

**The Development and Testing of an Oral Health-Related
Quality of Life Measure for Children/Adolescents with
Down Syndrome
(OH-QOLADS)**

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



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Abstract

Background: Down Syndrome is the most common genetic cause of intellectual disabilities. In addition, it is the most common chromosomal anomaly among live-born infants. Individuals with this condition exhibit special oro-facial characteristics that increase their risk of oral conditions. The impact of oro-facial conditions on individuals may be related closely to their oral health (such as pain, discomfort, and in severe cases tooth loss), but can also extend to broader effects on personal relationships, emotional status and Quality of Life (QoL). However, there is very little research on the way oral health affects QoL of people with Down Syndrome.

Aim: The aim of this study was to develop and test an Oral Health-Related Quality of Life (OHRQoL) instrument among children/adolescents with Down Syndrome.

Methods: The study entailed two phases. In Phase One and in order to develop the instrument, interviews with 20 mothers of children with Down Syndrome were conducted to explore their perceptions of how oral health of their children impacted their lives (i.e. socially, emotionally) and the life of a family as a whole. Analysis of these interviews along with the literature review informed the formulation of the OHRQoL measure. In Phase Two, the developed measure was validated and tested among 97 mothers and their children with Down Syndrome. Clinical examination of oral health status of group of children/adolescents with Down Syndrome whose mothers answered the questionnaire was also conducted for the questionnaire validation purposes.

Results: Analysis of mothers' interviews helped in identifying the dimensions of impacts of child's oral health on different aspects of the child and family's QoL that resulted in a total of 20 items on child's OHRQoL, and 10 items on family's OHRQoL. Since the instrument is on its developmental stage and in order to capture any impact occurrence, each identified item was collected at two time frames: Ever-happened and happened Last-year. Results of phase-two showed that 82% of children had experienced at least one oral impact on their lives, and 77% of mothers reported at least one impact of their children's oral health on the family's QoL.

Results also showed that the developed measure has good psychometric properties; Cronbach's Alpha of the item-total correlation of the child's OHRQoL was 0.909 for 'Ever-happened', and 0.902 for 'Last-year'. And for the family's OHRQoL, the Cronbach's Alpha ranged from 0.828 'Ever-happened' to 0.807 for impacts experienced 'Last-year'. For construct validity, findings revealed significant correlations between subjective health indicators and child's OHRQoL and family's QoL. The new measure also showed its ability to discriminate between different clinical groups.

Conclusion: This is the first study to develop and validate an OHRQoL measure for use among children/adolescents with Down Syndrome. Oral health of children/adolescents with Down Syndrome had negative impacts on different aspects of their lives and that on their family. Further studies are needed to further validate this instrument to other cultures/populations, and explore the intensity of these impacts and how they might affect the rehabilitation process of the existing disability.

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LIST OF ABBREVIATIONS

COHQOL	Child Oral Health Quality of Life
COSMIN	COnsensus-based Standards for the selection of health Measurement Instruments
CPQ	Child Perceptions Questionnaire
ELDQOL	Learning Disabilities Quality of Life scale
FIS	Family Impact Scale
ECOHIS	Early Childhood Oral Health Impact Scale
HRQoL	Health-related Quality Of Life
HUI2	Health Utilities Index Mark 2
HUI3	Health Utilities Index Mark 3
ICF	The International Classification of Functioning, Disability and Health
ICHD	International Classification of Impairments, Disabilities, and Handicaps
IQ	Intelligent Quotient
MOH	Ministry of Health
OHIP	Oral health Impact Profile
OH-QOLADS	Oral Health-related Quality Of Life for Adolescents with Down Syndrome
OHQoL-UK	Questionnaire of the Oral Health-related Quality of Life in the UK
OHRQoL	Oral Health-Related Quality of Life
P-CPQ	Parental- Caregivers Perceptions Questionnaire
QoL	Quality of Life
QoL-Q	Quality of Life Questionnaire
QQoL	Questionnaire on Quality of Life
WHO	World Health Organization
WHOQOL-Dis	World Health Organization Quality of Life Disability group

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Chapter 1 INTRODUCTION

The number of people with disabilities is increasing in the world; mainly because of their higher survival rates from advances in medical and social care services (Freedman et al., 2004; Van Baar et al., 2005). Rates of disability are also increasing due to population ageing and increases in chronic health conditions (WHO, 2014). One consequence of the increased number of people with disability is that the need for costly medical and oral health care will also increase. Research has also shown that compared to the general population, people with disabilities experience poorer health and inferior access to high quality health services (Elliott et al. 2003; Graham 2005; Ouellette-Kuntz 2005; Krahn et al. 2006; Nocon 2006; Emerson & Baines 2010; O'Hara et al. 2010). There is urgent need to address this public health issue, and plan and implement effective health promoting activities to ensure better service provision, secure the lives of this vulnerable group of the community, and avoid the increasing gap in health.

There are diverse types of disabilities. This project focuses mainly on people with intellectual disabilities and more specifically individuals with Down Syndrome, because it is the most common genetic cause of intellectual disabilities (Van-Trotsenburg et al., 2006). This research will focus on their general health, oral health and their Oral Health-Related Quality of Life (OHRQoL). The reason for choosing people with Down Syndrome is that they have specific oro-facial characteristics that increase their risk of developing oral conditions. Systemic dysfunction among this population may predispose them to oral disease, and oral disease may in turn aggravate systemic diseases. Reviews on Down Syndrome reveal that people suffering from it are particularly prone to oro-facial disorders such as: periodontal disease, malocclusion and soft tissue disturbances such as protruded tongue or inverted lips (Desai & Fayetteville, 1997; Fiske & Shafik, 2001; Hennequin et al., 1999). These oro-facial conditions may impact on the people's oral health (such as pain, discomfort, and in severe cases tooth loss), but can also extend to broader effects on their personal relationships, emotional status and Quality of Life (QoL).

Despite the improvements in survival rates, people with Down Syndrome still face many and different quality of life issues including cultural, environmental, and economic challenges, and studies have shown that individuals with disabilities -

including those with intellectual disabilities and Down Syndrome- are at higher risk of experiencing poorer health and increased age-adjusted mortality compared to general population; some of these health conditions are to some extent preventable and unjust (Emerson et al., 2012; Emerson & Hatton, 2014; Heslop et al., 2014) therefore this represents an example of health inequity (Emerson, 2015). Studies on OHRQoL on the general population have revealed that oral health influences psychological wellbeing and life satisfaction (Christensen et al., 2011; Locker et al., 2000; Persson et al., 2009), and there is no reason to suggest that there is any difference for people with intellectual disabilities. However, this cannot be confirmed due to the scarcity of studies aimed at assessing OHRQoL among people with intellectual disabilities. In addition, there is no instrument developed specifically to measure the impact of oral health status on different aspects of QoL of people with intellectual challenges and /or those with Down Syndrome.

This research therefore aimed to develop and test a new measure to assess the OHRQoL of children/adolescents with Down Syndrome as a first step to understand how their oral health may affect their QoL and that of their family.

This thesis is structured as follows:

Chapter Two is a narrative review of the literature on definitions on disability. Concepts of Health-Related Quality of Life (HRQoL) and OHRQoL are also presented in this chapter. The chapter further details the results of a review of the literature on HRQoL and OHRQoL among people with intellectual disabilities including those with Down Syndrome. A conceptual model of QoL of individuals with disabilities is presented in this chapter as well; along with a theoretical model that guided this research. Finally, the aim and objectives of the research are presented.

Chapter Three describes the methods and materials used within the research; this includes a detailed description of the methods used in both phases of the study. Statistical methods and data analysis strategy for the study are also presented in this chapter.

Chapter Four presents the research results of Phase One (qualitative), as well as Phase Two (quantitative) elements of the study.

Chapter Five discusses the research findings and highlights the strengths and limitations of the study. It then presents the key conclusions and summarises recommendations for policy and research.

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Chapter 2 LITERATURE REVIEW

2.1 Introduction

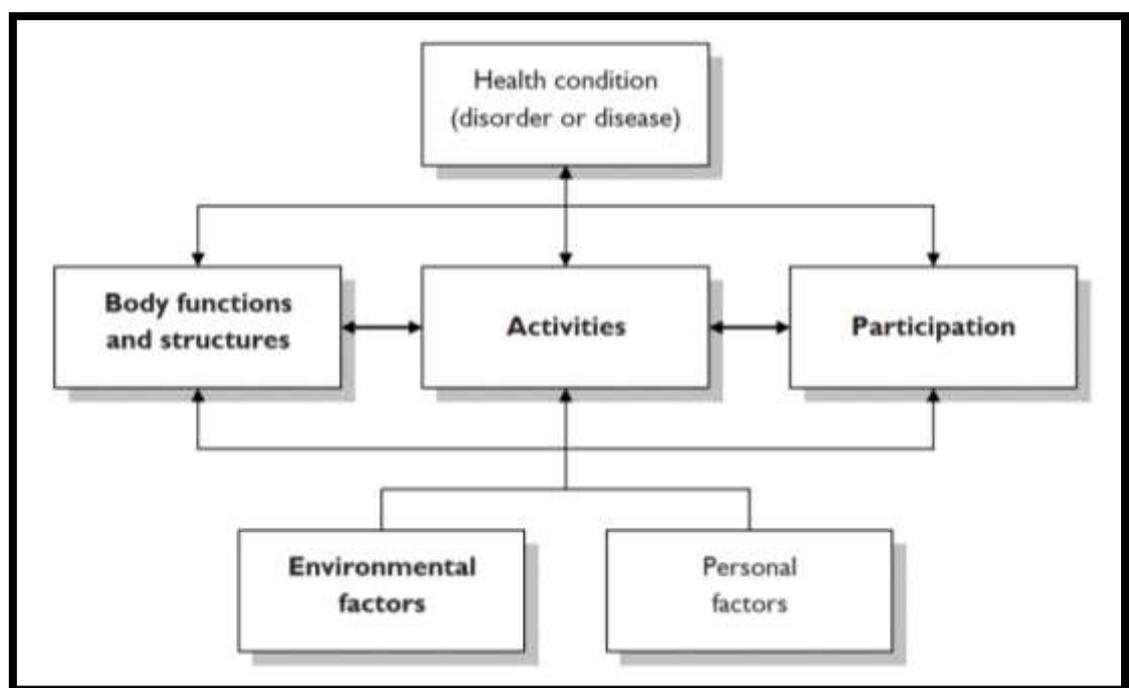
People with disabilities may suffer more from the consequences of oral diseases than those without such disabilities or impairments. Not only can this cause physical problems, but it can potentially have a wider reaching impact as poor oral health can have a negative effect on self-esteem, quality of life and general health (Benyamini et al., 2004; Sheiham, 2005). This literature review starts with a brief overview of the definitions, diversity, and statistics on disability. Then it focuses more on the definition of intellectual disability, its types and causes, and explores the differences between terminologies such as intellectual disabilities, learning disabilities, and learning difficulties. A review of the existing literature on the general health, as well as oral health of individuals with intellectual disabilities is also presented, with some examples of these conditions among people with Down Syndrome since this study focused on people with this disability. The review also presents the research in the field of QoL and individuals with intellectual disabilities and this is followed by studies on OHRQoL among people with intellectual disabilities including those with Down Syndrome.

2.2 Overview of Concepts and Definitions of Disability

2.2.1 What is disability?

Disability is an umbrella term for impairment of body function or structure, activity limitations or restrictions in social participation. It is a very complex, multidimensional and contested term, reflecting effects on body function and structure, the activities people engage in, and their participation in all areas of life. In general, it refers to an individual's capacity to function within a given social and environmental context (Freedman et al., 2004). This definition views disability from individual/medical perspectives, as well as structural/social perspectives, and therefore disability should be viewed neither as purely medical nor as purely social in nature. The International Classification of Functioning, Disability and Health (ICF) (WHO, 2001), considers functioning and disability as a dynamic interaction

between health conditions (diseases, disorders, or injuries) and contextual factors. The ICF identifies the three levels of human functioning; functioning at the level of body or body part (body function), the whole person (activities), and the whole person in social context (participation). In ICF, disability therefore involves dysfunction at one or more of these levels: impairments, activity limitations, and participation restrictions, (Figure 2.1) (WHO, 2001).



**Figure 2-1 International Classification of Functioning, Disability and Health, ICF
(WHO, 2001)**

The contextual factors consist of internal personal factors and external environmental factors. Personal factors include gender, age, coping style, social background, education, past and current experience, profession, overall behaviour pattern, character and other factors that influence how the individual experiences disability. A person's external environment has a large impact on the experience and extent of disability; it can either disable people with health problems (i.e. inaccessible buildings for wheelchair users), or negatively affect their participation in social,

economic, political, and cultural life. It may also affect the health of individuals directly by increasing their risk for some health problems, and hence for disability (i.e. water sanitation, air pollution). In addition, environmental factors include a wider set of issues such as an effective enforcement of laws and regulations, and better information on environments and their accessibility. They also include cultural differences that have a role in viewing disability. For example, negative attitudes, ignorance and prejudice can produce barriers in all domains.

2.2.2 Models of disability

A number of “disability models” have been suggested. The two distinct and most commonly used models are:

1. Medical/ Deficit or individual model and
2. Social/ structural model.

The two models differ significantly in the way they view and define “disability”. In the medical model, disability is viewed as a problem of the person (Kiesler, 1999), directly caused by disease, trauma, or other health conditions, which consequently require sustained medical care provided in the form of individual treatment by professionals. The medical model is the most well known in contrast to the recently developed social model (Smart & Smart, 2006). In the medical model, management of the disability is aimed at a "cure" or the individual's adjustment and behavioural change that would lead to an effective cure. In this model, medical care is viewed as the main issue, and at the political level, the principal response is that of modifying or reforming healthcare policy.

In contrast to the medical model, the social model of disability sees the issue of "disability" as a socially created problem and a matter of the full integration of individuals into society. In this model, disability is not a characteristic of an individual, but rather a complex pool of conditions, many of which are created by the surrounding physical, social and political environment. Hence, the management of the problem requires social action and is the collective responsibility of society at large to make the environmental modifications necessary for the full participation of

people with disabilities in all aspects of social life (Llewellyn & Hogan, 2000). The issue is both cultural and ideological, requiring individual, community, and large-scale social change. From this perspective, equal access for someone with an impairment or disability is a human rights issue of major concern (Marks, 1997). It should be noted that the social model does not deny the existence of impairments that may affect the daily lives of people with disabilities, but it shifts the emphasis towards the barriers that affect participation (Disability Rights Commission, 2002), Figure 2.2.

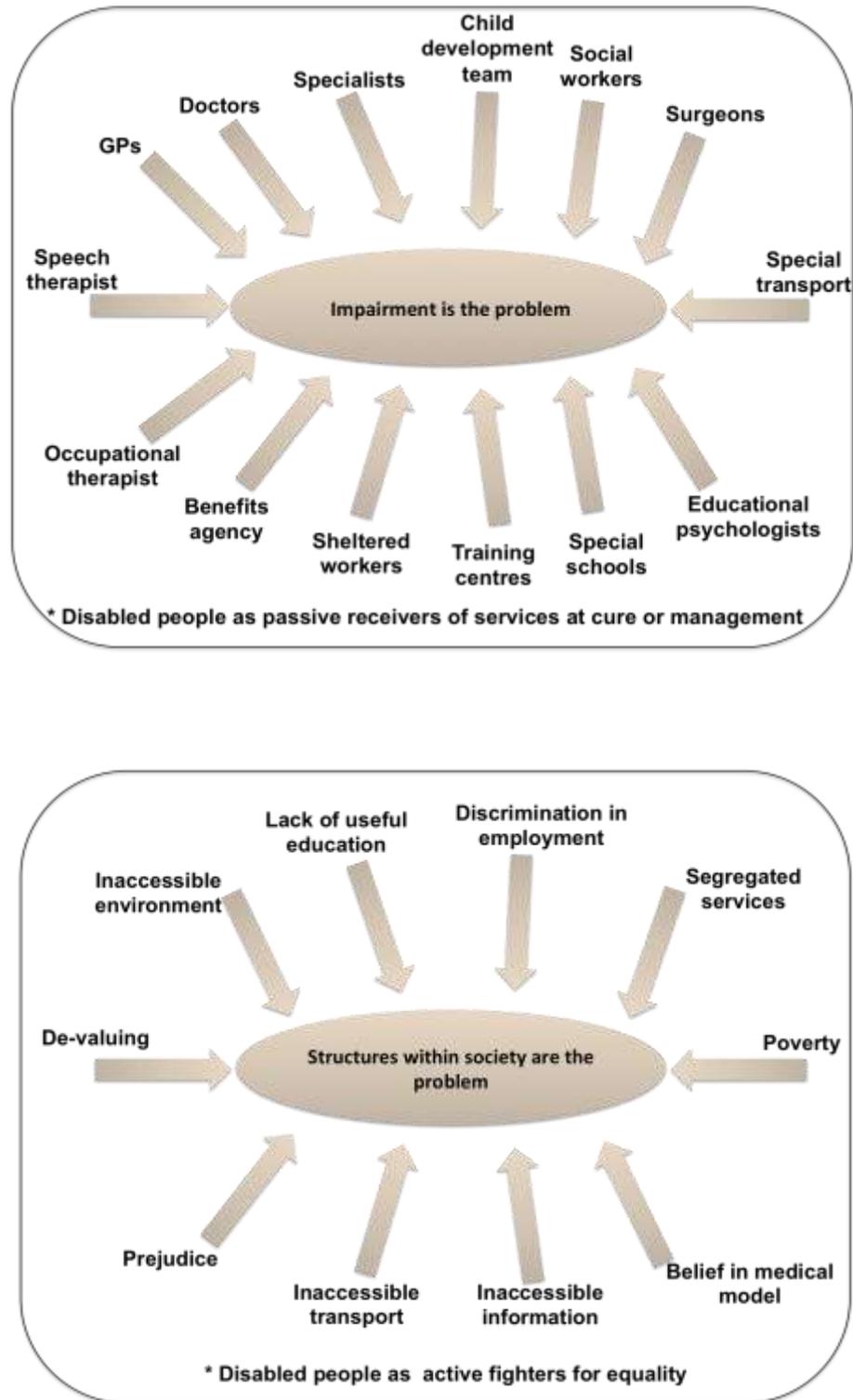


Figure 2-2 Medical and Social Models of Disability

(Adapted from <http://attitudes2disability.wordpress.com>)

2.2.3 Epidemiology of disability

Due to the complexity of defining disability and inconsistency in methods used in data collection, it has been difficult to have a definitive estimate of its prevalence. Using multiple surveys from more than 100 countries, the available data indicates that there are around 785 (15.6% according to the World Health Survey) to 975 (19.4% according to the Global Burden of Disease) million persons 15 years and older living with a disability; based on 2010 population estimates (6.9 billion with 1.86 billion under 15 years) (WHO, 2011). Of these, the World Health Survey estimates that 110 million people (2.2%) have very significant difficulties in functioning (WHO, 2011), while the World Health Organization Global Burden of Disease study estimates that 190 million (3.8%) have “severe disability” – the equivalent of disability inferred for conditions such as quadriplegia, severe depression, or blindness. When children are included, over a billion people (or about 15% of the world’s population) were estimated to be living with disability (WHO, 2011), (Appendix 1).

2.3 Overview of Intellectual Disability

2.3.1 Definition

There are a number of ways of defining and classifying intellectual disability and they are all open to different interpretations. The terms learning disability, intellectual disability, and mental retardation have been defined and understood differently in different countries and even within various regions of the same country. Different countries use different terminologies to describe the same group of people. The term ‘learning disability’ itself can be confusing and it has had many different labels over time, and continues to be referred to by different terms such as; ‘mental retardation’, ‘special needs’, ‘mental handicap’ and ‘intellectual impairment or disability’ (Kelly, 2002). The United Kingdom is the only country that uses the term ‘learning disability’ whereas an increasing number of international organizations and countries use the term ‘intellectual disability’ instead (Emerson & Heslop, 2010). This term has recently replaced the term ‘mental retardation’ as the term to be used by academics and clinicians in the US and a number of other

countries such as Canada and Australia (Reid, 1997; AAIDD, 2010). The current accepted international terms include ‘intellectual disability’ or ‘developmental disability’. Other terms such as ‘mental handicap’, ‘learning difficulty’ and ‘mental retardation’ have been used in the past and are still acceptable in some places but may cause offence in others (BILD, 2011).

The term ‘intellectual disability’ should be considered interchangeable with the UK term ‘learning disability’ which has been defined by the World Health Organization as “a developmental disability that first appears in children under the age of 18, with an intellectual functioning level (as measured by standard tests for intelligent quotient, IQ) that is well below average and significant limitations in daily living skills (adaptive functioning)” (WHO, 1996). This definition encompasses people with a broad range of disabilities, which can include for example, Down Syndrome, Attention Deficit/Hyperactivity Disorder, Cerebral Palsy or Autistic Spectrum Disorder.

2.3.2 Severity of intellectual disabilities

Severity of intellectual disabilities differs according to the level of IQ of the individual;

1. Mild

Approximate IQ ranges from 50 to 69 (in adults, mental age from 9 to under 12 years). Many adults will be able to work and maintain good social relationships, and contribute to society; however, this results in some learning difficulties.

2. Moderate

Approximate IQ ranges from 35 to 49 (in adults, mental age from 6 to under 9 years). Likely to result in marked developmental delays in childhood, but most can learn to develop some degree of independence in self-care and acquire adequate communication and academic skills. Adults will need varying degrees of support to live and work in the community

3. Severe

Approximate IQ ranges from 20 to 34 (in adults, mental age from 3 to under 6 years), and it is likely to result in continuous need for on-going support throughout life.

4. Profound

In individuals with an IQ that is under 20 (in adults, mental age below 3 years). This results in severe limitation in self-care, communication and mobility (WHO, 1996).

There is also a group of people described as having ‘profound and multiple intellectual disabilities’ (PMID) and are characterized by a complex range of severe physical and intellectual disabilities with IQ below 25 and a lack of functional skills (Kelly, 2002). Acknowledging the differences between terms, the following section will focus on some health issues experienced by people with intellectual disabilities (internationally) or learning disabilities according to the UK way of defining this disability.

2.3.3 Causes of intellectual disabilities

Intellectual disability is caused by problems during brain development before, during or after birth.

Before birth, damage to central nervous system e.g.: accident or illness of mother while pregnant (malnutrition, drugs, alcohol, diseases) or genetic syndromes (Down Syndrome; Fragile X Syndrome)

During birth, for example in cases of premature birth, not enough oxygen during birth/hypoxia, birth difficulties or infections in the womb

After birth, that occurs in cases of illness or accident during early childhood (head injury, epilepsy, meningitis), or because of the effect of environmental factors (lead/mercury poisoning, malnutrition, social deprivation) (Hodapp & Dykens, 2003; Harris, 2006).

The most common causes of intellectual disability are associated with inherited conditions such as chromosomal abnormalities. Down Syndrome and Fragile X Syndrome, epilepsy, cerebral palsy are not intellectual disabilities in themselves, but people with these conditions are likely to have an accompanying intellectual disabilities (Bray, 2003; Krahn et al., 2006).

Aetiology of intellectual disabilities can be outlined in a more complex and integrated way as outlined in table 2.1. The American Association of Intellectual and Developmental disabilities (AAIDD, 2010) described how many factors (Biomedical, social, behavioural, and educational) could interact across time resulting in an intellectual disability.

Table 2-1 The risk factors of intellectual disabilities

Timing	Biomedical	Social	Behavioural	Educational
Prenatal	<ol style="list-style-type: none"> 1. Chromosomal disorders 2. Single-gene disorders 3. Syndromes 4. Metabolic disorders 5. Cerebral dysgenesis 6. Maternal illness 7. Parental age 	<ol style="list-style-type: none"> 1. Poverty 2. Maternal malnutrition 3. Domestic violence 4. Lack of access to prenatal care 	<ol style="list-style-type: none"> 1. Parental drug use 2. Parental alcohol use 3. Parental smoking 4. Parental immaturity 	<ol style="list-style-type: none"> 1. Parental cognitive disability without supports 2. Lack of preparation for parenthood
Perinatal	<ol style="list-style-type: none"> 1. Prematurity 2. Birth injury 3. Neonatal disorders 	<ol style="list-style-type: none"> 1. Lack of access to prenatal care 	<ol style="list-style-type: none"> 1. Parental rejection of caretaking 2. Parental abandonment of child 	<ol style="list-style-type: none"> 1. Lack of medical referral for intervention services at discharge
Postnatal	<ol style="list-style-type: none"> 1. Traumatic brain injury 2. Malnutrition 3. Meningoencephalitis 4. Seizure disorders 5. Degenerative disorders 	<ol style="list-style-type: none"> 1. Impaired child-caregiver interaction 2. Lack of adequate stimulation 3. Family poverty 4. Chronic illness in the family 5. Institutionalisation 	<ol style="list-style-type: none"> 1. Child abuse and neglect 2. Domestic violence 3. Inadequate safety measures 4. Social deprivation 5. Difficult child behaviours 	<ol style="list-style-type: none"> 1. Impaired parenting 2. Delayed diagnosis 3. Inadequate early intervention services 4. Inadequate special education services 5. Inadequate family support

Table from the AAIDD manual 'Intellectual Disability: Definition, Classification, and Systems of Supports (11th ed)', 2010 pg 60.
(The AAIDD Ad Hoc Committee on Terminology and Classification, 2010)

2.3.4 Prevalence of intellectual disabilities

Due to differences in definitions and methods of data collection used in studies aimed at measuring the prevalence of individuals with intellectual disabilities, differences in estimates across studies also exist. In the few studies conducted so far, rates of about 2% have often been found (Zigler & Hodapp, 1986). Other studies, especially those employing registries or hospital records, have more often reported rates from below 1% to 1.5% (Larson et al., 2001) of the total general population. One relatively recent meta-analysis of all literature on the estimate of intellectual disabilities at a population level and published between 1980 and 2009 showed that the prevalence of intellectual disability across studies included in the meta-analysis was 10.37/1000 population (Maulik et al., 2011). The estimates varied according to income group of the country of origin, the age group of the study population, and the study design. The highest rates were seen in low- and middle-income countries. The estimate varied also according to the methods used for identification of cases. The finding revealed that the prevalence is higher among studies based on children/adolescents, compared to those on adults (Maulik et al., 2011) Table 2.2.

Table 2-2 The prevalence of intellectual disabilities (Maulik et al., 2011)

Proportion of studies and pooled prevalence estimates per 1000 population by subgroups (N = 52).				
	N	% ^a	Prevalence/1000 population ^b	95% CI of prevalence rate
<i>Income group of country</i>				
Low-income	6	11.5	16.41	11.14–21.68
Middle-income	17	32.7	15.94	13.56–18.32
High-income	29	55.8	9.21	8.46–9.96
<i>Type of population targeted</i>				
Rural	8	15.4	19.88	13.60–26.17
Urban	1	1.9	7.0	6.12–7.87
Urban slum/mixed rural-urban	17	32.7	21.23	16.34–26.11
Regional/provincial	23	44.2	7.85	6.98–8.71
National	3	5.8	6.23	5.48–6.98
<i>Age-group of study population</i>				
Adult	5	9.6	4.94	3.66–6.22
Child/adolescent	35	67.3	18.30	15.17–21.43
Both adult and child/adolescent	12	23.1	5.04	4.07–6.01
<i>Type of study</i>				
Cross-sectional	41	78.9	9.69	8.76–10.63
Cohort	11	21.1	13.21	10.70–15.72
<i>Sampling strategy used to gather data</i>				
Key informant report	1	1.9	2.61	–1.00–6.23
School based study	2	3.9	7.04	6.35–7.73
Hospital data or administrative registry	30	57.7	9.35	8.60–10.10
Random household survey	19	36.5	15.78	13.73–17.86
<i>Measure used for diagnosis</i>				
Psychological assessment	30	57.7	14.30	12.70–15.91
DSM/ICD	12	23.1	8.68	7.89–9.48
AAIDD/ICF/some disability criteria	10	19.2	6.41	4.89–7.93

^a Values have been rounded so may not add up to 100%.

^b Estimates based on meta-analysis using random effects model.

2.3.5 Health differences between the general population and people with intellectual disabilities

2.3.5.1 General health status

Several studies conducted have revealed that people with intellectual disabilities have poorer health than their non-disabled peers, and in addition to inequalities in health status, they also experience inequality in medical care utilization compared to the general population (Elliott et al. 2003; Graham 2005; Ouellette-Kuntz 2005; Krahn et al. 2006; Nocon 2006; Emerson & Baines 2010; O’Hara et al. 2010). A recent review by McCarthy & O’Hara (2011) revealed that people with intellectual disabilities die 15 years younger than people without intellectual disabilities and they also have poorer health outcomes for a number of conditions, including respiratory diseases, epilepsy, and oral health.

The differences in health are mainly because of: increased mortality, increased morbidity and greater exposure to health damaging determinants, such as poverty. The differences in health care use that occurs amongst people with disabilities are because of unequal access to services (Kerr, 2004). Table 2.3 highlights key inequalities issues supported by some examples of differences in health and health care between people with intellectual disabilities and the general population (Kerr, 2004).

Table 2-3 Inequalities in health among people with intellectual disabilities (Kerr, 2004)

Area of inequality	Example in people with intellectual disabilities
Increased mortality	Lower life expectancy
Increased morbidity	High levels of epilepsy, sensory impairment and behavioural disorders
Increased in negative determinants of health	High levels of obesity and underweight; low employment; fewer social connections
Access to services	Low rates of uptake of health promotion
Quality of services	High prescription rates of antipsychotic medication with no evidence of psychosis; high rates of unrecognised disease identified on health screening

2.3.5.2 Health damaging behaviours

People with intellectual disabilities are more prone to undertaking certain health damaging behaviours than the general population. A study by Emerson (2010) collected self-reported data from people with mild intellectual disabilities indicated that individuals who did not access intellectual disability services are more likely to smoke and less likely to access some health services and promotion activities than those who do use these services. Maag et al. (1994) also found that adolescents with intellectual disabilities were significantly more likely to use tobacco and marijuana but found no differences in alcohol use between the study groups. This is comparable to the findings of Blum et al., (2001) who analysed a nationally representative sample of young people in the US and found for most of the health behaviours studied (smoking, marijuana use, suicide attempt), adolescents with disabilities were higher than their peers in reporting poor health behaviours. Another study by Robertson et al., (2000) among people with intellectual disabilities in residential settings reported the prevalence of risk factors and variables that predict the presence of these risk factors and stated that compared with the general population, the study sample were underweight, had poorer dietary habits and performed less physical activity.

In summary the literature on general health of individuals with intellectual disabilities concluded that this segment of population has significantly poorer health and they are more exposed to health-risk factors than the general population.

2.3.5.3 Oral health

Although there has been a marked overall improvement in the overall oral health status among children over the past few decades (Petersen et al., 2005), this might not be the case for children with disabilities. Poor oral health conditions and unmet oral health needs are often considered as a probable source of health inequalities in persons with neuro-motor and intellectual deficiencies compared to the general populations (Hennequin et al., 2008). Studies among people with intellectual disabilities indicated that they have problems such as poor oral and denture hygiene, more untreated dental caries and higher prevalence of gingivitis and other

periodontal diseases (Andres & Davis, 2010). They also have different treatment patterns with fewer filled teeth and instead more extracted teeth than the general population (Andres & Davis, 2010) (Appendix 2).

Some oral health issues among individuals with intellectual disabilities

a) Oral health conditions

In a systematic analysis of the overall global burden of oral conditions in 1990-2010, oral conditions remained highly prevalent affecting 3.9 billion people (Marcenes et al., 2013). In this study results showed that untreated caries in permanent teeth was the most prevalent condition evaluated for the entire Global Burden of Disease 2010 Study, whereas severe periodontitis and untreated caries in deciduous teeth were the 6th and 10th most prevalent conditions (Marcenes et al., 2013). In addition to the high cost of treating dental diseases (Petersen et al., 2005), oral diseases have considerable negative impacts on individuals' lives. They can affect the daily activities and result in short or long-term health loss. Studies conducted among children and aimed at assessing the impacts of oral diseases and conditions on their QoL revealed an array of undesirable impacts that ranges from experience of dental pain to loss of schoolwork or social isolation. This section briefly reviews studies on aspects of oral health and oral health-related issues among individuals with intellectual disabilities.

Dental Caries. Many studies have been conducted to evaluate the oral health of people with intellectual disabilities. However, each study has its own methodology and definition of the study sample that make it difficult to conduct a meta-analysis. A relatively recent systematic review of oral health among people with intellectual disabilities highlighted that most of studies that examined caries rates concluded that the rates in people with disabilities were either the same as the general population or even lower (Andres & Davis, 2010). Whilst levels of dental caries were not found to be higher, people with intellectual disabilities experienced poorer oral health outcomes; they had higher levels of untreated dental caries, higher numbers of missing teeth, but fewer filled teeth (Andres & Davis, 2010).

Periodontal Disease. The Andres & Davis (2010) review also considered studies aimed at assessing periodontal/ gingival health of people with intellectual disabilities. The methodology of assessing periodontal health differed across studies (i.e. bleeding on probing, pocket depth, calculus, etc.) but they all found strong evidence to indicate that people with intellectual disabilities had higher prevalence and greater severity of periodontal disease than the general population (Andres & Davis, 2010).

b) Oral hygiene status

Andres and Davis (2010) in their review showed that the in majority of studies reviewed, people with intellectual disabilities had poorer oral hygiene than the general population, and no studies demonstrated or suggested better oral hygiene among them. The review also outlined that the oral health status and dental service use of adults with intellectual disabilities varied according to the place of residence; they found that people living in community settings having poorer oral hygiene and higher levels of untreated dental caries compared to their counterparts in residential care.

c) Utilization of dental services

Havercamp et al., (2004) compared data on health status, health-risk behaviours, chronic health conditions and utilisation of care across non-disabled, disabled and developmentally disabled (including those with intellectual disabilities) using data from a national survey. They found that there were significant inequalities in oral health care; individuals with developmental disabilities were more likely than non-disabled group not to have had their teeth cleaned in the past five years or never to have had their teeth cleaned. Studies also showed that the utilization of dental care services differs according to the type of housing. A study showed that a change from institutional living to community-based housing for a group of adults with intellectual disabilities was associated with changes in dental attendance and treatment patterns (Stanfield et al., 2003), with those living in the community-based settings were less likely to receive regular dental examination and operative dental treatment than they previously received when in long-term hospital care. Tiller et al.,

(2001) also found that subjects living in community settings were significantly less likely to have a dentist and to use community dental services than their residential counterparts who were more likely to attend only when having trouble.

2.3.5.4 Possible explanations of frequent oral health problems among people with intellectual disabilities

As mentioned earlier, people with intellectual disabilities have in general poorer oral health and much higher levels of untreated oral disease and poorer oral hygiene than those without disabilities. Therefore, they are at additional risk, and hence they require greater attention in maintaining good oral hygiene (Lange et al., 2000; Department of Health, 2001) for several reasons. Firstly, many people with intellectual disabilities have conditions that have inherent risks to oral health. For example, people with Down Syndrome are more likely to breathe through their mouths, which can compromise oral hygiene, and people with cerebral palsy are subject to dental abrasion from gastro-esophageal reflux. Secondly, difficulties caregivers face in meeting the nutritional needs of people with multiple disabilities, which may include the necessity for high-energy sugary food supplements, and laxatives that increases the risk of dental caries (BSDOH, 2001). In addition, oral hygiene practices tend not to be given a very high priority in services for profoundly disabled individuals due to other pressing demands (Griffiths and Boyle, 2005). Carers may not have the appropriate skills to maintain oral hygiene levels of those they are caring for (BSDOH, 2001; Tiller et al, 2001), which can be compounded by the behavioral and communication difficulties of service users. Another very important factor that might result in delay of seeking dental treatment is the inability of many people with intellectual disabilities to complain of dental or gingival pain, this if combined with low levels of parental/carers awareness might lead to poor oral health.

Poor access to oral care plays an important role, resulting either from factors that inhibit access to generic health care (for example fear, ignorance, lack of appropriate health promotion) or from a misinterpretation of normalization, which can result in

rejection of the medical model of care by health care professionals and subsequent neglect of physical needs (BSDOH, 2001). The lack of specialist training for dentists to meet the needs of this group (BSDOH, 2001) might also affect their oral health status.

Finally, fragmentation of dental services (Rawlinson, 2001) is a major factor that might negatively impacts on individuals with disabilities' oral health. In general, health care services for people with chronic, long-term conditions are always fragmented when it should be coordinated around their needs. The fragmented care provision might also lead to conflicting health messages that in turn affect individuals' cooperation. This highlights the need for providing an integrated care in which general and oral health services work alongside with other services provided to those individuals.

In summary, people with intellectual disabilities typically have poorer oral health compared to the general population. This is due to many factors some of them related to the existing disability, while other factors are due to the type and quality of services provided, and/or level of care and awareness provided by parents, carers or care services.

2.4 Overview of Down Syndrome

As this study focuses on children/adolescent with Down Syndrome, the next section covers some important general and oral health issues affecting this group. Down Syndrome is a common genetic disorder that ranges in severity and is usually associated with varying degrees of intellectual disabilities, and some medical and physical conditions. Research has shown that Down Syndrome is the most common genetic cause of intellectual disabilities (Van-Trotsenburg et al., 2006) in which most people with this disorder have mild or moderate intellectual disability while small percentage could be severely affected. In addition, it is the most common chromosomal anomaly among live-born infants with an incidence of 1:600 to 1:900 (Yang et al., 2002; Canfield et al., 2006). Down Syndrome occurs as a result of an aberration on the 21st chromosome that occurs prior to fertilization or during

gestation (Crocker, 2006; Nehring & Betz, 2010). There are three general chromosomal profiles of Down Syndrome that are all based on abnormality on the 21st chromosome; these include 1) nondisjunction, 2) translocation, and 3) mosaicism.

Down Syndrome occurs in different races, nationalities, and across diverse socioeconomic groups without discrimination (Nehring & Betz, 2010). Individuals with Down Syndrome are characterised by special physical features such as small head, ears, and mouth; a low nasal bridge; upward slanting eyes, and epicanthal folds; they are also characterized by a small stature (Jorde, 2010; Zigman, 2013). The birth prevalence of people with Down Syndrome has remained relatively stable during the last decade (Collins et al., 2008) but survival rates have improved (Weijerman et al., 2008). In developed countries, life expectancy has increased to nearly 60 years of age during the past two generations (Janicki et al., 1999; Merrick, 2000). Despite the improvements in the survival rates, people with Down Syndrome still face different quality of life issues; including cultural, environmental, and economic challenges and they are still at higher risk of health inequalities compared to mainstream population.

2.4.1 Health needs of individuals with Down Syndrome

2.4.1.1 General health status among people with Down Syndrome

a) Cardiac conditions. Congenital heart defects are common in individuals with Down Syndrome in which they have an increased risk (40-50%) of structural defects of valvular disorders (Bosch, 2003; Nehring & Betz, 2010). This is important to dentistry because historically in order to prevent systemic bacterial endocarditis, individuals with valvular diseases were strongly recommended to prophylactically take antibiotics prior to dental procedures (Bosch, 2003; Cohen, 1999).

b) Compromised endocrine system. It is well known that individuals with Down Syndrome have an increased prevalence of autoimmune disorders affecting both endocrine and non-endocrine organs (Karlsson et al., 1998). The most common endocrine problem among children with Down Syndrome is hypothyroidism. Hypothyroidism can occur at any time from infancy through to adulthood. It is estimated that approximately 10-20% of children with Down Syndrome have

congenital or acquired thyroid disease (Pueschel & Pezzullo, 1985), while studies among adults vary widely, but the incidence of thyroid disease among them is believed to be between 13% and 50% (Botero et al., 2006). Children with Down Syndrome are also at an increased risk of diabetes since studies have shown that they are more prone to develop Type 1 diabetes mellitus (Anwar et al., 1998; Van Goor & Massa, 1997) compared to other children, however, they do not appear to be at an increased risk of Type 2 diabetes (Esbensen, 2010).

c) Respiratory disease. Individuals with Down Syndrome have more frequent respiratory infections, and more reported obstructive airway disease mainly because of the narrowed airways, and impaired immune system (Bosch, 2003; Cohen, 1999; McCarron et al., 2005)

d) Sleep disorders. The most common sleep disorder occurs among individuals with Down Syndrome is the obstructive sleep apnea; other sleep disorders such as light sleeping and frequent waking are also common (Bosch 2003; Cohen, 1999)

e) Hearing problems. Studies showed that around 40-80% of children with Down Syndrome have middle ear involvement, often resulting in a hearing deficit (Bosch 2003; Cohen, 1999; Schwartz & Schwartz, 1978), and this could happen as results of conductive hearing loss or sensori-neural hearing loss. Hearing problems may further complicate poor communication skills.

f) Visual impairments. Studies have shown that children with Down Syndrome frequently have eye problems including strabismus, glaucoma, cataracts, keratoconus, and nystagmus (Bosch 2003; Cohen, 1999).

2.4.1.2 Oral health status among people with Down Syndrome

In general, individuals with Down Syndrome do not have any unique oral health problems although some of the problems they experience tend to be severe and frequent in nature (Hennequin et al., 1999). The most prevalent oral health problem among people with Down Syndrome is periodontal disease. It is usually rapid and

destructive resulting in the loss of teeth during their early adulthood. Increased risk of tooth loss does not occur as a result of periodontal diseases alone; many factors such as poor oral hygiene, malocclusion, bruxism or conical-shaped teeth roots might contribute to premature tooth loss (Pilcher, 1998).

In contrast, people with Down Syndrome tend to have lower levels of dental caries compared to the general population (Hennequin et al., 1999). This could be due to many factors such as delayed eruption of teeth, wider spaces between teeth, high incidence of congenitally missing teeth (hypodontia) and smaller sized teeth.

Malocclusion is very common amongst this group because of delayed eruption of teeth and the underdevelopment of the maxilla, in addition to other factors related to the tonicity of the facial musculature. Individuals with Down Syndrome have specific oro-facial features and tooth anomalies that increase their risk of developing oral health problems (Fischer-Brandies, 1988; Roizen, 2007). Figure 2.3 outlines the interaction between oro-facial characteristics of people with Down Syndrome and their systemic manifestation that might leads to the development of dental diseases (Hennequin et al., 1999).

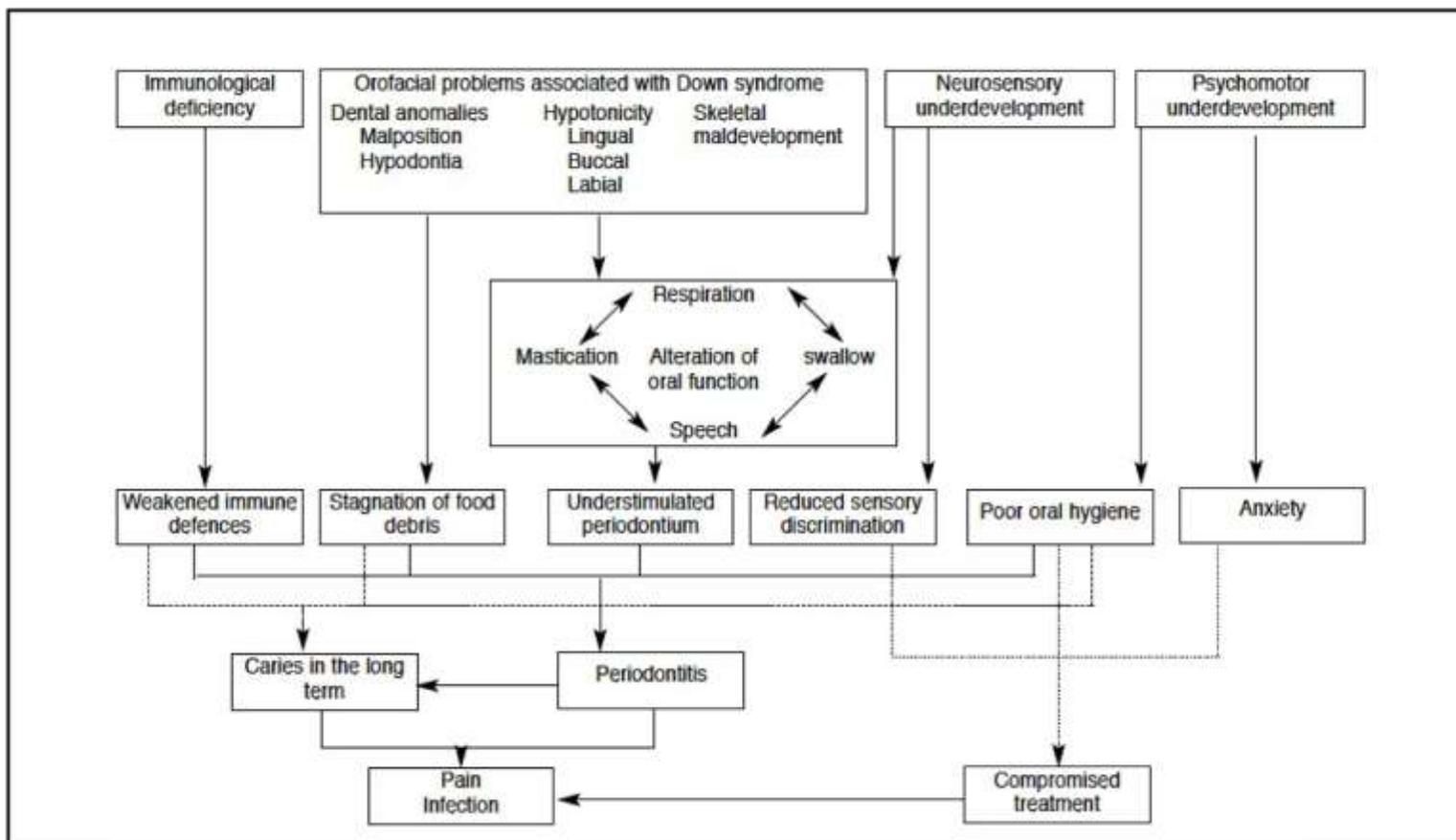


Figure 2-3 Interaction between orofacial and systemic manifestations of Down Syndrome in the development of dental disease and compromised oral function (Hennequin, 1999)

In summary, people with Down Syndrome vary in severity of accompanied intellectual disabilities and health conditions. However, they share characteristic phenotype, which increases their susceptibility to develop health problems compared to people without the syndrome. They do not have unique oral problems, although they have special oro-facial characteristics that increase their risk of developing oral diseases at an early age, or at a more severe level. Their increased risk of developing oral conditions might have an impact on their lives but this issue needs more research to establish if oral diseases do have a significant impact or not.

2.4.1.3 Why is it important to assess the impact of oral health of individuals with Down Syndrome on their quality of life?

