

THE IMPACT OF HYPODONTIA ON THE ORAL HEALTH-RELATED  
QUALITY OF LIFE IN CHILDREN

By

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## **Abstract**

**Aims:** The purpose of this cross-sectional survey was to evaluate the psychosocial impact of hypodontia in children and to investigate the potential influence of gender, socioeconomic status, severity of hypodontia and the number of retained deciduous teeth on their quality of life.

**Method:** A total of 86 children (36 male, 50 female) with hypodontia, aged 11-14 years were recruited from the Birmingham Dental Hospital, United Kingdom. Thirty subjects without hypodontia and having a low treatment need acted as controls. Children completed the validated Child Perceptions Questionnaire (CPQ) and their parents completed the Parental-Caregiver Perceptions Questionnaire (P-CPQ).

**Results:** The median number of missing teeth in the sample population was 6. There were significant differences in the oral symptoms, functional limitations and the social and emotional well-being reported between the hypodontia and control groups. The overall CPQ scores were significantly higher in children with hypodontia ( $p < 0.001$ ). No significant correlation was detected between the number of missing teeth and the quality of life score. There was no influence found on the CPQ score from gender, socioeconomic status, the site of hypodontia or the presence of retained deciduous teeth. There was moderate correlation between parental and child reported quality of life.

**Conclusions:** Hypodontia can have a significant psychosocial impact on the quality of life of children. This study has implications for our understanding of the effect of hypodontia on the quality of life of children and their parents.

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## Contents

Chapter One: Literature review.....	1
Chapter Two: Method.....	45
Chapter Three: Results.....	53
Chapter Four: Discussion.....	68
References.....	80
Appendices.....	90

## Detailed Contents

<b>Chapter One: Literature review</b> .....	<b>1</b>
1.1 Introduction.....	2
1.2 Prevalence.....	2
1.2.1 Racial differences.....	3
1.2.2 Gender differences.....	3
1.2.3 Tooth predilection.....	3
1.2.4 Severity of hypodontia.....	5
1.3 Aetiology of hypodontia.....	5
1.3.1 Genetic influences.....	5
1.3.2 Environmental factors.....	7
1.3.3 Associated syndromes and systemic conditions.....	7
1.4 Associated skeletal features.....	8
1.4.1 Skeletal pattern.....	8
1.5 Associated dental anomalies.....	9
1.5.1 Microdontia.....	10
1.5.2 Impacted permanent canines.....	10
1.5.3 Maxillary canine/first premolar transpositions.....	11
1.5.4 Taurodontism.....	11
1.5.5 Retained and/or submerged deciduous teeth.....	11
1.6 The management of hypodontia.....	12
1.7 Oral health-related Quality of Life (OHRQoL).....	14
1.7.1 Quality of life (QoL).....	14
1.7.2 OHRQoL.....	14
1.7.3 The relevance of OHRQoL.....	15

1.7.4 Assessment of OHRQoL	16
1.7.5 HRQoL instruments	18
1.7.6 The Child Perceptions Questionnaire	20
1.8 The use of proxies to report on QoL	28
1.8.1 The Parental-Caregiver Child Perceptions Questionnaire	31
1.9 The impact of oral conditions on HRQoL	32
1.9.1 The impact of malocclusion on OHRQoL	33
1.9.2 The impact of hypodontia on OHRQoL	39
1.10 Aims of the study	44
<b>Chapter two: Method</b>	<b>45</b>
2.1 Ethical approval	46
2.2 Study participants	46
2.3 Sample size calculation	47
2.4 Method	48
2.5 Statistical Analysis	50
<b>Chapter three: Results</b>	<b>53</b>
3.1 Characteristics of the sample	54
3.2 Skeletal and dental features	56
3.3 Global health ratings	57
3.4 OHRQoL in subjects with hypodontia	57
3.5 The effect of gender, ethnicity and social deprivation on CPQ score	60
3.6 The effect of retained deciduous teeth on OHRQoL	62
3.7 The effect of site of hypodontia on OHRQoL	62
3.8 Family history of hypodontia and OHRQoL	63
3.9 Associated dental features and OHRQoL	64

3.10 The relationships between the number of missing teeth OHRQoL .....	64
3.11 Correlation between parental and child reported OHRQoL .....	65
3.2 Regression analysis .....	66
<b>Chapter four: Discussion</b> .....	<b>68</b>
4.1 Discussion .....	69
4.2 Conclusions .....	79
References .....	80
Appendices .....	91

### **Legend to Illustrations**

Figure 3.1 The number of missing teeth .....	64
Figure 3.2 Fractional polynomial to show distribution of quality of life scores .....	67

### **Legend to Tables**

Table 3.1 Characteristics of the sample .....	55
Table 3.2 Prevalence of missing teeth by tooth type .....	56
Table 3.3 Prevalence of impacts in CPQ according to group .....	57
Table 3.4 Summary statistics for each domain for children and parents .....	58
Table 3.5 Comparison of CPQ scores in subjects with and without hypodontia .....	59
Table 3.6 Comparison of CPQ scores between mild and severe hypodontia .....	59
Table 3.7 Comparison of CPQ scores between males and females .....	60
Table 3.8 Comparison of CPQ scores between ethnic groups .....	61
Table 3.9 OHRQoL scores in subjects with and without retained deciduous teeth .....	62
Table 3.10 OHRQoL scores for anterior and posterior hypodontia .....	63
Table 3.11 OHRQoL scores in subjects with and without a positive family history of hypodontia .....	63
Table 3.12 OHRQoL scores in subjects with and without other dental features .....	64
Table 3.13 Spearman rank correlation between Child CPQ scores and the number of missing teeth in the total sample and within the hypodontia group .....	65
Table 3.14 Spearman rank correlation between child OHRQoL scores and the corresponding parent scores .....	66

## Appendices

1.	Invitation letter to participants.....	91
2.	Children's information sheet.....	92
3.	Parent/guardian information sheet.....	94
4.	Children's consent form.....	96
5.	Parental consent form.....	97
6.	Hypodontia data collection proforma.....	98
7.	Child Perceptions Questionnaire.....	99
8.	Parental-Caregiver Perceptions Questionnaire.....	108
9.	Summary statistics for CPQ by categories of group.....	123
10.	Summary statistics for P-CPQ by categories of group.....	125

**Chapter One**  
**Literature Review**

## **1. Literature review**

### **1.1 Introduction:**

Hypodontia is defined as the developmental absence of one or more primary or secondary teeth, excluding the third molars (Goodman *et al*, 1994). It is the most common congenital dental anomaly and presents a complex clinical problem. Hypodontia can arise *de novo* but may be apart of a syndrome or associated with other dental anomalies. Oligodontia is the term used to describe the absence of six or more teeth and typically occurs with syndromes or severe anomalies. Anodontia is the complete absence of any dental structures (Meon, 1992) and is usually associated with hypohidrotic ectodermal dysplasia.

### **1.2 Prevalence**

The reported prevalence of hypodontia from retrospective studies shows wide variation between the samples studied. This can be partly explained by differences in the population studied. Difference in the age of the sample groups may account for some of the variation in the reported prevalence of the condition: permanent teeth may not have calcified in younger patients and in older children teeth may have been extracted. Reported prevalence rates also vary according to the method of assessment of hypodontia; those utilising radiographs may report a greater prevalence than studies in which only clinical assessment is undertaken.

Hypodontia most frequently affects the permanent dentition. Agenesis of the primary dentition is rare, and if present is usually seen in the incisor region: it is invariably

associated with agenesis of the succeeding permanent teeth (Meon, 1992, Nik-Hussien, 1989).

### **1.2.1 Racial differences**

The prevalence of hypodontia shows wide variation between racial groups, ranging from 2.6% in Saudi Arabia (Salama and Megid, 1994) to 11.3% (O'Dowling and McNamara, 1990) in Ireland. The prevalence in the United Kingdom was found to be 4.3% by Rose (1966) and 4.5% by Brook (1974). The prevalence of hypodontia in the primary dentition is lower than in the permanent dentition and varies between 0.5% in the Icelandic population (Magnusson, 1984) to 2.4% in the Japanese population (Yoneza *et al*, 1997).

### **1.2.2 Gender differences**

A higher prevalence of hypodontia has been reported in females with a ratio of 3:2 (Brook, 1974; Bergstrom, 1977). A similar gender difference has been documented with microdontia, which is closely associated with hypodontia.

### **1.2.3 Tooth predilection**

Several studies have reported that symmetrical hypodontia is more common (Bergstrom, 1977; Lai and Seow, 1989) and that teeth are more commonly missing from the maxillary arch than the mandibular arch (Cua-Benward *et al*, 1992; Wong *et al*, 2006).

The most common missing teeth in the primary dentition are the mandibular and maxillary lateral incisors. Daugaard-Jensen *et al* (1997) in a radiographic survey of 213 children with absent teeth in the primary dentition demonstrated that hypodontia was also a feature in the permanent dentition, but that the pattern was different, with premolars more commonly missing in the permanent dentition and with more units absent.

In the permanent dentition the most common missing tooth is the third molar. The reported prevalence of missing third molars is between 9 and 20% (Bishara and Andreasen, 1983). The reported prevalence for other teeth varies according to the population studied. In Caucasians the most frequently missing teeth are the mandibular second premolars and the maxillary lateral incisors. In the United Kingdom the most common missing tooth is the mandibular second premolar (Rose, 1966; Brook, 1974). Grahnen (1956) reported a prevalence of 2.8% for missing mandibular premolars, followed by 1.6% for maxillary lateral incisors, 0.23% for maxillary second premolars and 0.08% for mandibular incisors. In Swedish and Asian populations the mandibular incisor is the most commonly missing tooth (Davis, 1987). Endo *et al* (2006) reported the mandibular second premolar to be the most common missing tooth in a Japanese study but found an increased incidence of severe hypodontia and agenesis of the mandibular lateral incisor in cases of even minor hypodontia. There have been case reports of missing permanent canines but this is rare.

#### **1.2.4 Severity of hypodontia**

The majority of patients have one or two missing teeth. Ten per cent present with four or more missing teeth and less than 1% will have severe hypodontia with six or more missing teeth (Larmour *et al*, 2005). Cases with severe hypodontia are more likely to be associated with a syndrome.

### **1.3 Aetiology of hypodontia**

Hypodontia is a multi-factorial condition with both genetic and environmental factors implicated in the aetiology. It is a consequence of space limitation, physical obstruction or disruption of the dental lamina, functional abnormalities of the dental epithelium or failure of initiation of the underlying mesenchyme (Nunn *et al*, 2003).

#### **1.3.1 Genetic factors**

A number of twin and family studies have confirmed the role of genetics in the aetiology of hypodontia but there is controversy regarding whether it is due to a single gene defect or if it is a result of a polygenic trait. Hypodontia can follow autosomal dominant, autosomal recessive or x-linked patterns of inheritance.

In 1984, Brook conducted a large family study and suggested that in the majority of cases hypodontia has a polygenic inheritance pattern with relatives close to the proband having a higher risk of hypodontia. The proportion of relatives affected depended on the severity of the condition and environmental factors. Another study showed that the prevalence of hypodontia was 50% in siblings or parents of children with hypodontia, compared to 6% in the general population (Grahnen, 1956). In contradiction to the work

by Brook (1984), a study by Burzynski (1983) suggested that hypodontia is caused by a single gene defect inherited in an autosomal dominant fashion but with variable gene penetration.

It has been proposed that tooth shape and position are determined by homeobox genes expressed in the neural crest cells derived from mesenchyme of the branchial arches. A number of these different homeobox genes have been implicated in the aetiology of hypodontia including Msx1, Msx2, Pax9, (Cobourne, 2007) Dlx1, Dlx2, Lhx6, Lhx7 and Axin2 (Mostowska *et al*, 2006).

Studies in transgenic mice have identified homeobox genes Msx1 and Msx2 to have a role in tooth development with Msx1 being more important in specification and induction, and Msx2 responsible for the later development of tooth buds. Mice with the Msx1 gene knocked out had complete failure of tooth development, arresting at the bud stage (Satokata and Maas, 1994). Msx1 has been shown to be responsible for a specific type of severe tooth agenesis and is not implicated in the more common cases of incisor or premolar agenesis suggesting that the different presentations have different aetiologies (Lidral and Reising, 2002).

Pax9 is another homeobox gene, essential during the later stages of tooth development. Mice with mutations in the Pax9 gene exhibit arrested tooth development at the bud stage.

### **1.3.2 Environmental factors**

Studies on twins have demonstrated a variable expression of hypodontia in monozygotic twin pairs, confirming that environmental factors can modify phenotype expression. The environmental factors implicated in the aetiology of hypodontia include infections, for example Rubella, drugs, metabolic or hormonal disturbances and irradiation.

### **1.3.3 Associated syndromes and systemic conditions**

Hypodontia most commonly occurs in its non-syndromic familial form, as an isolated trait, appearing sporadically or in a familial fashion (Cobourne, 2007). However, it can also occur accompanying a genetic disease as part of a recognised clinical syndrome. A large number of syndromes have associated with hypodontia. These are discussed below.

#### **Ectodermal dysplasia**

Ectodermal dysplasia (ED) is the condition most commonly linked with hypodontia. It occurs as a result of disturbances in the ectoderm of the developing embryo and can be inherited either as a sex-linked or autosomal dominant condition with an incidence of 1 in 100,000. The x-linked condition classically consists of the triad of hypohydrosis, hypotrichosis and hypodontia. In the most severe cases complete anodontia is observed, but more commonly patients present with severe hypodontia.

In any patient presenting with severe hypodontia, particularly when the central incisors and the first molars are absent, a possible association of ED should be considered even if only discreet other features of the syndrome are present.

### **Cleft lip and palate**

Cleft lip and palate is the most common craniofacial abnormality with an incidence of 1:700 live births in the UK. The incidence of hypodontia is higher in this group than in the general population. A prevalence rate of 37% has been reported with increasing prevalence with increasing severity of the cleft (Laatikainen and Ranta, 1994). The maxillary lateral incisor in the cleft site is the most frequently affected tooth, most likely owing to a localised disturbance of the dental lamina.

### **Van Der Woude syndrome**

This is an autosomal dominant disorder comprising cleft lip and palate, paramedian lip pits and severe hypodontia. The teeth missing in order of frequency are the upper second premolars, lower second premolars and the upper lateral incisors (Rizos and Spyropoulos, 2004).

### **Downs Syndrome**

Patients with Downs syndrome have a higher prevalence of hypodontia compared to the general population with a reported prevalence of up to 63% (Kumaska *et al*, 1997). The maxillary lateral incisors are the most commonly missing teeth.

### **1.4 Associated skeletal features**

A typical facial appearance is often observed in patients with hypodontia, possibly associated with growth disturbances (Worsaae *et al*, 2007). Typical extra-oral features include:

- Bimaxillary retrognathism
- Midface hypoplasia
- Anterior rotation of mandible
- Decreased vertical and transverse dimensions of the dental arches
- Reduced lower anterior face height
- Increased naso-labial angle
- Deep labio-mental fold

#### **1.4.1 Skeletal pattern**

The literature presents conflicting findings in relation to skeletal pattern. Some studies have shown that patients with a hypodontia are more likely to present with a class III skeletal pattern and a retrognathic maxilla, with increasing severity as the number of missing teeth increases (Sarnas and Rune, 1983). Conversely other studies have reported that patients with hypodontia have a normal skeletal I relationship (Yuksel and Ucem, 1997). In the vertical dimension hypodontia patients tend towards reduced lower anterior face height as a consequence of limited alveolar bone growth (Bondarets and McDonald, 2000). This characteristic becomes more marked with increased severity of hypodontia (Chung *et al*, 2000). Another study (Cua-Benward *et al*, 1992) showed the highest prevalence of hypodontia in Class II malocclusions - accounting for 52.3% of the sample - followed by class I (33.7%) and class III (14%).

