his own emotions. The adolescent with Asperger syndrome from Phase two of the interview reported that he had random mood swings in which he first feels happy and it changed to pain and misery in a moment. In addition, another parent described that the length of each emotion that a child felt was varied. Feelings of fear, anger and upset tended to be instant, but feelings of happiness and sadness were more likely to last longer. There may be difficulty in capturing the general characteristics of mental states in children and adolescents with ASDs.

Children and adolescents with ASDs are characterised by atypical behavioural traits, which affect their responses to the QOL questionnaire. *Behaviour* refers to a particular way that the child or adolescent answers the questionnaire. Two peculiar behaviours of "repetition" and "preference" which are typical features of ASDs were observed and reported throughout Phase two of the interviews. One pair of parents described,

*"He's got a routine of clicking a happy box because that just it becomes a habit. It's about a routine and a pattern that is responded to."*

In response to this feedback, the order of the response scales was slightly altered in every question. After a couple of trials of this version, one of the parents commented,

*"Do you change these orders around, don't you? Actually it's a good point. He (the child) would do that (choose the same answer)."*

Furthermore, children and adolescents with ASDs tend to prefer positive to negative responses, such as "a lot" rather than "not at all", "smiley face" rather than "sad face", or "yes" rather than "no". Two parents from Phase two of the interviews reported that the child always answered "yes" for everything. The parent also mentioned the child liked to choose "a lot" and "smiley face". These findings demonstrate that the presentation of the questions needs to be considered to avoid repetitive behaviours and preference but to internalise the children and adolescents with ASDs to respond each question.

Children and adolescents with ASDs have unique perception, which impacts on their feelings and behaviours. Predominant visual dependence, impairs in emotional

recognition and atypical behaviours were issues of conducting the QOL questionnaire in children and adolescents with ASDs. The use of visuals in each item may cause misinterpretation of the questions because the children and adolescents may not be familiar with the pictures or the pictures may not represent their real situation. Children and adolescents with ASDs may take questions as a task of matching facial expressions in the response scales to the pictures in the question, and they do not reflect their own feelings. Likewise, behaviours of repetition and preference in children and adolescents with ASDs may result in their habitually choosing the same option. Therefore, the presentation and administration of each question for children and adolescents with ASDs are important components of getting the children engaged in the questionnaire, and helping the children and adolescents understand the questions and reflect their thoughts.

#### **CONTENT ANALYSIS OF FOCUS GROUPS AND INTERVIEWS: PHASE ONE**

The appropriateness of the original PedsQL questionnaire in children and adolescents with ASDs or MID was examined in Phase one. Five categories of *rewording*, *responses*, *presentation*, *irrelevance* and *omission* were identified (see Table 4.3). *Rewording* refers to the wording in the original PedsQL that need to be changed into different words or phrases or even a new sentence. Parents reported that children and adolescents with ASDs might face a difficulty in understanding a word or phrase which has multiple meanings such as “hard”. In particular, autistic children with low cognitive functioning tend to interpret the meaning of each word literally. For instance, the word of “hard” might be perceived as “something that is difficult to bend or break” instead of “difficult to do, understand or answer”. This hallmark of literal thinking, in other words, concrete thinking which is related to the theme of *concreteness* would be a major obstacle to comprehending metaphorical terms in the original PedsQL questionnaire. Therefore, it is suitable for children and adolescents with ASDs to use a simple statement with a lucid account.

A *response* refers to an answer to a question in which options such as dichotomous or rating scales are included. The young child version of the original PedsQL has three Likert scales of “not at all”, “sometimes” and “a lot”, and the child report has five-point

Likert scales of “never”, “almost never”, “sometimes”, “often”, and “almost always”. One parent described that a five-point scale was too varied and too difficult to select one of them. Other parents suggested that “yes-no question” such as “Do you...?” or “Can you...?” would be more reasonable for children and adolescents with ASDs. All questions in the original PedsQL questionnaire arrange response scales in the same place, that is “not at all” in the left side, “sometimes” in the middle”, and “a lot” in the right side. Children and adolescents with ASDs have atypical repetitive behaviour of making a routine or habit, which may influence their selection of a response scale. Parents reported that the order of the response scales needed to be altered in order not to simply select the same scale. Not only the order of the response scales but the presentation of the questionnaire or questions was also an important factor. *Presentation* refers to the way in which the questionnaire or each question is shown or described to the children. The original PedsQL questionnaire is a one-page paper-and-pencil questionnaire questions in which 23 questions are listed consecutively. Two parents pointed out that it is preferable for children and adolescents with ASDs to present one question at a time. In addition, a parent recommended that a question be showed as a declarative rather than interrogative sentence.

The original PedsQL questionnaire was designed for children and adolescents with and without acute and chronic health conditions, but not specifically for children and adolescents with ASDs. The findings demonstrated that there were unrelated or missing aspects of their QOL in the original PedsQL. *Irrelevance* refers to a question that is not important to or associated with the child and adolescent. Most of the questions in the domain of social functioning were regarded as irrelevant to the children and adolescents with ASDs. In particular, children and adolescents with low-functioning autism and some with Asperger syndrome were unlikely to socially interact with their friends. *Omission* refers to elements that are important to the child’s or adolescent’s QOL but are not included in the questionnaire. One parent highlighted that the significance of “sense of belonging” in the child’s life was not included in the original PedsQL. A general impression of the original PedsQL questionnaire were favourable such as “straightforward”, “relatively easy”, “not too long” and “good for New Zealand kids”. However, there were still pitfalls that children and adolescents with ASDs were able to

complete the self-report of the original PedsQL, referring to a child's comment of "a little hard".

**Table 4.3**

*Categories from Content Analysis of the Original PedsQL Questionnaire*

Category	Definition	Subcategory	Examples of quotations
Rewording	The wording in the original PedsQL that need to be changed into different words or phrases or even a new sentence	Physical Functioning	"Use of the word 'hard' could be difficult for some children to understand."
		Emotional Functioning	"'Angry' instead of 'mad'. "; "'Trouble' may be a difficult word to understand in the context of sleeping."
		Social Functioning	"'Get along with' would be perhaps changed to ' to be friends with'. "; "The word of 'tease' would be changed to 'friendly' or 'unfriendly' perhaps. "; "The children might struggle with 'keep up'."
		School Functioning	"It may be useful to separate 'classwork' from 'homework' in terms of keeping up with 'schoolwork'."

Responses	An answer to a question in which options such as dichotomous or rating scales are included	Yes-No	"'Hard for you' would be changed to 'Can you...?'. "; "The question ('Is it hard for you...?') could be reworded to 'Do you...?'."
		Rating Scale	"A 5-point scale would be too hard and too many."
		Order	"Mixing up the smiley face answers between questions so there is less risk of the children simply selecting the same face as an answer."; "Need to mix things up so that the smiley is not always on the left."
Presentation	The way in which the questionnaire or each question is shown or described to the children	One-by-One	"One question at a time"; "Present questions as individual questions."
		Declarative Sentence	"Presenting questions as sentences, for example, '[NAME] is happy when he/she runs'."
Irrelevance	A question that is not important to or associated with the child	Social Functioning	"These questions are not relevant for low-functioning children with autism because they don't want to play. However, children with Asperger's want to have friends."
		School Functioning	"Are they supposed to remember things? (about the question of 'Do you forget things?')"