The impact of oro-facial conditions on individuals' daily lives may be related closely to their oral health (such as pain, discomfort, and in severe cases tooth loss), but can also extend to broader effects on personal relationships, emotional status and quality of life. Studies on OHRQoL in the general population indicate that oral health influences psychological wellbeing and satisfaction in the general population (Locker et al., 2000; Persson et al., 2009; Christensen et al., 2011). There is no reason to suggest that there is any difference for people with intellectual disabilities/Down Syndrome. The following part of the review covers definitions and basic issues in QoL research, and highlights considerations in assessing QoL among adults and children with intellectual disabilities. The review will then outline studies conducted to assess HRQoL. The review will then cover the key concepts of OHRQoL and will summarize the studies conducted to assess the impacts of oral health of individuals with intellectual disabilities on different aspects of their lives.

2.5 Overview of QoL/HRQoL

After the World Health Organization (WHO) defined health as “a state of complete physical, mental and social well-being and not merely the absence of disease” (WHO, 1948), there was increased interest in QoL assessment. Since that time efforts were made to develop reliable and valid measures. However, the progress in this direction was slow because of the difficulties in obtaining this kind of data and making the definition operational. QoL is a broad concept that covers a variety of aspects of individual’s life.

2.5.1 Definition of QoL and HRQoL

QoL is a broad concept that encompasses both medical and non-medical aspects, including physical functioning (ability to perform daily activities), psychological functioning (emotional and mental wellbeing), social functioning (relationship with others or participation in social activities), and perception of health status, pain and overall satisfaction with life (Gill & Feinstein, 1994; McKenna & Whalley, 1998; Sanders et al., 1998; Orley et al., 1998). A group of researchers of WHO defined HRQoL as “An individual’s perception of their position in life in the context of the culture and value systems in which they live and in relation to their personal goals, expectations, standards and concerns” (WHOQOL Group, 1995). Although the concept of QoL is in no way fixed or stable, there is some agreement around the key dimensions of QoL (Lefort & Fraser, 2002) that are summarised in Table 2.4.

Table 2-4 Quality of Life Dimensions and Indicators

Core QoL dimensions (Keith, 2001)	QoL indicators (Schalock, 1996)
Emotional wellbeing Interpersonal relations Material relationships Personal development Physical development Self-determination Social inclusion Rights	Happiness, safety, spirituality Family, friendship, intimacy Ownership, employment, shelter, finance Competence, personal perception, skills Education, health Choices, autonomy, control Acceptance, roles, community activity Voting, personal privacy, ownership

Thus, QoL encompasses overall general wellbeing that comprises objective descriptors and subjective evaluations of physical, material, social, and emotional wellbeing together with the extent of personal development and purposeful activity, all weighted by a personal set of values (Felce & Perry, 1995).

HRQoL is one aspect of individual's overall QoL, and refers to physical, psychological and social domains of health that are influenced by individual's experiences, beliefs, expectations and perceptions (Patrick et al., 1973; Brook et al., 1979; Brook et al., 1983). HRQoL measures can be used to provide data on objective assessments of health status, and/or subjective perceptions of health (Levine & Croog, 1984; Bergner, 1989; Patrick & Erickson, 1993). The ultimate aim of HRQoL assessment is to measure the impacts on or changes in physical, functional, psychological, and social health after experiencing any disease or health condition.

Researchers interested in measuring HRQoL face various challenges linked to its theory and method, and a number of definitions exist. Patrick & Erickson (1987) defined HRQoL as "the level of well-being and satisfaction associated with events or conditions in a person's life as influenced by disease, accidents or treatments". This considers not only the functional ability but also degree of satisfaction derived from the performance of social roles and activities.

HRQoL questionnaires can be used to measure cross-sectional differences in HRQoL between patients at a point in time (discriminative instruments) or longitudinally to measure changes in HRQoL within patients during a period of time (evaluative instruments). Two basic approaches to HRQoL measurement are available: generic instruments that provide a summary of HRQoL; and disease-specific instruments that focus on problems associated with specific condition, disease, patient groups or areas of function. In addition, there are many modes of administering HRQoL measures, which include; interview, telephone, self-reported or surrogate responders (proxy), each method of administration has its weaknesses and strengths (Guyatt et al., 1993).

2.5.2 Are individuals with intellectual disabilities capable of providing valid QoL/HRQoL reports?

Though outcome indicators in people with intellectual disabilities have been described to include: age at death, hospital admission and readmission rates, up-take of services, additional handicaps and consumer satisfaction (Jenkins, 1990), there is an important outcome measure, *Quality of Life*, that has been used as a challenging concept in the field of intellectual disabilities research. Moreover, various measures have been developed previously to evaluate the QoL of people with intellectual disabilities (Ager, 1990; Cummins, 1993; Schalock & Keith, 1993). Some of the QoL measures for individuals with intellectual disabilities were developed as proxy measure while few were developed as a self-report. In the intellectual disability literature, the vast majority of respondent-based health assessments use proxies in preference of self-report (Boulton et al., 2006; Chien et al., 2006; Cui et al., 2008; Lau et al., 2006; Sabaz et al., 2001), and that is mainly because of the challenges presented in terms of limited communication skills and difficulties with comprehension (Lloyd et al., 2006). Therefore, proxy measures from either parents or caregivers are usually used instead, although there are some issues concerning the accuracy of proxy assessments. The idea is that a third-party respondent who lives with the individual with intellectual disability can provide a response and give a reliable perception on their behalf. Evidence on the efficacy of the proxy measures is mixed, and the accuracy of proxy's reporting varies according to the proxy's relationship to the individual with intellectual disability (Perkins, 2007; Schwartz & Rabinovitz, 2003) and type of domain being measured (Andresen et al., 2001; Arlt et al., 2008). Studies from the QoL literature suggest that the use of proxy reports among people with intellectual disability should be limited to objective aspects of life, however they can be considered inappropriate when analyzing subjective aspects of life of individuals with intellectual disability (Ramcharan & Grant, 2001; Schalock et al., 2002; Stancliffe, 1999).

Response bias has also been well documented in self-report measures among people with intellectual disabilities (Sigelman et al., 1980; Verri et al., 1999). Adolescents and adults with intellectual disability are susceptible to presentation bias and have a tendency to choose the most positive response alternative (Verri et al., 1999; Schalock et al. 2002). This is known as "Disability paradox" (Albrecht and

Devlieger, 1999), where QoL is routinely rated higher by people with disabilities even though life conditions would be considered undesirable by most objective criteria and social conventions. This is potential bias in the already complicated construct of QoL, and emphasizes the need to carefully study the impact of health and health-related conditions on the QoL of individuals with disabilities in order to improve our understanding of their needs and therefore, enhance services provision.

2.5.3 Assessing QoL/HRQoL among people with intellectual disabilities

QoL measurement among people with intellectual disabilities presents a unique challenge since obtaining details directly from subjects is difficult and sometimes inaccurate, the use of proxy indicators (such as caregivers or parents) does not always give the real picture, in addition, relying only on objective or hard data provides an incomplete picture.

Within the last decades, studies have shown how people with certain degrees of intellectual disabilities have increasingly come to be viewed as being a reliable source of information on issues affecting their lives, experiences, feelings and views (Stalker, 1998). Therefore, whenever possible individuals themselves should be asked for their opinion on their life, although, methodological considerations should be taken into accounts for self-report in people with limited intellectual abilities. For example, the QoL instrument should be chosen according to the underlying conceptual hypotheses, simplicity, clarity, use of pictures, length, and should be kept as basic as possible. In addition, it is extremely important to assess the level of intellectual disabilities before starting the QoL questionnaires (formal competency evaluation) to ensure that the individual is able to answer the questionnaire by him/herself otherwise proxy measures become the only option available (White-Koning et al., 2005). In principle, self-report QoL information can be collected directly from individuals with intellectual disability considering the characteristics of the population and using an appropriate measure; however, the use of proxy measure is more practical and realistic option.

2.5.4 QoL/HRQoL studies conducted among people with intellectual disabilities

2.5.4.1 Assessment of QoL/HRQoL among adults with intellectual disabilities

In the field of intellectual disability research, some aspects of QoL have been extensively researched (Brown & Brown, 2005). Cummins (1997) conducted a review of all QoL measures developed for use among individuals with intellectual disabilities, and in that review, he included all measures that can at least partially be answered by people who have an intellectual disability, and excluded other measures that are solely reliant on proxy responses. This indicates that actively including individuals with intellectual disability in the research process has been a priority for some time.

Cummins's (1997) search revealed 13 self-rated QoL measures designed and validated for use among this specific group, however, the measures were not equally useful in assessing individuals' QoL, and they were presented in a rank-order of utility. For example some of the measures were developed to assess the effect of normalization/residential on people with intellectual disability QoL, while others were related to services, and some aimed at assessing only objective aspects of QoL. Others aimed at measuring purely the subjective aspects of QoL, while a few aimed at measuring both subjective & objective aspects of individuals with intellectual disabilities QoL. The review concluded that the most widely used measures of QoL among people with intellectual disabilities were: the Quality of Life Questionnaire (QOL-Q; Schalock & Keith, 1993), and the Comprehensive Quality of Life Scale Intellectual Disability (ComQoL-ID; Cummins et al., 1997).

In this review, Cummins (1997) suggested a list of criteria essential in the construction of an adequate instrument to be used among people with intellectual disability;

- 1) subjective and objective aspects of QoL should be measured;
- 2) each subjective and objective dimension should be measured by a number of life domains which in aggregate should present the total QoL construct;
- 3) measures of domain satisfaction should be weighted by the importance of each domain to the individual;

- 4) the instrument should have adequate reliability, validity and sensitivity;
- 5) the scale should be equally applicable to non-disabled people thus ensuring normative comparison of the life quality;
- 6) the response mode and choice of answers should reflect psychometric theory and strike a balance between reliability and sensitivity;
- 7) the instrument should be brief, simple to administer, and easy to score and finally,
- 8) a pre-test should be used to establish that respondents could comprehend the questions (Cummins, 1997).

There are also several studies aimed at assessing level of agreements between self-report of individuals with intellectual disabilities and their proxies. For example, Stancliffe (1999) in a cross sectional study aimed at assessing the agreements between self-reports of 63 adults with intellectual disability and caregiver proxy responses using QOL-Q, and found substantial positive correlations and no significant difference between self and caregiver reports. However, Stancliffe (1999) noted that proxy measures are not substitute for consumer self-reports and the two data sources should not be treated interchangeably; he also noted that the differences between these two types of reports are not signs of unreliability but rather an indication of different perceptions by the two groups. Another study aimed at assessing the agreement between self and proxy (parents and staff) measures of a more subjective aspect of QoL, *life satisfaction*, using the Life Satisfaction Scale (LSS) showed that the life satisfaction reports of patients with intellectual disabilities and caregivers were positively correlated. However, caregiver ratings were significantly higher than subject ratings and no such divergence were found between subject's and parent's reports, so it was suggested that parent's should be selected in preference to caregivers in assessing life satisfaction (Schwartz & Rabinovitz, 2003).

QoL in intellectual disability research was also used as an outcome measure to evaluate changes on QoL after relocation and/or community integration. A study by Bhaumik et al. (2011) aimed to assess QoL of 51 adults with intellectual disabilities and complex health problems following a move from long-stay hospital to community settings using the Questionnaire on Quality of Life (QQoL). Results found that QoL improved between baseline and 6 months follow-up but leveled off at 1-year follow-up. These findings were similar to a previous longitudinal study by

Dagnan et al. (1998) where the QoL scale was also used as a measure to assess the QoL of 29 older people with intellectual disabilities who left hospital to live in ordinary houses; results showed a continued improvement in scores in the first 41 months of living in the community with an eventual levelling or reduction in scores later on. Cooper and Picton (2000) also reported the long-term effects of relocation on a sample of 45 people with an intellectual disability who moved from an institution to community and/or other institutions and followed them up for 3 years. They used the QoL-Q to study the long-term effects of relocation and found that relocation was associated with improved QoL outcome.

All previously summarised studies were conducted to assess the general concept of QoL. Little research has directly examined the concept of HRQoL among individuals with intellectual disabilities. However, literature suggests a high risk of experiencing negative impact of health status on various aspects of life in individuals with intellectual disabilities, such as emotional distress (Svetaz et al., 2000), and social isolation (Jackson et al., 1987). A study conducted to assess the HRQoL among 68 college undergraduates (age range 18-29) with self-reported learning disabilities using the self-reported SF-36 which is a 36-item, generic and most widely used measure of HRQoL (Davis et al., 2009). Although no details were given on the validation of this measure (SF-36) to use among the study sample, results indicated that individuals having a learning disability experienced significantly poorer emotional wellbeing compared to those who did not report having such a disability. In 2010, Power, Green, and the World Health Organization Quality of Life Disability group (WHOQOL-Dis) group conducted research that aimed at adapting the generic version of the WHOQOL measure for use with adults with physical or intellectual disabilities and then test its use in a series of cross-cultural field trials, using similar procedures used to develop the generic World Health Organization Quality of Life-100 (WHOQOL-100) & World Health Organization Quality of Life-Brief (WHOQOL-BREF) (Power et al., 2010). The adaptation study consisted of the development of a supplementary module that resulted in 12-items that can be added to the existing World Health Organization Quality of Life (WHOQOL) instruments. A study to assess the agreement level between self and proxy reports of HRQoL of people with intellectual disabilities conducted among 614 adults as well as two different samples of proxies including both professional carers and relatives using

WHOQOL-Dis, the study concluded that there is a good agreement between the person-proxy quality of life assessment (Schmidt et al., 2010).

2.5.4.2 Assessment of QoL/HRQoL among children with intellectual disabilities

As mentioned earlier, individuals with intellectual disabilities should be included in the process of evaluation of different aspects of their lives, even children, when possible (Stalker, 1998). Many studies found that relatives and clinicians have the tendency to underestimate a child's QoL compared to how the child rates it; also there seems to be a lower level of agreement between proxies and children when dealing with the more subjective dimensions of QoL such as social or psychological domains (Britto et al., 2004; Eiser & Morse, 2001; Ennett et al., 1991; Levi & Drotar, 1999; Parsons et al., 1999; Varni et al., 1995).

QoL in children with intellectual disabilities has not been examined extensively. Aspects such as pain (van Dongen et al., 2002) or emotional and behavioural problems (Dekker et al., 2002) have been assessed in a few studies using proxy reports. However self-reported fears (Ramirez & Kratochwill, 1997) and anxiety (Sarfare & Aman, 1996) were assessed and compared in children with and without intellectual problems, but in both studies a formal procedure to evaluate their cognitive ability was set up before the assessment itself. In the literature, there are a number of QoL/HRQoL measures that have been accepted to use among children with an intellectual disabilities. Several measures were developed to assess HRQoL among children with epilepsy. A review of subjective measures aimed at assessing QoL of children and adolescents with epilepsy revealed five condition-specific measures (Cowan & Baker, 2004);

- Quality of Life in Epilepsy Inventory for Adolescents (QOLIE-AD-48) (Cramer et al., 1999),
- Quality of Life for Children with Epilepsy (QOLCE) (Sabaz et al., 2000),
- Health-Related Quality of Life in Children with Epilepsy measure (Ronen et al., 2003),
- Impact of Childhood Neurological Disability scale (ICND) (Camfield et al., 2003),
- Epilepsy and Learning Disabilities Quality of Life scale (ELDQOL) (Baker & Jacoby, 1997).

Buck et al. (2007) conducted a re-evaluation of the ELDQOL scale and concluded that it was a reliable and valid instrument for assessing QoL in children and young adults with epilepsy and learning disabilities. This measure was aimed at informal/formal carers of children with both severe epilepsy and learning disabilities.

Since the research of this thesis focuses on children/adolescents with Down Syndrome, this part of the review of the literature will specifically focus on HRQoL of individuals with Down Syndrome and their families.

Bertoli et al., (2011) conducted a study of HRQoL of 518 individuals of all ages with Down Syndrome in Rome, Italy. Although the study does not specify that the HRQoL was measured, some aspects generally consistent with the variables defining HRQoL were assessed, such as daily activities, and social integrations. The research identified the association of health and function with an individual's QoL. A questionnaire was developed which could be answered by either the participant with Down Syndrome or by a proxy-respondent if necessary. The HRQoL of older participants with Down Syndrome was "very poor" because of health problems, limited social relationships, restricted educational and employment opportunities, and lack of independence. However, the study investigators did not provide evidence supporting the reliability and validity of questions used.

In the Netherlands, the HRQoL of children with Down Syndrome was assessed in two separate studies using the validated Netherlands Organisation for Applied Scientific Research Academic Medical Centre (TNO-AZL) Children's Quality of Life Parent Form questionnaire (TACQOL). This instrument was created for the parental measurement of pain; symptoms of disease or disability; autonomy; and functioning of motor ability, cognition, social aspects, positive emotional aspects, and negative emotional aspects in children (Vogels et al., 1998). The first study investigated behavior and HRQoL of eight-year-old children with Down Syndrome (van Gameren-Oosterom et al., 2011). Results showed that the study sample had more emotional and behavioral problems and less favorable HRQoL compared to children from the general population. The children with Down Syndrome had lower HRQoL in domains of cognition, social function, independence, and gross motor

skills than the comparison mainstream children. However, there was no significant difference in physical complaints between the groups of children (van Gameren-Oosterom et al., 2011). A second study focused on the HRQOL of children with Down Syndrome who had recurrent respiratory infections, and findings revealed that they had lower HRQOL in aspects of social functioning, independence, motor skills, and physical well-being subscales compared to the control group, which was comprised of children with Down Syndrome without recurrent respiratory infections (Verstegen et al., 2013).

A recent Chinese cross sectional study was conducted to validate and assess HRQoL among 109 children and adults with Down Syndrome (Mok et al., 2014). Health Utilities Index Mark 2 (HUI2) and Health Utilities Index Mark 3 (HUI3) were used as measures of HRQoL. These instruments are non-disease specific indices applicable to individuals over the age of 5, and have been shown to be responsive to various chronic diseases such as rheumatoid arthritis (Drake et al., 1996), Type 2 diabetes (Maddigan et al., 2004), and stroke (Grootendorst et al., 2000), in the general population. HUI2 and HUI3 are two independent yet complementary systems measuring HRQoL. Researchers concluded that these measures were valid for use among Chinese individuals with Down Syndrome, and they found that individuals with Down Syndrome had a lower HRQoL as compared to the general population (Mok et al., 2014). Results also showed that a significant graded relationship existed showing that when the number of health problems increased, the HRQoL decreased. (Mok et al., 2014).

Raising a child with Down Syndrome can also have a substantial impact on family's QoL. Concerning HRQoL in parents of children with Down Syndrome, studies have shown that parents tend to experience lower HRQoL regarding vitality, leisure (social functioning, daily activities, and recreation), and mental- or psychological health (Buzatto & Beresin, 2008; Hatzmann et al., 2008; Hedov et al., 2000; Murphy et al., 2000; Oliveira & Limongi, 2011). Predictors of family QoL have also been studied and factors that were found to be associated with lower HRQoL show that these factors concern mainly a child's functioning and psychosocial variables. A lower household income, higher levels of functional impairment and behavioral problems of the child, less social support, maladaptive coping style of parents, less

participation in health promoting activities, and poorer professional support were all related to lower HRQoL outcomes (Bourke-Taylor et al., 2012; Browne & Bramston, 1998; Davis & Gavidia-Payne, 2009; Khanna et al., 2011; Lin et al., 2009). A recent study aimed to explore which socio-demographics, child functioning and psychosocial variables were related to HRQoL domains in parents of children with Down Syndrome concluded that psychosocial variables mainly social support and time pressures rather than socio-demographics or child functioning showed the most consistent and powerful relations to the HRQoL of parents of children with Down Syndrome (Marchal et al., 2013).

In summary, interest in general QoL has a long history among individuals with intellectual disabilities, especially adults; this interest resulted in the development of several measures of QoL. The HRQoL literature in this field is less developed, but studies conducted among adults and children with different intellectual disabilities showed that their QoL is affected by their health conditions. Using generic HRQoL measures, which were developed with mainstream children, studies showed that children with Down Syndrome had worse HRQoL compared to the general population. Studies also showed that the health of children with Down Syndrome has undesirable effects on their families' QoL. These broad findings highlight the need for good quality studies to go more in-depth in the assessment of the HRQoL of individuals with intellectual disabilities/Down Syndrome in order to understand its nature, contributors, and therefore intervene accordingly.

2.6 Overview of OHRQoL

This part of the review briefly covers definitions and basic concepts in OHRQoL and summarizes some of the studies conducted to assess OHRQoL among people with intellectual disabilities.

2.6.1 Definition of OHRQoL

For many years, clinical examinations were considered to be the main criterion in investigating the oral health status ignoring the importance of subjective aspects of oral health assessments. In the late 1970s and early 1980s, researchers reported the

need for a more comprehensive measure that captures the social and psychological impact of oral conditions (Cohen & Jago, 1976; Sheiham & Croog, 1981; Locker, 1988; Reisine, 1981, 1988a, 1988b). Over the last decades, assessing the OHRQoL has been widely advocated as an adjunct to clinical examinations documenting the full impact of oral disorders (Slade, 1998). Researchers have focused on the effects of poor oral health, not only on general health, but also on people's day-to-day functioning, wellbeing, and ability to carry out activities of daily living. Several studies acknowledged that poor oral health is an important cause of negative impacts on daily performance and QoL (Adulyanon & Sheiham, 1997; Barbosa et al., 2009; Foster-Page et al., 2005; Goursand et al., 2008; Jokovic et al., 2002; Locker, 2007; Locker & Allen, 2002; McGrath & Bedi, 2003).

There are many variations in the approach to defining OHRQoL. It was initially defined as "the impact of oral conditions on daily functioning" (Slade, 1998). A few years later, in a paper evaluating OHRQoL outcomes in elderly people, Locker and colleagues redefined OHRQoL as "the symptoms and functional and psychosocial impacts that emanate from oral diseases and disorders" (Locker et al., 2002). In this definition, the authors did not give importance to other predictors of OHRQoL, such as contextual factors. Other researchers defined OHRQoL as "the absence of negative impacts of oral conditions on social life and a positive sense of dentofacial self-confidence" (Inglehart & Bagramian, 2002). This definition embraced the central dimensions of OHRQoL proposed by Gift and Atchison in 1995. These suggest that OHRQoL be defined as a person's assessment of how functional factors, psychological factors, social factors, and experience of pain/discomfort affect his or her wellbeing, Figure 2.4.

Locker and colleagues (2005) re-defined OHRQoL emphasizing the role interaction between and among oral health conditions, social and contextual factors (Locker et al., 2005), and the rest of the body (Atchison et al., 2006). Thus, OHRQoL is a multidimensional construct that includes a subjective evaluation of the individual's oral health, functional and psychosocial wellbeing. Therefore OHRQoL is defined as "The impact of oral diseases and disorders on aspects of everyday life that a patient or person values, that are of sufficient magnitude, in terms of frequency, severity or

duration to affect their experience and perception of their life overall” (Locker and Allen, 2007).

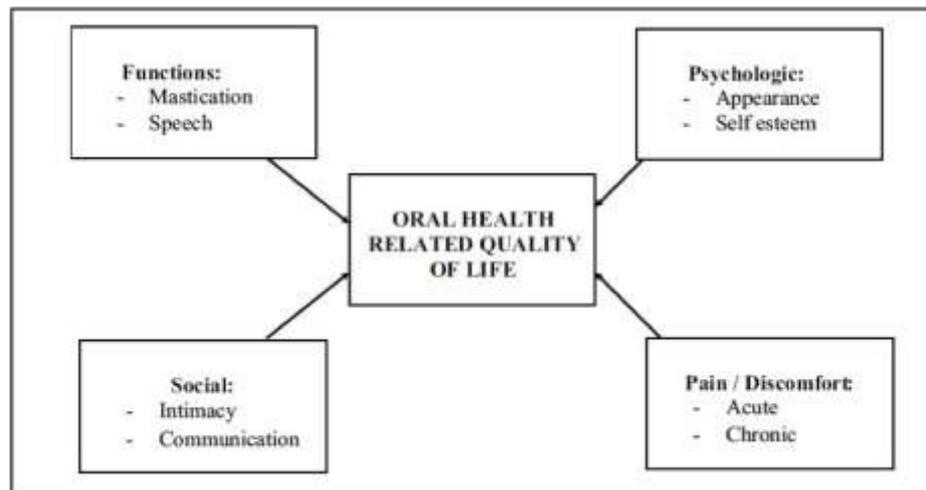


Figure 2-4 Factors associated with OHRQoL (Inglehart & Bagramian, 2002)

The majority of OHRQoL indices are most commonly developed for use among adults and older people (Adulyanon & Sheiham, 1997; Locker & Allen, 2002; Atchison & Dolan, 1990; Cornell et al., 1997; Leao & Sheiham, 1996; McGrath & Bedi, 2003; Sheiham et al., 2001; Slade, 1998; Strauss & Hunt, 1993). More recently researchers have developed instruments specifically designed to assess OHRQoL in children (Jokovic et al., 2002; Locker et al., 2002; Jokovic et al., 2003; Jokovic et al., 2004; Gherunpong et al., 2004; Filstrup et al., 2003; Broder et al., 2007; Pahel et al., 2007; Huntington et al., 2011; Tsakos et al., 2012). These measures were designed primarily to assess the undesirable impacts of oral conditions on individuals by defining how oral status affects daily activities such as speaking, eating, smiling, learning, emotional, and social wellbeing. Therefore, OHRQoL measures allow for the shift from traditional biomedical methods of assessing oral health to broader physical and psychosocial criteria that are closer to the actual health perceptions and needs in the population (Sischo & Border, 2011).

2.6.2 Children's OHRQoL

For over a decade, interest in children's OHRQoL resulted in the development of several measures (Table 2.5). Measures differ in dimensions, age of targeted children, and method of reporting OHRQoL (either by children themselves, or by proxy), but they share the concept of measuring how oral health affects aspects of daily living of children, and a couple of measures also include some questions on the potential impact of child's oral health on his/her family life. A recent review of research on OHRQoL in children showed that the most frequently used measure is the Child Perceptions Questionnaire (CPQ) (Gilchrist et al., 2014).

Table 2-5 Oral Health-Related Quality of Life measures for children

Measure	Author/Year	Aim	Dimensions
CPQ11-14 * Child Perception Questionnaire	Jokovic et al., 2002	The impact of oral and oro-facial conditions	Oral symptoms Functional limitations Emotional well-being Social well-being
FIS * Family Impact Scale	Locker et al., 2002	The family impact of oral and oro-facial disorders	Parental/ family activities Parental emotions Family conflict
P-CPQ * Parental-Caregivers Perceptions Questionnaire	Jokovic et al., 2003	Parental/care-givers perception of the oral health-related quality of life for children	Oral symptoms Functional limitations Emotional well-being Social well-being
CPQ8-10 * Child Perception Questionnaire	Jokovic et al., 2004	The impact of oral and oro-facial condition	Oral symptoms Functional limitations Emotional well-being Social well-being
MOHRQOL Michigan Oral Health-Related Quality of Life Scale	Filstrup et al., 2003	The effects of early childhood caries on children's oral health-related quality of life	Functional aspects Pain/discomfort, Psychological aspects Social aspects
Child-OIDP Child Oral Impact on Daily Performance	Gherunpong et al., 2004	The serious oral impact on children's ability to perform daily activities	Eating Speaking Cleaning mouth Sleeping Emotion Smiling Study Social contact
ECOHIS Early Childhood Oral Health Impact Scale	Pahel et al., 2007	The impact of oral health problems and related treatment experiences on the quality of life of preschool age children (3 to 5 years old) and their	Child symptoms Child function Child psychological Child self-image/ social interaction Parent distress

		families.	Family function
COHIP Child Oral Health Impact Profile	Broder et al., 2007	Oral health related quality of life in children with a broad age range (8–15 years) that include positive as well as negative aspects: parallel forms exist for the child and caregiver	Oral health Functional well-being Social-emotional well- being School environment Self-image
POQL Pediatric Oral Health-Related Quality of Life	Huntington et al., 2011	A brief measure of oral health-related quality of life (OHQL) in children with a particular focus on input from parents and children from low- income or minority populations	Social Role functioning Physical Emotional
SOHO-5 Scale of Oral Health Outcomes	Tsakos et al., 2012	Self-reported oral health related quality of life measure for 5-year-old children	Eating Drinking Speaking Playing Smiling (because teeth hurt) Smiling (because of the way teeth look) Sleeping

* Canadian researchers have developed the Child Oral Health Quality of Life (COHQoL) questionnaires, which include the Parental- Caregiver Perceptions Questionnaires (P-CPQ) and the Family Impact Scale (FIS) for children aged 6–14 years, and three Child Perceptions Questionnaires for children aged 6 to 7 (CPQ6–7), 8 to 10 (CPQ8–10), and 11 to 14 (CPQ11–14) years of age

All above summarized measures were developed and validated for use among mainstream children in order to assess the impacts of oral health on children's QoL. None of these measures aimed at assessing OHRQoL among children with disabilities. It should be noted that some of the presented measures (i.e. CPQ, Child-OIDP) have been extensively adapted to be used in different cultures/ settings.

2.6.3 Family impact of child's oral health

Sheiham and Croog (1981) raised the issue of the family impact of oral and oro-facial conditions in the early 1980s, when they described the psychosocial impact of dental diseases on individuals and societies, and highlighted that a broad series of family life areas might be affected by the presence of dental diseases among its members (Sheiham & Croog, 1981). For example, evidence indicates that early childhood caries results in lost workdays for caregivers who have to stay at home to take care of their child, or spend time and money in accessing dental care (Gift et al., 1992). Years later, Osman and Silverman (1996) recommended that outcomes of oral and oro-facial conditions should be addressed very closely in children from two broad perceptions. First, is the impact of oral health on the child's QoL, and second is the impact of child's condition on the family. There are a number of reasons for including the family impact on child's HRQoL measures. One of them is the central role played by the family in child health, and the likelihood that chronic illness in a child will impact on the family to some degree. In addition, the fact that health care interventions often address parental needs and concerns as well as the child's, and the fact that parental reports of a child's health may be influenced by the degree to which the parent is physically or emotionally affected by the child's condition (Rothman et al., 1991).

Furthermore, there is strong evidence in the literature that parents or caregivers of young children experience significant QoL issues because of their children's health problems and treatment experiences (Locker et al, 2002; Juniper et al., 1996). Therefore, family impact is essential in order to assess this caregiver-burden, and measurement of child's OHRQoL should be assessed from the perspective of both the child, as well as the family.

Researchers in the field of OHRQoL research have developed and evaluated a few measures to assess the family impact of child's oral and oro-facial conditions (Locker et al., 2002; Pahel et al., 2007). The first was Family Impact Scale (FIS), which is one component of the Child Oral Health Quality Of Life Instrument (COHQOL). This instrument was designed to assess the OHRQoL of children aged

6–14 years with oral and oro-facial conditions. The second relevant measure is the Early Childhood Oral Health Impact Scale (ECOHIS), which was developed to assess the impact of oral health problems and related treatment experiences on the QoL of preschool age children (3 to 5 years old) and their families (Pahel et al., 2007). The findings of these studies demonstrated the pervasive effects of child's oral health conditions on the functioning of the family as a whole. Appendix 3 presents items of family quality of life included in both instruments.

2.6.4 OHRQoL studies among people with intellectual disabilities

This section reviews the few studies aimed at assessing OHRQoL or aspects of it among adults and children with intellectual disabilities; studies reviewed include some among people with Down Syndrome, autism, and cerebral palsy.

2.6.4.1 Assessment of OHRQoL among adults with intellectual disabilities

Stanfield et al. (1999) studied the impact of oral health on the QoL of adults with intellectual disabilities using the Questionnaire of the Oral Health-related Quality of Life in the UK (OHQoL-UK) instrument. The questionnaire was administered to carers of people with intellectual disabilities and concluded that oral health significantly influenced the QoL of adults with intellectual disabilities. OHQoL-UK is a battery of 16 questions, which takes into account the impact of oral health on life quality. This questionnaire was originally developed based on a general UK population's perceptions of how oral health affects life quality; however, there was no mention whether the researchers tried to validate the instrument to assess its suitability for their study population.

A report aimed at investigating the oral health and dental service use of adults with intellectual disabilities in Sheffield, and to explore their experiences and perceptions of dental services by actively involving them in the research (Hall et al., 2011). The research consisted of two studies, one quantitative and one qualitative. The self-reported questions about the impact of the mouth on everyday life were included in the quantitative part, and were conducted among a sample of 628 adults with intellectual disabilities. The first question was concerned with the overall health of

the teeth, lips, jaws and mouth. Three more questions asked about the impact of oral health in terms of the frequency, in the last 12 months, of pain, discomfort when eating and being self-conscious (Hall et al., 2011). Results showed that approximately one third of the study sample reported that their oral health was fair, poor or very poor. Similar numbers reported the occasional or more frequent experience of toothache and discomfort when eating because of problems with their teeth, mouth or dentures in the last 12 months. With regards to self-consciousness, 13% of the individuals with intellectual disabilities reported being occasionally or more often self-conscious because of their oral health. In this survey, researchers relied on some common questions to assess the impact of oral health on individuals with intellectual disabilities QoL. No specific, validated measure of OHRQoL was used, and no criteria was mentioned about the selection of the above three specific questions of OHRQoL. Therefore, the results gave a very general conclusion of the oral health impact of individuals with intellectual disabilities on their everyday lives.

An Australian cross-sectional survey of carers of 18– 44 years old with physical and intellectual disabilities was conducted to assess the impact of oral health on the QoL of individuals with disabilities. Researchers used some questions from the 49-item Oral health Impact Profile (OHIP); they used four out of seven conceptual domains, and they explored one question for each domain of impact; psychological disability (trouble sleeping), physical pain (pain and discomfort), physical disability (problem eating), and social disability (being irritable). There was no mention in the methods of the validation used to assess the questions selected but the authors assumed that observable domains like function (problem eating) or social issues (irritability) were more likely to be valid, however this was not assessed for the specific population of people with intellectual disability. The study concluded that the prevalence of negative impact from a dental problem on individual items like diet, sleep, behavior, and pain and discomfort was low. However, more than one in 10 care recipients reported that they experienced one or more negative impacts during the last year (Pradhan, 2013).

2.6.4.2 Assessment of OHRQoL among children with intellectual disabilities

A Brazilian cross-sectional study aimed at determining the prevalence of periodontal disease among a group of children and adolescents with Down Syndrome, and the possible effect of this condition on their QoL by interviewing their mothers (Amaral-Loureiro et al., 2007). To measure the impact on QoL, researchers adapted the Oral Health Impact Profile (OHIP-14). However, this measure is validated to use as a self-report measure of OHRQoL among mainstream adults population but not as a proxy measure for children with degree of intellectual disabilities. The child was also present during the interview, and whenever possible was encouraged to answer with the mother. OHIP-14 was used as a basic reference, and modification to facilitate the conduct of the interview and to examine the understanding was made, although no details of modifications were mentioned, and no indication of a standard adaptation method was done. The study concluded that periodontal conditions had negative effects on the QoL of people with Down Syndrome, and these effects were increased by the increase in the disease severity (Amaral-Loureiro et al., 2007).

Another Brazilian exploratory study interviewed 19 mothers of children and adolescents with Down Syndrome investigated broadly the mothers' perceptions concerning the general and oral health of their children with Down Syndrome and their opinion on the impact of oral health on the life of their children. The interviews were conducted in an open-ended, in-depth manner to give flexibility and broadness to the study objectives and no standard, validated measure of children OHRQoL was used. Also no topic guide was used with broad themes to ensure full coverage and consistency across interviews. Although some mothers reported the issue of social acceptance, mainly relating to aesthetic concerns or halitosis, there were no clear findings on the possible impacts of child's oral health on their QoL from mothers' perceptions. According to the interviews, results also showed that overall health and oral health entailed specificities associated with the absence of illness, the performance of daily activities, and feelings of wellbeing (Oliveira et al., 2010a). The findings of limited impact of oral health on the child's QoL might have occurred because of the lack of depth and consistency across all interviews and lack of use of specific OHRQoL measures, therefore, these findings should be interpreted with a degree of caution.

A study conducted in Saudi Arabia aimed at assessing OHRQoL of group of children with autism and intellectual disability, and to compare this to non-disabled siblings of same age group (Pani et al., 2013). The study also aimed at assessing the impact of child's oral health on the family. P-CPQ and FIS were used to assess OHRQoL of 59 children and their families, which they were cross-matched for socioeconomic status and age of the child with families with non-disabled children. Linguistic validation and translation of the English version of the questionnaires was conducted, but not reported in detail. The questionnaires were completed by one of the parents in families with an autistic child; the parent completed two questionnaires, one for the autistic child and the other for the unaffected sibling. When the parental perception scores were compared between autistic children, their unaffected siblings, and children from families without an autistic child, it was found that the overall P-CPQ scores for autistic children were significantly higher than those for families with unaffected siblings, especially in the domains of functional limitation, emotional wellbeing and social wellbeing. Results on the FIS also showed a significant difference between families in which families with autistic children had significantly higher scores in the domains of parental emotion and family finances.

OHRQoL of a group of autistic children was also assessed and compared with the same age-group of non-autistic children using P-CPQ. The study was conducted among 135 Indian autistic children, in which they were matched with 135 non-autistic peers; and parents were asked to fill out the questionnaire (Yashoda & Puranik, 2014). Limited details were given on the methodology used and no explanation of the translation and adaptation of the P-CPQ were provided, although it was mentioned that the questionnaire was administered in a local language after ensuring linguistic validity by a back translation method. OHRQoL scores of autistic children were significantly higher indicating poorer OHRQoL compared to children without autism especially in the functional limitation domain (Yashoda & Puranik, 2014).

Another study of OHRQoL of children with cerebral palsy was conducted in Brazil (Abanto et al., 2014). Researchers collected data from 60 parents using the 47-item questionnaire that combines the validated Brazilian version of the P-CPQ, and the FIS components of the COHQOL instrument. They concluded that oral health

conditions, mainly dental caries experience and presence of bruxism, were strongly associated with a negative impact on OHRQoL of children with cerebral palsy, mainly on the emotional well-being domain, and on their parents but no details were provided about the impacts on different aspects of family's QoL (Abanto et al., 2014).

In summary, this review of the literature on the impact of oral health on different aspects of QoL among people with intellectual disabilities has highlighted a limited number of studies and many with some methodological limitations. In addition, there appears to be no measure of OHRQoL developed and validated for use among this target population. Results of these studies indicate that oral health does have an impact on the life of individuals with intellectual disabilities, but little effort was placed to investigate the impact of oral health of individuals with intellectual disabilities on their families or caregivers, and to extensively assess the determinants of OHRQoL among this group.

Down Syndrome and Oral Health-Related Quality of Life

When assessing OHRQoL among children/adolescents with Down Syndrome there are some factors that should be considered. First, their oral health status might impact on their QoL through: a) disease such as dental caries or periodontal diseases that also affect mainstream children, or b) oral conditions such as protruded tongue, dribbling of the saliva which are characteristics of children with Down Syndrome. Second, the disability by itself is a chronic condition, and its impact on individual's QoL might be stronger than the impact of oral health problems that are usually acute and/or last for a short period (e.g. toothache), so the presence of the disability and its associated characteristics could have a direct impact on OHRQoL. For example, some disabilities such as Down Syndrome carry with them special oro-facial characteristics that are usually untreatable and cannot be completely eliminated and/or treated such as protruded tongue, therefore, they are considered as lifelong or chronic conditions, so OHRQoL measures which are developed to be used among mainstream populations might not be suitable or sensitive enough to capture the

OHRQoL among individuals with disabilities. In addition, knowing that children go through stages of development, their perceptions change and their understanding and evaluation of health might also change; this aspect is not fully understood in mainstream adolescence age. The developmental changes, including psychological, among adolescents with Down Syndrome might differ from mainstream adolescents, and might have an impact on their evaluation or perception of OHRQoL. All these factors collectively highlight the complexity of assessing OHRQoL among individuals with Down Syndrome.

2.7 Gaps in the literature

Although the oral health of people with disabilities is often assumed to be poor, the quality of the evidence underpinning this assumption is contested and very limited (Hennequin et al., 2008). Despite numerous studies reporting poor oral health in people with disabilities, many of the studies have limitations that cast doubts on the validity of the findings. These include the absence of control groups, use of small sample sizes or lack of comprehensive evaluation of oral conditions. It should also be noted that studies aimed to assess oral health have used different methods or have aimed to measure the prevalence of a specific dental disease, such as dental caries or periodontal disease (Shaw et al., 1990; Rodriguez-Vazquez et al., 2002; Nunn et al., 1993), while others have focused on the existence of anatomical deficiencies (Oreland et al., 1987; Fischer-Brandies, 1988; Hobson et al., 2005), traumatic injuries (Shyama et al., 2001) or some aspects of functional incapacity (Frazier & Friedman, 1996; Spender et al., 1996; Dos-Santos & Nogueira, 2005; Hennequin et al., 2005), making it difficult to make a comprehensive comparison across these studies. This topic therefore needs more research that is sound methodologically to be able to generalize such findings among this specific segment of the population.

For decades, it has been advocated that comprehensive evaluation of oral health should incorporate both objective, as well as subjective indicators. Locker (1988) and McEntee (2006) have proposed that the concept of oral health is much more complex and embraces all aspects of health related to the mouth, jaw, teeth, throat and all related tissues. OHRQoL instruments aim to offer a global measure of the concept of health, although their use is not always feasible by the majority of people with intellectual disability (Slade & Spencer, 1994; Allison et al., 1999; Foster-Page et al., 2005). Thus, several proxy questionnaires have been developed for use among people with special health conditions (Allison & Hennequin, 2000; Baker & Jacoby, 1997; Bertoli et al., 2011; Dekker et al., 2002; van Dongen et al., 2002; van Gameren-Oosterom et al., 2011; Verstegen et al., 2013). However, they are very specific and cannot be used widely across all disability groups. It is also important to outline the conceptual, developmental, and methodological challenges of assessing OHRQoL among people with intellectual disabilities, which demand a careful assessment of the suitability of previously developed OHRQoL among mainstream

populations. The lack of appropriate OHRQoL measure to be used specifically among people with intellectual disabilities has consequently made it difficult to assess how oral health conditions might impact on their QoL and that of their families, and therefore, impossible to compare OHRQoL between groups of individuals with other types of disabilities. This study aimed at adding to the literature by developing and testing an OHRQoL measure to be used for children and adolescents with intellectual disabilities and mainly those with Down Syndrome.

2.8 Theoretical frameworks

Previously developed OHRQoL measures relied mainly on Locker's model of measuring the consequences of oral diseases (Locker, 1988), (Figure 2.5). This was based on the first WHO classification of consequences of diseases known as International Classification of Impairments, Disabilities, and Handicaps (ICIDH) (WHO, 1980), (Figure 2.6). Locker's model illustrates oral health in a unidirectional relationship between oral disease, disability and handicap modelled by pain and functional limitations. This model was very influential in the area of oral health research. However, it has several limitations that need to be taken into account. First, it does not take into account the dynamic progressive nature of oral health and disease as occurs with any other health status, and does not account for the fluctuation in health status that might occur along the course of experiencing oral conditions. Second, although the model recognises that impairment does not necessarily cause disability or handicap, it does not accommodate for individuals who can stop, minimise or even reverse the progress of disease through many individual's experiences such as coping and adaptation (Brondani & MacEntee, 2014), or adjusting to disease by changing their acceptance of their situation that usually occurs in some individuals after a period of time of experiencing the disability. In addition, Locker's model did not account for either the positive or negative impact of the environment, as indicated in the ICF approach (Figure 2.1), in the course of disease.

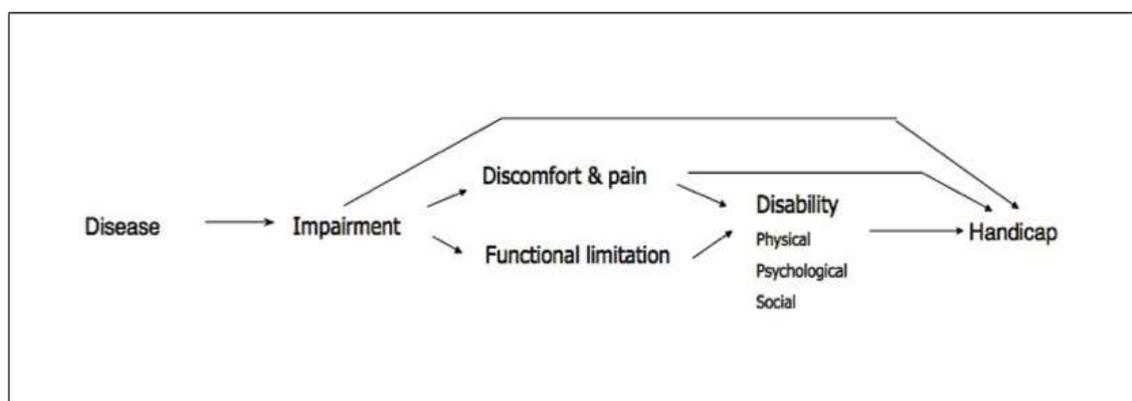


Figure 2-5 Conceptual model for measuring oral health (Locker, 1988)

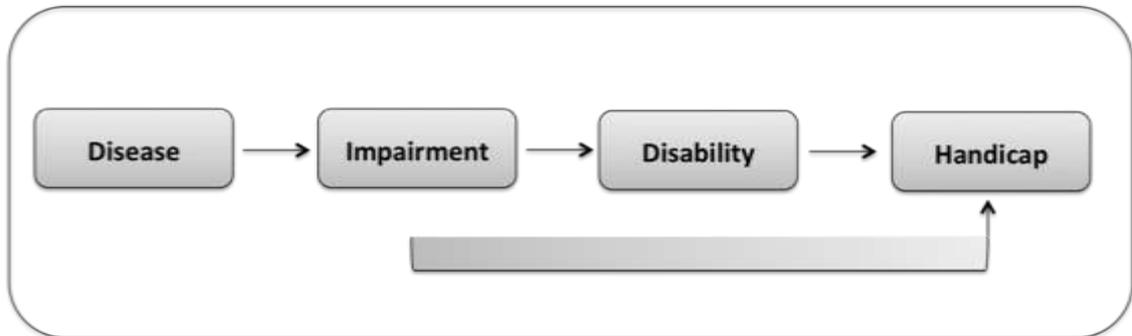


Figure 2-6 International classification of Impairments, Disabilities, and Handicaps (WHO, 1980)

Both models, ICIDH and Locker's, are presenting the consequences of disease at different levels; at the organ, individual, and social levels, but not the ultimate outcome that WHO definition of health encompasses, namely HRQoL. HRQoL is a broad concept that includes both clinical and social paradigms. This has been captured in the health outcomes model (Wilson and Cleary, 1995), this model linked clinical variables with HRQoL, and developed a causal model with clear distinctions between common approaches used to assess HRQoL, (Figure 2.7).