#### **1.5 Associated dental anomalies**

Anomalies in tooth number are often associated with other dental anomalies (Cobourne, 2007). These include:

- Microdontia
- Conical crown shape usually affecting the incisors and canines
- Enamel hypoplasia
- Delayed dental development
- Delayed eruption
- Molar taurodontism
- Prolonged retention of primary teeth
- Infraocclusion of primary teeth
- Tooth impaction
- Ectopic eruption and transpositions
- Lack of alveolar bone

### **1.5.1 Microdontia**

Hypodontia has a strong association with microdontia. Congenitally absent lateral incisors are often associated with a diminutive contralateral tooth. This may indicate a common genetic aetiology between the two conditions. This, combined with hypodontia can result in multiple diastemas and rotations of the adjacent teeth.

### **1.5.2 Impacted permanent canines**

There is an increased incidence of impacted maxillary canines with hypodontia. This is most likely related to a reduced dimension or absence of the maxillary lateral incisor. It was proposed in the guidance theory (Becker *et al*, 1984) that in cases of diminutive or absent lateral incisors the eruption guidance for the maxillary canine is lost. Studies have reported that canine impaction is 13 times more likely in cases with missing lateral

incisors and that the incidence of impaction in hypodontia cases is 26 – 42% (Brin *et al*, 1986; Peck *et al*, 1996).

### **1.5.3 Maxillary canine/first premolar transpositions**

It has been reported that in 50% of patients with a canine and premolar transposition have hypodontia and therefore the two conditions may share similar genetic aetiologies (Peck *et al*, 1993).

### **1.5.4 Tauradontism**

Taurodontism is the term used to describe teeth with an enlarged and vertically elongated pulp chamber extending down into the roots. Taurodontism predominantly affects the mandibular molars and is reported to have an increased prevalence in patients with hypodontia (Seow and Lai, 1989).

### **1.5.5 Retained and/or submerged deciduous teeth**

Hypodontia of the permanent dentition is characterised by retention of the deciduous teeth beyond the normal age of shedding. Haselden *et al*, (2001) looked at root resorption in retained deciduous teeth and found the survival of deciduous first molars to be poor. The life span of deciduous second molars was unpredictable with some showing very good survival rates.

Retained primary teeth preserve alveolar bone in the hypodontia site, which is important if implant supported restorations are going to form part of definitive management. They

also act as a space maintainer and prevent undesirable drift of adjacent teeth should it be deemed preferable to maintain the hypodontia space.

Retained deciduous teeth may become ankylosed and infraoccluded over time. In the absence of the permanent successor resorption of the deciduous tooth is delayed (Haselden *et al*, 2001). This has been reported to be the most frequent dental anomaly associated with hypodontia (Kirzioglu *et al*, 2005).

### **1.6 The management of hypodontia**

The successful management of hypodontia requires a multidisciplinary approach with input from paediatric dentistry, restorative dentistry, orthodontics and oral surgery aided by diagnostic set-ups (Larmour *et al*, 2005). Broad treatment options are to accept, redistribute or close the spaces in sites where teeth are absent. The decision is dependent on a number of patient and dental factors including: (Forgei *et al*, 2005; Thind *et al*, 2005)

- Patient factors
  - Patient concerns and motivation
  - Medical history
  - Social history
  - Financial position
- Facial features
  - Overall skeletal pattern
  - Soft tissue pattern
  - Lip line

- Gingival aesthetics
- Dental features
  - Malocclusion
  - Number and site of teeth to be replaced
  - Tooth size
  - Adjacent tooth condition
  - Adjacent tooth position
  - Periodontal condition of adjacent teeth
  - Amount of alveolar bone

The advantages of space closure are the permanence of the finished result, improved gingival contour and potentially reduced costs. However, teeth adjacent to the site of hypodontia may require crown modification to resemble the teeth that they are replacing. This may include recontouring, composite build-ups, localised tooth bleaching and crown lengthening (Rosa and Zachrisson, 2001). These modifications have short and long-term financial implications and restorations will require maintenance.

Space-opening will require prosthetic replacement of the missing teeth, which may be a fixed or removable option. The fixed option, generally preferred by patients, includes conventional or resin-retained bridges and implant-supported prosthesis. Prosthetic replacement of teeth requires long-term maintenance and can be expensive for patients.

## **1.7 Oral health-related quality of life (OHRQoL)**

### **1.7.1 Quality of life (QoL)**

Over the past two decades research has been carried out on the impact of medical conditions on the quality of life of patients. The World Health Organisation Quality of Life (WHOQoL) group defined QoL as the “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 goals, expectations, standards and concerns’. QoL is complex and multidimensional and has been shown to be related to OHRQoL (Taylor *et al*, 2009).

### **1.7.2 OHRQoL**

Oral health has been defined as the standard of oral and related tissues that allows individuals to eat, speak and socialise without active disease, discomfort or embarrassment and contributes to general well-being (DoH, 1994).

Locker and Allen (2007) further defined OHRQoL as “the impact of oral disorders on aspects of everyday life that are important to patients and persons, with those impacts being of sufficient magnitude, whether in terms of severity, frequency or duration to affect an individual’s perception of their life overall.” OHRQoL has also been defined as the “cyclical and self-renewing interaction between the relevance and impact of oral-health in everyday life” (Gregory *et al*, 2005).

OHRQoL encompasses different domains including survival of the dentition, absence of disease or symptoms, appropriate physical functioning, absence of pain and discomfort, emotional functioning associated with smiling, social functioning, satisfaction with oral

health and an absence of social or cultural disadvantages due to oral status (Cunningham and Hunt, 2001).

### **1.7.3 The relevance of OHRQoL**

Over recent years increasing importance has been placed on quality assurance and clinical effectiveness in the National Health Service (NHS) and with the introduction of clinical governance greater emphasis has been placed on patient involvement. The 1997 White Paper, *The New NHS* suggested that we should look at improvements in health in terms of fairer access to care, quality and outcomes of treatment and the views of the patients (Secretary of State for Health, 1997). Since patient involvement has become important it is necessary to utilise measures, which not only reflect the clinical problems but also the patient's experiences. Understanding the impact on QoL is important for several reasons. It provides insight into potential consequences of the condition on the day-to-day lives of patients and facilitates management of such cases.

At present the severity of dental malocclusion and the treatment outcome are usually assessed using occlusal indices and cephalometric measurements. Occlusal indices are used to rank the severity of the malocclusion and to grade treatment outcome. The sole use of these traditional methods of assessing oral health with clinical indices, which concentrate on the absence of disease, have been criticised as they do not consider the patient's perceptions from an aesthetic, functional or psychological point of view. In the UK, orthodontic need is assessed utilising the Index of Orthodontic Treatment Need (IOTN). This has a dental health component and an aesthetic component. Patients with hypodontia are scored on the dental health component as IOTN 5h if there are two or

more missing teeth in one or more quadrant or 4h if there is one missing tooth in one or more quadrant. This index places patients with hypodontia in the category of great need and entitles them to treatment on the NHS. The IOTN and other traditional indices represent a unidimensional aspect of oral health and do not consider the effect of hypodontia on the patient's quality of life. A key strength of QoL measures is that they embody the notion that the patient's perspective has equal importance to that of the clinician and should be considered when evaluating the consequences of disease and treatment outcomes.

#### **1.7.4 Assessment of OHRQoL**

QoL instruments were developed to help evaluate both the physical and psychosocial impact of disease. Assessing the impact of disease on an individual can improve communication between patients, parents and the dental team. It offers an insight into the consequences of adverse oral health conditions on children's lives and the lives of their families and provides information on the consequences of the problem for the patient on daily basis (Cunnigham and Hunt, 2001).

OHRQoL can also provide assistance in needs assessment, prioritisation of care and evaluation of outcomes for clinicians. In addition, they serve as an adjunct to clinical measures in evaluating the outcomes of treatment and in the development of clinical guidelines. Information compiled from QoL studies may also help with the development of health policies.

The purpose of the HRQoL instrument is not just to measure the presence and severity of disease symptoms but to also show the impact of the disease on that individual (Cunningham and Hunt, 2001). Over the last two decades the number of validated measures to assess psychological health has risen. Several measures of OHRQoL have been developed for adult populations. These include the General Oral Health Assessment Index and the Oral Health Impact Profile. Measures designed for assessing adult OHRQoL may not be suitable for children because of lack of validity. Assessing QoL in children is challenging and therefore, it was previously assessed using parents' views because of concerns that children's responses to a questionnaire would not meet psychometric standards due to limitations in children's cognitive capacities and communication (Barbosa and Gavião, 2008a).

Childhood is a period of significant change in psychosocial awareness and in their dental and facial features. Also children's cognitive development varies such that the wording of questions and their relevance and meaning to children of similar ages can differ. Developmental changes in a child over time can invalidate repeat measurement (Eiser *et al*, 2000). In addition, a child's concept of oral health and well-being is influenced by a number of variables including gender and social class: Locker *et al* (2007b) found that children from low-income households had poorer QoL than those from high-income households.

There is an argument therefore to minimise these errors by asking a parent or guardian to report on the quality of life of the child. This raises the question of how well proxy reports represent the views of the child and parental awareness (McGrath *et al*, 2004).

Recently a number of instruments to obtain quality of life information from children have been developed which take into account developmental differences. At the age of 11 years children enter a period of adolescence characterised by increasing centrality of the peer group, clique dynamics and their pre-occupation with other's views of themselves (Bee, 1998). This illustrates the point that it is not appropriate to have a single questionnaire designed for children of all ages. Age specific questionnaires are recommended for different age ranges: 6-7, 8-10 and 11-14 year olds. These age groups are considered fairly homogenous in terms of cognitive development (Jokovic *et al*, 2002).

It is imperative that the validity and reliability of instruments to be used in children be tested amongst child populations. It is also important that measures are as brief as possible and contain the minimum number of items to assess the QoL whilst minimising the burden on participants (Slade, 1997).

### **1.7.5 HRQoL instruments**

From the viewpoint of contemporary definitions of oral health, radiographic and clinical measures such as IOTN and the Peer Assessment Rating Index (PAR), have serious limitations. Clinical measures may only reflect the opinion of the orthodontist without taking into account the problems experienced by the patient (O'Brien *et al*, 2006).

Instruments used to measure HRQoL can be categorised as disease-specific or generic. Generic instruments allow for comparisons between different conditions or populations. These can be further categorised into two types: health profiles and preference-based

index measures. The latter provides a single score or index, which represents the total impact of physical, emotional and social well-being on QoL. The former consist of multiple items that are grouped into domains of health and functioning. This allows clinicians and researchers information on the impact of diseases on different aspects of HRQoL (Connolly and Johnson, 1999).

Instruments can be either administered by a researcher or be self-administered by the patient either in a clinical setting or by mail. Administration by a researcher reduces the number of missed responses and errors, but requires more resources and may introduce researcher bias. A compromise is to have a supervised self-completed questionnaire.

In selecting a QoL instrument it is necessary to consider its psychometric properties. That is the ability of the instrument to measure psychological constructs. Two important features, necessary in any QoL instrument are validity and reliability (Connolly and Johnson, 1999). Validity determines if the instrument measures what it purports to measure. This can be further divided into content, criterion and construct validity. Content validity assesses if an instrument samples all the relevant domains. Criterion validity is the correlation of a scale with some other measure of the disease under study. Construct validity looks at the relationship of a variable to other variables. A discriminatory instrument is able to determine differences in HRQoL as they are related to their disease.

Reliability is the proportion of variance that is attributable to the true score of the latent variable. It is evaluated with internal consistency and test-retest, which looks at repeat

measurements over time. Internal consistency assesses the agreement of items in instruments with multiple items and confirms that the individual items are related and measure the same thing.

A literature review conducted by Barbosa and Gavião (2008a) identified 12 studies that had used a validated OHRQoL instrument. From this they identified only three well-validated QoL instruments. These included two age-specific versions of the Child Perceptions Questionnaire (CPQ): one suitable for 8-10 year olds and the other designed for 11-14 year olds; the child-oral impacts on daily performances (Child-OIDP) and Child Oral Health Impact Profile (COHIP). These authors concluded that when appropriate questionnaires are used, valid and reliable information on the OHRQoL can be obtained from children and it is appropriate to supplement normative indices to identify patients with psychosocial need. The COHIP was developed using the item pool used in the development of the CPQ but it deviates from the CPQ by inclusion of positive health constructs (Broder et al, 2007). The questionnaire consists of 34 items divided into 5 domains including oral health, functional well-being, social-emotional well-being, school environment and self-image. It has been validated in the United States (Broder and Wilson-Genderson, 2007; Dunlow *et al*, 2007). Testing of this instrument showed discriminant and convergent validity and excellent test-retest reliability.

#### **1.7.6 The Child Perceptions Questionnaire**

The CPQ forms one component of the Child Oral Health Quality of Life (COHQoL) questionnaire, which consists of a number of measures for children and their parents. It

was originally developed by Jokovic *et al* (2002) at the Faculty of Dentistry, University of Toronto with both orthodontic and paediatric dentistry patients. The aim was to develop a measure for a range of dental, oral and oro-facial disorders for use in clinical trials and evaluation studies (Jokovic *et al*, 2002). It consists of three age-specific versions of the CPQ. One designed for 6-7 year olds, one for 8-10 year olds and another for 11-14 year olds. It also consists of a Parental-Caregiver Perceptions Questionnaire (P-CPQ) and a Family Impact Scale. These measures are generic and may be used for a number of oral conditions.

The COHQoL questionnaires were developed using the theory of measurement and scale development. This is the process for development and evaluation of OHQoL measures as outlined by Guyatt *et al* (1986). Items for the instrument were developed using a two-stage process. Initially a preliminary pool of 46 questions covering a range of health domains were developed after reviewing current oral health and child health instruments. The relevance, clarity and comprehensiveness of the questions were evaluated in a content study by an expert panel of health professionals who regularly treated children with oral conditions. Input was also sought from parents of children with various oral conditions. In-depth interviews were conducted with 11 children. Following this a modified question list was developed.

Questions for the final instrument were selected using an item study. This seeks to rank items that are of greatest importance to the subjects who are completing the questionnaire. Participants involved in the development and evaluation of the COHQoL were from three groups, each suffering from a different clinical condition: dental

disease (primarily dental caries), malocclusion and oro-facial disorders (primarily cleft lip/palate).

The CPQ is a self-administered questionnaire originally composed of 37 items divided into four domains encompassing oral symptoms, functional limitations, emotional well-being and social well-being (peer interaction, schooling and leisure activities). The questions ask about the frequency of events experienced by children in the last 3 months, with the following options: never = 0; once or twice = 1; sometimes = 2; often = 3 and everyday or almost every day = 4. Summing the response codes generates domain scores and an overall CPQ score. The CPQ also includes global ratings of the child's oral health and the extent to which the oral condition affects his or her life. The first question is "would you say that the health of your teeth, lips, jaws and mouth is...?" The response options range from 'excellent' to 'poor'. The second question is "How much does the condition of your teeth affect your life overall?" The response ranges from 'not at all' to 'very much'.

The CPQ for 8-10 year olds is divided into 3 components: "oral health status awareness", "oral social self-image" and "social confidence and well-being". Positive associations between the OHRQoL as determined by this instrument and self-esteem have been shown (Humphris *et al*, 2005). The reliability and construct validity for the CPQ 8-10 year olds has also been confirmed by Humphris *et al* (2005) who found that it has acceptable reliability, substantial internal consistency (Cronbach's alpha = 0.87) and excellent test-retest reliability. Summary measures correlated with the global health ratings indicating acceptable criterion validity (Marshmann *et al*, 2005).

The performance of the CPQ 11-14 year olds was assessed in a validity and reliability study (Jokovic *et al*, 2002). It performed well as a discriminative measure and was able to distinguish between the three study groups producing significant differences in the CPQ scores. Children with oro-facial conditions had the highest score and the paediatric group the lowest. They also found significant correlations between CPQ scores and global ratings of health and overall well-being. The Cronbach's alpha for the whole sample was 0.91 demonstrating excellent internal consistency. Cronbach's alpha for the different domains ranged from 0.64 to 0.86 and intra class correlations from 0.79 to 0.88. Although, there was some variability of social well-being with time, overall the test-retest reliability was acceptable. The overall Intraclass Correlation Coefficient was 0.90 indicating almost perfect agreement. They found that the impact of child oral and orofacial conditions and on function and psychosocial well-being is substantial in children aged 11-14 years and concluded that children are able to give psychometrically acceptable accounts of impact on quality of life. The questionnaire performed well as a discriminant measure (Jokovic *et al*, 2002).