Omission	Elements that are important to the child's quality of life but are not included in the questionnaire	Social Functioning	"If a questionnaire were to be designed from scratch, it would have a question about 'sense of belonging'."
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## CONTENT ANALYSIS OF INTERVIEWS: PHASE TWO

In Phase two, the appropriateness of the modified PedsQL questionnaire in children and adolescents with ASDs or MID was examined. Each question in the modified PedsQL questionnaire included pictures and was presented one by one in a page. Four categories of *rewording*, *responses*, *irrelevance* and *omission* were recognised (see Table 4.4). *Rewording* refers to the wording in the modified PedsQL that need to be changed into different words or phrases or even a new sentence. A phrase of Question 6 in physical functioning, “to help in the house” was rephrased by most parents to something like “to tidy up” or “to mop and vacuum”. A parent explained that this phrase was too abstract for the children and adolescents to understand it. The word of “friendly” was also ambiguous according to one parent. Instead of asking children and adolescents with ASDs “how other kids behaved to you”, it is understandable to ask them “what other kids did with you” such as “talking” or “playing”. Again, concrete concepts and description are necessary for children and adolescents with ASDs.

A *response* refers to an answer to a question in which options such as dichotomous or rating scales are included. Response options in the modified PedsQL which consisted of three-point Likert scales with three distinct facial expressions, namely smiley, neutral and sad faces were presented below each item and picture. In response to the first interview with parents, the description of each scale was adjusted depending on the nature of the questions such as “very easy”, “sometimes easy” and “not easy”. The order of response scales was also slightly changed in every question. The format of the modified PedsQL questionnaire was firmly established after the second interview in order to obtain consistent ideas of further revisions of the questionnaire. In agreement with the findings in Phase one, two parents reported “yes-no question” is more suitable for children and adolescents with ASDs. On the other hand, one parent pointed out that there were no alternative options such as “not applicable” and “do not know” which are usually included in a survey.

In Phase two, irrelevant and missing facets of the modified PedsQL were clearly demonstrated. *Irrelevance* refers to a question that is not important to or associated with the child, and *omission* stands for elements that are important to the child’s or



adolescent's QOL but are not included in the questionnaire. Most children and adolescents in the interviews went to special schools, and a few studied at mainstream schools. The majority of parents agreed that the children usually do not receive homework. Compared to the findings from Phase one, more views on unique characteristics of social interactions in children and adolescents with ASDs were attained. As consistent with the findings of the theme of *relationships*, irrelevant aspects of the modified PedsQL questionnaire were interactions with peers and friends while questions about interactions with family and adults particularly teachers and teachers' aides who are important persons for the child's life were left out. Clarifying the type of individual activities and the level of activity and engagement were also viewed as a critical component of QOL in children and adolescents with ASDs in terms of social and physical perspectives.

**Table 4.4**

*Categories from Content Analysis of the Modified PedsQL Questionnaire*

Category	Definition	Subcategory	Examples of quotations
Rewording	The wording in the modified PedsQL that need to be changed into different words or phrases or even a new sentence	Physical Functioning	The phrase of 'to help in the house' was rephrased to 'to tidy up', 'to make your own bed' or 'to mop and vacuum' by parents.
		Social Functioning	"I (the parent) always say 'who did he (the child) talk to today' because with us it's all around talking anyway and other kids will talk to him."; The word of 'friendly' was changed to 'nice' by a parent.
		School Functioning	"I (the parent) think 'what you learned at school' rather than 'was taught'."

Responses	An answer to a question in which options such as dichotomous or rating scales are included	Yes-No	"The three categories is actually quite hard. Probably it would be easy 'yes' or 'no', and I (the parent) would think he (the child) would get that right, too."; "This is more 'yes-no' question."
		Varied Options	"The same answers for every question. It's easier but it doesn't mean that the children actually interpreting the question."; "I (the parent) noticed that the different section obviously scale is the same, right? But it varies, right? Depending on the section, I think, differs some, right?"
		Alternative Options	Another option such as 'not applicable', 'cannot answer', 'do not want to answer', 'do not know', or 'maybe' were suggested by parents.
		Order	"I (the parent) think you changed the order of these all the time, didn't you? Definitely."; "It is better to swap the orders around."; "Can you change these order around, don't you? Actually it's a good point."
Irrelevance	A question that is not important to or associated with the child	School Functioning	"It's too stressful but well that's what they suggest is don't try and do homework with kids on the spectrum, it's all, the day is over, forget it, just have some down time."; "He (the child) doesn't have homework."; "There is no schoolwork at home."

Omission	Elements that are important to the child's quality of life but are not included in the questionnaire	Social Functioning	"What about interaction with adults in his world, teachers or other caregivers such an important persons in the child's life."; "Content of 'interaction with others' such as adults, teachers or teacher aides."; "I (the parent) don't think that there's actually nothing in there about 'family'."; "I (the parent) think if you do something on family as well like 'Do you have brothers or sisters?' and 'Do you play sports with (your family)?'"
		Activity	"I (the parent) guess you could tailor it to the individual activities that the child likes doing."; "It should include 'how many times' to identify the level of activity and to ascertain how often the child is actually doing it."; "Probably something like 'What do you do in your spare time?' to make things a bit more interesting."

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In Phase two, completion rate and time for completion of the modified PedsQL were also measured (see Table 4.5). Except for one adolescent who had low-functioning ASDs and was not able to answer the questionnaire, seven children and adolescents completed the questionnaire without omitting any of the questions. Mean and standard deviation of time for completion were  $10.78 \pm 7.18$  (ranged 4.87 to 21.35) minutes.

Throughout Phase two of the interviews, enthusiastic impression and high satisfaction of the modified PedsQL questionnaire were obtained such as “pretty good”, “straightforward”, “just perfect”, and “better than the original (questionnaire)”. Parents reported that the modified PedsQL questionnaire provided them with a new viewpoint on their children and adolescents. One parent commented,

*“The first time I think (for the child) to complete (a questionnaire)... It (the modified PedsQL questionnaire) gave me every information (of the child) as well. That is good.”*

Another parent remarked,

*“I was quite surprised that he (the child) could make time for what he likes and come and do this (the modified PedsQL questionnaire).”*

The modified PedsQL questionnaire also fascinated parents to know their children’s and adolescents’ answers regarding QOL of the children and adolescents, and to see how the children and adolescents internalised themselves. It was also reported that consultation with parents to tailor the questionnaire to each child and adolescent would be essential. However, the presence of parents and their observation of the children and adolescents may prevent from reflecting the children’s and adolescents’ own perspectives, due to the fact that parents assisted the children and adolescents in suggesting answers which are more likely to be “proxy-reports”. This issue would be a major obstacle in terms of “self-reported quality of life”.