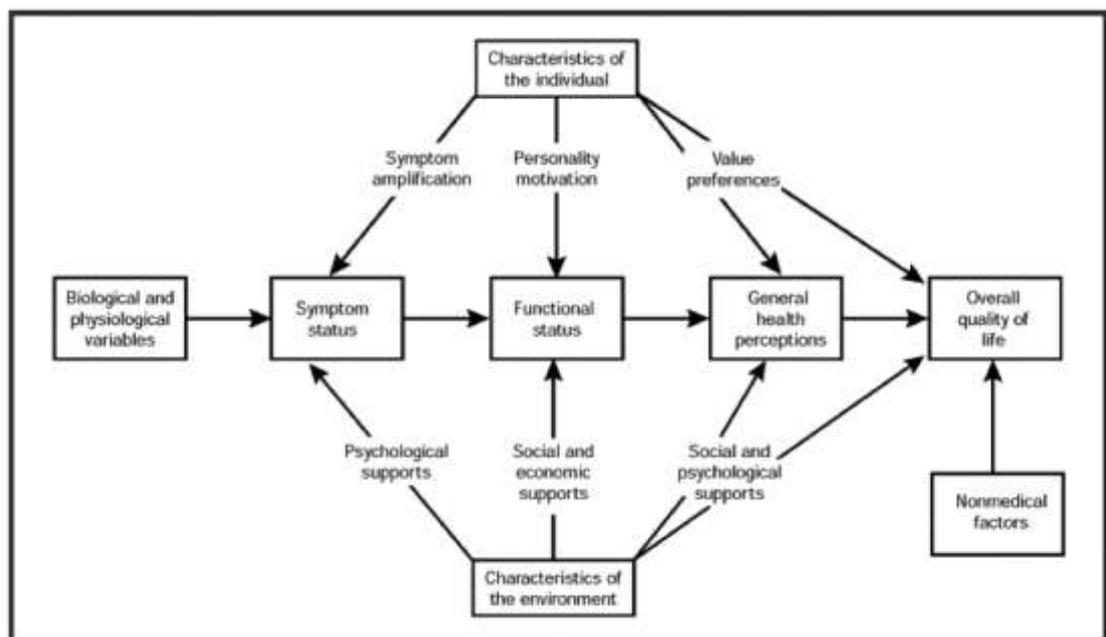


Figure 2-7 Wilson and Cleary model of health-related quality of life (Wilson & Cleary, 1995)

With the better understanding of disability, and because the interest is more toward the social model of disability that emphasises the role of environmental factors in health outcomes, a more holistic framework is needed that captures the synergistic and/or antagonistic factors that might change the previous rather simplistic linear relationship between health and QoL. Ferrans and her colleagues (2005) revised the Wilson and Cleary model; they concentrated mainly on the contextual factors that were originally included in Wilson and Cleary model but were not discussed. Their revised model is based on the ecological model of McLeroy and colleagues (McLeroy et al., 1988), as modified by Eyler et al. (2002), to clarify the multiple layers of influence on health outcomes at both individual and environmental levels in HRQoL (Ferrans et al., 2005). McLeroy and colleagues' model indicates five levels of influence: (a) intrapersonal factors, (b) interpersonal factors, (c) institutional factors, (d) community factors, and (e) public policy.

In the revised model, out of these five levels of contextual factors, intrapersonal factors are the individual characteristics, and the remaining four levels are the environmental characteristics. Individual characteristics are categorized as demographic, developmental, psychological, and biological factors. Characteristics of the environment are categorized as either social or physical. Therefore, the main changes from the original Wilson and Cleary model are: (a) indicating that biological function is influenced by characteristics of both individuals and environments; (b) deleting nonmedical factors; and (c) deleting the labels on the arrows that tend to restrict characterization of the relationships, (Figure 2.8). This way of categorization is broad enough to include any factor that might have an impact on the ultimate health outcome, which is QoL.

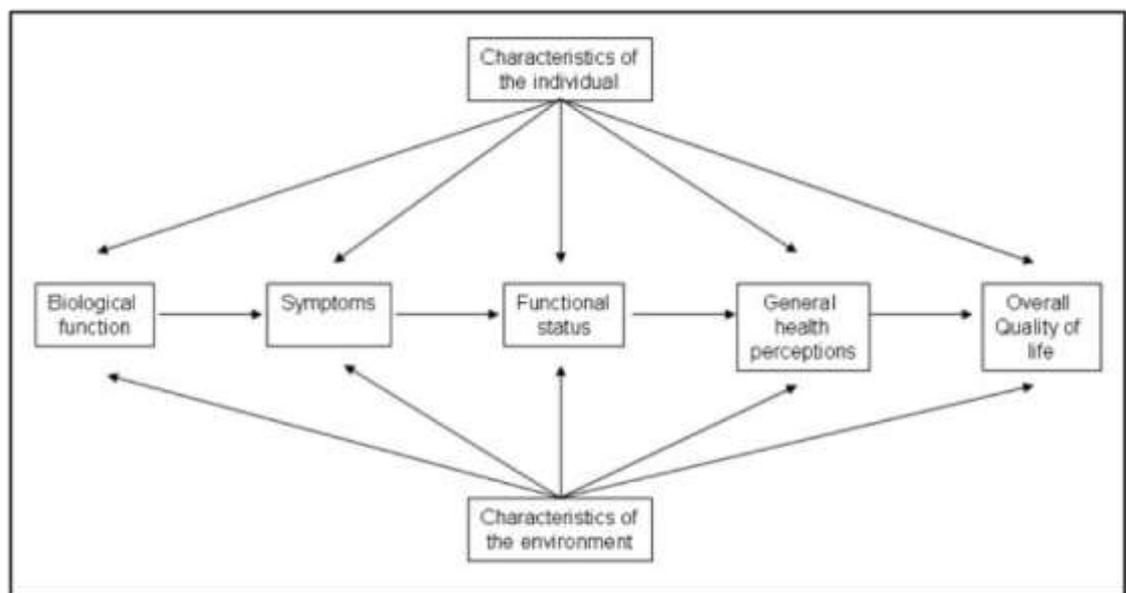


Figure 2-8 Revised model of Wilson and Cleary HRQoL (Ferrans et al., 2005)

This research aimed at developing an OHRQoL measure for children/adolescents with Down Syndrome, and intended to base it on the latest World Health Organization classification of health and health-related states, known as the International Classification of Functioning, Disability, and Health (ICF) (WHO, 2001), (Figure 2.9), in order to overcome the limitations imbedded in the previously used model, ICIDH.

ICF is a multidimensional and complex model in which a combination of biological (e.g. health condition), psychological (e.g. personal factors), and social factors (e.g. environmental factors) contribute to health in various contexts (Brondani & MacEntee, 2014), and in a bi-directional way understanding the fluctuating nature of health status in some cases. With a focus on social roles, the ICF offers an integrated and holistic view that is intricately linked to health and quality of life as an individual's perceptions in the context of their culture and value systems, and their personal goals, standards and concerns (WHO, 2001). The WHO recognizes the large social variance associated with personal factors in the ICF.

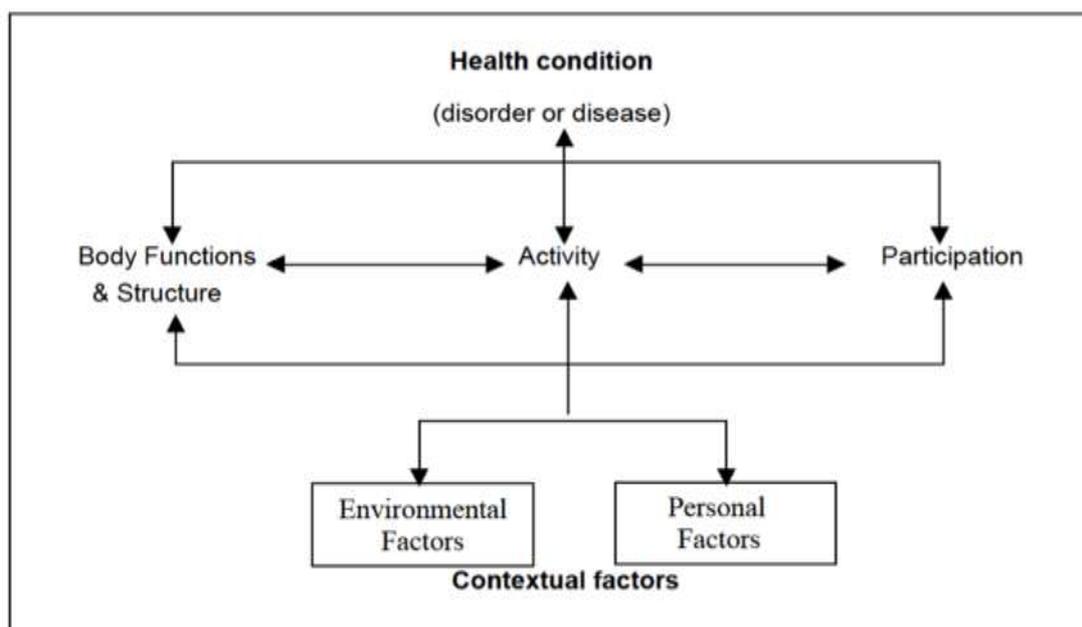


Figure 2-9 International Classification of Functioning, disability, and health, ICF (WHO, 2001)

Therefore, the modified version of Wilson and Cleary model, and ICF are now very similar in capturing the important role of both personal and environmental factors on health outcomes.

Oral health researchers have critiqued ICF as a broad conceptual framework for models and measures of oral health, and a more recent model of oral health has been developed based upon the ICF (McEntee, 2006). This new oral health model demonstrates the dynamic bio-psychosocial aspects of function and disablement of the mouth and was further refined by Brondani et al. (2007) to represent the constituents of health, not illness, and accommodate current views of health as a dynamic phenomenon of adjustment, coping and adaptation, Figure 2.10.

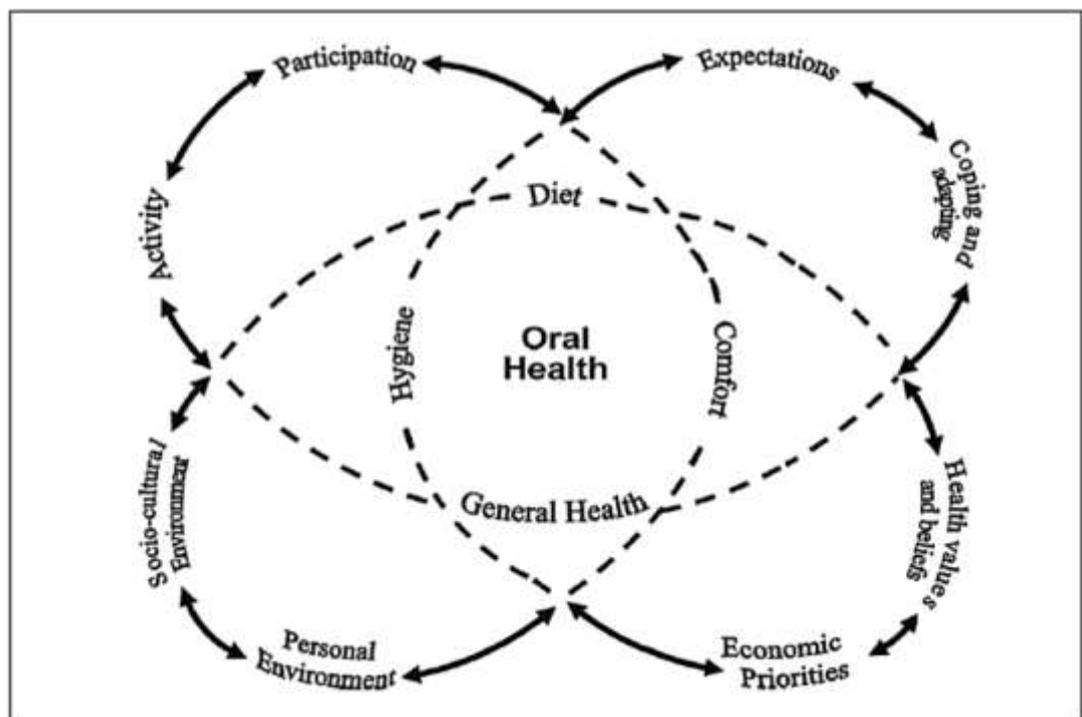


Figure 2-10 Refined model of the key components relating to oral health (Brondani et al., 2007)

The main advantages of this revised model are that it represents oral health in a more dynamic way and it reflects the views of lay people in their health conditions and the process of portraying oral health. This may help understanding oral health in a more positive way even in the presence of dysfunction than previous more linear displays of oral disease and its consequences that have been used for a long time in research and mainly reflect objective views. As this revised model was developed to accommodate older people's views that had experienced some sort of oral

impairments due to oral diseases and/or ageing process, the model might not be completely applicable to other segments of the population. Therefore, the model seems partially suitable for use as a base to assess oral health among people with varieties of disability, but some modifications are needed according to the type and nature of oral health problem studied. It should be noted as well that the model did not give any directions to the proposed links, and hence, it is very difficult to use it as an operationalized model of oral health, its consequences and mediating/moderating factors.

Do we need a new OHRQoL framework for people with Down Syndrome?

Limitations of the existing and widely used HRQoL model (Wilson and Cleary) necessitate the need for adjustment before using it as a framework for health outcome measures applicable across different segments of populations. For example, the Wilson & Cleary model of HRQoL does not capture the impact of existing disability on different aspects of an individual's life. This demands a modification of the existing model, such as adapting the revised version of Wilson and Cleary HRQoL model (Ferrans et al., 2005). This can be achieved for example by adding disability and its related biological and physiological factors to the individual characteristics, and stigma attached to disability, and its social acceptance to environmental characteristics.

In addition, individuals with disabilities are believed to have lower QoL than their able-bodied peers. Kottke (1982) expressed this view when he stated; "the disabled patient has a greater problem in achieving a satisfactory quality of life. He has lost, or possibly never had, the physical capacity for the necessary responses to establish and maintain the relationships, interactions, and participation that healthy persons have." However, research evidence presents a more complex picture. In practice, the difference in the patients' perceptions of personal health, well-being and life satisfaction are often discordant with their objective health status and disability (Albrecht & Higgins, 1977; Albrecht, 1994), and with other views confirming the phenomenon of the 'disability paradox'. This paradox is encapsulated in the following question: 'why do many people with serious and persistent disabilities

report that they experience a good or excellent QoL when to most external observers (such as objective measures) these people seem to live an undesirable daily existence'?' (Albrecht & Devlieger, 1999). In a study aimed to understand factors that contribute to such a paradox, Albrecht and Devlieger (1999) concluded that establishing and maintaining a sense of balance between the body, mind and spirit and with the individual's social context and environment can lead to this phenomenon. However, they noted that this explains only part of the disability paradox and they called for further studies to assess why such a paradox work with some individuals but not others.

The case might be similar when considering OHRQoL, however, this has not been fully investigated yet, and relevant research needs to be conducted. Figure 2.11 represents the potential impact of the presence of disability and its associated conditions on patient's QoL. This figure represents the factors such as health factors that partly contribute to overall quality of life but not the whole construct of QoL.

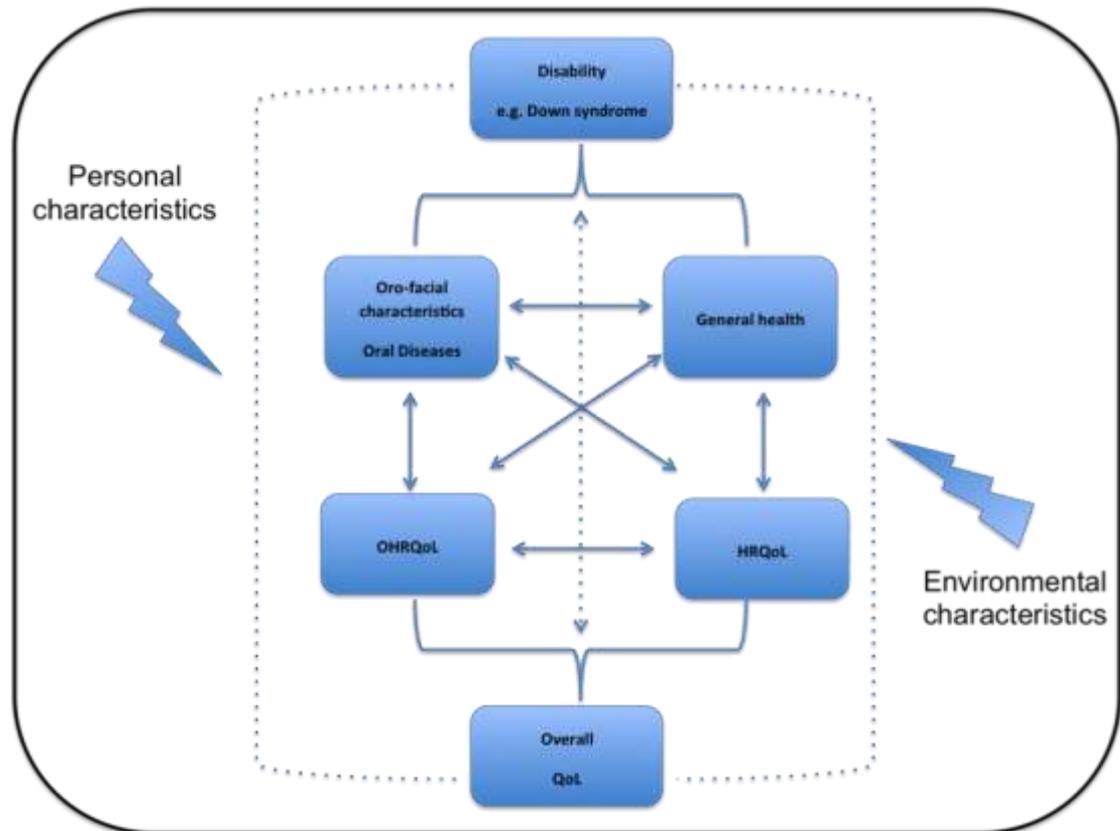


Figure 2-11 Impacts of the presence of disability on individual's QoL

This hypothesised framework emphasizes the importance of personal and environmental factors that are highly relevant among individuals with disabilities and/or chronic conditions, who are at risk of negative psychosocial sequelae as a result of their existing condition ‘disability’, not only the sequelae of disease itself.

The framework is too broad for the aim of this study, although it was important to highlight the wider range of factors that might impact on individuals’ QoL. It was also vital to show how factors (physical, psychological, environmental) interact with each other, possibly also in different ways. This broad conceptual framework guided the study and also helped in visualizing the very complex, interrelated nature of QoL in people with disabilities and/or chronic conditions. More research is needed to facilitate understanding the sequences of health conditions in actual life, and determine the factors and pathways that lead to better or worse QoL outcome.

In the field of measuring QoL among individuals with intellectual disability the case is even more challenging for different reasons. First, those people need special considerations if they are chosen to participate by themselves (e.g. pre-evaluation of the intellectual abilities). Second, if proxy measures were used, and this is the situation in many cases especially among those with severe intellectual disabilities, it is important to assess the level of agreement between actual self-reported and proxy measures. It is also of prime importance to assess the proxy's psychological state and other factors that might influence proxy reports (e.g. acceptance concept and its impact on the mother's perceptions and expectations).

Figure 2.12, presents the theoretical framework of this project, however, the main aim is not to investigate the indicators and predictors of the OHRQoL among individuals with Down Syndrome, but the initial step of understanding the perceptions of mothers about their children OHRQoL, and come up with the most reflective dimensions of OHRQoL of children/adolescents with Down Syndrome.

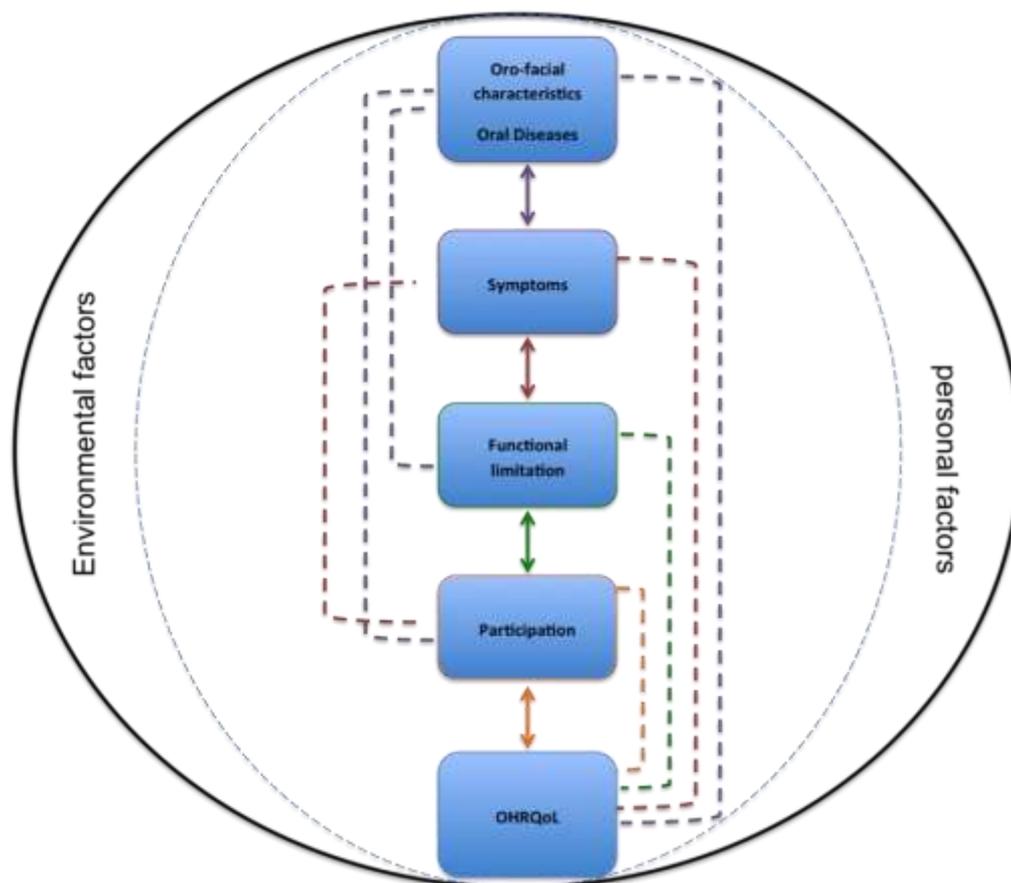


Figure 2-12 Conceptual model of OHRQoL for individuals with disability (adapted from Wilson & Cleary, 1995)

As it shown in figure 2.12, the impact can occur between any level of disease outcomes, and the direction of arrows does not necessitate a linear one-way relationship, but fluctuation can occur depending on the interaction between the various factors. No arrows are drawn from personal and environmental factors assuming that such factors can have an impact at any level, and with each other.

Personal factors in this model refer to demographic, psychological, biological, and developmental factors. This broad inclusion of such factors can accommodate for many variables such as the impact of the presence of disability and its related physiological (biological) changes, and/or the effect of developmental stage of perceiving and reporting OHRQoL. The model represents also the environmental

factors that include both social and physical environmental characteristics. This is highly important to consider when assessing quality of life for individuals with disability because enabling or disabling environments can have a greater impact on the quality of life of individuals with disabilities than the disease itself. For example, in case of children with Down Syndrome in which they are characterized by difficulty speaking, if the social environment does stigmatize the child, then this could be the reason behind poor reports of OHRQoL, not the condition itself; and therefore the intervention should be directed toward the broader aspect (social environment).

What does the model add to previous OHRQoL measures?

All previous OHRQoL measures of children were developed for use among the mainstream populations; however some have been used in the few studies to assess OHRQoL among children with types of disabilities such as cerebral palsy and Down Syndrome without investigating the actual and relevant dimensions of OHRQoL from patients' perspectives. This shows the need to investigate dimensions of OHRQoL among children with chronic conditions or disabilities such as Down Syndrome to be able to understand their perceptions and concerns of their OHRQoL and thereafter decide whether we need to develop a new measure or simply adapt an existing one.

This study model highlights the importance of both personal and environmental factors (contextual factors) at different levels of disease outcome as this was clearly stressed at the latest International Classification of Functioning, Disability, and Health (ICF) (WHO, 2001), and the revised Wilson and Cleary model of HRQoL (Ferrans et al., 2005).

2.9 Aims and objectives

Overall aim

To develop and test an Oral Health-Related Quality of Life measure for children/Adolescents with Down Syndrome (OH-QOLADS).

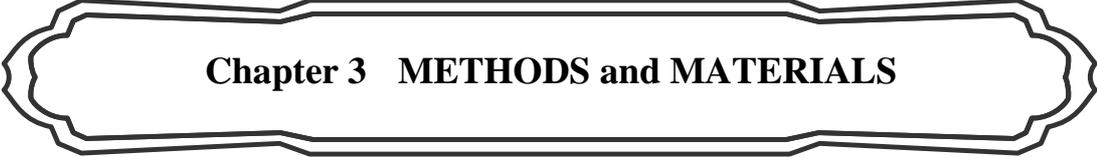
To achieve this aim, study was conducted in two phases;

Objectives of phase-one:

- 1- To explore mothers' views concerning the oral health status of their children/adolescents with Down Syndrome
- 2- To investigate mothers' perceptions of how oral health might impact on the QoL of children with Down Syndrome
- 3- To examine mothers' perceptions of how oral health conditions of their children with Down Syndrome might impact on the family's QoL

Objectives of phase-two:

- 1- To develop an OH-QOLADS measure based on the results from phase one and a literature review
- 2- To pilot test the developed OH-QOLADS measure to assess its applicability and suitability in a sample of mothers of children with Down Syndrome
- 3- To assess the reliability (internal consistency and test-retest reliability) of the developed OH-QOLADS measure on a sample of children/adolescents with Down Syndrome
- 4- To assess the face, content, construct, and discriminant validity of the developed OH-QOLADS on a sample of children/adolescents with Down Syndrome.

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Chapter 3 METHODS and MATERIALS

3.1 Introduction

This chapter covers the methods and materials used to carry out the study. Before describing the study methodology, the next section will briefly provide a broad overview of the Kingdom of Saudi Arabia, where the study was conducted.

Overview of the Kingdom of Saudi Arabia

Saudi Arabia is the biggest country in the Middle East, occupying most of the Arabian Peninsula in Southeast Asia. Saudi Arabia has been categorized in the upper middle-income group, which would increase the assumption that there is a fairly high standard of living within the Kingdom. The 2011 census placed the population of Saudi Arabia at 28.3 million, compared with 22.6 million in 2004 (Central Department of Statistics and Information, 2011a). The annual population growth rate for 2004 to 2010 was 3.2% per annum (Central Department of Statistics and Information, 2011b).

Healthcare system in Saudi Arabia

The healthcare system in Saudi Arabia can be classified as a national health care system in which the government provides health care services through a number of government agencies. However, there is a growing role and increased participation from the private sector in the provision of health care services. The Ministry of Health (MOH) is the body entitled to provide preventive, curative and rehabilitative health care for Saudi population. The MOH is considered the lead Government agency responsible for the management, planning, financing and regulating of the health care sector. The MOH also undertakes the overall supervision and follow-up of health care related activities carried out by the private sector. Therefore, the MOH can be viewed as a national health service for the entire population. Figure 3.1 shows the current structure of health services in the country.

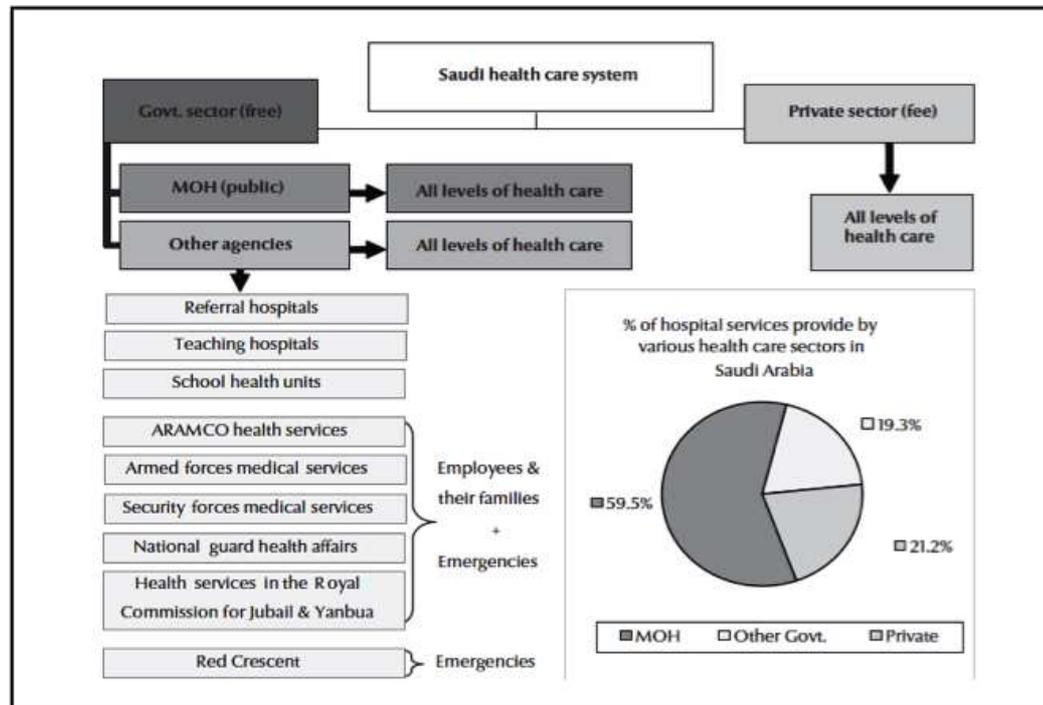


Figure 3-1 Current structure of health care sectors in Saudi Arabia (Health Statistical year Book, 2009)

Disability in Saudi Arabia

As per the Labor and Workman Law of Saudi Arabia, a person with a disability is defined as “any person whose capacity to achieve and continue a suitable job has actually diminished as a result of a physical or mental infirmity” (Labor and Workman Law, 1969). As it is indicated in this definition, disability in Saudi includes both developmental and acquired aspects, which are the result of either poor health condition or trauma. As a result of the lack of a common measure, different definitions of disability across existing studies aimed at assessing disability levels in the country, and the limited epidemiological research in Saudi Arabia, it is difficult to assess the exact burden of disability in the country although it is commonly acknowledged that the burden is generally high due to social and environmental

factors (Al-Jadid, 2013). Despite the growing awareness, research to determine the pattern of disabilities in Saudi Arabia is still limited (Al-Hazmi et al., 2003). A study from Qaseem region reported that the incidence of physical disability (1.7%) was higher in children as compared to intellectual disability (1.4%) (AlSekait, 1993). A national survey that was conducted among 60,630 children showed that 6.33% children were reported as having a disability. The survey also reported that the highest ratio of children with disabilities was in the Jazan region (9.9%) while Riyadh had the lowest (4.36%). The most common disability was physical disability (3%) followed by intellectual disability (1.8%) (Al-Hazmy et al., 2004). Furthermore, the highest proportion of disability was found among children of disabled parents, later in life pregnancies and mothers who had not received medical care and vaccination during pregnancy (Al-Hazmy et al., 2004). According to some studies conducted in the region, road traffic accidents, stroke, cerebral palsy, head and spinal cord injuries, infection and inflammation were considered as the major causes of mortality, hospitalization and chronic disabilities (Central Department of Statistics and Information, 2011a; Kingdom of Saudi Arabia Healthcare Overview, 2012). Disabilities associated with genetic causes are also significant (Al Essa et al., 1997).

In Saudi Arabia, similar rates of live Down Syndrome births have been found with a prevalence of 1 in 554 live births (Niazi et al., 1995). However, there is no clear data on the exact number of individuals with Down Syndrome, their age, and distribution.

Policies and practices on disability in Saudi Arabia

Saudi Arabia is based on the Islamic Sharia law, which emphasizes human rights and the right of persons with disabilities to live with dignity (Al-Jadid, 2013). In 1987, the legislation of disability passed as the first legislation for people with disabilities in Kingdom of Saudi Arabia. The disability legislation contains important provisions that assures people with disabilities have equal rights to those of other people in society (Ministry of Health Care, 2010). In 2000, the disability code was passed by the Saudi government to pledge that people with disabilities have access to free and appropriate medical, psychological, social, educational, and rehabilitation services through public agencies (The Provision Code for Persons with Disabilities in

Kingdom of Saudi Arabia, 2004). The above guiding principles support the equal rights of individuals with disabilities in obtaining free and appropriate education and medical facilities. However, these laws were passed a decade ago and not practiced well in the country. The lack of the effective implementation has created in a gap between the framework of these laws and the provision of services, resulting in a lack of many essential services for persons with disabilities (Al-Jadid, 2013).

Healthcare for people with disability in Saudi Arabia

Over the last two decades, the Ministry of Health has established numerous rehabilitative services for persons with disabilities and other residents in the country. A majority of these programmes offer physical, occupational, speech and hearing therapy as well as prosthetic and orthotic services within the existing health care service system and infrastructure. Rehabilitation programmes and facilities, as an integral part of health care delivery services, have received due attention by government authorities, with services being made available to all citizens and residents (Al-Jadid, 2013).

Greater attention has been placed on the health care services of people with disabilities rather than on their education and training, and there is very little attention given to helping people with disabilities gain employment. In addition, institutions for persons with disabilities are largely available in urban rather than rural areas, with an uneven distribution of facilities irrelative to persons with disabilities distribution (Al-Jadid, 2013). However, balanced distribution of facilities requires a robust estimate of individuals with disabilities' actual distribution and needs.

Clinical oral health status of Saudi children

Studies on the prevalence of dental caries at the country level indicated that Saudi Arabia is experiencing an 'epidemic' of dental caries that mandate immediate collaborative, systematic, and multi-level interventions (Marghalani et al., 2014). A recent systematic review of dental caries prevalence in Saudi Arabia concluded that childhood dental caries is a serious dental public health problem that warrants immediate attention of the government and dental profession officials. Results

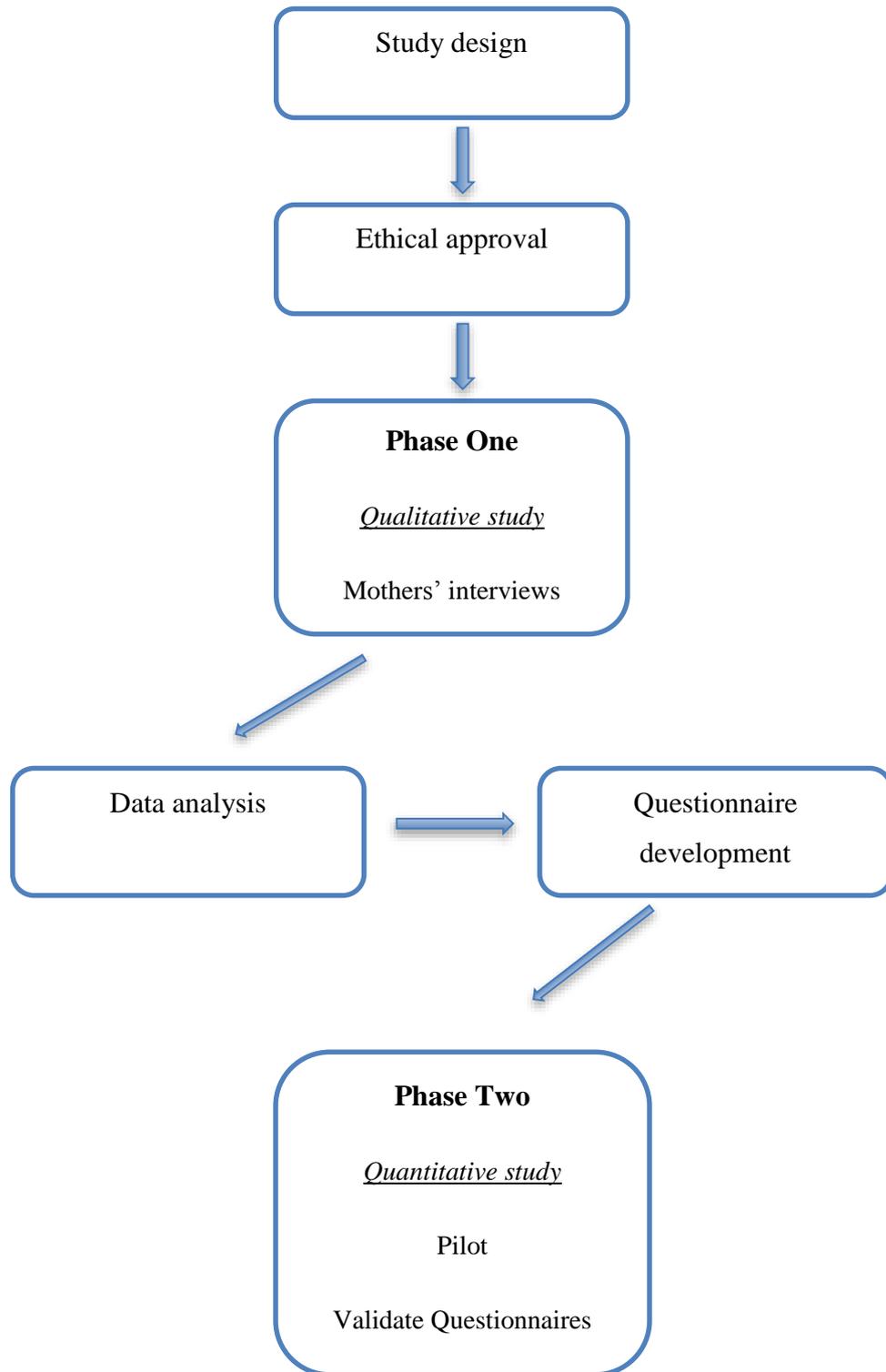
showed that the national prevalence is estimated to be 80% in the primary dentition with a mean dmft score of 5.0, and it is estimated to be 70% for children's permanent dentition with a mean DMFT score of 3.5 (Al Agili, 2013). The former presented data are for mainstream children; however the research on the prevalence of dental diseases and conditions among Saudi individuals with disabilities is scarce although studies suggested that people with disabilities are at greater risk for poorer oral health.

3.2 Study Design

The overall aim of this research was to develop an OHRQoL measure for children/adolescents with Down Syndrome. In order to achieve this aim, a cross-sectional, two-phased study was undertaken. The study followed a mixed method approach (qualitative and quantitative).

In phase one, a qualitative approach (in-depth interviews) was used to identify the dimensions of OHRQoL for this specific group (children with Down Syndrome) from the mothers' perspective. Then data collected from the interviews was analysed, and with the review of existing OHRQoL literature, a questionnaire about the impact of oral health on the QoL of children with Down Syndrome was formulated. The second phase aimed at validating the developed questionnaire. Figure 3.2, presents the study sequence.

Figure 3-2 Flowchart of the study sequence



3.3 Ethical Approval

Ethical approval was obtained from the Research Ethics Committee of University College London (Appendix 4), as well as King Saud University (Appendix 5). In addition, a permission to conduct the study and access families with children with Down Syndrome was granted from both the Ministry of Social Affairs (Appendix 6), and the Ministry of Education (Appendix 7). The principals of the centres who agreed to participate received a thorough explanation of the study in person or by telephone call, and written summary of the study describing its aims was also provided (Appendix 8). Information sheets and consent forms for both phases were sent to all participating families (Appendix 9).

3.4 Initial Contact and Arrangements with Down Syndrome Centres

Before starting the study, a search for Down Syndrome centres in Riyadh city of Saudi Arabia was conducted. Four main centres providing care to children with Down Syndrome in Riyadh were identified and contacted by telephone and, having indicated their willingness to take part in the study, formal letters were sent explaining the purpose of the study and asking them to take part (Appendix 8). Approval to access other special needs centres was sought from the Ministry of Social Affairs (including Al-Wafa rehabilitation centre, and Al-Khatwa society for special needs). The Ministry of Education was also contacted to get approval to access government schools with inclusive education programmes and institutions, which provide intellectual rehabilitation.

3.5 Phase One (Qualitative Study)

3.5.1 Training on qualitative research methods

In order to improve the researcher's skills in qualitative research methods, and before starting the phase one data collection, selected courses in qualitative research skills (Introduction to qualitative research, Qualitative research design, Depth interviewing skills, and the analysis of qualitative data) were attended at the National Centre for

Social Research (NatCen). Training in NVivo analytic software was also undertaken to facilitate a thorough analysis of the phase one interviews in order to identify the dimensions of OHRQoL of children/adolescents with Down Syndrome from their mothers' perspective

3.5.2 Sample selection

Since qualitative research does not aim for generalization of findings, the sample was selected and determined in a way that allowed the study objectives to be met (Bryman, 2012; Patton, 2015). The focus of phase one was to explore the views and opinions of mothers of children/adolescents with Down Syndrome about how oral health might impact on their QoL. Therefore, a purposive sampling technique was followed in which all data were collected from mothers caring for their children with Down Syndrome. A purposive sampling is defined as "selecting information-rich cases to study cases that by their nature and substance will illuminate the inquiry question being investigated" (Patton, 2015).

A sample of mothers was selected in collaboration with two Down Syndrome day-care centres in Riyadh city (Down Syndrome Charitable Association, and SAUT Down Syndrome). The mothers were selected at different ages, levels of education and ages of their children, to consider variations in their perceptions that might occur due to these characteristics (see justifications below). The selected mothers were the principle carers of the individuals with Down Syndrome.

3.5.2.1 Justification of qualitative sample selection

In purposive sampling, participants are selected because they are information-rich whose answers will yield insights and in-depth understanding and illuminate the questions under study (Bryman, 2012; Patton, 2015). In order to account for any differences in mothers' perceptions that may occur because of their different age or educational level, mothers were selected at two age groups (35 and below, and 36 and above at time of giving birth to child with Down Syndrome). The mothers' educational level was also considered so that the sample was a mix of mothers with

no school or equivalent qualifications, and some with qualifications beyond school level. In terms of the age of the children, the sample consisted of mothers with younger adolescents (12-15 years) and mothers with older adolescents (16-18 years). The rationale for these selection criteria is explained below:

1- Age of mothers: It is well known that the risk of having a child with Down Syndrome strongly correlates with mother's age (Huether et al, 1998; Strauss et al, 2013). So the reason for dividing mother's age as 35 and below, and 36 and above was to explore the different views and experiences of young mothers (with less risk of having child with Down Syndrome) versus older mothers.

2- Education level of mothers: the reason for selecting mothers from different socio-economic levels (measured by level of education: no qualification, school or equivalent, and qualifications beyond school level) was to map the diversity in oral health status as well as perceptions of its impacts on QoL. Studies have shown the presence of social gradients in oral health with poorer conditions among more disadvantaged groups (Sabbah et al, 2007; Geyer et al, 2010). The level of mothers' education might also affects their perceptions of oral health and its impact on the QoL of their children, and that of the family as a whole.

3- Age of the child: since the study aimed at children/adolescents (12-18 years old) with Down Syndrome, the selected sample was divided into two categories: 12-15 years and 16-18 years. For all teenagers, including those with Down Syndrome, adolescence is a period of development characterised by a shift from dependence to independence, but younger teenagers (12-15 years) with Down Syndrome may still need a high degree of support for almost all aspects of their daily lives, including personal daily care activities. In later adolescence (16-18 years) many young people with Down Syndrome make significant progress as they begin to take more responsibility for their daily lives, and therefore become more independent. To cover the range of differences that may occur during the teenage years, mothers were selected considering their children's development age.

3.5.3 Sampling matrix

The key indicator for sample size was data saturation, though it is almost impossible to guarantee the saturation since each participant has a characteristic that might add to the question studies, in addition to the different views and opinions of interviewees in achieving data saturation. In order to minimize this kind of error, the researcher (interviewer) received training in qualitative data and interviewing skills in addition to careful reviewing of the existing literature.

The sample size in this study was 20 interviews. This number was reasonable for the purpose of exploring mothers' experiences and opinions concerning their children's oral health, and how their oral health impact on the QoL of the child and that of the family as a whole. However, it was planned that the number might be increased until themes start to re-occur and new categories or explanations stopped emerging "data saturation" (Mason, 2010). The characteristics of the interviewed mothers are listed in table 3.1.

Table 3-1 Phase 1 sample matrix

	Girls	Boys
Age of mothers		
20-35	4 – 6	4 – 6
36+	4 – 6	4 – 6
Education level of mother		
No qualification	2 – 5	2 – 5
School or equivalent	3 – 5	3 – 5
Post school	3 – 4	3 – 4
Age of child		
12-15	4 – 6	4 – 6
16-18	4 – 6	4 – 6
Total	10	10

3.5.4 Interview topic guide

3.5.4.1 Topic guide development

After the relevant information for this study was clearly defined and before data collection started, a topic guide was prepared (Appendix 10). The topic guide was used to increase the comprehensiveness of the data and makes data collection somewhat systematic for each interviewee, and to keep the interview focus around the study objectives (Patton, 2015) The topic guide was developed after reviewing the relevant literature (a priori), and the themes that were incorporated were closely aligned with the research objectives (Mays & Pope, 2000). The structure of the topic guide was designed around these information themes, while taking care of the flow and manner of the questions, figure 3.3.

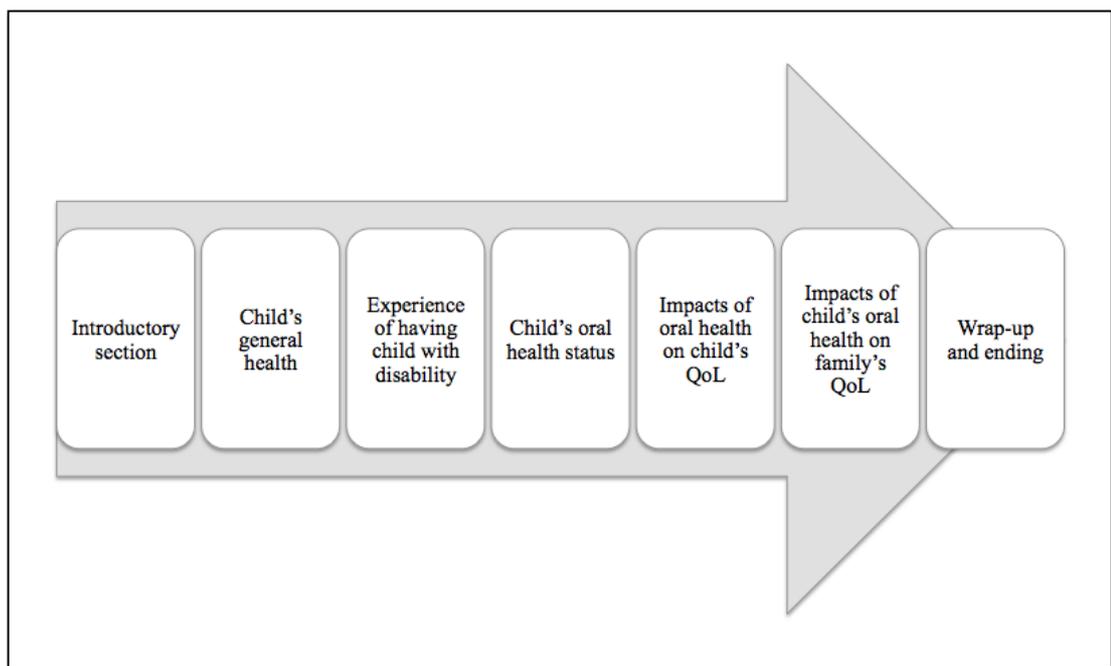


Figure 3-3 Structure and flow of the topic guide

3.5.4.2 Topic guide translation

The process of translating the topic guide began soon after it was finalized. The guide was translated into Arabic for ease of data collection. Then, a person fluent in

both languages translated the topic guide back into English to check the accuracy of the translation, and any inconsistencies were sorted out by discussion between the researcher and translator. Asking mothers about their experiences in having child with disability and how they coped with it is very emotional aspect to investigate or ask about. Therefore, it was not the first part to be explored in the interview.