The CPQ has also been shown to be valid and reliable in a UK orthodontic population: Marshman *et al* (2005) tested the CPQ on 89 children from Sheffield. The children ranged from relatively healthy to having a variety of oral conditions including dental caries, malocclusion, gingival conditions and enamel opacities. Participants who failed to answer more than one seventh of the questionnaire were excluded. Criterion validity was examined by comparing the global rating of oral health to the overall CPQ. Construct validity was assessed by testing association between the CPQ scores and the

clinical data. Internal consistency was tested by Cronbach's alpha and test-retest reliability by means of Intraclass Correlation Coefficient. They found that summary measures of CPQ related well with global oral health rating and impact on life overall demonstrating acceptable criterion validity. The number of impacts correlated with the number of missing teeth and the missing teeth due to caries.

The CPQ has also been validated as a measure of the impact of malocclusion on children's oral health by O'Brein *et al* (2006) in a longitudinal study on a sample of school children from Manchester, UK. They found that CPQ scores corresponded to differences in the need for orthodontic treatment as measured by the IOTN. Scores were greater for children who thought that their teeth needed straightening. They found no relationship between CPQ score and the IOTN aesthetic component.

Locker *et al* (2007c) carried out a study on 141 children to assess the association between CPQ scores and clinical and self-perceived measures of malocclusion. Children were asked to complete the long and short-forms of the CPQ to assess concerns about the condition of their teeth. Study models were taken and rated according to the PAR Index and the Dental Aesthetic Index (DAI), an indicator of malocclusion severity. Correlations between CPQ and the orthodontic indices ranged from 0.26-0.31. There was a clear gradient in CPQ scores with increasing PAR scores but this was less clear for the DAI. The association between CPQ and children's self-ratings of oral health was significant. Scores from both the short and long forms of the questionnaire showed positive correlations with the PAR and DAI suggesting that the CPQ was sensitive to variations in the severity of malocclusions.

Foster Page *et al* (2005) also found a clear gradient in the mean CPQ mean with four categories of treatment need: minor/none, definite, severe and handicapping. In contrast, de Oliveria *et al* (2008) found that a child's perceived need for orthodontic treatment was supported by OHQoL measures but not by an objective measure of malocclusion.

A possible reason for a lack of correlation between clinical indicators and OHRQoL measures is that they are not measuring the same parameter. In addition, there is no significant correlation between PAR and ICON with the number of missing teeth (Shelton *et al*, 2008) which has been shown to be related to OHRQoL (Wong *et al* (2006). Inconsistencies between normative and subjective perceptions of malocclusion highlight the broader conceptual distinction between disease and health. They demonstrate the inadequacy of clinical measures to assess people's feeling and satisfaction with their teeth. Tsakos (2008) proposed applying the socio-dental approach to health, which requires the assessment of normative need, subjective perceptions of QoL and health behaviours.

A limitation of the CPQ is that it does not elicit the specific cause for the QoL impact, which may be due to a number of oral conditions. Another potential disadvantage is that it does not include positively worded items or positive health concepts, like those found in the COHIP. The need for positive items in QoL studies has been investigated by Locker *et al* (2007d). This study assessed the performance of negatively and positively worded items in questionnaires to measure child and parent perceptions of child

OHRQoL. The results indicated a greater number of ‘Don’t know’ responses and missing values for the positively worded items (39.1%) compared to negatively worded items (16.3%). A similar difference was seen in parental responses (49% and 10.2% respectively). The mean parental and child scores were significantly higher for the positive items than the negative items and resulted in a higher prevalence of impacts. Further, there was poor agreement between positively worded and negatively worded items for both parents and children. Agreement between child and parental scores was better for negatively-worded items. The findings of this study suggest that negative and positive items may not be measuring the same construct and that in the context of health-related quality of life questionnaires, positively worded items do not function well. This would suggest that it is better to use a scale in which all items are unidirectional.

The length of the original CPQ may also limit its use in clinical settings because of the burden placed on respondents and lengthy data analysis. A short form would broaden its application; reduce the time taken for and financial costs of data collection and interpretation. It would also reduce the risk of total and individual item non-response. To facilitate its use in clinical settings and population-based health surveys, the CPQ for 11-14 year olds was shortened and tested for cross-sectional validity and reliability and compared with the original instrument in terms of measurement sensitivity and discriminative properties (Jokovic *et al*, 2006). No guidelines have been published on how short a questionnaire should be. Four items is considered to be the minimum that is necessary to control for random error and allow analysis within different domains.

Jokovic *et al* (2006) reduced the CPQ to a 16-item version and an 8-item measure with two items per domain, however, the latter would not allow within-domain analysis. Item impact and stepwise regression methods were used to produce two versions of each short-form. In the regression method, the dependent variable was the overall CPQ score and the independent variable was the individual question. The advantage of the item impact approach is that it selects items, which are of most importance to the people who will be completing the questionnaire.

Criterion validity, construct validity and internal consistency of the short forms were tested on the responses of 123 children from paediatric dentistry, orthodontics and a group with oro-facial conditions, predominantly cleft lip and palate. Sixty-five children of the 125 completed the questionnaire two weeks later to provide data on test retest reliability. The results demonstrated that the short forms developed had good criterion validity and almost perfect correlation with the long forms of the questionnaire. Overall the short forms showed discriminant construct validity as they were able to detect differences in the three study groups but this was not statistically significant for the 16-item version developed using regression analysis. Even with the reduced number of items the CPQ demonstrated considerable measurement sensitivity and good correlational construct validity as all short forms were significantly correlated with ratings for oral health and overall well-being. All questionnaires showed substantial internal consistency and high test-retest reliability. The Cronbach's alpha for the 16-item version developed using the impact method was 0.83 and the Intraclass Correlation Coefficient was 0.77.

## **1.8 The use of proxies to report on QoL**

As previously discussed, assessing the impact of health status on QoL in children is complex because childhood is a time of change in physical appearance and psychosocial awareness. Studies have suggested that difficulties can be minimised by having a proxy: a parent or guardian (Theunissen *et al*, 1998). However, using proxies has disadvantages in that children may give different accounts of their health status compared to their parent or caregiver. Caregivers may over or underestimate the importance of certain aspects, as they do not observe school-aged children throughout the day. One reason for looking at parent-child agreement is to assess if the parent can be used as a proxy.

Studies to date have shown conflicting findings. One study found low agreement (Le Coq *et al*, 2000) compared to others, which have demonstrated moderate correlation and that correlation varies with the domain being assessed (Theunissen *et al*, 1998). There is better correlation with functional limitation than emotional and social well-being. Pantell and Lewis (1987) also found high correlation between parent and child when measuring concepts of functional status but poorer correlation with emotion or pain.

A study by Jokovic *et al* (2004) found good correlation between P-CPQ and CPQ 11-14 year olds. Wilson-Genderson *et al* (2007) assessed child-caregiver concordance regarding OHRQoL using the COHIP. They found low to moderate correlation between child and caregiver for the sample overall. Both the paediatric and orthodontic groups were more likely to agree and disagree with the caregivers in a similar manner. Children in the craniofacial group were more likely to report better QoL compared to their

parents. They found no differences in concordance between caregiver and child ratings on the child's OHRQoL associated with age, gender or ethnicity.

The agreement between mothers' and fathers' scores and child scores was investigated by Zhang *et al* (2007b) using 71 sets of parents and their children. Children had lower overall QoL scores than mothers and fathers. Fathers had higher mean overall CPQ and domain scores than mothers but this was not statistically significant. At the group level mothers and fathers overestimated the impact of malocclusion on all domains except oral functional limitation. Mothers and fathers overestimated the impact on emotional and social well-being by similar amounts. This suggests that either parent's view will suffice as complementary. At the individual level there was poor agreement between parents and their children, and mothers and fathers tended to disagree on their child's OHRQoL. At the group level both mothers and fathers tend to agree on perception of their child's oral health status but other research has shown lower knowledge for fathers than mothers (Jokovic *et al*, 2004).

Jokovic *et al* (2003) carried out a study to assess agreement between mothers and children concerning the child's OHRQoL. A total of 42 pairs of mothers and children completed the P-CPQ and CPQ. They found that for overall scores there was substantial agreement between mother and child pairs but the Intra Class Correlation for the emotional and social well-being subscales showed only moderate correlation. Children reported an average worse OHRQoL than their mothers with an overall score of 26.2 versus 22.6 but there was no systematic under-reporting in mothers' assessments. The Intraclass Correlation Coefficients for emotional and social well-being were lower than

oral symptoms and functional limitations. This is similar to the findings in the Child Dental Health Survey in which pain was the most frequently reported oral impact that parents were aware of (Nuttall *et al*, 2006). These findings can be explained by the fact that emotional and social well-being domains address issues that mothers may not have insight into. They concluded that mothers should not be used as proxies for their children when considering individual ratings.

Differences in child and parental reports may reflect true differences in perspectives but may also be due to a lack of insight of parents into the lives of their children. It may be that parents' knowledge is lacking with regard to relationships and feelings outside the home (Jokovic *et al*, 2004). It is important to remember that the concepts measured by the parental and child instruments are not identical. The CPQ measures the child's perception of his/her OHRQoL whereas the P-CPQ measures the parent's perception of OHRQoL.

Even though parents' reports may be incomplete due to lack of knowledge in certain areas they still provide useful information, even when children's responses are available. Valid and reliable information can still be obtained from parents using appropriate questionnaires (Barbosa and Gavião, 2008c). The relatively low correlation of some of the subscales shows that parents cannot simply be used as proxies, however, because of the role they play in the decision making of the healthcare of their child making their assessments should still be sought. Parallel reporting is increasingly recommended in assessing QoL in children.

### **1.8.1 The Parental-Caregiver Perceptions Questionnaire (P-CPQ)**

The P-CPQ forms another component of the COHQoL questionnaire. It was also developed using the methodology advocated by Guyatt *et al* (1986) to allow parallel parent-child reporting and examine the extent to which parental assessments correspond to those obtained from children. It is a self-completed questionnaire for parents/guardians of children with the oral condition under study. The scoring system for the responses is the same as the CPQ but in addition it also includes a 'Don't Know' (DK) response. The reason for the inclusion of DK response is that forced responses are invalid and increase random error. Allowing DK responses reduces the number of missing values and allows exploration of parent's knowledge of their children's OHRQoL.

A study by Jokovic *et al* (2004) evaluated four methods of dealing with the DK response when calculating domain and overall scores. These were:

1. Deletion in which only questions without DK responses were included for the analysis
2. Imputation of item means in which the mean item score for the entire sample was inserted
3. Replacement of the DK response with a score of zero
4. Adjustment made to the overall score according to the number of items with a DK response

DK responses represented 5.8% of the total number of responses with 46.6% of parents selecting one or more DK response. The child's age had an independent effect on the number of DKs, with parents of children in the 11-14 year old group having twice as

many DK responses compared to the 8-10 year old group. This may reflect the fact that as children get older they spend less time under parental supervision and are less likely to share experiences with their parents. The majority of DK responses were in the social well-being and emotional well-being domains (Jokovic *et al*, 2004).

Overall, the P-CPQ was shown to have good construct validity, internal consistency and test-retest reliability. The four methods of managing DK responses did not affect the correlation of the overall P-CPQ score with global health ratings. The level of the agreement between parental and child reports on the overall scale was also unaffected by the different methods of managing DK responses (Jokovic *et al*, 2004).

### **1.9 The impact of oral conditions HRQoL**

Recently there has been greater emphasis on oral health-related quality of life research. A recent review of the literature suggested that there is a relationship between oral health and OHRQoL (Barbosa and Gavião, 2008b). Some studies included in the review only reported a weak correlation between oral conditions and OHRQoL, which was attributed to low disease levels in the sample, the diseases causing only low impacts or variation in the impact on OHRQoL according to culture and education. They also reported that dental caries and fluorosis were both highly correlated with reduced OHRQoL. They concluded that patients with craniofacial conditions including cleft lip and palate reported negative impacts on their QoL. Six out of the seven studies in the review, which investigated the impact of malocclusion, found a significant association between QoL and malocclusion. Gherunpong *et al* (2004) have shown that periodontal problems also affect children's OHRQoL.

### **1.9.1 The impact of malocclusion on OHRQoL**

In 1962 the World Health Organisation pronounced that a dental anomaly should be regarded as requiring treatment if the defect is likely to impact on an individual's physical or emotional well-being. With recent changes in the NHS it has become increasingly important to demonstrate the benefits of treatment and the impact on health outcomes to patients and purchasers of dental services. Due to the high demand for orthodontic treatment, it may be assumed that the benefits of orthodontic treatment are self-evident but a review in the 1970s (Shaw *et al*, 1980a) failed to demonstrate the benefit on oral health or psychological well-being. This review was the stimulus for significant research, which commenced in 1981 by Shaw *et al* (2007). They conducted a 20-year prospective longitudinal cohort study to investigate the effects of malocclusion on oral health and social well-being. 1018 children aged 11-12 years underwent extensive health and psychosocial well-being assessment.

The initial results (Kenealy *et al*, 1988) supported a previously proposed relationship between attractiveness and teacher expectations (Shaw *et al*, 1982) and between attractiveness and self-esteem (Kenealy *et al*, 1991). The study did not, however, demonstrate that children with a visible malocclusion were likely to be socially or psychologically disadvantaged. The results need to be interpreted with caution as the psychological effects reported are dependent on the measures available at the time of the study. Three hundred and thirty seven participants from the original sample were followed-up at 20 years. Subjects with a need for orthodontic treatment who had obtained orthodontic care had better tooth alignment, were more satisfied with their

occlusion, had higher self-esteem and better QoL when compared to those with untreated needs. However, when baseline self-esteem was accounted for there were no significant differences between the two groups and orthodontic treatment was concluded to have little positive impact on psychological health and a lack of treatment when there was a prior need did not lead to psychological issues in adulthood (Shaw *et al*, 2007). The possibility of bias in this study due to the high dropout rate cannot be excluded and failure to demonstrate a positive psychological change may be a reflection of the standard of treatment.

Facial and dental appearance has become increasingly important in today's society. The media in general reinforces the message that "beauty is good" (Kiyak, 2008). Fashion models on television and in magazines display teeth that are perfectly aligned. These popular images, which are often achieved with software image manipulation, can generate dissatisfaction and self-criticism amongst viewers. Attractive children are perceived by others to be more intelligent and have better social interaction and receive more positive treatment (Langlois *et al*, 2000). There is some evidence, which suggests that patients with unaesthetic occlusal traits can attract unfavourable social responses such as nicknames, harassment and teasing from school children (Shaw *et al*, 1980b). Deviation from facial and dental norms in children may also have an unfavourable affect on self-esteem and self-confidence (Shaw *et al*, 1991; Shaw *et al*, 1986). We are aware that children who experience teasing have lower self-esteem and are less sure of them selves (Lansdown *et al*, 1991). Self-esteem is further affected by QoL. Shaw *et al*, (2007) reported that 65% of the variance in self-esteem could be explained by perception of quality of life, and the perception of attractiveness.

Studies have consistently reported improvements in oral health-related quality of life in orthognathic patients (Hatch *et al*, 1998; Motegi *et al*, 2003; Lee *et al*, 2008 Esperao *et al*, 2010). The evidence for routine non-surgical cases is conflicting. Zhang *et al* (2006) reviewed the literature on the impact of malocclusion on OHRQoL. They concluded that the evidence regarding the impact of malocclusion and orthodontic treatment is conflicting. Taylor *et al* (2009) found no significant differences in the COHQoL scores in children needing orthodontic treatment as assessed according to the Index of Orthodontic Complexity and Need and those who did not. This supports the view that the complexity of the malocclusion does not influence the OHRQoL score. Patients who had received a course of interceptive treatment reported better oral health than those who had received no treatment but there was no significant difference in the general QoL score or the COHQoL.