**Table 4.5***Completion Rate and Time for Completion of the Modified PedsQL Questionnaire*

<b>Child</b>	<b>Age</b>	<b>Completion Rate (%)</b>	<b>Time for Completion (minute)</b>
1	6	100	21.00
2	7	100	21.35
3	16	100	06.37
4	10	100	06.55
5	10	100	06.92
6	14	0	Not applicable <sup>a</sup>
7	6	100	8.42
8	15	100	4.87

*Note.* <sup>a</sup> The children had low-functioning ASDs.

Overall, the perception of QOL in children and adolescents with ASDs or MID and their characters were different from those in typically developing children and adolescents. Specifically, deficits in social interactions and relationships, unique perceptions, and cognitive disability significantly impact on their QOL which, in turn, affect their interpretation of and attitudes towards the original PedsQL questionnaire. The wording, format and presentation of the original PedsQL questionnaire were less appropriate for children with ASDs or MID, and we recognised the need for developing the original PedsQL questionnaire. The modification of the original PedsQL questionnaire included rewording from abstract to concrete accounts; using visuals; changing the order of response scales; and presenting individual question at a time. Although positive feedback from parents as well as children and adolescents was obtained in Phase two, wording and response options in the modified PedsQL need to be improved.

## **CHAPTER 5: DISCUSSION**

The purpose of this research was to initiate the development of a tool that could be used by children and adolescents with autism spectrum disorders (ASDs) specifically autism and Asperger syndrome or mild intellectual disability (MID) to communicate perspectives of their quality of life (QOL). The exploration included (1) the determination of content validity of a child self-report QOL inventory, Pediatric Quality of Life Inventory (PedsQL), in children and adolescent with ASDs or MID; and (2) the development of a child self-report QOL inventory, PedsQL, to be appropriate for children and adolescents with ASDs. The present study consisted of two phases. In Phase one, the content validity of the PedsQL questionnaire was examined using qualitative methods of focus groups and interviews with children and adolescents with ASDs or MID, parents and special school teachers. In Phase two, the original PedsQL questionnaire was re-developed using the findings from Phase one and adapted for use in children and adolescents with ASDs or MID. After the amendment of the original PedsQL questionnaire, the modified PedsQL questionnaire was utilised for children and adolescents with ASDs and interviews with the children and their parents were conducted. Due to the fact that the majority of the children and adolescents participated in Phase one and two were primarily diagnosed as ASDs, the data were analysed mainly in relation to the characteristics of ASDs. Nonetheless, some of the children and adolescents with ASDs also had MID. We predicted that the content validity of a child self-report of the original PedsQL questionnaire among children and adolescents with ASDs or MID would be low due to their impaired cognitive processes and language abilities. We also predicted that there would be a need to reword or redesign the original PedsQL questionnaire for use in children and adolescents with ASDs.

### **CONTENT VALIDITY OF THE PEDSQL CHILD SELF-REPORT IN CHILDREN AND ADOLESCENTS WITH ASDS OR MID**

Through thematic analysis of the data from Phase one and two, three themes of *concreteness*, *relationships* and *perception* were identified. The overall findings from Phase one and two indicated that child self-report used of the original PedsQL questionnaire was not appropriate for children and adolescents with ASDs or MID due

to a dearth of consideration to their unique nature of social dysfunction, cognitive and language impairments, and deficits in emotional recognition. As regards problems of self-reports in individuals with autism and other mental health conditions, Iarocci and McDonald (2006) demonstrated that characteristics of autism including peculiar use of language, specific hindrances to working memory, and perseveration on a subject, may limit the ability of individuals with autism to accurately report and have insight on their sensory experiences. In spite of this limitation, the value of children's own perspectives obtained from self-reports needs to be prioritised in terms of children's rights to express their opinions which should be respected, protected and fulfilled (Cremeens et al., 2006; UNICEF, 2005). In the present study, the aims of thematic analysis of the focus groups and interviews were to identify the perception of QOL measurement in children and adolescents with ASDs or MID; and to analyse the interpretation of and attitudes towards the PedsQL questionnaires in children and adolescents with ASDs or MID.

### **Important Aspects of QOL in Children and Adolescents with ASDs or MID**

Parents and one adolescent with Asperger syndrome reported that most children and adolescents with ASDs and some with MID experienced difficulty with social interactions and relationships. Through interactions with others, children start to build a "theory of mind" which refers to a coherent understanding of mental states of others such as thoughts, beliefs, desires and intentions (White, Hayes, & Livesey, 2010a). Individuals with autism, however, have a deficit in theory of mind, which may be interpreted as their particular insensitivity to social stimuli (Ropar & Peebles, 2007). In the present study, social interactions of children with ASDs were classified into four elements: interactions with "friends", "family", "adults" and "community". The majority of parents of children and adolescents with ASDs demonstrated that their children and adolescents had significant difficulties in social interactions with their friends or peers but had more interactive play with their siblings and family, and more communication with adults particularly their teachers and teacher aides. A few studies reported that children and adolescents with ASDs had at least one friend, and their relationships are often superficial but deep nor reciprocal (Carrington et al., 2003; Church et al., 2000; Travis, 1998). Church et al. (2000) found that middle school age children with Asperger syndrome established and maintained good relationships with



grandparents and other adults such as teachers with whom the children often interact at school.

Social difficulties specifically deficits in social skills and social competence are characterised as a primary feature of ASDs (Knott et al., 2006; Rao et al., 2008; Stichter et al., 2012). Social skills refer to basic abilities that allow an individual to engage in interactions, such as “eye contact” (Knott et al., 2006; Stichter et al., 2012). Social competence refers to an individual’s ability to effectively and independently engage in social interactions, and make and sustain relationships with others (Knott et al., 2006; Stichter et al., 2012). An individual with social competence is able to effectively adjust oneself to different contexts, using various social skills and behaviours (White et al., 2010c). Social impairments, including restricted interests, deficits in reciprocal communication and developmentally strained peer relationships, lead to limited social interactions among children and adolescents with ASDs (Stichter et al., 2012). These social difficulties for individuals with ASDs originate in infancy, become more with age, and remain across their lifetime (Ozonoff et al., 2005; Stichter et al., 2012). Moreover, they may affect other areas of overall functioning, such as academic and emotional development (Rao et al., 2008; Stichter et al., 2012). Therefore, it is critical to identify the content, quality and patterning of social interactions and relationships in children and adolescents with ASDs as parameters of their QOL.

Another idiosyncratic feature of relationships in children and adolescents with ASDs was the inclination to be by themselves, which refers to the state of spending time for self without any interactions with others. Abnormalities in the system of self-in-relation-to-other in individuals with autism represent separation between understanding of the self and understanding of others (Hobson, 2012; Travis, 1998). Kanner found that children with autism, around the age of six appeared to lack comprehension of the meaning of an ‘I’ in relation to a ‘you’ (Hobson, 2012). In the present study, some parents and one adolescent reported that the importance and preference of the time and activities for self instead of socialising with others. Church et al. (2000) and Jennes-Coussens et al. (2006) reported that adolescents with Asperger syndrome were likely to stay in their room alone, watch TV, play on the computer or video games, listen to music, or do reading. Similarly primary school children with high-functioning autism

preferred to spend a lot of time inside with computers and TV rather than participating in outdoor activities and a variety of sports with others (Church & Coplan, 1995).