3.5.5 Fieldwork preparation

The phase one data collection took place in December 2012. Table 3.2 shows the timetable of the fieldwork data collection.

Table 3-2 Fieldwork timetable of phase 1

Activity \ Week	Week 1 3 rd to 5 th Dec 2012	Week 2 8 th to 12 th Dec	Week 3 15 th to 19 th Dec	Week 4 22 nd to 26 th Dec	Week 5 29 th Dec to 2 nd Jan 2013	Week 6 5 th to 7 th Jan
Arrangements	*	*	*	*	*	*
Visit to Centers		*		*		
Interviews with Mothers		*	*	*	*	*

Interviews and data collection were conducted at two centres catering for children with Down Syndrome:

- The Down Syndrome Charitable Association (DSCA) contacted the families and arranged directly with mothers who consented to take part in the study, and arranged the date and time of the interviews. (All interviews were carried out at DSCA School in a quiet and isolated room.)

- The SAUT centre decided to contact the families first, describe the aims of the study and ask them to participate. Then only the approved consent forms were returned with contact numbers. After that, the researcher arranged interview locations and times with the mothers. All interviews were conducted at SAUT school in a quiet and isolated room known as the “school clinic”, while one of the interviews was carried out at the mother’s request, at her place of work.

3.5.6 Data collection procedures

One researcher (AJ) interviewed the selected mothers separately. Before each interview, the purpose and method of the study were explained and an assurance of complete confidentiality and anonymity was given. Participants were invited to ask any questions regarding the purpose of the study before signing a written consent form.

The interviews were reflexive, with the interviewer exploring topics raised by the participants who were encouraged to speak about their perceptions, experiences, and thoughts regarding oral health status and its impact on their children’s QoL and also on the family. A topic guide was used to ensure all areas of interest were explored.

3.5.7 Data management and data analysis

All data was recorded using an Olympus recorder (WS 811), in addition to supplementary notes that were taken during the interviews to record important points and/or non-verbal data (emotions, facial expressions, body language and interviewee mood). After each interview, the recorded conversation was transferred to a computer and saved under a serial number that matched the one on the notes of the same participant (containing place of the interview, date, time, and the length of the interview). Interviews were then transcribed verbatim by a specialized transcriber and reviewed by the researcher. Transcriptions were then translated into English for further analysis. To validate the translation process, two different persons who were fluent in both languages translated several transcripts to English. The comparison in the translated transcripts revealed no major differences.

All interviews were then analysed by the researcher to identify common themes. The themes represented key issues and concepts that were raised by the respondents, and also reflected the aims and objectives of the study. To validate the themes identified by the researcher (AJ), several transcripts were read and coded by another researcher (RGW).

For thorough and detailed analysis, the 'Framework' analytic method (Ritchie & Spencer, 1993) was applied systematically to all transcripts and the themes were assembled into a series of thematic charts. The context of the information was retained and the page of the transcript was noted so that it could be possible to return to a transcript in order to explore a point more or extract a quotation. A matrix of themes and respondents was compiled and used to map the range and nature of phenomena, and to identify associations between themes with a view to providing explanations for the findings (Pope et al., 2000). Using this method, the accounts of all mothers' views and opinions were explored within a common analytical framework. The ordering of the data in this way helped to highlight the full range of views, experiences and behaviours expressed and influences that underpin them.

The framework method involves five separate but interconnected stages that include:

- Familiarisation,
- Identification of themes,
- Indexing,
- Charting, and
- Mapping and Interpretation.

1- Familiarisation: whole or partial transcription and reading of the data.

2- Identification of the thematic framework: this is the initial coding framework, which is developed both from *a priori* issues (i.e. the literature review on OHRQoL) and from emerging issues from the familiarisation stage. This thematic framework was developed and refined during subsequent stages.

3- Indexing: the process of applying the thematic framework to the data, to abstract all relevant data.

4- Charting: data across all interviews were combined in one chart or matrix. This chart is devised according to the previous established framework. This allows comparison between respondents within the same heading or theme.

5- Mapping: at this stage pieces of data set can be mapped together and analysed as a whole to make interpretations.

3.5.8 Quality assessment

The coding process was validated by another researcher (RGW) in order to minimise interpretation bias in identifying and finalising the themes/codes from mothers' speech. All transcripts were coded using predefined codes (Table 3.3 & 3.4) that were explored thoroughly and updated along the analysis process.

Table 3-3 Code description of the study

Oral Health Condition	<p>Any condition related to the mouth and its contents including teeth and the gums (gingiva) and their supporting connective tissues, ligaments, and bone, the hard and soft palate, the soft mucosal tissue lining of the mouth and throat, the tongue, the lips, the salivary glands, the chewing muscles, and the upper and lower jaws.</p> <p>Oral functional conditions like speaking</p>
Oral Health-Related Quality of Life	Any impact of child's oral health on different aspects of his/her life (e.g. school's homework, friendship)
Physiological impact	Any impact of oral health on physiological aspects of the mouth like pain, discomfort either acute or chronic
Impacts on daily living activities/ functional	Any impact of child's oral health on different daily activities such as: playing, eating, speaking
Psychological/ emotional impact	Any impact of child's oral health on his/her psychological or emotional state such as: anxiety, depression, or embarrassment
Social impact	Any impact of child's oral health on his/her social life such as: relationship with friends, chatting with other people
Family's Quality of Life	Any impact of child's oral health on different aspects of family's life that includes; family activities, relationships, or emotions

Table 3-4 Code list used for the study

Original	Expanded
<p style="text-align: center;"><u>Child's OHROoL</u></p> <p>1- Physiological impact</p> <p>2- Impacts on daily activities</p> <p>3- Psychological/ emotional impacts</p> <p>4- Social impacts</p> <p style="text-align: center;"><u>Family's QoL</u></p> <p>1- Emotions</p> <p>2- Activities</p> <p>3- Conflicts</p>	<p style="text-align: center;"><u>Child's OHROoL</u></p> <p><i>Physiological impact</i> Pain Discomfort</p> <p><i>Impacts on daily activities</i> Eating/ drinking Sleeping Talking/ Speaking Homework Playing Brushing teeth</p> <p><i>Emotional impacts</i> Crying Stop laughing Get quite Upset Shyness Inferior to other people Embarrassed Introvert Confidence Nervous Angry</p> <p><i>Social impacts</i> Play with friends Relationships with friends Isolation</p> <p><i>Behavioral impact</i> Stubbornness</p> <p style="text-align: center;"><u>Family's QoL</u></p> <p><i>Family emotions</i> Depressed Angry Upset Distress Self-blame/ guilt Worry Irritated Afraid that child in pain</p> <p><i>Family activities</i> Cancel scheduled activity (family gathering, meetings, parties) Job time Time off other family members (mother, partner or</p>

	<p>siblings) Sleeping</p> <p><i>Family conflicts</i> With other family members if they need to go to dental appointment Other siblings might become jealous if more time was given when child in pain (toothache)</p>
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3.6 Phase Two (Quantitative Study)

3.6.1 Questionnaire development

The questionnaire was developed using the process described by Guyatt et al. (1987), and Juniper et al. (1996). A review of existing OHRQoL measures for children that included measures of the family impact of child chronic conditions (i.e. Child-OIDP, CPQ, P-CPQ, FIS, COHIP) was used to form the preliminary pool of items. In addition, items emerged from the phase one interviews were added to ensure the comprehensiveness of the item pool, as shown in Figure 3.4. In addition, some questions were used to assess the perceptions of mothers about the oral health status of their children. The questionnaire included some information about demographic data, the child's general health and other basic questions (Appendix 11).

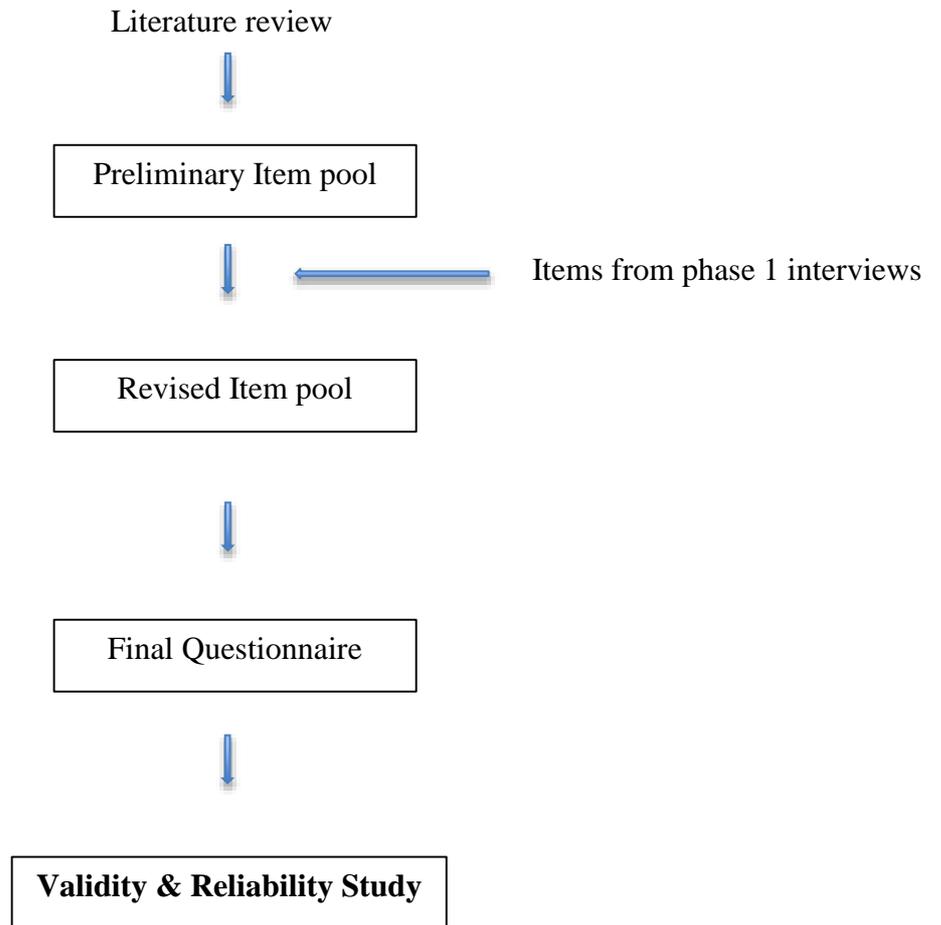


Figure 3-4 Summary of the process used to develop the questionnaire

3.6.2 Linguistic validation plan

After finalizing the questionnaire and before starting the validation process, the questionnaire went through a linguistic validation process, since the main data collection was conducted in Arabic. The translation was conducted using the standard linguistic validation procedures (Guillemin et al., 1993; Acquadro et al., 2008). The process comprises a series of steps (Figure 3.5); the original questionnaire was forward translated to Arabic by two separate translators, and the consensus was translated back into English by two different translators, then the results were discussed with the questionnaire developer/researcher to check if the concepts remained unchanged.

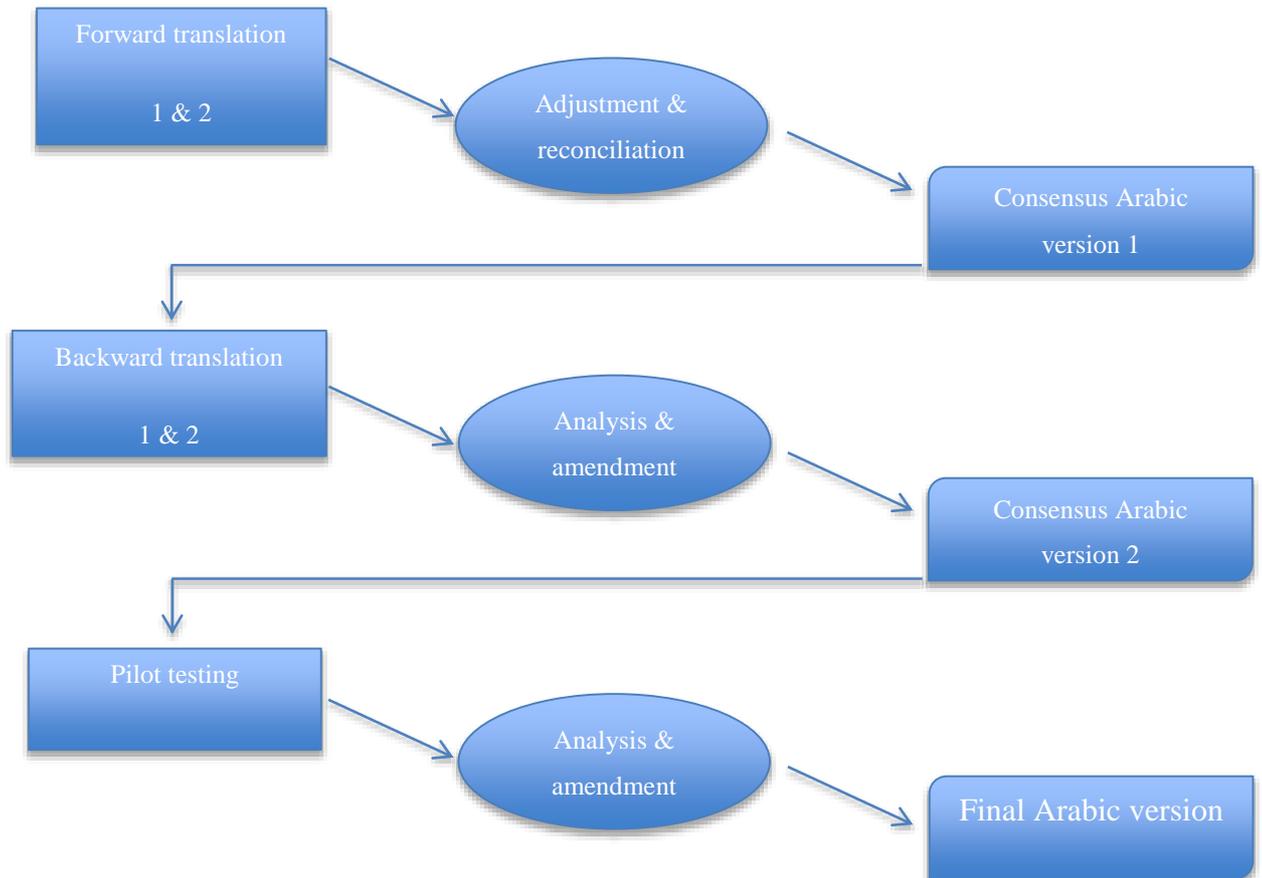


Figure 3-5 Linguistic validation and translation process of the developed OHRQoL questionnaire

3.6.2.1 Forward Translation Process

The questionnaire went through “forward” translation by two independent people who were:

- Briefed about the nature and purpose of the work
- Native speakers of Arabic language
- Fluent in English
- Residing in Saudi Arabia (where the study was conducted)

The forward translation followed two specific stages that include:

- 1- Translation stage: each translator produced a translation of the original questionnaire from English into Arabic without mutual consultation.
- 2- Reconciliation stage: because it was difficult for the original translators to meet and establish the consensus version, the principal researcher reviewed the two versions of forward translations, compared them with the original, and established a consensus version.

3.6.2.2 Backward Translation Process

Two people then independently translated the consensus “backward” version from Arabic into English. Because it was difficult to find translators who had English as their mother tongue and were fluent in lay Arabic, the translation was performed by:

- One person fluent in both languages, with experience in the field of oral health-related quality of life, and living in Saudi Arabia.
- Another person who is fluent in both languages as well and had some experience of the backward translation process, living in Saudi Arabia.

- To perform this step of linguistic validation, both translators had no knowledge of the original (developed) English version of the questionnaire, and were not involved in the forward translation stage.

The backward translation passed through these stages:

- 1- Translation: translators worked independently on translating the Arabic version to English, which reflected it as closely as possible. The translators had no contact with each other, and had no knowledge of the original English text.
- 2- Following the same procedure used for the forward translation process, both translated versions were further reconciled by the principal researcher into a common consensus backward translated version.
- 3- Analysis stage, in which the principle researcher compared the consensus backward translation, the original English questionnaire, with the consensus Arabic version. For each item the researcher analysed the backward translation and determined whether it appropriately reflected the consensus Arabic language version. Any discrepancies between the consensus backward translation and the original questionnaire were carefully examined. There was only one component of the questionnaire that was changed in terms of wording compared to the original version. The term “teased” used in the original questionnaire has been translated to “harassed, bullied, or called names” in the backward translation.
- 4- Review and discussion with other research members (supervisors) stage: at this step, the backward translated version was carefully reviewed with another member of the research team (RGW) to determine whether it accurately reflected the original questionnaire, and to highlight possible discrepancies of the original questionnaire. At this stage, we discussed the concept of teasing, and decided to use both “teased/bullied” in the final version.

3.6.3 Clinical examination

Using the WHO methods (1997), the clinical examination was carried out at day-care centers, and was conducted in a regular chair, under normal daylight conditions, using disposable instruments (mouth mirror and probe). No radiographs were taken. The examination included (Appendix 12):

1- Caries level

Using DMFT/dmft index (decayed, missing and filled surface) (Klein et al 1938) and PUFA index (pulpal involvement, ulceration, fistula and abscess) (Monse et al 2010).

2- Plaque index (PII): using Silness and Løe index (1964)

Measurement of plaque index was used on selected teeth (index teeth) with no substitution for any missing tooth. Partially erupted teeth, retained root, and third molars were excluded. All surfaces (mesial, distal, buccal, and lingual) were measured.

3- Gingival index (GI): using Løe and Silness index (1963)

Using a blunt probe, gingival index was measured on selected teeth (index teeth) with no substitution for any missing tooth. Partially erupted teeth, retained root, and third molars were excluded. All surfaces (mesial, distal, buccal, and lingual) were measured.

4- Malocclusion

The clinical examination recorded aspects of overbite, overjet, and crossbite. The criteria for the occlusion diagnoses were based on studies by the World Health Organization (WHO, 1997). When at least one condition described below was diagnosed, the subject was classified as exhibiting malocclusion stemming from a variation in vertical or transversal occlusion, see Table 3.5.

Table 3-5 Criteria used for the diagnosis of malocclusion

Occlusion criteria	Description
Over-jet	<p>Protrusion: Incisal edge of maxillary incisor more prominent toward the vestibular face of the corresponding mandibular incisor (over 3 mm) (excessive over-jet)</p> <p>Anterior cross-bite: Mandibular incisors in front of maxillary incisors (negative over-jet)</p> <p>Absent: Anterior open bite, edge-to-edge bite, or absence of anterior teeth</p>
Over-bite	<p>Deep overbite: Maxillary teeth cover more than 3 mm of the vestibular surface of the mandibular teeth (excessive overbite)</p> <p>Anterior open bite: No contact between maxillary and mandibular anterior teeth (absent over-jet)</p> <p>Edge-to-edge bite: Incisal surfaces of maxillary teeth touch the incisal surfaces of mandibular teeth (no overbite)</p> <p>Absent: Anterior cross-bite or absence of anterior teeth</p>
Posterior cross-bite	<p>Posterior teeth of the maxillary arch are displaced to the palatine region in relation to the mandibular teeth either unilaterally or bilaterally</p>

5- Other oral health conditions

Other oral health conditions were also assessed such as protrusion of the tongue at time of examination. This was assessed visually as the child walked into the examination room. Dribbling at time of examination was also recorded as present or absent. Presence or absence of visible untreated missing and/or decayed anterior teeth was also recorded, and this was recorded because a study showed that schoolchildren with untreated fractured anterior teeth experienced a higher socio-dental impact on their daily living than children with no traumatic dental injury (Ilma de Souza Cortes et al., 2002).

A copy of the summary of clinical examination's findings of each child was sent to

his/her family to take the necessary action, pointing that this is only a simple epidemiological examination and findings could change if proper clinical examinations including radiographs were conducted (Appendix 13).

3.6.4 Inclusion and Exclusion criteria

Inclusion

1. Confirmed diagnosis of Down Syndrome
2. 12-18 years old children
3. Children/adolescents attending a Down Syndrome day-care centres
4. Children whose parents agreed to participate in the study

Exclusion

1. Children/adolescents with multiple disabilities
2. Children/adolescents with severe form of intellectual disabilities
3. Children/adolescents with complicated medical conditions or taking medications (such as current cardiac problems, severe upper respiratory tract infection, or under the use of antibiotic medication)

3.6.5 Sample size

Researchers recommend that for a validation study, convenience samples are sufficient with a sample size of 50 to 200 participants (Stewart et al., 1992). Therefore, 100 participants were planned for the main data collection, and in collaboration with Down Syndrome centres, all mothers of children 12-18 years old who are attending/have attended Down Syndrome centres were invited to take part in the study.

3.6.6 Fieldwork of phase two

As shown in table 3.6, fieldwork included preparing the materials (including printing material and clinical examination instruments), contacting the centres and doing follow-up work, and collecting data took around four months. Participants were selected from different centres/ schools, and clinical examinations as well as interview-based data collection were conducted in different settings, Table 3.7.

Table 3-6 Phase Two data collection timeline

Month Task	September 2013				October 2013				November 2013				December 2013			
	1	2	3	4*	1	2	3	4**	1	2	3	4	1	2	3	4
Preparing materials & instruments																
Preparation & centres' follow-up																
Pilot stage																
Analysis of pilot stage																
Main data collection																

* National Day 23/09/2013 (schools closed)

** Al-Adha Eid break 10/10/2013 till 21/10/2013 (schools closed)

Table 3-7 Outline of Phase Two data collection settings

Agencies contacted	<p>- Ministry of Social Affairs</p> <ol style="list-style-type: none"> 1. Down Syndrome Charitable Association (DSCA) 2. SAUT Down Syndrome society (AlNahdah charitable society) 3. National Centre for Early Intervention (DS) 4. Saudi Centre for Down Syndrome 5. Al-Wafa Rehabilitation Centre 6. Al-Khatwa Society for special needs <p>- Ministry of Education</p> <ol style="list-style-type: none"> 1. Schools with inclusive education system (boys & girls) 2. Institutes of intellectual rehabilitation (2 for boys & 2 for girls) <p>- Several Governmental Hospitals</p> <ol style="list-style-type: none"> 1. Dental College at King Saud University 2. King Saud Medical City 3. Al-Yamama Hospital (Ministry of Health) 4. Prince Sultan Military Medical City 5. National Guard Hospital 6. King Faisal Specialist Hospital and Research Centre
Agencies approved	<p>- Ministry of Social Affairs</p> <ol style="list-style-type: none"> 1. Down Syndrome Charitable Association (DSCA) 2. SAUT Down Syndrome society (AlNahdah charitable society) 3. National Centre for Early Intervention (DS) 4. Saudi Centre for Down Syndrome 5. Al-Wafa Rehabilitation Centre 6. Al-Khatwa Society for special needs <p>- Ministry of Education</p> <ol style="list-style-type: none"> 1. Schools with inclusive education system (boys & girls) 2. Institutes of intellectual rehabilitation (girls) <p>- Several Governmental Hospitals</p> <ol style="list-style-type: none"> 1. Dental College at King Saud University 2. Al-Yamama Hospital (Ministry of Health)
Settings of data collection	<p>All clinical and interview-based data were collected in the centres approved to take part. Families from inclusive education schools (mainly boys) were invited to the Dental College at King Saud University, or Al-Yamama Hospital according to their preference.</p>

3.6.7 Pilot testing stage

Prior to the main data collection, a pilot study was conducted to assess the appropriateness, clarity, and feasibility of the administration of the developed questionnaire under fieldwork conditions. Ten interviews with mothers of children (12-18 years old) with Down Syndrome were conducted, at SAUT Down Syndrome centre, Riyadh city of Saudi Arabia.

After each interview, mothers were informally asked to fill in a feedback form to evaluate the questionnaire, and assess its appropriateness (Appendix 14). They were asked about the length of the questionnaire, the clarity of the wording, and if there was anything relevant that was not included in the questionnaire. No major problems were faced or pointed out by participants, and therefore no modifications were required.

The pilot study confirmed the feasibility of the planned methodology; it also gave an indication of the time required to complete the questionnaire using a structured interview method.

3.6.8 Main data collection

With the help of Down Syndrome centres' administration staff (which are all under the supervision of Ministry of Social Affairs), the information sheets and consent forms (Appendix 9) were sent to all mothers of 12-18 years old children attending/have attended these institutions (excluding those who participated at phase one and pilot stage of phase two), inviting them and their children to take part in the study. The number of participants from the four Down syndrome centres was small, and to increase the sample size and reach the proposed target population (around 100 participants), other bodies were included, such as the Ministry of Education (to approach children with Down Syndrome attending schools with inclusive education system), and some large governmental hospitals (in order to get access to their Down

Syndrome clinics). The Dental College at King Saud University was also contacted to get access to the list of patients with Down Syndrome attending their paediatric dentistry clinics.

A separate dental clinic was provided by the Dental College (King Saud University), and Al-Yamama Hospital (Ministry of Health). These were booked twice a week for three months to conduct the interviews and clinical examinations.

3.6.9 Test-retest reliability

After their initial participation, mothers were informally asked if they were willing to take part in another follow-up interview and if they would allow their children to be examined again within 2-3 weeks of their initial participation. The researcher then repeated the work (structured interviews and clinical examinations) on 10% of the sample (10 children & their mothers).

3.6.10 Intra-examiner reliability

Intra-examiner reliability was assessed from the repeated work on 10% of the study sample. Results showed a high level of agreement with Intra-class Correlation Coefficient (ICC) 0.91 or more on the repeated measures. This is mainly because the same examiner (AJ) collected the data within a short period of time between the initial and re-test examination (2-3 weeks). Some of the disagreements were unavoidable; for example in the plaque index, in which some of the participants in the initial records were recorded to have moderate accumulation of plaque but in the re-test the plaque index was shown to have improved slightly. This might have occurred as an immediate consequence of increased oral health awareness on part of the participants.

3.6.11 Data analysis

Statistical analysis was carried out using STATA statistical package version 13.

3.6.12 Descriptive analysis

Frequency distribution of demographic data including age and sex of children examined, as well as marital status, educational levels, and employment status of their mothers were analysed. Clinical data analysis included frequency distributions of some oral health status (protruded tongue, dribbling, and presence of unrestored decayed or missing anterior teeth), as well as means, standard deviations, and quartiles of decayed, pulpally involved, and filled teeth in primary and permanent dentition. Data on plaque index and gingival index along with malocclusion status (over-jet, overbite, and posterior cross-bite) were also presented. Subjective health and QoL data analysis included the frequency distributions of some general health conditions and oral health status, self-assessed general and oral health, and child and family's OHRQoL data (prevalence and severity).

3.6.13 Psychometric testing of the developed instrument

The psychometric properties of the newly developed OHRQoL questionnaire, OH-QOLADS, were assessed for reliability and validity. In order for assessments to be sound, they must be free of bias and distortion. Reliability and validity are two concepts that are important for defining and measuring bias and distortion. Table 3.8 summarises the definitions of reliability and validity tests used for psychometric testing of the developed questionnaire, OH-QOLADS.

Table 3-8 Definitions on of reliability and validity tests (Streiner et al., 2015)

Reliability	
Test reliability refers to the degree to which a test is consistent and stable in measuring what it is intended to measure	
External reliability/Test-retest / Reproducibility	A measure of reliability obtained by administering the same test twice over a period of time to a group of individuals. The scores from Time 1 and Time 2 are then correlated in order to evaluate the test for stability over time.
Internal consistency reliability	A measure of reliability used to evaluate the degree to which different test items that probe the same construct produce similar results.
Validity	
Test validity refers to the degree to which the test actually measures what it claims to measure	
Face Validity	Indicates whether the measure appears to be assessing the intended construct under study.
Content validity	Assess whether the instrument sample all relevant or important content or domains. Face and content validities are technical description of the judgment of experts of whether the scale appears appropriate for the intended purpose.
Criterion validity	Is used to correlates test results with another criterion of interest. For example, correlate the findings of developed measure to already existing measure.
Construct validity	Is used to assess the attribute we are measuring to some other attribute by a hypothesis. For example, we hypothesized that the OHRQoL is related to child's oral health and therefore results should reflect that OH-QOLADS associated with perceived oral health.
Discriminant validity	Is used to assess the ability of developed questionnaire to discriminate between different groups. For example, the questionnaire should be able to differentiate between individuals with and without dental caries.
Convergent validity	Is used to assess how closely the new measure is related to other variables and other measures of the same construct to which it should be related. For example if our theory suggest that child's oral health and OHRQoL are supposed to be related to family's QoL, then scores of family's OH-QOLADS should correlate with child's global rating of QoL and child's OH-QOLADS.

Assessment of this questionnaire considered the COnsensus-based Standards for the selection of health Measurement Instruments (COSMIN) recommendations in assessing any health-related patient-reported outcome measures (HR-PRO). The COSMIN initiative have produced a checklist to evaluate the methodological quality of studies on measurement properties, and came out with a list of properties that can be used to assess the quality of health outcome measures (Mokkink et al., 2010), presented in the figure 3.6 below.



Figure 3-6 COSMIN taxonomy of relationships of measurement properties (Mokkink et al., 2010)

The first step to produce evidence that the developed instrument/questionnaire is of any value, it is necessary to gather evidence that the developed questionnaire is measuring something in a reproducible manner, this is known as reliability and is expressed as a number between 0 and 1; with 0 indicating no reliability, and 1 indicating perfect reliability. There are many number of ways in which reliability can be obtained (Streiner et al., 2015). In our analysis reproducibility was assessed with test-retest reliability (section 3.6.9).

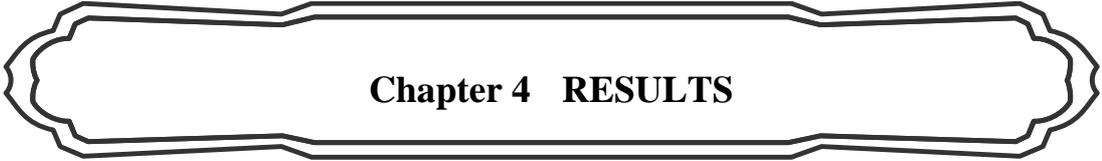
To assess **external/test-retest reliability**, the developed questionnaire was administered on two occasions (with an interval of three weeks between them) to a convenience subsample of mothers. Test-retest reliability was assessed by Kappa coefficient, using data from respondents who reported no dental visits or change in their child's oral health status during the 2-3 weeks interval between initial and follow-up assessments. **Internal consistency reliability** is based on the single administration of the questionnaire and was assessed by Cronbach's alpha, item-total and inter-item correlations.

Content and face validity were based on the findings of the interviews with mothers (phase one) and the literature review. The idea of assessing instrument validity is to make sure that it is measuring what it is intended to measure. Since there is no 'gold standard' measure of OHRQoL that can be used to test any newly developed measure based on it, it is difficult to assess the criterion validity of our developed measure, OH-QOLADS. Therefore, validation process relied mainly on **construct validity** that was assessed by linking the attribute we were measuring, OHRQoL, to some other related attributes including: a) mothers' perceptions of the child's perceived general health, b) perceived oral health of the child, and c) the overall extent that oral problems affected child's QoL. These were assessed through both Kruskal-Wallis tests and Spearman's rank correlation coefficients. Construct validity of Family's QoL section of the developed questionnaire was also assessed using Kruskal-Wallis tests and Spearman's rank correlations by comparing family QoL to: a) mothers' perceptions of the child's perceived general health; b) perceived oral health; and c) overall extent that oral problems affected family QoL.

Discriminant validity was assessed by testing the following hypothesis: differences in child OHRQoL would exist between the patient and healthy groups. We used a variety of patient groups with different oral health problems (presence of carious teeth, pulpal involved teeth, malocclusion, plaque index, gingival index, bad breath, and toothache) with the assumption being that children with the oral health condition would have higher OHRQoL and therefore poorer OHRQoL than their healthy counterparts. The discriminant validity was assessed using Mann-Whitney test.

Convergent validity was evaluated based on Spearman's rank correlations. The correlations used family QoL with both global child's QoL rating and child OHRQoL. We hypothesized that family QoL would be significantly correlated to global child QoL rating. We also hypothesized that the child and family sections of the OHRQoL would be significantly correlated because parents' assessment of their child's oral health was likely to be closely related to parental perceptions of the effect of their child's oral health on their family.

In summary, this chapter presented the methodology used to develop and test the OH-QOLADS instrument that aims at assessing the impacts of children with Down Syndrome oral health on them and their families' QoL. The study utilized a mixed method approach of qualitative interviews of mothers of children with Down Syndrome to develop the instrument, and then validate the OH-QOLADS on another sample. The validation analysis was based on the assessment of psychometric properties that are widely used on OHRQoL research and recommended by COSMIN. The next chapter presents the results of the whole study including both phases.

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Chapter 4 RESULTS

4.1 Introduction

The results chapter presents findings of both phases of the study in two separate sections. The first section presents the results of the phase one exploratory study of OHRQoL among children/adolescents with Down Syndrome from their mothers' perspective. The study was qualitative in nature involving interviews with a purposive sample of 20 mothers who were direct carers of their children with Down Syndrome. The second section presents the findings of phase two, which focused on the validation of the questionnaire assessing the OHRQL of children and adolescents with Down Syndrome.

4.2 Phase One

4.2.1 Introduction

After transcribing them verbatim, all interviews were analysed using thematic analysis, and results were classified broadly into five sections. The first segment summarised mothers' views on their children's general health while the second presented briefly their experiences of having a child with disability. The third section of this analysis showed the mothers' opinions regarding their children's oral health. After that, mothers' opinions of how the oral health of their children impacted on different aspects of their lives were presented. Finally, the findings on the mothers' opinions of how oral health conditions of their children impacted on the family QoL were summarized.

4.2.2 Sample characteristics

The sample consisted of 20 mothers of 12-18 years-old (boys & girls) children with Down Syndrome. The majority of the mothers interviewed had either school level educational qualifications or no education at all. The age of mothers interviewed ranged from 40-63 years old, and the majority gave birth to their children with Down Syndrome at an age of 36 years old or older (Table 4.1).

Table 4-1. Profile matrix of the study sample

Case	ID	Mother's age	Mother's level of education	Child's gender	Child's age
1	DB1	36+	No qualification	Boy	14
2	DG2	36+	Post school	Girl	12
3	DG3	36+	No qualification	Girl	16
4	*DGG4	20-35	School or equivalent	2 Girls	12 & 16
5	DG5	20-35	Post school	Girl	13
6	DG6	36+	No qualification	Girl	15
7	DG7	36+	School or equivalent	Girl	16
8	DG8	36+	Post school	Girl	17
9	DG9	20-35	No qualification	Girl	14
10	DG10	20-35	No qualification	Girl	16
11	DB11	36+	School or equivalent	Boy	16
12	DB12	36+	School or equivalent	Boy	13
13	DG13	36+	Post school	Girl	12
14	DB14	20-35	School or equivalent	Boy	18
15	**DG15	20-35	No qualification	2 Girls	18
16	SG1	36+	No qualification	Girl	16
17	SB2	20-35	Post school	Boy	16
18	***SGB3	36+	School or equivalent	Girl & Boy	22 & 17
19	SB4	20-35	School or equivalent	Boy	12
20	SB5	36+	No qualification	Boy	13

* Mother of two sisters with DS

** Mother of twin with DS

*** Mother of sister and brother with DS

4.2.3 Child's general health

This section summarizes the child's general health from their mothers' perspective. The mothers were asked about the overall health status of their children, and then if their children had/have any specific health conditions such as heart problems, endocrine system conditions (e.g. thyroid gland malfunction or diabetes), and sensory conditions (e.g. vision or hearing problems).

The child's general health was a key priority of all mothers. The majority of mothers felt that their children were in good general health. However, some of the children previously suffered heart problems when they were young, and a few had had heart surgery. However, they all had no current heart conditions.

For example, when a mother was asked about her child's heart conditions, she reported that:

‘Thank God, He had a septal defect that... but it healed on its own’
(DB12)

Results also showed that some of the children had thyroid gland problems that were controlled by thyroxin replacement therapy.

‘Now he's only being treated for thyroid malfunction’ (SB5)

Problems on the sensory system were reported when some mothers stated vision problems and the use of eyeglasses, in addition to some problems related to hearing especially recurrent ear infections. In this regards a mother of 13 years old boy said:

‘Thanks God, his Health is fine, only the eyes aren't fine’ (DB12)

Almost all children had good access to health care services (in governmental hospitals mainly through their schools), and had regular medical checks. However, some mothers stopped visiting the governmental hospitals for a variety of reasons including: long waiting lists; looking for better quality of care; transportation issues; avoiding people's comments and behaviors while waiting for their turn at waiting areas especially when the waiting times were lengthy.

Many mothers reported their concerns about their children's weight. They stated that especially at this stage (teenage years), it was difficult to control the child's appetite, and mainly their desire to eat sweets and high calories food. A mother said that even when she tried to stop her daughter, she did not succeed because her daughter had grown up and she knew where and how to reach such foods, she said:

‘She likes sweets, but we don't let her. She has a good appetite for everything’ (SG1)

Another mother of a 14 years old girl said:

‘She's fat, she likes carbohydrates, she loves burgers and junk food very much, and she just loves candy so much. Whenever she finds them she doesn't stop eating’ (DG9)

Knowing that children with Down Syndrome are prone to increased body weight, some of the participants expressed concerns about maintaining adequate physical activities that played an important role in weight gain, and therefore general health. In this regard, for example a mother of 16 years old girl reported:

‘Her weight has increased slightly, because she does not move. She does not stand and move.... Now that she has grown up a bit,.... She does not move as much as when she was young. She sits in front of the TV all the time’ (SG1)

In summary, the results showed that the mothers considered their children to have generally good health. Nonetheless, a few mothers reported health concerns such as sensory problems and weight issues.

4.2.4 Experience of having child with disability

This section presents results on mothers' experiences of having a child with a disability. A majority of mothers reported that they knew about the presence of their children's condition either at the day of delivery or a couple of days later. However, some did not know about it for a couple of months or longer. Mothers' reports of the

diagnosis of the disability showed that their immediate responses included denial, sadness, and depression. Discussion revealed that these responses reduced with time and amongst some mothers were completely resolved. In addition, some mothers reported that they went to more than one doctor to make sure of their child's condition.

When mothers were asked about how the presence of child with Down Syndrome within the family changed their lives, in general mothers were happy with their children, and they viewed it as a good, yet not easy experience. Below are some quotes reflects the mothers' feelings of having a child with Down Syndrome:

'I swear he was the reason behind everything good, the reason behind guidance, tranquillity, quietness, stability, religion, all thanks and praises to God' (DB14)

'I swear it added nothing except more love and care to him' (DB11)

'He grows on you. He is a sweet heart. Everybody loves him, but sometimes you feel like he is a responsibility, you fear for him' (SB2)

Almost all the mothers experienced some difficulties and struggles when their children were very young, most of them did not know about this disability before the birth, and did not know how to care for their children. It was also difficult for them to find the relevant information, and many mothers struggled to find informative books.

'It was a great shock!... I mean we hadn't...we hadn't... We hadn't heard about that... before! We didn't have the background...' (SG1)

Mothers also talked about the society and how unfair, unhelpful people were, in addition to the fact that some mothers became isolated from the surrounding community to avoid their unfair judgments and endless questions.

Some said that at the time they needed support and education about the disability, the whole community was also in need of education about disability. They felt that the

problem was always bigger when they went outside with their children because of the attitude and reaction of people which was difficult not only for mothers, but for child and siblings too. Social stigma experienced by mothers created a main source of struggles and increased the burden on parents especially when their child was very young. Mothers' reports reflect how families started to adjust to the presence of disability with time, although the negative influence of people attitudes and the feelings of stigmatization remained in some cases. Below are some of mother's quotes regarding the people's attitude in regards to disability:

'When we go to a family reunion ...someone asks the other kids loudly not to annoy (child) because she is sick, she is poor, don't hit her. They also give her two toys or two gifts..... All these destroy her feelings' (DG2)

'Sometimes I didn't go, sometimes I left her home. People gave her a terrible look' (DG9)

However, defensive behaviour and denial of the presence of disability was evident among some participants. For example a mother of a 15 year old girl insisted that her daughter had nothing different than other children, and she totally denied the fact that she had a daughter with Down Syndrome, despite the fact that she was bringing her daughter to a special Down Syndrome school on a daily bases.

'As I have told you before, thank God, (child's) life is as ordinary as any of my other children's lives. -----, and you can write these words down, as I don't have anything else to add. Her situation is normal, and we treat her as a normal person living with us... she has nothing different than her siblings' (DG6)

Findings also indicated that the presence of child with disability caused conflict or tension among family members. When participants were asked about the strategy they used to deal with their children, the majority of them said being patient, tolerant, and having a strong belief in God was the approach that helped them a lot. To get over the external environment and other people's negative attitudes and questions, some mothers tried to avoid going out and mixing with others, especially at the

beginning when they first knew about the disability and until they became strong enough to face negative attitudes, and be able to adapt to the situation.

The results of the mothers' interviews revealed that having a child with a disability was not an easy experience although some reported it as a gift from "God" that filled their lives with happiness and tranquillity mainly when their children got older. Mothers' reports outlined the important role of other factors such as lack of awareness about such disabilities and professional guidance that they needed. The issue of environmental factors such as negative social attitudes was also reported giving the importance of such factors in the experience of having a child with disability.

4.2.5 Child's oral health

Findings of the oral health status from mothers' reports were grouped into: an overall evaluation of the child's oral health, and their evaluation of specific oral health conditions such as tooth decay or gum disease, bad breath, tongue condition, and speech difficulties.

When mothers were initially asked about the oral health of their children, a majority reported that their children's teeth/mouth were healthy, although some mothers reported experience of dental caries. Results also showed that some children received dental treatment when families were advised to visit a dental clinic as a result of the schools' physician advice or when the child started complaining from toothache. While, a few mothers were aware of the presence of dental problems, it appeared that experiencing dental caries was not seen as a problem for the majority to be concerned about. This could be a result of many reasons such as the perception that dental problems are not as important as the child's general health especially if it compared to heart conditions for example; or could be a result of lack of awareness of how oral health problems such as caries can lead to severe pain or infections.

Some mothers reported that they had never been to dentist before because they thought their children's teeth had no problems and they have never complained. Results also indicated the problem of halitosis or bad breath, and the presence of active gum disease that was reported by mothers as gum bleeding with tooth brushing. Below are some quotes from mothers' interviews regarding their children oral health:

'Her only problem is teeth decay' (DG13)

'She has no gum disease, but there are holes. I don't know how many teeth, as she has got one here and one there' (DG6)

'He had it for a long time (bad odour), when he wakes up there's a horrible abnormal smell' (DB12)

'But her gum hurts.... Here.... I try to brush it for her, but she refuses because of the pain and prefers to do it herself, sometimes with her hands' (DGG4)

Results also showed some concern about orthodontic treatment as one participant asked for treatment for her daughter since she was concerned about the appearance of her teeth, and also said that the daughter asked for that.

'She (daughter) cries there (dental clinic)..... I want to take her to the doctor to have orthodontics. I want to take her, and she says I want to do orthodontic treatment' (DG6)

Some mothers did report the problem of dribbling and/or their child having a relatively bigger, or protruded tongue when they were younger but these problems reduced a lot, and were almost resolved in some cases with time as a result of the early interventions received from Down Syndrome schools.

'(Tongue) inside her mouth..... no saliva dripping..... When she was young, she used to infuse balloons and blow soap bubbles and chew gums...she had very good training' (DG2)

The main oral health problem that was reported by mothers was difficulty speaking or pronouncing words. They felt that although, there was an improvement in the clarity of the child's speech with time, the improvement in the child's ability to speak and learn new words was very slow or limited. A mother of a 16 years old boy said:

‘He understands speech, but he can’t speak very well. The people..... who come from outside, aren't in close contact with him, may not understand him and what he is trying to say’ (DB11)

4.2.6 Impacts of child's oral health on his/her quality of life

This section of the interviews investigated the mothers' opinions on how their children's oral health affected different aspects of their lives such as their daily activities or their social relationships. As interviews indicated, it appeared that it was difficult for mothers to recognize the potential impact of the child's oral health on different aspects of his/her life. When they were initially asked, generally, the main answer was that there was no impact other than complaining from pain if they experienced dental problem such as caries. However, through gentle probing and further assessment of their views about other potential impacts that their children's oral health might cause, the mothers revealed a relatively wide range of impacts on various aspects of the child's life (Figure 4.1).

4.2.6.1 Physiological pain

Although the majority of respondents reported good oral health status, they mentioned that their children suffered from pain when they experienced dental caries and continued to complain from pain till treated. The severity of pain experienced by the child varied according to the mothers' reports.

‘If she feels pain, you see that her figure has changed and she is not at ease. She weeps sometimes, and so you know that she is not feeling well...’ (SG1)

‘She used to avoid laughing due to her toothache’ (DG2)

‘Yes I gave her analgesics all the time (because of toothache)’ (DG13)

From mothers’ reports, it appears that pain as a result of dental caries can lead to other impacts such as functional limitation or emotional impacts (i.e. avoid laughing) this can be seen in the following paragraphs.

4.2.6.2 Impacts on daily activities

Experience of dental pain as a result of dental problems such as caries also affected children’s daily activities like doing their schools’ homework, their sleeping pattern, and their eating habits. Playing with their friends and siblings was also affected.

‘Yes. He stops eating and yells “my teeth, my teeth’ (DB12)

‘Yes, (if they experience toothache),,,, they did not play with their friends’ (DGG15)

‘Yes sure... we went to the hospital when she had pain, which tired me and delayed her from school’ (DG5)

‘He doesn’t play. He gets very quiet and I know,,, he kept biting his finger to go to sleep’ (SB4)

4.2.6.3 Emotional impacts

In most cases, the main problem that impacted negatively on the child’s psychology was difficulty speaking or having unclear speech. Mothers believed that this difficulty caused their children to become depressed and angry if they were not understood. Because of this problem children became more shy and avoided talking in front of strangers to avoid being judged accordingly.

‘The source of her suffering is that she wants to speak fluently.....
She is shy because she is unable to speak well’ (DG2)

‘Yeah and he hides his shyness, he feels that he is inferior to other people, but I usually say you are old you are man you are... he feels embarrassed and he becomes a little introvert’ (DB14)

Experience of dental pain also had an impact on the child’s emotions. Some mothers reported that their children used to cry, stopped laughing, and became angry if they had dental pain. They also recognized the improvement on the child’s mood after appropriate dental treatment, and how they became better when the oral symptoms had been removed.