Some recent studies have shown a link between malocclusion and OHRQoL (Foster Page *et al*, 2005). They observed an impact on emotional and social well-being among orthodontic patients aged 11-14 years. Mandall *et al* (2000) found greater psychosocial impact with increasing severity of malocclusion. Studies have also shown that OHRQoL is correlated with self-perceived need. Children who expressed a concern about their teeth had worse emotional and social impacts (Kok *et al*, 2004). A longitudinal study on a sample of school children from Manchester, UK reported that malocclusion may have a significant impact on the QoL of children. They used the CPQ to show that OHRQoL was lower for the more severe grades of the IOTN dhc and for children who thought that their teeth needed straightening (O'Brien *et al*, 2006).

The differences were in the emotional and social well-being domains. This may be because the most common reason for seeking orthodontic treatment is to address aesthetics and malocclusion is unlikely to result in oral symptoms and functional limitations.

A Brazilian study on a sample of 225 subjects aged 12-15 years old reported that patients who sought orthodontic treatment had worse OHQoL as evaluated by the OHIP than subjects who had never sought orthodontic treatment. They also had more severe malocclusions as assessed by IOTN and greater aesthetic impairment. Those that sought orthodontic treatment were 3.1 times more likely to have worse QoL than those who did not. No gender differences were observed in the number of impacts overall (Feu *et al*, 2010) and there was no effect from socio-economic status.

Another study showed that pre-treatment CPQ scores in a group of orthodontic patients were low but wearing fixed appliances increased the CPQ score. There was an increase in oral symptoms and functional limitations but an improvement in emotional well-being over the 6 months study period. The greatest deterioration was in the first week, which, may be attributed to discomfort and oral symptoms, such as soft tissue trauma that may arise from fixed appliances being placed (Zhang *et al*, 2007a).

Some studies have shown that the type of malocclusion can have an impact on QoL (Traebert *et al*, 2007) whereas others have found no differences between the types of malocclusion (Johal *et al*, 2007; O'Brien *et al*, 2007). O'Brien *et al* (2007) evaluated the CPQ for children with malocclusion in a cross-sectional survey of UK children aged

11-14 years. The malocclusion group consisted of individuals presenting with any one of three occlusal traits including crowding, overjet and hypodontia. They found a significant difference between the OHRQoL in the malocclusion and acceptable occlusion groups in the total CPQ scores. There were no statistically significant differences between the malocclusion subgroups. At the subscale level these differences were only significant for emotional and social well-being domains and not for oral symptoms and functional limitations. There was significant correlation between the total CPQ scores and overall well-being and patient satisfaction. They concluded that different occlusal traits have a negative impact on the OHRQoL of an adolescent.

Johal *et al* (2006) assessed the impact of two occlusal traits on the quality of life of children aged 13-15 years. They sampled 30 patients with an increased overjet and 30 patients with spacing in the upper labial segment. Thirty patients with a class I incisor relationship and a well-aligned upper labial segment were recruited as a control group. The participants and their parents were asked to complete the CPQ and the P-CPQ respectively. There was a highly significant difference in the CPQ scores between children in the control group and the malocclusion groups demonstrating that an increased overjet or spacing in the upper anterior segment can impact on OHRQoL. No difference was detected between children in the increased overjet and spaced dentition groups demonstrating that both malocclusion traits have highly similar significant impacts. Parents of children in the increased overjet group and spaced dentition groups reported greater impacts on QoL than parents of children in the control group. There was no difference in parental reported OHRQoL between both study groups suggesting

that both an increased overjet and spacing have a significant negative impact on the OHRQoL of the children and their families.

A lower OHRQoL does not always equate to a greater desire for treatment. Researchers in the UK administered the CPQ and the Aesthetic Component of the IOTN. The AC scores of children were lower than those given by dentists but only 35% of patients who rated their AC negatively wanted to undergo treatment (Kok *et al*, 2004). The correlation between the CPQ and self-rated AC was significant but low.

As studies have demonstrated only a modest relationship between clinical indicators and CPQ (Marshman *et al*, 2005, Locker *et al*, 2007c), it is reasonable to assume that the relationship between malocclusion and OHRQoL is mediated by other factors. Agou *et al* (2008) examined the relationship between self-esteem and OHRQoL in a sample of children attending an orthodontic screening appointment. Children completed the CPQ and a self-esteem subscale of the Child Health Questionnaire. The DAI was used to determine clinical need. They found a tenuous relationship between DAI and CPQ suggesting that increasing severity of malocclusion does not always lead to a direct increase in CPQ score. Patients with higher self-esteem reported better OHRQoL whereas the impact of malocclusion on children with low self-esteem was substantial.

The relationship between clinical variables and HRQoL is not direct. They are mediated by personal, social and environmental factors and child development. The child may not be able to relate oral health, illness and quality of life. A literature review found good construct validity in all child perceptions of OHRQoL but also reported that children's

understanding of oral health is also affected by a number of variables including age, gender, race and education (Barbosa and Gavião, 2008a). Mandall *et al* (2000) evaluated the effect of ethnicity, social deprivation and normative orthodontic need on self-perceived aesthetics and need for treatment. They found children with a higher clinical need for treatment perceived themselves as worse off than their peers with lower need. They found that children who were socially deprived had a greater aesthetic impacts score as measured on an Oral Aesthetic Subjective Impact Scale (OASIS) but this was not an important variable with respect to self-perception. Ethnicity did not influence orthodontic aesthetic self-perception.

Another variable, which, has been shown to affect the QoL score, is age-related experiences (Gherunpong *et al*, 2004). Shedding of deciduous teeth or space due to unerupted permanent teeth may result in high prevalence of oral impacts therefore poor OHRQoL may reflect part of the natural process of tooth exfoliation (Weintraub, 1998). The influence of individual personality traits on QoL cannot be ignored and may be able to explain why some patients with minor problems seek treatment whereas others are willing to accept a severe deviation from the norm.

### **1.9.2 The impact of hypodontia on OHRQoL**

There have only been a few studies investigating the functional, social and behavioural implications for the patient and his/her immediate family. Hobkirk *et al* (1994) conducted a retrospective study looking at the concerns of 451 patients with hypodontia. The most common complaints were spacing, poor aesthetics and some of the patients were aware of missing teeth. Only 8.7% of patients reported functional problems.

Wong *et al* (2006) conducted the first QoL study into the impact of hypodontia. They used the CPQ in a study of 25 patients with severe hypodontia, with 4 or more missing teeth, who were attending the Paediatric and Orthodontic Unit at Prince Phillip Dental Hospital, University of Hong Kong. The number of missing teeth ranged from 4 to 20 with a mean of 8.9. Overall there was a predilection for the maxilla with the most common missing tooth being the upper lateral incisor. The most common missing tooth in the mandible was an incisor, which is unsurprising as patients in this study were of southern Chinese origin, and a higher prevalence of missing mandibular incisors has been reported in this ethnic group.

All of the children reported one or more impacts as evaluated by the CPQ. All children reported oral symptoms as a result of hypodontia, 88% reported functional limitations, 88% had some impact on emotional well-being. One hundred per cent of their sample reported one or more social impacts. The mean CPQ score was 29.0 and they found a significant association between the number of missing teeth and the CPQ score. They found that the overall correlation between the number of missing permanent teeth and the overall CPQ score was 0.54. When the retained primary teeth were accounted for the correlation was 0.94.

A further study by Locker *et al*, (2010) also demonstrated the impact of hypodontia on OHRQoL. They carried out a study on children aged 11-14 years also utilising the CPQ as their instrument for assessment. Patients were recruited from the orthodontic clinics at the Hospital for Sick Children and the Bloorview MacMillan Children's Centre

Travelling Clinics, Canada. They employed a convenience sampling approach and recruited 36 children. The number of missing teeth ranged from 1-14 with a mean number of missing teeth of 6.8. Two thirds of the group had six or more missing teeth. They found premolars to be the most common missing teeth (58%) followed by anterior teeth (26%). 5.6% of children rated their oral health as excellent and 25% as very good. Thirty-six per cent rated their oral health to be either fair or poor. Fifty-eighty per cent of patients said that their teeth/mouth had no or very little effect on their life overall but 11% said their life was 'very much' affected. Seventy-eight per cent of children reported experiencing one or more impact 'Often' or 'Everyday/Almost everyday'. The main impacts in their study were related to function with 61% reporting some functional limitations. 28% experienced oral symptoms, 19% reported some impact on emotional well-being and 17% on social well-being. The mean overall CPQ score was 22.3 (standard deviation = 14) with a range of 4 to 69. They found no gender or age related differences in the prevalence of impacts or severity scores. A study by Wong *et al* (2006) they found no significant correlation between the number of missing teeth and overall and sub-scale scores. They divided their sample into two groups according to which children would be eligible for public funding for their orthodontic treatment (five or more missing teeth) and those that would not be (less than missing teeth). They found no differences in the total and domain scores between the two groups.

In a study by Locker *et al* (2010) a high prevalence of functional and psychosocial impacts in subjects with hypodontia was reported. The impact of hypodontia reported in this study is, however, lower than that reported by Wong *et al* (2006) in which, 100% of subjects reported one or more impacts overall and one or more impacts in the social

well-being domain. This may be because the study by Wong *et al* (2006) used a more lenient threshold for defining prevalence. They included items scored 'sometimes' in reporting the prevalence of an impact, however, the mean CPQ score was also higher, 29.0 compared to 22.3 in the study by Locker *et al* (2010). Disparity may be explained by cultural and demographic differences between the groups. Locker *et al* (2010) compared their results to the CPQ scores and impacts reported by subjects in an earlier study (Jokovic *et al*, 2002) with dental caries and malocclusion. They found that hypodontia had a greater impact on OHRQoL than the latter two conditions. The impacts reported for dental caries utilising the CPQ was 43.7% and 61.5% for malocclusion. The impact of hypodontia was lower than for oro-facial conditions in which 84.6% of children with clefts of the lip and/or palate reported an impact. Comparisons in these studies have to be interpreted with care as they are based on small, convenience samples.

Both the above studies employed a convenience sampling approach without a sample size calculation. The Hong Kong study (Wong *et al*, 2006) had no control group for comparison and the Canadian study used a group from a previous study to make comparisons. The findings of these studies, therefore, have to be interpreted with caution.

A more robust cross-sectional survey to determine the psychosocial impact of hypodontia using the CPQ was conducted by Laing *et al*, 2010 in London. They sampled 62 children aged 11-16 years with hypodontia. Sixty-one children without hypodontia but with an IOTN dhc score of 4 or 5 were assigned to a control group. The

mean number of missing teeth was 4.52. They found no statistically significant difference in the CPQ scores between the hypodontia (total CPQ = 26.8) and routine orthodontic groups (total CPQ = 28.5). There was some association between the number of missing teeth and difficulty chewing. In this study hypodontia did not affect the psychosocial status of patients as compared to other malocclusion traits.

The impact of hypodontia on the OHRQoL of adults has been considered in a sample of adults aged 16-25 years (Meaney *et al*, 2011). They found that with age patients became more aware of their condition. All participants in the study who had received treatment were satisfied with the outcome and reported reduced anxiety about the appearance of their teeth once treatment was nearly complete.

### **1.10 Aims of the study**

The aim of this study was to investigate the impact of hypodontia on the oral health-related quality of life in children.

The null hypotheses were:

- (i) There is no difference in the oral health-related quality of life scores reported in children with and without hypodontia.
- (ii) There is no difference in the quality of life scores reported between children with hypodontia and that reported by their parents.
- (iii) There is no correlation between the quality of life scores and the number of missing teeth.
- (iv) There is no difference in the OHRQoL scores in hypodontia patients with and without retained deciduous teeth.

## **Chapter 2**

### **Method**

## **2. Method**

### **2.1 Ethical approval**

This research protocol was granted ethical approval by The Black Country Research Ethics Committee (REC reference number 09/H1202/74). Research and Development approval was obtained from the University of Birmingham.

### **2.2 Study Participants**

This was a cross-sectional survey of children presenting with hypodontia. All participants were recruited by the principle researcher (SK) from September 2009 to April 2011. Consecutive children attending the multidisciplinary hypodontia clinic at the Birmingham Dental Hospital who satisfied the inclusion criteria were invited to participate in the study by letter (appendix 1). Each subject underwent an orthodontic assessment by a single investigator (S.K.). The criteria for inclusion in the study were:

- patients between 11 and 14 years of age;
- radiographically confirmed hypodontia of at least two teeth excluding the third molars;
- willing to participate in the study and
- English speaking

Children presenting with less than 6 missing teeth were assigned to the mild hypodontia group and those presenting with 6 or more missing teeth to the severe hypodontia category.

A second group was selected to serve as a control group. This group were recruited from orthodontic new patient clinics at Birmingham Dental Hospital. The inclusion criteria for the control group were:

- patients between 11 and 14 years of age;
- presence of all permanent teeth;
- IOTN dental health component score of 2 or 3 and
- willing to participate in the study

Exclusion criteria for all children in the study were:

- associated medical history or craniofacial anomaly;
- previous orthodontic treatment;
- previous restorative treatment to address hypodontia;
- restorations in the upper labial segment;
- not accompanied by a parent or guardian or non-English speaking;
- other dental problems including dental caries, periodontal disease and enamel and dental defects and
- unwilling to participate in the study

### **2.3 Sample size calculation**

A sample size calculation proposed a sample of 28 patients in each of the 3 groups to determine a minimum effect size of 0.75 for the difference in QoL score between hypodontia and no hypodontia and between mild and severe hypodontia. Cohen (1969) defined a medium effect size as 0.5 and a large effect size as 0.8. Norman *et al*, 2003

also concluded that the threshold of discrimination for changes in HRQoL is approximately half a standard deviation.

Power was set at the 80% level with  $\alpha=0.05$ . Accounting for the fact that incomplete questionnaires would have to be excluded from the final analysis, participants were recruited until at least 30 patients were obtained in each group.

## **2.4 Method**

The study was explained verbally to each patient and the parent/legal guardian. Written information sheets outlining details of the study were provided for the child (appendix 2) and the parent (appendix 3). If the parent/legal guardian and the patient were willing to taking part in the study written consent was obtained from both the child (appendix 4) and the parent (appendix 5).

The following demographic details were recorded:

- Age
- Gender
- Ethnic group
- Postcode

Data on gender, age and ethnic group were obtained to evaluate any confounding effect on the relationships between clinical status and OHRQoL. The postcode was used to obtain the patient's Index of Multiple Deprivation Score using the Office for National Statistics ([www.neighbourhood.statistics.gov.uk](http://www.neighbourhood.statistics.gov.uk)). This is based on the Indices of Deprivation 2010, which provides a relative measure of deprivation in small areas

across England. It is composed of 38 indicators, which are grouped into 7 domains to produce an overall index. The domains are income, employment, health, education, crime, access to services and living environment. The country is divided into homogenous small areas of relatively even size (around 1500 people) known as lower super output areas (LSOA). A deprivation score is calculated for each area. The scores can also be used to rank the LSOAs according to their deprivation score. An area has a higher deprivation score than another one if the proportion of people living in that area is classed as more deprived (Communities and Local Government, 2011).

The following clinical details were recorded using a proforma (appendix 6).

- Skeletal pattern
- Incisor relationship
- Total number of missing teeth
- Site of missing teeth
- Family history of hypodontia
- Presence of retained primary teeth
- Other dental features including increased overjet, increased overbite, microdontia, spacing, hypoplasia, abnormal morphology

Each child was asked to complete the shortened form of the CPQ for children aged 11-14 years (appendix 7). The CPQ consists of 17 questions divided into 4 health domains: oral symptoms, functional limitations, emotional well-being and social well-being. The questions assessed the child's opinions and the perceived views of peers about his or her dental appearance. It also covers behavioural problems at home and at school. The

response options and scores range from never 0; once or twice 1; sometimes 2; often 3 and everyday or almost every day; 4. The scores for each domain are added together to give a total quality of life score.

The format of the study was a supervised self-completed questionnaire to be completed on the clinic at the time of the appointment. One parent/guardian of each of the patients in the hypodontia group was asked to complete the P-CPQ (appendix 8).

Subjects were given as much time as required to complete the questionnaire and were reassured that the results would remain anonymous. Children and parents were requested not to confer when completing the questionnaire.