For children and adolescents with ASDs, non-interactive activities may be more enjoyable than interactive play with friends or peers which is problematic most of the time for them. In the original PedsQL questionnaire, however, there are no items regarding non-interactive activities for self. It is reasonable to infer that the significance of an individual's activities in children with ASDs needs to be highlighted as one of the primary domain of their QOL. These unique characteristics of social functioning in children and adolescents with ASDs demonstrate the need for identifying their concepts of social interactions and relationships as well as their favourable activities which are likely to be non-interactive manner. Despite parents' desires for their children and adolescents to socially interact with others, the children and adolescents themselves may not be keen on or have difficulties with socialisation. In fact, it is evident that there were significant disparities between self-reports from children with ASDs and parent proxy-reports on social domains of QOL (Kamp-Becker et al., 2011; Sheldrick et al., 2011; Shipman et al., 2011).

### **Cognition and Perception of the PedsQL Self-Reports in Children and Adolescents with ASDs or MID**

Atypical cognitive process and deficits in language in children and adolescents with ASDs or MID considerably influence their abilities to self-report their QOL. According to Bell (2007), there are four important cognitive processes to respond a question; (1) comprehension of the question including an understanding of the terms and the task in the question; (2) retrieval of the necessary information from memory; (3) evaluation of the information; and (4) communication with the response. The present study reveals that children with ASDs tended to think concretely and have difficulty with interpreting abstract concepts, terms and phrases. Hobson (2012) reported that concrete thinking in individuals with autism may result from their cognitive impairments of establishing a conceptual and categorical framework of reference which is converted from the instant and unconnected context of experience. The patterning of concrete thinking in individuals with ASDs is more likely to depend on external circumstances rather than inner experiences (Hill et al., 2004). Children with autism, for example, often described

their experiences in relation to physical conditions rather than mental or emotional states (Lombardo, Barnes, Wheelwright, & Baron-Cohen, 2007).

Impaired working memory and executive dysfunction are other notable factors that contribute to cognitive deficits in individuals with ASDs (Iarocci & McDonald, 2006; Ozonoff et al., 2005; Stichter et al., 2012). Working memory refers to the structures and processes used for temporarily storing and manipulating information; and executive function is defined as the ability to control and coordinate other cognitive processes including planning, working memory, attention, inhibition and cognitive flexibility (Ozonoff et al., 2005; Ozonoff et al., 1991). Impairment in executive function is related to lack in anticipation of long-term consequences of behaviour, and difficulties with self-reflecting and self-monitoring (Ozonoff et al., 1991). Dysregulation of executive functioning among children and adolescents with ASDs may affect their abilities to evaluate and report their satisfaction of their QOL. Moreover, children with ASDs had difficulty with interpreting language in flexible manner, in which they cannot understand the meaning of a word with anything other than literal meaning (Church & Coplan, 1995; Happe, 1993).

It is also reported that children with autism had problems in verbal communication specifically in the use of pronouns (Hobson et al., 2006). The findings showed that the use and selection of words in the original PedsQL questionnaire needed to be improved for children and adolescents with ASDs, specifically careful considerations of unique cognitive process (i.e. concrete thinking) and impaired language ability (i.e. literal meaning of a terms). Furthermore, in-depth and simple accounts of each item would be required for children and adolescents with ASDs to comprehend the meaning of each question. Unique characteristics of perception in children and adolescents with ASDs also impacted on the original PedsQL questionnaire. In the present study, three subthemes of “visuals”, “feelings”, and “behaviour” were identified under the theme of *perception*. Among individuals with autism, visual thinking is superior to verbal thinking (Grandin, 2000). Visual thinking is defined as the ability to perceive and think about visual objects and spatial relationships in two and three dimensions while verbal thinking refers to the ability to use language to store and categorise memories as associated events (Grandin, 2000). Considering that the unimpaired system of visual

thinking and impaired language ability in individuals with ASDs, the use of pictures to depict the description of each question in a questionnaire may be advantageous.

Emotion recognition is one of the primary deficits in individuals with ASDs (Stichter et al., 2012), which had considerable effect on the capability of responding to self-reports. Emotion recognition is defined as the ability to identify affective expressions and following emotions in the self and others (Stichter et al., 2012). Although children with ASDs are able to recognise and express simple emotions such as happiness, sadness, and anger, it is hard to interpret and convey feelings relating to their episodes in a coherent manner (Losh & Capps, 2006). The present study found that the children and adolescents with ASDs were able to report their emotions (i.e. fear, sadness, anger and happiness) and their feelings responding to each question in the modified PedQL questionnaire. The nature of their mental states was instable and fluctuated in a day. Parents described that they were sometimes uncertain about the children's feelings and emotions. These findings highlighted that self-reports would be a privileged method to access to the children's and adolescents' own inner states.

However, there were a few problems regarding the use of emotive languages and facial expressions. Parents from Phase one of the interviews reported that displaying only emotive language such as "scared", "sad", "angry" and "happy" was difficult for children and adolescents with ASDs to understand the state of each emotion. This issue might be solved by presenting a pictorial facial expression of each emotion together with a script. Use of facial expressions used in response options was sometimes problematic for children and adolescents with ASDs because their own feelings and the facial expressions did not always match each other. For example, in the modified PedsQL questionnaire, social interactions with other kids were viewed as a "positive" position. Therefore, if a child has "a lot" interactions, the response of "a lot" is shown with "smiley" face. As it was discussed in the previous section, the children and adolescents with ASDs often become distressed about social interactions. They do not necessarily feel happy like "smiley" face when they communicate with others. This mismatch between the child's real feeling and the facial expression shown in response scales needs to be resolved in future studies in order not to cause misinterpretation from children and adolescents with ASDs.

The present study found that attitudes of the children and adolescents with ASDs towards the modified PedsQL questionnaire were relevant to core symptoms of ASDs. The behaviour of individuals with ASDs seems rigid and inflexible (Church & Coplan, 1995; Ozonoff et al., 1991), and obsessive and compulsive (Church et al., 2000). Individual with ASDs often have routine and rituals in precise detail and feel extremely anxious or frustrated over trivial changes (Ozonoff et al., 1991). They also focus on restricted interests or engage in stereotyped and repetitive behaviours (Church et al., 2000; Ozonoff et al., 1991; Stichter et al., 2012). Consistent with these features, parents reported that children and adolescents with ASDs made a routine or pattern to select the same response or place such as “smiley” face or “in the right” side regardless of their own feelings. Some children and adolescents preferred “positive” answers such as “a lot” or “yes” rather than “negative” such as “not at all” or “no”. It was feasible to prevent repetitive behaviour in children and adolescents with ASDs by rearranging the order of response scales, which have already trialled in the modified PedsQL questionnaire. Nonetheless, it is not clear from the present findings whether their repetitive behaviour and preference influence their responses.