‘Yes (if she has toothache)... she gets nervous so quickly’ (DG3)

‘Her mood changes and she cries’ (DG7)

‘After 2 days of the surgery (dental treatment), after recovery, even before she finished the antibiotic, she became much..... much better, laughs, moves and write on the board’ (DG2)

4.2.6.4 Social impacts

Experience of dental pain by some children affected their social relationships from mothers’ opinions as some reported that their children used to stop playing and interacting with their friends if they were in pain. The problem of unclear speech reported by mothers resulted in many social impacts as well. It affected the children’s ability to make friendships with others, this can be seen when mothers thought that their children tried to select their friends carefully and they tried to become friends only with their relatives, who knew them very well and are able to understand their talks, or made new friendships with an older children who were able to understand them in order to avoid being bullied by youngsters. For example, a mother of 12 years old girl reported that:

‘Yes, there are some children who repeat her words and that causes her pain, for example, I say let’s go to (name) her cousin,,,,,,,,,,,,, she refuses,

her friendships are with those older than her, college girls, high school girls, she loves them, when they come she communicates, gets her laptop, iPad and use them. Those younger than her no, because they criticize her, (saying) you can't count, you say 6 in a wrong way, you don't know' (DG2)

4.2.6.5 Behavioral impact

Oral health conditions, especially problems with speaking, were reported by a participant to cause unfavourable behavioural consequences such as stubbornness, mainly if the child wanted to say something and found the person in front of him/her did not understand what he/she was trying to say.

'Yes it does (affect her), it causes a stubborn problem, if she said a word she doesn't change it ever, the consultant who I used to take her to said this is a demonstration that you don't understand her, she is stubborn to attract your attention,,, we understand that, but yet it is a problem'
(DG2)

From this analysis, it seems that children's oral health has various impacts on different aspects of their lives according to their mothers' reports. In addition, these different impacts of child's oral health appear to operate at different levels and may interact with each other.

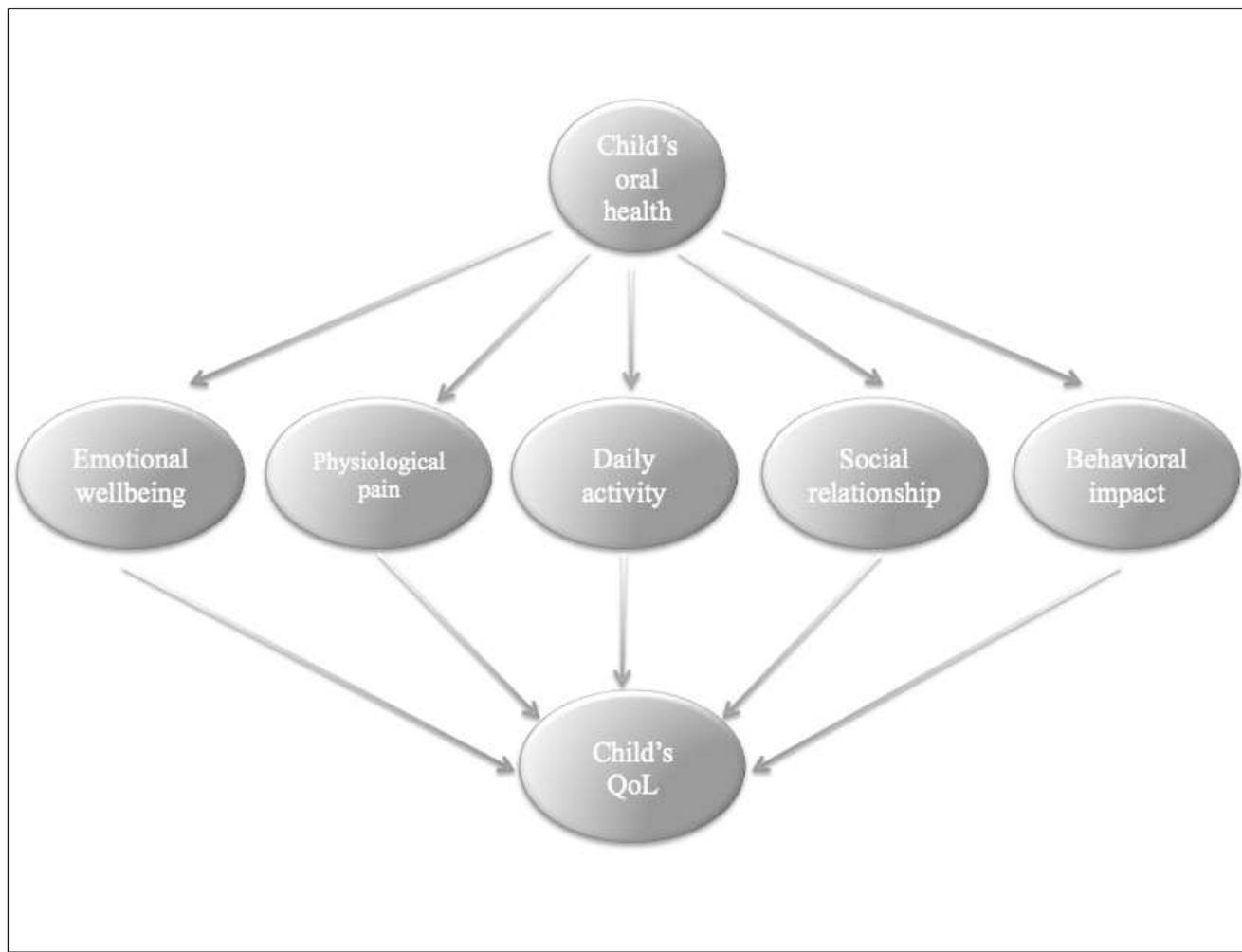


Figure 4-1. Impacts of child's oral health on his/her QoL

4.2.7 Influences of child's oral health on family's quality of life

The last part of the topic guide explored in the interviews how the child's oral health impacted on the broader family's QoL. The general question used to explore mothers' views in this regard was 'How do you think you, your partner, or siblings have been affected by his/her oral health conditions?'. When this question was asked, almost all mothers were not able to answer it easily. Therefore, the question was broken down into three distinct sections and was worded more simply: how do you think your child's oral health affected the family's... a) emotions, b) activities, and c) conflicts (Figure 4.2).

4.2.7.1 Family emotions

From the interviews, it appears that the mothers were more emotionally affected by their children's' situation than any other family member. When their children had dental pain, mothers became very emotionally affected. They reported mood changes, they felt that the pain was hurting them, as well as their child, they felt irritated, angry, depressed, and preferred to be socially isolated until their child recovered and felt better. Some mothers also reported the feeling of self-blame and guilt if their child experienced dental pain, and they felt they did not provide enough care to prevent their child from suffering.

'Yeah, I get upset and people look at him, but I don't care' (DB14)

'I blame myself for neglecting her and not brushing her teeth. I say to myself that I must be doing something wrong' (DG9)

'We all got worried a bit' ... 'I feel the pain like it was in my teeth'
(SB5)

4.2.7.2 Family activities

Many mothers reported that they changed their planned activities if their child got some sort of pain including toothache. Their sleeping patterns were disturbed as well

(e.g., shift to sleep with the child, having sleepless nights), and they tried to avoid going out with families and friends if their child was in pain. Mothers also reported the fact that child's caring (in case suffered from toothache or any source of pain) took time and resulted in neglecting themselves and other family members. For example, some mothers reported that if the child felt pain (i.e. toothache) they spend time with the child ignoring other family members such as siblings, and also some mothers reported that they cared less than usual for themselves until they made sure that the child pain was alleviated.

'Yeah, I change it sometimes (scheduled activity), for example, if his father is going outside he goes with him, but if his father was busy I take his role.....' (DB14)

'We cancel it,, if something could happen I give her analgesics, but what if the pain re-occur and I am not with her' (DG13)

'Yes, to an extent I cancel an important meeting' (DG2)

4.2.7.3 Family conflict

Asking mothers if their children's oral health caused any kind of family conflicts showed that oral health had no strong or evident impact on this, except mothers who reported that arguing with other family member especially the father or older brother might happen if they needed to take the child to a dental appointment and no one else was available to take them there.

'Not really, no, but yes sometimes if I need to take her to the clinic or something like that' (SG1)

Some answers revealed that the child's oral health (especially experience of pain/toothache) might cause some arguments with other siblings; and they started feeling jealous as the child's with Down Syndrome occupied the mother until the dental pain/problems ceased.

'Yes,, they get jealous of her... but I tell them she's ill... they say I only care about her' (DG9)

So analysis of data on this regards revealed that child's oral health have an evident impacts on family QoL especially on activities and emotions domains. It appeared that mothers were the most affected family members and this might be due to the nature of mother relationship with her child or also as a result of interviewing mothers only but no other family members which might masks an important impacts of the child's oral health on his/her family's QoL.

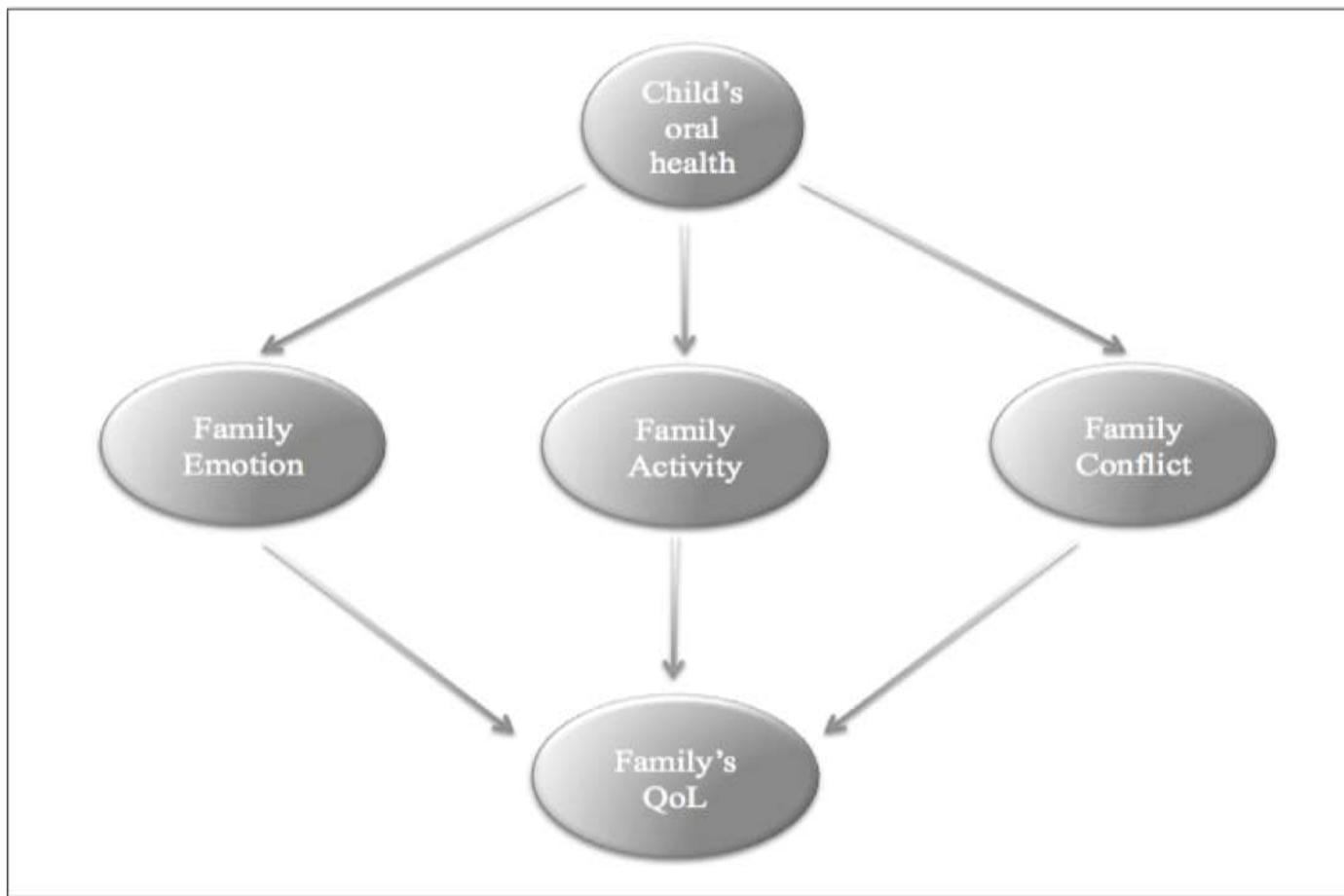


Figure 4-2 Impacts of child's oral health on family's QoL

From the data presented above, the child's oral health appeared to have considerable impacts on the child with Down Syndrome and his/her family's QoL. These findings were used to develop the OHRQoL questionnaire that was tested and validated at phase-two of this study. The next section presents findings on the developed questionnaire and its psychometric properties tested at phase two.

4.3 Phase Two

4.3.1 Introduction

This section presents the results from phase two of the study. It starts with the process of questionnaire development. Then findings of the validation study are presented into three sections: **A)** Pilot testing stage that reports the findings of the pilot testing of the developed questionnaire (OH-QOLADS). This was conducted with 10 mothers and their children in order to assess the appropriateness and feasibility of the planned procedures and developed questionnaire; **B)** The descriptive data analysis provides an overview of the sample with regards to the demographic status of the children, their clinical oral health status, and subjective judgements of their overall oral and general health status. This section also examines the prevalence of oral health impacts on the QoL of both the children and their families, as well as findings of the overall/global rating of oral health impacts on child and family; and **C)** The validation analysis, which presents the main findings of phase two. Both the internal consistency reliability and the external (test-retest) reliability are presented in this section, alongside analysis on testing for validity that includes data on face and content validity, construct and discriminant validity.

A total of 107 mothers and their children with DS participated at phase two of this study. Ten participants (comprising 10 mothers and 10 children) were involved in the pilot stage, and the remaining 97 were included in the main validation study. Details of the sample are fully described in section 4.3.4

4.3.2 Questionnaire development

After analysis of all interviews, items of the child's OHRQoL as well as family's OHRQoL were identified and supplemented by items from previously validated child and family's OHRQoL measures, mainly CPQ (Jokovic et al., 2002) and Child-OIDP (Gherunpong et al., 2004). The developed OHRQoL measure, OH-QOLADS, consisted of 20 items on Child's OHRQoL that covered physiological pain, impacts on daily activities, emotional, and social dimensions of the child's life. While the family's QoL section contained 10 items on the emotional state, daily activities, and conflicts that might occur as a results of the child's oral health status (Table 4.2). The questionnaire contained questions assessing the occurrence and severity of each item (as minor, moderate or severe), and assessed the occurrence of each item in two different time periods (ever happened, and in the last 12 months). Since the questionnaire (OH-QOLADS) was in its development stage, all items that seemed to be related were listed. After discussing the list of items with the research team, some were regrouped as shown in the table below.

Table 4-2 Items of OH-QOLADS

Section	Items
Child's OHRQoL	<p>Physiological pain</p> <p>Impacts on daily activities (Eating, Speaking, Brushing teeth, Sleeping, School work, Playing)</p> <p>Emotional impacts (Crying/ Feeling upset, Stop laughing, Being quiet, Becoming shy/ introvert, Feeling embarrassed, becoming less confident, Being conscious about his/her mouth, Being angry, behaving in stubborn manner)</p> <p>Social impacts (Withdraw him/herself from</p>

	family, Withdraw him/herself from friend, been excluded, been teased/ bullied)
Family's OHRQoL	<p>Emotional impacts (Feeling depressed/ distressed/ upset, feeling of self-blame/ guilt, being worried, feeling angry)</p> <p>Impacts on family's activities (cancelling scheduled activity, affect their job, time off from other family members, affect sleeping pattern)</p> <p>Impacts on family's conflict (Arguing with other family member, other sibling being jealous)</p>

A section about the demographic details of the child and mother was also incorporated into the questionnaire, along with some questions about the mothers' perceptions of their child's general and oral health status. Therefore, the main questionnaire consisted of two sections; first concerning the demographic and general and oral health status of the child, and a second section that included both child and family's OHRQoL assessments (please see appendix 11 for full version of the questionnaire used in the main study data collection).

4.3.3 Pilot testing of the questionnaire

This stage was conducted prior to the main data collection in order to assess the newly developed questionnaire. Ten interviews with mothers of children with Down Syndrome were conducted at one of the participating Down Syndrome schools (SAUT), those mothers were not involved at phase one study and were also excluded from the validation stage. SAUT school randomly contacted number of mothers of 12-18 year-old children with Down Syndrome, explained the study purpose and invited them to take part in the study.

A clinical examination of the children's oral health was also performed to check the feasibility of the planned procedures. Data on the length of the interviews (for answering the questionnaire) were recorded and showed that each interview lasted for about 20-27 minutes.

After each interview, mothers were asked about the length of the interview, the clarity of the wording used, and the comprehensiveness of the questionnaire. They all reported that the questionnaire's length was acceptable. They also thought that the questionnaire was clear, understandable, and there was no need for further clarification or adjustment. When they were asked about the comprehensiveness of the questionnaire, they reported that the questionnaire accounted for all relevant areas, and therefore nothing else should be added.

An oral examination was conducted in daylight using disposable oral examination instruments in a room at the clinic. The examination took around 10-12 minutes to complete. The oral examination was relatively straightforward to conduct, although at times it was challenging to measure the gingival index of some children. Therefore, findings of the pilot stage confirmed the feasibility of the planned methodology, and it also gave an indication of the time required to complete the questionnaire using a structured interview method.

4.3.4 Descriptive data analysis of the main study (validation stage)

4.3.4.1 Demographic characteristics of the study sample

Table 4.3 shows the demographic data of both the children and their mothers who participated in phase two of the study. The sample included a slightly higher proportion of girls, almost 57%, than boys. The ages of the children sampled ranged from 12 to 18 years old, and the average age was 15 years. The average number of siblings among this sample of Saudi Arabian children was 7 brothers and sisters. The majority of children included in this study were the only members in their family with a disability, as only 11% reported that they had another family member with some kind of disability, whether “physical or developmental”.

The average age of mothers was 50 years, the majority of which were married (86%), with only 5% divorced and around 8% widowed. Almost 90% of them were not in employment and only 7% worked in full-time jobs. Around 20% of mothers participated did not receive any education, the majority had school level education (around 60%), and approximately 20% had post-secondary levels of education.

Table 4-3. Socio-demographic characteristics of the study sample

Variable	Number N= 97	Percentage % Or Mean (SD)
<u>Child</u>		
Gender		
Girls	55	56.7
Boys	42	43.3
Age	97	15.2 (2.3)
Number of siblings	97	7.1 (2.6)
Other sibling with disability		
Yes	11	11.34
No	88	88.66
<u>Mother</u>		
Age	97	50.1 (6.3)
Marital status		
Married	84	86.6
Divorced	5	5.2
Widowed	8	8.3
Employment status		
Working full-time	7	7.2
Working part-time	3	3.1
Not working	87	89.7
Education level		
Non-educated	20	20.6
Primary school	33	34.0
Intermediate school	15	15.5
High school	10	10.3
University	18	18.6
Post-graduate	1	1.0

4.3.4.2 Perceived general health status of child (mother's assessment)

Information about the general health status of the child is presented in Table 4.4. Mothers rated the general health status of their children using a five-point scale, and almost half of the sample (48%) reported that their children had a good level of general health. In addition, around 22% thought that their children had a very good general health, while 5% said it was poor. Table 4.3 also presents the findings on child's specific health conditions as diagnosed by a doctor. The conditions included in this table are known to be common among children with Down Syndrome. Approximately 50% of mothers reported that their children suffered from heart problems during their lifetime. Results also showed that the occurrence of sensory problems, particularly visual problems, was high (53%). 40% of the sample reported that they are currently taking medication.

Table 4-4. Child's general health status (mothers' reports)

Variable	Girls Number (%) N=55	Boys Number (%) N=42	Total Number (%) N=97
Overall general health status			
Very poor	0 (0.00)	0 (0.00)	0 (0.00)
Poor	3 (5.45)	2 (4.76)	5 (5.15)
Fair	16 (29.09)	7 (16.67)	23 (23.71)
Good	23 (41.82)	24 (57.14)	47 (48.45)
Very good	13 (23.64)	9 (21.43)	22 (22.68)
Diagnosed medical conditions			
Heart problems	26 (47.27)	23 (54.76)	49 (50.51)
Thyroid gland disorder	19 (34.55)	10 (23.81)	29 (29.90)
Diabetes	2 (3.64)	1 (2.38)	3 (3.09)
Visual problems	32 (58.18)	20 (47.62)	52 (53.61)
Hearing problems	4 (7.27)	4 (9.52)	8 (8.28)
Others (kidney problems, psychotic problems, etc.)	11 (20.00)	2 (4.76)	13 (13.40)
Medication use *	27 (49.09)	12 (28.57)	39 (40.21)
Number of medications taken *	0.33 (0.57)	0.65 (0.84)	0.52 (0.75)

* Mean & Standard deviation (SD)

Table 4.5 presents information on the child's access to the health care system. Half of the sample was registered with a physician, and 42% had regular check-ups with their doctor. 50% of them visited physician only for treatment of specific problems (only with troubles). On asking mothers about how difficult it was to find a physician who can accept and treat their children, 30% responded either that it was difficult or very difficult (26% and 4% respectively), while 49% said it was easy.

Table 4-5. Access to general health care services by gender

Variable	Girls Number (%) N=55	Boys Number (%) N=42	Total Number (%) N=97
Registered with a physician			
Yes	29 (52.7)	20 (47.6)	49 (50.5)
No	26 (47.3)	22 (52.4)	48 (49.5)
Nature of visit to physician's			
Regular	23 (41.8)	18 (42.9)	41 (42.3)
Occasional	5 (9.1)	2 (4.7)	7 (7.2)
Only with troubles	27 (49.1)	22 (52.4)	49 (50.5)
Difficulty of finding a physician willing to treat child			
Very difficult	2 (3.6)	2 (4.8)	4 (4.1)
Difficult	14 (25.5)	12 (28.6)	26 (26.8)
Neither difficult nor easy	12 (21.8)	6 (14.3)	18 (18.6)
Easy	27 (49.1)	21 (50.0)	48 (49.5)
Very easy	0 (0.0)	1 (2.4)	1 (1.0)

4.3.4.3 Clinical oral health status

Data on the following section was classified by gender to give extra details on the sample characteristics; however, all presented data were not statistically significantly different across boys and girls. This section presents the oral health status of 97 boys and girls with Down Syndrome. Table 4.6 summarises the overall oral health status of the children examined. Protrusion of the tongue, dribbling of saliva at time of examination and the presence of visible anterior unrestored carious or missing teeth were all assessed visually, and these criteria were absent in almost all children (96% and above), both boys and girls.

Table 4-6. Other oral characteristics at time of examination, classified by gender

Variable	Girls Number (%) N= 55	Boys Number (%) N= 42	Total Number (%) N= 97
Protruded tongue	2 (3.6)	1 (2.4)	3 (3.1)
Dribbling	0 (0.0)	1 (2.4)	1 (1.0)
Anterior decayed/missed	1 (1.8)	0 (0.0)	1 (1.0)

The children's malocclusion status (over-jet, over-bite, and posterior cross-bite) was also assessed (Table 4.7). Over-jet classification was as follows: normal, protrusion, anterior cross-bite, or absent (this category comprised anterior open-bite, edge-to-edge bite, or an absence of anterior teeth). Only 18% of the sample was in the normal over-jet category while almost 60% were in the absent category. With regards to over-bite, cases were classified as having normal over-bite relationship, deep over-bite, anterior open-bite, edge-to-edge, or absent relationship in case of having anterior cross-bite, or absence of anterior teeth. Around half of the sample (50.52%)

had edge-to-edge relationship, and only 14 % had a normal over-bite. Posterior cross-bite presented in majority of children (73%). Data on malocclusion status among participated children appears to show similarities across boys and girls.

Table 4-7. Child's malocclusion status, classified by gender

Variable	Girls Number (%) N= 55	Boys Number (%) N= 42	Total Number (%) N= 97
Over-jet			
Normal	10 (18.2)	8 (19.1)	18 (18.6)
Protrusion	4 (7.3)	1 (2.4)	5 (5.1)
Anterior cross-bite	5 (9.1)	13 (30.9)	18 (18.6)
Absent	36 (65.4)	20 (47.6)	56 (57.7)
Over-bite			
Normal	8 (14.5)	6 (14.3)	14 (14.4)
Deep over-bite	6 (10.9)	3 (7.1)	9 (9.3)
Anterior open-bite	9 (16.4)	5 (11.9)	14 (14.4)
Edge-to-edge	28 (50.9)	21 (50.0)	49 (50.5)
Absent	4 (7.8)	7 (16.7)	11 (11.3)
Posterior cross-bite			
Present	38 (69.1)	33 (78.6)	71 (73.2)
Absent	17 (30.9)	9 (21.4)	26 (26.8)

Information on caries experience was also collected. Table 4.8 shows the means, standard deviations (SD), and quartiles of carious teeth, pulpally involved teeth, and filled teeth of both the primary and permanent dentition. Findings of caries experience were stratified by age (12-15 and 16-18 years old) to show the distribution of dental diseases across the two different age groups. Caries level was high among the study sample with a total mean of 1.4 for primary teeth and 3.7 for permanent carious teeth. The caries level was high in permanent teeth among older adolescents. The mean of pulpal-involved teeth was high (0.5, SD 1.3) in primary dentition of young age group, and higher (0.8, SD 2.1) in permanent dentition of older group. The data also shows that the mean of filled teeth was higher in permanent teeth in the older age group (1.3, SD 2.3) compared to the younger group.

Table 4-8. Child's caries experience, stratified by age

Variable	12 to 15 years old N= 49		16 to 18 years old N= 48		Total N=97	
	Mean (SD) Quartiles (25,50,75)		Mean (SD) Quartiles (25,50,75)		Mean (SD) Quartiles (25,50,75)	
	Primary	Permanent	Primary	Permanent	Primary	Permanent
Carious teeth	1.0 (1.5) 0, 0, 1	2.6 (3.2) 0, 1, 4	0.4 (1.1) 0, 0, 0	3.8 (4.1) 0, 2.5, 6	0.7 (1.4) 0, 0, 1	3.2 (3.7) 0, 2, 5
Pulpal-involved teeth	0.5 (1.3) 0, 0, 0	0.1 (0.4) 0, 0, 0	0.1 (0.3) 0, 0, 0	0.8 (2.1) 0, 0, 0	0.3 (1.0) 0, 0, 0	0.5 (1.6) 0, 0, 0
Filled teeth	0.5 (1.2) 0, 0, 0	0.4 (1.0) 0, 0, 0	0.2 (0.7) 0, 0, 0	1.3 (2.3) 0, 0, 2	0.3 (1.0) 0, 0, 0	0.8 (1.8) 0, 0, 1

Table 4.9 presents the findings of periodontal health status of the participating children. The majority of the children had a mild plaque index (73%), while around 27% of them had moderate plaque accumulation. With regards to gingival index, almost half of children had mild gingivitis and the other half had moderate. The prevalence of mild and moderate plaque accumulation and gingivitis was similar in both boys and girls. None of the participants were categorised in the severe categories for either plaque or gingival index.

Table 4-9. Child's periodontal health status, classified by gender

Variable	Girls Number (%) N= 55	Boys Number (%) N= 42	Total Number (%) N=97
Plaque index			
Mild	41 (74.6)	30 (71.4)	71 (73.2)
Moderate	14 (25.4)	12 (28.6)	26 (26.8)
Severe	0 (0.0)	0 (0.0)	0 (0.0)
Gingival index			
Mild	26 (47.3)	24 (57.1)	50 (51.6)
Moderate	29 (52.7)	18 (42.9)	47 (48.4)
Severe	0 (0.0)	0 (0.0)	0 (0.0)

4.3.4.4 Perceived oral health status of the child

This section presents some subjective oral health findings based upon the mothers' perception. Using a five point scale, the majority of mothers perceived that the oral health of their children was generally fair (43%), 25% of them reported it to be good, while only 5% thought it was very good. Around 26% rated it as poor or very poor. Data on children's experiences of oral health conditions in the last 12 months showed that more than half of mothers reported mouth breathing as a problem, and a minority (7%) reported the problem of dribbling in the last 12 months. Nearly half (46%) indicated that tooth alignment was a problem for their children. A considerable number of mothers also believed that their children had caries (57.7%), and 46% reported that their children had complained from toothache. Around one-third of participants reported bleeding gums, bad breath, and a habit of tooth grinding as other problems. Nearly half (44%) of mothers thought that their children had enlarged tongues, but only 19% reported that the tongue was protruding outside the mouth. The majority of mothers (70%) said that their children experienced difficulty speaking, while only 17% reported difficulties with chewing. The prevalence of these oral health conditions was similar among boys and girls, with only minimal differences in some cases (Table 4.10).

Table 4-10. Subjective assessment of the child's oral health status (mothers' reports), classified by gender

Variable	Girls Number (%) N=55	Boys Number (%) N=42	Total Number (%) N=97
Overall oral health status			
Very poor	4 (7.3)	1 (2.4)	5 (5.2)
Poor	7 (12.7)	13 (30.9)	20 (20.6)
Fair	27 (49.1)	15 (35.7)	42 (43.3)
Good	15 (27.3)	10 (23.8)	25 (25.8)
Very good	2 (3.6)	3 (7.1)	5 (5.1)
Oral health condition			
Mouth breathing	28 (50.9)	25 (59.5)	53 (54.6)
Dribbling	5 (9.1)	2 (4.8)	7 (7.2)
Crooked teeth	21 (38.2)	24 (57.1)	45 (46.4)
Tooth decay	34 (61.8)	22 (52.4)	56 (57.7)
Toothache	27 (49.1)	18 (42.9)	45 (46.4)
Bleeding gums	21 (38.2)	12 (29.3)	33 (34.4)
Bad breath	17 (30.9)	18 (42.9)	35 (36.1)
Tooth grinding	16 (29.1)	17 (40.5)	33 (34.0)
Enlarged tongue	27 (49.1)	16 (38.1)	43 (44.3)
Protruding tongue	9 (16.4)	10 (23.8)	19 (19.6)
Difficulty speaking	35 (63.6)	33 (78.6)	68 (70.1)
Difficulty chewing	8 (14.5)	9 (21.4)	17 (17.5)

Table 4.11 presents findings on the child's previous dental visits, and types of treatment received. Almost 60% of mothers said that their children had visited the dentist in more than 3 occasions; however, 15% reported that they had never been to a dentist before. Almost two-thirds (63%) reported that their child visited the dentist only to seek treatment (problem oriented), while only 9% regularly going to the dentist (prevention oriented). Almost half of the participated mothers reported that finding a dentist who was willing to treat their children with Down Syndrome was difficult or very difficult (36% and 15% respectively). Asking mothers about the type of dental treatment their children received, they reported that around 50% received fillings and extractions. Results on the treatment under general anaesthesia were high in which 33% received dental intervention under such treatment. A very small number said that their children have ever received any form of preventive dental intervention such as fissure sealants (6%).

Table 4-11. Summary of dental attendance patterns, characteristics, and treatment received, classified by gender

Variable	Girls Number (%) N=55	Boys Number (%) N=42	Total Number (%) N=97
Number of previous dental Visits			
Never been to dentist	7 (12.7)	8 (19.1)	15 (15.5)
Once	2 (3.6)	5 (11.9)	7 (7.2)
Two to three times	11 (20.0)	5 (11.9)	16 (16.5)
More than three times	35 (63.6)	24 (57.1)	59 (60.8)
Nature of dental visit			
Regular	6 (10.9)	3 (7.1)	9 (9.3)
Occasional	6 (10.9)	6 (14.3)	12 (12.4)
Only to seek treatment	36 (65.5)	25 (59.5)	61 (62.9)
Not applicable (never visited)	7 (12.7)	8 (19.1)	15 (15.5)
Difficulty of finding a dentist			
Very difficult	9 (16.4)	6 (14.3)	15 (15.5)
Difficult	21 (38.2)	14 (33.3)	35 (36.1)
Neither difficult nor easy	6 (10.9)	6 (14.3)	12 (12.4)
Easy	14 (25.5)	9 (21.4)	23 (23.7)
Very easy	1 (1.8)	1 (2.4)	2 (2.1)
Never tried	4 (7.3)	6 (14.3)	10 (10.3)
Previous dental treatment			
Filling	26 (47.3)	20 (47.6)	46 (47.4)
Extraction	30 (54.6)	23 (54.8)	53 (54.6)
Treatment under general anaesthesia	16 (29.1)	16 (38.1)	32 (33.0)
Treatment under sedation	2 (3.6)	0 (0.0)	2 (2.1)
Preventive dental treatment (i.e. fissure sealant)	4 (7.3)	2 (4.8)	6 (6.2)
Braces	1 (1.8)	0 (0.0)	1 (1.0)
Crowns	3 (5.5)	2 (2.8)	5 (5.2)
Scale and polish	23 (41.8)	20 (47.6)	43 (44.3)
X-ray taken	26 (47.3)	23 (54.8)	49 (50.5)
Examination only (no treatment)	8 (14.6)	3 (7.1)	11 (11.3)

Findings on the children's tooth brushing behaviour are summarized in Table 4.12. Over a quarter (27%) of mothers reported that their children started brushing their teeth between the age of 2 and 4 years, and 26% between 4 and 6 years of age. More than a third (39%) began brushing at 6 years of age or older. The majority (69%) of mothers said that the child brushed his/her teeth unsupervised, while around 30% said that the child needed to be accompanied by an adult at time of tooth brushing. With regards to the frequency of tooth brushing, the majority of mothers reported that their children brushed their teeth one to two times a day (34% and 40% respectively), and 13% reported that brushing was done less than once a day.

Table 4-12. Tooth brushing behaviours, classified by gender

Variable	Girls Number (%) N=55	Boys Number (%) N=42	Total Number (%) N=97
Age at which the child began brushing			
Under 6 months of age	0 (0.0)	0 (0.0)	0 (0.0)
Between 6 months and 1 year of age	1 (1.8)	0 (0.0)	1 (1.0)
Between 1 and 2 years of age	3 (5.5)	1 (2.4)	4 (4.1)
Between 2 and 4 years of age	12 (21.8)	15 (35.7)	27 (27.8)
Between 4 and 6 years of age	17 (30.9)	9 (21.4)	26 (26.8)
6 years of age or older	22 (40.0)	16 (38.1)	38 (39.2)
Never brushed the teeth	0 (0.0)	1 (2.4)	1 (1.0)
Person/people responsible for tooth brushing			
Child	44 (80.0)	23 (54.7)	67 (69.1)
An adult	0 (0.0)	0 (0.0)	0 (0.0)
An adult and the child	11 (20.0)	18 (42.9)	29 (29.9)
Not applicable (never brushed)	0 (0.0)	1 (2.4)	1 (1.0)
Frequency of tooth brushing			
More than three times a day	0 (0.0)	0 (0.0)	0 (0.0)
Three times a day	8 (14.5)	3 (7.1)	11 (11.3)
Twice a day	26 (47.3)	13 (30.9)	39 (40.2)
Once a day	16 (29.1)	17 (40.5)	33 (34.0)
Less than once a day	5 (9.1)	8 (19.1)	13 (13.4)
Never	0 (0.0)	1 (2.4)	1 (1.0)

4.3.4.5 Child's OHRQoL

Mothers were asked about the prevalence of oral-related problems and/or conditions and their impacts on different aspects of the child's life; whether experienced at all, and if so, the occurrence of problems within the last twelve months.

The prevalence of oral health impacts is presented in Table 4.13. Overall, 82% of children had at some point experienced an impact of oral health problems on their life. Pain was the most frequently reported problem, with more than 75% of the sample reporting that their child had experienced it at some point in their life, while the majority said that the experience of pain had been moderate and/or severe. The second most reported impact was difficulty eating (44%). Oral health problems also appeared to have an impact on several daily activities such as speaking (15%), tooth brushing (21%), sleeping patterns (21%), education (14%), and playing (21%). Oral health conditions also appeared to have a negative impact on the emotional aspects of the child's life, with the most common emotional impacts being crying (38%), and being quiet (33%) because of oral health-related problems such as pain as a result of tooth decay. While some mothers also reported that the child became withdrawn from family (12%) and friends (8%) because of their oral health status, very few said that their children were excluded by their peers because of oral health-related conditions (2%). Around 15% said that their children had been teased because of their oral health. The total prevalence (in which participants were not mutually excluded) of such impact was as follows: minor severity was 56.7%, moderate was 57.7%, and severe was 35.1%. However, considering the most severe impact for each participant, 35.1% were reported to have experienced a severe impact, 31.9% a moderate impact while 15.5% experienced only minor oral health-related impacts.

Table 4.14 also presents child's OHRQoL within the last year. The oral health impacts experienced by children in the last year appeared to show very similar patterns as the findings of such impacts experienced throughout the course of their life (shown in the previous table). Pain as caused by dental problems had the highest prevalence with 50%, followed by problems eating that were reported in 26% of the study sample. Impacts on the emotional as well as social aspects of the child's life appeared to be common. The total prevalence of any oral health impacts experienced

in the last year with only minor severity was 43.3%, moderate was 41.2%, and severe was 18.6%. However, considering the most severe impact for each participant, 18.5% were reported to have experienced a severe impact, 26.8% a moderate impact while 15.5% experienced only minor oral health-related impacts.

Table 4-13. Prevalence of reported child's OHRQoL impacts (Ever-happened)

Type problem/impact experienced	Prevalence N (%)		Severity of the problem/impact N (%)		
	Never happened	Ever happened	Minor	Moderate	Severe
Physiological pain Pain	23 (23.7)	74 (76.3)	17 (17.5)	29 (29.9)	28 (28.9)
Activity Eating	54 (55.7)	43 (44.3)	18 (18.6)	14 (14.4)	11 (11.3)
Speaking	82 (84.5)	15 (15.5)	2 (2.1)	11 (11.3)	2 (2.1)
Tooth Brushing	76 (78.4)	21 (21.6)	9 (9.3)	7 (7.2)	5 (5.1)
Sleeping	76 (78.4)	21 (21.6)	3 (3.1)	11 (11.3)	7 (7.2)
Schooling	83 (85.6)	14 (14.4)	5 (5.1)	6 (6.2)	3 (3.1)
Playing	76 (78.4)	21 (21.6)	4 (4.1)	12 (12.4)	5 (5.1)
Emotional Crying	60 (61.8)	37 (38.2)	12 (12.4)	9 (9.3)	16 (16.5)
Ceasing to laugh	68 (70.1)	29 (29.9)	10 (10.3)	13 (13.4)	6 (6.2)
Being quiet	65 (67.0)	32 (33.0)	12 (12.4)	15 (15.5)	5 (5.1)
Shyness	83 (85.6)	14 (14.4)	6 (6.2)	6 (6.2)	2 (2.0)
Embarrassment	87 (89.6)	10 (10.4)	5 (5.2)	5 (5.2)	0 (0.0)
Lack of confidence	89 (91.7)	8 (8.3)	3 (3.1)	5 (5.2)	0 (0.0)
Self-consciousness	97 (100)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Anger	80 (82.5)	17 (17.5)	6 (6.2)	10 (10.3)	1 (1.0)
Stubbornness	91 (93.8)	6 (6.2)	4 (4.1)	2 (2.1)	0 (0.0)
Social Withdrawal from family	85 (87.6)	12 (12.4)	3 (3.1)	8 (8.3)	1 (1.0)
Withdrawal from friends	89 (91.8)	8 (8.2)	1 (1.0)	7 (7.2)	0 (0.0)
Exclusion by peers	95 (97.9)	2 (2.1)	1 (1.0)	1 (1.0)	0 (0.0)
Teasing/Bullying	82 (84.5)	15 (15.5)	6 (6.2)	9 (9.3)	0 (0.0)
Any	17 (17.5)	80 (82.5)	15 (15.5)	31 (31.9)	34 (35.1)

Table 4-14. Prevalence of reported child's OHRQoL impacts (Last-year)

Type problem/impact experienced	Prevalence N (%)		Severity of the problem/impact N (%)		
	Never happened	Happened last year	Minor	Moderate	Severe
Physiological pain Pain	48 (49.5)	49 (50.5)	15 (15.5)	20 (20.6)	14 (14.4)
Activity Eating	71 (73.2)	26 (26.8)	13 (13.4)	10 (10.3)	3 (3.1)
Speaking	87 (89.7)	10 (10.3)	2 (2.1)	7 (7.2)	1 (1.0)
Tooth Brushing	83 (85.5)	14 (14.5)	7 (7.2)	5 (5.2)	2 (2.1)
Sleeping	87 (89.7)	10 (10.3)	0 (0.0)	6 (6.2)	4 (4.1)
Schooling	90 (92.8)	7 (7.2)	4 (4.1)	1 (1.0)	2 (2.1)
Playing	84 (86.6)	13 (13.4)	4 (4.1)	7 (7.2)	2 (2.1)
Emotional Crying	74 (76.2)	23 (23.8)	8 (8.3)	7 (7.2)	8 (8.3)
Ceasing to laugh	80 (82.4)	17 (17.6)	6 (6.2)	6 (6.2)	5 (5.2)
Being quiet	79 (81.4)	18 (18.6)	6 (6.2)	8 (8.3)	4 (4.1)
Shyness	86 (88.6)	11 (11.4)	4 (4.1)	5 (5.2)	2 (2.1)
Embarrassment	89 (91.8)	8 (8.2)	4 (4.1)	4 (4.1)	0 (0.0)
Lack of confidence	91 (93.8)	6 (6.2)	2 (2.1)	4 (4.1)	0 (0.0)
Self-consciousness	97 (100)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Anger	82 (84.5)	15 (15.5)	6 (6.2)	8 (8.3)	1 (1.0)
Stubbornness	92 (94.9)	5 (5.1)	4 (4.1)	1 (1.0)	0 (0.0)
Social Withdrawal from family	90 (92.8)	7 (7.2)	1 (1.0)	5 (5.2)	1 (1.0)
Withdrawal from friends	93 (95.9)	4 (4.1)	1 (1.0)	3 (3.1)	0 (0.0)
Exclusion by peers	95 (98.0)	2 (2.0)	1 (1.0)	1 (1.0)	0 (0.0)
Teasing/Bullying	83 (85.5)	14 (14.5)	5 (5.2)	9 (9.3)	0 (0.0)
Any	38 (39.2)	59 (60.8)	15 (15.5)	26 (26.8)	18 (18.5)

4.3.4.6 Family's OHRQoL

The impacts of the child's oral health on different aspects of family's QoL are presented in tables 4.15 (Ever-happened) and 4.16 (Last-year). The results showed that child's oral health had strong impacts on emotional aspects of the family's QoL, mainly causing feelings of depression and distress. This was reported by 65% of the sample (table 4.15) of whom the majority reported that such feelings were severe (40%). Feelings of being worried about the child's oral health (55%) and the parents blaming themselves (referred to in table as 'self-blame') (52%) were also experienced by a majority. The findings also suggest that family sleeping patterns were negatively affected by the child's oral health problems, which was reported in around one-third of the sample (32%). The total prevalence (in which participants were not mutually excluded) of oral health impacts on family's QoL with minor severity was 27.8%, moderate was 54.6%, and severe was 50.5%. However, considering the most severe impact for each participant, 50.5% were reported to have experienced a severe impact, 20.6% a moderate impact while 6.2% experienced only minor oral health-related impacts.

As mentioned earlier, table 4.16 presents the findings of the impact of the child's oral health on family's QoL that had been experienced in the last year. Findings appeared to be similar to the previous table (Ever-happened) with slightly lower prevalence. The most commonly reported impacts were emotional, such as feeling depressed or distressed (50%), being worried about the child's oral health (46%) and feelings of self-blame in parents (41%) with varying degrees of severity. The impact of the child's oral health on family sleeping patterns was also high, with a prevalence of 20%. Results showed that the calculated total prevalence of any oral health impact on family quality of life that was experienced within the last year with minor severity was 21.6%, while moderate was 43.3%, and severe was 40.2%. In contrast, considering the most severe impact for each participant, 40.2% were reported to have experienced a severe impact, 16.5% a moderate impact while 6.2% experienced only minor oral health-related impacts.

Table 4-15. Prevalence of reported family's QoL (Ever-happened)

Type problem/impact experienced	Prevalence N (%)		Severity of the problem/impact N (%)		
	Never happened	Ever happened	Minor	Moderate	Severe
Activity					
Scheduled activity cancelled	82 (84.5)	15 (15.5)	7 (7.2)	5 (5.2)	3 (3.1)
Disruption to parent's employment	95 (98.0)	2 (2.0)	1 (1.0)	0 (0.0)	1 (1.0)
Isolation from other family members	80 (82.5)	17 (17.5)	5 (5.2)	9 (9.2)	3 (3.1)
Disruption to family sleeping patterns	66 (68.0)	31 (32.0)	5 (5.2)	17 (17.5)	9 (9.3)
Emotional					
Feelings of depression or distress	34 (35.0)	63 (65.0)	3 (3.1)	21 (21.7)	40 (40.2)
Self-blame/ Guilt	46 (47.4)	51 (52.6)	3 (3.1)	14 (14.4)	34 (35.1)
Worry	43 (44.3)	54 (55.7)	4 (4.1)	27 (27.9)	23 (23.7)
Anger	79 (81.4)	18 (18.6)	3 (3.1)	9 (9.3)	6 (6.2)
Conflict					
Arguments	82 (84.5)	15 (15.5)	8 (8.3)	6 (6.2)	1 (1.0)
Jealousy	94 (96.9)	3 (3.1)	1 (1.0)	0 (0.0)	2 (2.1)
Any	22 (22.7)	75 (77.3)	6 (6.2)	20 (20.6)	49 (50.5)

Table 4-16. Prevalence of reported family's QoL (Last-year)

Type problem/impact experienced	Prevalence N (%)		Severity of the problem/impact N (%)		
	Never happened	Happened Last-year	Minor	Moderate	Severe
Activity					
Scheduled activity cancelled	89 (91.7)	8 (8.3)	6 (6.2)	2 (2.1)	0 (0.0)
Disruption to parent's employment	96 (99.0)	1 (1.0)	1 (1.0)	0 (0.0)	0 (0.0)
Isolation from other family members	88 (90.7)	9 (9.3)	3 (3.1)	6 (6.2)	0 (0.0)
Disruption to family sleeping patterns	77 (79.4)	20 (20.6)	4 (4.1)	13 (13.4)	3 (3.1)
Emotional					
Feelings of depression or distress	48 (49.5)	49 (50.5)	3 (3.1)	16 (16.5)	30 (30.9)
Self-blame/ Guilt	57 (58.8)	40 (41.2)	3 (3.1)	11 (11.3)	26 (26.8)
Worry	52 (53.6)	45 (46.4)	4 (4.1)	22 (22.7)	19 (19.6)
Anger	82 (84.5)	15 (15.5)	3 (3.1)	8 (8.3)	4 (4.1)
Conflict					
Arguments	86 (88.6)	11 (11.4)	5 (5.2)	5 (5.2)	1 (1.0)
Jealousy	94 (96.9)	3 (3.1)	1 (1.0)	0 (0.0)	2 (2.1)
Any	36 (37.1)	61 (62.9)	6 (6.2)	16 (16.5)	39 (40.2)

4.3.4.7 Overall impact of child's oral health

Findings on the overall oral health impact on the child and family are presented in Table 4.17. Almost half of mothers (47%) thought that the child's oral health had no impact on their life, and 30% reported that the overall impact was very little. Very few mothers said that child's oral health had a more severe impact on their life (7% responded "a lot", and only 1% "very much"). Conversely, around 25% of mothers thought that the child's oral health had a severe impact on the family (18% responding "a lot", and 6% "very much"). So, according to these findings, mothers believed that the negative impact of their child's oral health was felt more strongly by the family as a whole, in comparison to the child.