## **2.5 Statistical analysis**

All participants were allocated an identification number and the coded data was entered onto a bespoke database (Microsoft Access, 2007) for analysis. Overall CPQ and domain additive scores for each child were calculated by summing the response codes for the individual items. Scores were also generated by counting the number of impacts reported 'everyday/almost every day' or 'often'. These scores allow the impact of hypodontia to be reported in terms of prevalence, whereas the mean overall CPQ and domain scores give an indication of severity.

Analysis of the data was conducted using Stata Statistical Software: Release 11 2009 (College Station, TX: Statacorp LP). The data was initially analysed using descriptive statistics. Differences in sample characteristics between the hypodontia groups and the

control group were evaluated using the Chi-square test and one-way ANOVA as appropriate. Normality of the distribution of the quality of life scores was checked using qq plots and the Shapiro-Wilk test. Non-parametric statistical methods were used as CPQ scores were not normally distributed. Mann-Whitney U tests were used to determine differences in the 4 domains and overall CPQ scores between the hypodontia and control group and between the mild and severe hypodontia groups. Mann-Whitney U tests were also used to test for differences in CPQ scores according to gender, ethnic group and between hypodontia affecting the anterior teeth and that confined only to the posterior teeth. The effects of retention of the deciduous teeth were investigated by comparing CPQ scores in patients with retained deciduous teeth with those in which the deciduous teeth had been shed.

Spearman rank correlation coefficients were calculated to evaluate the correlation between the number of missing teeth and the CPQ score and to investigate the correlation between social deprivation and CPQ score. Spearman rank correlation was also used to check for agreement between global oral health ratings and total CPQ and P-CQP scores. Correlation between parental and child scores was tested with Spearman Rank correlation.

Linear regression was used to evaluate the association between hypodontia and CPQ scores, adjusting for age, gender and ethnicity, as well as the association between number of missing teeth and QoL scores. Fractional polynomial regression was used to explore the dose-response function of this association. Furthermore, linear regression with interaction terms was used to evaluate whether the association between child and

parent CPQ scores was modified by gender of the parent, social deprivation or family history of hypodontia. For all linear regression analyses, CPQ scores were transformed using the square root to achieve normality. All statistical tests were two-sided at a significance level of  $\alpha=0.05$ .

## **Chapter 3**

### **Results**

### 3. Results

#### 3.1 Characteristics of the sample

Recruitment for this study commenced in October 2009 and was completed in April 2011. All patients approached agreed to participate in the study and completed the questionnaire on the clinic. Thus a 100% response rate for children was achieved. Eighty-four parents of the patients with hypodontia completed the P-CPQ questionnaire. 2 parents were unable to complete the questionnaire due to language barriers.

Table 3.1 demonstrates the socio-demographic characteristics of the sample. The sample comprised of 86 patients with hypodontia, 43 with severe hypodontia (18 male, 25 female) and 43 with mild hypodontia (18 male, 25 female). The mean age was 12.6 years in the mild hypodontia group and 12.4 years in the severe hypodontia group (range 11-14 years). There were no statistically significant differences between groups with regard to age, child and parent gender and social deprivation scores. There was a higher proportion of White British children in the hypodontia groups compared to the control group ( $p=0.036$ ). Compared to children with mild hypodontia, children with severe hypodontia were more likely to have a combination of missing posterior and anterior units ( $p=0.001$ ), more likely to have retained primary teeth ( $p=0.003$ ) and more likely to have a positive family history of hypodontia ( $p<0.001$ ).

**Table 3.1: Characteristics of the sample**

Parameter	Mild	Group Severe	Control	Total	p value
Gender, % (n)					
Male	42 (18)	42 (18)	43 (13)	42 (49)	1.00*
Female	58 (25)	58 (25)	57 (17)	58 (67)	
Age, mean (sd)	12.6 (1.1)	12.4 (1.2)	12.5 (1.0)		0.737**
Ethnicity, % (n)					
White British	93 (40)	86 (37)	67 (20)	84 (97)	0.036*
Pakistani	5 (2)	9 (4)	17 (5)	9 (11)	
Afro-Caribbean	2 (1)	2 (1)	17 (5)	6 (7)	
White European	0 (0)	2 (1)	0 (0)	1 (1)	
Parent, % (n)					
Father	21 (9)	33 (14)		27 (23)	0.221*
Mother	79 (33)	67 (28)		73 (61)	
Site, % (n)					
Combination	44 (19)	84 (36)		64 (55)	0.001*
Posterior	30 (13)	12 (5)		21 (18)	
Anterior	26 (11)	5 (2)		15 (13)	
Retained primary teeth, % (n)					
Yes	67 (29)	93 (40)		80 (69)	0.003*
No	33 (14)	7 (3)		20 (17)	
Other features, % (n)					
Yes	37 (16)	53 (23)		45 (39)	0.129*
No	63 (27)	47 (20)		55 (47)	
Family history, % (n)					
Yes	19 (8)	56 (24)		37(32)	<0.001*
No	81(35)	44(19)		63 (54)	
Deprivation score, mean (sd)	23.6 (17.5)	21.8 (15.7)			0.620**

\*Chi-squared test

\*\* Oneway Anova

### 3.2 Skeletal and dental features

The sample of hypodontia included an even spread of malocclusions, 29% (n=25) class I, 26% (n=22) class II div 1, 22% (n=19) class II div 2 and 23% (n=20) class III malocclusion.

The most common missing teeth (Table 3.2) were the upper lateral incisor (16.4%), the lower second premolars (16.0%) and the upper second premolars (13.6%). In this sample a total of 587 teeth were missing: 308 maxillary and 271 mandibular. 59% (n=51) of patients had missing all four third molars. Eighty per cent (n=69) of patients had at least one missing third molar but as the sample was children aged 11-14 years this could not be fully ascertained. Third molar development is often not radiographically evident until the patient reaches early adolescence. Patients with hypodontia may also have delayed dental development.

Forty-seven per cent of patients with hypodontia had other dental features including microdontia, hypoplasia, generalised spacing or a midline diastema.

**Table 3.2: Prevalence of missing teeth by tooth type**

Tooth	7	6	5	4	3	2	1
Upper % (n)	6.6 (39)	1.4 (8)	13.6 (80)	9.0 (53)	5.1 (30)	16.4 (96)	0.3 (2)
Lower % (n)	8.0 (47)	1.2 (7)	16.0 (94)	4.3 (25)	1.2 (7)	5.5 (32)	11.4 (67)

### 3.3 Global health ratings

Twenty nine per cent (n=25/86) of children in the hypodontia group rated the health of their teeth, lips and mouth (global oral health rating) as ‘fair’ or ‘poor’ compared to only 2 subjects in the control group. Ten per cent (n=9/86) of children with hypodontia and 10% of patients in the control group (n=3/30) reported that the condition of their teeth, lips, jaws or mouth affected their lives either ‘a lot’ or ‘very much’. Forty two per cent (n=36/86) in the hypodontia group were dissatisfied with their teeth compared to 16.7% in the control group (n=5/30).

Construct validity was assessed by comparing overall and domain scores. Spearman rank correlation showed that the overall CPQ score and the P-CPQ scores were correlated to the global oral health questions ( $\rho=0.59$ ,  $p<0.001$  for CPQ and  $\rho=0.515$ ,  $p<0.001$  for P-CPQ).

### 3.4 OHRQoL scores in subjects with hypodontia

Frequency tables were derived from responses scored as ‘often’ or ‘everyday’ (Table 3.3).

**Table 3.3 Prevalence of impacts in CPQ according to group**

	Control group	Mild hypodontia	Severe hypodontia
Oral Symptoms, % (n)	0 (0)	23 (10)	35 (15)
Functional Limitations, %(n)	3 (1)	21 (9)	40 (17)
Emotional well-being, % (n)	13 (4)	28 (12)	37 (16)
Social well-being, % (n)	20 (6)	28 (12)	30 (13)

The individual item scores for the CPQ (appendix 9) and the P-CPQ (appendix 10) were added together to produce a score for each domain. The domain scores for each group were summed to produce an overall CPQ or P-CPQ score (Table 3.4).

**Table 3.4: Summary statistics for each domain for children with hypodontia**

Domain	Children
Oral Symptoms <i>median (p25, p75)</i>	4 (2,6)
<i>range (min, max)</i>	(0,10)
Functional limitation <i>median (p25, p75)</i>	3 (2,6)
<i>range (min, max)</i>	(0, 10)
Emotional well-being <i>median (p25, p75)</i>	5 (2,9)
<i>range (min, max)</i>	(0, 19)
Social well-being <i>median (p25, p75)</i>	3 (1,5)
<i>range (min, max)</i>	(0, 15)
Total QoL score <i>median (p25, p75)</i>	16 (10,23)
<i>range (min, max)</i>	(0, 43)

Qq plots and the Shapiro-Wilk's test indicated that the children's ( $p < 0.001$ ) and parents' ( $p < 0.001$ ) CPQ scores were not normally distributed. Data transformation was carried out using the square root of the mean to allow regression analysis. This resulted in a normal distribution of the total CPQ ( $p = 0.74$ ) and P-CPQ scores ( $p = 0.28$ ). The median total CPQ score was 16 in the hypodontia group with a range of 0 to 43. There was one participant with a floor effect, that is an overall score of zero, but there were no participants with ceiling effects, scoring the maximum. Mann-Whitney U tests for 2 independent samples showed that this was significantly higher ( $p < 0.001$ ) than the control group: median CPQ: 8 (range 1 to 30). This difference was significant for all domains (Table 3.5).

**Table 3.5: Comparison of CPQ scores in subjects with and without hypodontia**

Domain	Hypodontia	Control	p-value*
Oral Symptoms <i>median (p25, p75)</i>	4 (2,6)	2 (1,4)	0.002
<i>range (min, max)</i>	(0,10)	(0, 7)	
Functional limitation <i>median (p25, p75)</i>	3 (2,6)	1 (0,3)	0.001
<i>range (min, max)</i>	(0,10)	(0, 6)	
Emotional well-being <i>median (p25, p75)</i>	5 (2,9)	2.5 (2,5)	0.009
<i>range (min, max)</i>	(0,19)	(0, 11)	
Social well-being <i>median (p25, p75)</i>	3 (1,5)	1.5 (0,4)	0.03
<i>range (min, max)</i>	(0, 15)	(0, 11)	
Total CPQ <i>median (p25, p75)</i>	16 (10,23)	8 (5,13)	<0.001
<i>range (min, max)</i>	(0, 43)	(1, 30)	

\*p-value for comparison between hypodontia and control group (Mann-Whitney U test)

The overall median CPQ (Table 3.6) score for the patients with severe hypodontia was not significantly greater than for patients with mild hypodontia (p=0.12). None of the domain scores were significantly different between the two groups.

**Table 3.6: Comparison of CPQ scores between mild and severe hypodontia**

Domain	Severe	Mild	p-value*
Oral Symptoms <i>median (p25, p75)</i>	4 (3,6)	4 (2,5)	0.42
<i>range (min, max)</i>	(0,10)	(0, 9)	
Functional limitation <i>median (p25, p75)</i>	4 (2,7)	3 (1,4)	0.07
<i>range (min, max)</i>	(0,10)	(0, 10)	
Emotional well-being <i>median (p25, p75)</i>	6 (3,9)	4 (1,9)	0.24
<i>range (min, max)</i>	(0,17)	(0, 19)	
Social well-being <i>median (p25, p75)</i>	3 (2,6)	2 (1,5)	0.24
<i>range (min, max)</i>	(0, 10)	(0, 15)	
Total CPQ <i>median (p25, p75)</i>	17 (13,27)	14 (8,21)	0.12
<i>range (min, max)</i>	(0, 36)	(2, 43)	

\*p-value for comparison between mild and severe hypodontia (Mann-Whitney U test)

### 3.5 The effect of gender, ethnicity and social deprivation on CPQ score

There was no significant difference ( $p=0.88$ ) in QoL scores between males and females (Table 3.7) in overall CPQ scores and domain scores.

**Table 3.7: Comparison of CPQ scores between males and females**

Domain	Male	Female	p-value*
Oral Symptoms <i>median (p25, p75)</i>	4.5(2.5, 5.5)	4 (2,6)	0.81
Functional limitation <i>median (p25, p75)</i>	3 (1, 6.5)	3 (2,5)	0.72
Emotional well-being <i>median (p25, p75)</i>	5 (1,9)	5 (2,9)	0.71
Social well-being <i>median (p25, p75)</i>	3 (2, 5.5)	2 (1,5)	0.48
Total CPQ <i>median (p25, p75)</i>	16 (10, 24.5)	16 (10,23)	0.88

\*p-value for comparison between males and females with hypodontia (Mann-Whitney U test)

Due to the small number of non-white subjects in the sample (Table 3.1) for the purpose of analysis of the CPQ score the patients were divided into two groups: white (including British and European whites) and non-white (including Pakistani and Afro-carribean). Two sample Mann-Whitney U tests showed that the overall CPQ scores were not significantly different ( $p=0.65$ ) between the two groups (Table 3.8).

**Table 3.8: Comparison of CPQ scores between ethnic groups**

Domain	White	Other	p-value*
Oral Symptoms <i>median (p25, p75)</i>	4 (2,6)	5 (3,8)	0.36
Functional limitation <i>median (p25, p75)</i>	2 (3, 6)	2 (0.5,4.5)	0.29
Emotional well-being <i>median (p25, p75)</i>	5 (2,9)	6 (2,8)	0.91
Social well-being <i>median (p25, p75)</i>	3 (1,6)	2 (1.5,3)	0.52
Total CPQ <i>median (p25, p75)</i>	16 (10, 23)	13.5 (7.5,26.5)	0.65

\*p-value for comparison of CPQ scores according to ethnicity (Mann-Whitney U test)

Spearman rank correlation between child CPQ scores and social deprivation showed that there was no significant correlation between the two for any domain: oral symptoms ( $\rho=0.006$ ,  $p=0.95$ ), functional limitation ( $\rho=-0.02$ ,  $p=0.82$ ), social well-being ( $\rho=0.09$ ,  $p=0.38$ ), emotional well-being ( $\rho=0.07$ ,  $p=0.53$ ) and for the overall score ( $\rho=0.08$ ,  $p=0.43$ ).

### 3.6 The effect of retained deciduous teeth on OHRQoL

The sample was divided into groups to assess the effects of retained deciduous teeth on the CPQ score. Although the overall CPQ score was lower in patients with retained deciduous teeth (Table 3.9) this was not statistically significant ( $p=0.73$ ).

**Table 3.9: OHRQoL scores in subjects with and without retained deciduous teeth**

Domain	Retained deciduous teeth		p-value*
	Yes	No	
Oral Symptoms <i>median (p25, p75)</i>	5 (3,6)	3 (2,5)	0.46
Functional limitation <i>median (p25, p75)</i>	2 (3,6)	3 (1,4)	0.90
Emotional well-being <i>median (p25, p75)</i>	5 (2,8)	6 (3,12)	0.19
Social well-being <i>median (p25, p75)</i>	3 (1,5)	2 (3,5)	0.69
Total CPQ <i>median (p25, p75)</i>	16 (10, 23)	15 (10,29)	0.73

\*p-value for comparison between children with and without retained deciduous teeth (Mann-Whitney U test)

### 3.7 The effect of site of hypodontia on OHRQoL

Subjects were categorised according to those that had missing anterior teeth and those with hypodontia in only the posterior segments (Table 3.10). Subjects with teeth missing in only the posterior sextants had a lower CPQ score compared with patients with hypodontia affecting the anterior sextants. This was statistically significant only for social well-being ( $p=0.04$ ) but the difference in overall CPQ scores was not significant ( $p=0.21$ ).