#### **DEVELOPMENT OF THE PEDSQL SELF-REPORTS FOR CHILDREN AND ADOLESCENTS WITH ASDs OR MID**

The aims of content analysis of the focus groups and interviews were to assess the appropriateness of the wording, format and presentation of the PedsQL questionnaire in children and adolescents with ASDs or MID; and to identify relevant and irrelevant dimensions of the PedsQL questionnaire to children and adolescents with ASDs or MID. Through content analysis of the focus groups and interviews from Phase one, we recognised the necessity for the development of the original PedsQL questionnaire. The original PedsQL questionnaire was modified in the wording, format and presentation to meet the needs of children and adolescents with ASDs. In Phase two, the modified PedsQL questionnaire was piloted on children and adolescents with ASDs and the interviews with the children or adolescents and their parents were analysed. Mean of time for completion ( $10.78 \pm 7.18$  minutes) of the modified PedsQL questionnaire was longer than the standard of four minutes for the original PedsQL (Varni, n.d.). However, the result was similar to another study of children with ASDs using the original PedsQL

questionnaire which reported between 7 and 10 minutes (Kuhlthau et al., 2010). With regard to completion rate, seven out of eight participants completed the questionnaire without leaving out any questions.

The categories of *rewording*, *responses* and *presentation* were related to impairments in cognitive functioning and language in children and adolescents with ASDs or MID. Individuals with ASDs have impaired weak central coherence which refers to the ability to integrate information from a variety of processes such as perception, attention, linguistic and semantic to draw higher-level of meaning (Iarocci & McDonald, 2006). In Phase one, parents reported that multiple information and complex structures which were used in the original PedsQL questionnaire would make children and adolescents with ASDs or MID confused. In particular, it was proposed that sentences be shown in a declarative rather than interrogative manner; fewer response scales be practical; and one question be shown at a time instead of listing many questions in one page.

Regarding linguistic issues, it was suggested that words and phrases be used in a literal manner which means the “defined” or “normal” meaning of the terms without implication and inference. Bell (2007) recommended that questionnaires for children should be short in length, consist of short sentences, use straightforward language, present in direct and specific manner, but avoid using complex structure and negative formulations. It is also evident that individuals with ASDs were not able to use language in a flexible manner (Church & Coplan, 1995; Happe, 1993). Carrington et al. (2003) found that a child with autism had difficulty in comprehending the words used in the question, and interviewers needed to prompt and reword questions in most interviews. In the present study, the original PedsQL questionnaire was reworded and reformatted, and pictures were inserted in each question. After Phase two, we recognised that a few phrases would be required to be reworded because expressions of the phrases were not concrete enough for children and adolescents with ASDs.

In terms of *responses*, new ideas were proposed from parents such as varied answers according to the type of questions, and alternative options if the question was “not applicable” to the child and adolescent. Parents were consistently reported that the type of “yes-no question” was more straightforward for children and adolescents with ASDs. In general, the format, wording and presentation of the original PedsQL questionnaire

were less appropriate for children and adolescents with ASDs or MID, and required improvement in order to pilot the questionnaire for the children and adolescents. The modified PedsQL questionnaire was more appropriate for children and adolescents with ASDs; however, feedback from parents and children and adolescents in Phase two demonstrated that the use of concrete wording and the type of response options may need further amendments to increase the accuracy of responses from the children and adolescents.

The domains of social and school functioning were the most discussed contextual issues with regard to *irrelevance* and *omission*. Basically the original and modified PedsQL questionnaire had the same interpretation of the questions in both domains of social and school functioning even though all of the questions in the original PedsQL questionnaire were re-worded in the modified PedsQL questionnaire. It was clearly reported that schoolwork between special and mainstream schools were different and there was no homework in special schools. Due to deficits in social interactions and relationships in children and adolescents with ASDs, the questions about social interactions with peers in the domain of social functioning were not relevant to children and adolescents with ASDs. Instead, questions about interactions with family and teachers or teacher aides seemed to be more relevant and important to their life. Moreover, non-interactive activities and their own time were pertinent aspects for children and adolescents with ASDs but missed in both questionnaires. These findings indicated that the content of the original and modified PedsQL questionnaire was not always relevant to QOL in children and adolescents with ASDs or MID. Supplemental QOL domains specific to children and adolescent with ASDs are required.

In summary, the content validity of a child self-report of the original PedsQL questionnaire among children and adolescents with ASDs or MID was low. The concept of the original PedsQL questionnaire covers most aspects of QOL in children and adolescents with ASDs or MID including physical, emotional, social and school functioning. However, the content of the original PedsQL questionnaire needed to be developed for children and adolescents with ASDs or MID as their cognition and perception are unique and different from typically developing children and adolescents.

The findings indicated that the core ASDs symptoms of impaired social interaction, communication, repetitive behaviour, and restrictive interests and activities considerably impact on QOL for this population, and consequently QOL measurement. This indicates that the PedsQL questionnaire may assess “health status” or “functional status” instead of “quality of life” due to the similarities between assessment criteria. In this regard, ASDs-specific QOL questionnaire may be required to develop, particularly on the domain of social functioning which focuses on “what they like to do” or “what they are interested in”. In addition, formatting and presentation style of the original PedsQL questionnaire was less suitable for children and adolescents with ASDs. The modified PedsQL questionnaire (see Appendix I for complete proofs), on the other hand, tailored the structure of the original PedsQL questionnaire for children and adolescents in terms of wording, formatting and presentation style. The modified PedsQL questionnaire gave parents fascinating insights into their children’s internalisation as well as QOL. Although most children and adolescents with ASDs completed the modified PedsQL questionnaire, presence of parents and parental assistance with the questionnaire would be the main drawback of self-reporting QOL. It is also acknowledged that the PedsQL questionnaire will only measure the items that it contains and may not capture other complex social phenomenon that accompanies these children and adolescents as well as families lives.



## **CHAPTER 6: CONCLUSION**

This research employed a qualitative descriptive approach to answer the following question: “What is an appropriate QOL measurement for children and adolescents with ASDs or MID?” From the review of literature, the most commonly used QOL measure in children and adolescents with ASDs, the PedsQL child self-report, was selected as the instrument of choice.

### **SUMMARY OF FINDINGS**

The findings showed that the original PedsQL questionnaire was not appropriate for children and adolescents with ASDs or MID compared to the modified PedsQL questionnaire. The main issues regarding the content validity of the original PedsQL questionnaire were (1) a number of missing and unrelated questions on the domain of social functioning; (2) use of abstract concepts and terms; and (3) monotonous format, response options and presentation style. The core symptoms of ASDs such as impaired social interaction, repetitive behaviours and limited interests are strongly associated with the key factors of social functioning in the PedsQL questionnaires. In this regard, the PedsQL questionnaires need to incorporate ASDs-specific or child-specific components to assess “quality of life” in children and adolescents with ASDs instead of their symptoms or functional impairments. Concrete thinking is superior to abstract thinking in children and adolescents with ASDs or MID due to their impairments in cognitive and language ability. Use of simple, concrete and explicit terms and phrases is vital for the PedsQL questionnaires. Visuals are also important elements in terms of the children’s and adolescents’ comprehension of and engagement with the PedsQL questionnaires. The format, response options and presentation of the modified PedsQL questionnaire (see Appendix I for complete proofs) were improved by use of pictures, facial expressions and one-by-one presentation, which enabled children and adolescents with ASDs to complete the modified PedsQL questionnaire. The development of the appropriate PedsQL self-report for children and adolescents with ASDs or MID may increase the accuracy of their self-appraisal and reflect their own views on QOL. However, pursuing an ‘appropriate’ questionnaire may result in creating a ‘non-generic’ but a ‘customised’ questionnaire for this population.