Table 4-17. Frequency distribution of global QoL rating

Rating	Number N=97	%
Overall impact on child		
Not at all	46	47.4
Very little	30	30.9
Some	13	13.4
A lot	17	7.2
Very much	1	1.0
Overall impact on family		
Not at all	34	35.1
Very little	17	17.5
Some	22	22.7
A lot	18	18.6
Very much	6	6.2

4.3.5 Psychometric testing of questionnaire

This section covers the psychometric testing process of the developed questionnaire, presenting findings on both reliability and validity assessments.

4.3.5.1 Instrument reliability

a) Internal consistency reliability

The **inter-item correlation** for the child's OHRQoL was assessed. The child's OHRQoL was recorded for two different time-frames; ever-happened and/or happened in the last year. The inter-item correlation coefficients for the 20 items "Ever happened" and "Last year" child section of OHRQoL was performed and presented in Tables 4.18 and 4.19 separately. Table 4.18 shows the relationships between items in the "Ever-happened" section, and the highest score was 0.96, which represented the relationship between the child being embarrassed and less confident because of their oral health status. A similar pattern of relationships between items was obtained when inter-item correlation coefficients of the "Last-year" part of the child OHRQoL was analysed, Table 4.19.

Table 4-18 Inter-item correlation for child's OHRQoL (Ever-happened)

	Pain	Eat	Speak	Brush	Sleep	School	Play	Cry	Stop laugh	Quiet	Shy	Embarrassed	Less confident	Conscious	Angry	Stubborn	Withdraw Family	Withdraw friend	Been excluded	Teased/Bullied	
Pain	1.00																				
Eat	0.49	1.00																			
Speak	0.30	0.59	1.00																		
Brush	0.33	0.67	0.37	1.00																	
Sleep	0.40	0.57	0.46	0.47	1.00																
School	0.27	0.39	0.31	0.40	0.60	1.00															
Play	0.40	0.60	0.45	0.51	0.50	0.49	1.00														
Cry	0.56	0.60	0.52	0.42	0.54	0.45	0.58	1.00													
Stop laugh	0.51	0.69	0.60	0.47	0.53	0.39	0.61	0.74	1.00												
Quiet	0.52	0.69	0.57	0.52	0.52	0.40	0.65	0.77	0.92	1.00											
Shy	0.18	0.45	0.43	0.46	0.52	0.25	0.44	0.42	0.48	0.44	1.00										
Embarrassed	0.01	0.20	0.29	0.08	0.38	0.24	0.20	0.24	0.24	0.18	0.66	1.00									
Less confident	0.05	0.24	0.32	0.10	0.41	0.26	0.23	0.28	0.27	0.22	0.63	0.96	1.00								
Conscious	-	-	-	-	-	-	-	-	-	-	-	-	-	-							
Angry	0.26	0.36	0.32	0.50	0.36	0.36	0.34	0.33	0.41	0.35	0.54	0.20	0.16	-	1.00						
Stubborn	0.21	0.36	0.46	0.47	0.39	0.16	0.32	0.38	0.37	0.33	0.45	0.17	0.19	-	0.46	1.00					
Withdraw Family	0.26	0.45	0.33	0.40	0.56	0.57	0.24	0.32	0.43	0.39	0.38	0.22	0.24	-	0.57	0.15	1.00				
Withdraw friend	0.15	0.35	0.40	0.19	0.51	0.39	0.12	0.29	0.33	0.28	0.25	0.31	0.33	-	0.38	0.22	0.79	1.00			
Been excluded	0.04	0.07	-0.06	0.04	-0.07	-0.05	-0.07	0.14	0.11	0.21	-0.05	-0.04	-0.04	-	-0.06	-0.03	-0.05	-0.04	1.00		
Teased/Bullied	0.13	0.15	0.06	0.05	0.15	0.13	0.13	0.10	0.14	0.13	0.03	0.08	0.10	-	-0.08	0.10	0.17	0.27	0.24	1.00	

Table 4-19 Inter-item correlation for child's OHRQoL (Last-year)

	Pain	Eat	Speak	Brush	Sleep	School	Play	Cry	Stop laugh	Quiet	Shy	Embarrassed	Less confident	Conscious	Angry	Stubborn	Withdraw Family	Withdraw friend	Been excluded	Teased/ Bullied
Pain	1.00																			
Eat	0.55	1.00																		
Speak	0.39	0.66	1.00																	
Brush	0.34	0.73	0.41	1.00																
Sleep	0.44	0.51	0.42	0.46	1.00															
School	0.31	0.17	-0.01	0.33	0.47	1.00														
Play	0.49	0.57	0.57	0.39	0.45	0.40	1.00													
Cry	0.70	0.58	0.53	0.41	0.40	0.31	0.50	1.00												
Stop laugh	0.56	0.68	0.58	0.42	0.47	0.33	0.56	0.72	1.00											
Quiet	0.57	0.68	0.54	0.50	0.44	0.30	0.60	0.74	0.91	1.00										
Shy	0.29	0.52	0.49	0.50	0.60	0.28	0.54	0.46	0.57	0.54	1.00									
Embarrassed	0.08	0.11	0.26	-0.03	0.31	0.16	0.21	0.16	0.26	0.20	0.62	1.00								
Less confident	0.11	0.14	0.29	-0.01	0.35	0.18	0.24	0.19	0.29	0.23	0.59	0.95	1.00							
Conscious	-	-	-	-	-	-	-	-	-	-	-	-	-	-						
Angry	0.42	0.52	0.32	0.53	0.44	0.31	0.44	0.42	0.50	0.44	0.51	0.07	0.02	-	1.00					
Stubborn	0.29	0.48	0.54	0.49	0.42	-0.05	0.41	0.47	0.45	0.41	0.52	0.11	0.12	-	0.42	1.00				
Withdraw Family	0.27	0.48	0.10	0.35	0.55	0.43	0.27	0.31	0.43	0.39	0.37	0.10	0.12	-	0.58	0.01	1.00			
Withdraw friend	0.20	0.29	0.22	0.01	0.55	0.06	0.10	0.17	0.20	0.20	0.21	0.21	0.22	-	0.19	0.06	0.64	1.00		
Been excluded	0.12	0.16	-0.05	0.09	-0.04	-0.03	-0.05	0.22	0.17	0.28	-0.04	-0.04	-0.04	-	-0.05	-0.03	-0.04	-0.03	1.00	
Teased/ Bullied	0.07	0.06	0.04	-0.07	0.10	0.04	0.06	0.09	0.13	0.13	-0.02	0.04	0.07	-	-0.16	-0.08	0.15	0.30	0.25	1.00

The **item-total statistics** for child's OHRQoL was also assessed. The item-scale correlations of the questionnaire for both the 'Ever-happened' and the 'Last-year' sections of the child's OHRQoL are listed in Tables 4.20 and 4.21 respectively. The Cronbach's Alpha for the 20 items 'Ever-happened' child's OHRQoL was 0.909 (Table 4.20) showing a very satisfactory coefficient results. The corrected item-total correlations ranged from 0.043 for the item 'been excluded' to 0.806 for 'ceasing of laugh' item. The Cronbach's Alpha was lower after deleting any of the items with the exception of "been excluded" and/or 'teasing/bullying' items when the alpha was slightly increased after dropping them.

Very similar results were obtained when data of child OHRQL 'Last year' part was analysed with an item total Cronbach's Alpha of 0.902, which is similar to the Cronbach's Alpha of child's OHRQoL 'Ever-happened' and shows a good correlation. The corrected item-total correlations ranged from 0.094 for "teased/bullied" item to 0.814 for 'stop laughing' item. There was also slight increase in Cronbach's Alpha after deleting both 'Exclusion by peers' and/or 'teasing/bullying' items, Table 4.21.

Table 4-20. Internal consistency reliability for child's OHRQoL: Item-total correlation coefficients, Alpha if item deleted, Cronbach's Alpha (Ever-happened)

Impact Ever happened	Corrected item-total correlation coefficients	Alpha if item deleted
Pain	0.503	0.908
Eating	0.766	0.898
Speaking	0.628	0.903
Tooth Brushing	0.610	0.903
Sleeping	0.722	0.899
Education	0.565	0.905
Playing	0.665	0.901
Crying	0.744	0.899
Ceasing to laugh	0.806	0.897
Being quiet	0.795	0.897
Shyness	0.610	0.903
Embarrassment	0.371	0.909
Lack of confidence	0.408	0.908
Self-consciousness	-	-
Anger	0.516	0.906
Stubbornness	0.477	0.908
Withdrawal from family	0.580	0.905
Withdrawal from friends	0.477	0.907
Exclusion by peers	0.043	0.912
Teasing/Bullying	0.167	0.913
Total Alpha 0.909		

Table 4-21. Internal consistency reliability for child's OHRQoL: Item-total correlation coefficient, Alpha if item deleted, Cronbach's Alpha (Last-year)

Impact Last year	Corrected item-total correlation coefficients	Alpha if item deleted
Pain	0.619	0.898
Eating	0.773	0.889
Speaking	0.613	0.895
Tooth Brushing	0.570	0.896
Sleeping	0.675	0.893
Education	0.395	0.900
Playing	0.663	0.893
Crying	0.732	0.891
Ceasing to laugh	0.814	0.888
Being quiet	0.801	0.888
Shyness	0.691	0.893
Embarrassment	0.313	0.902
Lack of confidence	0.342	0.902
Self-consciousness	-	-
Anger	0.578	0.896
Stubbornness	0.504	0.900
Withdrawal from family	0.520	0.898
Withdrawal from friends	0.347	0.902
Exclusion by peers	0.107	0.905
Teasing/Bullying	0.094	0.908
Total Alpha 0.902		

The **inter-item correlation** for family's OHRQoL was also assessed. The inter-item correlation coefficients for the 10 items 'Ever-happened' and "Last-year" family's OHRQoL was performed and presented in tables 4.22 and 4.23 separately. Table 4.22 shows the relationships between items in the "Ever happened" part, in which results showed that the highest score was 0.64, which represented the relationship between the scheduled family activities and isolation from others as a result of the child's oral health problems, and also between the mother feeling depression and worry, with a correlation score of 0.61. Similar patterns of relationships between items were found when inter-item correlation coefficients of the "Last-year" part of the family's OHRQoL was analysed, but a strong correlation was also found between the mother experiencing feelings of depression and self-blame and feeling worried about the child's oral health (see Table 4.23).

Table 4-22. Inter-item correlation for family's OHRQoL (Ever-happened)

	Activity	Employment	Isolation	Sleep	Depression	Self-blame	Worry	Angry	Argument	Jealousy
Activity	1.00									
Employment	0.28	1.00								
Isolation	0.64	0.32	1.00							
Sleep	0.51	0.25	0.63	1.00						
Depression	0.33	0.13	0.32	0.54	1.00					
Self-blame	0.37	0.15	0.33	0.46	0.59	1.00				
Worry	0.41	0.17	0.42	0.55	0.61	0.48	1.00			
Angry	0.37	0.27	0.57	0.54	0.32	0.38	0.41	1.00		
Argument	0.31	-0.05	0.34	0.36	0.22	0.32	0.20	0.21	1.00	
Jealousy	0.24	-0.02	0.02	0.09	0.11	0.14	0.11	0.08	0.05	1.00

Table 4-23. Inter-item correlation for famil't's OHRQoL (Last-year)

	Activity	Employment	Isolation	Sleep	Depression	Self-blame	Worry	Angry	Argument	Jealousy
Activity	1.00									
Employment	0.25	1.00								
Isolation	0.58	-0.03	1.00							
Sleep	0.54	0.20	0.59	1.00						
Depression	0.27	0.13	0.27	0.50	1.00					
Self-blame	0.35	0.15	0.31	0.47	0.63	1.00				
Worry	0.32	0.16	0.36	0.48	0.70	0.56	1.00			
Angry	0.31	-0.04	0.57	0.55	0.37	0.41	0.42	1.00		
Argument	0.36	-0.03	0.26	0.28	0.27	0.32	0.16	0.19	1.00	
Jealousy	0.15	-0.02	0.09	0.18	0.16	0.19	0.14	0.11	0.07	1.00

The **item-total statistics** for family's OHRQoL was assessed. Table 4.24 presents the item-scale correlations of the "Ever-happened" family's OHRQoL. The Cronbach's Alpha for the 10 items was 0.828, showing again a satisfactory level of correlation. The corrected item-total correlations ranged from 0.150 for "being jealous" to 0.730 for "sleeping pattern" item. The Cronbach's Alpha slightly increased after deleting "feel jealous" and/or "effect on job" items.

As shown in Table 4.25, similar results were obtained when data of family's OHRQoL "Last-year" part was analysed with an item total Cronbach's Alpha of 0.807, and corrected item-total correlations ranged from 0.199 for "feeling jealous", to 0.675 for "being worried". The Cronbach's Alpha scores were also slightly increased after deleting "feel jealous" and/or "effect on job" items.

Table 4-24. Internal consistency reliability for family's QoL: Item-total correlation coefficients, Alpha if item deleted, Cronbach's Alpha (Ever-happened)

Impact Ever happened	Corrected item-total correlation coefficients	Alpha if item deleted
Scheduled activity cancelled	0.589	0.809
Disruption to parent's employment	0.260	0.833
Isolation from other family members	0.623	0.805
Disruption to family sleeping patterns	0.730	0.788
Feelings of depression or distress	0.631	0.802
Self-blame/ Guilt	0.614	0.805
Worry	0.651	0.798
Anger	0.559	0.808
Arguments	0.358	0.826
Jealousy	0.150	0.837
Total Alpha 0.828		

Table 4-25. Internal consistency reliability for family's QoL: Item-total correlation coefficients, Alpha if item deleted, Cronbach's Alpha (Last-year)

Impact Last year	Corrected item-total correlation coefficients	Alpha if item deleted
Scheduled activity cancelled	0.505	0.798
Disruption to parent's employment	0.145	0.815
Isolation from other family members	0.522	0.792
Disruption to family sleeping patterns	0.673	0.768
Feelings of depression or distress	0.693	0.764
Self-blame/ Guilt	0.672	0.767
Worry	0.675	0.765
Anger	0.550	0.783
Arguments	0.333	0.804
Jealousy	0.199	0.812
Total Alpha 0.807		

b) External (Test-retest) reliability

Test-retest reliability for 10 participants, who took part again in two weeks after their initial participation, was performed (Appendix 15). The un-weighted and weighted Kappa coefficients were calculated for both sections of the developed questionnaire. The results were satisfactory in which the minimum weighted Kappa coefficient of “Ever-happened” child OHRQL was 0.76, and the minimum weighted Kappa coefficient of “Ever-happened” family’s OHRQoL was 0.60.

4.3.5.2 Instrument validity

a) Face and content validity

All items included in this questionnaire were derived from a literature review of OHRQoL studies and from phase-one qualitative interviews with mothers who were the primary carers of their children with Down Syndrome. At this stage, we tried to be inclusive of all possible items related to the OHRQoL of children with Down Syndrome. Mothers’ interviews provided useful insight into their concerns about the oral health of their children and identified common conditions related to their oral health and their potential impacts on child and family’s QoL.

Each item/question involved in the questionnaire fell into at least one content area/dimension (such as: emotional, activity/functional, social etc.). Each content area/dimension was presented by at least one item/question. According to literature review and mothers’ interviews, number of items involved in each content area reflected its actual importance or relevance to that area.

Face validity is so closely related to content validity (Streiner et al., 2015). To make sure that the questionnaire is measuring what it is intended to measure “face validity”, all items included were generated from existing literature and supplemented by findings of phase one mothers’ interviews.

b) Construct validity

With the absence of a gold standard or reference measure to assess the criterion validity of our developed instrument, we proposed underlying factors that might explain the relationships among various behaviours and attitudes, and therefore, help explain the observed correlations among variables.

Construct validity was assessed by comparing the child's OHRQoL to: a) perceived general health, b) perceived oral health, and c) the overall extent that oral problems affected child's QoL.

Child's OHRQoL

Table 4.26 presents the findings of "Ever-happened" child's OHRQoL construct validity results. Children with poor perceived general health from mothers' perceptions had higher child's OHRQoL score, indicating a worse QoL compared to children with a better rating of general health ($p=0.008$). Furthermore, the same pattern was found when correlating perceived oral health to child's OHRQoL, showing that children with worse perceived oral health had higher OHRQoL score, and therefore a worse QoL ($p = 0.007$). In terms of the overall extent to which the child's oral problems affected their QoL, findings revealed a clear gradient with worse children's OHRQoL (ie higher OH-QOLADS score) for each group of children whose mothers reported higher impact ($p < 0.001$).

Table 4-26. Association of child's OHRQoL with perceived health indicators (Ever-happened) N=97

Variable	N	Mean (SD)	Quartiles (25,50,75)	P-value*	r	P-value**
Perceived general health						
Very poor, Poor	5	14.2 (15.8)	3, 4, 26	0.029	0.269	0.008
Fair	23	10.9 (10.1)	4, 11, 14			
Good, Very good	69	6.4 (7.9)	1, 3, 9			
Perceived oral health						
Very poor, Poor	25	11.76 (11.8)	2, 10, 16	0.027	0.272	0.007
Fair	42	7.07 (7.0)	2, 4.5, 11			
Good, Very good	30	5.73 (8.8)	0, 2, 6			
Global child's OHRQoL rating						
Not at all	46	2.41 (3.2)	0, 1, 4	< 0.001	0.729	< 0.001
Very little	30	8.37 (5.7)	4, 7, 12			
Some	13	13.62 (9.5)	7, 11, 19			
A lot, Very much	8	28 (8.9)	22.5, 28.5, 35			

* Kruskal-Wallis Test

** Spearman test

Table 4.27 shows the findings of “Last-year” child’s OHRQoL construct validity. Children with poor perceived general health had higher OH-QOLADS indicating worse OHRQoL compared to children with better rating of general health, the gradient in the association was clear although the association was not statistically significant. The relationship between perceived oral health and child’s OHRQoL, showed that children with worse perceived oral health had higher OH-QOLADS, and therefore, worse OHRQoL ($p < 0.001$). In terms of the overall extent that the child’s oral problems affected their QoL, findings revealed that those who reported “a lot”, or very much” had the highest impacts on OHRQoL followed by those who reported “some” ($p < 0.001$).

Table 4-27. Association of child's OHRQoL with perceived health indicators (Last-year) N=97

Variable	N	Mean (SD)	Quartiles (25,50,75)	P- value*	r	P- value**
Perceived general health						
Very poor, Poor	5	13.4 (16.5)	1, 4, 26	0.200	0.178	0.081
Fair	23	6.22 (8.0)	0, 4, 11			
Good, Very good	69	3.83 (6.1)	0, 1, 6			
Perceived oral health						
Very poor, Poor	25	8.32 (10.3)	0, 5, 12	< 0.001	0.375	< 0.001
Fair	42	5.57 (7.1)	0, 3, 8			
Good, Very good	30	1.07 (1.8)	0, 0, 1			
Global child OHRQL rating						
Not at all	46	1.43 (2.7)	0, 0, 1	< 0.001	0.511	< 0.001
Very little	30	5.2 (5.0)	1, 4, 8			
Some	13	12.77 (9.5)	7, 11, 13			
A lot, Very much	8	10.75 (15.6)	0, 1.5, 23.5			

* Kruskal-Wallis Test

** Spearman test

c) Discriminant validity

An association between the “Ever-happened” category of the child’s OHRQoL and some clinical indicators was also analysed in order to assess the ability of the developed questionnaire to discriminate between different clinical groups (see Table 4.28). The relationship between child’s OHRQoL and decayed teeth was statistically significant ($p = 0.016$) showing that children with decayed teeth had in general higher OH-QOLADS score. The same pattern was observed when assessing the association between OHRQoL and pulpal-involved teeth, indicating that children with pulpal-involved teeth had higher OH-QOLADS score ($p = 0.007$), which means worse OHRQoL. The association between different malocclusion indicators and OHRQoL was not significant, and association between OHRQoL and the child having bad breath was also statistically not significant. However, the magnitude of the difference between groups indicates that the worse the oral condition the higher the impact on OHRQoL. As shown in the table below, periodontal status (both plaque and gingival indices) was significantly associated with higher OHRQoL ($p = 0.021$ and $p = 0.020$ respectively). The link between subjective experiences of toothache and the child’s OHRQoL was also assessed, and results showed a significant association; with those who experienced toothache had higher OHRQoL, and therefore worse QoL ($p < 0.001$). It should be noted as well that the differences were larger for more extreme groups (i.e. for those that refer to pulp involvement and those for toothache). It really makes sense to have bigger differences in those rather than on other clinical status such as malocclusion or gingival bleeding.

Table 4-28. Association of child's OHRQoL with some oral health indicators (Ever-happened) N=97

Variable	Number	Mean (SD)	Quartiles (25,50,75)	P-value
Caries				
Yes	69	8.46 (9.0)	2, 5, 12	0.016
No	28	6.39 (9.7)	0, 1.5, 8	
Pulpal involvement				
Yes	25	11.52 (10.8)	4, 8, 14	0.007
No	72	6.59 (8.2)	1, 3, 10	
Malocclusion				
<u>Over-jet</u>				
Normal	18	7.39 (9.1)	1, 4, 10	0.929
Deviated from normal	79	7.97 (9.2)	1, 5, 11	
<u>Over-bite</u>				
Normal	14	8.14 (9.7)	2, 6, 8	0.654
Deviated from normal	83	7.82 (9.1)	1, 4, 11	
<u>Posterior cross-bite</u>				
Present	71	7.93 (9.2)	2, 4, 11	0.876
Absent	26	7.69 (9.3)	1, 5, 11	
Plaque Index				
Mild	71	6.37 (7.4)	1, 3, 11	0.021
Moderate	26	11.96 (12.2)	2, 6.5, 16	
Gingival Index				
Mild	50	6.08 (7.6)	0, 3, 10	0.020
Moderate	47	9.76 (10.3)	2, 6, 13	
Bad breath				
Yes	35	8.40 (8.3)	2, 5, 11	0.277
No	62	7.56 (9.7)	1, 3, 11	
Toothache				
Yes	45	10.18 (9.1)	3, 7, 13	<0.001
No	52	5.86 (8.9)	0, 2, 8	

Table 4.29 shows the association between “Last-year” child’s OHRQoL and some clinical indicators to assess discriminant validity. Results were very similar to the findings in table 4.28 (association between “Ever-happened” child’s OHRQoL and the clinical indicators). The association between child’s OHRQoL and decayed and pulpal-involved teeth was highly significant ($p < 0.001$), showing that children with decayed teeth and pulpal-involved teeth had in general higher OHRQoL, indicating worse QoL. The association between different malocclusion indicators and OHRQoL was not significant, and the association between OHRQoL and child having bad breath was also statistically not significant, however the magnitude of difference between groups indicated that bad breath had negative impact on the child’s OHRQoL. Plaque and gingival indices were significantly associated with higher OHRQoL ($p = 0.013$ and $p = 0.015$ respectively). There was a significant association between subjective experiences of toothache and child’s OHRQoL, meaning that those who reported experiencing toothache had higher OHRQoL, and therefore, worse QoL ($p < 0.001$).

Table 4-29. Association of child's OHRQoL with some oral health indicators (Last-year) N=97

Variable	Number	Mean (SD)	Quartiles (25,50,75)	P-value
Caries				
Yes	69	6.15 (8.1)	0, 3, 10	< 0.001
No	28	1.75 (4.9)	0, 0, 1	
Pulpal involvement				
Yes	25	8.84 (9.6)	2, 6, 12	< 0.001
No	72	3.51 (6.2)	0, 1, 5	
Malocclusion				
<u>Over-jet</u>				
Normal	18	4.44 (8.8)	0, 0.5, 8	0.355
Deviated from normal	79	4.98 (7.3)	0, 2, 7	
<u>Over-bite</u>				
Normal	14	5.71 (9.6)	0, 1.5, 8	0.775
Deviated from normal	83	4.75 (7.2)	0, 1, 7	
<u>Posterior cross-bite</u>				
Present	71	4.20 (6.5)	0, 1, 6	0.545
Absent	26	6.77 (9.7)	0, 1.5, 11	
Plaque Index				
Mild	71	3.39 (5.1)	0, 1, 5	0.013
Moderate	26	8.96 (11.1)	0, 6, 13	
Gingival Index				
Mild	50	3.06 (5.0)	0, 1, 5	0.015
Moderate	47	6.83 (9.2)	0, 3, 11	
Bad breath				
Yes	35	6.14 (8.2)	0, 2, 11	0.125
No	62	4.18 (7.1)	0, 1, 6	
Toothache				
Yes	45	8.76 (9.1)	2, 7, 11	< 0.001
No	52	1.54 (3.4)	0, 0, 1	

Family's OHRQoL

This section presents the construct and discriminant validity of the impact of child's oral health on family's QoL (both Ever-happened and Last-year). Table 4.30 shows the results of Spearman's r correlation between "Ever-happened" family's QoL and mothers' perceptions of the child's perceived general health; perceived oral health; and overall extent that oral problems affected family's QoL. As shown in the Table, children with poor perceived general health had higher impacts on family's QoL compared to children with better rating of general health ($p = 0.006$). Results also showed a statistically significant association between perceived oral health and family's QoL, showing that children with worse perceived oral health had worse impact on family's QoL ($p < 0.001$). In terms of the overall extent that the child's oral problems affected family's QoL, findings revealed that mothers who thought that their children's oral health problems affected the overall family's QoL "a lot, or very much" had the highest impacts on family's OHRQoL followed by those who reported "some" ($p < 0.001$). The associations are characterised by a graded pattern with worse family's QoL (i.e. higher OH-QOLADS score) for each group of children whose mothers reported worse perceived health indicators.

Table 4-30. Association of family's OHRQoL with child's subjective health indicators (Ever-happened) N=97

Variable	N	Mean (SD)	Quartiles (25,50,75)	P-value*	r	P-value**
Perceived general health						
Very poor, Poor	5	12.0 (5.2)	9, 14, 15	0.016	0.279	0.006
Fair	23	8.04 (5.9)	4, 7, 11			
Good, Very good	69	5.42 (5.7)	0, 4, 9			
Perceived oral health						
Very poor, Poor	25	9.2 (6.0)	4, 9, 14	0.005	0.331	< 0.001
Fair	42	5.88 (4.7)	2, 6, 9			
Good, Very good	30	4.73 (6.7)	0, 2, 6			
Global family OHRQoL rating						
Not at all	34	1.59 (2.7)	0, 0, 2	<0.001	0.750	< 0.001
Very little	17	5.53 (3.9)	2, 6, 7			
Some	22	7.64 (4.4)	4, 8, 10			
A lot, Very much	24	12.63 (5.6)	8.5, 13, 17.5			

* Kruskal-Wallis Test

** Spearman test

Table 4.31 shows the findings of “Last-year” family’s OHRQoL construct validity. Mothers of children with poor perceived general health had higher impacts on family’s OHRQoL indicating worse QoL compared to children with better rating of general health ($p = 0.017$). Similar relationship was found between perceived oral health and family’s OHRQoL that showed children with worse perceived oral health had higher family’s OHRQoL ($p < 0.001$). In terms of the overall extent that child’s oral problems affected family’s QoL, findings revealed that those who are in “a lot, very much” category had the highest family’s OHRQoL followed by those who reported “some” ($p < 0.001$).

Table 4-31. Association of family's OHRQoL with child's subjective health indicators (Last-year) N=97

Variable	N	Mean (SD)	Quartiles (25,50,75)	P-value*	r	P-value**
Perceived general health						
Very poor, Poor	5	10.2 (4.4)	9, 9, 14	0.021	-0.242	0.017
Fair	23	5.91 (5.7)	0, 5, 9			
Good, Very good	69	3.87 (4.8)	0, 2, 7			
Perceived oral health						
Very poor, Poor	25	7.64 (5.3)	4, 9, 12	< 0.001	-0.419	< 0.001
Fair	42	4.62 (4.8)	0, 3.5, 9			
Good, Very good	30	2.3 (4.5)	0, 0, 3			
Global family OHRQoL rating						
Not at all	34	1.26 (2.6)	0, 0, 1	< 0.001	0.584	< 0.001
Very little	17	3.59 (4.0)	0, 2, 6			
Some	22	6.73 (4.7)	2, 5.5, 10			
A lot, Very much	24	8.41 (5.9)	4, 9, 13			

* Kruskal-Wallis Test

** Spearman test

In order to assess the discriminant validity of the family's OHRQoL, association between "Ever-happened" family's OHRQoL and some clinical indicators was analysed, Table 4.32. The relationship between family's OHRQoL and decayed teeth was not significant ($p = 0.098$) but results showed that children with decayed teeth had in general higher family's OHRQoL indicating worse QoL. Mothers of children with pulpal-involved teeth had higher family's OHRQoL scores, which means worse impact of the family's QoL ($p = 0.044$). There was no significant association between family's OHRQoL and different malocclusion indicators and subjective reports of bad breath. Periodontal status was significantly associated with higher impacts on family's QoL (plaque index $p = 0.002$, and gingival index $p = 0.014$). Association between subjective experience of toothache and family's QoL was also assessed, and results showed a significant association with those who experienced toothache had higher impacts on the family's OHRQoL ($p = 0.004$).

Table 4-32. Association of family's OHRQoL with child's oral health indicators (Ever-happened) N=97

Variable	Number	Mean (SD)	Quartiles (25,50,75)	P-value
Caries				
Yes	69	6.81 (5.7)	2, 6, 10	0.098
No	28	5.32 (6.4)	0, 3.5, 9	
Pulpal involvement				
Yes	25	7.92 (5.4)	4, 7, 11	0.044
No	72	5.85 (6.0)	0, 4, 9	
Malocclusion				
<u>Over-jet</u>				
Normal	18	6.44 (6.9)	0, 5, 13	0.640
Deviated from normal	79	6.37 (5.7)	2, 5, 9	
<u>Over-bite</u>				
Normal	14	6.78 (7.6)	0, 2.5, 15	0.668
Deviated from normal	83	6.31 (5.6)	2, 6, 9	
<u>Posterior cross-bite</u>				
Present	71	6.17 (5.9)	1, 5, 9	0.611
Absent	26	6.96 (6.1)	0, 6, 12	
Plaque Index				
Mild	71	5.21 (5.4)	0, 4, 9	0.002
Moderate	26	9.58 (6.3)	5, 9.5, 14	
Gingival Index				
Mild	50	5.14 (5.8)	0, 2.5, 9	0.014
Moderate	47	7.70 (5.8)	3, 6, 11	
Bad breath				
Yes	35	6.40 (5.0)	2, 6, 10	0.513
No	62	6.37 (6.4)	0, 5, 9	
Toothache				
Yes	45	7.73 (5.2)	3, 8, 11	0.004
No	52	5.21 (6.3)	0, 3.5, 7.5	

Discriminant validity of the “Last-year” family’s OHRQoL and some clinical indicators of child’s oral health are shown in Table 4.33. The relationship between decayed teeth and family’s OHRQoL was not significant ($p = 0.104$) although, the magnitude of difference between group categories indicates that the presence of dental caries associated with higher impacts on family’s OHRQoL. The relationship between family’s OHRQoL, and pulpal-involved teeth was significant ($p = 0.004$) showing that children with pulpal-involved teeth had in general higher impact on their family’s OHRQoL. The association between different malocclusion indicators and subjective reports of bad breath was not significant. Plaque and gingival indices were significantly associated with higher impacts on family’s OHRQoL ($p = 0.006$ and $p = 0.058$ respectively). There was a significant association between subjective experience of toothache and family’s OHRQoL with those who reported experienced toothache had higher impacts on family’s OHRQoL, and therefore, worse QoL ($p < 0.001$).

Table 4-33. Association of family's OHRQoL with child's oral health indicators (Last-year) N=97

Variable	Number	Mean (SD)	Quartiles (25,50,75)	P-value
Caries				
Yes	69	5.14 (5.3)	0, 4, 9	0.104
No	28	3.54 (5.0)	0, 1, 5.5	
Pulpal involvement				
Yes	25	7.12 (5.4)	2, 7, 11	0.004
No	72	3.83 (4.9)	0, 2, 8	
Malocclusion				
<u>Over-jet</u>				
Normal	18	4.33 (6.1)	0, 0, 9	0.292
Deviated from normal	79	4.76 (5.0)	0, 3, 9	
<u>Over-bite</u>				
Normal	14	5.00 (6.5)	0, 0, 12	0.669
Deviated from normal	83	4.63 (5.0)	0, 3, 9	
<u>Posterior cross-bite</u>				
Present	71	4.34 (4.7)	0, 3, 8	0.648
Absent	26	5.62 (6.3)	0, 3, 11	
Plaque Index				
Mild	71	3.76 (4.7)	0, 2, 7	0.006
Moderate	26	7.19 (5.7)	1, 8, 11	
Gingival Index				
Mild	50	3.72 (4.8)	0, 2, 8	0.058
Moderate	47	5.70 (5.5)	0, 5, 10	
Bad breath				
Yes	35	5.51 (5.0)	0, 4, 9	0.119
No	62	4.21 (5.3)	0, 2, 8	
Toothache				
Yes	45	6.44 (5.4)	2, 7, 10	< 0.001
No	52	3.15 (4.6)	0, 0.5, 4.5	

4.3.5.3 Association between child and family's QoL

Tables 4.34 and 4.35 present the correlations between child and family's QoL for "Ever-happened" for "Last-year" respectively. The correlation was assessed using family's OHRQoL with both global child's QoL and child's OHRQoL ratings. Results showed a significant association between family's OHRQoL and global child's QoL rating with those who reported that child's oral health had "a lot, very much" impact on the child's QoL had the highest impacts on family's QoL as well followed by those who reported "some" ($p < 0.001$). There was also a significant correlation between family and child's OHRQoL ($p < 0.001$).

Table 4-34. Association of family's OHRQoL with child's global rating of QoL and child's OHRQoL (Ever-happened)

Variable	N	Mean (SD)	Quartiles (25,50,75)	P-value	r	P-value***
Global child's QoL rating						
Not at all	46	2.19 (2.8)	0, 1, 4	< 0.001*	0.739	< 0.001
Very little	30	8.83 (5.1)	5, 7.5, 12			
Some	13	9.92 (5.3)	8, 9, 14			
A lot, Very much	8	15.5 (17.7)	12.5, 16, 18.5			
Child's OHRQoL						
No	17	2.29 (3.4)	0, 0, 5	< 0.001**	0.359	< 0.001
Yes	80	7.25 (6.0)	2, 6, 11			

* Kruskal-Wallis Test

** Mann-Whitney test

***Spearman test

Table 4-35. Association of family's OHRQoL with child's global rating of QoL and child's OHRQoL (Last-year)

Variable	N	Mean (SD)	Quartiles (25,50,75)	P-value	r	P-value***
Global child's QoL rating						
Not at all	46	1.78 (2.8)	0, 0, 3	< 0.001*	0.562	< 0.001
Very little	30	6.17 (5.3)	2, 5, 10			
Some	13	8.77 (4.4)	8, 9, 10			
A lot, Very much	8	9.13 (7.4)	1, 11, 15.5			
Child's OHRQoL						
No	38	1.55 (2.9)	0, 0, 2	< 0.001**	0.537	< 0.001
Yes	59	6.69 (5.4)	2, 6, 10			

* Kruskal-Wallis Test

** Mann-Whitney test

***Spearman test

In summary, the findings of the phase-two study showed satisfactory levels of reliability and validity of the developed questionnaire, OH-QOLADS. Cronbach's Alpha of the item-total correlation of the child's OHRQoL was 0.909 for 'Ever-happened', and 0.902 for 'Last-year'. And for the family's OHRQoL, the Cronbach's Alpha ranged from 0.828 'Ever-happened' to 0.807 for impacts experienced 'Last-year'. For construct validity, findings revealed significant correlations between subjective health indicators and child and family's OHRQoL, with clear gradient in the association indicating that the worse the subjective health indicator the higher the OH-QOLADS. The new measure also shows its ability to discriminate between different clinical groups. The findings showed that the more severe the oral problem (i.e. pulpal involvement, toothache) the worse the reports on OH-QOLADS.

The results logically indicated that the prevalence of reported impacts on children and families OHRQoL was higher in the 'Ever-happened' compared to 'last-year'. This shows the ability of the questionnaire to detect impacts experienced at different

time periods. It appears that pain is the most reported impact on children's OHRQoL, followed by impacts on functional aspects of the child's life (i.e. eating). With regards, to family's OHRQoL, the most affected aspects of the family's life was emotional aspects especially the feeling of depression or distress. The results also showed significant correlations between child and family's OHRQoL. The following chapter will discuss these findings with the relevant literature.

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Chapter 5 DISCUSSION

5.1 Summary of key findings

The Phase One study aimed to explore mothers' views concerning the oral health status of their children/adolescents with Down Syndrome, and how their oral health affected their QoL and that of the whole family. The interviews showed that mothers in general gave lower priority to oral health compared to the general health status of their children. They linked the occurrence of dental disease mainly to the presence of dental caries and/or toothache. It was also difficult for mothers to spontaneously respond to the questions of how the oral health of their children impacted on their QoL and that of the family. However, after probing on the potential different impacts of oral health, they revealed an array of impacts that oral health had on the child as well as family's QoL.

A new OHRQoL questionnaire, the OH-QOLADS, was developed after analysing data from the mothers' interviews along with the review of literature on children's OHRQoL and the literature on the oral health of children and adults with Down Syndrome. This ensured that items included were relevant for children with Down Syndrome, as the main concerns reported by their mothers were reflected in the questionnaire. The results of the pilot testing revealed the feasibility of the developed questionnaire and the planned procedure of clinical data collection.

The Phase Two study aimed at testing the developed questionnaire to assess its psychometric properties and the findings revealed overall satisfactory levels of reliability and validity (see section 4.3.6). The Cronbach's Alpha of the item-total correlation of the child's OHRQoL was 0.909 for 'Ever-happened', and 0.902 for 'Last-year'. For the family's OHRQoL, the Cronbach's Alpha ranged from 0.828 'Ever-happened' to 0.807 for impacts experienced 'Last-year'. For construct validity (see section 4.3.6.2), findings revealed significant correlations between perceived health indicators and child and family's OHRQoL, indicating that the poorer the perceived health indicators the higher the impacts on child's, as well as the family's OHRQoL (see tables 4.26, 4.27, 4.30, and 4.31).

Results also showed the ability of the questionnaire to discriminate between different groups with those experiencing poorer oral health (in terms of dental caries, pulpal involvement, plaque and gingival health) also having higher OH-QOLADS scores indicating worse QoL. Similar findings were also found in the family section of OHRQoL, indicating that poorer child's oral health was also associated with increased impact on family's life (see tables 4.28, 4.29, 4.32, and 4.33).

Findings of Phase Two revealed that children with oral-related problems and/or conditions tended to have poorer OHRQoL compared to children with no such problems according to the mothers' proxy reports. The most frequently reported oral impact was dental pain in which more than three quarters of mothers reported its occurrence at some point of the child's life (76.3%) and the majority of mothers thought that its severity was either moderate (29.9%) or severe (28.9%). The second most affected aspect of the child's life was related to difficulties eating due to oral problems. Emotional impacts were also reported as aspects affected by child's oral health. For the impact of the child's oral conditions on the QoL of the family, results showed a high prevalence of emotional impacts with feelings of depression or distress experienced by almost two-third of participated mothers.

5.2 Discussion of the findings with relevant literature

Findings of both phases are discussed in this section, and divided into four main parts; child's oral health, child's OHRQoL, family's OHRQoL, and results of the validation process of the newly developed OHRQoL questionnaire, OH-QOLADS.

5.2.1 Child with Down Syndrome oral health conditions

There were no specific oral-related problems reported by mothers. This is in line with the literature that indicates individuals with Down Syndrome do not have unique oral health problems (Hennequin et al., 1999). However, the severity of dental problems and also their responses to dental diseases might differ. For example the response to pain sensitivity among individuals with Down Syndrome is different than the general population where studies have revealed the delay in response to painful stimulus but confirmed the fact that individuals with Down Syndrome experience pain in the same way as the mainstream population (Hennequin et al., 2000). Mothers' reports along with clinical findings indicated the high prevalence of carious teeth as well as pulpal-involved teeth and that differs from the published literature on levels of dental caries among individuals with Down Syndrome, which indicates usually lower levels compared to mainstream populations (Hennequin et al., 1999). These contrasting results may be because this study was conducted in a country with high levels of dental caries (Marghalani et al., 2014). The levels of dental caries among the studied sample were comparable to mainstream children in Saudi Arabia. A recent systematic review of the prevalence, severity, and trends of dental caries among various Saudi populations indicated that approximately 91% had caries; the highest DMFT value was 7.35 among the mainstream children/adolescents ages 12-19 years (Al-Ansari, 2014). Therefore, the problem of dental caries among this sample of children was different to individuals with Down Syndrome internationally and in Western countries in particular, but comparable to the caries levels among mainstream children in Saudi Arabia. Results also showed that the mean of carious teeth was much higher than filled teeth. This is consistent

with the literature that showed that among people with intellectual disabilities, the number of filled and treated teeth was lower compared to general populations (Cumella et al., 2000; Hogan & White, 1982; Hinchcliffe et al., 1988; Shaw et al., 1990).

Some interviewed mothers reported the problem of periodontal diseases. They mentioned “bleeding gums” or “pain on the gums”. This was also supported by the findings of the quantitative phase of this study where high proportions of mothers reported periodontal bleeding. The clinical examination also revealed moderate levels of plaque and gingival indices in the majority of the sample examined. Individuals with Down Syndrome are at higher risk of developing periodontal disease than the general population (Hennequin et al., 1999). The cause of the high prevalence of periodontal disease among those with Down Syndrome is complex (Lopez-Perez et al., 2002) but interaction between many factors (systemic health, oral hygiene, oro-facial characteristics, along with the effect of wider environmental factors) might contribute to their elevated risk (Garcia et al., 2001).

Prevalence of malocclusion is also high among people with Down Syndrome; interaction between dental as well as general health factors contribute to this problem. For example, oral factors such as underdevelopment of maxillary arch, muscles hypotonia, missing teeth (hypodontia), and enlarged tongue all collectively contribute to this elevated risk of malocclusion (Backman et al., 2007; Oliveira et al., 2010b; Quintanilla et al., 2002). Variations in both vertical and transversal occlusions, identified mainly as anterior open bite, anteroposterior crossbite, and proclination of the anterior teeth also led to the malocclusion among individuals with Down Syndrome (Hennequin et al., 2000; Quintanilha et al., 2002; Venail et al., 2004). In the present study, mothers reported malocclusion as a problem, and clinical findings were in line with the literature showing that majority of children examined had abnormal overjet, overbite, and posterior crossbite. It is important to highlight the consequences of undesired occlusion as this might contribute to other oral problems such as periodontal diseases, and in severe cases might interfere with the child’s ability to speak properly. Malocclusion also played an important role in negatively affecting QoL and wellbeing on mainstream children (Liu et al., 2009; Zhang et al., 2006), and the case might be similar among people with Down

Syndrome.

Drooling can impose a significant disability on individuals and it has negative health and social consequences. In addition to the cosmetic effect that might interfere with psychosocial, physical and educational consequences, drooling can impair swallowing and masticatory function, and can produce peri-oral infections (Hedge et al., 2008; Kohler et al., 1984; Myer, 1989). Some interviewed mothers reported drooling as a problem. However, the prevalence was very low in the quantitative phase. This could be partly due to the age of children included in the study (12 – 18 years old). Usually at this age, drooling reduces dramatically depending on the physiotherapy and training the child received at a younger age, which help in increasing the tonicity of facial muscles, and therefore reduction in such condition. In contrast, its prevalence in the whole population of children with Down Syndrome might be expected to be higher. Many children in the present study had used special services at some point in their lives and therefore, were probably exposed to some interventions at an early age that targeted conditions such as drooling or tongue thrusting. This highlights the importance of early interventions, and the role of physiotherapy that might lead to an improvement in the health status of the child and therefore, the overall QoL and wellbeing. Collaborations between health professions should be the strategy to use in all special care schools and centres.

Another oral-related condition reported by mothers was difficulty speaking. But this might not be a typical oral health problem. Most children with Down Syndrome have difficulty speaking clearly (Buckley, 1993). The delayed language and speech development is a common condition among children with Down Syndrome (Rondal, 1988). This could be a result of their cognitive delay, hearing and visual defects, and motor delay as research in this area has indicated that difficulty with speaking among individuals with Down Syndrome is not usually a result of oral-related problems, however, some severe cases of malocclusion and/or oral muscle hypo-tonicity (such as tongue) could interfere with clarity of speech.

One of the main findings of mothers' interviews was their perception of their children's oral health, giving a lower priority to oral health compared to child's general health. The majority thought that their children had a healthy mouth.

Although mothers did not prioritize oral health, the findings of the validation study showed that the majority of mothers rated their children's oral health lower in importance than general health. This could be an indication of either: a) lack of awareness of the important role that oral health plays on the general health as well as wellbeing and life satisfaction, or b) that mothers did not give much concern to their children's oral health while they knew that the current oral health status may not be as good as it should be. The latter suggests that parents may not act to address an issue (which is in this case oral health related problems) because of competing priorities (such as general health problems, or social aspects of the child's life). This agrees with other studies that concluded oral health has a lower priority in the context of other social and medical challenges faced by individuals with disabilities (BSDOH, 2012).

The qualitative data in this study revealed that most mothers linked the presence or absence of oral problems with the existence of dental caries and toothache; this is comparable to the findings of previous research amongst mothers of children with Down Syndrome (Oliveira et al., 2010). This might be the part of the reason behind their pattern of dental visits in which results from the quantitative phase showed that around 15% of the sample have never been to see a dentist and almost two third of those who visited a dentist were seeking dental treatment indicating a treatment but not prevention oriented behavior (regular check-ups). So, the level of awareness on the importance of oral health might be a contributing factor to reduced dental attendance behaviours.