**Table 3.10: OHRQoL scores for anterior and posterior hypodontia**

Domain	Location of hypodontia		p-value*
	Anterior	Posterior	
Oral Symptoms <i>median (p25, p75)</i>	4 (2.5,6)	4.5 (2,5)	0.87
Functional limitation <i>median (p25, p75)</i>	3 (1.5,6)	3.5 (2,5)	0.99
Emotional well-being <i>median (p25, p75)</i>	6 (2,9)	4.5 (1,8)	0.37
Social well-being <i>median (p25, p75)</i>	3 (1.5,5.5)	1.5 (0,3)	0.04
Total CPQ <i>median (p25, p75)</i>	16.5 (10, 24)	12.5 (7,20)	0.21

\*p-value for comparison between anterior and posterior hypodontia (Mann-Whitney U test)

### 3.8 Family history of hypodontia and OHRQoL

Having a positive family history did not affect the domain or overall CPQ score (Table 3.11).

**Table 3.11: OHRQoL scores in subjects with and without a positive family history of hypodontia**

Domain	Family history		p-value*
	Positive	Negative	
Oral Symptoms <i>median (p25, p75)</i>	4 (2,5.5)	5 (3,6)	0.26
Functional limitation <i>median (p25, p75)</i>	3 (1.5,6.5)	2 (3,5)	0.89
Emotional well-being <i>median (p25, p75)</i>	6 (2.5,9)	5 (2,9)	0.62
Social well-being <i>median (p25, p75)</i>	3 (1.5,4.5)	2.5 (1,6)	0.82
Total CPQ <i>median (p25, p75)</i>	16.5 (9, 23)	15.5 (10,23)	0.97

\*p-value for comparison between participants with and without a family history of hypodontia (Mann-Whitney U test)

### 3.9 Associated dental features and OHRQoL

Subjects without other dental features reported a lower impact from hypodontia on their emotional well-being ( $p=0.02$ ). The presence of other dental features did not significantly increase the overall CPQ score (Table 3.12).

**Table 3.12: OHRQoL scores in subjects with and without other dental features**

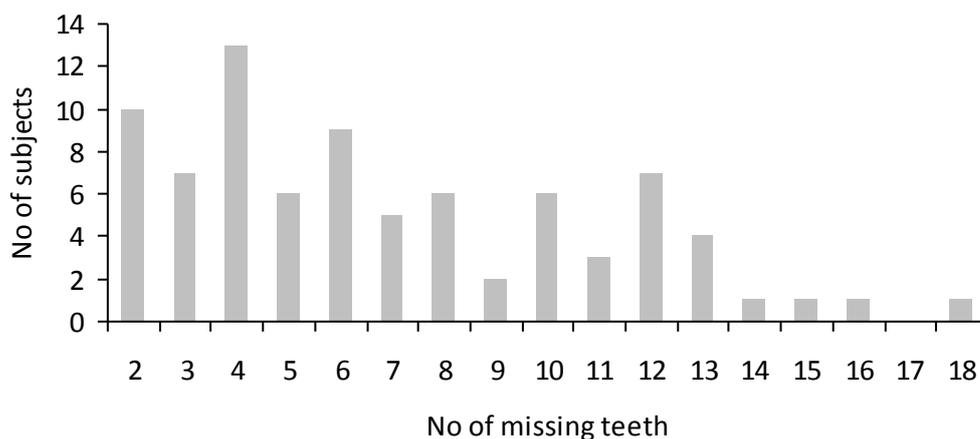
Domain	Other dental features		p-value*
	Yes	No	
Oral Symptoms <i>median (p25, p75)</i>	4 (3,7)	4 (2,5)	0.28
Functional limitation <i>median (p25, p75)</i>	3 (1,6)	2 (3,5)	0.75
Emotional well-being <i>median (p25, p75)</i>	7 (3,11)	4 (1,8)	0.02
Social well-being <i>median (p25, p75)</i>	3 (2,6)	2 (1,5)	0.21
Total CPQ <i>median (p25, p75)</i>	17 (13, 29)	14 (9,21)	0.07

\*p-value for comparison between subjects with and without other dental features (Mann-Whitney U test)

### 3.10 The relationship between the number of missing teeth OHRQoL

The mean number of missing teeth was 6.8 with a range of 2 to 18. The majority of patients had 6 missing teeth (Fig 3.1).

**Fig 3.1: The number of missing teeth**



Spearman rank correlation (Table 3.13) showed moderate correlation between the number of missing teeth and the overall CPQ scores within the total sample ( $\rho=0.351$ ,  $p<0.001$ ). There was poor correlation between number of missing teeth and CPQ scores within the hypodontia group ( $\rho=0.130$ ,  $p=0.233$  for overall score).

**Table 3.13: Spearman rank correlation between child CPQ scores (specific domains and overall score) and the number of missing teeth in the total sample and within the hypodontia group**

Domain	No of missing teeth	
	Total sample	Hypodontia group
Oral Symptoms	$\rho = 0.284$ $p = 0.002$	$\rho = -0.06$ $p = 0.57$
Functional limitation	$\rho = 0.281$ $p = 0.002$	$\rho = 0.09$ $p = 0.406$
Social well-being	$\rho = 0.270$ $p = 0.003$	$\rho = 0.146$ $p = 0.181$
Emotional well-being	$\rho = 0.249$ $p = 0.007$	$\rho = 0.180$ $p = 0.10$
Total CPQ score	$\rho = 0.351$ $p < 0.001$	$\rho = 0.130$ $p = 0.233$

### 3.11 Correlation between parental and child reported OHRQoL

Spearman rank correlation between the overall parental reported and the child OHRQoL scores (Table 3.14) was moderate ( $\rho=0.464$ ,  $p<0.001$ ). The correlation was weakest for functional limitation ( $\rho = 0.219$ ,  $p=0.05$ ). The correlation between mother's score ( $\rho=0.450$ ,  $p<0.001$ ) and father's score ( $\rho=0.488$ ,  $p=0.02$ ) was not significantly different ( $p=0.72$ ).

**Table 3.14: Spearman rank correlation between child OHRQoL scores (specific domains and overall score) and the corresponding parent scores**

Domain	Corresponding parent score
Oral Symptoms	rho = 0.517 p < 0.001
Functional limitation	rho = 0.219 p = 0.05
Emotional well-being	rho = 0.471 p < 0.001
Social well-being	rho = 0.254 p = 0.02
Total CPQ score	rho = 0.464 p < 0.001

The correlation between parental score and child score was not significantly affected by social deprivation ( $p=0.723$ ). Correlation between parental and child was similar for families with ( $\rho=0.514$ ) and without a positive family history of hypodontia ( $\rho=0.440$ ). This correlation was significant for both children with a positive family history ( $p=0.003$ ) and those without ( $p=0.001$ ).

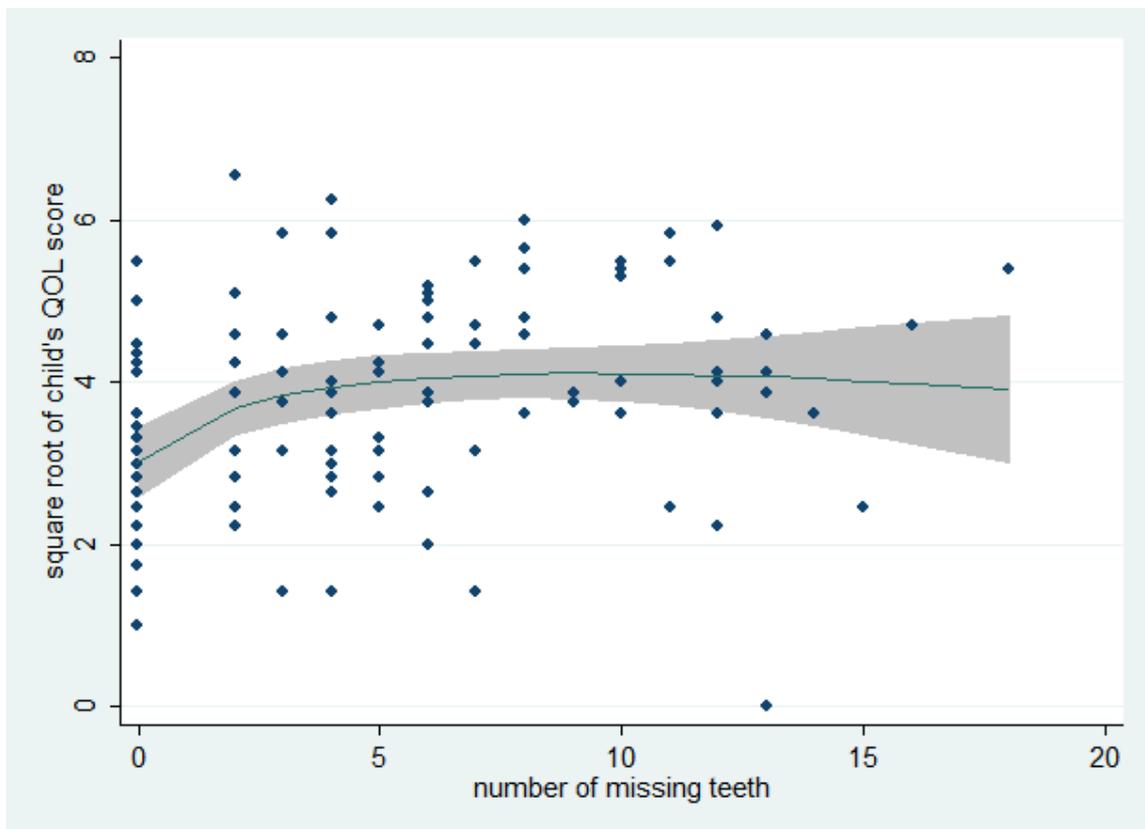
### 3.2 Regression analysis

The linear regression analysis indicated that the significant association between hypodontia and overall CPQ score (dependent variable) was independent of age, gender and ethnicity and that there was no confounding by these covariates (crude  $\beta$ -coefficient: -0.96,  $p<0.001$ , adjusted  $\beta$ -coefficient: -0.98,  $p=0.001$ ).

There was no association between number of missing teeth and overall CPQ score among children with hypodontia, independent of location of hypodontia (crude  $\beta$ -coefficient: 0.02,  $p=0.5$ , adjusted  $\beta$ -coefficient: 0.02,  $p=0.6$ ).

Fractional polynomial regression confirmed that there was a non-linear association between number of missing teeth and the CPQ score among all children. There is a qualitative jump in the QoL score between the control group and subjects with hypodontia (Fig 3.2). Within the hypodontia group, no association between number of missing teeth and CPQ score can be seen (Fig 3.2).

**Fig. 3.2 Fractional polynomial to show distribution of quality of life scores**



## **Chapter 4**

### **Discussion**

## 4. Discussion

### 4.1 Discussion

A cross-sectional study was conducted to investigate the psychosocial impact of hypodontia on a child sample. In the event 43 children with mild hypodontia, 43 with severe hypodontia and 30 without hypodontia were recruited for the study. The majority of patients in the hypodontia sample were female (Table 3.1), which is in agreement with other workers in the field (Shafi *et al*, 2008). The median number of missing teeth was 6 with a range of 2 to 18. The most common missing tooth, excluding third molars was the maxillary lateral incisor (Table 3.2). This is similar to previous studies (Shafi *et al*, 2008). Eighty per cent of patients with hypodontia had at least one missing third molar with 59% missing all four third molars. This prevalence is higher than reported in the general population (Brook, 1974) but may be because third molar development was not radiographically evident in the younger patients. In the present study patients presented with a full range of skeletal patterns although previous research has shown a predilection for specific skeletal patterns (Sarnas and Rune, 1983; Yuksel and Ucem, 1997).

Hypodontia is the most common congenital dental anomaly and a wealth of literature on the aetiology, prevalence and management of the condition has been published. However, to date, only a few studies have investigated the psychosocial impact of hypodontia (Wong *et al*, 2006; Laing *et al*, 2010, Locker *et al*, 2010). The results from the present study add weight to existing literature that hypodontia has a psychosocial impact (Wong *et al*, 2006; Locker *et al*, 2010).

The recommended minimum effect size suggested for quality of life studies is 0.5 (Cohen, 1969). The sample size calculation for this study was based on detecting a larger difference between the CPQ scores of 0.75. There have been few studies investigating the psychosocial effects of hypodontia. Wong *et al* (2006) and Locker *et al* (2010) found that hypodontia had a significant impact on the quality of life of children, however, there was no sample size calculation used in these studies and convenience sampling was used. More recently Laing *et al*, (2010) reported that hypodontia did not affect the quality of life when compared to a control group of routine orthodontic patients with a similar treatment need.

Patients with other dental conditions including caries, fluorosis and periodontal disease were excluded from the present study to avoid confounding (Gherunpong *et al* 2004; Barbosa and Gavião, 2008b). Patients with craniofacial conditions were excluded for the same reason (Jokovic *et al*, 2002; Barbosa and Gavião, 2008b). Six out of the seven studies, which have previously investigated the association between QoL and malocclusion, have reported a significant effect. Therefore, the present study recruited a control group without a significant malocclusion (Barbosa and Gavião, 2008b). This is similar to the control group used in the study by Johal *et al* (2007) as the purpose of this study was to investigate the effects of hypodontia as compared to no hypodontia.

The problems of evaluating QoL in children due to limitations in cognitive development and communication have meant that previously parents/guardians were used as proxies (Theunissen *et al*, 1998). This is affected by parental awareness and differences between child and parental reported OHRQoL exist (Jokovic *et al*, 2004). Studies have

shown that valid and meaningful information on OHRQoL can be obtained from children (Jokovic *et al*, 2002; Barbosa and Gavião, 2008a), providing that the psychometric properties of the instruments used have been tested on child populations. In the present study both parental and child reports of OHRQoL were sought to assess the relationship between the two and because research has shown that even incomplete parental reports still provide useful additional information (Barbosa and Gavião, 2008c). Parents also play an important role in the healthcare decisions of their child.

There is no gold standard for OHRQoL measurement (McGrath *et al*, 2004). In the present study, the short form of the CPQ was selected because it was originally developed for use in assessing the impact of oral conditions in adolescence a time when children often become more aware of their appearance and increasingly concerned with opinions of their peers (Jokovic *et al*, 2002). It has demonstrated criterion and construct validity and excellent reliability (Jokovic *et al*, 2002; Marshmann *et al*, 2005). The long form of the questionnaire has been shown to be valid in a number of different countries including Canada (Jokovic *et al*, 2002), Hong Kong (Wong *et al*, 2006) and the United Kingdom (O'Brien *et al*, 2006; Johal *et al*, 2007). The CPQ has also been validated for use in children with malocclusion (O'Brien *et al*, 2007) and specific occlusal traits (Johal *et al*, 2007). The CPQ has also been previously used to investigate the impact of hypodontia and this would allow meaningful comparison of the results of this study with other populations (Wong *et al*, 2006; Laing *et al*, 2010). The validated short form of the CPQ was also used to ensure a good response rate and reduce the number of missed responses (Jokovic *et al*, 2006).

The CPQ is not without limitations. It was not developed primarily for malocclusions. An OHRQoL measure specific to orthodontics may be more responsive or sensitive to clinically important changes in health. The CPQ does not include positive health concepts or open-ended questions, which may elicit experiences, not covered by the questionnaire. A further limitation of this questionnaire is that it does not elicit the specific cause for the QoL impact, which may be due to a number of oral conditions, particularly the spacing that is common in hypodontia and may not be due to functional impairment. No validated QoL instrument for malocclusion is currently available, although attempts have been made to develop this (Mandall *et al*, 2000).

Few studies have investigated the psychosocial impact of hypodontia in terms of quality of life. The present study provides additional data on the functional and psychosocial impacts of hypodontia in children and confirms the findings of other researchers (Wong *et al*, 2006; Locker *et al*, 2010). There were highly statistically significant differences between subjects with and without hypodontia. The median overall CPQ score was higher in subjects with hypodontia (CPQ=24) than the control group (CPQ=8). The impact on QoL is most likely as a result of spacing, which has been shown to result in poorer OHRQoL. The overall CPQ score is comparable to that previously reported (Wong *et al*, 2006; Laing *et al*, 2010). The differences between the hypodontia and non-hypodontia groups were highly statistically significant for the overall scores ( $p < 0.001$ ) and within the domains (Table 3.5).