## **LIMITATIONS**

There were two main limitations: Participants and procedure. The researchers attempted to provide a broader picture on the appropriateness of the PedsQL questionnaire for children and adolescents with cognitive disability in Phase one. However, the types of disorder in children and adolescents in Phase one were mostly autism spectrum disorders (ASDs) and slightly moderate to mild intellectual disability (MID). Therefore, the researchers decided to focus on children and adolescents with ASDs for participants in Phase two. All children and adolescents who participated in Phase two were primarily diagnosed as ASDs even though some of them also had intellectual impairments. In addition, most participants had comorbidities such as attention deficit hyperactivity disorder and global developmental delay. Although different types of intellectual disability were involved, the data from Phase one and two were analysed in relation to mainly ASDs and MID.

With regard to the research procedure, the facilitator and the interviewer were not the same in each focus group and interview due to the researchers' schedule. However, the procedure of the focus groups and interviews were discussed with the team to ensure the consistency across data analysis. Parental assistance was given to younger children and one adolescent completed the modified PedsQL questionnaire by himself. Although the purpose of this research was not collecting self-reports from children and adolescents with ASDs or MID, the findings could have been missed obtaining greater insight into the child's ability to complete the modified PedsQL questionnaire. Despite these limitations, the findings provided valuable perception and feedback on the development of the PedsQL self-report for children and adolescents with ASDs or MID.

## **FUTURE RESEARCH**

The findings from the present study indicate that further development of the PedsQL self-report for children and adolescents with ASDs may be required with respect to rewording, design of response options and ASDs-specific social domains. For future studies, it is also worthwhile to investigate different modes of administration such as the use of qualitative interviews and electronic devices. Considered that electronic devices

such as the iPad and DynaVox are utilised as educational tools for children and adolescents with ASDs, it may be practical to collect data using these devices.

It is noted that there have been media releases that in the American Psychiatric Association's new diagnostic manual, DSM-V, which will be published in May 2013, the term of "Asperger disorder (Asperger syndrome)" will be dropped and incorporated under the term of "autism spectrum disorder" (Rehel, 2012).

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## **GLOSSARY**

### **Autism Spectrum Disorders (ASDs):**

ASDs, also known as Pervasive Developmental Disorders (PDDs) include autistic disorder, Asperger disorder, pervasive developmental disorder not otherwise specified (PDD-NOS), Rett disorder, childhood disintegrative disorder and atypical autism (APA, 2000).

### **Children and Adolescents (Young People):**

The definition and use of “children” and “adolescents” were not clear and varied across literature. This research defined the age of “children” between five and 12 years, and “adolescents” or “young people” between 13 and 18 years old. However, some articles used the word of “children” who are aged above 12 years or “adolescents (young people)” above 18 years of age. Therefore, we used the same words that were utilised in the literature regardless of our definition of age. This research distinguished between “children” and “adolescents” as possible depending on the age of participants. This is because we recognised that age and developmental stages are important factors to determine the appropriateness of the PedsQL questionnaire.

### **Quality of Life (QOL):**

An individual’s perception of their position in life, in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns (WHOQOL Group, 1993, p. 153)

### **Health:**

A state of complete physical, mental and social well-being and not merely the absence of disease or infirmity (WHO, 1946, p. 1315)

### **Health-Related Quality of Life (HRQOL):**

The impact of health, illness and disorder on QOL (Eiser & Morse, 2001)

## **APPENDICES**

## APPENDIX A

### Ethical Approval



# MEMORANDUM

## Auckland University of Technology Ethics Committee (AUTEC)

To: Erica Hinckson  
 From: **Dr Rosemary Godbold** Executive Secretary, AUTEC  
 Date: 19 September 2011  
 Subject: Ethics Application Number 11/206 **SNAP! For health: Content validity of the PEDS-QL questionnaires in children with cognitive disability.**

Dear Erica

Thank you for providing written evidence as requested. I am pleased to advise that it satisfies the points raised by the Auckland University of Technology Ethics Committee (AUTEC) at their meeting on 22 August 2011 and I have approved your ethics application. This delegated approval is made in accordance with section 5.3.2.3 of AUTEC's *Applying for Ethics Approval: Guidelines and Procedures* and is subject to endorsement at AUTEC's meeting on 10 October 2011.

Your ethics application is approved for a period of three years until 19 September 2014.

I advise that as part of the ethics approval process, you are required to submit the following to AUTEC:

- A brief annual progress report using form EA2, which is available online through <http://www.aut.ac.nz/research/research-ethics/ethics>. When necessary this form may also be used to request an extension of the approval at least one month prior to its expiry on 19 September 2014;
- A brief report on the status of the project using form EA3, which is available online through <http://www.aut.ac.nz/research/research-ethics/ethics>. This report is to be submitted either when the approval expires on 19 September 2014 or on completion of the project, whichever comes sooner;

It is a condition of approval that AUTECH is notified of any adverse events or if the research does not commence. AUTECH approval needs to be sought for any alteration to the research, including any alteration of or addition to any documents that are provided to participants. You are reminded that, as applicant, you are responsible for ensuring that research undertaken under this approval occurs within the parameters outlined in the approved application.

Please note that AUTECH grants ethical approval only. If you require management approval from an institution or organisation for your research, then you will need to make the arrangements necessary to obtain this. Also, if your research is undertaken within a jurisdiction outside New Zealand, you will need to make the arrangements necessary to meet the legal and ethical requirements that apply within that jurisdiction.

When communicating with us about this application, we ask that you use the application number and study title to enable us to provide you with prompt service. Should you have any further enquiries regarding this matter, you are welcome to contact Charles Grinter, Ethics Coordinator, by email at [ethics@aut.ac.nz](mailto:ethics@aut.ac.nz) or by telephone on 921 9999 at extension 8860.

On behalf of AUTECH and myself, I wish you success with your research and look forward to reading about it in your reports.

Yours sincerely

Dr Rosemary Godbold  
**Executive Secretary**  
**Auckland University of Technology Ethics Committee**

Cc: Chris Krageloh, Tineke Water, Tania Brown

## APPENDIX B

### Child Information Sheet and Assent Form



## Child Information Sheet and Assent Form

(Parents/Legal Guardians please read this to your children)

## **SNAP for Health: Content validity of the PEDS-QL questionnaires**

Hello my name is Erika Ikeda!

I would like to spend some time with you and your family to ask some questions about a questionnaire on how you feel about your health. We would like to find out if the questions are easy to understand or not and what you think of them.