Dental attendance behavior may not rely only on the carers/patient's awareness or disease experience; there are other reasons that might act as barriers to dental care services utilization such as barriers related to availability, accessibility, acceptability, and quality (BSDOH, 2012). This can be seen in the qualitative phase where mothers reported some reasons for not accessing health services such as long waiting list in governmental hospitals, or quality of services provided. Difficulty in finding a dentist who is willing to treat children with Down Syndrome was also a problem reported by almost half of the participants. This is similar to the findings of a cross-sectional study of 119 individuals with disabilities in Saudi Arabia that reported half of those with disabilities faced difficulty accessing dental care (Al-Shehri, 2012). A

cross-sectional study in France revealed similar findings in which parents of children with Down Syndrome reported difficulties in findings both medical and dental services for their children with Down Syndrome compared to their non-Down Syndrome siblings (Allison et al., 2000). Other barriers to accessing dental care for those with disabilities in Saudi Arabia were reported to be: fear of dentists and/or cost of dental treatment (Al-Shehri, 2012). International studies assessing barriers to dental services revealed that the unmet dental needs among individuals with disabilities related to various barriers that can be broadly classified into factors related to patients and/or their carers, factors related to dental care services and professional service providers, physical barriers to accessing dental care, and cultural issues (BSDOH, 2012). Factors related to patients can be low expectations, fear of dental treatment (Band, 1997), lack of awareness among carers and/or financial barrier (Schultz et al., 2001). With regard to factors related to care services provisions include for example dentists' lack of time and/ or domiciliary equipment (Cumella et al., 2000; Edwards & Merry, 2002). A study aimed at assessing the views and experiences of parents and siblings of adults with Down Syndrome to oral health, using a combined qualitative and quantitative study demonstrated that the parents/carers highlighted a need for appropriate and timely oral health information early in their child's life, and access to dentists who were both experienced and sympathetic with good communication skills and good knowledge of Down Syndrome. The study also demonstrated a strong association between parental oral health beliefs and the dental care the person with Down Syndrome received (Kaye et al., 2005). Therefore it is important to assess the barriers to dental services especially among individuals with special needs as their risk to oral health problems and unmet treatment needs is increased.

5.2.2 Child's OHRQoL

To our knowledge, this is the first study to develop and test an OHRQoL measure for children/adolescents with Down Syndrome. Two studies are reported which assessed OHRQoL in individuals with Down Syndrome. One used an existing validated measure (OHIP-14) that was developed and tested for use amongst an adult general population (Amaral-Loureiro et al., 2007). The second study did not use a validated

OHRQoL measure but assessed OHRQoL using broad, in-depth interviews with mothers (Oliveira et al., 2010a). Although both studies showed that the oral health of children with Down Syndrome had a negative impact on their QoL, a more comprehensive and appropriate assessment is needed in order to provide valid and reliable results. Since both studies did not validate the approaches they used to fit the specificities of individuals with Down Syndrome, this might have a different impact, which might not be captured by the approaches they used.

The primary aim of this study was to develop and test an OHRQoL measure among adolescents with Down Syndrome. The measure was developed in two phases; an initial phase rooted in the literature review and semi-structured, in-depth interviews with mothers of 12-18 years old children with Down Syndrome that informed the identification and selection of themes and sub-themes that were used in the development process of the questionnaire. The following quantitative phase provided initial evidence on the psychometric properties of the new measure.

The findings of this study show that children with Down Syndrome do not have unique oral health problems and therefore it might not be necessary to develop a specific OHRQoL measure for them. However, they exhibit some specific conditions that might have impacts. Conditions such as drooling, protruded tongue, difficulty speaking, in addition to the presence of the disability of being a child with Down Syndrome. Studies revealed that chronic illnesses including developmental conditions could have a profound influence on various aspects of the development because it changes the developmental trajectory. A review of the literature on young people's experiences of living with chronic illness indicated that chronic illness affected various aspects of daily living, and therefore is likely to impinge on their QoL (Taylor et al., 2008a). In the review it was suggested that attributes of HRQoL from mainstream population are not sufficiently related to the developmentally important aspects of life for young people living with chronic illnesses. For example, certain aspects of living with an illness depend on the stage of adolescence, such as attitudes to the illness and strategy they used to make themselves more acceptable. Therefore, HRQoL in young people with chronic illness was defined as "a subjective, multidimensional and dynamic. It is unique to each individual young person and includes aspects of physical, psychological and social function. It is

dependent upon not only the stage of development but also the illness trajectory. This involves the achievement of goals and aspirations and the constraints imposed through ill health and treatment” (Taylor et al., 2008b). This is very important aspect to consider when assessing OHRQoL of individuals with disabilities or chronic conditions.

Although interest in OHRQoL started almost three decades ago, there is a lack of conceptual understanding and comprehensive evaluation of the oral impacts on different aspects of the life of individuals with disabilities. So, in OHRQoL research, careful attention should also be given to the fact that attributes of OHRQoL for people with chronic disabilities might differ than those of the mainstream population, and therefore more careful assessment should be carried out to make sure that any negative impact on different aspects of QoL is actually a result of the oral-related conditions and not due to the presence of disability by itself, discrimination, or other factors.

The lower priority of oral health, which has been reflected on mothers’ views, might lead to difficulty of mothers realising and spontaneously reporting the impact of their children’s oral health on their QoL. This is a very important finding, which showed how mothers’ concerns were mainly directed toward the child’s general health and their disability. This might affect mothers’ reports on their children’s OHRQoL compared to mothers of mainstream children, and therefore, using a generic OHRQoL developed for use among mainstream children might not be suitable assuming that their OHRQoL measures might not be sensitive enough to detect the actual impact of the child’s oral health. As shown in the results of Phase One interviews, mothers could not answer the section of OHRQoL easily. They could not easily see how their children’s oral health impacts on the child’s and/or family’s QoL, however, further probing revealed that child’s oral health resulted in a considerable and wide range of impacts.

Experience of physical pain in turn resulted in many negative consequences on different aspects of the child’s life. For example, the child’s experience of toothache resulted in a reduction of the child’s functioning ability such as eating and/or performing at school. Child’s emotional wellbeing was also affected by the

experience of dental pain. The child's social life was affected as well, for example when the child experienced dental pain; he/she preferred to be isolated from their friends and other family members, according to their mothers' reports. These findings were supported by the results of the quantitative phase where high number of mothers reported the experience of dental pain among their children (76.3% ever happened, and 50.5% experienced dental pain last year). The clinical examination in phase two revealed a high prevalence of dental caries and pulp-involved teeth and this was associated with the high reports of dental pain when the majority of mothers reported dental pain at moderate or severe levels. Another oral health related problem that resulted in many undesirable impacts on the child's QoL was problems with speaking. Mothers thought that child's inability to speak clearly resulted in many negative impacts on emotional, social, as well as behavioural aspects of child's life.

Impacts of oral health on individuals' daily activities have been the research interest for many years, and in the literature there are specific measures aimed at assessing that, such as Oral Impact on Daily Performance among adults (OIDP) (Adulyanon & Sheiham, 1996), and children (Child-OIDP) (Gherunpong et al., 2004). Oral problems are believed to impede the people's daily activities, such as their eating, speaking and even their schooling and/or working hours, in mainstream populations. The results from the mothers' interviews also revealed how the child's oral health affects different daily activities. The most reported activity that was affected negatively by child's oral health was eating. Tooth brushing, sleeping pattern, and playing were also reported by mothers to be affected negatively by the child's oral health. The results also showed that the child's oral health status affected the child's speaking ability, however it is difficult to confirm that the impact of the reduced speaking ability among children with Down Syndrome was purely a result of dental-related problem. The literature on language and speaking among people with Down Syndrome is large, and speaking deficiency experienced by them is a result of many factors (Buckley, 1993; Martin et al., 2009; Rondal, 1988). However, the results from the mothers' interviews suggest that a child's oral health might impact on his/her speaking ability in that the experience of dental pain reduced the child's desire to speak. Another activity that was reported by mothers to be affected by the child's oral health was schooling, and this is consistent with the literature that oral

health problems resulted in loss of working hours and reduction in school performance (Jackson et al., 2011; Blumenshine et al., 2008).

The emotional impact of oral health problems has been reported in previous studies among mainstream populations (Jokovic et al., 2002; Locker, 1997). Results showed that some oral disorders and/or conditions contributed to a wide range of emotional impact such as crying, embarrassment, and feeling angry. The main reason behind that were experiences of dental pain and/or difficulty speaking. The present validation study confirmed mothers' perceptions reported in Phase One where the results revealed high prevalence of some emotional impacts. Crying, being quiet, and stop laughing were all emotional responses reported by mothers and that occurred as a result of experiencing pain. Studies revealed that the emotional expression of pain could occur amongst individuals who are facing difficulties in expressing their feelings, such as those with intellectual disabilities, and experience of pain can alter behaviour (McGrath, 1993; Radovich et al., 1991). Mothers' reports on OHRQoL confirmed the negative emotional impacts of oral health of their children.

In the qualitative phase where mothers were asked to talk about how their children's oral health affected their QoL, a mother said that her child's behaviour changed as a result of his difficulty in speaking. She explained this as 'stubbornness behaviour'. In the quantitative phase, a few mothers also reported the effect of oral problems in inducing stubbornness behaviour; however, it is difficult to confirm that this impact was actually resulted from oral problems as mothers thought since adolescence age is always accompanied by stubbornness behaviour especially among children with Down Syndrome (Pueschel et al., 1991).

The interviews with mothers as well as their quantitative reports highlighted the negative social impacts that their child's oral health had caused. The most prevalent social impact was teasing/bullying by others that expectedly leads to social isolation. As previous research has shown, social participation and interaction is a strong predictor of satisfaction and QoL (Yanos et al., 2001). However, some of the strategies that parents of children with disabilities used to accommodate to their children's needs are restriction of their social life and making some changes in family routines (Seltzer et al., 2001). This approach was also found in the present

study where mothers were asked about how they coped with having a child with disability. This should be kept in consideration when analysing data about the impact of oral health on the social aspects of QoL since impact of reduced social interaction could be a result of disability and its consequences but not due to oral-related problems per se. Stigma associated with illness and disability often negatively affects the QoL of both individual with disability and parents/caregivers (Green, 2003; Green et al., 2005). In the present study the data indicated that many mothers attributed the experience of social isolation as an impact of the child's oral health, and in some cases they isolated their child to avoid being stigmatised by other people because of oral problems such as drooling, or appearance of protruded tongue. However, it is difficult to confirm the main reason of social isolation since the pre-existence of disability might play a role in the social isolation.

In summary, the findings of the impacts of child's oral health on their QoL were consistent with studies aimed to assess OHRQoL among mainstream child populations (Jokovic et al., 2002; Gherunpong et al., 2004; Jokovic et al. 2004; Patel et al. 2007; Broder et al. 2007; Huntington et al., 2011) showing that a child's oral health has a significant effect on physiological (pain), functional, and psychosocial aspects of child's life, as those with poor oral health experienced decreased QoL. In addition, the findings of this study showed that the child's oral health, specifically a reduced speaking ability, which is very common condition among individuals with Down Syndrome and those with intellectual disabilities in general, might affect the behavioral aspects of his/her life. Previous studies among non-intellectually disabled individuals did not reveal such a finding, and this might be because previous children's OHRQoL measures have not focused on the potential behavioral impacts of child's OHRQoL. However, the behavioral changes might be related to other reasons such as the developmental age (teenage) that is usually accompanied by behavioral and emotional disturbances (e.g., stubbornness). The results also showed that oral health problems led to more than one impact. Different levels of impacts were also noted; for example, one consequence of oral diseases such as dental pain led to other consequences such as social isolation or reduced daily activities. However, it is not easy to assess the hierarchical impacts of oral health from this data since the study did not aim at assessing this finding but exploring the different impacts of oral health of children with Down Syndrome on their lives and that of

their families. Further studies are needed to investigate this longitudinally and how preventing one impact could result in a substantial decrease on other undesirable impacts, and therefore improve the OHRQoL outcomes.

5.2.3 Impacts of the child's oral health on family's QoL

Impact of child's oral health on some aspects of family life has been reported in the literature since early 1980s (Sheiham & Croog, 1981). However, in the field of disability and oral health and how individuals with disabilities' oral health status impacts on their families, data are lacking. In relation to the QoL of the family, it is not easy to distinguish if the negative impacts on family's QoL occurred as a result of disability by itself, general health, or child's oral health status. In the current study, the interviewer asked all informants about the reasons behind each impact on family life to assure herself (mother) that all reported impacts were a result of child's oral health, and to rule out or minimise impacts attributed to other reasons.

Studies on the impacts of child's general health and family's QoL of children with disabilities showed the extent and severity of such impacts among family members (Buzatto & Beresin, 2008; Hatzmann et al., 2008; Hedov et al., 2000; Murphy et al., 2000; Marchal et al., 2013; Oliveira & Limongi, 2011). This was similar to the findings in this study in which mothers reported high prevalence of negative impacts on family as a result of some oral-related conditions such as experience of dental pain. Findings from the quantitative phase showed high reports of depression or distress, feeling of self-blame and guilt, as well as worrying about the child's oral conditions.

Family activities were also affected by the child's oral health according to mothers' reports. The results from the mothers' interviews showed that their children's oral health had a considerable impact and the majority of mothers reported that the child required more time and attention in case he/she experienced oral problems. However, the quantitative phase showed that disruption of sleeping patterns as a result of child's oral problems was the most reported family impact. Impacts on the family paid work was not highly reported and this might occur due to the fact that only mothers were interviewed while the working member of the family was the father, or

might be because the impact of child's oral health on parent's job was not strong enough to be noted by mothers. It should be acknowledged that interviewing mothers only might have resulted in underreporting the actual impacts of their child's oral health on the family's employment status and therefore, the respective results should be interpreted with caution.

Finally, the third aspect of family's life that was affected by the child's oral health was family conflict. Although it was not common across interviews, some mothers reported the fact that they might argue with other family members if they needed to go for a dental appointment. This is because women in Saudi are dependent on males to take them out since they do not drive. It is not possible to generalize this finding to other children with disabilities, unless they are resident in Saudi Arabia. Child being jealous, arguing or blaming parents because of oral health problem did not appear in this analysis.

Other impacts on aspects of family's QoL could be present as well, but were not pointed out by phase one informants. For example, previous studies of family OHRQoL scales reported that the child's oral health had negative impacts on family finance. This item was not found in phase one interviews, and that may be because; 1- mothers were not the responsible member in the family for its finance; 2- they were unaware of potential impact of oral health on the finance of the family; 3- they did not use private dental clinics, and therefore never thought of its financial impacts. Further research is needed to re-define items of family OHRQoL of individuals with Down Syndrome.

In summary, the findings were in line with other studies of the impact of child's oral health on the family's life amongst mainstream children (Locker et al., 2002; Pahel et al., 2007), however, it seems that the severity of the impacts on family is higher especially among mothers. The results also showed that the impact of the child's oral health on different domains of family's QoL varied between interviewees in terms of frequency and intensity. It was clear from mothers' reports that impacts on family members varied with the mother being mostly affected, and this is in accordance with other studies conducted among parents of children with different types of disabilities and revealed that the impairments on aspects of quality of life were more

evident among mothers compared to other family members (Allik et al., 2006; Hedov et al., 2000), although some of these impacts might pre-exist and be experienced because of the presence of the disability by itself and not as a result of the child's oral health conditions. It is also possible that the pre-existence of disability and its burden (especially on family emotions) might contribute to the high severity of the negative impacts of children's oral health on their mothers.

In addition to the direct impacts of child's oral health on his/her QoL and that of the family separately, Figure 5.1 shows that the family's QoL can be affected directly by child's oral health or indirectly through the negative impacts of oral health on the child's OHRQoL. For example, the family's QoL can be affected directly by the child's oral health (i.e. cancel a planned family activity as a consequence of child's dental problem). Indirect impacts on family's QoL might be experienced as results of negative impacts of oral health on the child's emotional wellbeing or social relationships that in turn impacts negatively on family's QoL.

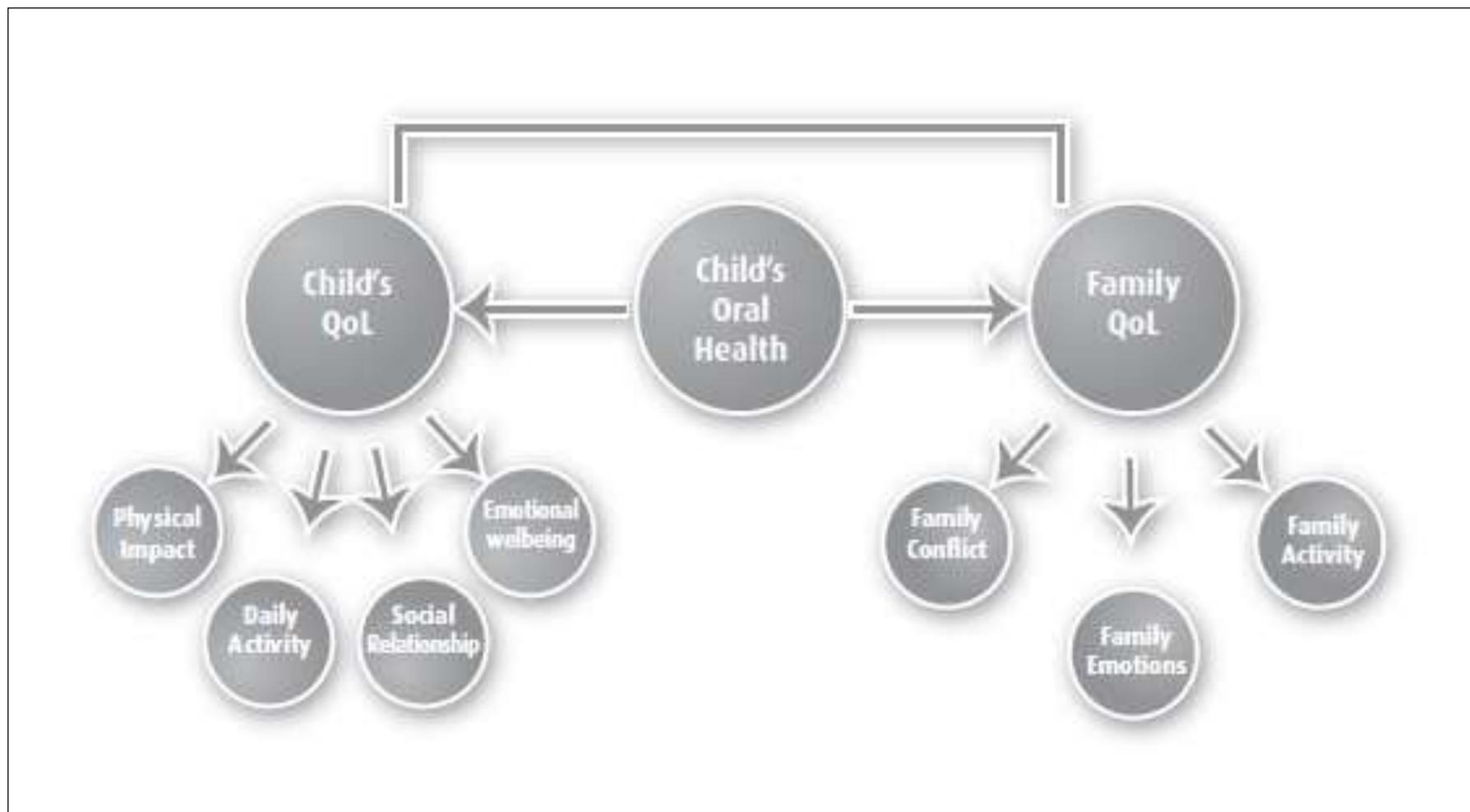


Figure 5.1. Impacts of Child's oral health on his/her QoL and that of the family as whole

5.2.4 Validation of the newly developed OHRQoL questionnaire, OH-QOLADS

Phase Two of the study aimed at assessing the psychometric properties of the newly developed questionnaire in which results were satisfactory. Internal consistency reliability was established through different statistical tests. The majority of inter-item correlations for the child section of OHRQoL were positive, some variables presented high correlation (for example, the correlation between embarrassment and being less confident was 0.96, and the correlation between stop laughing and being quiet was 0.92), and this might imply the potential redundancy of some of the items included. A negative weak correlation (close to zero) was also found in the variable 'been excluded' with the majority of other items; again, this raises concerns about this item potentially being redundant, however, at this stage of the study we aimed at including all items that may be relevant to the study population. Item-total correlation coefficients were above the recommended level of 0.2 (Streiner & Norman, 2003), except for the following items: exclusion by peers (0.043), and being teased or bullied by others (0.167). Cronbach's alpha was 0.909 (ever happened) and 0.902 (last year), above the arbitrary threshold of 0.70 (Kline, 1993).

Internal consistency reliability for the family section of OHRQoL showed a positive correlation between items except for the item on 'employment', which showed weak and sometimes negative correlations with the rest of the items. This might be due to the fact that the majority of participating mothers were not working; therefore, their child's oral health had no effect on their employment. Although what we aimed at is assessing impact of child's oral health on the employment of the whole family, this could underestimate the case. The Cronbach's alpha showed again satisfactory results of 0.828 (ever happened) and 0.807 (last year).

For construct validity, the child's OHRQoL scores were compared to different subjective measures (perceived general health, perceived oral health, and overall extent that oral problems affected child's QoL). All associations were significant and in the expected direction with higher child's OH-QOLADS scores, indicating worse OHRQoL, for the groups reporting worse perceptions. These consistent findings provide clear support for the validity of the new measure. The results also showed

clear gradients with every worse category in the exposures resulted in having higher OH-QOLADS and therefore worse OHRQoL. Furthermore, the discriminant validity of the measure was assessed by comparing OH-QOLADS scores with different indicators (clinically diagnosed caries, pulp involvement, malocclusion, plaque and gingival indices, and subjective report of bad breath and toothache) and results showed a significant association with those who experienced caries, pulp involvement, periodontal status, and toothache had higher OHRQoL score, and therefore worse quality of life, which means it practically demonstrates the ability of the developed measure to discriminate between different clinical groups. Results also show no association between malocclusion and child's OHRQoL this could be due to the fact that malocclusion (in mothers' perception) is not essential for children that have other more pressing issues, but there was a positive association between bad breath and child's OHRQoL with those reporting the presence of bad breath had higher OHRQoL indicating worse QoL, however this association was not significant.

Construct validity of family section of the OH-QOLADS was also assessed and showed significant association with mothers' reports on their child's perceived general health; perceived oral health; and overall extent that oral problems affected family QoL. Moreover, correlating the family section of the developed measure with different indicators to assess discriminant ability indicated the ability of the measure to discriminate between different clinical groups. Results showed that caries was not significantly associated with family's QoL but the experiences of pulp involvement and toothache were both correlated. This make sense because the presence of dental caries might not affect the family if it was in initial stages and the pain was not severe enough to cause any disturbances, but might impact on the family in case of severe caries that reach pulpal tissue and cause toothache.

In summary, results from the initial assessment of the developed questionnaire showed very satisfactory findings. Further studies should focus on further psychometric testing and complement its use for different purposes such as test its 'sensitivity to change' to facilitate its use in clinical settings and intervention studies.

5.3 Broader methodological issues

This study aimed to develop and test a measure of OHRQoL for children/adolescents with Down Syndrome, OH-QOLADS. There are some points that should be considered in relation to the methodology used:

5.3.1.1 Social context

It is important to consider the social context where the study was conducted in, and the nature of participants. The study was conducted in Riyadh city of Saudi Arabia. Saudi Arabia is the biggest country in the Middle East that is characterised by a high growth rate. According to national censuses the number of population is growing fast although at the moment there is no definite data on the prevalence of various types of disabilities at the country level. In 2000, the disability code was passed by the Saudi government to pledge that people with disabilities have access to free and appropriate medical, psychological, social, educational, and rehabilitation services through public agencies (The Provision Code for Persons with Disabilities in Kingdom of Saudi Arabia, 2004). The above guiding principles support the equal rights of individuals with disabilities in obtaining free and appropriate education and medical facilities. However, greater attention has been placed on enabling a person with disabilities to access health care services rather than education and training, and there is very little attention given to helping persons with disabilities gain employment and/or to wider participation issues.

Families of children with disabilities in Saudi Arabia receive help and support at an early stage from either public or private rehabilitation/educational centres. Depending on their levels of disabilities, children then continue their education at an inclusive education system in mainstream schools, or immediately start their training in vocational rehabilitation institutes. This study was conducted amongst mothers of children/adolescents with Down Syndrome (both boys and girls), whose children have access to either special care centres, or have access to an inclusive education system since it was easier to approach families with children with Down Syndrome from their schools and/or centres. This means that we missed other families who have no access to such facilities and they might have other views or opinions in regards to OHRQoL.

The study used mixed methods, which involved a qualitative phase where mothers were asked to talk about their experiences and opinions. It should be noted that this kind of research is not common in a country like Saudi Arabia where family privacy is a big issue, and mothers are not used to in-depth interviews. It is not common to speak about individuals' experiences in such a community especially in a sensitive topic. Mothers usually do not like to talk about their children's disability; therefore, it was not easy for mothers to fully express their opinions. This might affect the breadth and depth of the data obtained, and there might be other issues that were difficult for participating mothers to express.

Family conflicts and cultural differences should also be noted. For example, in Saudi Arabia, women are dependent on their male guardians -or drivers- to take them out for example they need somebody (male) to drive them to their appointments or any other places since women in this country are not allowed to drive by themselves. So, conflicts between family members (e.g. between mother and her husband) might occur if she needed to go out and take her child to attend for a dental appointment. This factor is cultural specific and might not be applied to other countries.

5.3.1.2 Other methodological considerations

The age of the children (12-18 years old) included in this study might have an impact on mothers' reports of oral health problems since at this age children would be expected to have a mixed dentition or newly erupted permanent teeth, and therefore, the problem of dental caries might be expected to be less obvious. In order to overcome the problem of under-reporting the oral health problems and their impacts, mothers were asked separately about any oral health problem that affected their children using two time frames; one had no specific time frame (problems ever experienced) and the other was confined to problems experienced in the last 12 months.

The issue of time recall was a challenge especially knowing that mothers showed lower priority to oral health status and therefore might not report it accurately. However mothers were able to report data on child's oral health and its impacts. In both qualitative and quantitative phases, data on child and family's OHRQoL were

collected at two time periods (ever happened, and happened in the last 12 month), to try to capture any possible impacts of the child's with Down Syndrome oral health. It was collected this way because there was no indication in the literature about the proper timeframe to use for OHRQoL among people with intellectual disabilities, and to make sure that we captured any impact experienced by them. And because mothers could confuse between times where impacts occurred, they were asked about each impact twice at different timeframe.

Psychological wellbeing of mothers and its effect on reporting is an important point that should be taken into consideration when using proxy reports. This might have an effect on the way the mothers used to report or express their opinions. For example, if the mother was in a low mood state or depressed this might impact on her answers in both ways, she may either over or under-estimate her answers. There are also other factors that might affect mothers' reporting, and if they exist, answers should be interpreted with caution. Some of these factors are: presence of other family member with any sort of chronic condition or disability.

5.4 Strengths and limitations of the study

Strengths

One of the main strengths of this study is that informants of Phase One interviews (mothers of children/ adolescents with Down Syndrome) were direct carers and the knowledge holders, and therefore expected to be in the best position to provide proxy reports as they should know their children's health status. In turn, this has facilitated generating items that were related to the OHRQoL of children with Down Syndrome. In addition, the mixed method approach of the study provided more meaningful data that allowed for triangulation of the results and discussion, and supporting our findings for both phases and relates them to the relevant literature.

This study is the first study that aims to develop a specific instrument to assess the impact of oral health on different aspects of life of children/adolescents with Down Syndrome. Therefore, the main strength of this study is the contribution of

information about OHRQoL of individuals with Down Syndrome to a field of research that is sparse. Findings from this study can inspire and support future investigations of the OHRQoL of individuals with Down Syndrome and those with other types of intellectual disabilities. In addition, the diversity of participated mothers and their children gave the opportunity to test OH-QOLADS across different individuals at different settings (i.e. inclusive education system, special needs schools, rehabilitation centres). It should also be noted that the collection of clinical data, and not only relying on perceived health status, added to the study and enhanced the validation process.

Limitations

Since the study was considered as a first step in understanding the impacts of oral-related problems and conditions of children/adolescents with Down Syndrome on the child and family's QoL, the sampling process excluded children with severe and/or multiple disabilities and this might mask important findings related to the topic. Limiting the interviews to mothers or direct carers of children with Down Syndrome might also result in masking some other oral impacts on other family members especially the siblings. In addition, actively including children with Down Syndrome might result on different perspectives, however, the main aim of this study as an initial step to understand their OHRQoL was exploring the dimensions of OHRQoL from the mothers' perspective; this initial step can be followed up by more inclusive methodologies. Another limitation of this study lies in the social context and this might result in perspectives that are specific to the culture studied. The aforementioned culture specific results in relation to the family conflict are a relevant example of this.

One of the current study limitations is using mothers as a proxy for their adolescents with Down Syndrome. Individuals with Down Syndrome vary in their levels of intellectual abilities and some of them with minor intellectual disabilities may provide a valid reports about their general health, oral health, and also their OHRQoL. However the main aim of the study was to explore the mothers' perceptions of their children's with Down Syndrome in regards to their oral health and OHRQoL to provide a more comprehensive view of mothers' concerns that

might not be captured by their children's reports due to their limited intellectual abilities. It is also of vital importance to use mothers' perception as the base on OHRQoL of children with Down Syndrome that future studies can build upon. Therefore, the aim of this study was directed towards mothers' perceptions with the hypothesis that mothers are usually the main carers of their children and the knowledge holder of their children's health status and well-being. It should also be noted that at this development stage, it was very important to collect qualitative data from informants who can capture and express all impacts of oral health on different aspects of life to develop a comprehensive and inclusive OH-QOLADS that can be adapted later as a self-report measure. Therefore, the aim was to target mothers of children/adolescents with Down Syndrome to avoid missing any important impacts that could be missed out if the qualitative interviews were conducted directly with the children. Future studies can build on the current work by including individuals with Down Syndrome and assessing if there are any related impacts that were not captured from mothers' reports. The involvement of young people with Down Syndrome in future research should also include their evaluation and reports on the ranking of the level of importance of such impacts on their lives to further develop the OH-QOLADS.

As shown in sections 2.5.2 & 2.5.3 in the literature review chapter, the majority of respondent-based health assessments developed for individuals with intellectual disabilities use proxies in preference to self-reports, and that is mainly because of the challenges presented in terms of limited communication skills and difficulties with comprehension (Lloyd et al., 2006). So in principle, self-report QoL information can be collected directly from individuals with intellectual disability considering the characteristics of the population and using an appropriate measure; however, the use of proxy measure is a more practical and realistic option.

In addition, studies among mainstream children indicated that proxy reports especially those collected from parents/mothers could provide valid reports on HRQoL & OHRQoL (Abanto et al., 2014; Jokovic et al., 2003). However, this does not underplay the importance of actively involving the children/adolescents themselves especially when it comes to their personal experiences and their health

and quality of life aspects, and future studies in the field of children/adolescents with intellectual disabilities should aim to actively include children with such disabilities.

It is also important to note that the developed OHRQoL questionnaire, OH-QOLADS, used in the validation study lacks an important item in the section of family's QoL, namely, financial impact. This item was not evident in mothers' reports of phase one for many reasons (explained in section 5.2.3 Family's QoL) but it is very crucial item in the family's QoL in general. Therefore, future studies addressing the financial aspects of family's QoL are needed.

5.5 Implications for research and practice

5.5.1 Implications for research

Initial findings of the developed measure showed satisfactory results of its validity and reliability testing. However, more studies are needed to further assess psychometric properties that are not addressed in this study such as responsiveness to change. Future studies are also needed to validate this measure across different age groups, and in people with different severities of Down Syndrome. Since this study was considered as the first step of assessing children with Down Syndrome OHRQoL and it was assessed by proxy reports. Further studies should also aim at developing the measure to be adapted and used as a self-report OH-QOLADS to help individuals with Down Syndrome express their opinions and actively involve them in research about their own feelings and perspectives of their lives.

The OH-QOLADS was developed and validated among group of mothers of adolescents with Down Syndrome residing in the Kingdom of Saudi Arabia. The measure shows its initial validity properties, however future work should aim at further development of the OH-QOLADS to confirm its suitability and applicability in different settings. It is important to outline the limitations that might emerge from developing such measure among a culture with its own characteristics and custom, Saudi Arabian culture. In this study we realised that some limitations exist due to cultural specific issues that in turn could affect the items included in the developed OH-QOLADS. Therefore, it is of prime importance to confirm OH-QOLADS items

through further studies among different cultures and settings. Although children and adolescents with Down Syndrome share specific oro-facial characteristics worldwide as indicated in the literature, their experiences of oral health impacts might differ within and between countries. Therefore, the developed measure can be further improved through many aspects such as testing the developed OH-QOLADS on different countries and different social settings to assess if there are any other impacts that were not present or evident among Saudi community (cultural specific and differences).

Future studies to document the prevalence of oral impacts of individuals with Down Syndrome are mandatory as this area is lacking. In order to provide the required oral health interventions to such group, a careful assessment of the oral health and its consequences using such measure is required.

5.5.2 Practice implications

Some oral conditions such as dental caries and/or periodontal diseases are chronic conditions that can affect children from a very young age. It is therefore important to measure their impacts on QoL, as they may affect his psychological, social and educational development. This is of prime importance especially among people with intellectual disabilities whose ability to learn face many challenges. Therefore, it is important to assess the impact of oral conditions and work on eliminating or reducing them by early, evidenced-based interventions (i.e. physiotherapy). In addition, this OH-QOLADS measure could be potentially in the future be used in clinical decision making to assess needs for treatment and the effectiveness of dental treatment and/or early intervention such as physiotherapy targeting drooling or tongue hypo-tonicity of children with Down Syndrome; thereby advancing the pediatric outcomes research agenda (Forrest et al., 2003). However, the measure cannot be used to assess intervention without the need to conduct further studies aimed at further assessing the psychometric properties (i.e. responsiveness to change). The developed measure could also potentially be a valuable outcome measure for evaluating oral health promotion programs and/or service initiatives for this segment of population (Watt et al., 2006).

The study found that many mothers reported that they had never been to dentist before, but the case was not the same regarding the general health in which majority of children had access to. This should encourage general health care providers to work in collaboration with dental professions, and apply a system that connect general and oral health. Efforts should also be directed to increase carers' awareness on the oral health and its role on the general wellbeing and satisfaction.

Knowing that individuals with Down Syndrome are at higher risk of developing systemic conditions that share the same risk factors as other diseases (i.e. obesity, diabetes), interventions adapting common risk factor approach might help in dramatic improvement of health condition (general and oral) of people with Down Syndrome. For example, prevention of periodontal disease among this segment can be achieved by proper education of children and their families, carers, schools and institutions representatives in which they accessed. Implementation of prevention in common risk factor approach might result in accelerated and desirable health improvements especially that children with Down Syndrome are usually at higher risk of developing systemic condition such as diabetes, and obesity in which they share common risk factors with oral diseases, therefore, reduction and control of sugar consumption for example might produce a significant reduction in many preventable health conditions including periodontal disease and dental caries.

5.5.3 Theoretical implications

Knowing that Down Syndrome is a chronic condition and as for other disabilities, adapting oral health models to a life course perspective should be considered in future research. This highlights the importance of updating and adapting current oral health models to accommodate the needs and demands of this segment of population.

5.5.4 Policy implications

The burden of oral diseases has a disproportionate impact on the poorer, less educated members of society, and the case might be the same, if not worse, among those with disabilities. Oral diseases are the fourth most expensive disease to treat (Petersen, 2008), and the cost of dental treatment is always higher among those with disabilities because they are more frequently left untreated until disease reaches

advanced levels which require higher cost treatments. Therefore, prevention policies implemented for mainstream population should also include those with disabilities, as they are at higher risk as well as having higher needs. Policies to improve health services utilizations and quality of services provision should also address those with disabilities as they face more challenges to access quality services.

5.6 Conclusions

Oral health conditions experienced by children/adolescents with Down Syndrome have undesirable impacts on different aspects of the child, as well as their family's QoL.

1. Results from the qualitative in-depth interviews showed a wide range of oral conditions that were reported by mothers and resulted on a range of impacts on the child's and family's QoL.
2. From the phase two study, the initial assessment of the developed OHRQoL questionnaire, OH-QOLADS, showed very satisfactory levels of its reliability and validity.
3. The oral health status (other than speech problems, chewing difficulties, drooling, and tongue conditions) and its impact on children with Down Syndrome are comparable to those among mainstream children of their age. Oral problems of adolescents with Down Syndrome have considerable negative effects on children's daily lives, as well as on the lives of their families.
4. The negative impacts especially on the emotional and social aspects of children's lives appear to be evident among Down Syndrome children. These may be further compounded by the existence of the disability and its consequences (such as stigmatisation and social isolation), however, this needs further investigation.
5. More studies are needed to further assess the psychometric properties of the developed OHRQoL questionnaire, gain understating of the magnitude of OHRQoL impacts among individuals with Down Syndrome and interpret them in a way that can be used to tailor the service provision so that it improves their general wellbeing and life satisfaction. Further work is also needed to examine whether items are missing from OH-QOLADS and needed to be added.

A decorative horizontal frame with a double-line border and a scalloped, ornate shape. The word "References" is centered within the frame.

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Appendices

Appendix 1

Prevalence rates of disability

Table 2.1. Disability prevalence rates for thresholds 40 and 50 derived from multidomain functioning levels in 59 countries, by country income level, sex, age, place of residence, and wealth

Population subgroup	Threshold of 40			Threshold of 50		
	Higher income countries (standard error)	Lower income countries (standard error)	All countries (standard error)	Higher income countries (standard error)	Lower income countries (standard error)	All countries (standard error)
Sex						
Male	9.1 (0.32)	13.8 (0.22)	12.0 (0.18)	1.0 (0.09)	1.7 (0.07)	1.4 (0.06)
Female	14.4 (0.32)	22.1 (0.24)	19.2 (0.19)	1.8 (0.10)	3.3 (0.10)	2.7 (0.07)
Age group						
18–49	6.4 (0.27)	10.4 (0.20)	8.9 (0.16)	0.5 (0.06)	0.8 (0.04)	0.7 (0.03)
50–59	15.9 (0.63)	23.4 (0.48)	20.6 (0.38)	1.7 (0.23)	2.7 (0.19)	2.4 (0.14)
60 and over	29.5 (0.66)	43.4 (0.47)	38.1 (0.38)	4.4 (0.25)	9.1 (0.27)	7.4 (0.19)
Place of residence						
Urban	11.3 (0.29)	16.5 (0.25)	14.6 (0.19)	1.2 (0.08)	2.2 (0.09)	2.0 (0.07)
Rural	12.3 (0.34)	18.6 (0.24)	16.4 (0.19)	1.7 (0.13)	2.6 (0.08)	2.3 (0.07)
Wealth quintile						
Q1(poorest)	17.6 (0.58)	22.4 (0.36)	20.7 (0.31)	2.4 (0.22)	3.6 (0.13)	3.2 (0.11)
Q2	13.2 (0.46)	19.7 (0.31)	17.4 (0.25)	1.8 (0.19)	2.5 (0.11)	2.3 (0.10)
Q3	11.6 (0.44)	18.3 (0.30)	15.9 (0.25)	1.1 (0.14)	2.1 (0.11)	1.8 (0.09)
Q4	8.8 (0.36)	16.2 (0.27)	13.6 (0.22)	0.8 (0.08)	2.3 (0.11)	1.7 (0.08)
Q5(richest)	6.5 (0.35)	13.3 (0.25)	11.0 (0.20)	0.5 (0.07)	1.6 (0.09)	1.2 (0.07)
Total	11.8 (0.24)	18.0 (0.19)	15.6 (0.15)	2.0 (0.13)	2.3 (0.09)	2.2 (0.07)

Note: Prevalence rates are standardized for age and sex. Countries are divided between low-income and high-income according to their 2004 gross national income (GNI) per capita (36). The dividing point is a GNI of US\$ 3255. Source (37).

A threshold of 40 on the scale 0–100 was set to include within estimates of disability, those experiencing significant difficulties in their everyday lives. A threshold of 50 was set to estimate the prevalence of persons experiencing very significant difficulties (WHO, 2011).

Appendix 2

Systematic review of oral health of people with intellectual disabilities (Anders & Davis, 2010)

Table 1. Included studies and summary of findings.						
Author (country)	Date	Score	Subjects	Controls/ comparison group	Measures	Findings
Hinchliffe (England)	1988	9	324 adults with mental retardation (MR)	165 age and gender-matched controls	DMFT, oral cleanliness, gingivitis, periodontal status, dentures	People with MR had poor oral hygiene, similar caries prevalence, more untreated caries, more gingivitis, more perio disease, worse OH, more edentulism, more traumatized teeth.
Sakellari (Greece)	2005	8	70 adolescents and young adults with Down syndrome (DS)	70 people with cerebral palsy, 121 age-matched controls	Probing depth, probing attachment level, bleeding on probing (BOP), hygiene, microbiology	People with DS had worse oral hygiene, more BOP, more severe periodontal destruction, earlier, heavier colonization with periodontal pathogens.
Zigmond (Israel)	2006	7	30 adults with DS	28 age-matched healthy controls	Plaque scores, BOP, probing depth, gingival recession, clinical attachment level, radiographic bone loss	People with DS had similar oral hygiene and gingival measures but severe periodontal disease. Prevalence, extent, and severity of periodontitis was significantly greater in DS group.
Cheng (China)	2007	7	65 adults with DS, age 17–42	65 age and gender-matched controls	DFT, plaque index (PI), BOP, pocket depths	People with DS had more plaque, fewer remaining teeth, more dental anomalies, fewer caries but more BOP, more severe periodontal disease, fewer filled teeth, more retained primary teeth.
Lopez-Perez (Mexico)	2002	7	32 people with DS, age 15–39	32 age and gender-matched controls	Simplified oral hygiene index (SOHI), gingival index (GI) attachment levels	DS group had more severe gingivitis but not periodontitis; greater extent of gingivitis and periodontitis, lower levels of calculus, similar plaque levels.
Shapira (Israel)	1991	7	12 institutionalized people with DS, age 20–48.	11 healthy and 12 non-DS-institutionalized MR age and gender-matched controls.	DMFT, community periodontal index of treatment needs (CPITN)	Periodontal treatment needs of DS and non-DS MR groups were higher than that of healthy controls. DS group had lowest caries experience. No correlation between salivary pH and caries levels found.
Seirawan (USA)	2008	6	102 institutionalized and noninstitutionalized adults with developmental disabilities (DD)	NHANES survey of general population	DMFT, PI, TMD, intraoral anomalies	Study group had higher caries prevalence than general population, poor oral hygiene, and higher DMF.
Oredugba (Nigeria)	2007	6	43 children and young adults with DS	43 age and gender-matched controls	SOHI, DMFT Angle's classification of malocclusion	DS group had poorer oral hygiene, no significant difference in caries prevalence, more malocclusion, more treatment needs.
Donnell (China)	2002	6	265 people with disabilities, age 25–35	Hong Kong adults in general population	DMFT, PI	Sample had poor oral hygiene, increasing with age. Sample had high number of missing teeth but lower DMFT than general population.
Cumella (England)	2000	6	50 adults with intellectual disabilities (ID)	UK adult dental survey of general population	DMFT, oral hygiene, traumatized teeth, general oral condition	Level of oral health depended on caregiver. Sample had poorer oral hygiene, more decayed and missing but fewer filled teeth, and high percentage of traumatized teeth.
Scott (Australia)	1998	6	101 adults with DD, age 21–53	Age-matched comparison group from National Oral Health Survey	DMFT, BOP, pocket depths, calculus, mucosal pathology, malocclusion	People with DD had more severe periodontal disease, more mucosal pathology and malocclusion but lower levels of calculus and lower levels of caries.

Table 1. Continued.

Author (country)	Date	Score	Subjects	Controls/ comparison group	Measures	Findings
Kendall (England)	1991	6	350 mentally handicapped adults	General population of adults in UK	DMFT, PI, calculus and gingivitis, WHO denture criteria	Study group had poor oral hygiene, extensive gingivitis, and high calculus prevalence but lower caries prevalence and lower rate of edentulism than comparison group.
Francis (England)	1991	6	195 handicapped adults, age 25–34	Dental health survey of general population in same region	DMF, dental cleanliness, calculus, periodontal condition, malocclusion, dentures	Study group had worse dental cleanliness and periodontal health. Study group had fewer filled teeth and more untreated caries.
Shaw (England)	1990	6	329 dentate mentally handicapped adults	General adult population of similar age	DMFT, PI, calculus, pocketing, BOP	Caries prevalence was lower in study group. Oral hygiene was poor, worst for people with physical disabilities as well as ID. More missing than filled teeth compared to general population.
Shapira (Israel)	1989	6	17 institutionalized people with autism, age 17–26	Pooled data of healthy age-matched adults from three sources	DMFS, DMFT, periodontal treatment need system	Adults with autism had severe periodontal problems, more missing teeth but lower rates of caries.
Rodríguez-Vasquez (Spain)	2002	5	166 institutionalized adults age 20–40 with mild-to-moderate DD	Spanish national survey	DMFT	Caries prevalence of entire sample was lower than that of adults in a national survey. Patients with DS had less caries, lower DMFT than other groups.
Lindemann (USA)	2001	5	325 adults with DD	NHANES III and one other study of adults	DMFT, "overall dental health"	Sixteen percent of subjects rated as having "good" and 78% "fair" oral health. Subjects had higher DMFT, similar rate of edentulism. 20% required urgent attention.
Gabre (Sweden)	2001	5	124 adults with ID	General population in Sweden	DMFS, reason for tooth loss	Longitudinal study showing majority of tooth loss in ID group was for periodontal reasons. Study group had lower caries incidence than general population but more tooth loss.
Gabre (Sweden)	1999	5	115 adults with MR	General adult population	Tooth loss, reasons for tooth loss	People with MR had fewer teeth and more tooth loss than the general population.
Whyman (New Zealand)	1995	5	207 intellectually handicapped, institutionalized adults	New Zealand study of oral health outcomes	DMFT, CPITN, SOHI, root caries index	Sample had more missing and decayed teeth but similar overall caries levels. Sample oral hygiene was poor, more edentulism than general population, more periodontal treatment needs.
Strauss (USA)	1985	5	233 disabled adults, 60% with MR	North Carolina norms from Dental Manpower study	DMFT, SOHI, Periodontal index	Study group had poorer oral hygiene scores, more calculus and debris. Study group had similar DMFT but more decayed and fewer missing teeth.
Nowak (USA)	1984	5	2,218 noninstitutionalized, handicapped people, age 16 or older	National statistics for general population	DMFT	Study group had similar caries index as general population. Study group has more missing and decayed than filled teeth.
Turner (Scotland)	2008	4	1,021 UK Special Olympians, mean age 28	Adult dental health survey of general population in UK	21 or more natural teeth, absence of fillings, no obvious untreated decay	Study group was more likely to be free from fillings and untreated decay than general population but in older age groups subjects were more likely to be having missing teeth.
Tiller (England)	2001	4	209 adults with learning disabilities, age 18–65	UK adult dental health survey of general population	DMFT, PI, calculus, denture assessment	Sample had more missing teeth and fewer filled teeth than general population, but same mean caries levels. Sample had high plaque levels.
Feldman (USA)	1997	4	713 Special Olympians, mean age 26	National Baseline Data from Healthy People 2000	DMFT, pain, gingivitis, fluorosis, malocclusion, hyperplasia, trauma	Study group had lower level of gingival health but also lower caries level than general population, fewer sealants. 13% of study group required urgent oral care.
Morton (England)	1977	4	90 mentally handicapped institutionalized adult females	Females in general adult population	Caries, oral hygiene, WHO criteria for periodontal disease, calculus, edentulism, DMF	Subjects had more missing and decayed teeth and fewer filled teeth than general population. Subjects had poor oral hygiene and high proportion of periodontal disease, 43% edentulous.
Reid (USA)	2003	3	9,620 Special Olympians, mean age 24	NHANES III survey of general population	DMFT, oral pain, gingivitis, sealants, fluorosis, oral injuries	It was unclear if study group had a higher prevalence of oral pain and untreated caries.