The present findings are in contrast to those obtained by Laing *et al* (2010). This may be due to a lower mean number of missing teeth (4.5) compared to the present study (6.8) and differences in the control groups. The present study used children with low

treatment need whereas Laing *et al* (2010) selected children with great need for orthodontic treatment. Other dental features in these patients such as crowding or increased overjet may have impacted on quality of life. An association between malocclusion and poorer OHRQoL has been reported (Mandall *et al*, 2000). A recent study showed that adolescents who never had orthodontic treatment but had a high need were 1.43 times more likely to report one or more impact on their lives compared to children who had completed orthodontic treatment (de Oliveria and Sheiham, 2003).

It is possible that a sample group derived from the dental hospital could have resulted in some bias in this study. Such patients may report an inherently perceived need, as they are self-selected in this respect at source. It was felt, however, that selecting a control group from the dental hospital should have some equipoise for this potential bias.

The present results show that hypodontia had an impact on overall health. Twenty-nine per cent of patients with hypodontia rated the health of their teeth, lips and mouth (global oral health rating) as 'fair' or 'poor' compared to only 7% subjects in the control group. Forty-two per cent (n=36/86) in the hypodontia group were dissatisfied with their teeth compared to 16.7% in the control group (n=5/30). The global health responses showed good correlation with the overall CPQ scores ( $\rho=0.59$ ), which confirmed the construct validity of the CPQ.

Of the hypodontia sample, 29% reported experiencing oral symptoms either 'often' or 'everyday/almost everyday', 30% had experienced functional limitations, 33% reported impacts on their emotional well-being and 29% reported impacts on their social well-

being. These figures are considerably lower than those reported in the study by Wong *et al*, (2006) in which 100% of patients had oral symptoms and impacts on their social well-being and 88% reported functional limitations and affects on social well-being. This may be explained by the fact that the sample described by Wong *et al* (2006) consisted of subjects with a greater mean number of missing teeth or the criteria the authors used for reporting an impact was more lenient. It is also possible that the differences could arise due to cultural differences or as a result of the number of retained deciduous teeth, which mask masticatory difficulties that may arise from missing posterior units and poor aesthetics from missing anterior teeth.

The overall CPQ score was greater in the severe hypodontia group compared to the mild hypodontia group but this was not statistically significant. There was no relationship between the number of missing teeth and the quality of life score. The findings of this study differ from the study by Wong *et al* (2006) in that there was no correlation between the number of missing teeth and the overall CPQ score. This may be explained by differences in the number or type of missing teeth in the study by Wong *et al* (2006) study compared to the present study. Interestingly, Locker *et al* (2010) also did not find any correlation. In the study by Laing *et al* (2010) functional impairment increased with the increased number of missing teeth, once the retained deciduous teeth were accounted for. A similar finding was not seen in the present study. This may be because patients with mild hypodontia, often have missing maxillary lateral incisors resulting in labial segment spacing, which has been shown to have a significant affect on OHRQoL (Johal *et al*, 2007). It may be there is plateau affect on the OHRQoL. The absence of further teeth may also be masked by retained deciduous teeth and may therefore not

have a further impact on the OHRQoL. Although previous studies have found a greater impact on OHRQoL when the retained deciduous teeth are accounted for (Wong *et al*, 2006), there was no statistically significant effect of retained primary teeth on the CPQ score in the present study.

The inconsistencies found between the clinical data and the CPQ may be a reflection of the psychometric properties of the questionnaire or may be due to the fact that impacts are mediated by a number of factors such as culture and social deprivation (Locker, 1992).

There was no difference in the CPQ scores between males and females confirming the findings of Locker *et al* (2010), but contradicting the findings of O'Brien *et al* (2006) who reported a greater CPQ score in girls. In the present study there was no affect from ethnicity or socioeconomic status. Other studies have also found no confounding effect from social deprivation (Marshman *et al*, 2005; O'Brien *et al*, 2006; Feu *et al*, 2010).

The CPQ score was greater in subjects with anterior teeth missing but this was not statistically significant except for in the social well-being domain. Anterior hypodontia is more likely to have an impact on aesthetics, which may lead to more teasing and lower self-esteem.

In the present study there was moderate correlation between parental and child OHRQoL scores. A significant agreement between parent and child on the impact on QoL was expected because the oral condition of the child also impinges on parental experience and on that of other family members. As expected the correlation was strongest for oral symptoms and weaker for functional limitations and emotional well-

being, although still statistically significant. Perfect correlation was not seen because even when parents feel they understand children's disease related experiences, their responses reflect the truth, as they perceive it. This may not be identical to that of their children.

These findings support previous literature and the idea that parental reports should be regarded as complementary rather than a substitute to obtaining information from children (Jokovic *et al*, 2004; Theunissen *et al*, 1998). Useful information may be lost if parental reports are not obtained. In agreement with the study by Jokovic *et al* (2007) there was no significant difference between mothers' and fathers' reports and there was no confounding affect from socio-economic status.

A positive family history of hypodontia was noted in 56% of children. Shafi *et al* (2008) reported a similar figure. Having a positive family history did not affect the OHRQoL. It is reasonable to assume that in families with a positive history a stronger correlation between parental and child reported OHRQoL would have been evident, however the results of this study showed no statistically significant difference. This may be because the small sample is underpowered to detect a difference or because hypodontia and the associated problems may not have been discussed amongst families.

The overall CPQ and P-CPQ scores showed moderate correlation with the global health ratings. Seventy per cent of children reported good, very good or excellent health of teeth. Despite this 42% of patients reported that the condition of their teeth had some impact on their life overall. The reason for lack of strong agreement may be because the

CPQ was not developed specifically to measure problems related to hypodontia and some of the questions in the oral symptoms and functional problems may not be relevant to patients with hypodontia. Akram *et al* (2011) are developing a condition-specific questionnaire for patients with hypodontia. The tool has demonstrated good face and content validity but requires further testing

Although, baseline data is important to evaluate changes in QoL (Zhang *et al*, 2007a), there is a need for more longitudinal cohort studies to evaluate the affect of treatment on QoL. Meaney *et al* (2011) showed that in adults, treatment for hypodontia, reduced anxiety related to dental appearance. The question remains on how best to evaluate the impact of a specific condition. Contemporary indices in isolation are limited and have demonstrated poor correlation to QoL measures. Marshmann *et al*, (2005) found no relationship between IOTN dhc and CPQ score. This may be because IOTN also places importance on components of the malocclusion that may not be relevant to patients such as crossbites. The inclusion of a QoL measure may be a useful adjunct to the clinical indices in demonstrating treatment need and outcome. De Oliveira *et al*, (2008) investigated the effect of addition of an HRQoL measure to IOTN. Combining the OHRQoL measures with the IOTN index did not predict the outcome of the consultation and showed a significant discrepancy between assessments based on IOTN and the child's own perceived need for treatment.

The findings in this study and that reported in other investigations have highlighted the negative effect that facial and dental aesthetics can have on a child's QoL. The concept of patient-centred care involves seeking patient's perceptions and opinions on their

clinical condition and its effect on their life overall. The findings of this and other studies (Wong *et al*, 2006; Johal *et al*, 2007; Locker *et al*, 2010) highlight the negative impact that facial and dental aesthetics can have on a child's OHRQoL.

It could be considered that one limitation of the present study was the fact that subjects were taken from only one clinical setting. Mays and Pope (2000) recommended that all types of cases and settings should be included in qualitative research to make the sample more representative. However, it should be borne in mind that for conditions such as hypodontia, which are not as prevalent as other dental conditions such as malocclusion and caries, population based sampling is difficult and recruitment is often, by necessity, limited to the clinics in which these patients are referred to or treated in. It should also be borne in mind that because of the cross-sectional nature of the study, the results observed are suggestive of an association rather than evidence of causation.

Care should be taken when interpreting the results of the present study because the minimal critical difference for CPQ has not yet been determined and the scores have not yet been categorised into low, average and high. Normative age and gender specific values for OHRQoL have been established for adults, which allow meaningful interpretation of results from QoL studies. It seems that age; social class and the number of teeth were the most important in determining the impact on QoL (McGrath & Bedi, 2002). To date such data is not available for children.

## **4.2 Conclusions:**

Hypodontia significantly affects the quality of life of children but the impact is not affected by the severity of hypodontia. This study has shown agreement between the child and their parent in relation to the impact on QoL. This is an important finding as it is generally accepted that parents have a significant role in their child's orthodontic treatment.

In the current health climate, some form of 'real life' outcome measure is required but contemporary indices do not address OHRQoL. QoL instruments such as the CPQ can be useful means of obtaining the relevant information. Addressing reported impacts may also help to improve patient satisfaction with the treatment provided and service provision.

Future research comparing a hypodontia group with both a group of non-orthodontic and routine orthodontic patients utilising a hypodontia-specific measure, could help develop our understanding of these issues further.

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## Appendices

## **Appendix 1: Invitation letter to participants**

### Letter inviting subjects to take part in a study to assess the impact of missing teeth on the quality of life v1.3 13/5/2009

Dear Patient and Parent,

I am a registrar in Orthodontics and as part of my studies I am undertaking a project into the problems of hypodontia (missing teeth).

I am asking you to be involved because some of your child's teeth are missing.

If you agree to take part you will both be asked to complete a questionnaire. The answers you provide to the questions will not be shown to anyone else and will not affect your treatment. At no point will your name or contact details appear on the forms. I hope this provides reassurance that the information gained will be kept safe.

You do not have to take part if you do not wish to do so. If you would rather not answer the questions it will not affect the treatment you receive but I hope that you will and help me to learn more about the problems experienced by patients, such as yourself, with missing teeth.

This project is being supervised by Mr A Dhopaktar, Consultant in Orthodontics. If you would like to know more about it, please feel free to ask me any questions.

Thank you for your help.

Yours sincerely

Sheena Kotecha  
**Specialist Registrar in Orthodontics**

## **Appendix 2: Children's information sheet**

### **CHILDREN'S INFORMATION SHEET v1.3 6/6/2009**

**Title: A study to look at the problems of hypodontia (missing teeth) on the quality of life**

#### **PART 1: The project**

We are asking if you would take part in a research project to look at the problems associated with missing teeth.

Before you decide if you want to join in it's important to understand why the research is being done and what it will involve for you. So please consider this leaflet carefully.

#### **Why have I been invited to take part?**

You have been asked to take part because you have one or more missing teeth. Other children with missing teeth will also be asked to take part.

#### **Do I have to take part?**

No. It is up to you. If you do I will ask you to sign a form saying you are happy to take part. You will be given a copy of this information sheet and your signed form to keep. You are free to stop taking part at any time during the research without giving a reason. If you decide to stop, this will not affect the treatment you are receiving.

#### **What will happen to me if I take part?**

If you agree to take part you will be asked to answer a few questions. This should take about 15 minutes. No extra appointments will be needed.

#### **Contact details**

If you have any questions you can ask me: Sheena Kotecha.

**Thank you for reading so far – if you are still interested, please go to Part 2:**



## **Appendix 3: Parent/guardian information sheet**

### **PARENT/GUARDIAN INFORMATION SHEET v1.3 6/6/2009**

*We would like to invite you to take part in a research study. Before you decide you need to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully. Talk to others about the study if you wish. (Part 1 tells you the purpose of this study and what will happen to you if you take part. Part 2 gives you more detailed information about the conduct of the study).*

#### **Title: A study to assess the impact of hypodontia (missing teeth) on the quality of life**

#### **PART 1: The project**

##### **Why are we doing this research?**

Many children suffer with missing teeth. There has been a lot of research into the causes and treatment of missing teeth but the profession have not considered the impact that this has on the way children feel because of their missing teeth.

##### **Why have we asked your child to participate?**

We are inviting you and your child to take part in this study because your child has some missing teeth. Participation is entirely voluntary and your child's treatment will not be affected if you decide not to participate.

##### **What is involved?**

Once you have verbally agreed to participate we will obtain written consent from you and your child.

You will then both be asked to complete a questionnaire. I will be present if you have any queries.

No additional appointments are required and answering the questionnaires will take a maximum of 15 minutes.

At no point will any treatment be withheld. You may withdraw your child from the study at any time without consequence to the quality of care your child will receive.

##### **Contact Details:**

For further information about the study or for any concerns please contact:

Miss Sheena Kotecha Tel: [REDACTED]

Mrs Sarah Mckaig Tel [REDACTED]

Alternatively you may contact the paediatric or the orthodontic department on the usual number.

## **Part 2: Additional information**

### **What happens following completion of the study?**

Ideally we would like you to complete the questionnaire before your appointment. The study team will not need to contact you again however you would be able to speak to us at any time regarding the study if you wish.

### **What if there is a problem or something goes wrong?**

If you have any problems these will be seen to immediately. If you are worried about the treatment received or the way you have been treated then you may contact the study team or speak to the consultants at any time.

### **Confidentiality**

All of the information that is collected regarding the participants, during the course of the research, will be kept strictly confidential. You will not be asked to provide any personal details. Information that has been provided will be anonymised so you and your child cannot be identified from it.

### **Who has reviewed the study?**

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee to protect your safety, rights, wellbeing and dignity. This group have approved this piece of research.

### **Organisation and funding**

This study is being funded by the University of Birmingham.

### **Complaints**

If you require further advice or have concerns, independent of the research team, then you may contact the Patient Advice and Liaison Service in the first instance. They can give advice, provide information on NHS services, listen to your concerns and help to sort out problems on your behalf.

Your PALS representatives are:

██████████ (Birmingham Dental Hospital)      Tel: ██████████

**Thank you for reading this – please ask if you have any questions.**

**Appendix 4: Children's consent form**

**CHILDREN'S CONSENT FORM v1.2 31/03/2009  
(to be completed by the participant)**

**A study to investigate the problems of missing teeth**

Please answer the following:

Have you read (or had read to you) about this project? Yes/No

Has somebody else explained this project to you? Yes/No

Do you understand what this project is about? Yes/No

Have you asked all the questions you want? Yes/No

Have you had your questions answered in a way you understand? Yes/No

Do you understand it's OK to stop taking part at any time? Yes/No

Are you happy to take part? Yes/N

If **any** answers are 'no' or you **don't** want to take part, **don't** sign your name!

If you do want to take part, please write your name and today's date

Your name \_\_\_\_\_

Date \_\_\_\_\_

The doctor who explained this project to you needs to sign too:

Print Name \_\_\_\_\_

Sign \_\_\_\_\_

Date \_\_\_\_\_

Thank you for your help.

**Appendix 5: Parental consent form**

**PARENTAL CONSENT FORM v1.2 31/03/2009  
(to be completed by the participant)**

**A study to investigate the impact of hypodontia on quality of life**

1. I confirm that I have read and understand the information sheet dated.....  
(version.....) for the above study. Yes/No
2. I have had the opportunity to consider the information, ask questions and  
have had these answered satisfactorily. Yes/No
3. I understand that my participation is voluntary and that I am free to  
withdraw at any time without giving any reason, without the treatment offered  
to my child or my legal rights being affected. Yes/No
4. I consent to my child completing the questionnaire and taking part in the study Yes/No
5. I agree to take part in the above study. Yes/No

\_\_\_\_\_  
Name of Patient      Date      Signature

\_\_\_\_\_  
Name of Person      Date      Signature  
taking consent

When completed, 1 for patient; 1 for researcher site file; 1 (original) to be kept in medical notes

## Appendix 6: Hypodontia data collection proforma

### Hypodontia Data Collection Proforma

#### **Demographic details**

Patient number .....

Gender .....

Age (yrs & months) .....

Ethnicity .....

Postcode .....

Family history yes/no

#### **Dental assessment:**

Number of missing teeth .....

Site Ant. Teeth/post teeth/combination

Missing teeth .....

Missing third molars yes/no

Teeth present (including deciduous teeth) .....

Previous treatment .....

#### **Occlusal details:**

Skeletal pattern .....

Incisor relationship .....

OJ .....

#### **Other features (Including microdontia, spacing, midline diastema, hypoplasia):**

.....

## Appendix 7: Child Perceptions Questionnaire

Today's Date

### CHILD ORAL HEALTH QUESTIONNAIRE

*Hello,*

Thanks for agreeing to help us with our study!

This study is being done so that there will be more understanding about problems children may have because of their **teeth, mouth, lips and jaws**. By answering the questions, you will help us learn more about young people's experiences.