Is that okay? Please circle how you feel about this



Or



I will ask you to fill out a short questionnaire about how you are feeling at the moment about your health. I will ask you to tell me what you think about my questionnaire.

Is this okay with you?



Or



If you have any concerns regarding the project do not hesitate to contact myself or the project supervisor.

Erika Ikeda

[eikeda@aut.ac.nz](mailto:eikeda@aut.ac.nz)

021-259-3897

Dr. Erica Hinckson

[erica.hinckson@aut.ac.nz](mailto:erica.hinckson@aut.ac.nz)

921-9999 ext 7224

**Approved by the Auckland University of Technology Ethics Committee on 19 September 2011, AUTECH Reference number 11/206**

Thank you for completing this form – would you please ask your parent/legal guardian to sign here and cut this off and give to your teacher tomorrow at school.

(Signature)

.....

(Date)

.....

If you feel that you understand what the project is about please give this form back to the researcher.

## APPENDIX C

## Parent Information Sheet

# Parent Information Sheet

**Date Information Sheet Produced:**

8<sup>th</sup> August 2011

**Project Title**

SNAP! for Health: Content validity of the PEDS-QL™ questionnaires in children with cognitive disabilities

**An Invitation**

My name is Dr Erica Hinckson and together with the research team (Dr Chris Krageloh, Dr Tineke Water and Erika Ikeda) would like to invite you and possibly your child / young person to take part in a research project. The aim of this research is to assess the appropriateness of a quality of life questionnaire. You and your child / young person's participation are voluntary and even if you choose to participate you may withdraw from the study at any time with no adverse consequences.

**What is the purpose of this research?**

This project is part of a larger project titled *Snap! for Health: A family and school approach for special needs children*. The aim of *Snap! for Health* is to establish a programme that will help improve health and wellbeing through physical activity and nutrition of children with special needs within the family and school environment. During the Snap project we will be using questionnaires to assess families and children / young people's perceptions of their quality of life. To make sure the questionnaires are suitable, we would like to receive your feedback and input on the appropriateness of the questions asked and changes that could be made.

**How was I identified and why am I being invited to participate in this research?**

You will have heard about this study through your child's school or through an organisation related to children / young people with cognitive disability. You have been invited to participate because you have a child or young person who has a cognitive disability and you are interested and willing to participate in this study.

**What will happen in this research?**

We would like to invite you to attend a focus group session. Prior to commencing the focus group session we will ask you to complete two quality of life questionnaires. In the focus group session we will ask you to give us feedback on the appropriateness of the questionnaires. We will use this feedback to amend the questionnaires. Following the focus group session we may ask if you would be willing to join a panel to assess the appropriateness of the amended questionnaires.

We may also ask if the questionnaires can be trialled with your child / young person. If you agree to this we would ask that you explain the study to your child / young person and seek assent for their involvement. A researcher would come and conduct an individual interview with you and your child / young person in a place of your choice. The researcher would also check with your child / young person that they were happy to participate. Your child / young person would be asked to complete the questionnaire and then the researcher would ask them about what they thought of the questions. The researcher would also ask you what you thought of the questionnaire.

### **What are the discomforts and risks?**

The discomforts and risks as a parent / family member in the focus group interviews and completing the questionnaires is that it may highlight areas of your life that you are not happy with at present. You may find this uncomfortable.

The discomforts for your child / young person are that they may feel uncomfortable with a change in routine or the unfamiliar face of the researcher. They may also feel a range of feelings (such as frustration) if the questions in the questionnaire are not clear.

### **How will these discomforts and risks be alleviated?**

As a parent or family member if you do experience discomfort or stress when completing the questionnaire (due to the issues this might raise) three free counselling sessions can be offered via AUT counselling services (09 921 9998-North Shore Campus). You do not need to answer any question that you do not wish to and may withdraw from the study at any time up until the completion of data collection.

Your child / young person will have a family member with them and if they are uncomfortable with the interview another interview can be rescheduled. You or they may also choose to not answer any question or to withdraw from the study at any time (up until the completion of data) with no adverse consequences. The researchers will try to meet you and your child / young person prior to interview so that the researcher is a familiar face.

### **What are the benefits?**

Although quality of life questionnaires have been used in children / young people and families who have a chronic disease they have not been used in children / young people with cognitive disabilities and their families. The benefits would be to know if these questionnaires are appropriate so that they can be used in SNAP for Health study or other studies. Although there may be no direct benefits to you, there may be a benefit to the development of programs that will support children / young people and their families in healthy life styles. You may find it useful to have a say in the development of such programmes.

### **How will my privacy be protected?**

All information that you give us will be treated as confidential. Only the researchers will know who you are, have access to the data you have given them, and will make sure that the data is kept in a secure place. In the final report and any publications of the findings, your name will be substituted with a pseudonym.



### **What are the costs of participating in this research?**

The main cost of participating in this study is your personal time for attending the focus group session (60-90 minutes); possibly a second session should you agree to join the panel to assess the amended questionnaire (60 minutes); and possibly if you agree an individual interview with you and your child / young person (60 minutes)

### **How do I agree to participate in this research?**

You have two weeks to decide if you would like to participate. If you would like to participate then you can contact the researcher on the details below. When you come to the focus groups and / or individual interviews the researcher will check again that you and your child / young person are happy to be part of the study. If you agree the researcher will ask you to sign a consent form (for you and / or your child / young person) and for your child / young person to sign an assent form.

### **Will I receive feedback on the results of this research?**

You will have the opportunity to view and approve the data gathered from you prior to the completion of the study. If you would like to receive a copy of the final report you can indicate this on the consent form.

### **What do I do if I have concerns about this research?**

Any concerns regarding the nature of this project should be notified in the first instance to the Project Supervisor, Dr. Erica Hinckson, [erica.hinckson@aut.ac.nz](mailto:erica.hinckson@aut.ac.nz), 921 9999

Concerns regarding the conduct of the research should be notified to the Executive Secretary, AUTEK, Rosemary Godbold, [rosemary.godbold@aut.ac.nz](mailto:rosemary.godbold@aut.ac.nz), 921 9999

### **Whom do I contact for further information about this research?**

#### ***Project Supervisor Contact Details:***

Dr. Erica Hinckson, [erica.hinckson@aut.ac.nz](mailto:erica.hinckson@aut.ac.nz), 921 9999

Approved by the Auckland University of Technology Ethics Committee on 19 September 2011, AUTEK Reference number 11/206

## APPENDIX D

### Principal Information Sheet

# Principal Information Sheet



#### Date Information Sheet Produced:

8<sup>th</sup> August 2011

#### Project Title

SNAP! for Health: Content validity of the PEDS-QL™ questionnaires in children with cognitive disabilities

#### An Invitation

My name is Dr Erica Hinckson and together with the research team (Dr Chris Krageloh, Dr Tineke Water and Tania Brown) would like to invite you and possibly your child / young person to take part in a research project. The aim of this research is to project that assess the appropriateness of a quality of life questionnaire. Your participation is voluntary and even if you choose to participate you may withdraw from the study at any time with no adverse consequences.