DMFT, decayed, missing, filled teeth; DFT, decayed, filled teeth; DMFS, decayed, missing, filled surfaces; OH, oral hygiene; NHANES, National Health and Nutrition Examination Survey; TMD, Temporomandibular disorder.

Appendix 3

Items of the family impact of child's oral health conditions

Family impact scale (FIS) (Locker et al. 2002)

Parental/family activity

- Have you or the other parent taken time off work?
- Has your child required more attention from you or the other parent?
- Have you or the other parent had less time for yourselves or other family members?
- Has your sleep or that of the other parent been disrupted?
- Have family activities been interrupted?

Parental emotions

- Have you or the other parent been upset?
- Have you or the other parent felt guilty?
- Have you or the other parent worried that your child will have fewer life opportunities?
- Have you felt uncomfortable in public places?

Family conflict

- Has your child argued with you or the other parent?
- Has your child been jealous of you or other family members?
- Has your child's condition caused disagreement or conflict in the family?
- Has your child blamed you or the other parent?

Financial burden

- Has your child's condition caused financial difficulties for your family?

Early childhood oral health impact scale (ECOHIS) (Pahel et al. 2007).

Parent distress domain

How often have you or another family member.....because of your child's dental problems or dental treatments?

- Been **upset**
- Felt **guilty**

Family function domain

How often....

- Have you or another family member taken **time off from** workbecause of your child's dental problems or dental treatments
- Has your child had dental problems or dental treatments that had a **financial impact** on your family?

Appendix 4

Ethical approval UCL

<p>2UCL RESEARCH ETHICS COMMITTEE GRADUATE SCHOOL OFFICE</p>	
<p>Dr Tsakos Department of Epidemiology and Public Health 1-19 Torrington Place UCL</p>	
<p>18 July 2012</p>	
<p>Dear Dr Tsakos</p>	
<p><u>Notification of Ethical Approval</u> <u>Project ID: 4047/001: Developing and testing an oral health-related quality of life measure for children/adolescents with Down Syndrome</u></p>	
<p>I am pleased to confirm that your study has been approved by the UCL Research Ethics Committee for the duration of the project i.e. until April 2014 subject to:</p>	
<ul style="list-style-type: none"> (a) Inclusion of a statement in the parental information sheet informing the child's family that they will be provided with a copy of the summary of clinical examination's findings. (b) Removal of the sentence "If (s)he withdraws we would reserve the right to include any information that has been recorded prior to withdrawal" from the information sheet. The Committee felt that if someone withdraws from the study it would be unethical to maintain records of the information that person has provided up to the point of withdrawal. (c) Inclusion of a data protection disclaimer in the various information sheets and consent forms. 	
<p>Approval is also subject to the following conditions:</p>	
<ol style="list-style-type: none"> 1. You must seek Chair's approval for proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature. Each research project is reviewed separately and if there are significant changes to the research protocol you should seek confirmation of continued ethical approval by completing the 'Amendment Approval Request Form'. 	
<p>The form identified above can be accessed by logging on to the ethics website homepage: http://www.grad.ucl.ac.uk/ethics/ and clicking on the button marked 'Key Responsibilities of the Researcher Following Approval'.</p>	
<ol style="list-style-type: none"> 2. It is your responsibility to report to the Committee any unanticipated problems or adverse events involving risks to participants or others. Both non-serious and serious adverse events must be reported. 	
<p><u>Reporting Non-Serious Adverse Events</u> For non-serious adverse events you will need to inform Helen Dougal, Ethics Committee Administrator (ethics@ucl.ac.uk), within ten days of an adverse incident occurring and provide a full written report that should include any amendments to the participant information sheet and study protocol. The Chair or</p>	

Vice-Chair of the Ethics Committee will confirm that the incident is non-serious and report to the Committee at the next meeting. The final view of the Committee will be communicated to you.

Reporting Serious Adverse Events

The Ethics Committee should be notified of all serious adverse events via the Ethics Committee Administrator immediately the incident occurs. Where the adverse incident is unexpected and serious, the Chair or Vice-Chair will decide whether the study should be terminated pending the opinion of an independent expert. The adverse event will be considered at the next Committee meeting and a decision will be made on the need to change the information leaflet and/or study protocol.

On completion of the research you must submit a brief report (a maximum of two sides of A4) of your findings/concluding comments to the Committee, which includes in particular issues relating to the ethical implications of the research.

With best wishes for the research.

Yours sincerely

Professor John Foreman
Chair of the UCL Research Ethics Committee

Co: AlBandary AlJameel

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Appendix 5

Ethical approval KSU

Appendix 6

Ministry of Social Affairs letter

Appendix 7

Ministry of Education letter

Appendix 8

Study description



Study Description

Study title

Developing and Testing an Oral Health-Related Quality of Life Measure for Children/Adolescents with Down syndrome

Study aims and objectives

Many researchers have investigated the impact of oral health conditions on individuals' quality of life; and they found that poor oral health status impact negatively on the various aspects of life (functional, psychological as well as social relationship). Knowing that there are no previous studies, and no specific questionnaire to measure the impact of oral health status on the quality of life of people with Down syndrome (DS), the aim of this study was to develop and test a questionnaire to help assessing the oral health related quality of life among 12 to 18 year-old children with Down syndrome from their mothers' perception. Accordingly, this study consisted of two phases; phase one was designed to explore the mothers perceptions about the oral health status of their children and how this could impact on their quality of life, so an interview with around (20) mothers of children with Down syndrome was conducted to help developing the questionnaire.

The second phase then aimed at testing and validating the developed oral health related quality of life questionnaire. So, the developed questionnaire needs to be administered to mothers of 12 to 18 year-old children with DS. And as a process of its validation, an oral examination of the children oral health condition is needed as well.

Sample of phase-two

A minimum of 100 mothers of 12 to 18 year-old children with DS will be selected to participate in the study with their children.

Ethical approval

The study has been approved by University College London's (UCL) Research Ethics Committee and King Saud University's (KSU) Research Ethics Committee.

Researcher

Appendix 9

Information sheet and Consent form

Project's title
Developing and Testing an Oral Health-Related Quality of Life Measure for Children/Adolescents
with Down syndrome



**Information sheet for the Semi-structured
Interview**



ALBandary ALJameel

Dear Mother,

You are being invited to take part in a research study. Before you decide to take part or not, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully.

Purpose of the study

We are developing a new questionnaire to measure some aspects of Oral Health-Related Quality of Life (OHRQoL) of children/adolescents with Down syndrome (DS). To do this first we want to talk to group of mothers of children/adolescents with DS about their experiences and difficulties they encounter and their thoughts about the oral health of their kids and how this could impact on their quality of life and that of the whole family.

Who are the researchers and who is funding the research?

The research is conducted by AlBandary AlJameel, and this research is not funded.

Why have you been chosen?

We are looking for volunteers who are mothers of children/adolescents with DS as they have first-hand experience of caring for children with DS including their oral health.

What do you have to do?

We would like you to take part in an interview discussion. We will talk to you about your perception of how the oral health status of your child could impact on his/her quality of life (functional, social, emotional, etc.). The interview will be audio-taped so that we have a record of what was said. You may also be asked to complete some questionnaires that we are developing for use later in the study.

What will happen to the information that you give?

The transcript of the interview and any questionnaires will only be accessible to members of the research team, and will be kept securely, in strict accordance with the data protection act. They will not be used for any other purpose. An analysis of the information will form part of our report at the end of the study and will be published in academic journals. Please let us know if you like to see a copy of the articles.

Will your taking part be confidential?

Be assured that all recorded information will be kept strictly confidential and only the research staff will have access to collected information. Your identifying information will not appear on any study report – all results will be reported as statistical summaries only. By completing and returning this form, you are giving us your consent that the personal information you provide will only be used for the purposes of this project.

Possible risks or inconveniences

Apart from the approximate 50-60 minutes needed for the interview, we can foresee no risks for you.

What are the benefits in taking part?

The benefits for taking part are that you will be sharing your experiences that could help others learning from them and also help improving our understanding of the impacts of oral health on children with DS. This in turn, could help improving the services provided to them.

What if you wish to withdraw?

Your participation is entirely voluntary and you can withdraw at any time you wish, without giving a reason. However, if you withdraw we would reserve the right to include any information that you give prior to leaving the group.

Ethics review

The research has been reviewed and approved by University College London's (UCL) Research Ethics Committee and King Saud University's (KSU) Research Ethics Committee.

**Contact information**

If you like further information about the study please do not hesitate to contact us at the following:

Researcher

**Thank you for reading this
Consent form for the interview**

Development of an instrument to measure Oral Health-Related Quality of Life
Among children/adolescents with Down syndrome

	Please tick
1. I confirm that I have read and understood the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	<input type="checkbox"/>
2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving a reason.	<input type="checkbox"/>
3. I agree to take part in the study.	<input type="checkbox"/>

Name of participant _____
Signature _____

Date _____



I have explained the project and the implications of the interview and I believe that the consent is informed and that she understands the implications of participation.

Name of researcher _____
Signature of researcher _____

Date _____

2 copies: 1 for participant and 1 for research file

Appendix 10

Topic Guide

OHRQoL-Down syndrome: Mother's Perspective

Aims & objectives:

The overall aim of this study is to assess in-depth the mother's perception of the impact of oral health conditions on their children's quality of life and family as whole. Experiences of and views about OHRQoL among children with Down syndrome are of particular interest.

This will include exploring all aspects of their experiences including:

- General Health status of the child
- Experience of having child with disability & any coping mechanism
- Oral health condition of the child
- Impacts of oral health conditions on child's quality of life
- Impacts of oral health conditions on family's quality of life

Introduction

Aim: to introduce the research and set the context for the proceeding discussion.

- Introduction to the study & my self
- Thank the interviewee for participation
- Ask the interviewee to answer openly and freely
- Tell the interviewee that there will be no right or wrong answers, all opinions are valid and helpful
- Ask permission to record the interview, and reasons for recording
- Tell how long this discussion will last
- Let the interviewee know that they can have a break if she needed during the discussion, voluntary nature of participation & right to withdraw
- Reassure that the responses are confidential
- Any questions they have

Background on the study

As you know the aim of this study is to develop and test a questionnaire to assess the impact of oral conditions on the quality of life of children/adolescents with Down syndrome. In order to reach this aim we need to conduct this interview with you (mother) because your opinion and experience as being the carer of your child will help us in developing the questionnaire that suits children with DS.

1. Background and personal circumstances (Icebreaker)

Aim: to introduce respondents and highlight any key background issues

- Tell me about your self
 - For information like: Name, Age, Kind of relation to the child (Mother or guardian), Marital status, Occupation (Ask if she works outside home), Education
- Tell me about your family and children
 - Possible prompts include
 - Number of children, ages, names etc.
 - How long lived in this area etc.
- Other interest and activities

2. General health

Aim: to establish a general idea of the child's general health

- Tell me about your child/ how is his/her health
OR
- Tell me about your child's general health
 - How is he/she doing – is he/she fit and healthy

3. Presence of the disability

Aim: to investigate how respondent feel about experience of having child with disability & map how do they cope with it

- What does it mean to the family to have child with disability (or alternatively 'what has 'child's name disability meant for the family
 - Possible prompts
 - When did you know about your child condition (probe here gently for 'uncertainty' with condition, child's future, what will happen, what to expect. Probe very gently on what immediate and wider family think about a child with disability
 - From where do you get advice (probe here for Mother's perception of her information needs)
 - What are your needs (perhaps be a bit more explicit here?)
 - Impact of child's disability regime on family dynamics, relationships and routines
 - Economic impact

- As a family, how do you cope with your child's disability/ what is the coping mechanism you use

Managing child's health care and social care regime?

Satisfaction with family support, other social support and health and social care

4. Oral health

Aim: to establish a general idea of the child oral health

- Tell me a bit about his/her oral health – how is his/her mouth, teeth and gums
 - Does he/she has any particular problem with his/her mouth
 - Possible prompts
 - Pain/discomfort etc.
 - Tooth decay
 - Bleeding gums
 - Bad breath etc.
 - Appearance/ fractured & unrestored ant. tooth
 - Enlarged tongue, dribbling or speech problem

5. Impacts of oral health on child's QoL

Aim: to establish how mothers perceive the impact of their children's oral health on QoL

- Describe how your child's oral health affects different aspects of his/her life
 - Possible prompts (PROBE WELL)
 - Daily function and activities such as sleeping, eating, playing, etc.
 - Emotional state – mood, upset, tense, anxious, unhappy etc.
 - Being with family, friends etc.
 - Social aspect, i.e. smiling, how others view appearance?

6. Impacts of child's oral health on family's QoL

Aim: to assess how children oral health impacts on their families' QoL

- How do you think you, your partner, or other kids have been affected by his/her oral health conditions **GENERAL**
- OR
- Could you tell me and explain how your child's oral health affects the family's activity?

- Possible prompts
 - Time off work
 - Interrupted family activities
 - Sleep disruption
 - More attention
 - Had less time for yourself or other family member
- How much care do you think your child's oral health needs, Does this have any impact on the family's activities/ relationships
- How do you think your child's oral health affects the emotions of you and your partner?
 - Possible prompts
 - Felt guilty
 - Upset
 - Worried that your child will have fewer life opportunities
 - Uncomfortable in public places
 - And how you overcome these emotions?
- Explain how your child's oral health does cause any family conflict
 - And how you solve it
 - Possible prompts
 - Child argued with you or other family member
 - Being jealous of you or other family member
 - Disagreement or conflict in the family
 - The child blame you or the other parent

Wrap up & ending

Summarize the conversation and what has been discussed in the interview and then ask:

- What do you feel have been the most important things that we have spoken about?
- Is there anything else that you would like to discuss?
- Or any question you would like to ask?
- Reassure them about the confidentiality & thank them for participation
- Ask if they would like to be informed of the outcomes of this research (address)

* In each interview note: Date, Time, Place of interview, & Reference No.

Appendix 11

Arabic questionnaire



إستبانة صحة الفم الجزء الأول

التاريخ:

الباحثة: د. البندري حسن الجميل

نشكر لكم مشاركتكم في هذه الدراسة ونحيطكم علما بان جميع المعلومات المقدمة سيتم التعامل معها بسرية تامة

- سوف يساعدنا هذا الإستبيان في زيادة مستوى الفهم عن صحة فم وأسنان طفلك، وتأثيرها على حياة الطفل والعائلة كذلك
- سوف تستغرق المقابلة 25 دقيقة تقريبا
- لا توجد إجابات صحيحة أو خاطئة، لذا يرجى الإجابة بصراحة
- تسأل معظم هذه الأسئلة عن ما إذا كانت مشاكل طفلك المتعلقة بالفم والأسنان (في حال وجودها) قد أثرت على أنشطه الطفل اليومية وكذلك العائلة، لذلك نرجوا التأكد قبل الإجابة أن الأثر الحاصل هو نتيجة لصحة الفم والأسنان وليس لسبب اخر
- جميع الإجابات سيتم التعامل معها بسرية تامة

مقدمة حول الدراسة

كما تعلمين فإن الهدف من هذه الدراسة يكمن في تطوير استبيان لقياس بعض جوانب الحياة المتعلقة بصحة الفم والأسنان للأطفال/ المراهقين الحاملين لمتلازمة دارون، ولتحقيق هذا الهدف فإننا نحتاج لجمع بعض المعلومات منك (الأم) لكون رأيك وخبرتك يشكلان المساعد الأكبر لتحقيق أهداف هذه الدراسة.

الرقم التسلسلي

البيانات العامة

(١) الطفل

- ما اسم طفلك المصاب بمتلازمة داون؟ _____
- هل طفلك ذكر أم أنثى؟
 - ذكر
 - أنثى
- كم عمره / عمرها؟ _____
- هل لطفلك أخوة أو أخوات، كم يبلغون من العمر؟ _____
- هل هو الطفل الوحيد في العائلة المصاب بهذه الإعاقة؟
 - نعم
 - لا

(٢) الأم

- ما أسمك؟ _____
- كم عمرك؟ _____
- ماهي حالتك الإجتماعية؟
 - متزوجة
 - مطلقة
 - أرملة
- هل تعملين؟
 - نعم، أعمل بدوام كامل
 - نعم، أعمل بدوام جزئي
 - لا أعمل

- ماهو المستوى التعليمي الحاصلة عليه؟
 - غير متعلمة
 - المرحلة الابتدائية
 - المرحلة المتوسطة
 - المرحلة الثانوية
 - المرحلة الجامعية
 - دراسات عليا

الصحة العامة

- كيف تقيمين مستوى الصحة العامة لطفلك؟

- سيئ جداً
- سيئ
- معتدل
- جيد
- جيد جداً

- هل يوجد لطفلك طبيب عام؟
 - نعم
 - لا

- هل يزور طفلك عادة الطبيب.....

- لإجراء الفحص الطبي الدوري
- لإجراء الفحوصات بين الحين والآخر (مرة واحدة في العام أو أقل)
- فقط عندما يكون لديه مشكلة ما

- ما مدى صعوبة العثور على طبيب لرعاية طفلك؟

- صعب جداً
- صعب
- ليس صعباً أو سهلاً / متوسط
- سهل
- سهل جداً

- هل سبق وأن تم تشخيص طفلك بواسطة الطبيب بأي من الحالات الصحية التالية؟

الحالة	نعم	لا	توضيح
مشاكل في القلب			
أمراض الغدة الدرقية			
مرض السكري			
مشاكل في البصر (العمى أو العمى الجزئي)			
مشاكل في السمع (الصمم أو الصمم الجزئي)			
أي حالات أخرى			

- هل يستخدم طفلك أي دواء؟

نعم

لا

إذا كانت الإجابة بنعم، لماذا؟ _____

عدد الأدوية المستخدمة _____

أسماء الأدوية (إن أمكن): _____

صحة الفم

- برأيك، كيف تقيّمين مستوى صحة أسنان طفلك (يشمل ذلك الفم والأسنان)؟

- سيئ جداً
 سيئ
 محتدل
 جيد
 جيد جداً

- خلال الـ ١٢ شهراً الماضية، هل عانى طفلك من إي من مشاكل/ حالات الفم التالية:

توضيح	لا	نعم	حالة صحة الفم
			التنفّس عن طريق الفم
			تقاطر/ سيلان اللعاب
			إلتواء/ إعوجاج في الأسنان
			تسوس في الأسنان
			ألم في الأسنان
			نزف اللثة
			رائحة كريهة من الفم
			طحن الأسنان
			تضخم في حجم اللسان
			تدلي/ خروج اللسان من الفم
			صعوبة في التحدث
			صعوبة في المضغ
			أي حالات أخرى

زيارة طبيب الأسنان

- هل سبق وأن زار طفلك طبيب الأسنان؟

- نعم، مرة واحدة
- نعم، مرتين أو ثلاث مرات
- نعم، أكثر من ثلاث مرات
- لا، لم يسبق وأن زار طبيب الأسنان - انتقل إلى الفقرة التالية

- هل يزور طفلك عادة طبيب الأسنان...

- لإجراء الفحص الدوري (فحص طبي كل ستة أشهر أو نحو ذلك)
- لإجراء الفحوصات بين الحين والآخر (فحص طبي مرة كل سنة أو أقل)
- فقط عندما يعاني من مشاكل في الأسنان

- هل سبق وأن تلقى طفلك في أي وقت مضى أي شكل من أشكال العلاج التالية :

- حشوة الأسنان
- خلع الأسنان
- التخدير العام قبل علاج الأسنان (الطفل غير واع)
- التخدير عند طبيب الأسنان للتهديئة قبل أو أثناء وجود فحص طبي أو علاج (لا يزال طفل واع)
- العلاج بغرض منع تسوس الأسنان، مثل وضع مادة واقية
- تقويم أو تعديل الأسنان
- إصلاح الأضرار التي لحقت الأسنان إثر السقوط أو إصابة أخرى
- التنظيف والتلميع
- الأشعة السينية للأسنان أو الفم
- معالجة أخرى (حدد: _____)
- لا يوجد علاج، لم يحصل إلا على فحص طبي

- ما مدى صعوبة إيجاد طبيب أسنان معالج لطفلك؟

- صعب جدا
- صعب
- سهل
- سهل جدا
- لم أحاول أبداً

العناية بأسنان طفلك

- كم كان عمر طفلك عندما بدأ تنظيف أسنانه بواسطة الفرشاة، أو تم استخدامها من قبل آخرين (مثل الوالدين) لتنظيف أسنانه؟

- أقل من ٦ أشهر
- بين ٦ أشهر وسنة
- بين سنة إلى سنتين
- بين سنتين إلى أربع سنوات
- بين أربع إلى ست سنوات
- ٦ سنوات فما فوق
- طفلي لا ينظف أسنانه بالفرشاة، ولا يقوم أحد آخر بتنظيفها له (مثل الوالدين) - انتقل إلى الجزء التالي

- من يقوم عادة بتنظيف أسنان طفلك بالفرشاة هذه الأيام؟

- طفلك
- شخص بالغ (مثل الوالدين، معلمة)
- شخص بالغ وطفلك معا

- عادةً، كم مرة في اليوم يقوم طفلك بتنظيف أسنانه بالفرشاة (أو في حال تنظيفها له من قبل شخص آخر)؟

- أكثر من ثلاث مرات في اليوم
- ثلاث مرات في اليوم
- مرتين في اليوم
- مرة واحدة في اليوم
- أقل من مرة في اليوم
- أبداً، لا ينظف أسنانه بالفرشاة

الجزء الثاني

صحة الفم وعلاقتها بجودة الحياة

للتذكير مرة أخرى:

- لا يوجد أجوبة صحيحة أو خاطئة، لذا يرجى الإجابة بصراحة
- تسأل معظم هذه الأسئلة عن ما إذا كانت مشاكل طفلك المتعلقة بالفم والأسنان (في حال وجودها) قد أثرت على أنشطة الطفل اليومية وكذلك العائلة، لذلك نرجوا التأكد قبل الإجابة أن الأثر الحاصل هو نتيجة لصحة الفم والأسنان وليس لسبب آخر

الاستبيان الخاص بالطفل

هل سبق وأن أثرت صحة فم وأسنان طفلك على أحد الجوانب التالية من حياته:

التأثير على الطفل	سبق أن حدث ٣ ٢ ١ ٠	حدث في السنة الماضية ٣ ٢ ١ ٠	ما سبب ذلك من وجهة نظرك؟
الجسدي <ul style="list-style-type: none"> • ألم / إنزعاج 	_____	_____	
النشاط اليومي <ul style="list-style-type: none"> • الأكل • التحدث/ الكلام • تنظيف الأسنان • النوم • الواجبات المدرسية • اللعب 	_____	_____	
الجانب العاطفي <ul style="list-style-type: none"> • البكاء أو الشعور بالضيق • التوقف عن الضحك • الهدوء • الخجل أو الإنطواء • الشعور بالإحراج • الشعور بعدم الثقة بالنفس • الوعي بالمشاكل المتعلقة بالفم • الغضب • التصرف بعناد 	_____	_____	
الجانب الاجتماعي <ul style="list-style-type: none"> • الإنسحاب من العلاقات الأسرية • الإنسحاب من الأصدقاء • إستبعاده من قبل أصدقائه • إغاضته وتلقيبه من قبل الآخرين 	_____	_____	
أخرى، يرجى التوضيح	_____		

* ٠ = لا على الإطلاق، ١ = حدث بشكل بسيط، ٢ = متوسط، ٣ = شديد

جودة الحياة الأسرية

هل سبق وأن أثرت صحة فم وأسنان طفلك على أحد الجوانب التالية من الحياة الأسرية:

التأثير على العائلة	سبق أن حدث ٣ ٢ ١ ٠	حدث في السنة الماضية ٣ ٢ ١ ٠	ما سبب ذلك من وجهة نظرك؟
الجانب العاطفي <ul style="list-style-type: none"> • إكتئاب أو ضيق، أو اضطراب • لوم الذات أو الشعور بالذنب • القلق • الغضب 	_____ _____ _____ _____	_____ _____ _____ _____	
النشاط اليومي <ul style="list-style-type: none"> • إلغاء نشاط مقرر • التأثير على العمل • الإنشغال عن أفراد العائلة الأخرى • التأثير على نمط/ طريقة النوم 	_____ _____ _____ _____	_____ _____ _____ _____	
النزاعات العائلية <ul style="list-style-type: none"> • المجادلة مع أحد أفراد العائلة (من أجل الذهاب لطبيب الأسنان مثلاً) • الشعور بالخيرة بين الأخوان 	_____ _____	_____ _____	
أخرى، يرجى التوضيح	_____ _____		

* ٠ = لا على الإطلاق، ١ = حدث بشكل بسيط، ٢ = متوسط، ٣ = شديد

الأسئلة النهائية العامة

- كم عدد المرات التي تم فيها مضايقة أو تلقيب طفلك من قبل الأطفال الآخرين بسبب صحة أسنانه وفمه؟

- أبدا، لم يسبق وأن تم تلقيبه
- مرة واحدة أو مرتين
- في بعض الأحيان
- في كثير من الأحيان
- كل يوم تقريبا

- هل تعتقد أن ذلك قد يكون له تأثير سلبي على طفلك؟

- لا، ليس له تأثير على الإطلاق
- تأثير بسيط
- تأثير متوسط
- تأثير كبير
- تأثير كبير جدا

توضيح _____

- هل تعتقد أن يكون لذلك تأثير سلبي على العائلة بأي حال من الأحوال؟

- لا، ليس له تأثير على الإطلاق
- تأثير بسيط
- تأثير متوسط
- تأثير كبير
- تأثير كبير جدا

توضيح _____

- برأيك، ما هو السبب الرئيسي الذي يؤدي إلى مضايقة أو تلقيب طفلك؟

- بشكل عام، إلى أي مدى أسهمت الحالة الصحية لعم طفلك في التأثير سلبا على حياته؟

- لم تؤثر على الإطلاق
- أثرت بشكل بسيط
- أثرت بشكل متوسط
- أثرت بشكل كبير
- أثرت بشكل كبير جدا

- بشكل عام، إلى أي مدى أسهمت الحالة الصحية لغم طفلك في التأثير سلباً على الحياة الأسرية؟

- لم تؤثر على الإطلاق
- أثرت بشكل بسيط
- أثرت بشكل متوسط
- أثرت بشكل كبير
- أثرت بشكل كبير جداً

- هل هناك شيء آخر ترغبين التحدث عنه أو اضافته فيما يتعلق بصحة أسنان طفلك أو بشكل عام؟

English questionnaire



Oral Health Questionnaire Part 1

Researcher: Dr. Albandary Hassan Aljameel

Date:

Demographic, General and Oral health questions

Thank you for agreeing to help us and participate in our study

- This questionnaire will help us understand more about your child's oral health condition and its effect on your child and family
- The interview will take 20-25 minutes
- There is no right or wrong answers, so please try to answer as honestly as you can
- Please remember that we are interested to know how the health of your child's mouth affects his/her daily activity and routine, so please remind yourself before answering any question "if the particular effect happens because of the dental health that is the health of your child's mouth and teeth"
- All of your responses will be treated in the strictest confidence

Introduction & Background on the study

As you know the aim of this study is to evaluate a questionnaire to assess the impact of oral conditions on the quality of life of children/adolescents with Down syndrome. In order to reach this aim we need to collect some information from you (mother) because your opinion and experience as being the carer of your child will help us to reach our aim.

Reference number

Demographic Data

A. Child

- What is the name of your child with Down syndrome? _____
- Is your child boy or girl?
 - Boy Girl
- How old is he/she? _____
- Does he/she have brothers and sisters, how old are they? _____
- Is he/she the only child with a disability in the family?
 - Yes No

B. Mother

- What is your name? _____
- How old are you? _____
- What is your marital status?
 - Married
 - Divorced
 - Widowed
- May we check, at the moment are you doing any paid work, either full or part time?
 - Yes, full-time
 - Yes, part-time
 - No, I do not work
- What is your level of education?
 - No education
 - Primary school
 - Intermediate school
 - Secondary school
 - University (e.g. Bachelor)
 - Postgraduate (e.g. PhD, MSc)

General Health

- **How would you rate the general health of your child?**
 - Very poor
 - Poor
 - Fair
 - Good
 - Very good

- **Does your child have a physician?**
 - Yes No

- **Does your child usually go to the physician ...**
 - For a regular check-up
 - For an occasional check-up (a check-up once a year or less)
 - Only when he/she has trouble

- **How difficult is it to find a doctor to care for your child?**
 - Very difficult
 - Difficult
 - Neither difficult nor easy
 - Easy
 - Very easy

- **Has your child ever been diagnosed by a doctor with any of these medical conditions?**

Condition	Yes	No	Explain
Heart problems			
Thyroid gland disease			
Diabetes			
Visual Problems (blind or partially blind)			
Hearing Problems (deaf or partially deaf)			
Any other condition			

- **Does your child take any medication?**
 - Yes No

If yes, for what? _____

Number of medication/s _____

Names (if possible) _____

Oral Health

- In your opinion, how would you rate the dental health (that is the health of the mouth and teeth) of your child?

- Very poor
- Poor
- Fair
- Good
- Very good

- In the last 12 months, has your child had any of the following Oral conditions/problems?

Oral condition	Yes	No	Explain
Mouth Breathing			
Drooling / Dribbling			
Crooked teeth			
Tooth decay			
Toothache			
Bleeding gum			
Bad breath			
Tooth grinding			
Enlarged tongue			
Protruded tongue			
Speaking difficulty			
Chewing difficulty			
Any other conditions			

Visiting the dentist

- Has your child ever been to a dentist?
 - Yes, once
 - Yes, 2 or 3 times
 - Yes, more than three times
 - No, he have never visited a dentist → **Go to the next section**
- Does your child usually go to the dentist ...
 - For a regular check-up (a check-up every six months or so)
 - For an occasional check-up (a check-up once a year or less)
 - Only when he/she has trouble with his/her teeth

- **Has your child ever received any form of the following treatments (You can choose more than one answer)...**
 - Teeth filled
 - Teeth taken out
 - A general anesthetic before dental treatment (child is unconscious)
 - Sedation at the dentist to calm him/her before or while having a check-up or treatment (child remains conscious)
 - Treatment to stop teeth decaying or going bad e.g. by painting or sealing the teeth
 - A brace fitted or adjusted
 - Repair of damage to teeth after a fall or other injury
 - Scale and polish
 - X-ray of teeth or mouth
 - Other treatment (specify: _____)
 - No treatment, only had a check-up

- **How difficult is to find a dentist to care for your child?**
 - Very difficult
 - Difficult
 - Neither difficult nor easy
 - Easy
 - Very easy
 - Never tried

Looking after your child's teeth

- **How old was your child when he/she started brushing his/her teeth or having them brushed for him/her?**
 - Under 6 months of age
 - Between 6 months and 1 year of age
 - Between 1 and 2 years of age
 - Between 2 and 4 years of age
 - Between 4 and 6 years of age
 - 6 years of age or older
 - My child does not brush their teeth or have them brushed for him/her → **Go to the next section**

- **Who usually brushes your child's teeth nowadays?**
 - Your child
 - An adult
 - An adult and your child together

- **How often does your child usually brush his/her teeth (or have them brushed for him/her)**
 - More than three times a day
 - Three times a day
 - Twice a day
 - Once a day
 - Less than once a day
 - Never

Oral Health Questionnaire Part 2

Oral Health-Related Quality of Life

Just to remind you again:

- There is no right or wrong answers, so please try to answer as honestly as you can
- Please remember that we are interested to know how the mouth health affects your child daily activity and routine, so please remind yourself before answering any question “if the particular effect happens because of the dental health that is the health of your child’s mouth and teeth”

Child's OHRQoL

- Have any of the following ever happened to your child because of his/her dental health (that is the health of the mouth and teeth)?

Impact on Child	Ever happened				Last year				What do you think the cause of it?
	0	1	2	3	0	1	2	3	
Physical <ul style="list-style-type: none"> ▪ Pain/ Discomfort 									
Activity <ul style="list-style-type: none"> ▪ Eating ▪ Speaking ▪ Brushing teeth ▪ Sleeping ▪ School homework ▪ Playing 									
Emotional <ul style="list-style-type: none"> ▪ Cry or feel upset ▪ Stop laughing ▪ Being quiet ▪ Become shy or introvert ▪ Feel embarrassed ▪ Become less confident ▪ Being conscious about his/her mouth ▪ Being angry ▪ Behave in stubborn manner 									
Social <ul style="list-style-type: none"> ▪ Withdraw him/herself from family relationships ▪ Withdraw him/herself from friends relationships ▪ Been excluded from friends ▪ Been teased/bullied by others 									
Others, specify	_____								

*Severity range: 0 = Not at all, 1 = Minor, 2 = Moderate, 3 = Severe

Family QoL

- Have any of the following affects the family life/ routine because of your child's dental health (that is the health of the mouth and teeth)?

Impact on Family	Ever happened				Last year				What do you think the cause of it?
	0	1	2	3	0	1	2	3	
Emotions <ul style="list-style-type: none"> ▪ Depressed, Distress, or Upset ▪ Self-blame/ guilt ▪ Worried ▪ Angry 									
Activities <ul style="list-style-type: none"> ▪ Cancel scheduled activity ▪ Affect your job ▪ Time-off from other family members ▪ Affect your sleeping pattern 									
Conflict <ul style="list-style-type: none"> ▪ Argue with other family member (e.g. to go to clinic) ▪ Other sibling being jealous 									
Others, specify									

*Severity range: 0 = Not at all, 1 = Minor, 2 = Moderate, 3 = Severe

General Final Questions

- How often has your child been teased/bullied by other children because of one of his/her oral health conditions?

- Never
- Once or twice
- Sometimes
- Often
- Every day or almost every day

- Do you think this could have any negative impact on your child?

- Not at all
- Very little
- Some
- A lot
- Very much

Explain _____

- Does this impact on the family by any means?

- Not at all
- Very little
- Some
- A lot
- Very much

Explain _____

- In your opinion, what is the main oral health condition that leads to teasing/bullying?

- To what extent does the condition of your child's dental health negatively affect his/her life overall?

- Not at all
- Very little
- Some
- A lot
- Very much

- To what extent does the condition of your child's dental health negatively affect the family life overall?

- Not at all
- Very little
- Some
- A lot
- Very much

- Is there anything else you would like to tell us about your child's dental health or in general?

Thank you for taking the time
to fill out this survey

Appendix 12



Oral Examination Form

Researcher: Dr. Albandary Hassan Aljameel

Date:

Reference number	
Name	
Age	
Gender	
Centre name	



Mouth Status

Condition	Present	Absent
Protruded tongue at time of examination		
Visible missing/decayed unrestored ant. teeth		
Dribbling at time of examination		

Malocclusion

Condition	Present	Absent
<p><i>Overjet</i></p> <ul style="list-style-type: none"> • Normal • Protrusion • Anterior cross bite • Absent 		
<p><i>Overbite</i></p> <ul style="list-style-type: none"> • Normal • Deep overbite • Anterior open bite • Absent • Edge-to-edge 		
Posterior cross bite		



Type of teeth

Permanent

Mixed

Caries experience

				55	54	53	52	51	61	62	63	64	65				
	18	17	16	15	14	13	12	11	21	22	23	24	25	26	27	28	
DMFT																	DMFT
PUFA																	PUFA
PUFA																	PUFA
DMFT																	DMFT
	48	47	46	45	44	43	42	41	31	32	33	34	35	36	37	38	
				85	84	83	82	81	71	72	73	74	75				

DMFT

	Primary	Permanent
Decayed		
Missing		
Filled		
Fractured		
Sound		

PUFA

	Primary	Permanent
Pulpal inv.		
Ulceration		
Fistula		
Abscess		

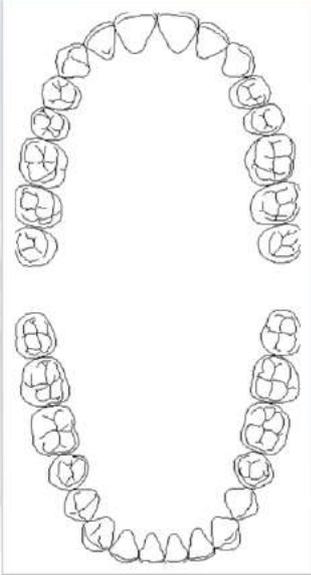


Appendix 13




نتيجة فحص الأسنان





الإسم:

تسوس

نعم

لا

التهاب في اللثة

نعم

لا

مشاكل أخرى

نعم

لا

التاريخ:

توقيع: 

الباحثة: د. البندري حسن الجميل

Appendix 14**Pilot study
Evaluation form****Ask mothers about:****1- Length of the interview**

- How long did it take?

- Is it of proper length, or it needs to be shortened?

2- Clarity of the wording used, if they recommend any other words

- Was there any difficult word/s that needed to be clarified?

- Do you recommend some modification/ adjustment of some words? If yes, please clarify?

3- Comprehensiveness of the questionnaire

- Does this questionnaire include all related areas?

- Is there anything you wish to add?

Appendix 15

External (Test-retest) reliability

Table 1. Test-retest κ coefficient for child’s section of OH-QOLADS (Ever happened)

Item	Absolute score difference				Number of observations	Un-Weighted κ	Weighted κ
	0	1	2	3			
Pain	9	1	0	0	10	0.859	0.927
Eating	9	1	0	0	10	0.825	0.911
Speaking	10	0	0	0	10	1	1
Tooth Brushing	9	1	0	0	10	0.756	0.821
Sleeping	10	0	0	0	10	1	1
Schooling	9	1	0	0	10	0.706	0.762
Playing	10	0	0	0	10	1	1
Crying	9	1	0	0	10	0.825	0.922
Ceasing to laugh	9	1	0	0	10	0.808	0.872
Being quiet	10	0	0	0	10	1	1
Shyness	10	0	0	0	10	1	1
Embarrassment	10	0	0	0	10	1	1
Lack of confidence	10	0	0	0	10	1	1
Self-consciousness	10	0	0	0	10	1	1
Anger	10	0	0	0	10	1	1
Stubbornness	10	0	0	0	10	1	1
Withdrawal from family	10	0	0	0	10	1	1
Withdrawal from friends	10	0	0	0	10	1	1
Exclusion by peers	10	0	0	0	10	1	1
Teasing/Bullying	10	0	0	0	10	1	1

Table 2. Test-retest κ coefficient for child’s OH-QOLADS (Last year)

Item	Absolute score difference				Number of observations	Un-Weighted κ	Weighted κ
	0	1	2	3			
Pain	9	1	0	0	10	0.831	0.914
Eating	9	1	0	0	10	0.811	0.906
Speaking	10	0	0	0	10	1	1
Tooth Brushing	9	1	0	0	10	0.630	0.773
Sleeping	10	0	0	0	10	1	1
Schooling	10	0	0	0	10	1	1
Playing	10	0	0	0	10	1	1
Crying	10	0	0	0	10	1	1
Ceasing to laugh	10	0	0	0	10	1	1
Being quiet	10	0	0	0	10	1	1
Shyness	10	0	0	0	10	1	1
Embarrassment	10	0	0	0	10	1	1
Lack of confidence	10	0	0	0	10	1	1
Self-consciousness	10	0	0	0	10	1	1
Anger	10	0	0	0	10	1	1
Stubbornness	10	0	0	0	10	1	1
Withdrawal from family	10	0	0	0	10	1	1
Withdrawal from friends	10	0	0	0	10	1	1
Exclusion by peers	10	0	0	0	10	1	1
Teasing/Bullying	10	0	0	0	10	1	1

Table 3. Test-retest κ coefficient for Family’s OH-QOLADS (Ever happened)

Item	Absolute score difference				Number of observations	Un-Weighted κ	Weighted κ
	0	1	2	3			
Scheduled activity cancelled	10	0	0	0	10	1	1
Disruption to parent’s employment	10	0	0	0	10	1	1
Isolation from other family members	10	0	0	0	10	1	1
Disruption to family sleeping patterns	9	1	0	0	10	0.821	0.865
Feeling of depression or distress	9	1	0	0	10	0.849	0.894
Self-blame/ Guilt	9	1	0	0	10	0.836	0.896
Worry	7	3	0	0	10	0.516	0.595
Anger	10	0	0	0	10	1	1
Arguments	10	0	0	0	10	1	1
Jealousy	10	0	0	0	10	1	1

Table 4. Test-retest K coefficient for Family’s OH-QOLADS (Last year)

Item	Absolute score difference				Number of observations	Un-Weighted κ	Weighted κ
	0	1	2	3			
Scheduled activity cancelled	10	0	0	0	10	1	1
Disruption to parent’s employment	10	0	0	0	10	1	1
Isolation from other family members	10	0	0	0	10	1	1
Disruption to family sleeping patterns	10	0	0	0	10	1	1
Feelings of depression or distress	9	1	0	0	10	0.836	0.896
Self-blame/ Guilt	9	1	0	0	10	0.836	0.896
Worry	7	3	0	0	10	0.508	0.605
Anger	10	0	0	0	10	1	1
Arguments	10	0	0	0	10	1	1
Jealousy	10	0	0	0	10	1	1

Poster 1

The Development and Testing of an Oral Health-Related Quality of Life Measure for Children / Adolescents with Down Syndrome





Background & Literature Review

People with intellectual disabilities experience a considerable segment of the population that might benefit with their history of higher disease prevalence rates after adaptive medical care, and several studies have shown that their life expectancy has been improved. It is feared that the presence of the disability by itself increases the opportunities of many factors such as health, education and social services. Maintaining health is very important for this people as it is for the general population in order to improve the quality of prolonged life. People with disabilities are entitled to the same standards of health and care as the general population. However, studies have shown that people with intellectual disabilities experience poorer general and oral health compared to the general population (1-5). Additionally, over the years, studies have confirmed that oral health is integral to the general health and therefore to the quality of life.

Studies on oral health related quality of life have revealed that oral health influences psychological wellbeing and satisfaction (6-10) in the general population, and there is no reason to suggest that there is any difference for people with intellectual disabilities. However, this cannot be confirmed due to the scarcity of the studies aimed at assessing oral health-related quality of life among this group.

It seems that there is a gap in the literature in terms of studying the impacts of oral health status on quality of life of people with intellectual disabilities.

Aims & Objectives

Overall aims

- To develop and test an oral health-related quality of life (OHRQoL) instrument/measure for children/adolescents with Down syndrome.

Objectives of phase 1:

- To explore the views of mothers of children/adolescents with Down syndrome concerning the oral health of their children.
- To assess the perception of how oral health might impact on the children with DS quality of life from the mother point of view.
- To assess the mother perception of how oral health condition of the children with DS might impact on the family's quality of life.

Objectives of phase 2:

- To develop an OHRQoL measure for children/adolescents with DS.
- To evaluate the developed OHRQoL measure.
- To explore, in addition to OHRQoL items, the mothers' perception of the oral health status of their children with DS.
- To assess OHRQoL and health status of group of children/adolescents with DS.
- To compare the findings of OHRQoL questionnaire to the mother perception.

What does disability/ Intellectual disability mean?





According to the World Health Organization, a disability is...
 "Any restriction or lack (resulting from any impairment) of ability to perform an activity in the manner or within the range considered normal for a human being"

Theoretical Model

Methodology

Study design (Mixed method approach)

Study sample: 12-14 years old children with Down syndrome & their mothers, selected from special Down syndrome centres/ schools of Riyadh city of Saudi Arabia.

Phase One Data Collection Methods: semi-structured interviews with 20 mothers of (12-14 years old) children with Down syndrome.

Phase Two Data Analysis: "Thematic" analysis method (11).

Phase Three Data Collection (12): questionnaire development from phase one findings, 50 to 100 children and their mothers will be selected to evaluate the developed questionnaire (12).

Phase Four Data Analysis: Psychometric properties of the "final" OHRQoL questionnaire will be assessed by means of a reliability and validity study.

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Poster 2



Impact of Oral Health on the Quality of Life of Adolescents with Down syndrome: Qualitative Exploration of Mothers' Perceptions

Al-Jameel AM¹, Wolf R², Taskiran G³, Daly B¹



Introduction

Down syndrome is the most common genetic cause of intellectual disabilities (van Triesenburg et al., 2004). In addition, it is the most common chromosomal anomaly among live-born infants with an incidence of 1:500 to 1:1000 (Yang et al., 2002; Cayrol et al., 2008). Individuals with the condition exhibit several clinical characteristics that increase their risk of oral conditions (Fleiss & Shufb, 2001; Harnequin et al., 2002). The impact of oral health conditions on individuals may be related directly to their oral health (such as pain, discomfort, and in severe cases tooth loss), but can also extend to broader effects on personal relationships, emotional status and quality of life. However, there is very little research on the way oral health affects the quality of life of people with Down syndrome. Such information is very important to guide the content provision and evaluation of oral health promotion programs and services initiatives for this segment of population. For example, it could be used in clinical decision making to assess the effectiveness of dental treatment and/or early intervention such as physiotherapy targeting chewing or tongue hypomotility of children with Down syndrome, thereby allowing the substance research agenda.

Aim

This study aimed to explore mothers' perceptions of oral health conditions of their children with Down syndrome, and also to explore how the oral problems impacted different aspects of the life of child with Down syndrome and his/her family.

Methods

Introduction

Child's general health

Perceptions of oral health conditions

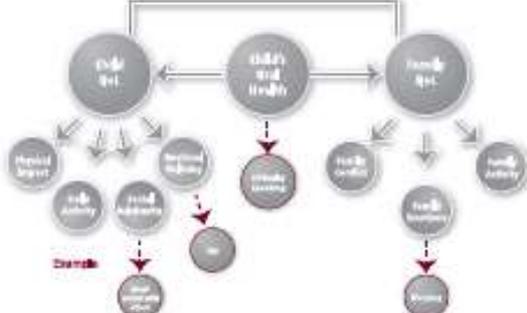
Child's oral health status

Impact of oral health on child

Impact of health on family

Wrap up and ending

Results



Conclusion

References

Acknowledgments

Further information