#### PLEASE REMEMBER:

- Don't write your name on the questionnaire
- This is **not a test** and there are no right or wrong answers
- Answer as **honestly** as you can. Don't talk to anyone about the questions when you are answering them. Your answers are **private**; no one you know will see them
- Read each question **carefully** and think about your experiences in the **past 3 months** when you answer
- Before you answer, ask yourself: **"Does this happen to me because of problems with my teeth, mouth, lips and jaws?"**
- Put an  in the box for the answer that is best for you

***FIRST, A FEW QUESTIONS ABOUT YOU***

**1. Would you say the health of your teeth, lips, jaws and mouth is:**

- Excellent
- Very good
- Good
- Fair
- Poor

**2. How much does the condition of your teeth, lips, jaws or mouth affect your life overall?**

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

**3. How satisfied are you with the appearance of your teeth?**

- Very satisfied
- Satisfied
- Neither satisfied or dissatisfied
- Dissatisfied
- Very dissatisfied

## ***QUESTIONS ABOUT ORAL PROBLEMS***

**In the past 3 months, how often have you had:**

**4. Pain in your teeth, lips, jaws or mouth?**

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

**5. Sores in your mouth?**

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

**6. Bad breath?**

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

**7. Food stuck in or between your teeth?**

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

**For the next question...**

*Has this happened because of your teeth, mouth, lips and jaws?*

**In the past 3 months, how often have you:**

**8. Taken longer than others to eat a meal?**

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

**In the past 3 months, because of your teeth, mouth, lips and jaws,  
how often has it been:**

**9. Difficult to bite or chew food like apples, corn on the cob or steak?**

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

**10. Difficult to say any words?**

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

**11. Difficult to drink or eat hot or cold foods?**

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

***QUESTIONS ABOUT FEELINGS***

*Have you had the feeling because of your teeth, mouth, lips and jaws?  
If you felt this way for another reason, answer 'Never'.*

**In the past 3 months, how often have you:**

**12. Felt irritable or frustrated?**

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

**13. Felt shy or embarrassed?**

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

**In the past 3 months, because of your teeth, mouth, lips and jaws,  
how often have you:**

**14. Been concerned what other people think about your teeth, mouth, lips and jaws?**

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

**15. Worried that you are not as good-looking as others?**

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

**16. Been upset?**

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

**QUESTIONS ABOUT YOUR SPARE-TIME ACTIVITIES  
& BEING WITH OTHER PEOPLE**

*Have you had these experiences because of your teeth, mouth, lips and jaws? If it was for another reason, answer 'Never'.*

**In the past 3 months, how often have you:**

**17. Avoided smiling or laughing when around other children?**

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

**18. Argued with other children or your family?**

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

**In the past 3 months, because of your teeth, mouth, lips and jaws,  
how often have:**

**19. Other children teased you or called you names?**

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

**20. Other children asked you questions about your teeth, lips, jaws or mouth?**

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

**THERE, IT'S FINISHED!**

**We appreciate the time and thought you have given to this questionnaire**

**THANK YOU FOR HELPING US**

Derived by  
Community Dental Health Services Research Unit  
Faculty of Dentistry, University of Toronto  
124 Edward Street, Toronto ON, M5G 1G6

Supported by: The Hospital for Sick Children Foundation

**Appendix 8: Parental-Caregiver Perceptions Questionnaire**

**COMMUNITY DENTAL HEALTH SERVICES RESEARCH  
UNIT FACULTY OF DENTISTRY UNIVERSITY OF TORONTO  
124 Edward Street Toronto, Ontario  
M5G 1G6**

**CHILD ORAL HEALTH QUESTIONNAIRE  
Parental report  
6-14 years**

**SUPPORTED BY THE HOSPITAL FOR SICK CHILDREN  
FOUNDATION**

## INSTRUCTIONS TO PARENTS

1. This questionnaire is about the effects of oral conditions on children's well-being and everyday life, and the effects on their families. We are interested in any condition that involves teeth, lips, mouth or jaws. **Please answer each question.**
2. To answer the question please put an  **in the box by the response.**
3. Please give the response that **best describes your child's experience**. If the question does not apply to your child, please answer with "Never".

**Example:** How often has your child had a hard time paying attention in school?

If your child has had a hard time paying attention in school because of problems with his/her teeth, lips, mouth or jaws, choose the appropriate response. If it has happened for other reasons, choose "Never".

<input type="checkbox"/>	<input type="checkbox"/>				
Never	Once or twice	Sometimes	Often	Everyday or almost everyday	Don't know

4. Please **do not discuss the questions with your child**, as we are interested only in the parents' perspective in this questionnaire.

**SECTION 1: Child's oral health and wellbeing**

**1. How would you rate the health of your child's teeth, lips, jaws and mouth?**

- Excellent       Very good       Good       Fair       Poor

**2. How much is your child's overall wellbeing affected by the condition of his/her teeth, lips, jaws or mouth?**

- Not at all       Very little       Some       A lot       Very much

**SECTION 2: The following questions ask about symptoms and discomfort that children may experience due to the condition of their teeth, lips, mouth and jaws**

*During the last 3 months, how often has your child had:*

**3. Pain in the teeth, lips, jaws or mouth?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**4. Bleeding gums?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**5. Sores in the mouth?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**6. Bad breath?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**7. Food stuck in the roof of the mouth?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**8. Food caught in or between the teeth?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**9. Difficulty biting or chewing foods such as fresh apple, corn on the cob or firm meat?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

***During the last 3 months, because of his/her teeth, lips, mouth, or jaws,  
how often has your child:***

**10. Breathed through the mouth?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**11. Had trouble sleeping?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**12. Had difficulty saying any words?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**13. Taken longer than others to eat a meal?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**14. Had difficulty drinking or eating hot or cold foods?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**15. Had difficulty eating foods he/she would like to eat?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**16. Had diet restricted to certain types of food (e.g. soft food)?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**SECTION 3: The following questions ask about the effects that the condition of children's teeth, lips, mouth and jaws may have on their feelings and everyday activities**

*During the last 3 months, because of his/her teeth, lips, mouth or jaws, how often has your child been:*

**17. Upset?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**18. Irritable or frustrated?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**19. Anxious or fearful?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

***During the last 3 months, because of his/her teeth, lips, mouth or jaws,  
how often has your child:***

**20. Missed school (e.g. pain, appointments, surgery)?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**21. Had a hard time paying attention in school?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**22. Not wanted to speak or read out loud in class?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**23. Not wanted to talk to other children?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**24. Avoided smiling or laughing when around other children?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

***During the last 3 months, because of his/her teeth, lips, mouth or jaws,  
how often has your child:***

**25. Worried that he/she is not as healthy as other people?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**26. Worried that he/she is different than other people?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**27. Worried that he/she is not as good-looking as other people?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**28. Acted shy or embarrassed?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**29. Been teased or called names by other children?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**30. Been left out by other children?**

Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**31. Not wanted or been unable to spend time with other children?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**32. Not wanted or been unable to participate in activities such as sports, clubs, drama, music, school trips?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**33. Worried that he/she has fewer friends?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

*During the last 3 months, how often has your child been:*

**34. Concerned what other people think about his/her teeth, lips, mouth or jaws?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**35. Asked questions by other children about his/her teeth, lips, mouth or jaws?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**SECTION 4: The following questions ask about effects that a child's oral condition may have on PARENTS AND OTHER FAMILY MEMBERS**

*During the last 3 months, because of your child's teeth, lips, mouth or jaws, how often have you or another family member:*

**36. Been upset?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**37. Had sleep disrupted?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**38. Felt guilty?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**39. Taken time off work (e.g. pain, appointments, surgery)?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**40. Had less time for yourself or the family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**41. Worried that your child will have fewer life opportunities (e.g. for dating, getting married, having children, getting a job he/she will like)?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**42. Felt uncomfortable in public places (e.g. stores, restaurants) with your child?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

*During the last 3 months, because of his/her teeth, lips, mouth, or jaws, how often has your child:*

**43. Been jealous of you or others in the family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**44. Blamed you or another person in the family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**45. Argued with you or others in the family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**46. Required more attention from you or others in the family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

***During the last 3 months, how often has the condition of your child's teeth, lips, mouth or jaws:***

**47. Interfered with family activities at home or elsewhere?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**48. Caused disagreement or conflict in your family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**49. Caused financial difficulties for your family?**

- Never       Once or twice       Sometimes       Often       Everyday or almost everyday       Don't know

**SECTION 5: Child's gender and age**

**a. Your child is:**

- MALE
- FEMALE

**b. Your child's age is: \_\_\_\_\_ YEARS**

**Questionnaire completed by:**

- MOTHER
- FATHER
- OTHER \_\_\_\_\_

**Date completed:**      \_\_\_\_\_ / \_\_\_\_\_ / \_\_\_\_\_  
   DAY            MONTH            YEAR

To test how good this questionnaire is at giving us the information we need, we would like a group of parents to complete it again.

Would you be willing to complete another copy of the questionnaire in the next 2 weeks?

**Yes**

**THANK YOU FOR YOUR PARTICIPATION !**

## Appendix 9: Summary statistics for CPQ by categories of group

### Summary statistics for CPQ by categories of group

Group	c1	c2	c3	c4	c5	c6	c7	c8	c9	c10
Severe,	2.2	1.5	2.5	1.1	1.0	0.9	1.5	1.6	1.9	0.7
mean	0.9	0.9	1.0	1.1	1.1	1.0	1.0	1.4	1.2	0.9
sd	1	0	1	0	0	0	0	0	0	0
min	2	1	2	0	0	0	1	0	0	0
p25	2	2	3	1	1	1	2	2	1	0
p50	3	2	3	2	2	1	2	3	2	1
p75	4	4	4	3	4	4	3	4	4	3
max										
	2.0	1.4	2.1	0.9	0.7	1.0	1.5	1.1	0.7	0.5
Mild,	1.0	0.9	1.1	0.8	0.7	1.1	1.1	1.2	0.9	1.0
mean	0	0	0	0	0	0	0	0	0	0
sd	2	1	1	0	0	0	1	0	0	0
min	2	1	2	1	1	1	1	1	1	0
p25	3	2	3	1	1	2	2	2	1	1
p50	4	4	4	3	3	4	4	4	3	4
p75										
max	1.5	1.3	1.8	0.7	0.4	0.7	1.0	0.7	0.5	0.2
	0.7	0.9	0.9	0.7	0.6	0.7	0.8	0.9	0.7	0.4
Control,	0	0	0	0	0	0	0	0	0	0
mean	1	1	1	0	0	0	0	0	0	0
sd	1.5	1	2	1	0	1	1	0	0	0
min	2	2	2	1	1	1	2	1	1	0
p25	3	4	4	2	2	2	2	3	2	1
p50										
p75										
max										

**Summary statistics for CPQ by categories of group**

Group	c11	c12	c13	c14	c15	c16	c17	c18	c19	c20
Severe,	0.7	1.2	1.4	1.6	1.3	1.1	1.3	0.9	0.5	0.7
mean	0.9	1.1	1.2	1.3	1.3	1.0	1.3	1.1	0.8	0.8
sd	0	0	0	0	0	0	0	0	0	0
min	0	0	0	0	0	0	0	0	0	0
p25	0	1	1	2	1	1	1	1	0	1
p50	1	2	2	2	2	2	2	2	1	1
p75	3	3	4	4	4	4	4	4	3	3
max	0.8	0.9	1.1	1.5	1.0	0.9	0.9	1.0	0.6	0.9
Mild,	0.9	1.1	1.3	1.4	1.2	1.2	1.2	1.2	1.1	1.1
mean	0	0	0	0	0	0	0	0	0	0
sd	0	0	0	0	0	0	0	0	0	0
min	1	1	1	1	1	0	0	1	0	1
p25	1	2	2	2	2	1	2	2	1	1
p50	4	4	4	4	4	4	4	4	4	4
p75										
max	0.6	0.4	0.5	1.0	0.8	0.4	0.9	0.5	0.4	0.5
Control,	0.8	0.6	0.9	0.9	0.8	0.6	1.1	0.8	0.9	0.7
mean	0	0	0	0	0	0	0	0	0	0
sd	0	1	0	1	0	0	0.5	0	0	0
min	1	1	1	1	1	1	1	1	0	1
p25	2	2	3	3	3	2	4	3	4	2
p50										
p75										
max										

## Appendix 2

### Summary statistics for PPQ by categories of group

Group	p1	p2	p3	p4	p5	p6	p7	p8	p9	p10	p11	p12	p13	p14	p15	p16	p17	p18	p19	p20	p21	p22	p23	p24	p25
Severe, mean	2.3	1.8	0.9	0.3	0.7	1.3	0.3	1.0	1.6	0.8	0.3	1.0	1.6	0.8	0.9	0.4	1.3	1.2	1.0	0.8	0.3	0.6	0.4	1.5	0.5
sd	1.2	0.8	1.0	0.6	0.9	1.3	0.7	1.0	1.4	1.2	0.7	1.2	1.6	1.2	1.1	0.9	1.1	1.0	0.9	0.9	0.8	1.0	0.7	1.4	0.9
min	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p25	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p50	2	2	0.5	0	0	1	0	1	2	0	0	0	2	0	0	0	1	1	1	1	0	0	0	1.5	0
p75	3	2	2	0	1	2	0	2	3	2	0	2	3	2	2	0	2	2	2	1	0	1	1	3	1
max	4	3	3	2	3	4	2	3	4	4	2	4	4	4	3	3	4	3	3	3	4	4	3	4	3
Mild, mean	2.1	1.8	0.5	0.6	0.7	0.9	0.4	1.1	0.9	0.9	0.3	0.4	0.5	0.3	0.3	0.2	1.4	1.3	1.1	0.8	0.2	0.5	0.3	0.9	0.8
sd	1.9	1.1	0.8	1.2	1.0	1.2	1.0	1.1	1.2	1.3	0.7	0.8	0.9	0.6	0.7	0.8	1.1	1.0	1.0	0.8	0.5	1.0	0.7	1.1	1.0
min	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p25	1	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p50	2	2	0	0	0	0	0	1	0	0	0	0	0	0	0	0	1	1	1	1	0	0	0	1	0
p75	3	3	1	1	1	2	0	2	2	2	0	0	0	0	0	0	2	2	2	1	0	1	0	2	2
max	4	4	2	4	4	4	4	3	4	4	3	3	3	2	3	4	4	3	3	3	2	4	3	3	4

**Summary statistics for PPQ by categories of group**

Group	p26	p27	p28	p29	p30	p31	p32	p33	p34	p35	p36	p37	p38	p39	p40	p41	p42	p43	p44	p45	p46	p47	p48	p49
Severe, mean	1.0	1.0	1.4	0.7	0.2	0.3	1.0	1.6	0.8	0.9	0.4	1.3	1.2	0.9	0.3	1.1	0.1	0.2	0.2	0.5	0.5	0.3	0.3	0.1
sd	0.9	1.1	1.2	1.0	0.7	0.7	1.2	1.6	1.2	1.1	0.9	1.1	1.0	1.0	0.8	1.4	0.5	0.5	0.7	0.9	0.9	0.7	0.7	0.6
min	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p25	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p50	1	0.5	1	0	0	0	0	0	1	0	1.5	0	0	1	0	0	0	0	0	0	0	0	0	0
p75	2	2	2	2	0	0	0	0	2	1	2	1	3	1	0	0	0	0	0	0	1	0	0	0
max	3	4	4	3	3	2	4	4	4	3	3	4	3	3	3	4	2	2	3	3	4	3	3	3
Mild, mean	1.2	1.5	1.4	0.8	0.5	0.3	0.3	0.4	1.5	0.9	1.0	0.5	1.2	0.9	0.4	1.0	0.2	0.5	0.4	0.5	0.4	0.2	0.2	0.2
sd	1.2	1.3	1.2	1.2	0.9	0.7	0.7	0.9	1.3	1.2	1.2	0.8	1.4	0.9	0.7	1.2	0.6	0.9	0.9	0.8	0.7	0.7	0.5	0.7
min	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p25	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
p50	1	1.5	1	0	0	0	0	0	1	0	0.5	0	0	1	0	0	0	0	0	0	0	0	0	0