#### What is the purpose of this research?

This project is part of a larger project titled *Snap! for Health: A family and school approach for special needs children*. The aim of *Snap! for Health* is to establish a programme that will help improve physical activity levels of children with special needs within the family and school environment. During the Snap project we will be using questionnaires to assess families and children / young people's perceptions of their quality of life. To make sure the questionnaires are suitable, we would like to receive your feedback and input on the appropriateness of the questions asked and changes that could be made.

#### How was I identified and why am I being invited to participate in this research?

You will have heard about this study through your school or through an organisation related to children / young people with cognitive disability. You have been invited to participate because you work with children or young people who have a cognitive disability and you are interested and willing to participate in this study.

#### What will happen in this research?

We would like to invite you to attend a focus group session. In the focus group session we will ask you to give us feedback on the appropriateness of the questionnaires. We will use this feedback to amend the questionnaires.

#### **What are the discomforts and risks?**

You do not have to answer any questions that you do not feel comfortable. You may withdraw from the study at any time up until the completion of data collection. Confidentiality will be maintained through the use of pseudonyms and aggregating of the data. There will be no risk of you being identified in the final report.

#### **How will these discomforts and risks be alleviated?**

If the discussion does raise any discomfort or stress for you the researchers the researchers have a list of other organisations they could refer you to. You do not need to answer any question that you do not wish to and may withdraw from the study at any time up until the completion of data collection

#### **What are the benefits?**

Although quality of life questionnaires have been used in children / young people and families who have a chronic disease they have not been used in children / young people with cognitive disabilities and their families. The benefits would be to know if these questionnaires are appropriate so that they can be used in SNAP for Health study and other studies. Although there may be no direct benefits to you, there may be a benefit to the development of programs that will support children / young people and their families in healthy life styles. You may find it useful to have a say in the development of such programs.

#### **How will my privacy be protected?**

All information that you give us will be treated as confidential. Only the researchers will know who you are, have access to the data you have given them, and will make sure that the data is kept in a secure place. In the final report and any publications of the findings, your name will be substituted with a pseudonym.

#### **What are the costs of participating in this research?**

The main cost of participating in this study is your personal time for attending the focus group session (60-90 minutes).

#### **How do I agree to participate in this research?**

You have two weeks to decide if you would like to participate. If you would like to participate then you can contact the researcher on the details below. When you come to the focus groups the researcher will check again that you are happy to be part of the study. If you agree the researcher will ask you to sign a consent form.

#### **Will I receive feedback on the results of this research?**

You will have the opportunity to view and approve the data gathered from you prior to the completion of the study. If you would like to receive a copy of the final report you can indicate this on the consent form.

#### **What do I do if I have concerns about this research?**

Any concerns regarding the nature of this project should be notified in the first instance to the Project Supervisor, Dr. Erica Hinckson, [erica.hinckson@aut.ac.nz](mailto:erica.hinckson@aut.ac.nz), 921 9999

Concerns regarding the conduct of the research should be notified to the Executive Secretary, AUTECH, Rosemary Godbold, rosemary.godbold@aut.ac.nz, 921 9999

**Whom do I contact for further information about this research?**

***Project Supervisor Contact Details:***

Dr. Erica Hinckson, erica.hinckson@aut.ac.nz, 921 9999

Approved by the Auckland University of Technology Ethics Committee on 19 September 2011, AUTECH Reference number 11/206

## *APPENDIX E*

### Permission to Access Students from the School Form

## Permission to Access Students from the School



*Project title:* **SNAP for Health: Content validity of the PEDS-QL™ questionnaires in children with cognitive disabilities.**

*Project Supervisor:* **Erica Hinckson**

I/We have read and understood the information provided about this research project (Information Sheet dated 8 August 2011).

- I/We understand that if I/we do not agree to participate in this research, no further information will be obtained.
- I/We have had an opportunity to ask questions and to have them answered.
- I/We agree to allow access to the school for the purposes of this research.

(Please see attached sheet)

Name of School:

.....

.....  
.....

Principal signature:

.....

Principal name:

.....

Principal Contact Details:

Email:.....

Phone:.....

Date:  
.....

***Approved by the Auckland University of Technology Ethics Committee on 19  
September 2011, AUTEC Reference number 11/206***

*Note: The School should retain a copy of this form.*

*APPENDIX F***Parent/Guardian Consent Form****Parent/Guardian Consent Form**

**Project title:** *SNAP! for health: Content validity of the PEDS-QL™ questionnaires in children with cognitive disability*

**Project Supervisor:** Dr Erica Hinckson

- ☐ I have read and understood the information provided about this research project in the Information Sheet.
- ☐ I have had an opportunity to ask questions and to have them answered.
- ☐ I understand that notes will be taken during the interviews and that they will also be audio-taped and transcribed.
- ☐ I understand that I may withdraw my child/young person and/or myself or any information that we have provided for this project at any time prior to completion of data collection, without being disadvantaged in any way.
- ☐ If my child/young person and/or I withdraw, I understand that all relevant information including tapes and transcripts, or parts thereof, will be destroyed.
- ☐ I agree to my child/young person taking part in this research.
- ☐ I wish to receive a copy of the report from the research (please tick one):

Yes ☐ No ☐

Child/children's name/s :

.....

Parent/Guardian's signature:

.....

Parent/Guardian's name:

.....

Parent/Guardian's Contact Details (if appropriate):

.....

.....

Date:

.....

***Approved by the Auckland University of Technology Ethics Committee on 19 September 2011, AUTEK Reference number 11/206***



## APPENDIX G

### Focus Group Consent Form

## Focus Group Consent Form



*Project title:* **SNAP! for Health: Content validity of the PEDS-QL™ questionnaires in children with cognitive disability**

*Project Supervisor:* **Dr Erica Hinckson**

*Researchers:* **Dr Chris Krageloh, Dr Tineke Water, Erika Ikeda**

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- ☐ I have read and understood the information provided about this research project in the Information Sheet.
- ☐ I have had an opportunity to ask questions and to have them answered.
- ☐ I understand that identity of my fellow participants and our discussions in the focus group is confidential to the group and I agree to keep this information confidential.
- ☐ I understand that notes will be taken during the focus group and that it will also be audio-taped and transcribed.
- ☐ I understand that I may withdraw myself or any information that I have provided for this project at any time prior to completion of data collection, without being disadvantaged in any way.
- ☐ If I withdraw, I understand that while it may not be possible to destroy all records of the focus group discussion of which I was part, the relevant information about myself including tapes and transcripts, or parts thereof, will not be used.
- ☐ I agree to take part in this research.
- ☐ I wish to receive a copy of the report from the research (please tick one):  
 Yes ☐ No ☐

Participant's signature:

.....

Participant's name:

.....

Participant's Contact Details (if appropriate):

.....

.....

Date:

.....

***Approved by the Auckland University of Technology Ethics Committee on 19 September 2011, AUTECH Reference number 11/206***

*Note: The Participant should retain a copy of this form.*

*APPENDIX H*

**Comparison of Items between the Original and Modified PedsQL Questionnaire**

**(Available upon request)**

*APPENDIX I***The Modified PedsQL Child Self-Report**  
**(Available upon request)**